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Assessment of malpractice litigation following spine surgery

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ABSTRACT

Medical litigation in spine surgery is a serious concern today, with a high volume of clinical negligence claims, substantial financial cost and significant burden, who is threatening the future of this surgery.

Classical spinal surgery should be performed with very well documented indication, in order to improve the expected results, with clear aims: decompression of the neural elements of the spine from tightness, stabilizing the spine to protect the nerves, eliminate the pain resulting from abnormal loading from the different movements.

Spinal surgery today means a wide analysis, understanding and realization of spinal decompression, also osteosynthesis and fusions, using high-performance gestures, with increased addressability especially in the elderly, for a varied pathology, which involves anaesthetic-surgical risks, complications. In such a context, surgical damage does not necessarily result from an error or from surgical misconduct and the surgeon is not always responsible for the damage in the absence of a proven fault in the legal sense.

The paper aims to briefly review the main problems, but also useful recommendations to meet various challenges, expectations, maintaining the quality of life of each patient, reducing risks of getting sued, also to increase the odds of a successful defence.

In conclusion: education, vigilance, improved patient-safety strategies, investigation, implementation and sharing of lessons learned from litigation claims remain important components of spinal surgeons training, to reduce future cases of negligence and improve patient care, quality of life, as many of the cases of successful litigation had a preventable cause.

Seneca: *Errare humanum est, sed perseverare diabolicum*

Murphy's law: If anything can go wrong, it will!

INTRODUCTION

Why spine surgery medico-legal aspects? Spinal surgery, made classic or minim invasive, means frequent, demanding high risks procedures, required for many people, especially for old people, performed for: trauma, degenerative, infectious, vascular, instability, functional procedures, especially for pain, as a condition: to improve the patients status, to avoid neurologic threatening of neurologic spine function.

Keywords

spine surgery,
complications,
malpractice,
medical litigation in spine
surgery,
informed consent,
medical malpractice stress
syndrome,
practical advices to avoid
spinal litigation



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Spinal surgeons, whether of orthopaedic or neurosurgical backgrounds, encountered several adverse events, despite skills, patients compliance and expectations (1).

DISCUSSION

Medical litigation in spine surgery - a high-risk malpractice specialty is particularly influenced by the current litigation climate and it means (1):

- **medical error** - a consequence of a failure of a planned action to be completed as intended or the use of a wrong plan to achieve an aim: special technologies to fulfil decompression, stabilization with motion respect, using standard or minim invasive procedures, not always available in a hospital; a physician *fails to act under the same circumstances to respect a standard of judgment, skill, diligence as any reasonable physician* (2-4). This standard will be ascertained by appointed experts who will contrast the liable medical conduct to existing practice guidelines, statements of professional organizations and societies, use of leading books, scientific treaties and published articles;
- **a breach of standard of care, negligence** - a doctor's failure to meet this standard, does not necessarily result in liability. Bad outcomes may result even with the best of medical care; also technically unsuccessful surgery does not automatically mean a breach of standard of care. It's important to explain that bad results don't equate with malpractice and good results don't mean that the patient has had the best of care! (4);
- **causation** covered a high range of causes pre, intra, postoperatively, to prove causal connection, the direct link between the negligence and damage;
- **damage**: proof of damage, directly resulted from the breach of the alleged standard of medical care

In 2005 alone, in the USA, 3,229 active certified neurosurgeons paid a total of \$28 million in malpractice claims, with the highest average payment per specialist surgeon \$465,000 (5).

1. Negligence claims in spinal surgery means more than 20,000 spinal operations/year in UK are the highest among other operations, more frequent

and expensive: +10% last 2 years (6). Spinal epidural abscess generates significantly higher plaintiff verdicts in cases with delay in diagnosis or treatment, in cases with paraplegic or quadriplegic patient (7).

2. In 2013 Hellsten et al. (8) mentioned adverse spinal events 17.4%, to a cost of \$21,000 per case, \$2.1 M per year, 1,171 bed days every year, more than 50% due to 'minor' events.
3. In 2019 for plaintiff verdicts, the mean indemnity payment was median \$753,057; for settlements, the mean indemnity payment was median \$547,935. A neurological deficit was associated with a significantly greater likelihood of a favorable outcome for the plaintiff - 52.8% versus 32.1% for plaintiffs without neurological deficit (9). The average time to a decision for defendant verdicts was 5.1 years; for plaintiff rulings, 5.0 years; for settlements, 3.4 years. Poor consent, misdiagnosis, delays in the diagnosis and the treatment of a surgical complication predict legal case outcomes favoring the plaintiff (6), also catastrophic complications are linked to large sums awarded to the plaintiff and are predictive of rulings against the physician.
4. For physician defendants, the costs of settlements are significantly less than those of losing in court (1). Incidence of spinal malpractice claims is increasing: 56% compared to 39% head and/or brain, 5% miscellaneous, with a multifactorial possible claim, many of them avoidable (10). Malpractice spinal claims are influenced by: the number and selection of patients, purpose of the procedure performed classic or minim invasive also by the social systems: compensation claims increasing the incidence rate, with a less patients intention to return to work as assurance payments are bigger (4).

In order for the doctor to be liable, the complaining patient must prove four things in the legal sense (6)(7):

- **duty** - did a physician-patient relationship exist?
- **breach of duty** - did the physician fail to meet the required standard of care - whether a body of responsible surgeons would have treated that patient the same way (Bolam test) the burden of the proof is on the plaintiff;

- **causation** - did the physician's breach cause the patient's injury?
- **damages** - did the patient incur medical expenses, pain, suffering, loss of wages as a result of the breach?

There are several causes to litigations:

A. Inappropriate decisions means:

- **poor communication despite patient's compliance, realistic expectation after surgery and the surgeon's skills** lack of respect for patient and relatives, for the surgical team, the institution prestige linked to: misdiagnosis, bad indications, "unindicated surgery" it is often referring to the lack of concordance between clinical, radiological ± neuro-physiological investigations, absence of marked psychosocial economic problems, failure of well conducted medical conservative treatment (1)(6);
- **poor documentation:** imaging's (mainly MRI) must be relatively recent (less than 3 months). In case of clinical changes or new clinical signs, MRI needs to be repeated before proceeding with the surgery (13);
- **inappropriate preoperative patient assessment:** comorbid conditions + the aging of the population - a growing concern for spine care in the 21st century (10). There are three major problems in elderly patients:
 1. *Failure due to wrong indication:* poly-morbid patient, multi-operated patient (also other than spine surgery), diabetic patient with neuropathy, severe osteoporosis: 700,000 of these occur in the spine - more than in the hip and wrist combined !, obese patient, Parkinson, depressed patient, physically non-active/active patient, patient in insurance battle waiting compensation. For elderly patients there are a very large pathology: metastatic and infectious diseases, degenerative spine: spinal stenosis, degenerative instability +/-adjacent segment problem (natural or iatrogenic), discogenic, cervical myelopathy, spondylolisthesis, arthritis, adult degenerative scoliosis, muscular insufficiency (fatty degeneration); difficult recovery (12);
 2. *Failure due to wrong biology* mostly patient and /or surgeon's judgement related; sometimes spinal surgery for geriatric patients unnecessary, too much, or too little;
 3. *Failure due to wrong biomechanics* mostly patient and also its biology and /or surgeon's judgement related (11).
- **surgeons' expertise:** a check list could be established and the steps are followed regularly before starting and during the surgery technique (sign in - prior to induction, time out - prior to skin incision, sign out - prior to drape removal), wrong judgement of own capabilities and skills, lack of expertise, wrong choices of technology (implants, instrumentation, surgical technique) + risks unexplained, failure to recognize the clinical signs and symptoms of complications, inappropriate delegation of procedures to junior doctors(13). Legal finding: the majority claims are from patients with no medical negligence, only 3% of patients who suffered negligence filed a claim. (6)

B. Failure to obtain consent (6):

Consent should be done by the consultant; to be legally valid for examination or operation should met three conditions: must be given knowingly, freely and voluntarily, the patient or legal entities must be capable of giving legal consent, the patient should be sufficiently informed to make a considered decision. It's mandatory to discuss immediately after surgical indication is sustained: surgery risks against conservative options; especially to avoid complications, expectations for patients with old, severe neurologic deficits, to patients with low resources to recuperate; ex: lumbar disc or stenosis surgery is for leg pain not back pain, surgical fusion as last resort is not wise, 10% are worse after fusion.

C. Wrong patient (14):

- **patient biology** - age, hypertension and cardio vascular disease, diabetes, patient over 60 years with hemorrhagic risks especially after Aspirin, Plavix, smoking and pulmonary disease; also ASA grad 1 - 9.1%, ASA grade 4 - 31.3%. In cases of elderly patients with spinal cord tumours (15) age increase in significant morbidity for more than 64, 5 fold; in cases with elderly patients with cervical discs and myelopathy, an operation performed to a patient aged > 65 yrs may increase mortality 14 fold, if the patient aged > 84 yrs mortality increase 44 fold.

- **adequate conservative treatment?** MRI if considering malignancy, sciatica/cauda equina syndr, infection, fracture, ankylosis, inflammatory disorders, referral for surgery (13), avoid operation delay (6)

- **planning operation: think always to do it right the first time!**

D. Wrong/stupid surgeon (14):

- **wrong-level surgery, wrong side:** always perform fluoroscopy before skin incision, before entering the canal; wrong positioning rate is estimated at 4.2%, but is found to be 15.7% on control CT;
- **wrong procedure: poor surgical performance:** inadequate decompression, **wrong technology** despite right procedure and patient
- **wise selection of radiation exposures in spinal surgery,** safe operating procedure achieving a good balance between patient care and radiation safety, minimize the high fluoro technique, always use ALARA concept: As Low as Reasonably Achievable (16)(17)
- **think to the best patient position:**
 - each position carries some degree of risk for haemodynamic and physiologic changes; also complications, risks magnified in anaesthetised patient due to loss of ability to compensate or communicate (18);
 - prone position during spinal surgery should be made with the patient's head raised, in order to minimize facial and per orbital edema, avoiding eyeball compression especially after prolonged position, which may generate peri-operative blindness and litigation both for surgeon and anesthesiologist, ex: 0.03% after spinal fusion (19);
 - all possible compression and traction points on the face, protecting neck position, trunk and limbs using protection pads to cover pressure points avoiding:
 - ulnar nerve compression at the elbow - the most common;
 - peroneal nerve injury at the fibular neck is possible in any patient positioning, inducing paresthesia or severe motor impairment with drop foot;
 - uni or bilateral femoral cutaneous nerve involvement results in meralgia paresthetica
 - brachial plexus stretch injury.

- **general anesthesia** (18) with care especially during intubation to elderly patients with myelopathy or severe cervical stenosis, hyper flexion or extension, using the Philadelphia collar with fiber optics intubation, even under intra operative monitoring; maintaining a stable hemodynamic, avoiding ischemia by hypo perfusion with medullary insults, compensating rapidly any blood loss during hemorrhagic procedures

E. Possible complications:

- **general causes** which can evolve to death (20-22): deep venous thrombosis, pulmonary embolism, cardio-respiratory failures, abdominal vessel injury if sudden unexplained drop in blood pressure, abnormal bleed coming from the disc space, ileus, sepsis with chest/urinary infections, operating room fires (19) in USA 50-100 events/yr, 1-2/yr fatal, fire triad, location: 34% airway, 28% head or face, 38% on/or inside patient, *heat/ignition source:* cautery/laser/fiber optic light cord, *fuel:* drapes/ET tube/alcohol prep/hair/O₂ build up under impervious drape, especially for cervical spine, *oxidizer:* oxygen/nitrous oxide/air.
- **acute or delayed neurologic deficits related to surgery** (23)(24):
 - direct iatrogenic medullary and radicular insults in procedures performed without spinal cord monitoring: MEPs, SSEPs, Nim Eclipse, or by hypo perfusion - arterial pressure < 80 mm Hg, especially in cases with myelopathy, medullary atrophy or ossification of the posterior longitudinal ligament estimated at 0.2 to 0.9%.
 - Cervical disk surgery with medullary decompression for myelopathy entails elevated risk, affecting the C5 root with deltoid impairment in 2.3 to 6.7% of cases (short sheath, subject to traction by medullary mobilization after decompression, inducing spinal cord retraction in surgical interventions restoring lordosis).
 - Complex surgical procedures for spinal deformity of the thoracic and lumbar spine, to correct: sagittal deformity - see three-column resection osteotomies with pedicle subtraction osteotomies and vertebral column resections may generate paraplegia 0,55% - 1,78%.
 - High grade lumbar spondylolisthesis may generate 11,8% with neurologic complications especially to the L5 root; cauda equina syndrome

following decompression for lumbar spinal stenosis, disc herniation: incidence of 2.8%.

- iatrogenic medullary and radicular insults could appear after poor positioning - estimated at 4.2%, but is found to be 15.7% on control CT or per- or postoperative implant mobilization: screws, inter body cages, a.s.o.
- subsequent stroke: estimated at 3.8% on the left and 1.8% on the right side after iatrogenic surgical trauma to the vertebral artery 0,3%;
- recurrent laryngeal nerve injury with dysphonia, as the most common postoperative complaint in anterior cervical approaches;
- Horner syndrome due to injury to the cervical sympathetic trunk is a very rare complication of anterior cervical decompression and fusion, the incidence rate is between 0, 2 to 4% mainly after revision surgery;
- retrograde ejaculation from damage to the hypo gastric plexus during anterior approach of the lumbar spine is estimated between 0, 42 to 4, 1%.
- **Local complications:**
 - implant related dysphagia in cervical disc herniation;
 - lymphocele after anterior lumbar interbody fusion;
 - dural tear (25-27) are very frequent during spine surgery (1-15%), versus 7,7% in cases with neurological complications, three-fold higher in revision surgery, may generate CSF collections (pseudomeningocele), also intracranial hypotension, acute subdural hematoma, tonsillary herniation; leak must be treated seriously and rapidly! Primary repair suturing using microtechnique, if larger than 3 mm, 6/0 suture, cover with: grafts (fat, muscle, fascia, blood patch), tissue sealants, fibrin glue, synthetic membrane; hermetic closure of the various planes, antibiotherapy are necessary, postoperative drainage after dural tear is controversial;
 - radiculopathy: recurrent disc prolapse, ectopic bone/stenosis, fragments end plate prep, symptomatic epidural adhesions, TLIF retraction, multiple surgeries, inflammatory BMP reaction, BMP Ectopic Bone (28)(29) in spinal fusion 92.8%, off label 85%, 16.6% ALIF, 30.0% PLIF/TLIF, 20.4% Post-lat, 13.6% Cervical, 3.9% T/L; no statistical correlation ectopic bone vs. increased leg pain, few cases of neurologic impairment of from ectopic bone;
 - bleeding (17)(28)(30) it can occur damaging the arterial feeder in a rich vascularized vertebral tumor,

through epidural varices, rarely by damage to an arterial vessel – see during discectomy. Post-operative bleeding could be: residual, discovered incidental or even symptomatic with different topography: epidural, paravertebral, even intradural. Main causes are: bad hemostasis, uncontrolled high blood pressure during operation, coagulopathy, drugs; ex. in cervical area is rare 0.2%-1.9%, may cause airways obstruction, requiring evacuation 0.1-0.4%; in lumbar area is about 5,6%. Best solutions are: meticulous dissecting and hemostasis, drain use, pay close attention to the patient's supervision even in ICU, quick evacuation;

- local seroma;
- instability (13)(17)(31-33) – avoid thinning the pars and aggressive facetectomy, junctional degeneration/instability (fusion disease);
- improper use of instrumentation (31-33): misplaced instrumentation - most frequent are lateral, may generate pedicle breaches 6.7% of screws; percutaneous fluoroscopically or navigation guided pedicle screw placement is safe and accurate, revision is rarely required; may increase the rate of CSF leakage, than without instrumentation 16% versus 3,5%, nonunion of the fusion or pseudoarthrosis, hardware malpositioning;
- infections (19)(28): especially in immunocompromise patients: diabetic, renal failure, HIV, etc, consider prophylactic antibiotherapy. More frequent infections appear as a complication of CSF leakage cases in nearly 1/4th of cases. There are several clinic entities: wound infection with postoperative abscess, spondylodiscitis, osteomyelitis, epidural collections, fungal meningitis: *Exserohilum Rostratum*, *Aspergillus Fumigatus*, *Cladosporium* from CSF after epidural steroid injection, 19 days post injection: fever, stiff neck;
- neuropathic pain, chronic pain corticalization, failed back surgery syndrome (13)(34)(35). Such entities can be generated by all previously mentioned complications. Patient selection is more important than most of the technical problems in FBSS (correct assessment is needed to get an accurate diagnosis, recurrent disc herniation or instability should be treated, no treatable cause, spinal cord stimulation may help improving patient's pain and functions;
- vertebroplasty may generate complications 3.7% of cases, kyphoplasty 0.3% of cases (17)(36): severe pulmonary embolism of PMMA:

hypertension, hypercapnia, loss of responsiveness, asymptomatic diffuse pulmonary embolism, neural and vascular complications after cement leakage inside the vertebral canal, silent leaks 30% - 70% of patients, most are undetected by the surgeon.

F. What kind of operation: classic or minimally invasive spine surgery. "MISS" is an acronym for the term: minimally invasive spine surgery, synonym: "LISS" - less invasive spine surgery, endoscopic spine surgery = efficient surgery with minimum of iatrogenic trauma, *but not minimal surgery*: minimal access & techniques spine surgery, percutaneous, even using microscope as a part of a less invasive technology (37-39). The current focus on "MISS" refer to several "trendy" procedures, in the early phase of a "concept shift" (39), still met with skepticism, disbelief, hostility by many neurosurgeons for safety reasons, but more accurate.

Such procedures are using advances in **access technology**: optical systems (endoscopes, microscopes), navigation guided surgery, specialized retractor systems, hybrids, robotics, **new generation of implants** suitable for minimal access by anterior/posterior surgery, **new solutions for fusion**: cages, bone harvesting tools, bone substitutes, 3-d prosthesis; designed to solve a pathological process as using standard open procedures for less pain, morbidity, disability, facilitates faster recovery, improves back muscle function.

WHAT DOES "MISS" MEAN TODAY?

Advantages (40,44):

- small incisions- more aesthetics, appealing
- some procedures can be performed as outpatient surgery;
- accurate fluoroscopic images with X ray exposure or by navigation-guidance;
- theoretical better quality of life: shorter hospital stay, structure-sparing, or perhaps structure-preserving, a "MISS" by products are operative time, reduced tissular distruction, less pressure on muscles using minimally invasive spinal retractors compared to open retractors and reduced scar (periradicular & skin), less blood loss, infection, disability; faster recovery, functional ability to return to normal activity, to work;
- long term pain control, minimal requirements for narcotic pain medications;

- may reduce or perhaps eliminate the development of adjacent segment disease;
- high expectations even to elderly, obese people with a complex spinal problem, such as deformity or trauma;

Drawbacks and limitations (45)(46):

- technically more demanding, longer operative time;
- less working space, extension and quality of direct spinal decompression, placing cross-links, long rods, less surface area of bone exposed for fusion cases
- limitations by pathology: possible spinal injuries associated with neurological deficits, see: spondyloptosis, severe multi-level stenosis, en-bloc removal of tumour masses, severe deformities;
- acute complications 10%, 12% reoperations: bleeding, 5.3% dural tear with CSF fistula, 2.6% fracture of an inferior facet - for lumbar spinal stenosis, TLIF may result in poor fusion without BMP, 10.5% transient neurological complications (47);
- radiation exposure with fluoroscopy (48) with poor radiological support - esp AP;
- education long steep learning curve, the intraoperative complication rate is highest between the 3rd and the 6th year of training;
- availability;
- cost: expensive hardware, hospital stay it could be economic (49)(50);
- "Maximal Intra-operative Surgical Stress" (51).

"MISS" recent evidence:

- **More frequent MISS indications** (12): degenerative disc disease - herniated disc, lumbar spinal stenosis, spinal deformities such as scoliosis, spinal infections, spinal instability including spondylolisthesis, vertebral compression fractures, reconstruction with internal fixation, resection of spinal or paraspinal tumours.
- **Performed with microscope or endoscope** there are several MISS procedures (12)(52)(53), ex:
 - *cervical procedures*: posterior foraminotomy, transarticular C1C2 screw fixation;
 - *thoracic procedures*: foraminal discectomies, percutaneous pedicle screw fixation;
 - *lumbar procedures*: discectomies, posterolateral interbody fusion PLIF, midline lumbar interbody fusion MIDLF, transforaminal lumbar interbody fusion TLIF, extreme lateral lumbar

interbody fusion DLIF (XLIF), anterior lumbar interbody fusion ALIF.

- **For discectomy:** micro discectomy (MD) vs tubular(52): no difference (the mean duration of surgery was 34 min shorter for conventional MD; the incidence of dural tear was 6.5% in MD group and 10.4% in early TD and decreased to 7.4% in late TD group, possible more leg pain, low-back pain, at 2 years with tubular) (53).
- **For lumbar spinal stenosis:** bilateral micro decompressive laminotomy, as minimally invasive endoscopic bilateral decompression with a unilateral approach (endo-BiDUA) for elderly patients (54)(55).
- **For posterior lumbar fusion:** significant advantages over traditional open procedures in the obese population, smaller incisions, less tissue trauma and quicker recovery, higher radiation exposure 84 s vs 37 s, may reduce or perhaps eliminate the development of adjacent segment disease (56-58). Revision TLIF identical results: less blood loss, less postoperative back pain at the second day postoperatively, same surgical time, higher radiation exposure 79 s vs 39 s (58).
- **Adult spinal deformity correction** with circumferential minimally invasive surgery and hybrid techniques result in overall reoperation rates of 27.9% and 33.8%, respectively, at minimum 2-year follow-up. Junctional failures are more common after hybrid approaches, while pseudarthrosis/fixation failures happen more often with circumferential minimally invasive surgery techniques. Early reoperations were less common than later returns to the operating room in both groups, but circumferential minimally invasive surgery demonstrated less risk of infection and early reoperation when compared with the hybrid group (60).

Practicing a good spinal surgery is not a guarantee to being hit with a medical malpractice lawsuit; nobody is immune from medical malpractice. Most spinal surgeons are not well prepared to deal with bad consequences of medical malpractice, they are perceived as a "wound to the heart", that may lead to frustrated, dissatisfied patients; also to several consequences: loss of reputation and significant supporters, lack of knowledge about the potential process and outcomes, loss of livelihood, control, assets.

Such situations may lead to "Medical Malpractice Stress Syndrome", expressed by *psychological symptoms*: anxiety, irritability, tiredness, restlessness, difficulty in concentration, excessive worry, may occupy more than 50% of working hours, tense muscles, insomnia, depression and by *physical illness*, at work and at home; dissatisfaction, to burnout of the medical practice, early retirement or even may generate extreme reaction, deep depression leading to suicide (1)(4)(59)(61).

Fear of lawsuits encourages defensive medicine, a defensive attitude, affecting patients (61): *to restrict practice*, not only to demanding procedures, also for emergency or trauma, *to move their practice to a state with "better" malpractice conditions*, in detriment of patients losing chances to receive the accurate treatment in the needed time; negatively influence any investment from the medical community, *to consider retirement, rather than continue to practice* in the face of increasing insurance costs, especially in that states with high malpractice claims and insurance premiums - so called "crisis" states.

CONCLUSIONS AND PRACTICAL ADVICES

Medical malpractice litigation in **spinal surgery** is one of the most stressful events of the life of any physician, but also a survivable and surmountable event to made you a stronger and better doctor, indeed, as Churchill said "there is only one answer to defeat, the victory". As a general remark, many claims in spinal surgery are avoidable (62-65); however, to reduce medical malpractice stress, vulnerability to potential litigation, negligence, **some advices should be retained:**

- good documentation and patient selection is required;
- the technique must be adapted both to the patient, to resources and facilities in your hospital;
- limit your activity, avoiding doing something outside of your expertise;

To reduce risk of getting sued it is necessary:

- never regret to lose effort and time to improve communication skills, communicating openly, explaining the patient to complete carefully the informed consent exposing clearly expectations, desired outcome, also possible complications. In more than 70% of litigations to improve situation, if a complication occurs, it's mandatory to justify in the record, explaining how, why it appeared;

- discuss non-surgical option, if necessary, thinking twice and choose without delay a procedure with a lower complication rate;
- achieve a safe and efficient operation .

To increase the odds of a successful defence:

- keep a good communication with the patients, their relatives; don't avoid the patients or hide the facts- all hidden information it will be found; most patients simply want to know what happened and most important knows you care (4), always remember: "the Court is after the Proof and not The Truth"; demonstrate *that what occurred was a complication rather than a deviation from the medical standard*;
- visit the patient more often! – convey empathy, welcome the patient, maintain eye contact, let them tell their story, work to help the patient recover, making him a very strong defendant *No attorney wants to put time and money necessary to pursue litigation into a case involving unavoidable complications, rather than negligence*;
- try to keep the patient away from the attorney's office to pursue the claim against you;
- in difficult cases, always think to an ancient Romanian saying: "it's better to accept a mutual agreement, instead of a fair judgment";
- avoid doing something new that you are not adequately trained for, operating in careless way, also possible conflicts of interest with the insurer;
- temperate young surgeon's enthusiasm - not a head of their surgical skill;
- be actively involved in the defense team, discuss your ideas and suggestions with your lawyer but follow their instructions;
- knowledge is power: support education, training or new techniques at any level & any age, professional support, local medical societies and associations, look for supportive 2-nd opinion from international professional medico-legal committees, seek advice from experienced colleagues, consultants, qualified malpractice lawyers experts, attend supportive educational meetings, enrol in risk management seminars, read available materials on litigation stress support, seek support education, training or new techniques at any level & any age, professional support, local medical societies and associations, look for supportive 2-nd opinion from international professional medico-legal committees.

- demonstrate that what occurred was a complication rather than a deviation from the medical standard.

CONFLICTS OF INTEREST

The authors of this paper state that they have no conflict of interests to disclosure.

ABBREVIATIONS

CT: computer tomography

MRI magnetic resonance imaging

ASA; American Society of Anaesthesiologists' classification of physical health

ALARA concept: As Low as Reasonably Achievable

TEP: pulmonary troboembolism

ET tube: endotracheal tube

MEPs: motor evoked potentials

SSEPs: somatosensory evoked potential

NimEclipse: a spinal and cranial neuro monitoring system (Medtronic)

BMP: human bone morphogenetic protein

ALIF: Anterior lumbar interbody fusion

PLIF/TLIF: posterior lumbar interbody fusion/thoracolumbar interbody fusion

FBSS: failed back surgery syndrome

PMMA: Poly methyl methacrylate

MISS: minimally invasive spine surgery

LISS: less invasive spine surgery

MD: micro discectomy

TB: tubular discectomy

endo-BiDUA: endoscopic bilateral decompression with a unilateral approach

REFERENCES

1. Daniels A.H., Ruttiman R. et al. - Malpractice litigation following spine surgery, Journal of Neurosurgery Spine 2017, 27(4):1-6.
2. Eloy JA, Svider PF, et al. - Comparison of plaintiff and defendant expert witness qualification in malpractice litigation in neurological surgery. J Neurosurg 2014, 120:185-190.
3. Daniels A.H., Ruttima R. et al. -Malpractice litigation following spine surgery, Journal of Neurosurgery Spine 2017, 27(4):1-6.
4. Iacob G, Majer C. – Reflections on medical malpractice stress disorders in Neurosurgery, Romanian Neurosurgery 2012, XIX 4: 247-250.
5. Betsy van Die - Study Analyzes How the Malpractice Environment Impacts Practicing Neurosurgeons, AANS Communications Department, 2008 28, (312) 949-3205.
6. Ameen A.A. - Negligence Claims in Spinal Surgery, 5 years Experience – 14th Dubai Spine Conference 2013.
7. DePasse J.M., Ruttiman R et al. - Assessment of malpractice claims due to spinal epidural abscess, J. Neurosurg. Spine 2017, 27(4), 476-480.

8. Hellesten E.K., Handbidge M.E. - An economic evaluation of perioperative adverse events associated with spinal surgery, *Spine J.* 2013, 13(1):44-53.
9. Ahmed S.A., DeFroda S. F. , et al. - Malpractice Litigation Following Traumatic Fracture, *J.Bone Joint Surg. Am* 2019, 3;101(7):e27.
10. Rovit R.L., Simon A.S. et al. - Neurosurgical experience with malpractice litigation: an analysis of closed claims against neurosurgeons in New York State, 1999 through 2003, *Neurosurgery* 2007 106(6):1108-1114.
11. Hamdan T.A. - Perils and Pitfalls in Cervical Spine Surgery, 14th Dubai Spine Conference 2013.
12. Aebi M. - Complex Spine Surgery in the Elderly and its Pitfalls and Complcations, 14th Dubai Spine Conference 2013.
13. Msaddi A.K., Majer C - Minimizing side effects in microdiscectomy, 14th Dubai Spine Conference.
14. Bolger C. - When Things go Wrong in Spinal Surgery, the spectre of the problem, 14th Dubai Spine Conference 2013.
15. Patil C., Patil T., et al. - Complications and outcomes after spinal cord tumor resection in the United States from 1993 to 2002, *Spinal Cord* 2008, 46, 375-379.
16. SamaraeeA.Al - Radiation Safety in Spine Surgery, 14th Dubai Spine Conference 2013.
17. Zerbi A. - Radiation exposure in spine surgery (surgeons and patients), 14th Dubai Spine Conference 2013.
18. Raof N. - Anesthesia and positioning complications, 14th Dubai Spine Conference 2013.
19. Wong D. - Patient Safety in Spine Surgery, 14th Dubai Spine Conference 2013.
20. DeVivo M.J., Black K.J. - Causes of death during the first 12 years after spinal cord injury, *Arch.Phys. Med. Rehabil.* 1993, 74 (3); 248-254.
21. Soden R.J., Walsh J. et al. - Causes of death after spinal cord injury, *Spinal Cord* 2000, 38(10): 604-610.
22. Connely N, Silverman D.G. - Review of Clinical Anesthesia, Lippincott Williams & Wilkins 2013, ch.4., 17-18.
23. Nanda A. - Complications in Neurosurgery, Elsevier 2019, 30, E-Book.
24. Roccamo G., Haydon J.H. - Medicine in the litigation process, Carswell, 1999, 395.
25. Andrews et al. -. Cerebellar Herniation and Infarction as a Complication of an Occult Postoperative Lumbar Dural Defect, *AJNR* 1995.
26. Sciubba,et al. - Acute Intracranial Subdural Hematoma Following a Lumbar CSF Leak Caused by Spine Surgery, *Spine (Phila Pa 1976)*. 2005 Dec 15;30(24):E730-2.
27. Wong et al. - Dural tears secondary to operations on the lumbar spine: Management and results, *J Bone Joint Surgery*1998, 1728-1732.
28. Stadler J.A., Wong A.P. et al. - Complications associated with posterior approaches in minimally invasive spine decompression, *Neurosurg. Clin N.Am* 2014, 25(2):233-245.
29. Haid R, Branch C Posterior lumbar interbody fusion using recombinant human bone morphogenetic protein type 2 with cylindrical interbody cages. *Spine J* 2004;4:527-539.
30. Sokolowski M.J., Garvey T.A, et al. - Prospective study of postoperative lumbar epidural hematoma, Incidence and Risk factors, *Spine* 2008, 33,108-113.
31. Abumi K. et al. - Complication of pedicle screws fixation in reconstructive surgery of cervical spine, *Spine* 2000, 25(8), 962-969.
32. Fessler R. et al. - Breach Rate of Percutaneous Lumbar Pedicle Screws Using Fluoroscopic Guidance, 14th Dubai Spine Conference 2013.
33. Alfawareh M. - Safety and Complication Avoidance in Cervical Spine Tumors Surgery, 14th Dubai Spine Conference 2013.
34. Iacob G. - The failed back surgery syndrome - Archives of the Balkan Medical Union, march 2009, vol. 44, 1, 51-55
35. Iacob G. - The failed back surgery syndrome, Conferinta Nationala a Asociatiei Romane pentru studiul durerii, Bucuresti, 26-27 octombrie 2017.
36. Sagi H.C., Yuan A.Y. - Vertebroplasty and Kyphoplasty in the treatment of osteoporotic vertebral compression fractures, in Haheer T.R, Merola A.A.- Surgical techniques for the spine, Thieme, 2003, ch.16, 79-85.
37. Szpalski M. - Does Size Matter ? What is the evidence for MISS, 14th Dubai Spine Conference 2013.
38. Jaikumar, S. et al. - Minimally Invasive Surgery of the Spine: History of Minimally Invasive Spine Surgery, *Neurosurgery* 2002, 51, 5, S2,1-S2-14.
39. Fessler R. G. - Minimally invasive spine surgery, *Neurosurgery*, 2002, 51,2, S2-iv.
40. Rosen DS, Ferguson SD, et al. - Obesity and self-reported outcome after minimally invasive lumbapapinal fusion surgery. *Neurosurgery* 2008, 63, 956-960.
41. Knight RQ et al. - Direct lateral lumbar interbody fusion for degenerative conditions: early complication profile. *J Spinal Disord Tech* 2009, 22,34-37.
42. Gorek et al. - Minimally invasive surgery of the spine: less is more. *Semin Spine Surg* 2013;23:2-8.
43. Bolger C - Minimally invasive spinal surgery, Is it worth the effort ? 14th Dubai Spine Conference2013.
44. Smith Z.A., Fessler R.G. - Paradigm changes in spine surgery—evolution of minimally invasive techniques, *Nature Reviews Neurology* 2012, 8, 443-450.
45. Park Y et Al. - The effect of a radiographic solid fusion on clinical outcomes after minimally invasive transforaminal lumbar interbody fusion. *Spine J.* 2011,11(3);:205-212.
46. Raco A. - Miss procedures, Advantages and limitations, 13th Dubai Spine Conference 2013.
47. Ikuta K., Tono O. - Surgical complications of microendoscopic procedures for lumbar spinal stenosis, *Minim Invasive Neurosurg.* 2007, 50(3), 145-149.
48. Bindal RK et al.- Surgeon and patient radiation exposure in minimally invasive transforaminal lumbar interbody fusion. *J Neurosurg Spine*, 2008 ,9(6),570-573.
49. Ritz JP. - MIS and the economics of it. Can MIS be cost efficient from a bussiness point of view?, *Chirurg.* 2007, 78(6), 501-504, 506-510.
50. Van den Akker ME et al. - Tubular discectomy vs

- conventional micro discectomy for the treatment of lumbar disk-related sciatica: cost utility analysis along side a double-blind randomized controlled trial, *Neurosurgery* 2011;69:829-35.
51. Hamid N. A. - Safety and Limitations of MIS in Lumbar Spine Surgery 14th Dubai Spine Conference 2013.
 52. Bhatia P.S., Chhabra H.S. et al. - Microdiscectomy or tubular discectomy: Is any of them a better option for management of lumbar disc prolapse, *J Craniovertebr Junction Spine* 2016, Jul-Sep; 7(3): 146-152.
 53. Arts M.P., Brand R. et al. - Tubular discectomy vs conventional microdiscectomy for the treatment of lumbar disk herniation: 2-year results of a double-blind randomized controlled trial, *Neurosurgery* 2011 Jul;69(1):135-144.
 54. Celik S.E. et al. - Microdecompressive laminotomy with a 5-year follow-up period for severe lumbar spinal stenosis *J Spinal Disord Tech*, 2010.
 55. Komatsu J., Muta T. et al. - Tubular surgery with the assistance of endoscopic surgery via a paramedian or midline approach for lumbar spinal canal stenosis at the L4/5 level, *Journal of Orthopaedic Surgery* 2018, 26(2), 1-8.
 56. Rosen D.S., Ferguson S.D. et al. - Obesity and self-reported outcome after minimally invasive lumbapinal fusion surgery, *Neurosurgery* 2008, 63, 956-960.
 57. Knight R.Q. et al - Direct lateral lumbar interbody fusion for degenerative conditions: early complication profile, *J Spinal Disord Tech* 2009, 22, 34-37.
 58. Wang J Zhou Y. et al. - Comparison of one-level minimally invasive and open transforaminal lumbar interbody fusion in degenerative and isthmic spondylolisthesis grades 1 and 2, *Eur Spine J* 2010, 19, 1780-1784.
 59. Makhni M.C., Park P.J. et al. - The medico-legal landscape of spine surgery: how do surgeons fare?, *Spine J.* 2018, Feb;18(2), 209-215.
 60. Eastlack RK, Srinivas R, et al. - Early and Late Reoperation Rates With Various MIS Techniques for Adult Spinal Deformity Correction, *Global Spine J.*, 2019;9(1):41-47.
 61. Lekovic G.P., Harrington T.R. - Litigation of missed cervical spine injuries in patients presenting with blunt traumatic injury, *Neurosurgery* 2007, 60:516-523.
 62. Fager C.A. - Malpractice issues in neurological surgery, *Surgical Neurology* 2006, 65(4): 416-421.
 63. Steele L., Mukherjee S. et al. - Extent of medicolegal burden in neurosurgery - An analysis of the National Health Service Litigation Authority Database, *Br J Neurosurg*, 2015;29(5):622-629.
 64. Hamdan A, Strachan RD et al. - Counting the cost of negligence in neurosurgery: Lessons to be learned from 10 years of claims in the NHS, *Br J Neurosurg*, 2015; 29(2):169-177.
 65. Banna SE, Catano A. - Complications of Spine Surgery and Litigations - Managing Malpractice Risk. *J ClinExpOrthop*. 2015, 1:3. doi: 10.4172/2471-8416.100003.



Concomitant orbital and intracranial abscess. A rare complication of sinusitis

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ABSTRACT

Background: Intracranial and orbital abscesses in combination together are rare complications of sinusitis. They can be life-threatening and can result in multiple sequelae.

Case presentation: A 9-year-old female presented with left periorbital swelling, gaze restriction and headache. Following scans, she underwent emergency endoscopic sinus surgery, evacuation of the intraorbital empyema and stereotactic minicraniectomy with the evacuation of the extradural empyema as a joint case. The patient recovered well and was discharged to complete intravenous antibiotics for 6 weeks.

Conclusion: In the pediatric population intracranial complications of acute sinusitis can have more devastating consequences. Therefore, prompt recognition and management are essential within a multidisciplinary team setting.

We also highlight the rarity of concomitant multi-site abscess formation and the need to be vigilant for same.

INTRODUCTION

Acute or chronic sinusitis complications can range from minor (headaches, flu symptoms) to loss of organ function (loss of vision) or can be life threatening (brain abscess). Early diagnosis and treatment is essential to prevent sequelae. A high index of suspicion is needed when assessing and treating the paediatric population in this setting.

In the paediatric setting paranasal sinuses have not completely developed until sometimes early in the teenage years. This is particularly the case particularly with frontal sinus development in the

Keywords

brain abscess,
craniectomy,
endoscopic sinus surgery,
infectious disease,
rhinosinusitis,
orbital abscess,
Streptococcus intermedius



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paediatric population. Indeed, in this population group, frontal sinuses are the commonest to cause significant complications, especially after the age of 10 (10,16). This is mainly due to their vicinity with the orbit and the orbital content but also with the brain and meninges.

The lamina papyracea (a thin bony plate that separates the orbital content from the nasal cavity) is very thin and often dehiscent, allowing for direct infection spread between the two anatomical regions. Orbital complications alone are more common in patients younger than 5 years of age (16). So preseptal and orbital cellulitis, followed by subperiosteal and orbital abscess are the most common complications secondary to acute or chronic sinusitis. This can lead to compromised vision. Cavernous sinus thrombosis is the most serious of all, with intracranial spread of infection and possible contralateral orbital symptoms. In the paediatric population, periorbital swelling is the most common sign that brings the patient to hospital or their family doctor's attention. This is usually associated with upper respiratory upset, fever and headache as the most common but nonspecific symptoms (16). Typical nasal symptoms seen in the adult population (nasal blockage or discharge, postnasal drip and pain over the cheeks or frontal headache) are sometimes lacking. Also a history of previous nasal complaints is absent in most cases (14).

Extension of the abscess to the intracranial cavity is usually gained through the valveless venous system, direct extension through a previous dehiscent or eroded skull base, or due to septic emboli. Subdural empyema is the most common intracranial but extra axial complication, followed by subdural and brain abscess. All are severe complications and can be life threatening or have major sequelae (hemiparesis, aphasia or epilepsy). In the paediatric population the consequences of such complications can affect patients for the rest of their life and can be debilitating and life changing for their families too.

Simultaneous intraorbital and intracranial complications secondary to acute sinusitis is very rare. This was reported as 9.3% of their patients over the age of 7 with intraorbital complications by Hermann (12).

A high index of suspicion for intracranial complications needs to be maintained and regular

assessment of the child is mandatory to check for any deterioration in their symptoms and signs or failure to improve.

Hermann et al (12) found that children with intraorbital complications of sinusitis benefit from performing a MRI brain early due to a higher association with intracranial infection in:

- children older than age 7,
- males,
- those with neurological status changes,
- frontal sinus opacification on CT,
- superior or lateral position of orbital abscess,
- intraorbital abscess needing drainage.

Sinus infection may progress to intracranial infection even after appropriate empiric antibiotic therapy has been initiated (14,17,19). Lack of early recognition and urgent treatment can be fatal. Empirical antibiotic treatment is generally broad-spectrum and started under local microbiological guidelines and supervision.

Timing of surgical intervention is a controversial subject, with multiple studies being in favour of early rather than late surgical intervention (14,17).

Intraorbital abscess formation needs to be drained in conjunction with an otorhinolaryngologist (ORL) performing endoscopic sinus surgery and intracranial abscess formation needs prompt neurosurgical input and craniotomy with drainage of the infection.

CASE PRESENTATION

A 9-year-old female patient presented to the emergency department with a 2 week history of flu symptoms and headaches. She complained of right upper eyelid oedema prior to presentation that subsided followed by persistent left periorbital oedema and ophthalmoplegia. Prior to her hospital presentation, our patient has been already on 5 days of oral antibiotics started by her family.

Her medical history was non-contributory with no history of nasal symptoms or allergic rhinitis, nor any history of atopy. The child lived with her family in America and was on holiday visiting her relatives.

An initial assessment showed pain on eye movement with no focal neurological signs with a Glasgow coma scale (GCS) of 15 out of 15. The patient had neither neck stiffness nor photophobia. She was afebrile with stable vital signs. Intravenous access

was obtained and routine bloods were sent to the laboratory.

She was admitted with a diagnosis of preseptal cellulitis for intravenous antibiotics as per our local guidelines.

On admission her inflammatory markers were elevated. White cell count was 18.2 [normal 6.0-18.0($\times 10^9/l$)], with neutrophilia of 13.9 [normal 1.0-8.5($\times 10^9/l$)]. Her C-reactive protein (CRP) was 162 (normal 0-10mg/l).

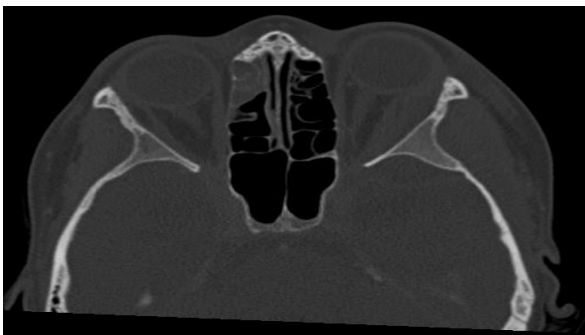
Despite 24 hours of intravenous Cefotaxime, she developed restriction of eye movement on elevation and downward gaze combined with drowsiness. Otorhinolaryngology was consulted and flexible nasal endoscopy did not reveal any intranasal pathology. An urgent ophthalmology consult was requested and vision was found normal in both eyes.

Our patient underwent urgent computer tomography (CT) of sinuses without contrast. The scan showed pansinusitis on the right side (maxillary, ethmoid and frontal sinuses) with left sided sinuses free of mucosal disease. Also she had a small left extraconal fluid collection (measuring 7x 15 x 25mm).

Pictures 1 and 2 shows the above findings on a coronal and axial CT cut:



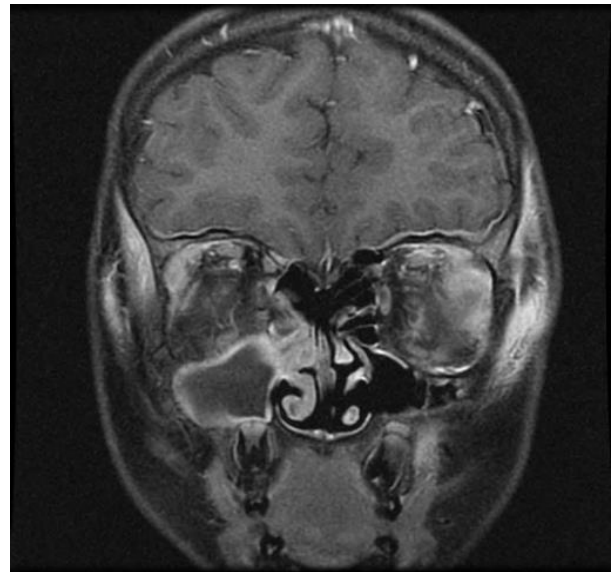
Picture 1. Coronal CT sinuses.



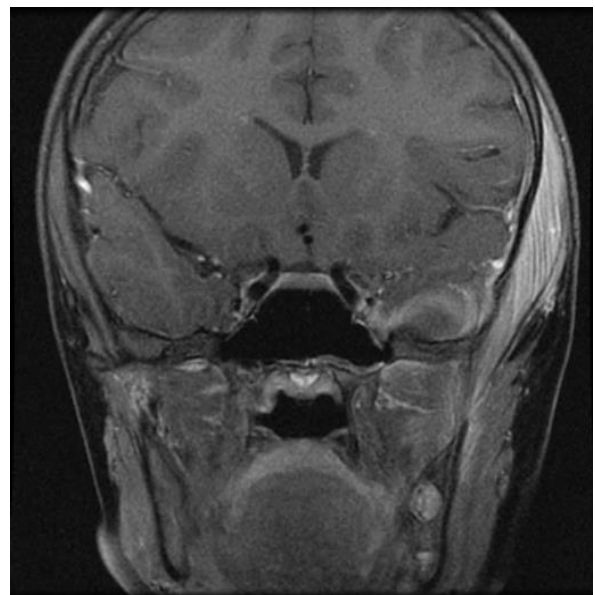
Picture 2. Axial CT sinuses.

Due to the high clinical suspicion of intracranial pathology and increased drowsiness an urgent magnetic resonance imaging (MRI) of brain with contrast was performed. This revealed the left orbital extraconal abscess with extension through the posterolateral orbital wall into the extradural space anterior to the left temporal lobe. The bone demonstrated osteomyelitis. There was also a small extracranial component adjacent to the pterion, with inflammatory changes in the left temporalis muscle.

All these findings are visible in the Pictures 3 and 4:



Picture 3. Coronal MRI showing left external orbital abscess.



Picture 4. Coronal MRI with contrast showing left extradural empyema.

After an urgent neurosurgical consult emergency surgery was planned as soon as possible. Endoscopic sinus surgery followed by left orbit abscess drainage and stereotactic mini-craniectomy with empyema evacuation was performed.

After the patient was prepped as per endoscopic sinus surgery protocol a left maxillary antrostomy was performed where frank pus was found. Similar findings were found in the left anterior ethmoidal sinus. On exploring the left posterior ethmoid only mucosa disease was found. Sinus surgery found normal sinuses on the right side despite the abnormal findings (pansinusitis) on the CT on this side.

The left external orbital abscess was drained via an external approach and a drain was left in situ.

A stereotactic mini-craniectomy was performed via a left curved pterional incision, followed by evacuation of the empyema and again a drain was placed. Pus for culture and sensitivity was sent from all 3 locations.

The patient tolerated the procedure well and no perioperative complications were encountered. No intensive care unit monitoring was needed postoperatively for our patient. She was commenced on intravenous Ceftriaxone and Metronidazole.

The approach to this case was a multidisciplinary one with Paediatricians, ORL, Neurosurgery, Ophthalmology, Infection Disease, microbiology and physiotherapy involved in this child's care.

Our patient recovered well from a neurosurgical and ORL point of view but was quite slow from an eye perspective. The intraorbital drain was removed after 12 hours and the intracranial drain after 24 hours. No temperature spikes were recorded postoperatively.

A repeated MRI brain was performed on day 2 postoperatively due to slow intraorbital progress and this revealed a small collection at the site of previous orbital abscess. Close ophthalmology review followed until resolution of the collection, 2 and half weeks after the surgery.

Culture from all sites grew *Streptococcus intermedius*. Inflammatory markers returned to normal values. A repeat MRI scan at 18 days postoperatively showed near complete resolution of the remaining collection.

The patient was hospitalized for 4 weeks duration and further intravenous antibiotics were arranged for 6 more weeks at home. During this period, our

patient was reviewed in multiple speciality clinics. She improved totally, with no neurological sequelae. A repeat MRI scan at 2 months postoperatively showed complete resolution of the collection and osteomyelitis.

Due to the fact that our patient lived in another country, further follow up was not possible but all images, documentation and advice on follow up were given to organise ORL follow up in her home country.

DISCUSSION

Simultaneous intraorbital and intracranial complications secondary to sinus disease are extremely rare. The incidence is unknown with only few cases in the literature (8,9,12,18,22) and a handful of case presentations from paediatric population (4,25).

Yeh presented the case of 17 years old male with longstanding orbital symptoms (1 month) treated with sinus surgery who developed 1 month after the treatment an inflammatory frontal mass needed excision with no neurological sequels (25).

Constantin et al (4) reported the case of a 12 years old male patient with sinusitis complicated later by orbital cellulitis ending developing frontal brain abscess. Sinus surgery and adenoidectomy was followed at 24 hours by neurosurgical intervention with collection excision.

Four retrospective studies presented the presented symptoms and management of their patients. Hermann et al (12) found a rate of 10% (4 patients) in his study of paediatric patients with simultaneous intraorbital and intracranial complications. All were males and the youngest was 14 years of age. They were treated surgically and all of the abscesses were found to be polymicrobial. He found that MRI helped identifying the intracranial complications after they were missed on CT. As a significant finding, in this study all patients had either a superior or lateral intraorbital located complication.

In his study Germiller et al (8) presented 7 cases of concomitant orbital and intracranial complications treated surgically with a focus on intracranial complications and outcome.

In her retrospective review of 10 patients younger than 18 years, Reynolds et al (18) found the youngest to be 7 years old and predominantly males. Also she

found that frontal lobe was involved in 8 out of 10 patients.

Goyita et al (9) found 14 paediatric patients to have intraorbital and intracranial complications, with a predominant male distribution in this group.

Our patient (age 9) was amongst the youngest ones from the previously reported cases.

An age greater than 7 was found to be associated with the development of sinogenic intracranial complication rather than intraorbital ones alone (5,14,17) but also with dual orbital and intracranial abscesses (11).

Our patient was a young female, although male adolescents were found to be more prone to intracranial complications alone and in association with intraorbital ones for reasons not yet elucidated (8,9,13,17,19,25). If there is a hormonal rather than an anatomical component it is to be discovered.

CRP, an inflammatory marker was found to be higher in children with brain empyema complicating rhinosinusitis than in uncomplicated rhinosinusitis, with a mean of 18.1 mg/dl (17).

While CT scan of the paranasal sinuses is the gold standard imaging modality in acute or chronic rhinosinusitis, used by surgeons worldwide to assess sinus disease, MRI brain with contrast remains the gold standard in identification of brain abscesses (9,12,21). This should be requested for patients with increased risk factors of intracranial disease or if any suspicion of intracranial complication exists. It should be performed early rather than later in the course of the disease.

Our patient's CT sinuses failed to show the true extent of the sinus disease. The scan revealed right sided pansinusitis and during endoscopic sinus surgery all these sinuses were free of disease. The left maxillary sinus seemed free of disease on the scan but frank pus was seen during surgery. Why there was such a non concordant finding it is not clear. The subsequent MRI brain showed clearly the presence and extent of the intracranial collection.

As demonstrated by multiple studies, CT sinuses findings are not absolutely accurate when compared with the endoscopic sinus surgery findings (2,20).

Another unique aspect of the case was the bacteria grown which was *Streptococcus intermedius*. The most common bacteria in uncomplicated sinusitis in children is *Haemophilus Influenzae* followed by *Streptococcus pneumoniae*, *Staphylococcus aureus*, *Staphylococcus epidermidis*,

and *Streptococcus pyogenes* (13,23). Mixed aerobic and anaerobic cultures are commonly seen also (12,23).

Streptococcus intermedius is a subspecies of *Streptococcus milleri*. It is highly pathogenic via multiple mechanisms. It is associated with deep, complicated abscesses including cerebral abscess as a complication of sinusitis (5,6,23). This could explain the aggressive course of this case.

Prompt recognition and aggressive medical and surgical treatment was the key to a complication free and smooth recovery. The role of early multidisciplinary surgical intervention was most useful in this case with all 3 specialities involved. Usually a third-generation cephalosporin plus metronidazole is most effective in fighting these types of complicated infections (7,24), but needs to be guided by local microbiology guidelines.

The role of surgery, although vital in some cases, still needs to be explored in finding the best technique and timing. The evidence in the literature is mainly based on retrospective reviews and case series. There is a lack of significant evidence in favour of one surgical treatment modality over the other (burr hole drainage, excision or stereotactic surgery) when analyzing their outcomes (1,15). The role and timing of endoscopic sinus surgery needs to be explored more, on larger samples to assess the full benefit these patients can gain. There seems to be some benefit in avoiding neurosurgical intervention if sinus surgery is performed first (3) but this was not reflected in all studies. We advocate that the focus of infection needs to be addressed first and at the same time as sinusitis being the „driver“ of the multicompartmental infection. While very small intracranial infections can be managed conservatively, any large intracranial collection needs to be evacuated or reduced due to the risk of spread, thrombosis and poor blood-brain barrier penetration of a lot of antibiotics.

Each case has to be individually evaluated and judged as no one procedure fits all.

Multidisciplinary input is common in paediatric practice but proved essential in this patient's care.

CONFLICTS OF INTEREST

Authors have no financial or other conflicts of interest to declare.

Authors have not received any financial sources of any kind and form.

REFERENCES

1. Arlotti M, et al. Consensus document on controversial issues for the treatment of infections of the central nervous system: bacterial brain abscesses. *Int J Infect Dis.* 2010;14:579-92.
2. Bair-Merritt M.H, Shah S.S, Zaoutis T.E, Bell L.M, Feudtner C, Suppurative Intracranial Complications Of Sinusitis In Previously Healthy Children *The Pediatric Infectious Disease Journal:* 2005, 24 (4):384-386.
3. Berdouk S, Pinto N, Fatal orbital cellulitis with intracranial complications: a case report, *Int J Emerg Med,* 2018, 11 (1), 51.
4. Constantin F, Niculescu PA, Petre O, et al. Orbital cellulitis and brain abscess - rare complications of maxillo-spheno-ethmoidal rhinosinusitis. *Rom J Ophthalmol,* 2017, 61 (2), 133-136.
5. Fabian J. S. van der Velden, Alexandra Battersby, Lucia Pareja-Cebrian, Nicholas Ross, Stephen L. Ball, and Marieke Emonts, Paediatric focal intracranial suppurative.
6. Faden, Howard S, Infections Associated with *Streptococcus intermedius* in Children, *The Pediatric Infectious Disease Journal,* 2016, 35(9):1047-1048.
7. Felsenstein S, Williams B, Shingadia D, Coxon L, Riordan A, Demetriades AK, Chandler CL, Bassi S, Koutoumanou E, Stapleton S, Sharland M, Bryant PA (2013) Clinical and microbiologic features guiding treatment recommendations for brain abscesses in children. *Pediatr Infect Dis J* 32(2):129-135.
8. Germiller J.A., Monin D.L, Sparano A.M, Lawrence W. C. T, Intracranial Complications of Sinusitis in Children and Adolescents and Their Outcomes, *Arch Otolaryngol Head Neck Surg.* 2006; 132(9):969-976.
9. Goytia V.K, Giannoni C.M, Edwards M.S, Intraorbital and Intracranial Extension of Sinusitis: Comparative Morbidity, *J Pediatr,* 2011, 158 (3), 486-91.
10. Hamdy El-Hakim, Anita C.Malik, Keith Aronyk, Edmund Ledi, Ravi Bhargava, The prevalence of intracranial complications in pediatric frontal sinusitis, *Int J Pediatr Otorhinolaryngol.* 70(8) (2006) 1383-1387.
11. Hartstein ME, Steinvurzel MD, Cohen CP. Intracranial abscess as a complication of subperiosteal abscess of the orbit. *Ophthalmic Plast Reconstr Surg.* 2001 Nov;17(6):398-403.
12. Herrmann B.W, Forsen J.W, Simultaneous intracranial and orbital complications of acute rhinosinusitis in children, *Int J Pediatr Otorhinolaryngol.* 68 (5) (2004) 619-625.
13. Jiannetto DF, Prett MF, Correlation between preoperative computed tomography and operative findings in functional endoscopic sinus surgery, *Laryngoscope.* 1995;105(9): 924-927.
14. Johnson DL, Markle BM, Wiedermann BL, Hanahan L. Treatment of intracranial abscesses associated with sinusitis in children and adolescents. *J Pediatr.* 1988 Jul;113(1 Pt 1):15-23.
15. Leys D, et al. Management of focal intracranial infections: is medical treatment better than surgery? *J Neurosurg Psychiatry,* 1990; 53:472-5.
16. Kou Y.F, Killeen D, Whittemore B, Farzal Z, Booth T, Swift D, Berg E, Mitchell R, Shah G, Intracranial Complications of Acute Sinusitis in Children: The Role of Endoscopic Sinus Surgery, *Int J Pediatr Otorhinolaryngol* 2018, 110(7):147-151.
17. Patel N.A, Garber D, Hu S, Kamat A, Systematic review and case report: Intracranial complications of pediatric sinusitis, *Int J Pediatr Otorhinolaryngol,* 2016 Jul; 86:200-12.
18. Reynolds DJ, Kodsí S.R, Rubin S.E, Rodgers I.R, Intracranial Infection Associated With Preseptal and Orbital Cellulitis in the Pediatric Patient *J AAPOS,* 2003, 7 (6), 413-7.
19. Rosenfeld EA, Rowley AH, Infectious intracranial complications of sinusitis, other than meningitis, in children: 12-year review. *Clin Infect Dis.* 1994 May;18(5):750-4.
20. Singhal P, Sonkhya N, Mishra P, Srivastava SP. Impact of anatomical and radiological findings for consideration of functional endoscopic sinus surgery. *Indian J Otolaryngol Head Neck Surg.* 2012;64(4):382-385. doi:10.1007/s12070-011-0351-2.
21. Skelton R, Maixner W, Isaacs D, Sinusitis-induced subdural empyema, *Arch Dis Child* 1992; 67:1478-80.
22. Traficante D, Riss A, Hochman S. Bifrontal brain abscesses secondary to orbital cellulitis and sinusitis extension. *Int J Emerg Med.* 2016;9(1):23.
23. van der Velden FJS, Battersby A, Pareja-Cebrian L, Ross N, Ball SL, Emonts M. Paediatric focal intracranial suppurative infection: a UK single-centre retrospective cohort study. *BMC Pediatr.* 2019;19(1):130. Published 2019 Apr 25. doi:10.1186/s12887-019-1486-7.
24. Widdrington JD, Bond H, Schwab U, Price DA, Schmid ML, McCarron B, Chadwick DR, Narayanan M, Williams J, Ong E, Pyogenic brain abscess and subdural empyema: presentation, management, and factors predicting outcome, *Infection.* 2018 Dec;46(6):785-792. doi: 10.1007/s15010-018-1182-9.
25. Yeh CH, Chen WC, Lin MS, et al. Intracranial brain abscess preceded by orbital cellulitis and sinusitis. *J Craniofac Surg.* 2010;21(3):934-6.



Presentation, management and outcomes of pituitary adenomas. A 10-year experience from a single tertiary neurosurgery centre

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ABSTRACT

Introduction: One of the most frequently encountered intracranial tumours are the pituitary adenomas, these accounting for 5% to 20%. Therapeutic strategies vary largely, from medical therapy to complex neurosurgical procedures. The transsphenoidal approach can solve most of the lesions of the pituitary area, as long as the invasion of the adjacent structures is not significant. The transcranial approach is indicated in tumours with extensive invasion

Materials and methods: We performed a retrospective study in the Bagdasar-Arseni Emergency Hospital Neurosurgery that aimed to analyze the demographics, signs and symptoms, therapeutic strategy, surgical approach, complications, and histopathology, from 2010 to 2019.

Results: The total number of hospitalization records, including the follow-up hospitalization and/or second surgery hospitalization, was of 1107. Furthermore, there were 704 unique patients. The most common signs and symptoms encountered for the first admission were headache (245 – 34.56%), optic chiasm deficits (153 – 21.58%), acromegaly (85 – 11.99%). However, the majority of patients (507 – 71.51%) presented with some sort of hormonal imbalance or diabetes insipidus. On the one hand, a number of 325 (45.84%) patients had non-surgical treatment. On the other hand, a total of 384 (54.16%) surgeries for pituitary tumours were performed in this period.

Discussion: In our study, the patients who underwent surgery benefited from either microsurgical transsphenoidal or transcranial surgeries. Even if the transsphenoidal approach was used far more, there was a greater relapse proportion in these patients. Transcranial surgery, even if followed by a far less proportion of relapse surgery, carried with it the burden of more days spent in hospital (most of the time twice as much as for the transsphenoidal patients).

Keywords

pituitary adenoma,
transcranial approach,
transsphenoidal approach



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Conclusion: Both transsphenoidal and transcranial approaches have advantages and disadvantages, thus the best strategy would be to tailor each surgery to each patient, keeping an open mind to all available approaches

INTRODUCTION

One of the most frequently encountered intracranial tumors are the pituitary adenomas, these accounting for 5% to 20% of all tumor lesions at this level. Most of the developing countries of the world estimate an incidence of about 20 cases per 100.000 population (1), while European developed countries agree on 3,9-4 new cases per 100.000 people each year, the prevalence hovering between 78 and 94 cases per 100.000 people.

While most of the tumors that originate in the pituitary gland are usually benign, the behavior of these lesions can be unpredictable. Thus, one can encounter anything from clinically silent, indolent tumors, all the way to extremely aggressive malignancies. Therefore, a meticulous understanding of the anatomy of the area and also of the specific physiology of the hypothalamo-pituitary complex is mandatory. (2) The therapeutic strategies vary largely, from medical therapy to complex neurosurgical procedures, depending on the aforementioned characteristics of the lesions.

As far as signs and symptoms are concerned, these again vary according to the nature of the tumor and the local extension. The clinician has to look for hormonal dysfunction, visual field deficits, decreased visual acuity, anterior pituitary dysfunction and headache. (3) The usual therapeutic strategy is to try to medically manage any case. If this is not enough or neurological/hormonal deficits occur, the obvious step is taken towards surgical management. At this stage, the neurosurgeon has to decide which surgical approach and technique best tailors every case: transcranial, transsphenoidal microsurgical or transsphenoidal endoscopic, as well as any combined approach that he or she might find adequate.

Regarding surgical approach used, one must look back to the era of Schloffer (1907), Hirsch (1910s) and Cushing (1920s). They were the ones who paved the road for the transsphenoidal approach for pituitary lesions. However, in the first half of the 20th century, due to lack of antibiotic therapy, the results were dismal. Hence, Cushing abandoned the technique and favored the transcranial approach. However,

with introduction of the operating microscope, endoscope and development of powerful antibiotics, the transsphenoidal approach soon regained attention. Nowadays, there is a general consensus that the transsphenoidal approach can solve most of the lesions of the pituitary area, as long as the invasion of the adjacent structures is not significant. Furthermore, an experienced neurosurgeon, more familiar with the transcranial approach, will usually have similar results by using this method. Therefore, as long as the lesion permits both surgical strategies, it usually comes down to surgeon preferences and experience. Non the less, the more complex the lesion, the more surgeons prefer the transcranial approach. (3).

MATERIALS AND METHODS

This is a retrospective study conducted in the Bagdasar Arseni Emergency Hospital Neurosurgery Departments 3 and 4 that aims to analyze the demographics, signs and symptoms, therapeutic strategy, surgery approach used, complications associated with these approaches, histopathology of pituitary tumors, over the course of 10 years, from January 2010 to December 2019.

To keep in line with confidentiality agreements and laws, each patient was coded and the original identification was removed. Data was collected directly from the electronic patient charts by the primary author.

Data included in the study were age, gender, number of days of hospitalization, number of hospitalizations for each patient, signs, symptoms, type of surgery (approach used), perioperative complications (CSF leak, hematoma, neurological deficits) and also death within period of hospitalization.

In order to be included in the study, the patients had to have diagnosis of pituitary tumor, be of either gender, with no limit in regard to age. The patient had to had been inbound in one of either Neurosurgery 3 or 4 Department and have an outgoing diagnosis of pituitary adenoma. All patients presenting with other sellar or parasellar masses were excluded (such as craniopharyngioma, epidermoid cyst, anterior circulation aneurysm).

RESULTS

The study included all patients presenting with pituitary tumors between January 2010 and

December 2019 on either of Department 3 or 4 of Neurosurgery in Bagdasar Arseni Hospital, Bucharest. Thus, 1107 hospitalization records were taken into account, including the follow-up hospitalization and/or second surgery hospitalization. Furthermore, there were 704 unique patients (personal identification numbers were analyzed).

The gender distribution was 243 (34.27%) women and 466 (65.73%) men (figure 1), minimum age was 14 and maximum was 84, with a mean age of 56.24 (62.77 years old for women and 52.77 years old for men) (figure 2).

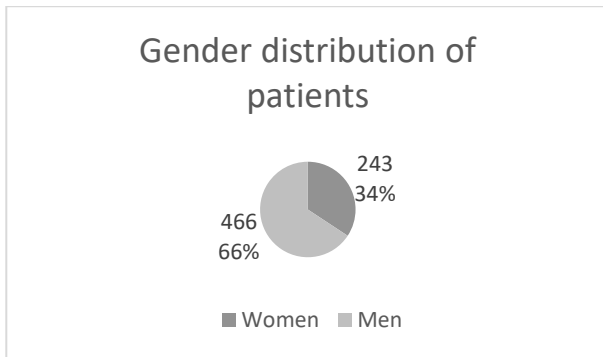


Figure 1.

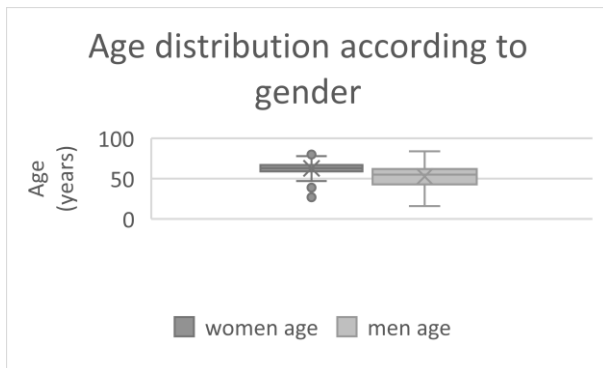


Figure 2.

The most common signs and symptoms encountered for the first admission were headache (245 – 34.56%), optic chiasm deficits (153 – 21.58%), acromegaly (85 – 11.99%). However, the majority of patients (507 – 71.51%) presented with some sort of hormonal imbalance or diabetes insipidus (figure 3).

On the one hand, a number of 325 (45.84%) patients had non-surgical treatment: they were either referred to an Endocrinology Department to begin conservative treatment, or a "watchful waiting" ensued after the first radiological evaluation (figure 4).

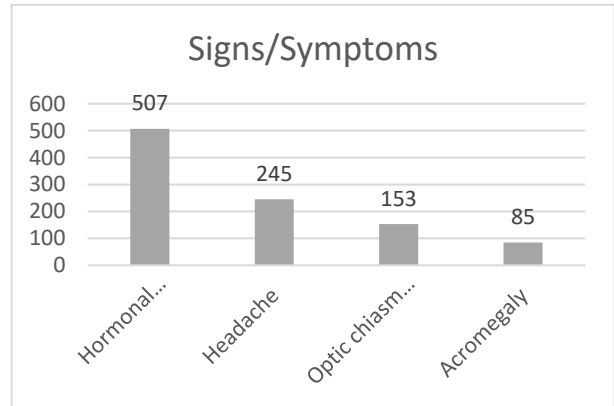


Figure 3.

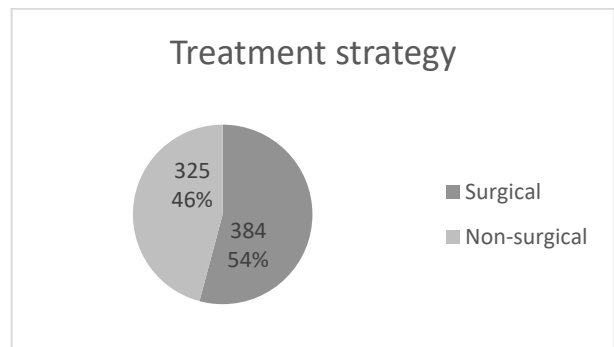


Figure 4.

On the other hand, a total of 384 (54.16%) surgeries for pituitary tumors were performed in this period as follows: 243 primary transsphenoidal surgeries, 36 primary transcranial surgeries, 62 transsphenoidal reinterventions, 18 transcranial interventions for relapse after transsphenoidal surgeries, 6 transsphenoidal surgeries after failed Gamma-Knife therapy, 8 combined approach surgeries (both transsphenoidal and transcranial) and 2 transcranial approaches after transcranial relapse (figure 5).

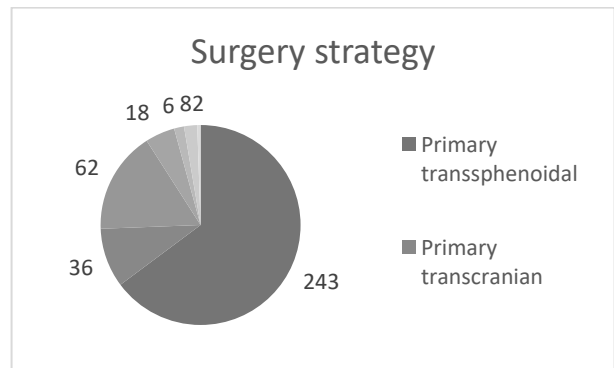


Figure 5.

Out of the patients who underwent surgery, 122 (31.77%) were macroadenomas, with extension ranging from only suprasellar, to one or both cavernous sinuses, anterior cranial fossa or third ventricle.

The histopathology examination revealed 95 GH secreting tumors (24.74%), 21 ACTH secreting tumors (5.46%), 18 prolactinomas (4.68%), 11 gonadotropin secreting hormones (2.86%). The rest were non secreting tumors (62.26%) (figure 6).

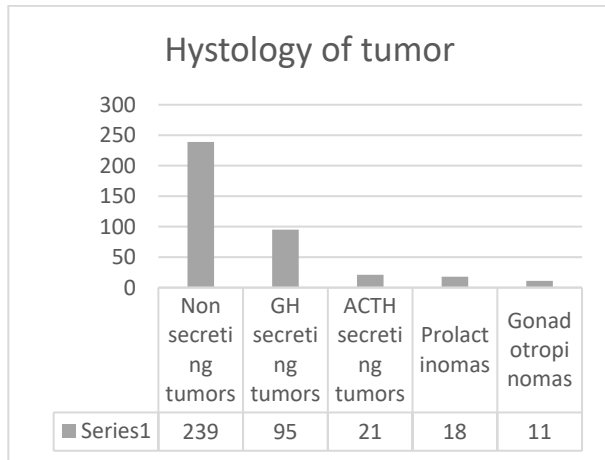


Figure 6.

The mean number of days of hospitalization was of 7.5 for the entire group, with an average of 7 days for the transsphenoidal approach and an average of 13 days for the transcranial approach (figure 7).

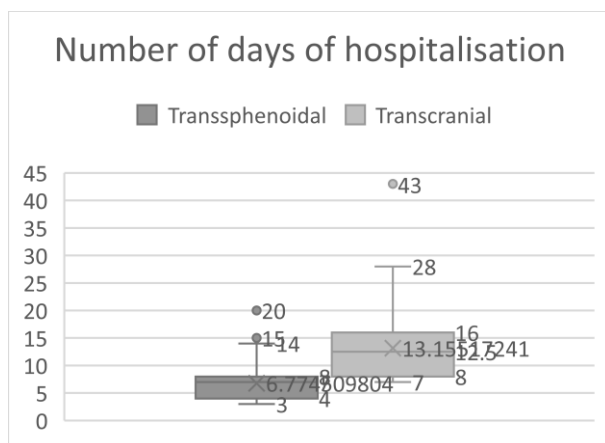


Figure 7.

The most common surgical complication encountered was pituitary insufficiency reported in 23 cases, while CSF leakage was observed in 17 cases.

For the total number of hospitalizations, 4 deaths were recorded: one after transsphenoidal surgery of a patient with a macroadenoma invading both cavernous sinuses, two patients who presented with acute hydrocephalus at admission and GCS less than 6, and one patient died of cardiac arrest in his sleep prior to surgery. No deaths were recorded for the transcranial group.

DISCUSSIONS

Hypothalamic/pituitary disorders represent a multifaceted pathology and, due to advancement of treatments, technology and understanding of physiopathology of the region, lately, not one type of strategy gets to be the gold standard. (2) Thus, recent understanding of complex mechanisms concerning cellular and molecular biology has brought medical treatment very high up in the list of strategies of treatment for pituitary tumors. Hence, almost half the patients who were admitted to our Neurosurgical Clinic benefited from a conservative treatment.

On the other hand, even though there is no consensus on the world stage of neurosurgery regarding the gold standard treatment in case of incidental discovered pituitary tumors, there is a different story when it comes to tumors which provoke neurological deficits of any sort, endocrine dysfunction beyond the drugs' capacity of compensating, or even pose an immediate threat to the patient's life. Most recent articles (4), (5) suggest a surgical strategy as first option in these cases, but this is where the consensus stops. There are mainly three types of surgical approaches: transcranial, microsurgical transsphenoidal and endoscopic transsphenoidal, each with its own advantages and disadvantages.

In our study, the patients who underwent surgery benefited from either microsurgical transsphenoidal or transcranial surgeries. Even if the transsphenoidal approach was used far more, there was a greater relapse proportion in these patients (figure 8). Furthermore, there were patients who underwent as many as 10 consecutive transsphenoidal surgeries in the course of the 10 years analyzed. Moreover, there were a number of patients who needed transcranial reintervention after the transsphenoidal approach did not offer the desired result. Only two patients underwent reintervention after transcranial approach, both of them with no relapse after the second surgery.

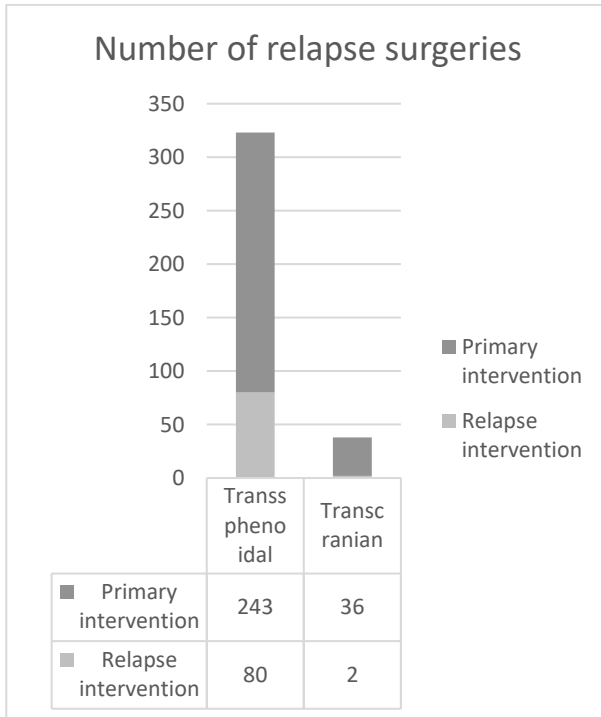


Figure 8.

On the other hand, transcranial surgeries, even if followed by a far less proportion of relapse surgery, carried with it the burden of more days spent in hospital (most of the time twice as much as for the transsphenoidal patients). Moreover, there were cases in which the transcranial approach was not enough for the tumor to be completely removed, in which case, a second transsphenoidal approach was used during the same hospitalization (the combined approach cases – 8 patients).

We also found a number of cases in which the first treatment option was Gamma-Knife therapy that proved not sufficient and a transsphenoidal approach was used in these cases.

In accordance with most of the studies published, our patient data base revealed a clear dominance of male over female patients and also a younger age of debut for the disease (or diagnosis) for men as opposed to women. Furthermore, due to the relative early diagnosis in the natural evolution of the disease, only 122 cases out of 384 operated patients presented with macroadenomas. Moreover, all of the patients who underwent transcranial approach presented with macroadenomas.

The most common symptoms that brought patients to the doctor were headache, optic chiasm deficits and body modification specific to GH

hypersecretion. However, there was quite a large proportion of patients (mostly microadenomas) who were diagnosed with a brain tumor after the hormonal imbalances were discovered or patients who underwent a CT scan for another pathology (for example: post-traumatic CT scans performed after car crashes that reveal pituitary tumors).

The most notable complications were CSF fistulae and pituitary insufficiency. These appeared in a total of less than 1% of all hospitalizations and less than 7% out of the patients who underwent surgery (17 fistulae, 23 pituitary insufficiency). Furthermore, all of the patients with CSF fistulae were part of the transsphenoidal approach group, none of the transcranial approach group presenting this complication.

Out of the total number of hospitalizations (1107), 4 deaths were reported, out of which only one directly related to the treatment option-transsphenoidal surgery, thus a death rate of less than 0.72 for the entire group.

CONCLUSIONS

Even with the advancements in molecular and cellular biology, surgery remains one of the most important aspects of pituitary tumors treatment. Even if there is still no general consensus on what the ideal approach should be, our study reveals that both transsphenoidal and transcranial approaches have advantages and disadvantages, thus the best strategy would be to tailor each surgery to each patient, keeping an open mind to all available approaches.

CONFLICTS OF INTEREST

The authors declare no conflict of interests

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REFERENCES

1. Presentation, Management, and Outcomes of Nonfunctioning Pituitary adenomas: An experience from a developing country. Das B, Batool S, Khoja A, et al. e5759, s.l.: Cureus 11(9), September 25, 2019. DOI: 10.7759/cureus.5759.
2. A Survey on Pituitary Surgery in Italy. Domenico Solari, Paolo Cappabianca et al. E1-E10, s.l. : World Neurosurgery, 2018. <https://doi.org/10.1016/j.wneu.2018.11.186>.
3. Complications associated with microscopic and endoscopic transsphenoidal pituitary surgery: experience of 1153 consecutive cases treated at a single

tertiary pituitary center. Matthew S Agam, BS, Gabriel Zada, MD et al. June 1, Los Angeles, California : Journal of Neurosurgery, Published online June 1, 2018. DOI: 10.3171/2017.12.JNS172318..

4. Endoscopic versus Microscopic Pituitary Adenoma Surgery: A Single-center Study. Ajler Pablo, Beltrame Sofia, Toscano Maximiliano, Fainstein Day Patricia, Campero Alvaro, Yampolsky Claudio, Carizzo Antonio. 4, s.l. : Neeurology India, 2019, Vol. 67. DOI: 10.4103/0028-3886.266241.
5. Comparison of Endoscopic Versus Microsurgical Resection of Pituitary Adenomas with Parasellar Extension and Evaluation of the Predictive Value of a Simple 4-Quadrant Radiologic Classification. Gianluca Trevisi, Vera Vigo, Maria Grazia Morena, Domenico Luca Grieco, Mario Rigante, Carmelo Anile, Annunziato Mangiola. E1-E6, s.l. : World Neurosurg., 2018. <https://doi.org/10.1016/j.wneu.2018.09.215>.



The impact of vertebral osteomyelitis on spinal stability and principles of surgical stabilization. Medical literature review

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ABSTRACT

Vertebral osteomyelitis (VO) is a disease that responds well to conservative treatment and antibiotherapy if diagnosed in an early stage. Due to the prolonged onset of this pathology, many cases are diagnosed in mid or late stages and require surgery. The surgical treatment is not yet standardized and may only mean decompression of the infectious outbreak without stabilization, or surgical decompression associated with stabilization.

Using only bone grafts for surgery or stabilizing the spine through segmental posterior and/or anterior instrumentation is accompanied by many controversies.

In this review, we focus on demonstrating that combining a well-conducted antibiotherapy with thorough debridement of the necrotic areas and using metal implants for spinal stabilization lowers the infection rates, provides an early pain-free mobilization of the patient and reduces hospitalization costs.

1. INTRODUCTION

Vertebral osteomyelitis (also called osteodiscitis) is an acute or chronic infection of the spine with direct iatrogenic or indirect (hematogenous) inoculation. It is a rare infection concentrated in the spine, the cases being so rare that only 2-4% of all bone infections are attributed to this disease. Vertebral osteomyelitis attacks the intervertebral disc and the two adjacent vertebrae, causing the destruction of the intervertebral space. The prognosis of the disease depends on several factors: the place where the infection is concentrated in the spine, the time elapsed between the initial onset and the treatment used, but also the approach used to treat the disease (1,5).

Historically we will cover the treatments applied to vertebral osteomyelitis over the years. The first measures taken were bed rest and symptomatic medication.

Laminectomy was the first attempt at surgical treatment, but it brought poor results in the approach to the anterior spine, and the

Keywords

c-reactive protein,
osteodiscitis,
spinal instrumentation,
spine surgery,
vertebral osteomyelitis



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main complication was postoperative instability (2, 11).

In 1911, Albee introduced a posterior bone fusion technique, but this was abandoned due to postoperative kyphosis (6).

Hodgson and Stock introduced the approach technique of the infectious outbreak, which directly resolves the infection and preserves the integrity of the anterior spine, preventing postoperative kyphosis (10).

After the introduction in the 1970s by Roy Camille of the vertebral fixation technique with transpedicular screws and rods, it became a standard in the surgical treatment of vertebral osteomyelitis (5, 8).

MATERIALS AND METHODS

For accomplishing this review, we enlisted a throughout research of database from "PubMed", "Scopus", "Web of science" using key words as "vertebral osteomyelitis", "spinal infection", "osteodiscitis", "spinal stabilization" and "spine surgery". Of the many existing studies, we reviewed 35 studies independent of time and date and from which we concluded this review, comparing different methods of treatment in vertebral osteomyelitis.

RESULTS

ETIOLOGY AND PATHOPHYSIOLOGY

The source of the infection was found in only 40% of cases and was represented by urinary tract infections, soft tissue, respiratory, intestinal, dental infections, endocarditis and penetrating trauma. The etiology of vertebral osteomyelitis consists of infections with *Staphylococcus aureus*, *Streptococcus*, *Mycobacterium tuberculosis*, etc.

The study by Nolla JM et al shows that acute VO affects mostly elderly patients with associated comorbidities. (27)

Most cases of osteodiscitis occur due to the inoculation of bacteria by hematogenous at the level of the intervertebral disc. Dermal inoculation is also present. The most common bacterial cause is *Staphylococcus Aureus* (32-67%). Occasionally, coagulase-negative staphylococci may be a cause with or without epidural involvement. Gram-negative organisms, such as *Escherichia coli* (21%), often from a urinary tract source are the second most commonly identified cause of infection.

Pseudomonas sp. are associated with approximately 6% of cases and should indicate a history of environmental water exposure or intravenous drug use (20).

In their study, Patzakis and coworkers included a 78 % of the patients as intravenous drug users and found *S. aureus* at a low prevalence as the main bacteria (17%), while *Pseudomonas aeruginosa* was the predominant etiologic agent (38%) (29).

Spinal infections lead to a spectrum of diseases with a varied clinical presentation. Vertebral infection usually results from bacterial insemination of the vertebral disc, which then spreads to adjacent vertebrae (vertebral osteomyelitis). A muscular abscess is frequently present in the paravertebral muscles. In 17% of cases, the infection at the disc level migrates into the epidural space, resulting in the epidural abscess. Its timely identification is essential, as 1 in 4 patients with this condition quickly develop paralysis (26).

Several studies describe the vertebral level involved and describe the following incidence of affection: lumbar spine (58%), thoracic spine (30%) and cervical spine (11%) (34, 28, 2, 26, 17). Vertebral defects were found in multiple levels, 6% were reported with continuous character and 3% reported with skipping multifocal involvement. A large number of multifocal involvement was reported in iv drug abusers (29).

SYMPTOMS. CLINICAL EVALUATION

A detailed neurological examination is essential for any patient suspected of having vertebral osteomyelitis. Objective neurological signs are rare, but when present can range from mild (radicular pain corresponding to a nerve root injury), moderate (motor weakness, sensory loss, urinary or intestinal dysfunction), to severe (paralysis).

In the presence of an abnormal neurological examination, the presence of a possible epidural abscess should be investigated, because the delay of the diagnosis can lead to the permanence of the deficit.

The accentuation of the neurological signs indicates the progression of the infection in the epidural space and the damage of the nerve roots or the compression of the spinal cord.

According to numerous studies, many patients present an insidious onset and unspecific symptomatology (neck or back pain, fever, painful

flexion/extension of the back and/or neurological deficits). Early diagnosis of this disease is important, but not always possible as 30-70% of the patients do not show signs of prior infection (5) and a delay of 2 to 6 months from the starting symptomatology to the diagnosis of this disease has been reported. (1, 11)

Local clinical examination, represented by the point of sensitivity to palpation can differentiate vertebral osteomyelitis from other differential diagnoses. There are no pathognomonic signs or symptoms to indicate the presence of this condition. A normal clinical examination does not rule out presence diagnosis of vertebral osteomyelitis. The classic triad consisting of fever, back pain and neurological deficit is specific to this condition. The potential consequences of undiagnosed vertebral osteomyelitis are devastating, so when referring to a differential diagnosis, laboratory and imaging tests are mandatory.

Thus, according to the studies of Butler et al and Frangen et al, the diagnosis should be supported by clinical, laboratory, and imaging findings (1, 11)

LABORATORY TESTS

Usual blood tests that help diagnose patients with suspected epidural abscess or vertebral osteomyelitis include: hemoleukogram, erythrocyte sedimentation rate (ESR), C-reactive protein (CRP). These tests are not completely reliable indicators for the presence of the disease and must be correlated with: clinical suspicion, patient history and imaging study. Leukocytosis is variable and does not indicate the severity of the disease. Increased ESR is important in the early diagnosis of patients with osteomyelitis.

CRP has a sensitivity of 84-100% in the case of the presence of epidural abscess and is a necessary analysis for the initial diagnosis.

However, according to Curry and coworkers, ESR and CRP are more useful than the white blood cell count (WBC), because a normal WBC does not exclude the presence of a spinal infection. (3)

Also, the positivity of blood cultures indicates the presence of a generalized infection, the most often isolated germ in most cases being *Staphylococcus aureus*.

As some studies show, up to 59% of positive blood cultures identify the etiological microorganism

in patients with monomicrobial pyogenic spondylodiscitis (24)

IMAGING

Radiological evaluation of a patient with vertebral osteomyelitis reveals changes only after a few weeks. The first obvious changes after a few days are local edema and loss of the psoas shadow. In the next 7-10 days, the reduction of the vertebral space and the erosion of the adjacent vertebral plates ("mirror image") are observed. Sclerosis at this level generally appears on the radiological image at 10-21 days and it may take 3 to 6 weeks from the onset of the symptoms for bony destruction of the vertebrae to become evident according to Jevtic V's study (20) (Figure 1). This results in a delay of the diagnosis.

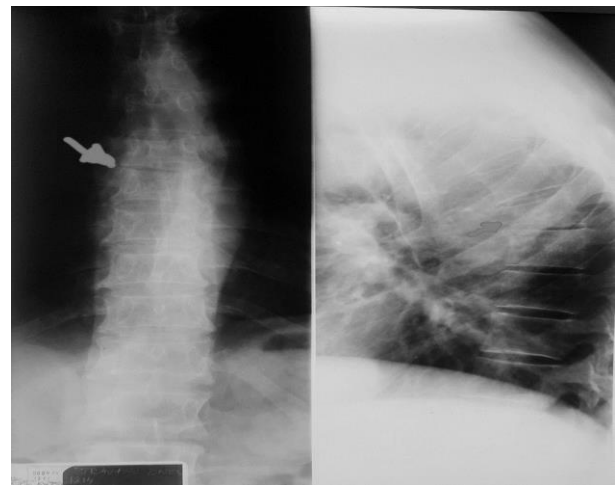


Figure 1. Preoperative thoracic spine radiograph: major narrowing of T7-T8 disc space (personal collection).

The gold standard for imaging diagnosis is MRI, which distinguishes hypointensity in T1, hyperintensity in T2 and capture of Gadolinium in the T1-weighted sequence. This investigation has a sensitivity of 96% and a specificity of 93% and is also used in the differential diagnosis of this pathology. (12, 13)

In case of an MRI contraindication, a CT scan is performed, which distinguishes bone abnormalities, abscess formation and the degree of bone damage. Another investigation that can be performed in the case of MRI contraindication is Tc99m Technetium Bone Scintigraphy, but it has a sensitivity of 90% and a much lower specificity than MRI. (17) (figures 2,3)

According to Hadjipavlou and coworkers, positive blood cultures and other paraclinical investigations

in association with clinical symptoms do not confirm the diagnosis of a spinal infection. The definitive diagnosis can be obtained by histopathological examination thorough a surgical or CT-guided biopsy. An exact diagnosis of this disease should be achieved in order to ensure a well-conducted management. (16, 17)

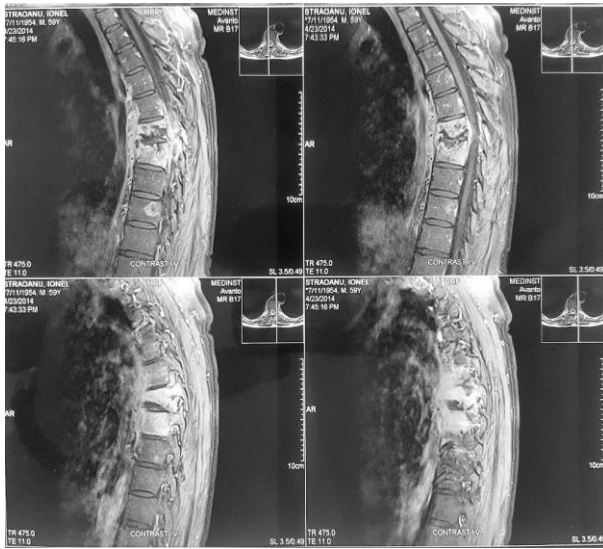


Figure 2. Thoracic MRI with intravenous contrast T2 sagittal plane: T7-T8 osteomyelitis with intraspinal abscess (personal collection).

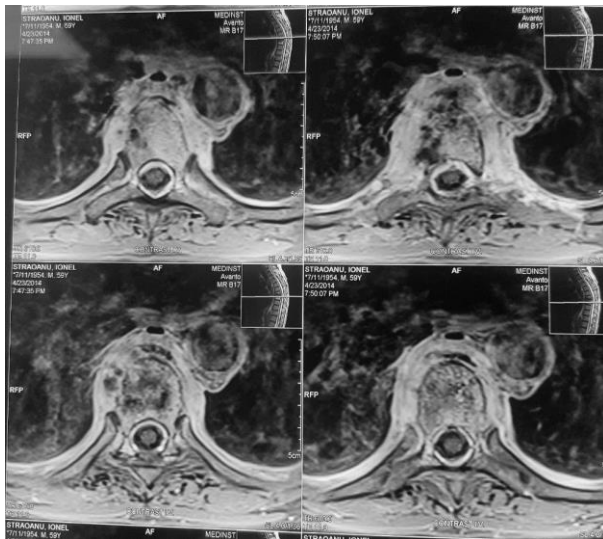


Figure 3. Thoracic MRI with intravenous contrast T2 transverse plane: T7-T8 osteomyelitis with intraspinal abscess (personal collection).

DIFFERENTIAL DIAGNOSIS

The main pathologies with which the differential diagnosis can be made are: vertebral tumors,

vertebral fracture and degenerative diseases of the spine (disc herniation, spinal canal stenosis, vertebral spondylolysis).

In a study conducted by Tyrell PN et al, it is demonstrated that a destructive bone lesion of the vertebrae associated with a preserved disc space with sharp endplates guides the diagnosis to a neoplastic infiltration, while a destructive bone lesion associated with an undefined vertebral body with loss of definition of the vertebral endplate with or without modification of disc height suggests an infection, which has a better prognosis than cancer. (35)

CONSERVATIVE AND MEDICAMENTOUS TREATMENT

Conservative treatment of vertebral osteomyelitis consists of prolonged bed rest and antibiotic therapy for 6 to 12 weeks.

During immobilization, the patient is placed in a corset or cervico-thoracic or thoraco-lumbar orthosis depending on the location of the lesion.

According to the studies of Fleege C et al and Skaf GS et al, the first line of treatment is a conservative approach. This choice is made especially for the patients with minor or no neurologic deficits and in case of severe associated comorbidities that limit surgery. Antibiotics with a large spectrum are used initially (Clindamycin + Ciprofloxacin or Cefotaxim + Flucloxacillin) in order to cover a wider area of potential pathogens. Afterwards, the antibiotic treatment is switched to an appropriate medication guided by the antibiogram. These last antibiotics are applied iv for 2-4 weeks or until the CRP is markedly dropped. In the final step of the treatment oral antibiotic is continued for a total of 6 to 12 weeks. This treatment is accompanied by prolonged bed rest and pain medication with or without orthosis for at least 6 weeks. (10, 33)

Medicamentous treatment is a symptomatic as well as an etiological treatment.

Symptomatic treatment addresses mainly pain, febrile syndrome and includes various types of analgesics and anti-inflammatory drugs.

The etiological treatment is addressed to the germ in question and is represented by third generation cephalosporins and / or Vancomycin for bacterial and tuberculostatic treatment in cases with Mycobacterium Tuberculosis (20).

Medicamentous treatment must also take into account the biological balance of each patient and in

this context, we refer to parenteral hydration with various osmolar solutions, parenteral nutrition in cases of severe sepsis, combating anemia in cases of severe sepsis through blood transfusions, hypoproteinemia and balancing the metabolic rate of the patient (17).

If conservative treatment fails and proper mobilization of the patients is not achieved surgery is then performed as many complications due to immobilization are critical to patients, especially to the elderly.

SURGICAL TREATMENT

Several studies suggest that the main indications for surgical treatment are represented by the following: severe lesional changes in vertebral endplates, formation of abscesses, biomechanical instability due to chronic osteomyelitis, appearance of neurological deficits, decompensated kyphosis, septic pseudarthrosis, pain that does not respond to pharmaceutical treatment, and non-responsive patients to conservative treatment. (4, 14, 30)

The surgical treatment has the following important objectives: evacuation of infection and debridement, decompression of the dural sac and spinal roots and vertebral fixation / fusion.

The main principle of the surgical treatment of this disease is radical and aggressive debridement of all unviable tissues and material according to a study conducted by Fayazi AH and coworkers. This study states that all necrotic and infected tissues are mandatory to be removed and all abscesses evacuated for a good management of the disease. (9)

The choice of surgical approach depends on the location of the lesion in the spine. Thus, the anterior cervical approach, the lateral cavitory approach and costotransversectomy for the thoracic spine, laminectomy, hemilaminectomy and foraminotomy for the lumbar spine are described.

Choosing between an anterior or posterior approach is debatable.

The studies of Emery SE, Fang D and Fukuoka M show that due to affection of the vertebral bodies and disc spaces the majority of surgeons incline to an anterior approach for direct access to the infection site and better reconstructive stabilization. (6, 7, 12)

Fayazi et al and Korovessis et al reveal that a posterior approach is used in order to drain the abscesses and for posterior stabilization of the spine

and that a combined approach needs to be chosen in relation to the surgical goals that are aimed to be achieved. (9, 22)

Several studies show that a combined approach results in a lower incidence of postoperative infection and revision surgery and a higher mortality rate with an isolated anterior approach for cervical spine lesions. The increased mortality rate of an anterior approach is also explained by a higher incidence of associated comorbidities of patients with cervical spine lesions. (4, 8, 13, 15, 23)

The vertebral fixation is performed using transpedicular screws and rods, but also the reconstruction techniques of the anterior column, with the help of fixed or expandable titanium cages. A relatively new method of treating this disease is represented by minimally invasive surgery.

Muckley T and coworkers demonstrated in their study that treating VO through video-assisted thoracoscopic surgery has advantages and disadvantages. The main advantages are debridement and surgical instrumentation of multiple levels can be achieved with minimal dissection of the surrounding soft tissues. The disadvantages of this technique are represented by a technically demanding procedure in which special instrumentation and experience of the surgeon are required. (25)

Hadjipavlou et al revealed in their study of 34 cases an immediate pain relief of 26 patients and stated that the most common long-term complications of the use of minimally invasive surgery are implant failure with axial pain and instability or severe kyphotic deformities. (16)

A controversial debate is between the use of single-stage surgery versus two-stage surgery.

Safran et al revealed in his study of 10 patients with osteodiscitis operated through single-stage surgery that it is a secure and efficient method of controlling the infection and reconstruction of the spine, shortening the hospitalization and mobilization time of the patient and also reducing hospitalization costs for the patient and the institute. The surgery is performed based on the experience of the surgeon and the overall condition of the patient, taking into consideration that medically unstable patients may not be able to withstand a second anesthesia, surgery or increased period of immobilization. (31)

Increased surgical debridement of a

compromised anterior column of the spine results in massive loss of bone tissue and increased axial instability, therefore it is mandatory to perform surgical reconstruction in order to prevent residual instability.

Bone grafting is required for the reconstruction of the anterior column either with autogenous bone graft or allograft. A study by Emery SE et al demonstrated that 19 patients operated for vertebral osteomyelitis through aggressive debridement and the use iliac bone grafting in the presence of an active infection had immediate pain relief and no septic complications 2 years after surgery. (6)

Many surgeons suggest that allografting can be used instead of autogenous grafting. Schuster et al conducted a study of 47 patients operated using allografts consecutive to surgical debridement, posterior instrumentation and iv antibiotics and demonstrated that it is a secure method of treating vertebral osteomyelitis. The main advantages of using allografts are lowering the operation time and morbidity associated with harvesting of the graft and the main disadvantages are the host's immune response and an increased risk of pathological transmission of diseases. (32)

Hodgson demonstrated in 1956 through his work on the treatment of Pott's paraplegia that anterior debridement and vertebral fusion with autogenous bone grafting rapidly correct axial alignment, however, long-term results show spinal instability with the collapse of the vertebral column, correction malalignment and formation of pseudarthrosis. (19) Therefore, the use of spinal instrumentation is mandatory in most cases associated with spinal instability. (Figures 4,5)

In their studies Hee et al. and Fayazi et al. reported that the use of a titanium mesh cage with posterior instrumentation for the reconstruction of the spine is an excellent method in improving the mobilization of the patient and a rapid fusion in patients that underwent surgery for active vertebral osteomyelitis. Therefore, they demonstrated that the patients had less postoperative complications, lower infection recurrence rates and improved spinal alignment and stability. The loss of kyphosis correction was noted due to collapse of the cage into the adjacent vertebrae, but overall the cases had a good prognosis because of the lower infection rates and early and pain free mobilization. (18, 9)



Figure 4. Intraoperative view showing spinal fixation using 8 transpedicular screws and rods after radical debridement (personal collection),

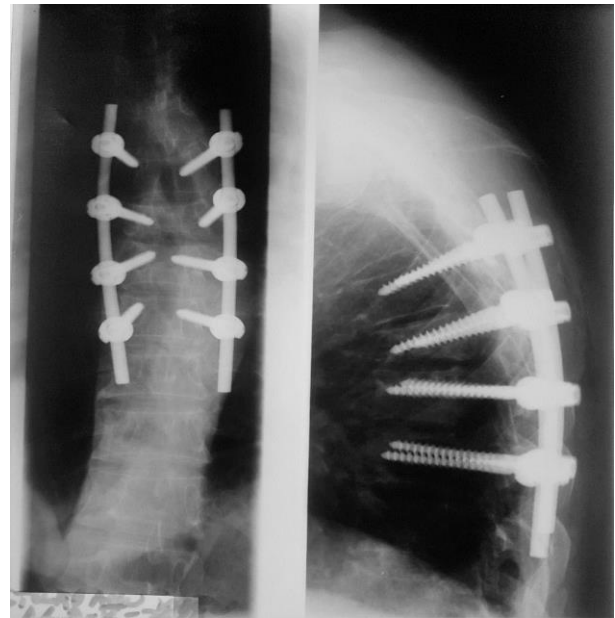


Figure 5. Postoperative radiographic control: stabilization with 8 transpedicular screws and 2 titanium rods (personal collection).

CONCLUSION

Most cases of vertebral osteomyelitis respond well to conservative treatment if diagnosed in an early stage. In cases that are unmanageable by conservative treatment, surgery is mandatory. Aggressive debridement of all necrotic tissues is needed along with surgical stabilization of the spine to ensure a good management of the disease. Vertebral fixation and fusion ensure immediate vertebral stability, which allows the patient to

mobilize quickly in the first days after surgery. This is beneficial for neurological recovery, starting physical therapy and preventing complications associated with prolonged bed rest. The patient can be discharged from the surgery department in the first week after the operation after wound healing and thread extraction, which allows rapid transfer to the infectious diseases department where prolonged and targeted antibiotic treatment will be continued. This reduces hospital stay and costs per patient. This paper will demonstrate that the combination of surgical treatment represented by decompression and fixation-fusion vertebral with targeted antibiotic treatment eliminates local recurrence and allows the patient to quickly return to a normal socio-professional life.

REFERENCES

- Butler JS, Shelly MJ, Timlin M, Powderly WG, O'Byrne JM. Nontuberculous pyogenic spinal infection in adults: a 12-year experience from a tertiary referral center. *Spine*. 2006 Nov 1;31(23):2695-700.
- Chelsom J, Solberg CO. Vertebral osteomyelitis at a Norwegian university hospital 1987-97: clinical features, laboratory findings and outcome. *Scandinavian journal of infectious diseases*. 1998 Jan 1;30(2):147-51.
- Curry Jr WT, Hoh BL, Amin-Hanjani S, Eskandar EN. Spinal epidural abscess: clinical presentation, management, and outcome. *Surgical neurology*. 2005 Apr 1;63(4):364-71.
- Dimar JR, Carreon LY, Glassman SD, Campbell MJ, Hartman MJ, Johnson JR. Treatment of pyogenic vertebral osteomyelitis with anterior debridement and fusion followed by delayed posterior spinal fusion. *Spine*. 2004 Feb 1;29(3):326-32.
- Duarte RM, Vaccaro AR. Spinal infection: state of the art and management algorithm. *European Spine Journal*. 2013 Dec 1;22(12):2787-99.
- Emery SE, Chan DP, Woodward HR. Treatment of hematogenous pyogenic vertebral osteomyelitis with anterior debridement and primary bone grafting. *Spine*. 1989 Mar;14(3):284-91.
- Fang D, Cheung KM, Dos IR, Lee YK, Leong JC. Pyogenic vertebral osteomyelitis: treatment by anterior spinal debridement and fusion. *Journal of spinal disorders*. 1994 Apr;7(2):173-80.
- Faraj AA, Webb JK. Spinal instrumentation for primary pyogenic infection. Report of 31 patients. *Acta orthopaedica belgica*. 2000 Jun;66(3):242-7.
- Fayazi AH, Ludwig SC, Dabbah M, Butler RB, Gelb DE. Preliminary results of staged anterior debridement and reconstruction using titanium mesh cages in the treatment of thoracolumbar vertebral osteomyelitis. *The Spine Journal*. 2004 Jul 1;4(4):388-95.
- Fleege C, Wichelhaus TA, Rauschmann M. Systemic and local antibiotic therapy of conservative and operative treatment of spondylodiscitis. *Der Orthopade*. 2012 Sep;41(9):727-35.
- Frangen TM, Källicke T, Gottwald M, Andereya S, Andress HJ, Russe OJ, Müller EJ, Muhr G, Schinkel C. Surgical management of spondylodiscitis. An analysis of 78 cases. *Der Unfallchirurg*. 2006 Sep;109(9):743-53.
- Fukuoka M, Aoki Y, Hayashi S, Nagasawa K, Aita K, Hotokebuchi T, Satoh T. Pyogenic vertebral osteomyelitis caused by *Prevotella intermedia*. *Journal of infection and chemotherapy*. 2002 Jan 1;8(2):182-4.
- Fukuta S, Miyamoto K, Masuda T, Hosoe H, Kodama H, Nishimoto H, Sakaeda H, Shimizu K. Two-stage (posterior and anterior) surgical treatment using posterior spinal instrumentation for pyogenic and tuberculous spondylitis. *Spine*. 2003 Aug 1;28(15):E302-8.
- Gasbarrini AL, Bertoldi E, Mazzetti M, Fini L, Terzi S, Gonella F, Mirabile L, Barbanti Brodano G, Furno A, Gasbarrini A, Boriani S. Clinical features, diagnostic and therapeutic approaches to haematogenous vertebral osteomyelitis. *Eur Rev Med Pharmacol Sci*. 2005 Jan;9(1):53-66.
- Graziano GP, Sidhu KS. Salvage reconstruction in acute and late sequelae from pyogenic thoracolumbar infection. *Journal of spinal disorders*. 1993 Jun;6(3):199-207.
- Hadjipavlou AG, Katonis PK, Gaitanis IN, Muffoletto AJ, Tzermiadianos MN, Crow W. Percutaneous transpedicular discectomy and drainage in pyogenic spondylodiscitis. *European Spine Journal*. 2004 Dec 1;13(8):707-13.
- Hadjipavlou AG, Mader JT, Necessary JT, Muffoletto AJ. Hematogenous pyogenic spinal infections and their surgical management. *Spine*. 2000 Jul 1;25(13):1668-79.
- Hee HT, Majd ME, Holt RT, Pienkowski D. Better treatment of vertebral osteomyelitis using posterior stabilization and titanium mesh cages. *Clinical Spine Surgery*. 2002 Apr 1;15(2):149-56.
- Hodgson AR, Stock FE. Anterior spinal fusion a preliminary communication on the radical treatment of Pott's disease and Pott's paraplegia. *British journal of surgery*. 1956 Nov;44(185):266-75.
- Jevtic V. Vertebral infection. *European Radiology Supplements*. 2004 Mar 1;14(3):E43-52.
- Karadimas EJ, Bungler C, Lindblad BE, Hansen ES, Høy K, Helmig P, Kannerup AS, Niedermann B. Spondylodiscitis. A retrospective study of 163 patients. *Acta orthopaedica*. 2008 Jan 1;79(5):650-9.
- Korovessis P, Sidiropoulos P, Piperos G, Karagiannis A. Spinal epidural abscess complicated closed vertebral fracture: a case report and review of literature. *Spine*. 1993 Apr 1;18(5):671-4.
- Lee MC, Wang MY, Fessler RG, Liauw J, Kim DH. Instrumentation in patients with spinal infection. *Neurosurgical focus*. 2004 Dec 1;17(6):1-6.
- Lener S, Hartmann S, Barbagallo GM, Certo F, Thomé C,

- Tschugg A. Management of spinal infection: a review of the literature. *Acta neurochirurgica*. 2018 Mar 1;160(3):487-96.
25. Mückley T, Schütz T, Schmidt MH, Potulski M, Bühren V, Beisse R. The role of thoracoscopic spinal surgery in the management of pyogenic vertebral osteomyelitis. *Spine*. 2004 Jun 1;29(11):E227-33.
 26. Nather A, David V, Hee HT, Thambiah J. Pyogenic vertebral osteomyelitis: a review of 14 cases. *Journal of Orthopaedic Surgery*. 2005 Dec;13(3):240-4.
 27. Nolla JM, Ariza J, Gómez-Vaquero C, Fiter J, Bermejo J, Valverde J, et al. Spontaneous pyogenic vertebral osteomyelitis in nondrug users. *Semin Arthritis Rheum* 2002;31:271-8.
 28. Osenbach RK, Hitchon PW, Menezes AH. Diagnosis and management of pyogenic vertebral osteomyelitis in adults. *Surgical neurology*. 1990 Apr 1;33(4):266-75.
 29. Patzakis MJ, Rao SA, Wilkins JE, Moore TM, Harvey PJ. Analysis of 61 cases of vertebral osteomyelitis. *Clinical orthopaedics and related research*. 1991 Mar(264):178-83.
 30. Priest DH, Peacock JE. Hematogenous vertebral osteomyelitis due to *Staphylococcus aureus* in the adult: clinical features and therapeutic outcomes. *Southern medical journal*. 2005 Sep 1;98(9):854-63.
 31. Safran O, Rand N, Kaplan L, Sagiv S, Floman Y. Sequential or simultaneous, same-day anterior decompression and posterior stabilization in the management of vertebral osteomyelitis of the lumbar spine. *Spine*. 1998 Sep 1;23(17):1885-90.
 32. Schuster JM, Avellino AM, Mann FA, Girouard AA, Grady MS, Newell DW, Winn HR, Chapman JR, Mirza SK. Use of structural allografts in spinal osteomyelitis: a review of 47 cases. *Journal of Neurosurgery: Spine*. 2000 Jul 1;93(1):8-14.
 33. Skaf GS, Domloj NT, Fehlings MG, Bouclaous CH, Sabbagh AS, Kanafani ZA, Kanj SS. Pyogenic spondylodiscitis: an overview. *Journal of infection and public health*. 2010 Jan 1;3(1):5-16.
 34. Torda AJ, Gottlieb T, Bradbury R. Pyogenic vertebral osteomyelitis: analysis of 20 cases and review. *Clinical Infectious Diseases*. 1995 Feb 1;20(2):320-8.
 35. Tyrrell PN, Cassar-Pullicino VN, McCall IW. Spinal infection. *European radiology*. 1999 Jul 1;9(6):1066-77.



Historical vignette: The first brain surgery performed by the first woman neurosurgeon in Romania, Dr. Sofia Ionescu-Ogrezeanu

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ABSTRACT

Introduction. Sofia Ionescu-Ogrezeanu (b. April 25, 1920, Fălticeni - d. March 21, 2008, Bucharest), also known as the Lady of Romanian Neurosurgery, became the first woman neurosurgeon in the world after performing a brain surgery during World War II, a fact recognized as a world premiere during the 13th World Congress of the World Federation of Neurosurgical Societies (WFNS) in Morocco, in 2005. [1]

Materials and methods. Sofia Ionescu is the first woman neurosurgeon. She was born on the fields of Bucovina, in Fălticeni, and became part of the "golden team" of the Romanian neurosurgery of the pioneering period. The decisive moment of her career took place in 1944, during the war when she was forced to perform an emergency operation on a child, a victim of the bombing.

The article coagulates the reports regarding the description of the first brain surgery performed by the first woman neurosurgeon with the continuous activity integrated with the field of neurosurgery. Both specialized articles, biographical books, and television interviews were used as references.

Results. The operation performed in the fifth year of faculty was the first step of a journey of 47 years of neurosurgical career, practised with high morality and devotion. The sacrifice of the pioneer of the first woman neurosurgeon was recognized in the press in the country and abroad, as well as by the recognition of different titles and distinctions.

Keywords

Neurosurgeon Sofia Ionescu
(Ogrezeanu),
first brain surgery 1944.
first woman neurosurgeon.
neurosurgical history



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INTRODUCTION

Dr. Sofia Ionescu's professional career reveals the secret of the art of reversing the meaning of obstacles; according to the idea advocated by Marcus Aurelius, year 170: "The impediment to action advances action. What stands in the way becomes the way." Thus, she performed her integrated neurosurgical activity during 47 years, in the socio-political context of the Second World War. She was part of the team that made the pioneering sacrifice in the Romanian neurosurgery, also called the "gold team"; together with Prof. Dr. Dimitrie Bagdasar, the founder of the neurosurgical specialty in Romania, formed by Prof. Dr. Harvey Williams Cushing in Boston, Dr. Constantin Arseni and Dr. Ionel Ionescu, who later became her husband and together they had two children [2],[3].

MATERIALS AND METHODS

In Dr. Sofia Ionescu we discover a personality who practices great virtues, such as: *courage* - to be the first woman neurosurgeon in Romania and internationally renowned; *reason* - to constantly perform the balance exercise in affective gymnastics and to practice for the first time in 1944, in the fifth year of study at the Faculty of Medicine, the drill procedure, in order to save the life of a boy in a coma, the victim of the bombings of World War II; *ingenuity and creativity* - to use in 1968 a sterile urinary well for external ventricular drainage, subsequently standardized procedure; *compassion* - to the fellow patients, who she gave pieces of soul and days of their lives; *humility* - to offer the sacrifice of her own existence in order to perpetuate neurosurgery as a specialty in Romania, fulfilling the last wish of Prof. Dr. Dumitru Bagdasar. [4]

About her first brain operation, which enrolled her in the history of world neurosurgery, she speaks in an interview with Eugenia Vodă, within the program "Professionals" of the Romanian Television 1 (TVR1) in 2000, reporting: "This operation decided life for 47 years ahead, while I was in neurosurgery, and brought me to 180 degrees compared to what I had planned, a quiet life as an internist in my hometown, Fălticeni. Because at some point it was necessary to perform an emergency operation on a child after a bombing and he had no one to do it. Dr. Bagdasar had a suppuration in his hand, the other two secondary doctors, Dr. Arseni and Dr. Ionescu, each had a similar impediment to enter the operation, and the child was

actually dying in front of us. And then Bagdasar, who was very impressed, said. Who can do this?" He asked the intern who was a neurologist. And he said "No. No way." shaking his head. And then he asked me, too, and I agreed. Although, in my mind, I thought "If he watches me operating, my hand will be shaking and he will look at me curiously", as it was the first neurosurgery operation. Dr. Bagdasar, at the end of the operation, told me "Miss, you have the necessary skills. Please stay with us".



Figure 1. Dr. Sofia Ionescu during surgery.

Also, in the biographical book "Neodina binelui - Neurosurgeon Sofia Ionescu", written by Rodica Simionescu, published in 1998, at Alas Publishing House, Călărași, there is a chapter that sets out the steps of initiating the operations in neurosurgery, fragments: "The year 1944. Among the wounded emergency patients in the hospital at the neurosurgery department, a little boy is brought into a coma. Dr. Bagdasar consults him and decides that the child should be operated on immediately. The only valid persons were the intern Sofia OGREZEANU and the intern Constantin CREȚAN. If they do not intervene urgently, the child dies in front of them. Who can do a tripod? She defeats her chosen spirit, professional conscience and, of course, courage and states that she wants to operate. She operated safely and quickly as if she had been doing this for ever. She was operating on a child. She was

performing brain surgery. No woman had done this before. She went into the operation with only one thought: to save the child. Extraordinary peace, perfect peace and clarity have seized her soul. Perfect concentration. Her mind works without any emotion. Everything is just calm and precision. That's how it was. That's how she always operated. Professor Bagdasar, dr. Ionel Ionescu, the staff of the operating room followed her breathlessly. She succeeded. Dr. Bagdasar immediately noticed that she had the skills and asks her to stay in neurosurgery. Thus, Prof. D. Bagdasar baptized an exceptional physician, blessed a name that would remain in the history of Romanian medicine in the honor gallery." [5]

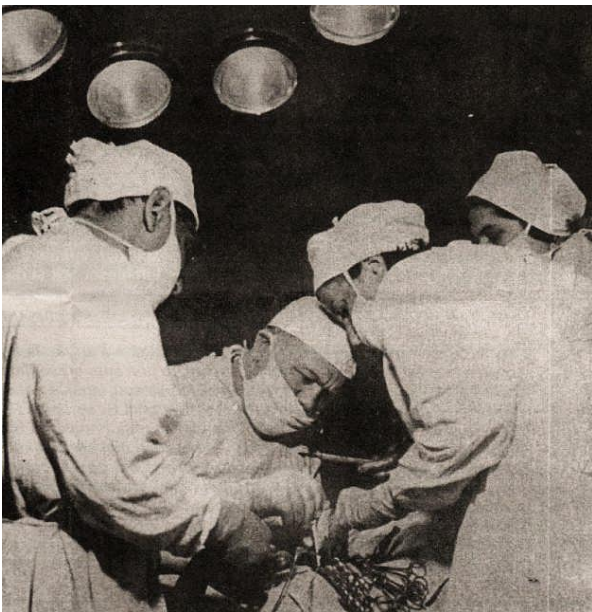


Figure 2. "The Golden Team" during brain surgery.

Prof. Dr. Sebastian Nicolau, Head of the Oncopediatrics Section at Fundeni Hospital, referred to the worldwide validation of the operation performed by Dr. Sofia Ionescu, in the article "Dr. Sofia Ionescu - Primum Inter Pares - the first woman neurosurgeon in Romania", published in the *Reper iatro-istorice J.M.B* magazine: *"The undeniable truth has been brought to light after long iatrics-historical research of over 30 years, on September 17, 2005, at the World Congress of Neurosurgery in Morocco (Marrakesh), which confirmed that Dr. Sofia Ionescu is the first woman neurosurgeon in the world. Until then, it was believed that Dr. Eisenhardt from the U.S.A. holds this title of honor, but it has been proven that the one who practiced medicine in the first decades of the*

twentieth century was an anatomopathologist and not a neurosurgeon."[6].

In the specialized literature, in the article entitled "Sofia Ionescu, the First Woman Neurosurgeon in the World", published in the journal "World Neurosurgery" in March, 2013, it is also mentioned the place occupied by Dr. Sofia Ionescu internationally: *"The nomination as first woman neurosurgeon took place in Marrakech during the 2005 WFNS Congress. Despite the fact that some claim Mrs. Diana Beck to be the first woman neurosurgeon in the World, our theory suggests otherwise. While the first documented surgical intervention performed by Mrs. Diana Beck dates since 1952, Mrs Sofia Ionescu operated for the first time on a human brain as early as 1944. Furthermore, Mrs. Diana Beck's actions surfaced in the World in the year 1947, long after the war had ended and Sofia Ionescu became a neurosurgeon. Last but not least, during the Second World War Mrs. Diana Beck had occupied the position of consultant for neurosurgery and not a fully entitled neurosurgeon."*[1]

The years that followed the early initiation into the mysteries of neurosurgery were devoted entirely to the perpetuation of this specialty at the beginning, the work of Dr. Sofia Ionescu being recognized in 1943 by the Sign of Distinction of the Red Cross. In 1972 she received the Medal for the proclamation of the Republic (25 years) decree no. 480, and in 1996 the Diploma of Honor A.N.F.D.U.R. for exceptional merits. In the General Assembly of the Academy of Medical Sciences she was elected HONORED MEMBER of the Academy, the Ing. Elisa Leonida Zamfirescu Award and the diploma of honor were offered by the National Confederation of Women in Romania for outstanding merits in promoting Romanian science and technology and everything else. In the same year, 1996, she became a Member of the Romanian Society for the History of Medicine. [7],[8].

The intraoperative effort has been doubled by the publication of scientific papers in journals in the country and abroad, such as *Acta Chirurgica Belgica* 1958, *Journal de Chirurgie* 1958, *Psychiatry, Neurology, Neurosurgery* 1960 Amsterdam, *Wiener Klinische Wochenschr* 1962, *Journal of Neurosurgery* 1967, *Rev. Roumaine de Neurosurgery* 1970, *Neurosurgery Stuttgart* 1970 and others, in a total of over 120 articles. Also, Dr. Sofia Ionescu is recognized in the national and foreign press as the first woman neurosurgeon.[9]



Figure 3. Dr Sofia Ionescu, The Lady of Neurosurgery.

CONCLUSION

At the World Congress of Neurosurgery in Morocco, Marrakech it was confirmed that Dr. Sofia Ionescu is the first woman neurosurgeon in the world.[10]

The life model of the Lady of International Neurosurgery, can be regarded as a true *guide of Life Coaching* through the objective judgment, the altruistic action and the power of acceptance that it manifests. The stories coagulated in the book illustrate the art of relentless zeal and ingenuity

CONFLICTS OF INTEREST

The authors declare that the article content was composed in the absence of any commercial or financial relationships that could be construed as potential conflict of interest.

REFERENCES

1. Ciurea AV, Moisa H, Mohan D, Sofia Ionescu. The first woman neurosurgeon in the World. *World Neurosurg.* 80(5):650-3, 2013.
2. Ciurea AV. *Istoria Neurochirurgiei Române*. Editura Viața Medicală Românească, București, 1995.
3. Aldea H: Famous neurosurgeons [in Romanian]. Bucharest: Glasul Bucovinei Publishing House; 1993.
4. Dumitrescu C. *Lungul drum prin mii de nopti albe*. Flacăra, 1987.
5. Simionescu Rodica. *Neodiha binelui - Neurochirurg Sofia Ionescu*. Călărași: Atlas Publishing; 1998.
6. Nicolau S. Doctor Sofia Ionescu – *Primum Inter Pares – Prima Femeie Neurochirurg din România*. *Repere iatroistorice*, 84-6, București, 1945.
7. Arseni C, Aldea H. *Milestones in the history of Romanian neurosurgery* [in Romanian]. Bucharest: Romanian Academy Publishing House; 1988.
8. OGREZEANU Irina, CIUREA AV. Sofia Ionescu – *O biografie de excepție*. *Textbook of neurosurgery*, Bucharest. Romanian Medical Publishing House; vol.1:71-5, 2010.
9. GLIKES CE. An account of the life and achievements of Miss Diana Beck, neurosurgeon (1902 1956). *Neurosurgery* 3:738-742, 2008.
10. OGREZEANU Irina. *Women in neurosurgery. Romania*. The 13th WFNS Congress, Marrakech, Morocco, June 19 24, 2005. Oral presentation mentioned in the abstract book of the Congress.



Incidence of vertebral osteomyelitis and benefits of spinal stabilization in infection. Retrospective analysis of 94 cases in 5 years

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ABSTRACT

In order to evaluate the etiology, characteristics and outcome of the surgical treatment of vertebral osteomyelitis cases in our hospital, patients with vertebral osteomyelitis between January 2014 and December 2018 were included in the study. Clinical and paraclinical data of the patients were collected from the medical records of the patients. Of the 164 patients diagnosed with vertebral osteomyelitis in our clinic 94 underwent surgery and only these last ones were included in the study. Of these 94 patients, 18 cases were diagnosed with tuberculous osteomyelitis and 76 with nontuberculous osteomyelitis. The age of the majority of patients ranged from 40 to 80 years with a peak of incidence between 61-70 years. All of the patients had back pain and regional tenderness of the affected area and many cases presented neurological deficits. The most common involved area affected in our cases was the thoracic spine. Magnetic resonance imaging is the examination that reveals the degree of involvement and excludes other pathologies from the differential diagnosis. The majority of patients had several comorbidities which we included in the study. From all the patients included 76 % underwent surgery with decompression and spinal stabilization using titanium instrumentation and 24 % underwent surgery with only spinal decompression. We also evaluated number hospitalization days of the operated cases. An early diagnosis seems crucial for a fast recovery, lowering the hospitalization costs of both the patient and the hospital and preventing sequelae development.

INTRODUCTION

The inflammation of the intervertebral disk and the surrounding vertebra is defined as vertebral osteomyelitis (VO) or spondylodiscitis. Etiologically it has been divided into 2 groups: tuberculous and nontuberculous osteomyelitis. Adults are the most affected in the general population, most patients diagnosed with this disease being over 50 years old. The clinical signs and symptoms are unspecific and this results in a late diagnose of this pathology. (1, 4, 12)

Keywords

decompression,
osteodiscitis,
spinal fixation surgery,
tuberculous infection,
vertebral osteomyelitis



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The lumbar vertebral bodies of the spine are the most often affected according to literature, being followed by the thoracic and cervical (rare) areas. The clinical presentation of VO is consisted of: acute, subacute or chronic. Tenderness and local regional pain are the most common symptoms in 90% of the patients. Fever is an inconstant sign, being present in 50% of the cases. Neurological deficits may exist when the spinal cord or nerve roots are affected through compression. (6)

The most isolated organism that causes vertebral osteomyelitis is *Staphylococcus aureus* (skin, invasive maneuvers and various percutaneous catheters may be an entry portal), being followed by *Escherichia coli*, Coagulase-negative staphylococci, *Propionibacterium acnes*. Gram-negative aerobic bacteria and *Candida spp* are common microorganisms are discovered in iv drug abusers and immunosuppressed patients. These represent various etiologies for nontuberculous osteomyelitis, while *Mycobacterium Tuberculosis* is the main factor for developing tuberculous (TB) vertebral osteomyelitis. (1, 7, 10, 13, 16)

Biopsy of the tissue has a high diagnostic accuracy. Leukocytosis is found in 50 % of cases and has a low diagnostic sensitivity. A raised erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP) levels are highly sensitive. A good clinical response to treatment is noted by lowering of the CRP levels. (1, 4, 9, 16)

Imaging studies used for the diagnosis of VO are represented by radiography, scintigraphy, CT scan and MRI. The MRI has high diagnostic sensitivity and specificity. For obtaining an anatomopathological and microbiological based diagnostic CT-guided fine-needle aspiration biopsy (FNAB) is used and has a sensitivity of 50%. (1)

The aim of the treatment is to eradicate the infection, relieve the pain and restore the neurological status of the patient. (1)

Our purpose of the study was to describe a series of vertebral osteomyelitis cases in our hospital, to differentiate de factors that cause or predispose to the formation of this infection and show the benefits of a surgical stabilization treatment for axial stability and early mobilization.

METHODS

Type of study

We conducted a retrospective descriptive study, in

which we included patients operated for osteomyelitis / primary vertebral osteodiscitis in the neurosurgery clinic of the Emergency Clinical Hospital Bagdasar - Arseni for a period of 5 years (January 1, 2014 – December 31, 2018). The study is based on the experience of the Neurosurgery II and IV departments of the hospital. It is the only study conducted in our country over a longer period of time and on a large number of patients, with the theme of multimodal treatment of vertebral osteomyelitis.

We had the method of retrospective revision of the electronic observation sheets, of the patients' files taken from the hospital archive and the operative protocols of the patients with osteomyelitis / primary vertebral osteodiscitis operated in the previously mentioned period. Based on the information obtained, we created a database using the Microsoft Office Excel 2007 program. The statistical analysis was performed using the IBM SPSS Statistics 20 program. We created a database that can be taken as a starting point in establishing medical and surgical treatment protocols for patients with vertebral osteomyelitis.

On a national level, there is no evidence of reported cases of vertebral osteomyelitis and there are no protocols for unitary treatment of this condition.

As inclusion criteria, only patients with osteomyelitis/osteodiscitis operated between January 1, 2014 - December 31, 2018 were included in the study.

Patients with vertebral osteomyelitis who received only conservative treatment without surgery or patients with osteodiscitis secondary to operated disc herniation were excluded from our study.

The variables introduced in the statistical analysis were nominal, dichotomous (binary, bimodal), ordinal and quantitative (measurable) discontinuous (discrete). Depending on the variables studied, we performed uni, bi and multivariate statistical analyzes.

RESULTS

Between January 1, 2014 and December 31, 2018, 164 patients were hospitalized and diagnosed with Vertebral Osteodiscitis in the Neurosurgery Clinic of the Bagdasar-Arseni Emergency Clinical Hospital. Of

these, 94 patients (57.31%) underwent surgery for vertebral osteomyelitis.

Sexual distribution

The distribution by sex was slightly in favor of males, 52 patients being male, representing 55% were operated for vertebral osteomyelitis, compared to 42 female patients, representing 42% operated for the same pathology. The sex ratio of M / F was 1.23 (Figure 1).

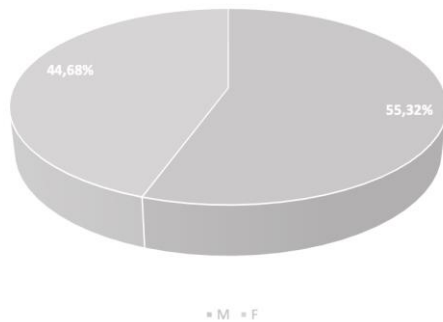


Figure 1. Sexual distribution of the patients.

Age distribution

In the studied group we distributed the patients by decades of age between 40 and 80 years. The vast majority of patients were in the age range of 51-70 years, respectively 27 patients (29% of cases) in the range of 51-60 years and 32 patients (35% of cases) in the range of 61-70 years. This is justified by the fact that these age groups include relatively active patients, who also perform physical exertion, but who have a personal history that may promote the occurrence of vertebral osteomyelitis.

The rest of the patients are distributed in lower percentages in the interval under 40 years old being 7 patients (8% of cases), and between 41-50 years old and over 70 years old 14 patients (15% of cases) in each group (Figure 2).

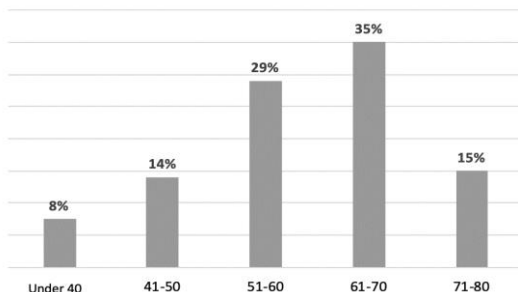


Figure 2. Age distribution of the patients.

Duration of hospitalization of patients in the study group

On the studied group, the hospitalization period varied between 0-10 days, 11-20 days, 21-30 days and respectively hospitalized patients for a period of over 30 days. Among the patients studied, the highest percentage was represented by those hospitalized for less than 20 days, who opted for vertebral fixation with titanium instrumentation, in 55 cases (70%), followed by those hospitalized between 21-30 days, in a number of 20 cases (21%) and respectively those hospitalized for a period longer than 30 days, in a number of 4 patients (4%), in which only the vertebral decompression was practiced (Figure 3).

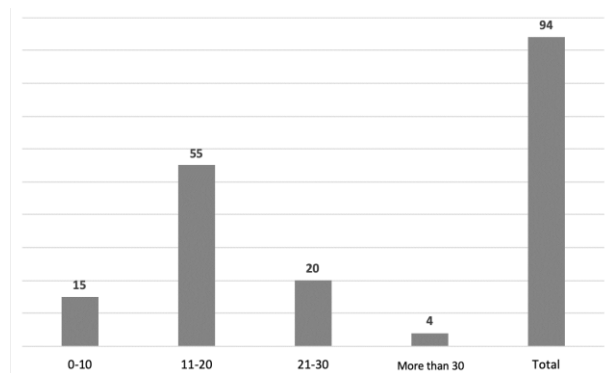


Figure 3. Number of hospitalization days of the patients.

Secondary Diagnostics

From the point of view of the pathology associated with patients operated for Vertebral Osteomyelitis, a number of 25 cases (26.6%) had diabetes, 10 cases had a personal history of pulmonary TB (10.6%), 8 cases had had hepatitis B or C virus infection (8.5%).

Out of the total number of patients, 43 cases (45.7%) were diagnosed and operated for Vertebral Osteomyelitis on the background of an immunosuppression already present due to the pathologies mentioned above (Figure 4).

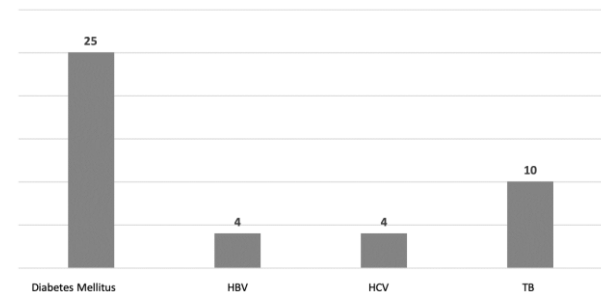


Figure 4. Associated pathologies of the patients.

Clinical Examination

Lumbar back pain that translates to vertebral instability syndrome is present in all patients with vertebral osteomyelitis. The neurological syndrome occurs in 57 of patients with vertebral osteomyelitis in the study group (60.6%). Of these, Frankel C and D type paraparesis dominates the clinical picture, Frankel C 25 cases (26.5%) and Frankel D 24 cases (25.5%). 5 patients from the studied group were Frankel type A (5.3%), and 3 patients type Frankel B (3.2%) (Figure 5).

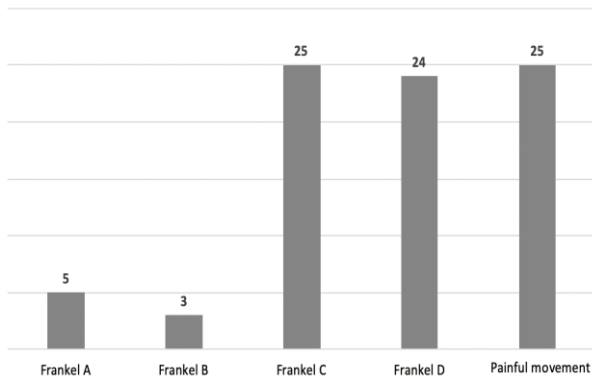


Figure 5. Preoperative status of the patients according to Frankel grading system.

Localization of the vertebral lesion

In the studied group, the vertebral seat of the inflammatory lesion predominates in the thoracic and lumbar region, thoracic osteomyelitis is found in 42 patients (45% of cases), lumbar localization in 40 patients (42% of cases) and cervical in the remaining 12 patients. (13% of cases). (Figure 6)

This is important in choosing the surgical approach and considering the importance of spinal fixation during surgery.

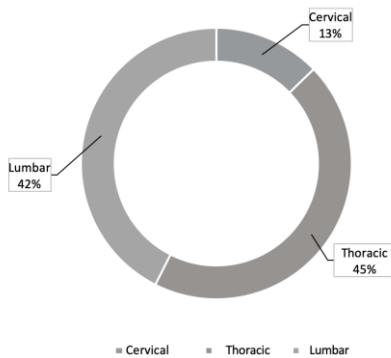


Figure 6. Incidence of the affected spinal region in the patients studied.

SURGICAL TREATMENT

The surgical treatment has as important objectives: evacuation of infection and debridement, decompression of the dural sac and spinal roots and vertebral fixation / fusion.

Surgical decompression involves debridement of the inflammatory focus and evacuation of pus from the intervertebral disc space, curettage of cartilaginous and bony necrotic-inflammatory tissue, evacuation of intra- and paraspinal abscesses, and decompression of the dural sac and spinal nerves.

The vertebral fixation at the level of the thoracic and lumbar spine is performed using transpedicular screws and titanium rods, but also the reconstruction techniques of the anterior spine, with the help of fixed or expandable titanium cages. The use of titanium instrumentation in vertebral osteomyelitis is a method in continuous debate, but recent studies show the advantages of vertebral stabilization and the compatibility of titanium with infection.

The group we studied includes patients operated on 2 neurosurgery departments in the Bagdasar Arseni Emergency Clinical Hospital by several surgical teams. Some surgeons have chosen only surgical decompression alone as a method. In addition to decompression, others also opted for spinal fixation with titanium instrumentation.

Thus, in the studied group, 72 patients (24%) benefited from internal fixation with titanium instrumentation, while in the remaining 22 cases (76%) only the evacuation of the infectious outbreak and the decompression of the nervous elements were performed, without metal fixation. (Figure 7)

The aim of this study is to perform a comparative analysis of postoperative results in patients operated for vertebral osteomyelitis in which metal fixation was performed compared to those without metal fixation.

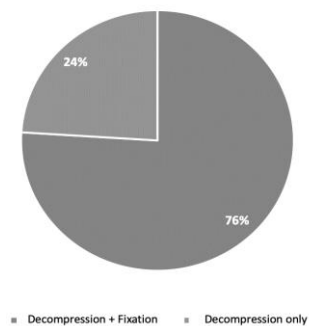


Figure 7. Type of surgical intervention performed on the studied patients.

POSTOPERATIVE MOBILIZATION

Due to the improvement of the implantation techniques of the titanium fixing systems (stronger screws, more elastic rods), the period of postoperative immobilization of the patient decreased significantly. This refers to the degree of vertebral stability obtained after surgery, the degree of preoperative neurological injury and the general pathology associated with vertebral osteomyelitis (diabetes, chronic hepatitis, heart failure, COPD).

The period in which the postoperative mobilization took place varied between 5 and 14 days, this being possible in 75 of (80% of cases). In 19 patients (20% of cases) early mobilization was not possible, requiring postoperative bed rest, which ranged from 6 to 8 weeks. (Figure 8)

Early mobilization of patients was corroborated in particular with the achievement of spinal fixation at the same time as spinal decompression.

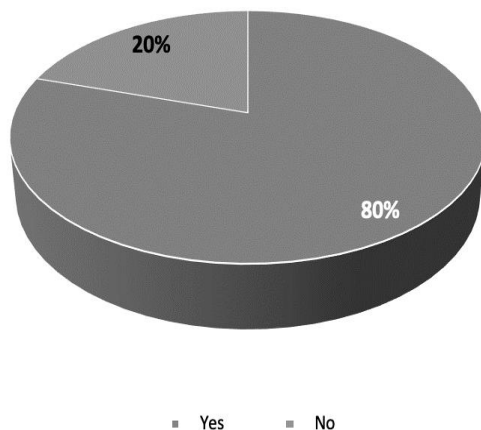


Figure 8. Early postoperative mobilization of the patients within 5 to 14 days.

HISTOPATHOLOGICAL RESULTS

In absolutely all patients, biopsy was collected from the intervertebral disc, adjacent bone plateaus and evacuated spinal abscesses. The goal was to establish the etiology of the infectious outbreak for proper antibiotic therapy. Therefore, the diagnosis of specific granulomatous osteomyelitis (TB) was found in 18 patients (19% of cases), and that of nonspecific osteomyelitis in 76 patients (81% of cases) (Figure 9).

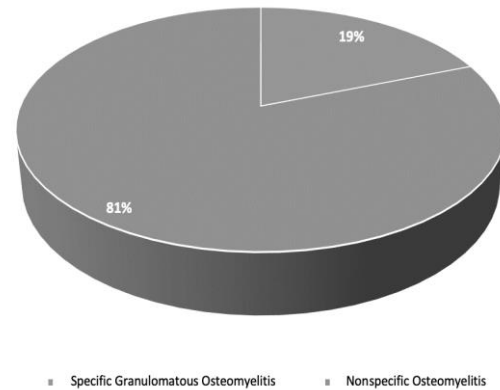


Figure 9. Incidence of Tuberculous Osteomyelitis in the studied cases.

DISCUSSION

Vertebral osteomyelitis is a rare disease with an incidence of 5.3 cases per million in one year (3,11). The incidence is growing because of the increasing number of elderly patients with severe comorbidities. The invasive options used for the diagnosis and treatment of this disease (3,16). The diagnosis is in most of the cases delayed due to the prolonged onset of this disease (11).

Tuberculous osteomyelitis is a disease that can rapidly alter the patient's prognosis, especially if it becomes infected and coexists with pyogenic osteomyelitis (14,15). Non-granulomatous VO has a higher incidence than TB VO and this is concluded in our study as well. Specific granulomatous osteomyelitis was detected in 19% of cases, the rest having a nonspecific anatomopathological result (81% of cases).

Men are more affected than women by VO (1,11). The disease may be diagnosed at any age, but it has a higher incidence in elderly patients (14). In our study the sex ratio between males and females was 1.23. We showed that the average age was between 61-70 years. Age groups were dominated by the following percentages: patients between 61-70 years (35%), 51-60 years (29%), 71-80 years (15%), 41-50 years (14%) and under 40 (14%). This demonstrates the increased frequency of vertebral osteomyelitis in elderly patients.

In terms of patient characteristics, it is described in previous papers that diabetes mellitus and chronic

renal insufficiency are the most frequent comorbidities in patients with hematogenous related vertebral osteomyelitis (2, 5, 8). In our study, the most common personal pathologies were diabetes mellitus (26.6%), pulmonary TB (10.6%) and hepatic viral infections with HBV and HCV (8%). It has been shown in the study group that patients with a deficient immune system are prone to the appearance of vertebral osteomyelitis.

The lumbar spine is the most common affected level (45-50%), followed by the thoracic, cervical and sacral region. In our study we revealed that the location of the lesion is predominantly thoracic (45%), followed by the lumbar location (42%) and then the cervical location (13%). The thoracic spine is the most common site of vertebral osteomyelitis in the patients studied.

Back pain and muscle spasm of the paravertebral muscles are the most common symptoms of the patients. Fever is present in 10-45 % of the cases (3, 11). Other clinical findings are represented by: decreased spine motion, paresthesia, anorexia, alteration of the patient's general state, swelling of the affected region and tenderness at palpation. If the infection spreads into the epidural space this results in the manifestation of neurological deficits (11). In our study, the vast majority of patients have lumbar back pain and spinal instability in the clinical picture. Neurological syndrome occurs in 60.6% of all cases studied, this is due to the late presentation at hospital.

In the study we conducted, decompression of the dural sac and nerve elements, followed by spinal fixation with titanium instrumentation in the same surgery (76% of cases) shows a rapid postoperative mobilization and a shortening of the hospitalization period, compared to patients who underwent only for surgical decompression (24% of cases).

Postoperative mobilization was possible in 80% of patients being done from the first 5-14 days after surgery, which demonstrates the effectiveness of using titanium instrumentation for spinal fixation after spinal decompression.

The average length of hospitalization of patients was less than 20 days. Out of the total of 94 cases studied, a number of 70 cases had a hospitalization time of less than 20 days, 20 cases with hospitalization between 21-30 days and respectively 4 patients with a hospitalization duration of over 30 of days. In the study group, patients treated with

spinal fixation with titanium instrumentation had a shorter hospitalization time than those operated without titanium instrumentation.

Following the study we conducted, we can relate that titanium instrumentation vertebral fixation for instability caused by osteomyelitis allows early mobilization of patients, faster healing of the inflammatory outbreak, reduction of postoperative complications and decreased hospitalization days.

CONCLUSION

Vertebral osteomyelitis occurs in both men and women, with a predisposition for the male population.

In addition to old age, other diseases, such as diabetes, chronic kidney failure with dialysis pose a risk for increased incidence. Although vertebral osteomyelitis is found in elderly patients, the infection is frequently reported, especially in patients over 60, who are associated with various diseases, such as diabetes, hepatitis or pulmonary TB.

The symptoms can be diverse, the dominant symptom is back pain. This translates the pain given by the infection / inflammation but also the spinal pain of vertebral instability. Neurological symptoms may range from radiculopathy to spinal cord or ponytail syndrome.

The site of the vertebral lesion can be anywhere on the spine, but the thoracic location predominates, then the lumbar one.

The surgical treatment aims at the debridement of the inflammatory focus and the evacuation of the pus, the decompression of the dural sac and of the nervous elements. The treatment of spinal instability using fixation-fusion and vertebral reconstruction techniques is a controversial topic, but studies in the literature show the compatibility of instrumentation with infection. The choice of surgical approach depends on the location of the lesion in the spine.

Early mobilization after surgery is an important goal that can be achieved in patients with metal fixation with titanium instrumentation, without postoperative vertebral instability. Early mobilization, especially in elderly patients with various associated pathologies, can significantly reduce the rate of complications associated with prolonged immobilization.

REFERENCES

1. Barbari EF, Steckelberg JM, Osmon DR (2005) Osteomyelitis. In: Mandell GL, Bennett JE, Dolin R (eds) Principles and practice of infectious diseases, vol 1, 6th edn. Churchill Livingstone, Philadelphia, pp 1322–1332.
2. Carragee EJ: Pyogenic vertebral osteomyelitis. *J Bone Joint Surg Am* 1997, 79:874-80.
3. Colmenero JD, Jiménez-Mejías ME, Sánchez-Lora FJ, Reguera JM, Palomino Nicas J, Martos F et al (1997) Pyogenic, tuberculous, and brucellar vertebral osteomyelitis: a descriptive and comparative study of 219 cases. *Ann Rheum Dis* 56:709–715.
4. Currier BL, Eismont FJ (1992) Infections of the spine. In: Rothman RH, Simeone FA (eds) *The Spine*, 3rd edn, vol 2. W.B Saunders Company, Philadelphia, pp 1319–1380.
5. Hadjipavlou AG, Mader JT, Necessary JT, Muffoletto AJ: Hematogenous pyogenic spinal infections and their surgical management. *Spine (Phila Pa 1976)* 2000, 25:1668-79.
6. Jaramillo-de la Torre JJ, Bohinski RJ, Kuntz C (2006) Vertebral osteomyelitis. *Neurosurg Clin N Am* 17:339–351.
7. Jensen AG, Espersen F, Skinhoj P, Frimodt-Moeller N (1998) Bacteremic *Staphylococcus aureus* spondylitis. *Arch Intern Med* 158:509–517.
8. Krogsgaard MR, Wagn P, Bengtsson J: Epidemiology of acute vertebral osteomyelitis in Denmark: 137 cases in Denmark 1978-1982, compared to cases reported to the National Patient Register 1991-1993. *Acta Orthop Scand* 1998, 69:513-7.
9. Lew DP, Waldvogel FA (2004) Osteomyelitis. *Lancet* 364:369–379.
10. Mc Henry MC, Easley KA, Locker GA (2002) Vertebral osteomyelitis: Long-term outcome for 253 patients from 7 Cleveland-area hospitals. *CID* 34:1342–1350.
11. Modic MT, Feiglin DH, Piraino DW, Boumpfrey FR, Weinstein MA, Duchesneau PM, Rehm SU. Vertebral osteomyelitis: assessment using MR. *Radiology*. 1985 Oct;157(1):157-66.
12. Mylona E, Samarkos M, Kakalou E, Fanourgiakis F, Skoutelis A (2009) Pyogenic vertebral osteomyelitis: a systematic review of clinical characteristics. *Sem Arthritis Rheum* 39:10–17.
13. Pirest DH, Peacock JE (2005) Hematogenous vertebral osteomyelitis due to *Staphylococcus aureus* in the adult: clinical features and therapeutic outcomes. *South Med J* 98:854–862.
14. Tsiodras S, Falagas ME (2006) Clinical assessment and medical treatment of spinal infections. *Clin Orthop Relat Res* 444:38–50.
15. Weisz RD, Errico TJ (2000) Spinal infections diagnosis and treatment. *Bull Hosp Joint Dis* 59:40–46
16. Zimmerli W (2010) Vertebral osteomyelitis. *N Engl J Med* 362:1022–1029.



Clinical spectrum of paediatric head injury. A prospective study from tribal region

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ABSTRACT

Introduction: Traumatic Brain injury is considered as a major health problem which causes frequent deaths and disabilities in paediatric population with special concern to tribal regions of developing countries like India where etiology of traumatic brain injury in the paediatric population fall from height dominant over the road traffic accident as a major.

Aim and objective: The aim is to analyse the epidemiology, mechanism, clinical presentation, severity and outcome of paediatric head injury in the tribal region of northern India that could help to make preventive policies to improve their care.

Material methods: It is a prospective observational study of 345 children of up to 18 years of age admitted under Department of Neurosurgery from October 2017 to April 2019.

Results: The study population comprised of 345 paediatric patients. Mean age was 9.25 years. 36.81% patients were in 1-6-year age group and male to female ratio was 2.45. The most common cause for trauma was fall from height in 179(52%) cases followed by RTA in 141(41%) cases. The most common radiological finding was depressed skull fractures in 97(50%) cases. There was 35% mortality in severe head injury patients.

Conclusion: This study through some light on the different scenario of head injury in Tribal regions of Developing country and will help to formulate effective strategies for prevention and better care of the patients.

INTRODUCTION

Traumatic brain injury (TBI) is a leading cause of death and disability in children worldwide. [1] Young children are at relatively high risk of minimal and mild traumatic head injuries. An increase in the more severe and fatal traumatic brain injuries has been found in late adolescence. Paediatric TBI has different Pathophysiology due to higher vascularity, plasticity and less rigidity of scalp. Paediatric brain has less degree of myelination which related to brain capacity to absorption of traumatic forces and increase the susceptibility to TBI.[2] TBI is classified as mild (Glasgow Coma Scale [GCS] 13-15), moderate (GCS 9-12), or severe (GCS 3-8).[3]

The study focuses on understanding the etiology, clinical presentation, treatment options, and outcome of these patients with special concern to tribal regions of developing countries like India

Keywords
tribal region,
paediatric traumatic brain
injury



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where fall from height dominant over the road traffic accident as a major etiology of traumatic brain injury in paediatric population.[4] Thus there is a critical need for effective fall and traffic accidents prevention strategies for children, and we should give attention to the predicting factors for more effective care of such patients

MATERIAL METHODS

It is a prospective observational study of 345 children of upto 18 years of age admitted under Department of Neurosurgery from October 2017 to April 2019. Study was started after obtaining the permission from ethical committee of the hospital. Informed consent was obtained from the parent/guardian/relative of the patient. A detailed clinical history obtained from the parents/guardian/relative admitted in the hospital with head injury.

ANALYSIS

Statistical analysis was performed using the collected data on incidence and clinical– radiologic correlation. Analyses included the age and gender distribution of the Children, the cause and location of the injury, medical status, and the part of the head injured and type of injury and the treatment provided. A comparison of types of head injuries sustained by gender, age, and cause was also carried out.

RESULTS

The study population comprised of 345 paediatric patients aged Between 2 months to 18 years with a mean age of 9.25 years. There were 245 males (71.01%) and 100 females (28.98%) with male to female ratio of 2.45:1.

Age	No	%	Male	Female	Good outcome	Poor outcome
<1yr	24	6.95	14	10	18(75%)	6(25%)
1-6yrs	127	36.81	95	32	108(85.03%)	19(14.07%)
7-12yrs	120	34.78	84	36	110 (91.7%)	10 (8.3%)
13-18yrs	74	21.44	52	22	62(83.8%)	12(16.2%)
TOTAL	345	100%	245(71.1%)	100(28.9%)	298	47

Table 1. Outcome according to age.

The most common cause for trauma was fall from height in 179(52%) cases followed by RTA in 141(41%) cases, Bull horns in 11 (3%) cases; assault 7 (2%) cases.

Mechanism	No	%	Mild	Moderate	Sever
Fall	179	52	93(52%)	73(41%)	13(7%)
RTA	141	41	38(27%)	72(51%)	31(22%)
Assault	7	2	5(72%)	1(14%)	1(14%)
Sport	7	2	7(100%)		
Bull horn	11	3%	8(73%)	2(18%)	1(10%)

Table 2. Mode of injury.

RTA: Road traffic accident

Severity of injury was decided with GCS score at the time of admission and it was mild injury in 218 (63.19%), moderate in 92 (26.67%) and sever in 35 (10.14%) cases. Outcome of injury was good in mild head injury group and poor outcome was associated with severe head injury group.

GCS	Severity	No	Good outcome	Poor outcome	Mortality
13-15	Mild	218	210(96.33%)	8(3.67%)	None
9-12	Moderate	92	60(65.22%)	22(23.91%)	10(10.87%)
3-8	sever	35	9(25.71%)	15(42.86%)	11(31.43%)

Table 3. Severity and outcome.

CT scan findings were positive in 195 cases and it was normal in 150 cases. Most common radiological finding was Depressed skull fractures in 97(50%) cases, Extradural hematoma in 29 (15%), Subdural hematoma in 20 (10%), contusion in 23 (12%), ICH in 6 (3%), IVH in 2 (1%) cases. (Figure 1 and 2)



Figure 1. NCCT Head of 8 yrs old child admitted with h/o of fall from roof with GCS E2V3M5 Pupils b/l 3mm reacting to light.

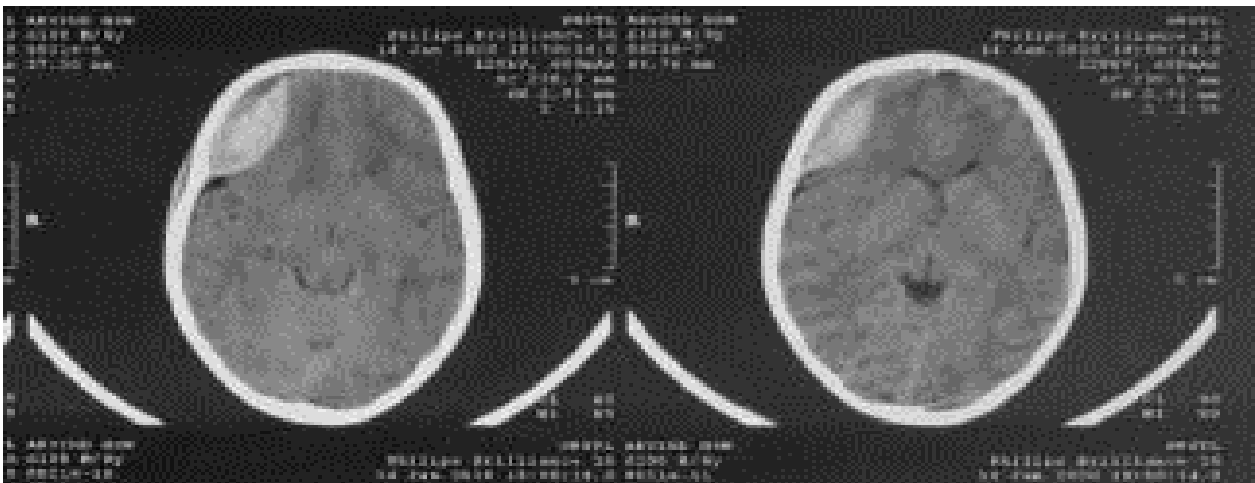


Figure 2. NCCT Head of 6 yrs male child admitted with h/o fall from tree and GCS at admission E4V3M3, Pupils b/l 3 mm reacting, after evacuation of EDH Child improved and at time of discharge GCS was E4V5M6.

CT Finding	No	%	Surgery	Conservative
Skull fractures	97	50%	40	57
EDH	29	15%	10	19
SDH	20	10%	8	12
Contusions	23	12%	11	12
ICH	6	3%	2	4
IVH	2	1%	0	2
Edema	18	9%	0	18
Total	195	100%	71	124

Table 4. Radiology positive in 195 cases and negative in 150 cases.

EDH: Extradural Hematoma, SDH: Subdural Hematoma, ICH: Intracerebral Hematoma, IVH: Intra ventricular Hematoma

Out of 195 patients 71 patients' required surgery and rest 124 patients were managed conservatively. Fracture debridement or elevation done in 40 patients, Hematoma (EDH+ICH) evacuation was done in 12 patients, Contusectomy done in 11 patients and decompressive craniectomy done in 8 patients. (Figure-3 and 4)

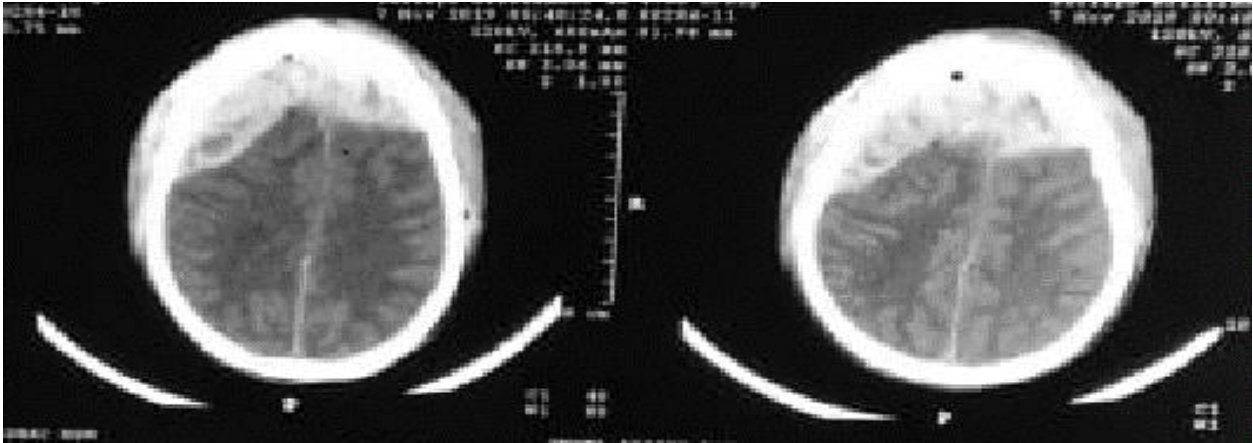


Figure 3a. NCCT Head of 15yrs male admitted with h/o RTA with GCS E2V1M4 Pupils b/l 3mm reacting to light undergone bifrontal craniectomy with evacuation of EHD. He discharged with GCS E4V5M6 and under gone 3D Mold customized cranioplasty after 4 months.

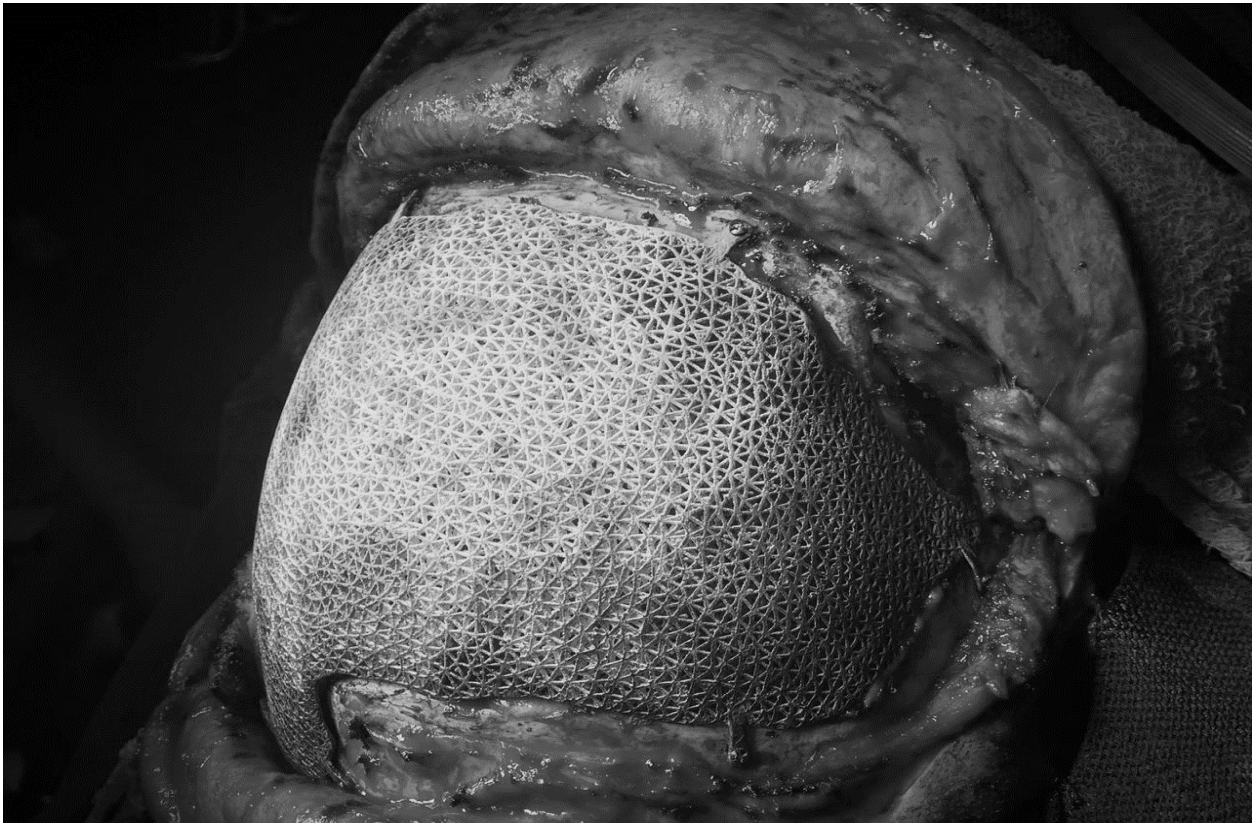


Figure 3b. Intraoperative photograph of 3D Customised cranioplasty flap of above-mentioned patient.

Intervention	Total	Good outcome	Poor outcome
Hematoma evacuation	10(EDH) +2(ICH)	10(83.33%)	2(16.67%)
Contusectomy	11	5(45.4%)	6(54.5%)

Decompressive craniectomy	8	3(37.5%)	5(62.5%)
Fracture debridement or elevation	40	35(87.5%)	5(12.5%)
Total	71	53(74.65%)	18(25.35%)

Table 5. Intervention and outcome.

EDH: Extradural Hematoma, ICH: intracerebral Hematoma

While doing the survey for associated injuries, out of 345 patients 105 patients have associated injuries and most common associated injury was facial injury in 42 patients followed by long bone fracture in 24 patients, chest injury in 10 patients, multiple injury in 16 patients, spinal injury in 8 patients and abdominal injury in 5 patients.

Associated injury	Good outcome	Poor outcome	Total
Nil	226(94.17%)	14(5.83%)	240
Facial injury	38(90.48%)	4(9.52%)	42
Limb fracture	22(91.67%)	2(8.33%)	24
Spinal injury	5(62.5%)	3(37.5%)	8
Chest injury	8(80%)	2(20%)	10
Abdominal injury	4(80%)	1(20%)	5
Multiple injuries	14(87.5%)	2(12.5%)	16
Total			345

Table 6. Associated injury and its outcome.

DISCUSSION

In India children below 18 years of age constitute about 40% of the total population [5]. Traumatic brain injury is listed as one of the most common cause of death in pediatric population. Our study on pediatric head injuries show male preponderance which also conformed in various studies [6].

In our study fall from height 179(52%) was the most common cause of pediatric head injury [7]. This peculiarly occurs due to fall from tree, unguarded rooftop while playing. This was followed by RTA 141 (41%), bull horn 11(3%), assault 7(2%) and sport related injury 7 (2%) [8].

Initial GCS score was the single most important factor affecting the outcome as described by Beca et al [9].

The patient who had a GCS of 13-15 (218) had a poor outcome in 8 (3.67%), followed by GCS of 9-12 (92) who had a poor outcome in 22(23.91%) followed by GCS of 8 or less than 8 (35) who had poor outcome in 15(42.86%) which is similar as reported by Astrand R et al [10].

Out of 345 patients in our study, CT scan findings were positive in 195 cases and it was normal study in 150 cases. We found isolated skull bone fracture as most common CT findings in 97 (50%) cases, it was

associated with good outcome (87.5%) similar results were described by Suresh et al [11]. Hematoma evacuation (EDH, ICH) was associated with good outcome in 83.3% and poor outcome in 16.67%, contusion was associated in good outcome in 45.4% and poor outcome in 54.6% cases and decompressive craniectomy was associated with poor outcome in 62.5% cases. Tomberg et al also found the similar outcome in their study [12].

In our study, we found that 69.6% patient have isolated head injury with good outcome in 94.17% patient. Facial injury was seen in 12.17%, limb fracture in 7%, spinal injury in 2.3%, chest injury in 3%, abdominal injury in 1.5% and multiple injuries seen in 4.6% patients. Paret et al reported chest trauma in (62%), limb fracture in (32%), facial injury in (20%), and multiple injuries in (48%) cases. This difference is because we include all the children in our study irrespective to severity of the injury while author include only severe cases of head injury in pediatric patient [13].

The overall outcome in our study was death in 21(6.09%), vegetative state in 10(2.90%), severe disability in 12(3.48%) and good outcome in 279(80.86%) of the cases which was similar to study conducted by Abrar Ahad Wani et al [14].

GOS N (%)
Death (1) 21(6.09%)
Vegetative (2) 10 (2.9%)
Sever disability (3) 23 (6.67%)
Moderate disability (4) 12 (3.48%) Normal (5) 279 (80.86%)

Table 7. Glasgow outcome scale.

GOS: Glasgow Outcome Scale

CONCLUSION

Our study highlights the different scenario of pediatric head injury in Tribal regions of Developing country, where even the minor head injuries are referred to tertiary care hospitals which can be easily managed by treating physicians. The findings of our study have implications for development of public health policy with especial reference to tribal regions of developing country. Where more than half of pediatric head injury which are minor in nature can be prevented by just increasing public awareness.

CONFLICTS OF INTEREST

Financial support and sponsorship: Nil

Conflicts of interest: There are no conflicts of interest.

REFERENCES

- Jagannathan J, Okonkwo DO, Yeoh HK, Dumont AS, Saulle D, Haizlip J, et al. Long term outcomes and prognostic factors in pediatric patients with severe traumatic brain injury and elevated intracranial pressure. *J Neurosurg Paediatr.* 2008; 2:237-9. [Pub Med: 18831656].
- Goldsmith W, Plunkett J: A biomechanical analysis of the causes of traumatic brain injury in infants and children. *Am J Forensic Med Pathol* 25: 89-100, 2004.
- Jennet B. Epidemiology of head injury. *Arch Dis Child* 1998; 78:403-6.
- Adirim TA, Wright JL, Lee E, Lomax TA, Chamberlain JM. Injury surveillance in a pediatric emergency department. *Am J Emerg Med.* 1999; 17:499-503. [Pub Med: 10530522].
- International Institute for Population Studies. Mumbai, India: NF, India; 1998-9.
- Mahapatra AK. Head injury in children. In: Mahapatra AK, Kamal R, Editors. *A Text Book of Head Injury.* Delhi: Modern Publication; 2004.p. 156-70.
- Sambasivan M. Epidemiology-Pediatric head injuries. *Neurol India* 1995; 43:57-8.
- Osmond MH, Brennan-Barnes M, Shephard AL. A 4-year review of Severe pediatric trauma in Eastern Ontario: A descriptive analysis. *J Trauma* 2002; 52:8-12. Beca J, Cox PN, Taylor MJ, Bohn D, Butt W, Logan WJ, et al.
- Somatosensory evoked potentials for Prediction of outcome in acute severe brain injury. *J Pediatr.* 1995; 126:44-9. [Pub Med: 7815222].
- Astrand R, Undén J, Hesselgard K, Reinstrup P, and Romner B. Clinical factors associated with intracranial complications after pediatric traumatic head injury: An observational study of children submitted to a neurosurgical referral unit. *Pediatr Neurosurg.* 2010; 46:101-9. [Pub Med: 20664236].
- Suresh HS, Praharaj SS, Indira Devi B, Shukla D, Sastry Kolluri VR. Prognosis in children with head Injury: An analysis of 340 patients. *Neurol India.* 2003; 51:16-8. [Pub Med: 12865508].
- Tomber g T, Rink U, Tikk A. Computerized tomography and prognosis in pediatric head injury. *Acta Neurochir (Wien)* 1996; 138:543-48.
- Paret G, Ben Abraham R, Berman S, Vardi A, Harel R, Manisterski Y, et al. Head injuries in children - Clinical characteristics as prognostic factors. *Harefuah.* 1999; 136:677-81. 755. [PubMed: 10955086].
- Abrar Ahad Wani, Arif Hussain Sarmast, Muzaffar Ahangar pediatric head injury: A study of 403 cases in a tertiary care hospital in a developing country. *J Pediatr Neurosci.* 2017 Oct-Dec; 12(4): 332-337.doi: 10.4103/JPN_80_17.



Coccydynia - a medical condition with multiple causes and different management strategies. A local experience

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ABSTRACT

Background: Coccydynia is a common complaint which is usually self-limited and tolerable. Although most of these patients respond dramatically to conservative measures, some patients need more complex plane of treatment. In these conditions, the cause of coccydynia can be complex and multifactorial.

Patients and methods: A total of 8 cases were included in the study. All cases received medical treatment for 9 months before surgery. Complete coccygectomy was performed for all cases. Post-operative outcomes were measured according to the degree of pain relief.

Results: The mean age of the included cases was 43.87 years (range, 39 – 52). A total of 5 females (62.5%) and 3 males (37.5%) were included. The mean preoperative VAS score was 9 (range, 8 – 10), while post-operatively, it decreased significantly down to 2 (range, 1 – 3). Excellent postoperative outcomes were achieved in 6 cases (75%) whereas good outcomes were obtained in the remaining 2 cases (25%).

Conclusion: Although conservative medical measures are effective for treating coccydynia, but surgical excision is still a valid treatment option in resistant cases.

INTRODUCTION

Although being a small bone the coccyx has many important functions. As we all know it gives insertion for many muscles, ligaments, and tendons which play a main role in the pelvic floor support and actively share in voluntary bowel control. It is also considered one limb of the tripod—in addition to the two ischial tuberosities—that represents weight-bearing support to a person while sitting [14].

Coccydynia is pain in the region of the coccyx. Simpson first announced the definition in 1859, but reports about this pain dated back to the 16th century. Although this condition is well known since a long time, its management may be a dilemma and a real controversial issue in resistant cases as it is multifactorial in nature. Several organic and psychological causes contribute to its occurrence. Most of these

Keywords
coccydynia,
coccygectomy,
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cases resolve within a short time with or without conservative measures, but for a small percent of these patients, the condition may become chronic and debilitating [19].

The accurate incidence of this condition has not been known exactly; however, high risk factors for coccydynia include obesity and female sex. Female to male ratio is about five times. Children are less likely to develop this condition than adolescents and adults. Marked rapid loss of weight as in bariatric surgery may increase the risk of this condition due to loss of subcutaneous fat and mechanical cushioning [12].

Coccydynia is commonly caused by external or internal trauma. The external trauma is usually caused by a backwards fall, childbirth, especially associated with obstructed or instrumented labor, sitting for long time on hard, or uncomfortable seats. Nontraumatic coccydynia may be caused by degenerative affection of joint or disc disease, infection and variants of coccygeal morphology. Coccydynia may be caused by nonorganic etiologies, such as somatization and other psychological disorders [13].

Although most cases respond to conservative treatment, few patients fail to show a good response and hence, surgery may be an option [17].

Historically, the surgical management of such condition has been discussed with caution as it was associated with poor results and high complication rates. Nevertheless, better outcomes have been reported recently [18].

This study was conducted at Mansoura University Hospitals aiming to evaluate the surgical outcomes of coccygectomy for coccydynea refractory to conservative treatment.

PATIENT AND METHODS

Study design

This is a prospective study that was conducted at Mansoura University Hospitals during the period between December 2016 and December 2018.

Study cases

A total of 8 cases (n = 8) with coccydynia who showed no response to 9-month duration of failed all conservative measures including physical therapy, coccydynia exercise, medications, injections, and even psychotherapy were included in our study. All of our cases had a history of trauma to the coccyx.

Inclusion criteria

Patients older than 18 years, no contraindication for anaesthesia or surgery, having pain at the region of coccyx, tenderness over the same area, or the presence of coccygeal abnormalities.

Exclusion criteria

Patients younger than 18 years, and patients who are not candidates for anaesthesia or operation were excluded from our study.

Patient consent

A pre-operative written informed consent was obtained from all cases after the explanation of advantages and drawbacks of the surgical approach. Moreover, the study was approved by the local ethical committee.

Patient preparation

All cases were subjected to complete history taking, thorough physical examination, and routine laboratory investigations. Digital rectal examination was also performed to exclude the presence of rectal pathology. During rectal examination the coccyx was grasped between the forefinger and thumb then we started to manipulate the coccyx to detect pain, hypermobility or hypomobility of the sacrococcygeal joint. The reported normal range of movement is about 13 degrees [10].

Besides, an X-ray was ordered for all cases to assess the lumbar and sacrococcygeal regions. CT sacrum and coccyx with 3D reconstruction or MRI were done in some cases to exclude inflammatory or infiltrative lesions.

Operative procedure

Patients were instructed to take a low residue diet for four days prior to surgery and an enema was done the night before surgery. Unsayn 1.5 gm i.v. was administered on call for surgery and repeated every 8 hours for 48 hours after surgery. All surgeries were done in prone position. The surgical strategy was similar to that technique described by Key [9]. The buttocks were separated and strapped laterally with adhesive plaster to open the cheeks of the buttocks for wide exposure. about 1 cm superior to the gluteal cleft, a 5cm skin incision was created at the midline. After skin incision we dissected through the layers down to the posterior surface of the coccyx. Then we bluntly dissected to expose the coccygeal tip. Then

we exposed and cut the anococcygeal ligament and then we elevated the coccygeal tip. Then we dissected and cut the attachment of the coccygeus and iliococcygeus muscle from the coccyx while taking great care not to injure the rectum. Then the coccyx was totally removed.

After meticulous hemostasis, we removed the straps retracting the buttock cheeks. The wound was closed in layers taking care to minimize the dead space as much as possible. Suction drains were left for at least forty-eight hours postoperatively.

Post-operative care

Early ambulation was encouraged, and post-operative pain was managed by paracetamol or NSAIDs. All cases were discharged on the 1st or 2nd post-operative day. CT and X rays were done postoperatively to ensure total excision of the coccyx.

Follow up

Regular follow up visits were arranged for our cases for one year after surgery (1 week after surgery, then after 1 month, 3 months, 6 months, and finally 12 months). Their current level of pain was assessed via VAS score. Moreover, cases were asked to evaluate their general pan symptoms as; complete relief, significant improvement, moderate relief, unchanged, or worsened. Surgical outcome was also measured according to table (1).

Outcome	Criteria
Excellent	Complete absence or significant pain improvement and VAS ≤ 2/10.
Good	Significant pain improvement and VAS ≤ 3/10.
Satisfactory	Moderate pain improvement with VAS ≤ 6/10.
Poor	Unchanged symptoms or VAS > 6/10.

Table 1. Outcome categories [1].

Statistical analysis

The collected data were coded, processed and analysed using the SPSS (Statistical Package for Social Sciences) version 22 for Windows® (IBM, SPSS Inc, Chicago, IL, USA).

Normally distributed quantitative data (VAS score) were expressed as mean (range) and

comparison between preoperative and post-operative values was compared using paired samples t-test. P value (< 0.05) was considered significant.

RESULTS

The mean age of the included cases was 43.87 years (range, 39 – 52). We included 5 females (62.5%) and 3 males (37.5%). The mean pre-operative VAS score was 9 (range, 8 – 10), while post-operatively, it decreased significantly down to 2 (range, 1 – 3) (p < 0.001). Excellent post-operative outcomes were achieved in 6 cases (75%) whereas good outcomes were obtained in the remaining 2 cases (25%). In this study, there were two cases of wound infection. One of them had superficial infection which improved on antibiotics and frequent dressing. The other case had deep wound infection and sinus discharging pus and she underwent surgical debridement and repair by plastic surgeon. These data are illustrated at table (2).

Case No.	Age	Gender	Preoperative VAS	Post-operative VAS	Outcome
1	43	Female	8	2	Excellent
2	48	Female	9	1	Excellent
3	52	Female	9	2	Excellent
4	40	Female	10	3	Good
5	44	Female	8	2	Excellent
6	39	Male	10	2	Excellent
7	42	Male	9	3	Good
8	43	Male	9	1	Excellent

Table 2. Overview of the included cases.



Figure 1. Sagittal CT Sacrum and Coccyx in patient with post traumatic anterior angulation of the coccyx and Coccydynia.

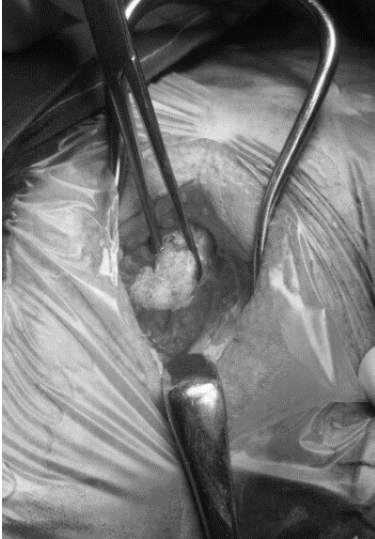


Figure 2.
Intraoperative exposure of the coccyx during Coccygectomy.

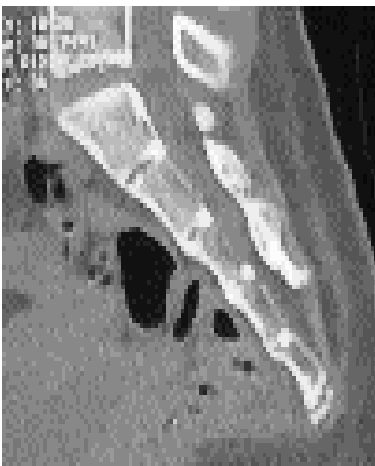


Figure 3.
Sagittal CT Sacrum and Coccyx after coccygectomy.

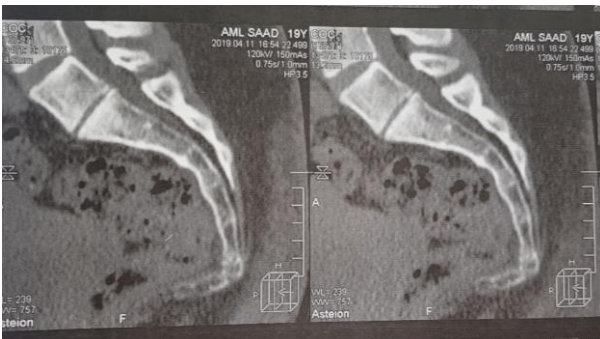


Figure 4. Preoperative Sagittal CT Sacrum and Coccyx in patient with showing anterior angulation of the coccyx.

DISCUSSION

Coccydynia seems to be emerging as a major medical condition in the current millennium due to the increasing number of populations that spend a long time setting in front of computers. Many patients complain of severe pain during sexual

intercourse or defecation. Pain and tenderness over the coccyx is usually diagnostic for this condition [7].



Figure 5. The excised coccyx.

First line of conservative measures includes NSAIDs, sitting aids, hot fomentations, and physiotherapy. These conservative measures are effective in about 60% to 66% of patients. When these measures fail, fluoroscopic or US guided local steroid administration and coccygeal manipulation showed success rates ranging between 75% and 85%. So surgical excision of the Coccyx is still needed to treat the resistant cases not responding to previous measures [16].

This study was conducted at Mansoura University Hospitals aiming to evaluate the role of coccygectomy in the management of coccydynia. We included a total of 8 cases with a mean age of 43.87 years (range, 39 – 52).

Another study handling the same perspective included 31 cases with coccydynia with a mean age of 41.5 years (range, 20 – 65) [4].

In the current study, we included 5 females (62.5%) and 3 males (37.5%). other studies reported that this disease has a higher prevalence in women [6].

On the other hand, another study conducted by Antoniadis and his colleagues included 6 males (60%) and 4 females (40%) [1]. This contradicts with our study results.

Regarding outcomes in the current study, the mean pre-operative VAS score was 9 (range, 8 – 10),

while post-operatively, it decreased significantly down to 2 (range, 1 – 3). Excellent post-operative outcomes were achieved in 6 cases (75%) whereas good outcomes were obtained in the remaining 2 cases (25%).

Another study also confirmed our findings as it reported a marked decrease in VAS score, and all cases included have achieved good or excellent outcomes (100%) [1].

This correlates with also with other studies that stated that more than 90% of cases were having a good or very good surgical outcomes after coccygectomy [11, 21, 22].

Cheng and his associates reported that excellent outcomes were achieved in 20 cases (64.5%) while good outcomes were present in 7 cases (22.6%). Additionally, 3 cases had moderate outcome (9.7%) whereas only one case reported poor outcome (3.2%) [4].

Another recent study reported that there was a significant decrease in VAS score following operation (2.25 vs. 9.62 preoperatively – $p < 0.001$). Moreover, excellent or good outcomes were achieved in 87.5% of the included cases [18].

Of note, some authors have reported that better surgical outcomes could be achieved in patients with traumatic coccydynia compared with idiopathic cases [2, 14]. However, others have found no difference between the surgical outcomes of the two types [15].

Another larger study also reported that the Short Form 36 (SF-36), the Oswestry Disability Index (ODI) and the visual analogue scale (VAS) showed a significant improvement after surgery ($p < 0.0001$). Successful outcome was obtained in 70.4% of cases while failure occurred in 25.5% of cases. The rest of cases were lost during follow up [7].

Cebesoy et al. reported no infection in 21 patients all of whom received prophylactic antibiotics for 5 days [3]. Doursounian et al. had no infection in his series of 80 patients all of whom received two prophylactic antibiotics over 48 hours and preoperative rectal enema [5].

Although coccygectomy appears to be a technically easy procedure, multiple complications have been documented in the literature. Its rate ranges between 0 and 50% [3, 20].

The most common encountered complication is wound infection, followed by healing problems

(dehiscence) [1]. Other rare complications may include rectal injury [8].

In the study conducted by Cheng and his colleagues, post-operative wound infection was encountered in 2 cases (6.45%) [4].

Other reports assume that the infection rates associated with coccygectomy have ranged from 14% to 30%, including superficial and deep wound infections and dehiscence [21]. This is comparable to our results as we had two cases of wound infection 25%.

The main drawback of this study is the small number of cases and short duration of follow up. Therefore, additional studies including more cases with longer follow up periods should be conducted.

CONCLUSION

Although conservative medical measures are effective for treating coccydynia but surgical excision is still a valid treatment option in resistant cases.

AUTHORS CONTRIBUTIONS

This work was carried out in collaboration between all authors. They designed the study and researched literature. They approved the protocol. All the surgeries were done by the same surgical team. They all shared in data collection and manuscript finalization. All authors read and approved the final manuscript.

REFERENCES

1. Antoniadis A, Ulrich N H-B, and Senyurt H: Coccygectomy as a surgical option in the treatment of chronic traumatic coccygodynia: a single-center experience and literature review. *Asian spine journal* 2014, 8(6),705.
2. Bayne O, Bateman J E, and Cameron H U: The influence of etiology on the results of coccygectomy. *Clinical orthopaedics and related research* 1984, (190),266-72.
3. Cebesoy O, Guclu B, Kose K, Basarir K, Guner D, and Us A: Coccygectomy for coccygodynia: do we really have to wait? *Injury* 2007, 38(10),1183-8.
4. Cheng S-w, Chen Q-y, Lin Z-q, Wei W, Zhang W, Kou D-q, Yue S, Ying X-z, Cheng X-j, and LÜ C-z: Coccygectomy for stubborn coccydynia. *Chinese Journal of Traumatology (English Edition)* 2011, 14(1),25-8.
5. Doursounian L, Maigne J-Y, Cherrier B, and Pacanowski J: Prevention of post-coccygectomy infection in a series of 136 coccygectomies. *International orthopaedics* 2011, 35(6),877-81.
6. Fogel G R, Cunningham III P Y, and Esses S I: Coccygodynia: evaluation and management. *JAAOS- Journal of the American Academy of Orthopaedic Surgeons* 2004, 12(1),49-54.
7. Hanley E, Ode G, Jackson Iii J, and Seymour R:

- Coccygectomy for patients with chronic coccydynia: a prospective, observational study of 98 patients. *The bone & joint journal* 2016, 98(4),526-33.
8. Karadimas E J, Trypsiannis G, and Giannoudis P V: Surgical treatment of coccygodynia: an analytic review of the literature. *European Spine Journal* 2011, 20(5),698-705.
 9. KEY J A: Operative treatment of coccygodynia. *JBJS* 1937, 19(3),759-64.
 10. Maigne J-Y, Doursounian L, and Chatellier G: Causes and mechanisms of common coccydynia: role of body mass index and coccygeal trauma. *Spine* 2000, 25(23),3072-9.
 11. Maigne J-Y, Lagauche D, and Doursounian L: Instability of the coccyx in coccydynia. *The Journal of bone and joint surgery British volume* 2000, 82(7),1038-41.
 12. Maigne J, Pigeau I, Aguer N, Doursounian L, and Chatellier G: Chronic coccydynia in adolescents. A series of 53 patients. *European journal of physical and rehabilitation medicine* 2011.
 13. Nathan S, Fisher B, and Roberts C: Coccydynia: a review of pathoanatomy, aetiology, treatment and outcome. *The Journal of bone and joint surgery British volume* 2010, 92(12),1622-7.
 14. Pennekamp P H, Kraft C N, Stütz A, Wallny T, Schmitt O, and Diedrich O: Coccygectomy for coccygodynia: does pathogenesis matter? *Journal of Trauma and Acute Care Surgery* 2005, 59(6),1414-9.
 15. Postacchini F, and Massobrio M: Idiopathic coccygodynia. Analysis of fifty-one operative cases and a radiographic study of the normal coccyx. *The Journal of bone and joint surgery American volume* 1983, 65(8),1116-24.
 16. Ramsey M L, Toohey J S, Neidre A, Stromberg L J, and Roberts D A: Coccygodynia: treatment. *Orthopedics* 2003, 26(4),403-5.
 17. Saleh I, and Reksoprodjo A Y: Coccygeal excision for treatment of coccyx instability. *Jurnal Orthopaedi dan Traumatologi Indonesia-The Journal of Indonesian Orthopaedic & Traumatology* 2018, 1(3).
 18. Sarmast A H, Kirmani A R, and Bhat A R: Coccygectomy for coccygodynia: A single center experience over 5 years. *Asian journal of neurosurgery* 2018, 13(2),277.
 19. Simpson J: Coccygodynia and diseases and deformities of the coccyx. *Med Times Gaz* 1859, 40,1-7.
 20. Traub S, Glaser J, and Manino B: Coccygectomy for the treatment of therapy-resistant coccygodynia. *Journal of surgical orthopaedic advances* 2009, 18(3),147-9.
 21. Wood K B, and Mehbod A A: Operative treatment for coccygodynia. *Clinical Spine Surgery* 2004, 17(6),511-5.
 22. Zayer M: Coccygodynia. *The ulster medical journal* 1996, 65(1),58.



Outcome analysis of upper and lower limb motor functions after anterior cervical discectomy and fusion for degenerative cervical disc disease

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ABSTRACT

Background: Anterior cervical discectomy and fusion (ACDF) is the most commonly performed surgical procedure for symptomatic cervical disc disease. In this study, we analysed the upper and lower limb motor functions after ACDF for disc prolapse in patients with degenerative cervical disc disease.

Methods: One hundred consecutive adult patients who underwent ACDF for single or two-level cervical disc prolapse during the study period (October 2015 to October 2017) were included in the study.

Results: Preoperative motor deficits in limbs were noted in 73% (73/100) of the patients. Enhance recovery of motor deficits was noted in 72.6% (53/73) of these patients and persisting motor deficits in the remaining patients (20/73- 27.4%). Five patients (5/27- 18.5%) without any preoperative motor deficits developed motor deficits after ACDF. Detailed pre and postoperative (at the time of discharge) motor power (graded by MRC grade) in all 4 limbs (Shoulder abduction / adduction / flexion / extension, elbow flexion / extension, wrist flexion / extension, hip abduction / adduction / flexion / extension, knee flexion/extension, ankle flexion/extension) was recorded. Statistically significant improvement in motor power (as recorded at the time of discharge) was noted in all the tested muscle groups after ACDF.

Conclusion: Early improvement in preoperative motor deficits can be expected in the majority of the patients with cervical PIVD following ACDF.

Keywords

anterior cervical discectomy and fusion, outcome, cervical disc degeneration



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INTRODUCTION

ACDF is most commonly done to treat a symptomatic cervical PIVD. (1, 16, 20, 22, 23, 25) ACDF is a safe procedure and is rarely associated with post-operative complications. (2, 7, 9, 12, 17) Significant proportion of patients shows remarkable recovery in motor deficits following ACDF. (11, 16) Authors in this publication analysed in detail the early recovery of motor deficits following ACDF for single or two level degenerative cervical PIVD.

MATERIAL AND METHODS

One hundred consecutive adult patients who underwent ACDF for degenerative cervical PIVD during the study period (October 2015 to October 2017) were included in the study. Patients with traumatic PIVD were excluded. Approval from the institutional ethical committee was taken for this study. Detailed pre and postoperative (At the time of discharge) motor power (graded by MRC grade except hand grip which was subjectively graded from 0-100%) in all 4 limbs (Shoulder abduction / adduction / flexion / extension, elbow flexion / extension, wrist flexion / extension, hip abduction / adduction / flexion / extension, knee flexion / extension, ankle flexion / extension) were analysed.

Statistical analysis

Data analysis was done by using statistical software SPSS Statistics Version 24.0. Descriptive statistics including mean and standard deviation for continuous variables, and frequency and percentage for categorical variables were used for data expression. Appropriate tests like Chi-square test, Wilcoxon signed rank test etc. were used for checking statistically significant correlation. A probability (P) value of <0.05 was considered significant.

RESULTS

Preoperative motor deficits in limbs were noted in 73% (73/100) of the patients. At the time of discharge, enhance recovery of motor deficits was noted in 72.6% (53/73) of these patients and persisting motor deficits in the remaining patients (20/73- 27.4%). Five patients (5/27- 18.5%) without any preoperative motor deficits developed motor deficits after ACDF. Pre and postoperative (At the time of discharge) motor power in all 4 limbs is compared in Tables 1-4. Statistically significant improvement in motor power at the time of discharge was recorded in all the tested muscle groups after ACDF.

Right upper limb	Preoperative (n=100)				Postoperative (n=100)				P-value	
	Mean [#]	Median [#]	Range [#]	IQR	Mean [#]	Median [#]	Range [#]	IQR		
Shoulder										
Shoulder abduction	3.80±1.4	4	0-5	2	4.15±1.29	5	0-5	1	0.001 (S)*	
Shoulder adduction	3.80±1.4	4	0-5	2	4.16±1.29	5	0-5	1	0.001 (S)*	
Shoulder flexion	3.81±1.4	4	0-5	2	4.18±1.29	5	0-5	1	0.001 (S)*	
Shoulder extension	3.80±1.4	4	0-5	2	4.17±1.32	5	0-5	1	0.001 (S)*	

Elbow										
Elbow flexion	3.73±1.5	4	0-5	1		4.17±1.32	5	0-5	1	0.001 (S)*
Elbow extension	3.71±1.5	4	0-5	2		4.16±1.33	5	0-5	1	0.001 (S)*
Wrist										
Wrist flexion	3.71±1.6	4	0-5	1		4.14±1.27	5	0-5	1	0.001 (S)*
Wrist extension	3.72±1.6	4	0-5	2		4.14±1.29	5	0-5	1	0.001 (S)*
Right hand grip	Preoperative (Number of patients)				Postoperative (Number of patients)				P-value	
0-25%	11				4				0.001 (S)†	
25-50%	11				13					
50-75%	29				22					
75-100%	49				61					

Table 1. Comparison of preoperative and postoperative motor power in right upper limb.

#Power graded according to MRC grade.

*Obtained Using Wilcoxon signed rank test; † Obtained using Chi square test; S: Significant.

Left upper limb	Preoperative (n=100)				Postoperative (n=100)				P-value*	
	Mean#	Median#	Range#	IQR	Mean#	Median#	Range#	IQR		
Shoulder										
Shoulder abduction	3.75±1.41	4	0-5	2	4.12±1.36	5	0-5	1	0.001 (S)*	
Shoulder adduction	3.74±1.41	4	0-5	2	4.14±1.34	5	0-5	1	0.001 (S)*	
Shoulder flexion	3.72±1.42	4	0-5	2	4.13±1.34	5	0-5	1	0.001 (S)*	
Shoulder extension	3.72±1.45	4	0-5	2	4.13±1.32	5	0-5	1	0.001 (S)*	
Elbow										

Knee flexion	3.40±1.86	4	0-5	2	3.90±1.60	5	0-5	2	0.001(S)
Knee extension	3.38±1.86	4	0-5	2	3.90±1.59	5	0-5	2	0.001(S)
Ankle									
Ankle flexion	3.33±1.85	4	0-5	2	3.84±1.58	4.5	0-5	2	0.001(S)
Ankle extension	3.35±1.86	4	0-5	2	3.85±1.58	5	0-5	2	0.001(S)

Table 3. Comparison of preoperative and postoperative motor power in right lower limb.

#Power graded according to MRC grade

*Obtained Using Wilcoxon signed rank test; S: Significant

Left lower limb	Preoperative (n=100)				Postoperative (n=100)				P-value*
	Mean	Median	Range	IQR	Mean	Median	Range	IQR	
Hip									
Hip abduction	3.32±1.86	4	0-5	2	3.98±1.53	5	0-5	1	0.001 (S)
Hip adduction	3.33±1.87	4	0-5	2	3.99±1.52	5	0-5	1	0.001 (S)
Hip flexion	3.32±1.86	4	0-5	2	4.00±1.49	5	0-5	1	0.001 (S)
Hip extension	3.29±1.84	4	0-5	2	3.98±1.49	5	0-5	1	0.001 (S)
Knee									
Knee flexion	3.27±1.83	4	0-5	2	3.95±1.52	5	0-5	1	0.001 (S)
Knee extension	3.27±1.84	4	0-5	2	3.94±1.51	5	0-5	1	0.001 (S)
Ankle									
Ankle flexion	3.22±1.89	4	0-5	3	3.88±1.55	5	0-5	2	0.001 (S)
Ankle extension	3.20±1.89	4	0-5	3	3.89±1.56	5	0-5	2	0.001 (S)

Table 4. Comparison of preoperative and postoperative motor power in right lower limb.

#Power graded according to MRC grade

*Obtained Using Wilcoxon signed rank test; S: Significant

DISCUSSION

Cervical PIVD is a common degenerative disc disease affecting millions of people. (24) Cervical disc herniation can occur as a result of ageing, wear and tear, or sudden stress from an accident.(5) Majority of these patients present with neck pain radiating to upper limbs. (24) Other presenting symptoms include motor deficits, stiffness in limbs, sensory deficits, paresthasias in limbs etc. (16, 24) Majority of the patients presenting with only neck pain or radicular pain can be managed with medication as

and conservative measures like physiotherapy, cervical collar etc. Patients with significant pain not responding to conservative measures and patients with neurological deficits like sensory/motor deficits and bladder symptoms respond well to surgery. (10, 16, 21)

ACDF is a common surgical procedure performed for symptomatic degenerative cervical disc disease. (6, 16) It helps to relieve the pressure on nerve roots and/or on the spinal cord, (14) thus resulting in improvement in various clinical symptoms including

neck pain, radicular pain, motor weakness, sensory symptoms, tightness in limbs and bladder disturbances. (10, 13, 16) Various complications reported with ACDF include dysphagia, hoarseness of voice, wound hematoma, graft migration, pseudoarthrosis, wound infection etc. (13) Patients can rarely have sensory or motor deficits after ACDF due to small risk of damage to the spinal cord, nerve roots or both. (10, 13, 16)

Improvement in neurological deficits ranging from 36-93% has been reported in various series after ACDF. (3, 13, 15, 16, 18) Lehman et al (16) reported preoperative motor deficits in 55% of the patients and reported recovery of these deficits in 95% of them at 1 year. Chiles et al (4) reported strength improvement rates ranging from 79.1% to 90.9% in various individual muscle groups of upper and lower limbs following ACDF. In the present study very high proportion of patients (73%) presented with motor deficits and early complete recovery of these deficits were noted in 72.6% (53/73) of these patients.

Majority of the studies on ACDF have graded neurological deficits using various scores like Nurick's grade, JOA, modified JOA scores etc. which combine both sensory and motor deficits. (4, 8, 16, 19) Detailed assessment of motor deficits with grading of motor power for various muscle groups has not been done in most of the studies on ACDF. (8, 16) In the present study we compared the preoperative motor power and early postoperative (at the time of discharge) motor power following ACDF in all major groups of muscles of lower and upper extremity and found that significant improvement in motor power in early postoperative period. Long term follow-up studies in patients following ACDF is required as they can develop new deficits secondary to adjacent segment disease. (16)

CONCLUSION

Early improvement in preoperative motor deficits can be expected in majority of the patients with degenerative cervical PIVD following ACDF.

REFERENCES

1. Bhaganagare AS, Nagesh SA, Shrihari BG, Naik V, Nagarjun MN, Pai BS: Management of cervical monoradiculopathy due to prolapsed intervertebral disc, an institutional experience. *Journal of craniovertebral junction & spine* 8:132, 2017.
2. Boakye M, Mummaneni PV, Garrett M, Rodts G, Haid R: Anterior cervical discectomy and fusion involving a polyetheretherketone spacer and bone morphogenetic protein. *Journal of Neurosurgery: Spine* 2:521-525, 2005.
3. Bohlman HH, Emery SE, Goodfellow DB, Jones PK: Robinson anterior cervical discectomy and arthrodesis for cervical radiculopathy. Long-term follow-up of one hundred and twenty-two patients. *JBJS* 75:1298-1307, 1993.
4. Chiles III BW, Leonard MA, Choudhri HF, Cooper PR: Cervical spondylotic myelopathy: patterns of neurological deficit and recovery after anterior cervical decompression. *Neurosurgery* 44:762-769, 1999.
5. Chung JY, Park J-B, Seo H-Y, Kim SK: Adjacent Segment Pathology after Anterior Cervical Fusion. *Asian Spine J* 10:582-592, 2016.
6. Dardis RM, Saxena A, Shad A, Chitnavis B, Gullan R (2012) *Disc Replacement Technologies in the Cervical and Lumbar Spine*. Elsevier, pp 1777-1788.
7. Davis RA: A long-term outcome study of 170 surgically treated patients with compressive cervical radiculopathy. *Surgical neurology* 46:523-533, 1996.
8. Emery SE, Bohlman HH, Bolesta MJ, Jones PK: Anterior cervical decompression and arthrodesis for the treatment of cervical spondylotic myelopathy. Two to seventeen-year follow-up. *JBJS* 80:941-951, 1998.
9. Fountas KN, Kapsalaki EZ, Nikolakakos LG, Smisson HF, Johnston KW, Grigorian AA, Lee GP, Robinson Jr JS: Anterior cervical discectomy and fusion associated complications. *Spine* 32:2310-2317, 2007.
10. Gore DR, Sepic SB: Anterior Discectomy and Fusion for Painful Cervical Disc Disease: A Report of 50 Patients With an Average Follow-up of 21 Years. *Spine* 23:2047-2051, 1998.
11. Hessler C, Boysen K, Westphal M, Regelsberger J: Functional and radiological outcome after ACDF in 67 cases. *Zeitschrift fur Orthopadie und Unfallchirurgie* 149:683-687, 2011.
12. Jacobs WC, Anderson PG, Limbeek J, Willems PC, Pavlov P: Single or double-level anterior interbody fusion techniques for cervical degenerative disc disease. *Cochrane Database Syst Rev* 4:CD, 2004.
13. Jagannathan J, Shaffrey CI, Oskouian RJ, Dumont AS, Herrold C, Sansur CA, Jane Sr JA: Radiographic and clinical outcomes following single-level anterior cervical discectomy and allograft fusion without plate placement or cervical collar. 2008.
14. Kim W-K (2011) *Role of Minimally Invasive Cervical Spine Surgery in the Aging Spine*. Elsevier, pp 198-203.
15. Lebl DR, Hughes A, Cammisa FP, O'Leary PF: Cervical spondylotic myelopathy: pathophysiology, clinical presentation, and treatment. *HSS journal* 7:170-178, 2011.
16. Lehmann CL, Buchowski JM, Stoker GE, Riew KD: Neurologic Recovery after Anterior Cervical Discectomy and Fusion. *Global Spine J* 4:41-46, 2014.
17. Levinthal R: Anterior cervical discectomy with and

- without fusion. Results, complications, and long-term follow-up. *Spine* 19:2343-2347, 1994.
18. Murrey D, Janssen M, Delamarter R, Goldstein J, Zigler J, Tay B, Darden B: Results of the prospective, randomized, controlled multicenter Food and Drug Administration investigational device exemption study of the ProDisc-C total disc replacement versus anterior discectomy and fusion for the treatment of 1-level symptomatic cervical disc disease. *The Spine Journal* 9:275-286, 2009.
 19. Nurjck S: The pathogenesis of the spinal cord disorder associated with cervical spondylosis. *Brain* 95:87-100, 1972.
 20. Orr RD, Zdeblick TA: Cervical Spondylotic Myelopathy: Approaches to Surgical Treatment. *Clinical orthopaedics and related research* 359:58-66, 1999.
 21. Saldua NS, Okafor C, Harris EB, Vaccaro AR (2011) The Role of Spinal Fusion and the Aging Spine: Stenosis without Deformity. Elsevier, pp 329-335.
 22. Truumees E, Herkowitz HN: Cervical spondylotic myelopathy and radiculopathy. *Instr Course Lect* 49:339-360, 2000.
 23. Wu T-k, Wang B-y, Deng M-d, Hong Y, Rong X, Chen H, Meng Y, Liu H: A comparison of anterior cervical discectomy and fusion combined with cervical disc arthroplasty and cervical disc arthroplasty for the treatment of skip-level cervical degenerative disc disease: A retrospective study. *Medicine* 96:2017.
 24. Yolas C, Ozdemir NG, Okay HO, Kanat A, Senol M, Atci IB, Yilmaz H, Coban MK, Yuksel MO, Kahraman U: Cervical disc hernia operations through posterior laminoforaminotomy. *Journal of Craniovertebral Junction and Spine* 7:91, 2016.
 25. Yu J, Ha Y, Shin JJ, Oh JK, Lee CK, Kim KN, Yoon DH: Influence of plate fixation on cervical height and alignment after one-or two-level anterior cervical discectomy and fusion. *British journal of neurosurgery*:1-8, 2017.



Prognostic factors of ruptured middle cerebral artery aneurysms treated with surgical clipping

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ABSTRACT

Background: Spontaneous subarachnoid haemorrhage is an emergent condition that leads to profound morbidity and mortality. It is mainly caused by a ruptured intracranial aneurysm. Herein we described the relationship of different factors and their impact on the outcome of patients who underwent a surgical clipping exclusively to the ruptured middle cerebral artery aneurysm.

Methods: We retrospectively analyzed the medical records of patients admitted to the Neurosurgery Teaching Hospital (NTH) in Baghdad for the period between January 2017-April 2019 of patients who underwent microsurgical clipping for ruptured middle cerebral artery aneurysm. Demographic, clinical, radiological and surgical data were extracted. A univariate analysis was used to illustrate the relationship between the variables and outcome which was assessed using the five scores of the Glasgow Outcome Scale where patients were dichotomized into two groups; favourable (IV + V) and unfavourable (I, II, III).

Results: Within this cohort, a total of 50 patients were studied and the analysis revealed that 92% (N=46) had a favourable outcome and 8% (N=4) had an unfavourable outcome at discharge. The in-hospital mortality was 6%(N=3). Factors that were significantly associated with poor outcome were giant aneurysms, the presence of other unruptured aneurysms, post-operative clinical vasospasm, presence of contralateral weakness, lower pre/post-operative Glasgow coma scores, higher Hunt and Hess, World Federation of Neurosurgical Societies (WFNS) and modified-WFNS grades.

Conclusion: The factors with a significant impact on the outcome of patients with surgically clipped ruptured middle cerebral artery aneurysms were GCS, WFNS, m-WFNS, H&H, contralateral muscle weakness, size of the aneurysm, presence of other unruptured aneurysms and clinical vasospasm.

INTRODUCTION

Subarachnoid hemorrhage (SAH) constitutes about 5% of all strokes. More than 80% of all Spontaneous SAH is due to ruptured intracranial aneurysms (7,13). CT scan is the best initial test for detecting SAH, CT

Keywords

intracranial aneurysm,
subarachnoid haemorrhage,
surgical clipping,
middle cerebral artery
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angiography (CTA) is often used to characterize the aneurysm. However, the catheter angiography is the best diagnostic modality to define the aneurysm characteristics preoperatively whenever possible (10,16).

Middle cerebral artery (MCA) aneurysms account for 20% of all intracranial aneurysms (26). Up to 81% of MCA aneurysms are located at the bifurcation. In addition, 30% present with symptoms related to mass effect (14). The MCA aneurysm is the most common aneurysm that co-present with intracerebral hematoma (ICH) (30-50%) due to its anatomic and hemodynamic features (21). Several grading scales have been used for ruptured SAH including Hunt and Hess (H&H), World federation of neurosurgical societies (WFNS) grading and modified WFNS which were reported to be valuable outcome prediction tools (1,2).

Complications of ruptured aneurysms can be categorized as early or late. Such complications include rebleeding, symptomatic vasospasm and hydrocephalus at the rates of 30%, 30% and 20%, respectively (4). Rebleeding is the most serious acute complication and is, at the same time, the most preventable (8). The rate of rebleeding has been declining significantly in association with the establishment of prompt management of SAH by early surgical clipping or endovascular coiling (16).

MCA aneurysm is the most feasible aneurysm for surgical clipping, owing to its accessible location as well as its morphology with a wide neck. Additionally, the MCA aneurysm is unfavorable to be treated with endovascular coiling due to its distal location that renders the difficult (3,9,15,23). In this study, we present a case series which is the first to be reported from Iraq regarding the surgical clipping of ruptured MCA aneurysms.

PATIENTS AND METHODS

In this retrospective cohort study, data were recruited from 50 patients with SAH attributed to a ruptured MCA aneurysm. All cases were treated with surgical clipping within 72 hours of aneurysm rupture at Neurosurgery Teaching Hospital in Baghdad/Iraq between January 2017 and April 2019. The inclusion criteria were as follows: age >18 years, confirmed diagnosis of aneurysmal SAH by CT-scan and CTA, ruptured MCA aneurysms and surgically treated groups. Cases of traumatic, idiopathic SAHs, unruptured aneurysms and non-MCA aneurysms

were excluded from this series. The analyzed parameters included: Age, sex, co-morbidities, reported sudden severe headache as part of the index presentation, pre or post-operative contralateral weakness (MRC grade), seizure, vomiting, neck stiffness, similar previous attacks, clinical vasospasm, pre or post-operative GCS (GCS categorized into; mild ≥ 13 , moderate 12-9 and severe ≤ 8), location and size of the MCA aneurysms, the presence of other unruptured aneurysms, ICH, aneurysmal remnant of post-operative CTA, size of the aneurysm (Giant >25mm, Large 11-25mm, Medium 5-10mm and Small <5mm), the need for temporary clipping, intraoperative rupture, m-WFNS scale, WFNS scale, H&H scale, Glasgow Outcome Scale at the time of discharge and at six-month follow-up period. At six months follow-up; two cases were lost to follow up in which they were excluded from the analysis.

Fisher exact test was used to evaluate the relationship between the aforementioned factors and GOS at both discharge and six-month intervals. Patients were divided into 2 categorical groups based on the 5 scores of the GOS into favorable (good recovery V+ moderate disability IV) and unfavorable (severe disability III + vegetative state II+ dead I) prognostic groups. The paired data of GOS at discharge and follow-up were also tested for any statistical difference using a Wilcoxon signed-rank test. The correlation between SAH grading scales and GOS was assessed using Kendall's tau correlation coefficient. The level of significance decided on p-Value. All statistical analysis was done using commercially available software (SPSS, Version 23.0).

RESULTS

The study sample included 50 patients, 68% (N=34) of whom were females with a female: male ratio of 2:1. The mean age was $40.32 \pm SD$, with 80% (N= 30) of the females being over 40 years of age and 93.8% (N=15) of the males being younger than 40 years of age. Results for patients parameters corresponding to the outcome are shown in tables 1,2 and 3. Overall, 92% (N=46) of the patients had favorable outcome at discharge (good recovery in 82% (N=41) and moderate disability in 10% (N=5)) while 8% (N=4) had unfavorable outcome including 2% (N=1) discharged with severe disability and 6% (N=3) pronounced dead. The mortality was attributed to

ischemic stroke in two patients and pulmonary embolism in one patient.

The Factors that were associated with unfavorable outcome (GOS scores of I to III) included giant aneurysms, the presence of other unruptured aneurysms, the need for temporary clipping during surgery, the presence of postoperative clinical vasospasm, the presence of pre/post-operative contralateral muscle weakness (Decreased MRC grading), lower GCS scores and higher H&H, WFNS and m-WFNS scores. In analyzing the correlation between these scales and GOS, m-WFNS had the best negative rank-order correlation coefficient with respect to higher GOS and the overall order was as: m-WFNS then WFNS then H&H. H&H showed the least negative correlation with respect to higher GOS (Table 4).

The remaining parameters were not considered as independent risk factors that influence the outcome because they showed no statistical

significance. Higher Pre-operative MRC scores were significantly associated with the absence of hematoma ($P = 0.035$). A higher post-operative MRC grade was notably associated with the absence of post-operative clinical vasospasm ($P < 0.001$). 64% ($N=32$) of patients presented with an ICH, 12.5% ($N=4$) of whom had unfavorable outcomes, compared to the patients without ICH where none of them had a poor outcome. The factors that showed statistical significance at discharge have also proven significant at six-month follow-up with the exception of temporary clipping which was not significant at the 6-month follow-up. Temporary clipping which was an infrequent adjunct applied only in 22% ($N= 11$) of all cases for a period of fewer than 10 minutes. 45.5% of patients in our study with temporary clipping developed vasospasm ($p = 0.017$). A Wilcoxon signed-rank test showed that the GOS at discharge differed significantly from the GOS at 6 months follow-up ($Z = -2.33, P = .031$).

Table 1 Patient's Characteristics

		Favorable outcome (%)	Unfavorable outcome (%)	Total No. of cases	P value at discharge	P value at follow-up
Age	≤40	18 (36%)	1 (2%)	19 (38%)	1.000	1.000
	>40	28 (56%)	3 (6%)	31 (62%)		
Sex	Male	14 (28%)	2 (4%)	16 (32%)	0.584	0.254
	Female	32 (64%)	(4%)2	34 (86%)		
Hypertension	Yes	15 (30%)	3 (6%)	16 (32%)	0.127	0.547
	No	31 (62%)	1 (2%)	32 (64%)		
Diabetes Mellitus	Yes	10 (20%)	2 (4%)	14 (24%)	0.240	0.150
	No	36 (72%)	2 (4%)	38 (76%)		
Sudden severe headache	Yes	44 (88%)	3 (6%)	47 (94%)	0.226	0.180
	No	2 (4%)	1 (2%)	3 (6%)		
Vomiting	Yes	43 (86%)	3 (6%)	46 (92%)	0.291	1.000
	No	3 (6%)	1 (2%)	4 (8%)		

Table 1 Patient's Charecteristics

Neck stiffness	Yes	44 (88%)	3 (6%)	47 (94%)	0.226	0.180
	No	2 (4%)	1 (2%)	3 (6%)		
History of previous attack	Yes	2 (4%)	1 (2%)	3 (6%)	0.226	0.180
	No	44 (88%)	3 (6%)	47 (94%)		
Pre-op seizure	Yes	8 (16%)	1 (2%)	9 (18%)	0.560	0.472
	No	38 (76%)	3 (6%)	41 (82%)		
Post-op seizure	Yes	4 (8%)	1 (2%)	5 (10%)	0.353	0.286
	No	42 (84%)	3 (6%)	45 (90%)		
Post-op clinical vasospasm	Yes	5 (10%)	4 (8%)	9 (18%)	0.001*	0.005*
	No	41 (82%)	-	41 (82%)		
Pre-op MRC grading	0	-	1 (2%)	1 (2%)	0.002*	0.004*
	1	3 (6%)	2 (4%)	5 (10%)		
	2	4 (8%)	1 (2%)	5 (10%)		
	3	1 (2%)	-	1 (2%)		
	Normal	38 (76%)	-	38 (76%)		
Post-op MRC grading	0	-	2 (4%)	2 (4%)	< 0.001*	< 0.001*
	1	-	1 (2%)	1 (2%)		
	3	2 (4%)	-	2 (4%)		
	Normal	44 (88%)	1 (2%)	45 (90%)		
Initial GCS	Mild	42 (84%)	1 (2%)	43 (86%)	< 0.001*	< 0.001*
	Moderate	4 (8%)	-	4 (8%)		
	Severe	-	3 (6%)	3 (6%)		
Post-op GCS	Mild	46 (92%)	1 (2%)	47 (94%)	< 0.001*	< 0.001*
	Severe	-	3 (6%)	3 (6%)		

MRC,medical research council ; GCS,glasgow coma scale ; pre-/post-op, pre-/post-operative

Significance was determined according to chi-square test.
* Statstically significant

Table 2 Radiological & surgical Data						
		Favorable outcome (%)	Unfavorable outcome (%)	Total No. of cases	P value at discharge	P value at follow-up
Aneurysm location	Right MCA bifurcation	27 (54%)	4 (8%)	31 (62%)	0.445	0.624
	Left MCA bifurcation	16 (32%)	-	16 (32%)		
	Right M2	1 (2%)	-	1 (2%)		
	Right M2 bifurcation	1 (2%)	-	1 (2%)		
	Left M2	1 (2%)	-	1(2%)		
Aneurysm size	Small	8 (16%)	1 (2%)	9 (18%)	0.004*	0.003*
	Medium	30 (60%)	-	30 (60%)		
	Large	7 (14%)	1 (2%)	8 (16%)		
	Giant	1 (2%)	2 (4%)	3 (6%)		
Other unruptured aneurysm	Yes	-	2 (4%)	2 (4%)	0.005*	0.003*
	No	46 (92%)	2 (4%)	48 (96%)		
CT-ICH	Yes	28 (56%)	4 (8%)	32 (64%)	0.283	0.543
	No	18 (36%)	-	18 (36%)		
ICH in dominant hemisphere	Yes	12 (37.5%)	2 (6.3%)	14 (43.8%)	1000	576
	No	16 (50%)	2 (6.3%)	18 (56.3%)		
Temporary clipping	Yes	8 (16%)	3 (6%)	11 (22%)	0.029*	0.127
	No	38 (76%)	1 (2%)	39 (78%)		
Intra-op rupture	Yes	24 (48%)	3 (6%)	27 (54%)	0.614	1.000
	No	22 (44%)	1 (2%)	23 (46%)		
Remnant of Post-op CTA	Yes	1 (2%)	1 (2%)	2 (4%)	0.155	1.000
	No	45 (90%)	3 (6%)	48 (96%)		

Table 2 Radiological & surgical Data					
	Favorable outcome (%)	Unfavorable outcome (%)	Total No. of cases	P value at discharge	P value at follow-up
CT, computed tomography; ICH, intracerebral hematoma; CTA; computed tomography angiography					
Significance was determined according to chi-square test.					
Statistically significant					

Table 3 Neurological grading scales						
		Favorable outcome (%)	Unfavorable outcome (%)	Total No. of cases	P value at discharge	P value at follow-up
Hunt&Hess	Level II	38 (76%)	-	38 (76%)	0.002**	0.013*
	Level IV	8 (16%)	4 (8%)	12 (24%)		
WFNS	I	37 (74%)	-	37 (74%)	0.002*	0.006*
	II	1 (2%)	-	1 (2%)		
	III	4 (8%)	1 (2%)	5 (10%)		
	IV	4 (8%)	3 (6%)	7 (14%)		
Modified -WFNS	I	37 (74%)	-	37 (74%)	0.002*	0.003*
	II	5 (10%)	1 (2%)	6 (12%)		
	IV	4 (8%)	3 (6%)	7 (14%)		
WFNS; world federation of neurosurgical societies						
Significance was determined according to chi-square test.						
* Statistically significant						

Table 4 Rank-order correlation coefficients of different grading systems on higher GOS

Rank order	Correlation coefficient	P value
modified-WFNS	- 0.56	< 0.001
WFNS	- 0.55	< 0.001
Hunt&Hess	- 0.54	< 0.001
WFNS; world federation of neurosurgical societies,		

DISCUSSION

Aneurysmal SAH is one of the most serious and often deadly phenomena (12). MCA aneurysm is one of the commonest causes of aneurysmal SAH (6). It is commonly located at the bifurcation and constitutes 81% of all unruptured MCA aneurysm and 87% of the ruptured ones (24).

Patient's characteristics: Our data showed a female predominance with a female: male ratio of 2:1. Although there appears to be a tendency towards a more favorable outcome in both females and younger patients, this failed to achieve statistical significance in our study; a finding that was also reported by similar studies (11,17,24). The presence of co-morbidities is often cited as a significant risk factor for aneurysm rupture, but their prognostic significance is still uncertain (14,25). In our data, concomitant diseases, history of previous attacks and seizures were not found to be significantly associated with an unfavorable outcome.

Vasospasm is classified as one of the major complications associated with MCA aneurysm clipping (26). In this study, we exclusively evaluated post-operative clinical vasospasm which was defined as a deterioration in the neurologic status (speech or motor) along with the presence of new ischemic CT changes or CT-angiography-defined vessels spasm. Vasospasm was only evident in 18%(N= 9) of patients and was noted to be associated with worse outcomes. Motor weakness is also a known neurological complication that follows aneurysmal SAH with an incidence of 14-29% (12). Hereby, we evaluated the presence of contralateral limb weakness both pre and post-operatively, using the MRC grading score. All patients showed a more

marked weakness in the upper limbs compared to the lower limbs which reflects the anatomical territory supplied by the MCA. Patients with Pre or post-operative MRC grading scores for muscle power of (0,1) showed a significant tendency toward a poorer outcome and those with no weakness showed a significant tendency toward a better prognosis. Additionally, higher pre-operative MRC scores were associated with the absence of hematoma whereas post-operative MRC was associated with postoperative clinical vasospasm. These findings were consistent with other studies reporting vasospasm and cerebral ischemia as the most common mechanisms that may lead to motor weakness in aneurysmal SAH (12).

Cerebral vasospasm is directly linked to cerebral ischemia and patients may present with stroke-like symptoms including motor weakness, hence low MRC scores are not uncommon in patients with cerebral vasospasm. The presence of lower (severe) pre or post-operative GCS scores was found to be significantly associated with a lower GOS in contrast to the higher (mild) GCS scores which were significantly associated with higher GOS.

Radiological and aneurysmal characteristics: Aneurysm characteristics such as aneurysm location, size and the presence of other unruptured aneurysms are important factors that impact the prognosis of ruptured cranial aneurysms (14,25). Most reports inversely correlate aneurysmal size with the outcome (11). Our data showed that giant aneurysms were associated with unfavorable outcome whereas medium-sized aneurysms were associated with a more favorable outcome. Notably, Ruggeri et al reported that the diameter of the

aneurysm didn't significantly affect the outcome which is in contrast to what most reports (19). Our study took the side of the aneurysm (right vs left) into account. 62% of the aneurysms were located on the right MCA bifurcation and were noted to be associated with unfavorable outcomes; this observation did not, however, achieve statistical significance. This can be attributed to the fact that most of the cases in our sample are right-handed, with a left dominant hemisphere. However, Brawanski et al reported that the side of the aneurysm did not affect the outcome directly, but rather determined the side of infarction (5). In addition to the aneurysm size and location, the presence of other unruptured aneurysms was found to significantly associated with an unfavorable outcome ($P = 0.005$). This finding is in contrast to the study performed by Rodriguez-Hernandez et al who stated that the number of aneurysms did not impact the outcome; although this series did not take into account the status of the aneurysm whether it was ruptured or not (25). MCA aneurysm is known to be the most common intracranial aneurysm that is associated with an ICH (24). In a paper by Shimoda et al which studied 47 patients presented with ruptured MCA aneurysm associated with a hematoma, it was found that 42% ($N=20$) had an ICH in the dominant hemisphere that led to a poor outcome but was reported to be non-significant similarly to the findings of this cohort (22).

Surgical parameters: Intra-operative rupture was reported as a factor that doesn't impact the surgical outcome (20). This finding is consistent with our results where intra-operative rupture showed a non-significant trend toward a poorer outcome. The application of temporary clipping was significantly associated with lower GOS at discharge but not at follow-up. This can be attributed to the fact that temporary clipping was noted to increase the risk of postoperative clinical vasospasm significantly where 45.5% of patients in our study with temporary clipping developed vasospasm ($p = 0.017$). Therefore, temporary clipping was found to be a dependent factor regarding follow-up. We observed that the absence of post-operative CTA remnants led to a favorable outcome but this was not statistically significant.

Grading scales: WFNS, mWFNS, and H&H are commonly used neurological scales that aid in surgical decision and predict the outcome of patients

with aneurysmal SAH (1). H&H scale showed the least negative correlation in respect to higher GOS since it relies only on general subjective terms that depend on the judgment and interpretation, leading to a blurring of the lines between grades which in return would increase the inter-rater reliability (2,18). Therefore, this scale is more useful in the selection of surgical candidates than outcome prediction. WFNS scale is superior to H&H as it depends on both GCS and the absence/presence of neurologic deficits. When both WFNS and m-WFNS were compared in terms of correlation with GOS, m-WFNS correlated slightly stronger. Regarding m-WFNS, none of the cases in our study scored grade III, 12% ($N= 6$) scored grade II and the rest scored grade I and IV. Hence grades II and III are the only different grades between the 2 scales, explaining the slight preference of m-WFNS over WFNS (Table 4).

Limitations: First, this is a retrospective cohort from a single neurosurgical center with a small sample size that only included univariate analysis, making the conclusions non-generalizable. Second, the absence of intravascular coiling facilities made surgical clipping the only available treatment option in Iraq, limiting our ability in comparing the results to those of intravascular coiling.

CONCLUSION

Factors that may predict poorer outcomes in ruptured MCA aneurysm Clipping include: Higher mWFNS, WFNS, and H&H scores, lower GCS scores, the presence of clinical vasospasm, the presence of hemiparesis, the presence of other unruptured aneurysms and aneurysm size (giant aneurysms). m-WFNS was found to be slightly superior to the other neurologic scales in predicting the outcome.

CONFLICTS OF INTEREST

The authors declare no conflict of interests

ABBREVIATIONS

CT: Computerized Tomography

MCA: middle cerebral artery

SAH: subarachnoid haemorrhage

H and H: Hunt and Hess scale

WFNS: World Federation of Neurosurgical Societies scale

REFERENCES

1. Adams Jr HP. Clinical scales to assess patients with stroke. *In* Stroke 2011 Jan 1 (pp. 307-333). WB Saunders.

2. Aggarwal A, Dhandapani S, Praneeth K, Sodhi HB, Pal SS, Gaudihalli S, Khandelwal N, Mukherjee KK, Tewari MK, Gupta SK, Mathuriya SN. Comparative evaluation of H&H and WFNS grading scales with modified H&H (sans systemic disease): A study on 1000 patients with subarachnoid hemorrhage. *Neurosurgical review*. 2018 Jan 1;41(1):241-7.
3. Alreshidi M, Cote DJ, Dasenbrock HH, Acosta M, Can A, Doucette J, Simjian T, Hulou MM, Wheeler LA, Huang K, Zaidi HA. Coiling versus microsurgical clipping in the treatment of unruptured middle cerebral artery aneurysms: a meta-analysis. *Neurosurgery*. 2018 Feb 9;83(5):879-89.
4. Bakker NA, Metzemaekers JD, Groen RJ, Mooij JJ, Van Dijk JM. International subarachnoid aneurysm trial 2009: endovascular coiling of ruptured intracranial aneurysms has no significant advantage over neurosurgical clipping. *Neurosurgery*. 2010 May 1;66(5):961-2.
5. Brawanski N, Kashefiolasl S, Won SY, Tritt S, Berkefeld J, Senft C, Seifert V, Konczalla J. Does aneurysm side influence the infarction side and patients' outcome after subarachnoid hemorrhage?. *PLoS one*. 2019;14(11).
6. Chyatte D, Porterfield R. Nuances of middle cerebral artery aneurysm microsurgery. *Neurosurgery*. 2001 Feb 1;48(2):339-46.
7. Dabilgou AA, Drave A, Kyelem JM, Naon L, Napon C, Kabore J. Spontaneous Subarachnoid Haemorrhage in Neurological Setting in Burkina Faso: Clinical Profile, Causes, and Mortality Risk Factors. *Neurology research international*. 2019;2019.
8. Danière F, Gascou G, de Champfleure NM, Machi P, Leboucq N, Riquelme C, Ruiz C, Bonafé A, Costalat V. Complications and follow up of subarachnoid hemorrhages. *Diagnostic and interventional imaging*. 2015 Jul 1;96(7-8):677-86.
9. de Gans K, Nieuwkamp DJ, Rinkel GJ, Algra A. Timing of aneurysm surgery in subarachnoid hemorrhage: a systemic review of the literature. *Neurosurgery*. 2002;50:336-40.
10. Đilvesi Đ, Cigić T, Papić V, Horvat I, Karan M, Vuleković P. The Fisher grade in predicting a degree of cerebral vasospasm in patients after intracranial aneurysm rupture. *Vojnosanitetski pregled*. 2016;73(4):349-52.
11. Hamdan A, Barnes J, Mitchell P. Subarachnoid hemorrhage and the female sex: analysis of risk factors, aneurysm characteristics, and outcomes. *Journal of neurosurgery*. 2014 Dec 1;121(6):1367-73.
12. Jang SH, Do Lee H. The pathogenic mechanisms of motor weakness following aneurysmal subarachnoid hemorrhage: A review. *Neurology Asia*. 2017 Sep 1;22(3).
13. Javadvpour M, Silver N. Subarachnoid haemorrhage (spontaneous aneurysmal). *BMJ clinical evidence*. 2009;2009.
14. Oh JW, Lee JY, Lee MS, Jung HH, Whang K, Brain Research Group. The meaning of the prognostic factors in ruptured middle cerebral artery aneurysm with intracerebral hemorrhage. *Journal of Korean Neurosurgical Society*. 2012 Aug;52(2):80.
15. Qin S, Sun X, Hui P, Li J, Wang Z. Analysis on prognostic factors of patients with ruptured intracranial aneurysms. *Int J Clin Exp Med*. 2017 Jan 1;10(1):1303-8.
16. Rahmanian A, Derakhshan N, Alibai EA. Outcome of In-Hospital Rebleeding and Early Aneurysm Rupture at the Referral Center. *Iranian Journal of Neurosurgery*. 2018 Jul 15;4(2):93-100.
17. Rodríguez-Hernández A, Sughrue ME, Akhavan S, Habdank-Kolaczowski J, Lawton MT. Current management of middle cerebral artery aneurysms: surgical results with a "clip first" policy. *Neurosurgery*. 2012 Nov 30;72(3):415-27.
18. Rosen DS, Macdonald RL. Subarachnoid hemorrhage grading scales. *Neurocritical care*. 2005 Apr 1;2(2):110-8.
19. Ruggeri AG. Ruptured Middle Cerebral Artery Aneurysms: Retrospective Study and Multivariate Analysis of 105 Patients Treated by Surgical Clipping. *Journal of Neurosurgery Research and Reviews*. 2017 Nov 25;1(1).
20. Sandalcioğlu IE, Schoch B, Regel JP, Wanke I, Gasser T, Forsting M, Stolke D, Wiedemayer H. Does intraoperative aneurysm rupture influence outcome? Analysis of 169 patients. *Clinical neurology and neurosurgery*. 2004 Mar 1;106(2):88-92.
21. Santiago-Dieppa DR, Pannell JS, Khalessi AA. Endovascular and surgical options for ruptured middle cerebral artery aneurysms: review of the literature. *Stroke research and treatment*. 2014;2014.
22. Shimoda M, Oda S, Mamata Y, Tsugane R, Sato O. Surgical indications in patients with an intracerebral hemorrhage due to ruptured middle cerebral artery aneurysm. *Journal of neurosurgery*. 1997 Aug 1;87(2):170-5.
23. Taha MM, Alawamry A, Abdelbary TH. Outcome of microsurgical clipping of anterior circulation aneurysms during the period of vasospasm: single center experience in Egypt. *Egyptian Journal of Neurosurgery*. 2019 Dec 1;34(1):5.
24. Ulm AJ, Fautheree GL, Tanriover N, Russo A, Albanese E, Rhoton Jr AL, Mericle RA, Lewis SB. Microsurgical and angiographic anatomy of middle cerebral artery aneurysms: prevalence and significance of early branch aneurysms. *Operative Neurosurgery*. 2008 May 1;62(suppl_5):ONS344-53.
25. Wang GX, Yu JY, Wen L, Zhang L, Mou KJ, Zhang D. Risk factors for the rupture of middle cerebral artery bifurcation aneurysms using CT angiography. *PLoS One*. 2016 Dec 15;11(12):e0166654.
26. Yang W, Huang J. Treatment of middle cerebral artery (MCA) aneurysms: a review of the literature. *Chinese Neurosurgical Journal*. 2015 Dec;1(1):1.



The efficacy of adalimumab on experimentally induced spinal cord ischemia-reperfusion injury

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ABSTRACT

Objective: Paraplegia is a dangerous complication of thoracoabdominal aortic surgery. Various studies have been conducted on the prevention of this complication and some spinal cord protection methods have been proposed. However, there is not any modality that prevent the development of paraplegia certainly. In the I / R period, primary injury triggers secondary injury due to increased inflammation, apoptosis and free radical formation. In this study, we evaluated that the neuroprotective effect of adalimumab in spinal cord ischemia-reperfusion injury.

Materials and Methods: In total, 24 adult New Zealand rabbits were divided into three groups: Group 1, control; Group 2, ischemia-reperfusion by infrarenal aortic clamping; Group 3, adalimumab treated followed by ischemia. Tissue and plasma tumor necrosis factor alpha, interleukin 6, interleukin 10, thiobarbituric acid reactive substance, total oxidant status and total antioxidant status levels were analyzed as a marker of inflammation and oxidation. Histopathological evaluation of the tissues was performed, and apoptosis was evaluated by TUNNEL method.

Results: I/R injury significantly increases plasma and spinal cord tissue at TNF alpha, TOS, TBARS, IL6 levels and reduces plasma and spinal cord tissue to TAS and IL10 levels. Adalimumab treatment significantly reduces plasma and spinal cord tissue to TNF alpha, TOS, TBARS, IL6 and increases plasma and tissue to TAS and IL10 levels.

Conclusion: Adalimumab treatment significantly reduces the spinal cord neuronal damage score and the number of apoptotic cells. This paper aims to demonstrate the important neuroprotective effects of adalimumab on rabbit spinal cord I/R injury.

1. INTRODUCTION

Spinal cord reperfusion injury is described as cell death of neurons although improvement of blood supply of spinal cord after ischemia. It usually occurs because of oxygen free radical-induced lipid peroxida-

Keywords

spinal,
ischemia-reperfusion,
neuroprotection,
inflammation,
adalimumab



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tio, leukocyte activation, inflammation and neuronal apoptosis. Although many clinical treatments of spinal cord injury have been provided in recent years, the results are far from satisfactory [1]. Paraplegia is a serious complication that can occur in a patient undergoing thoracoabdominal aortic surgery and has been seen in 2.4-40% of the patients [2]. Factors that cause this complication are: impaired arterial blood flow and I/R injury of the spinal cord due to collateral supplier damage, perfusion insufficiency, prolonged aortic cross-clamping time, intraoperative proximal hypertension, high cerebrospinal fluid (CSF) pressure and postoperative hypotension [3]. Neurological injury that occurs after impact is called primary injury. Primary injury triggers secondary injury with increased inflammation, apoptosis and the formation of free radicals that have serious effects on outcome. Various pharmacological agents have been used to prevent secondary damage at different success rates [4]. However, there is no complete treatment of secondary injuries.

Tumor necrosis factor is an important immunoregulator molecule that act important role in central nervous system events such as stroke [5]. There are two type of TNF molecule. First type of TNF molecule is transmembrane TNF which acts through cell-to-cell contact to initiate juxtracrine signaling and is important not only for cellular transmission in the natural immune system but also for functional improvement and axonal preservation [6,7]. The second type of TNF molecule is soluble TNF that acts in a paracrine manner and is a substantial mediator of both acute and chronic inflammation [8].

Adalimumab is a potent TNF alpha blocker. It suppresses TNF- α and IL-6, initiate to reduction or inhibition of the inflammatory process [9]. Many studies have shown that TNF- α and IL-6 are elevated during I/R and that they are responsible for activation of the cell death by apoptosis [10]. Recently, the neuroprotective impacts of anti-TNF treatment have been studied in a model of focal cerebral ischemia and beneficial effects have been stated [11].

The aim of this study was to investigate the neuroprotective impacts of adalimumab on rabbit spinal cord I/R injury model.

2. MATERIALS AND METHODS

This research was carried out in the Experimental

Medicine Application and Research Center of Necmettin Erbakan University. The experimental protocol was assessed and confirmed by the Ethics Review Committee of Necmettin Erbakan University. The animals were kept at a room temperature (18–21 °C) and fed on a standard diet. A 12 -h light-dark cycle (08:00 –20:00 hours light/20:01–07:59 hours dark) was preserved. The animals were able to get as much food and water as they wanted.

2.1 Groups

Twenty-four adult New Zealand rabbits were randomly separated into three groups: Group 1, control group (n=8); Group 2, ischemia-reperfusion (I/R) group and group 3 (n=8), I/R injury+ adalimumab (40 mg/kg, ip, single dose) treatment group.

All rabbits were anesthetized by intramuscular (i. m.) injection of ketamine (50 mg/kg) (Ketalar, Parke-Davis, Eczacıbaşı, Istanbul, Turkey) and xylazine (10 mg/kg) (Rompun, Bayer, Istanbul, Turkey) and permitted to breathe during the procedure. An intravenous catheter was placed in the auricular vein of the animals and preoperative cefazolin 10 mg/kg (Cefamezin, Eczacıbaşı, Istanbul, Turkey) was given as a single dose. As maintenance, 0.9% NaCl (20 ml/h) was given throughout the experiment.

All rabbits underwent laparotomy in supine position. Aortic cross-clamp was not applied to group 1. In group 2 and 3 the abdominal aorta was detected and dissected carefully from the beginning of the left renal artery by transperitoneal approach. Five minutes before occlusion, 100 IU/ kg heparin was given intravenously. The aorta was then cross-clamped using an aneurysm clip with a closing force of 70 grams (Yasargil FE 721, Aesculap). The clipping site was just below the origin of the left renal artery. Pulsation of the femoral artery disappeared after occlusion. The aneurysm clip was removed 30 minutes later, and aortic pulsation was restored. Neither aortic nor caval hemorrhage were observed during surgery. Before closure, rabbits of group 3 received single dose intraperitoneal 40 mg/kg adalimumab treatment. After the closure of laparotomy all rabbits awoke and returned to their cages. The animals were followed neurologically, and motor inefficiency and recovery rates were recorded. Seventy-two hours later, all rabbits were re-anesthetized by intramuscular (i. m.) injection of ketamine (50 mg/kg) (Ketalar, Parke-Davis, Eczacıbaşı, Istanbul, Turkey) and xylazine (10 mg/kg)

(Rompun, Bayer, Istanbul, Turkey). Blood samples were taken from auricular veins for biochemical examination. For histopathological examination spinal cord samples were taken from lumbar spinal cord segments between L4-L6 by laminectomies and the rabbits were sacrificed.

2.2 Biochemical Analysis

Venous blood samples were collected by centrifugation at 4° C and 1,000 g for 10 minutes to remove plasma. Plasma samples were kept at -80 ° C until the parameters were studied.

Spinal cord tissue samples were provided in pH 7.4 50 mM phosphate buffer and kept at -80 ° C until they were analyzed. The thawed tissue samples were weighed and homogenized in ice using a mechanical homogenizer and an ultrasonic homogenizer in a 10fold (w / v) cold phosphate buffer (50 mM, pH: 7.4). The supernatants were separated by centrifuging the homogenates for 10 min at 4 ° C and 10.000 g. Pierce bicinchoninic acid-BCA (Thermo Scientific, Illinois, USA) was used to measure spectrophotometrically plasma and spinal cord tissue total oxidant (TOS) and antioxidant status (TAS) (Rel Assay Diagnostics, Gaziantep, Turkey), thiobarbituric acid reactive substances (TBARS) (Oxford Biomedical Research, Missouri, USA) and tissue protein levels. Plasma and spinal cord tissue IL-6, IL-10 and TNF alpha levels were examined by using ELISA antigens that were intrinsic to rabbits (Elabscience Biotechnology Co., Wuhan, China).

2.3 Histopathological Studies

Spinal cord samples were stabilized by 10% formaldehyde for two days and then embedded in paraffin blocks. After dehydration, coronal sections of the spinal cord segment were severed at a thickness of 4 µm and stained with hematoxylin and eosin (HE) in order to examine the structural changes. Gray matter was checked in five different areas in each section. Depending on the degree of inflammation, hemorrhage, axonal swelling, congestion, neuronal degeneration and vacuolization of the spinal cord, the light microscopic findings were graded on a scale ranging from 0 to 3, corresponding to "no change", "mild", "moderate" and "severe" changes, respectively. The histopathological score was calculated for each spinal cord sample [12].

Apoptotic cells were labeled using an ApopTag In Situ Apoptosis Detection Kit (Millipore). DNA fragments in spinal cord regions were altered by the action of terminal deoxynucleotidyl transferase. The manufacturer's instructions were followed during procedures. In each section five dark visual fields were randomly chosen, and the TUNEL-positive neurons and the total number of neurons in the selective visual fields was counted. TUNEL-positive index (the TUNEL-positive to whole neurons ratio) was computed. Eight sections from each group were used for measurement, and five high-powered visuals were indiscriminately picked from every section to carry out measurement of the TUNEL-positive indices [13].

2.4 Statistical analysis

Data were analyzed using SPSS (version 24.0, SPSS Inc.) and expressed as mean ± SD. Comparisons were made by the Kruskal-Wallis test. Differences among the groups were evaluated by the Mann-Whitney U test. A $p < 0.05$ was considered statistically significant. Histopathological score and TUNEL positive cell count were contrasted using a one-way analysis of variance (ANOVA) with TUKEY test.

3. RESULTS

3.1 Histopathological evaluation

I/R injury significantly increased the spinal cord neuronal damage score and apoptotic cell count. Adalimumab treatment statistically substantially decreased spinal cord neuronal injury score and apoptotic cell count ($p=0$). Large motor cells were observed in anterior horn of the spinal cord in the control group (Figure 1A). No changes were observed in the neurons. The most serious injury was seen in ischemia-reperfusion group in spinal cord in HE sections (Figure 1B, C, D). Necrosis, hemorrhage and congestion were noticed in ischemia-reperfusion group. Nissl substances disappeared in necrotic neurons. In addition, neuropil vacuolization and tissue loss were observed in the gray matter (Figure 1E, F, G). Compared with control group, it was noticed that histopathological score rose in ischemia-reperfusion group. Histopathological alterations and score significantly reduced in adalimumab treatment group (Fig 1H). Myelin swelling determined in white matter in ischemia group and the adalimumab group had less myelin swelling compared to the ischemia group. (Figure 2A, B, C). TUNEL positive cells count

increased in I/R group when compared with control group (Figure 3A, B). Adalimumab treatment

decreased TUNEL positive cells count (Figure 3C). Figure 3D showed differences among groups.

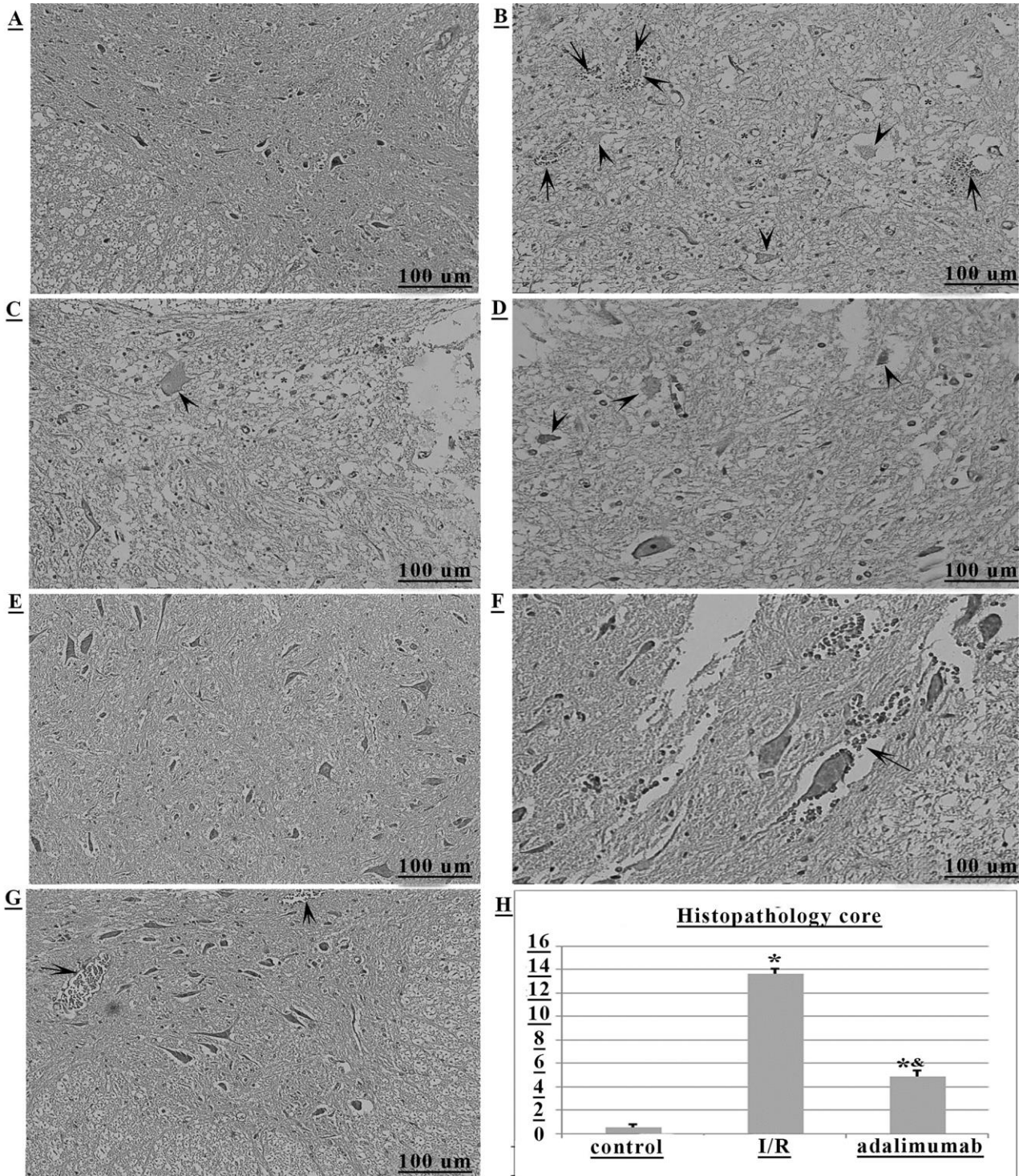


Figure 1. Histopathological photomicrographs of spinal cord tissue stained HE A: Control Group : Neurons in grey matter and White matter B: Necrotic neurons in Ischemia-Reperfusion Group (black arrow), Haemorrhage ve (arrows), *:vacuoles. C: chromatolyses in Ischemia-Reperfusion Group (arrow head), *:vacuoles. D: pyknosis in Ischemia-Reperfusion Group (arrows), necrotic neurons (arrows). E: Normal neurons in Adalimumab treatment group F: haemorrhage in Adalimumab treatment group (arrow). G: Congestion in Adalimumab treatment group (arrows). H: Histopathological assessment of spinal cord, (* P < 0.05, compared to group 1; &P < 0.05, compared to group 2).

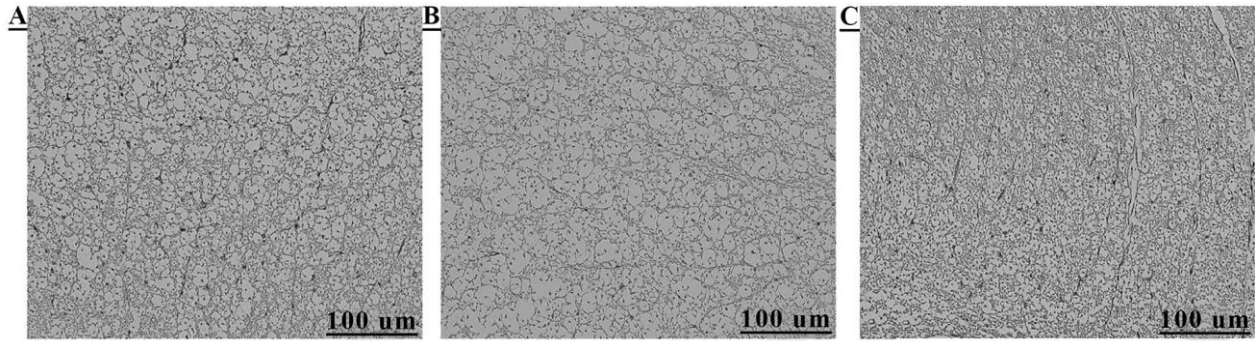


Figure 2. Histopathological photomicrographs of white matter stained HE A: Control Group. B: Ischemia-Reperfusion Group C: Adalimumab treatment group.

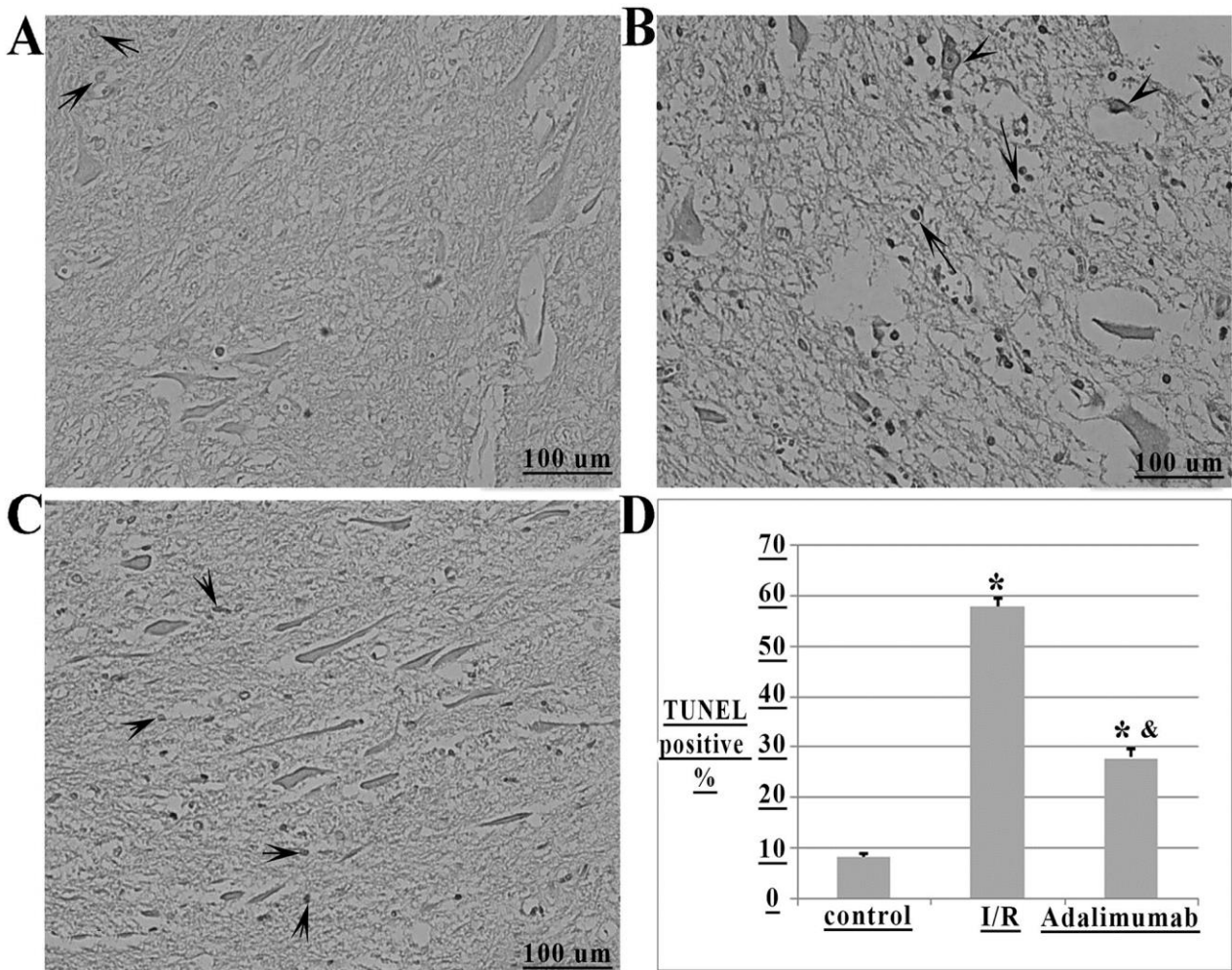


Figure 3. Representative spinal cord sections stained in the TUNEL assay. A: Control group, B: ischemia-Reperfusion group C: Adalimumab treatment group .D: Comparison of groups according to TUNEL-positive cells in spinal cord, (* P < 0.05, compared to group 1; &P < 0.05, compared to group 2).

3.2 Biochemical evaluation

I/R injury significantly increased the plasma and spinal cord tissue TNF alpha, TOS, TBARS, IL6 levels and reduced the plasma and spinal cord tissue TAS

and IL10 levels. Adalimumab treatment significantly decreased the plasma and spinal cord tissue TNF alpha, TOS, TBARS, IL6 levels and raised plasma and tissue TAS and IL10 levels (Figure 4, Figure 5).

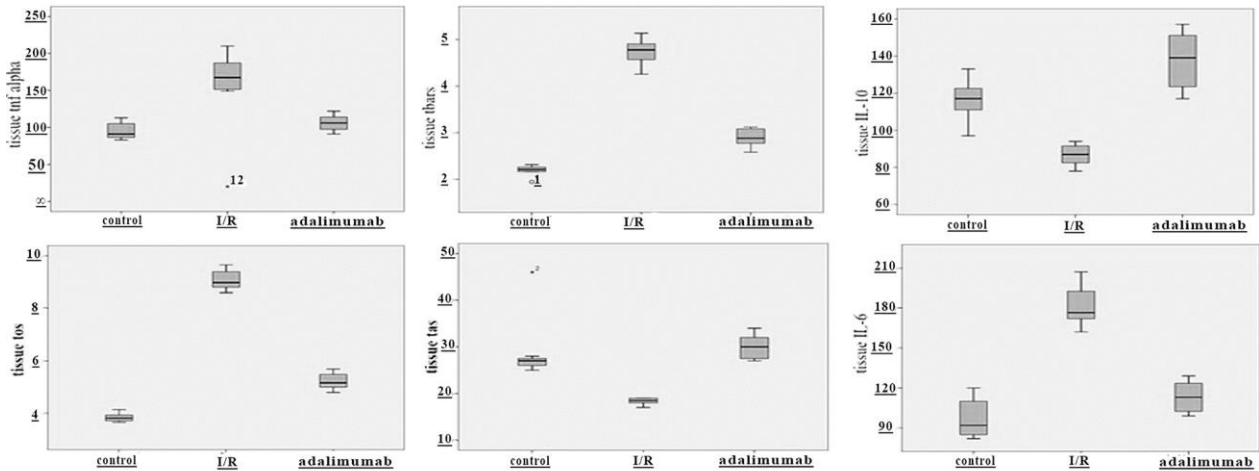


Figure 4. The effects of adalimumab on plasma levels of TAS,TOS,TBARS,TNF ALPHA,IL6,IL10.

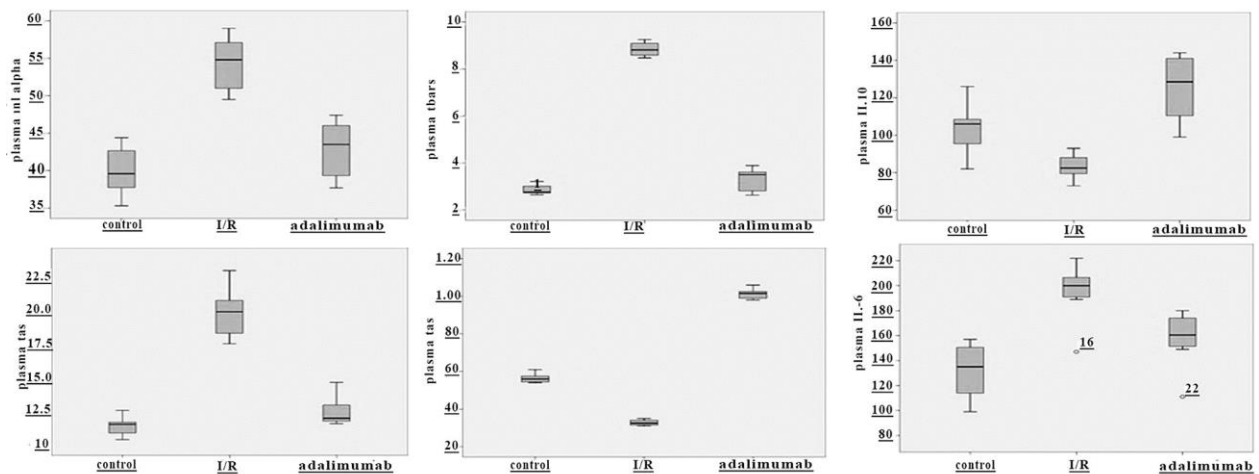


Figure 5. The effects of adalimumab on tissue levels of TAS,TOS,TBARS,TNF ALPHA,IL6,IL10.

4. DISCUSSION

Neural tissues can be said to be very sensitive to ischemia. I / R damage of the spinal cord during thoracoabdominal vascular surgery can cause serious discomfort such as paraplegia. Primary injury triggers secondary injury with increased inflammation, apoptosis and free radical formation during the I / R period [14]. Secondary injury may result in endothelial dysfunction and increase vascular permeability that promotes migration and activation of immune cells. These activated immune cells infiltrates the related area and secrete some proinflammatory cytokines. Enhanced inflammation causes reactive oxygen species produce and induces lipid peroxidation which causes injury in the ultrastructure of neural cell membranes and hinders their critical functions [15].

The spinal cord is very sensitive to ischemia because it has its own anatomical features. "The infrarenal aortic cross-clamp" method used in this study leads to severe spinal cord injury. It was first described by Liang et al. as an experimental method [16,17]. Rabbits have segmental blood supply in their lumbosacral spinal cord. Thus, rabbit model of spinal cord I/R injury is commonly used. It is clear that there are many causes of paraplegia. Long-term ischemia, interruption of critical intercostal and lumbar arteries, decrease in spinal cord perfusion pressure and postoperative reperfusion injury are some of these reasons [18]. Therefore, this method is thought to be appropriate to imitate the complications of aortic surgery.

High levels of plasma and spinal cord tissue TNF alpha, TOS, TBARS and IL6 in I/R group signifies

increased inflammation and oxidative stress. Neuronal damage score and apoptotic cell count increase after I/R injury. Adalimumab treatment significantly improves biochemical and histopathological adverse impacts of I/R injury.

5. CONCLUSION

In this study, it was found that adalimumab had significant neuroprotective effects on rabbit spinal cord I/R injury. After I / R injury, high inflammation and oxidative stress were successfully reversed by adalimumab, and the worse effects of biochemical, histopathological and neurological I / R damage were mitigated. Further studies are needed to carry out this treatment in clinical practices.

REFERENCES

- Kieffer E, Chiche L, Cormier E, Guegan E. Recurrent spinal cord ischemia after endovascular stent grafting for chronic traumatic aneurysm of the aortic isthmus. *Journal of Vascular Surgery* 2007; 45: 831–833.
- Coselli JS, LeMaire SA, Conklin LD, Koksoy C, Schmittling ZC. Morbidity and mortality after extent II thoracoabdominal aortic aneurysm repair. *Ann Thorac Surg.* 2002; 73(4):1107-15, [http://dx.doi.org/10.1016/S0003-4975\(02\)03370-2](http://dx.doi.org/10.1016/S0003-4975(02)03370-2).
- Kocogullari CU, Becit N, Erkut B, Keleş MS, Ceviz M, Ates A, Gündoğdu C, Kaygın MA, Kocak H. Prevention of reperfusion injury of the spinal cord in aortic surgery: an experimental study. *Surg Today* 2008; 38(3): 237–44.
- Junk AK, Mammis A, Savitz SI, Singh M, Roth S, Malhotra S, Rosenbaum PS, Cerami A, Brines M, Rosenbaum DM. Erythropoietin administration protects retinal neurons from acute ischemia-reperfusion injury. *Proc Natl Acad Sci USA* 2002; 99: 10659– 10664.
- Lambertsen KL, Biber K, Finsen B. Inflammatory cytokines in experimental and human stroke. *J Cereb Blood Flow Metab* 2012; 32: 1677–1698.
- Alexopoulou L, Kranidioti K, Xanthoulea S, Denis M, Kotanidou A, Douni E, Blackshear PJ, Kontoyiannis DL, Kollias G. Transmembrane TNF protects mutant mice against intracellular bacterial infections, chronic inflammation and autoimmunity. *Eur J Immunol* 2006; 36: 2768–2780.
- Brambilla R, Ashbaugh JJ, Magliozzi R, Dellarole A, Karmally S, Szymkowski DE, Bethea JR. Inhibition of soluble tumour necrosis factor is therapeutic in experimental autoimmune encephalomyelitis and promotes axon preservation and remyelination. *Brain* 2011; 134: 2736–2754.
- Ruuls SR, Hoek RM, Ngo VN, McNeil T, Lucian LA, Janatpour MJ, Korner H, Scheerens H, Hessel EM, Cyster JG, McEvoy LM, Sedgwick JD. Membrane-bound TNF supports secondary lymphoid organ structure but is subservient to secreted TNF in driving autoimmune inflammation. *Immunity* 2001; 15:5 33–543.
- McGovern JL, Nguyen DX, Notley CA, Mauri C, Isenberg DA, Ehrenstein MR. Th17 cells are restrained by Treg cells via the inhibition of interleukin-6 in patients with rheumatoid arthritis responding to anti-tumor necrosis factor antibody therapy. *Arthritis Rheum* 2012; 64: 3129–3138.
- Xu YF, Liu M, Peng B, Che JP, Zhang HM, Yan Y, Wang GC, Wu YC, Zheng JH. Protective effects of SP600125 on renal ischemia-reperfusion injury in rats. *J Surg Res* 2011; 169: 77–84.
- Clausen BH, Degn M, Martin NA, Couch Y, Karimi L, Ormhøj M, Mortensen ML. Systemically administered anti-TNF therapy ameliorates functional outcomes after focal cerebral ischemia. *J Neuroinflammation.* 2014; 12: 11:203. doi: 10.1186/s12974-014-0203-6.
- Kishimoto T, Akira S, Narazaki M, Taga T. Interleukin-6 family of cytokines and gp130. 1995; 86:1243-54.
- Gürer B, Kertmen H, Kasim E, Yilmaz ER, Kanat BH, Sargon MF, Arikok AT, Ergüder BI, Sekerci Z. Neuroprotective effects of testosterone on ischemia/reperfusion injury of the rabbit spinal cord. *Injury.* 2015; 46(2): 240-8.
- Klune JR, Tsung A. Molecular biology of liver ischemia/reperfusion injury: Established mechanisms and recent advancements. *Surg Clin North Am* 2001; 90: 665-77.
- Chan PH. Role of oxidants in ischemic brain damage. *Stroke.* 1996; 27(6): 1124-9.
- Liang Y, Yang Q-H, Yu X-D, Jiang D-M. Addictive effect of tetramethylpyrazine and deferoxamine in the treatment of spinal cord injury caused by aortic crossclamping in rats. *Spinal Cord* 2011; 49: 302-306; doi: 10.1038/sc.2010.113; published online 14 September 2010.
- Shechter R, London A, Varol C, Raposo C, Cusimano M, Yovel G, Rolls A, Mack M, Pluchino S, Martino G, Jung S, Schwartz M. Infiltrating blood-derived macrophages are vital cells playing an anti-inflammatory role in recovery from spinal cord injury in mice. *PLoS Med* 2009; 6(7): e1000113. doi:10.1371/journal.pmed.1000113.
- Sahin MA, Onan B, Guler A, Oztas E, Uysal B, Arslan S, Demirkilic U, Tatar H. Cilostazol, a type III phosphodiesterase inhibitor, reduces ischemia/reperfusion-induced spinal cord injury. *Heart Surg Forum* 2011; 14: 171-177.



Experience, challenges and lessons learnt from microsurgical clipping of intracranial aneurysms at an emerging neurosurgical centre

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ABSTRACT

Objectives: To share our experience, challenges faced, lessons learnt and analyze the results of surgical management by microsurgical clipping of intracranial aneurysms at an emerging neurosurgical centre at Guru Gobind Singh medical college and hospital (GGSMC&H) Faridkot, Punjab. India.

Material and Methods: This study includes all the patients who presented with the diagnosis of intracranial aneurysm on CT angiography and were treated with the microsurgical clipping, between March 2017 to April 2019.

Results: There was a total of 23 patients 11 female and 12 male. Age range 32 to 85 years. On admission 22 patients had SAH on CT scan and one was admitted after incidental detection of the aneurysm without SAH. The time interval between ictus and admission was 0-3 days in 13 patients, 3-14 days in 8 patients and more than 14 days in 1 patient. WFNS grade (gd) I-15 patients, gd II-2, gd III-2, gd IV-3 patients. Fisher gd I-nil, gd II-9, gd III-4, gd IV-9 patients. In 23 patients 27 Aneurysms were clipped. Distribution of location was Anterior Communicating-12, Distal Anterior Cerebral Artery- 4, Middle cerebral artery (MCA) Bifurcation-3, MCA trifurcation-1, Anterior Choroidal-1, Posterior Communicating (P-com) -1, Ophthalmic Internal Carotid Artery (OICA)-4 and three patients had associated multiple aneurysms. Size of aneurysms varied from < 02mm diameter in 2 patients, 2-25mm - 23 and, more than 25mm-2 aneurysms. There was intra op rupture in 2 cases. Post-operatively 2 patients developed hemiparesis, which recovered, nine patients developed vasospasm. Two patients developed chest related complications. One patient developed renal failure. There were 8 deaths. Patients are on follow up since March 2017 till date.

Conclusions: Intracranial aneurysms are challenging to manage due to their proximity to vital intracranial structures, and difficulty in securing intracranial proximal control. Thorough knowledge of intracranial anatomy of adjacent relations, arachnoid planes and skilful dissection is a key element for a successful outcome. Data collected from GGSMC & Hospital may not be representative of the entire state or country's population. Therefore, a large-scale data collection is necessary to create our own database to ascertain the risk factors and preventive measures that are exclusive to our state and nation.

INTRODUCTION

In simple words an aneurysm is an abnormal dilatation of an artery wall

Keywords

intracranial aneurysm,
microsurgical clipping,
pterional craniotomy,
subarachnoid haemorrhage,
vasospasm



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and in the brain commonly arises at branching points on a parent artery. Intracranial aneurysms (IAs) are usually revealed after they rupture, leading to subarachnoid hemorrhage (SAH). Usually the sentinel headache is the earliest warning sign of incipient rupture. Less frequently, they manifest themselves as a result of mass effect or are noticed incidentally during neuro-radiologic examinations performed for other diagnostic purposes.(6,9) Subarachnoid hemorrhage (SAH) due to the rupture of an intracranial aneurysm is a devastating event associated with high rates of mortality (40%–50%) and morbidity.(10, 20, 23, 13) The risk of intracranial aneurysm for human beings is 1 to 2% .(5)

The prime motive of intracranial aneurysm treatment is to exclude the aneurysm from the circulation; this can be achieved through two ways, microsurgical treatment or endovascular obliteration. Since ours is an emerging neurosurgical center where we have basic neurosurgical facilities, but lack digital subtraction angiography (DSA) and coiling facility. We are managing intracranial aneurysms with microsurgical clipping. The objective of this article is to share our experience acquired with the use of microsurgical obliteration of intracranial aneurysms at Guru Gobind Singh Medical College and Hospital Faridkot, Punjab, India.

MATERIAL AND METHODS

This study includes all the patients admitted to the neurosurgery department who presented with diagnosis of intracranial aneurysm and were treated with the microsurgical clipping, in the period between March 2017 and April 2019. Patients who presented with poor neurological status, with fixed dilated pupils and who did not give consent for surgery were excluded.

On their arrival at the emergency department, all the patients were treated according to the defined protocol for the treatment of subarachnoid hemorrhage in the hospital and were admitted to Neurosurgical intensive care units and managed depending on their neurologic state at the moment of admission, determined by using GCS, the Word Federation of Neurologic Surgeons (WFNS) scale and fisher grades of SAH on non contrast computed tomography (NCCT) Scan of brain.

All the patients suspected with intra cranial aneurysm were subjected to cerebral CT

angiography. Presence of aneurysm on CT angiogram was considered an essentiality to proceed for surgical treatment.

The aneurysmal sacs were labeled according to their number in single or multiple aneurysms, and distributed according to their location in the anterior segment or posterior segment of the circle of Willis', the projection and whether unilateral or bilateral. Outcome was evaluated using the modified Rankin scale (mRS). Patients are under follow-up since March 2017 till date.

RESULTS

There were a total 23 patients out of which 11 (47.82%) were female and 12 (52.17%) were male. Age range was 32 to 85 years. At the time of admission 22 patients had SAH on CT scan and one patient was admitted after incidental detection of aneurysm without SAH. Time of admission from time of ictus 0-3 days in 13 (59.09%) patients, 3-14 days in 8(36.36%) patients and more than 14 days in 1(04.54%) patient. WFNS gd I-15(68.18%), gd II-2 (09.09%), gd III-2 (09.09%), gd IV-3(13.36%) patients. Fisher gd I-nil, gd II-9 (40.90%), gd III-4 (18.18%), gd IV-9 (40.90%) patients. There were 31 aneurysms in total out of which 27 were clipped in 23 patients. Location wise anterior Communicating Artery (A-Com)- 12 , Distal Anterior Cerebral Artery DACA- 4, middle cerebral artery (MCA) Bifurcation-3, MCA Trifurcation-1, Anterior Choroidal-1, Posterior communicating (Pcom)-1, Ophthalmic internal carotid artery (ICA)-4 and three patients had associated multiple aneurysms. Size of aneurysms varied from < 02mm diameter in 2 patients, 2-25mm in 23 and, more than 25m in 2 patients (Table 1).

Sixteen 69.5% patients had comorbidities out of which 11 (68.75%) hypertension, one (06.25%) Diabetes, one (06.25%) Asthma, one (06.25%) had Chronic liver disease and in 1 patient (06.25%) Chronic kidney disease was diagnosed post operatively. There was intra op rupture in 2 (07.07%) cases. Post operatively 2 (8.69%) patients developed hemiparesis, which recovered over a period of time, nine (39.13%) patients developed vasospasm after surgery. Two (08.69%) patients developed chest related complications. In the study there were 8(34.78%) deaths. Follow up has ranged since March 2017 to till date.

Variable		No	%
Aneurysm Number	Single	18	78.26
	Multiple	5	21.73
Aneurysm side	Unilateral	21	91.30
	Bilateral	2	08.70
Size of the sac	<2mm	2	7.40
	2-25 mm	23	85.18
	>25 mm	2	7.40
Morphology of the sac	Secular	21	77.77
	Fusiform	NIL	0
	Multilobulated	6	22.23
Condition of the Sac	Ruptured	22	81.48
	Unruptured	5	18.52
A com aneurysms		12	44.44
	Superiorly projecting	4	33.33
	Anteriorly projecting	2	16.66
	Antero-superior	1	08.33
	Inferior	3	25
	Antero-Inferior	2	16.66

DACA aneurysms		4	14.81
	anterior	1	25
	Anterior superior	2	50
	Anterior toward opp side	1	25
	superior	Nil	0
MCA bifurcation Aneurysm		3	11.11
MCA trifurcation Aneurysms		1	03.70
Anterior choroidal Aneurysm		1	03.70
Ophthalmic seg Aneurysm		4	14.81
Communicating segment aneurysm		1	03.70
P-com Aneurysm		1	03.70

DISCUSSION

Intracranial aneurysms (IAs) are localized dilations of the cerebral arteries wall and are prone to rupture, resulting in bleeding. The overall prevalence of unruptured IAs is between 2% and 3.2% in the general population with a male to female ratio of 1:2.(19) Ruptured aneurysm is responsible for 85% of SAH which is one of the leading causes of haemorrhagic stroke.(18)

Junjie Zhao et al have described 4 Basic Types of intra cranial aneurysms (IAs).(25) (A)- Secular are the most common type of IAs. They resemble a round out pouching with well-defined aneurysmal domes and necks connecting to the parent vessel. Commonly they occur at bifurcation locations e.g. between the MCA and the posterior cerebral artery (PCA), between the anterior cerebral artery (ACA) and the anterior communicating artery (A Com) and between the bifurcations of MCA branches. (4) (B)- Micro aneurysms are IAs with diameters smaller than 2 mm.(11) (C)-Giant IAs are with diameters over 25 mm. The latter account for merely 5% of all IAs, but their prognosis is relatively dismal. (7- 17) Untreated giant IAs have over 50% risk in rupturing and 88% to 100% in mortality at 2-year follow-up. (2,12) We encountered two giant aneurysms. Due to their large size the mass effect alone can cause intracranial hypertension and neurological dysfunctions one of our patient with giant aneurysm presented with decreasing vision. (D)-Fusiform IA is widened and thinning segment of artery. According to Yahia et al, this dilatation must affect at least 270° of the lumen's circumference to be classified as fusiform.(24) Their surgical treatment is challenging due to the vital perforators located within the affected segment. We did not encounter any fusiform aneurysm.

In our Study the majority of patients were from the state of Punjab, (52.17%) male and (47.82%) female. A study conducted by Ab Ghani et al showed the mean age of affected patients as 48.9 years old, with 56.7% males and 43.3 % females.(1) A study by Rinkel et al., suggests that the incidence of intracranial aneurysm increases with age, and the most of the cases occur among patients in fifth decade of their life. (15) In our study the mean age of the patient was 50.3years. Male patients were more frequent than females, which is in contrast to the usual pattern of females being affected more than males. (16) However, this ratio does not seem to be applicable in all populations and may vary from state to state and country to country. (8)

In our study 69.5% patients had comorbidities out of which 68.75% hypertension, 06.25% Diabetes, 06.25% Asthma, 06.25% had Chronic liver disease and in 1 patient (06.25%) Chronic kidney disease was diagnosed post operatively. In the study population of Ab Ghani et al, 85.7% had hypertension. (1) Variation of blood pressure and hypertension are

known risk factors for the development of intracranial aneurysm.(21)

Intracranial arteries are comprised of the outer layer of the adventitia, a muscular media that maintains most of the vessel wall integrity and the inner layer of intima. As compare to other vessels in the body they do not have external elastic lamina, which is located between the adventitia and media leading to less elasticity of the media. They also have thinner adventitia compared to the extra cranial artery wall. These vessels exist in the subarachnoid space, which lack surrounding connective tissue to support the vessels. (1) These features predispose intracranial arteries to the formation of saccular aneurysms, commonly in hypertensive patients as the thin and less elastic vessel wall is subject to increased pressures, as well as in patients with congenital conditions which predispose to defects in the muscular layer of the arterial wall, such as autosomal dominant polycystic kidney disease, Ehler- Danlos syndrome fibromuscular dysplasia, and Marfan syndrome. (1, 15, 3)

Patients without history of hypertension in our study but having hypertension on presentation might have been undiagnosed with hypertension previously or it could be due to raised ICP. Common history among our patients was that they rarely go to the physician for routine checkup and those who do so, many of them had history of poor compliance of antihypertensive medication and no regular follow up. These facts accentuate the importance of hypertension screening and control.

Among the 23 patients who underwent microsurgical clipping, the majority were Fisher II, III or Fisher IV indicating that most patients only presented post intracranial aneurysm rupture with an added risk of vasospasm. It also reflects that incidental finding of intracranial aneurysm is still rare despite the improvement of our health services and increasing amount of imaging being done for other neurologic symptoms.

Post operatively two patients developed hemiparesis, which recovered, one patients developed chest related complication because of underlying bronchial asthma despite an improvement in GCS. However, based on the outcome in our observational study, within a span of three months after surgery, 8 (34.78%) had the mRS score of 0 with no symptoms at all, and 8 (34.78%) had the mRS score of 6 or had died. The cause of

death was post operative vasospasm in 4 (50%) patients. One (12.5%) of them had anaemia secondary to upper gastrointestinal bleed due to stress ulcer which was diagnosed on upper GI endoscopy. Postoperative chest infection in one (12.5%), chronic renal failure in one (12.5%) and pulmonary embolism in one (12.5%) patient. Literature says maximum risk of vasospasm is 3-14 days of ictus.(22,14) Three of our patients who died, developed vasospasm in first two weeks after ictus. After three months of discharge, four patients with mRS scores of 1 and 2 improved to the mRS score of 0, and after 6 months of discharge total 12 (52.17%) patient improve to MRS score 0. Others three are still on rehabilitation. This improvement was mainly due to the after care of patients, which comprises intensive rehabilitation and physiotherapy.

OUR INTERESTING CASES

Giant Aneurysm

Case 1: 45 year old female presented with right basal ganglia bleed CT angio revealed giant multi lobulated ophthalmic segment aneurysm. Patient underwent right pterional craniotomy with right neck control for proximal ICA. Because of its large size proximal and distal ICA was not visible. So, aneurysmal sac was punctured, with controlled suction aneurysmal sac was deflated that led to visualization of proximal, distal ICA and neck of the aneurysm, as a result we were able to apply permanent clip over the neck of the aneurysm. Post operatively patient was discharged without any deficit. Figure 1.

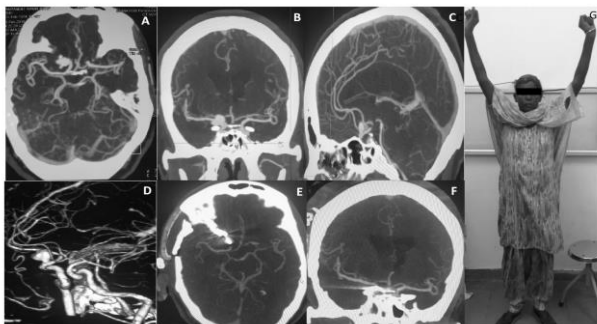


Figure 1. Computed tomography angiography of right ophthalmic aneurysm, axial view (A) showing multilobulated large aneurysm. Coronal view (B) showing the superiorly projecting aneurysm. Sagittal view (C) aneurysm with narrow neck. Three dimensional CT (D) showing large superiorly projecting ophthalmic aneurysms. Post op Computed tomography angiography (E and F) complete occlusion of aneurysm. (G) Patient is fine with no deficit.

Multiple Aneurysms

Case 2: 32 years old male patient presented with left anterior choroidal artery aneurysm and right MCA Bifurcation aneurysm. It was a challenge to manage both side aneurysms in a single surgery. We planned for two stage surgery based on SAH picture on NCCT brain and CT angio findings, we first clipped left anterior choroidal artery aneurysm. After dissection of the aneurysm the surgeon got engaged in selecting appropriate clip for few seconds, surgical field was out of his sight, aneurysm got ruptured and visible anatomy got distorted that led to difficult management of the ruptured aneurysm. The learning point for us was, if possible never let the aneurysm out of your vision after aneurysmal neck dissection from its surroundings and try to select the appropriate clip prior to final dissection. We clipped right MCA bifurcation aneurysm second stage successfully and patient was discharged with no deficit.

Case 3: 45 years male patient was presented with right MCA aneurysm and ophthalmic segment bleb. MCA aneurysm was clipped first and we faced difficulty in clip application over bleb, the clip slipped multiple times over bleb. We realized that we had limited number of small aneurysmal clips. Although in a small center like ours there are limited resources, surgeon should try to have back up of all sizes and shapes of clips before surgery. Figure 2.

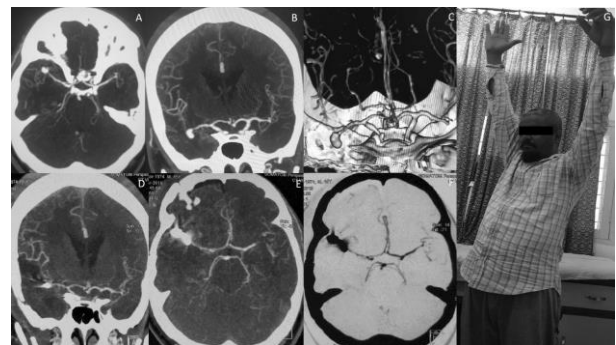


Figure 2. Computed tomography angiography of patient with multiple intracranial aneurysm (right middle cerebral artery bifurcation and right ophthalmic aneurysm), axial view (A) showing laterally projecting aneurysm right middle cerebral artery bifurcation aneurysm. Coronal view (B) and Three dimensional CT (C) showing the inferiorly projecting right MCA bifurcation and medially projecting right ophthalmic aneurysm. Post op Computed tomography angiography, coronal view (D), axial (F and G) showing complete occlusion of both aneurysms. (G) Patient is fine with no deficit.

Case 4: A 65 years old female patient was presented with left pcom (Bled), left ophthalmic segment, A-com and right MCA bifurcation aneurysm. All left sided aneurysms were clipped in the first surgery. Right MCA bifurcation Aneurysm was planned for second stage elective surgery but patient developed thrombocytopenia and surgery was deferred. Figure 3 A&B. Patient is asymptomatic and on regular follow up and is being worked up for intractable thrombocytopenia.

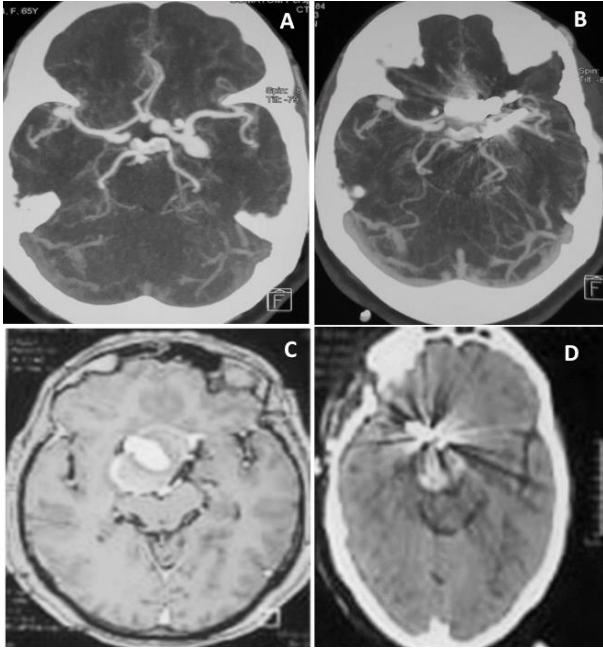


Figure 3. Axial view (A) showing the laterally projecting left PCOM , medially projecting left ophthalmic segment, ACOM and laterally projecting right MCA bifurcation aneurysm and (B) Post op Computed tomography angiography left PCOM, left ophthalmic segment and ACOM aneurysms T1 contrast MRI axial view (C) showing giant atherosclerotic right communicating segment aneurysm and non-contrast computed tomography, Axial view (D) showing post op clip artefact with occlusion of neck.

Missed aneurysm

Case 5: A 52 years old male patient was presented with SAH in left sylvian fissure and inter hemispheric fissure CT angiogram suggested left DACA aneurysm. In view of SAH location the aneurysm was approached through left inter hemispheric approach but we could not find aneurysm on the left side. We found aneurysm on right DACA after cutting the falx. Aneurysm was clipped and patient remained stable for one week and was planned for discharge but the night before discharge patient had a sudden fall in

GCS, immediate NCCT with CT angio Head was done, it showed diffuse SAH and Suggested right MCA aneurysm which was not visualized on the first CT Angiogram. We learnt that when ever there is discrepancy in the location of the aneurysm and the location of SAH on CT Angiogram, DSA should be done. Unfortunately, in our setup DSA facility is not available.

Aneurysms, which required ICA ligation.

Case 6: A 35 years old male patient was presented with left ophthalmic segment aneurysm, neck control was taken before starting surgery, during dissection of sylvian fissure aneurysm neck got avulsed probably due to traction during sylvian fissure dissection and brain started bulging, and patient became hypotensive. We could not control the bleeding with neck control, urgent left temporal and frontal lobe were removed and ICA just proximal to its bifurcation was clipped, In our opinion that was the best possible thing we could do in such a condition. Post op patient developed aphasia and right-side hemiplegia and is still on follow up and under rehabilitation. Again, we realized the importance of DSA in which we could appreciate the collateral supply and prepare our self for bypass. Reconstruction of ICA at the site of aneurysm with multiple clips and wrapping of it with muscle graft was our back up plan but, patient's deteriorating conditions did not allow us to attempt that.

Giant atherosclerotic communicating segment aneurysm which required ICA reconstruction

Case 7: A 55 years female presented with right eye vision loss having giant atherosclerotic right communicating segment aneurysm involving inferior half circumference of ICA. Aneurysmal sac medially-extending into the sella with compression on optic chiasma, laterally-adherent to the tent with compression of third nerve, posteriorly-adherent to basilar artery into the interpeduncular fossa. Proximal temporary clip was applied. Incision was given on aneurysmal neck both medial and lateral to ICA leaving 1cm of neck on each side contiguous to wall of ICA to reconstruct the inferior vessel wall. Major part of the neck and sac were atherosclerosed with calcification. Permanent fenestrated right-angled clip applied to reconstruct the ICA, which kept on slipping due of atherosclerosis of neck. So, atherosclerotic plaques were removed with Penfield

dissector and ICA was reconstructed with three permanent fenestrated right-angled clips. Medial and lateral part of sac was excised but part in the interpeduncular fossa couldn't be removed keeping in mind the risk of injury to basilar artery. Post operatively patient had right anterior choroidal artery infarct with medical research Council (MRC) Gd 3 power in left side. Patient was discharged on 7th post op day. Patient was put on anti-platelets and after three weeks she started with hematuria in view of which anti-platelets were stopped and patient developed massive pulmonary embolism and expired. Figure 3 C&D, Figure 4.

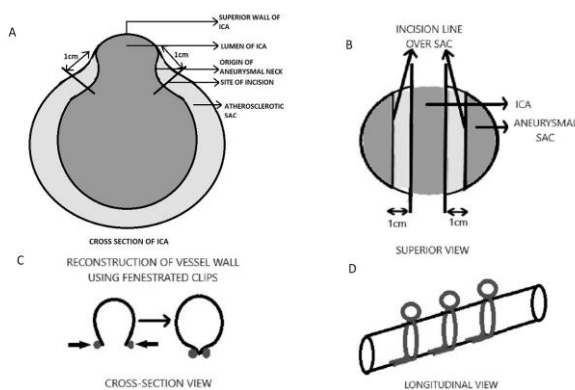


Figure 4. Sequential Artistic view of reconstruction of ICA in giant atherosclerotic communicating segment ICA aneurysm.

CONCLUSIONS

Intracranial aneurysms are challenging to manage due to their proximity to vital intracranial structures, the skull base, visual apparatus and difficulty in securing proximal control intra-cranially. They can be successfully clipped if the intimate anatomical relations of the aneurysm like the optic nerve, anterior clinoid process; optic strut, the dural ring and adjacent vessels are anatomically defined properly during surgery. Thorough knowledge of intracranial anatomy, adjacent relations, arachnoid planes and skillful dissection is a key element to successful outcome. Data collected from GGSMC & Hospital may not be representative of the entire state or country's population. Therefore, a larger-scale data collection is necessary to create our own database to ascertain the risk factors and preventive measures that are exclusive to our state and nation.

REFERENCES

1. Ailani AG, Saiful Azli MN, Regunath K, et al. Characteristics and Outcomes of Patients with anterior circulation intracranial aneurysm managed with clipping in Hospital Sungai Buloh. *Malaysian J Med Sci* 2016 Nov;23(6):113-117.
2. Barrow DL, Alleyne C. Natural history of giant intracranial aneurysms and indications for intervention. *Clin Neurosurg* 1995;42:214-244.
3. Batjer HH, Chandler JP, Getch CC, et al. Intracranial aneurysm. In: Rengachary SS, Ellenbogen RG, eds. *Principles of Neurosurgery*. Edinburgh: Elsevier Mosby Publishing; 2005:215-239.
4. Bharatha A, Yeung R, Durant D, et al. Comparison of computed tomography angiography with digital subtraction angiography in the assessment of clipped intracranial aneurysms. *J Comput Assist Tomogr* 2010;34(3):440-445.
5. Brown RD Jr, Broderick JP. Unruptured intracranial aneurysms: Epidemiology, natural history, management options, and familial screening. *Lancet Neurol* 2014;13:393-404.
6. Gasparotti R, Liserre R. Intracranial aneurysms. *Eur Radiol* 2005 Mar;15(3):441-447.
7. Gobble RM, Hoang H, Jafar J, et al. Extracranial-intracranial bypass: Resurrection of a nearly extinct operation. *J Vasc Surg* 2012;56(5):1303-1307.
8. Ingall T, Asplund K, Mahonen M, et al. A multinational comparison of subarachnoid hemorrhage epidemiology in the WHO MONICA stroke study. *Stroke* 2000;31(5):1054-1061.
9. Juvela S. Natural history of unruptured intracranial aneurysms: risks for aneurysm formation, growth, and rupture. *Acta Neurochir Suppl* 2002;82:27-30.
10. Juvela S. Prehemorrhage risk factors for fatal intracranial aneurysm rupture. *Stroke* 2003;34:1852-1858.
11. Karasawa H, Matsumoto H, Naito H, et al. Angiographically unrecognized microaneurysms: Intraoperative observation and operative technique. *Acta Neurochir (Wien)* 1997;139(5):416-419.
12. Lonjon M, Pennes F, Sedat J, et al. Epidemiology, genetic, natural history and clinical presentation of giant cerebral aneurysms. *Neurochirurgie* 2015;61(6):361-365.
13. Millán RD, Dempere-Marco L, Pozo JM, et al. Morphological Characterization of Intracranial Aneurysms Using 3-D Moment Invariants. *IEEE Trans Med Imaging* 2007;26(9):1270-1282.
14. Pasqualin A. Epidemiology and pathophysiology of cerebral vasospasm following subarachnoid hemorrhage. *J Neurosurg Sci* 1998;42:15-2.
15. Rinkel GJ, Djibuti M, Algra A, et al. Prevalence and risk of rupture of intracranial aneurysms: A systematic review. *Stroke* 1998;29(1):251-256.
16. Samandouras G. *The Neurosurgeon's Handbook*. Oxford University Press; 2010.
17. Sekhar LN, Duff JM, Kalavakonda C, et al. Cerebral revascularization using radial artery grafts for the treatment of complex intracranial aneurysms:

- techniques and outcomes for 17 patients. *Neurosurgery* 2001;49(3):646-658.
18. Van Gijn J, Kerr RS, Rinkel GJ. Subarachnoid haemorrhage. *Lancet* 2007;369(9558):306-318.
 19. Vlak MH, Algra A, Brandenburg R, et al. Prevalence of unruptured intracranial aneurysms, with emphasis on sex, age, comorbidity, country, and time period: systematic review and meta-analysis. *Lancet Neurol* 2011;10(7):626-636.
 20. W. Schievink. Intracranial aneurysms. *New Eng. J. Med* 1997;336:28-41.
 21. Wardlaw JM, White PM. The detection and management of unruptured intracranial aneurysms. *Brain* 2000;123:205-221.
 22. Weir B, Grace M, Hansen J, et al. Time Course of Vasospasm in Man. *J Neurosurg* 1978;48:173-178.
 23. Woo D, Broderick J. Genetics of intracranial aneurysm. *J Stroke Cerebrovascular Dis* 2002;11(5):230-240.
 24. Yahia AM, Gordon V, Whapham J, et al. Complications of neuroform stent in endovascular treatment of intracranial aneurysms. *Neurocrit Care* 2008;8(1):19-30.
 25. Zhao J, Lin H, Summers R, Yang M, et al. Current Treatment Strategies for Intracranial Aneurysms: An Overview. *Angiology* 2018;69(1):17-30.



Endoscopic third ventriculostomy for obstructive hydrocephalus

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ABSTRACT

Introduction: Obstructive hydrocephalus has long been managed by valve-regulated shunts. These shunts are associated with a myriad of short and long-term complications. This has fuelled interest in Endoscopic Third Ventriculostomy (ETV) which provides a more physiological pathway for cerebrospinal fluid (CSF) diversion while avoiding many shunt-related complications.

Aim: The objective of this study is to analyse the outcomes of ETV at our institution, focusing on the indications, success rates, and short-term complications.

Methods: Between July 2010 and September 2015, 47 patients with obstructive hydrocephalus underwent ETV at the Neurosurgery Teaching Hospital in Baghdad/Iraq. We retrospectively analysed the data of these patients using hospital health records. Simple statistics were performed using SPSS Version 20. A standardized surgical technique was employed in all cases.

Results: The mean age was 4.4 years (range 40 days - 38 years). The male: female ratio was 1.23:1 (55% males and 45% females). The most common cause of obstructive hydrocephalus in patients undergoing ETV was aqueductal stenosis (62%; N=29). The second most common cause was posterior fossa tumours (23%; N=11). The overall success rate for ETV was 68%. The net post-operative complication rate was 13% (N=6). CSF leak and seizures were the only two charted post-operative complications at the rates of 9%(N=4) and 4% (N=2), accordingly. No deaths were recorded.

Conclusion: ETV is a viable alternative to shunt insertion in a select group of patients with obstructive hydrocephalus, with acceptable success rate and safety profile.

INTRODUCTION

Hydrocephalus is a condition that results when excess cerebrospinal fluid (CSF) accumulates in the cerebral ventricles resulting in ventricular dilatation and raised intracranial pressure (Lu, Chen, Weng, & Xu, 2019). Hydrocephalus is divided into communicating and non-communicating

Keywords
endoscopic third
ventriculostomy,
obstructive hydrocephalus,
shunts,
CSF



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(Aka. obstructive) subtypes, as per Dandy's classification (Lu et al., 2019). Non-physiological CSF diversion using valve-regulated ventriculo-peritoneal and ventriculo-atrial shunts has dominated the field of hydrocephalus management over the past 45 years. However, shunts come with many complications including infections, displacement, and hardware failure. Failure rates are reported at 25-40 % in the first year then 5-10% per year thereafter (Deopujari, Karmarkar, & Shaikh, 2017). The significant morbidity imposed by these shunts and the need for frequent revisions has warranted the search for novel treatment options (Mugamba & Stagno, 2013).

First introduced by mixer in 1923, Endoscopic Third Ventriculostomy (ETV) has regained popularity in the last two decades as an attractive treatment option for hydrocephalus (Mixer, 1923). In fact, ETV for the treatment of obstructive hydrocephalus is currently the most commonly performed endoscopic neurosurgical procedure (M. Gangemi et al., 1999) (Hellwig et al., 2005). Desirable qualities of ETV include 1) its physiological mechanism of CSF diversion, 2) its minimally invasive nature and 3) the absence of long-term hardware and all its associated complications (Mugamba & Stagno, 2013).

Since its advent in the early 1990s, ETV has shown the highest success rate amongst patients with obstructive hydrocephalus caused by aqueductal stenosis, both congenital and acquired (Mugamba & Stagno, 2013). Nonetheless, both the surgical techniques and indications for ETV are constantly being refined. In the last few decades, many authors have been calling to expand the indications for ETV and numerous reports have been published to support this notion. (Beems & Grotenhuis, 2004) (Brockmeyer, Abtin, Carey, & Walker, 1998) (Choi, Kim, & Kim, 1999) (Cinalli et al., 1999) (Feng et al., 2004) (Grunert, Charalampaki, Hopf, & Filippi, 2003) (Scarrow, Levy, Pascucci, & Albright, 2000) (Schroeder, Niendorf, & Gaab, 2002) (Tisell, Almström, Stephensen, Tullberg, & Wikkelso, 2000).

In their systematic review and meta-analysis that compared the failure rates of obstructive hydrocephalus vs shunt insertion for patients with non-communicating hydrocephalus, Rasul et al concluded that ETV has statistically non-significant advantage over shunts; it does, however, appear to provide long-term survival benefit specifically in

patients with congenital aqueductal stenosis (Rasul, Marcus, Toma, Thorne, & Watkins, 2013).

In Iraq, ETV is still a relatively novel modality. In this paper, we aim to report our experience with ETV in the management of patients with obstructive hydrocephalus, which is one of the earliest reports in this field from our country.

PATIENTS AND METHODS

We conducted a retrospective analysis of a consecutive sample of 47 patients who underwent ETV for obstructive hydrocephalus at the Neurosurgery Teaching Hospital in Baghdad (NTH) for the period between July 2010 and September 2015. The aim of this study was to determine the success rate of ETV at our institution and to identify the main complications. A clinicoradiological criterion was implemented in our study. Patients were deemed candidates for ETV if they 1) presented with symptoms related to hydrocephalus such as raised ICP or enlarging head circumference and 2) had an MRI-confirmed diagnosis of obstructive hydrocephalus. Patients with communicating hydrocephalus were excluded from the study. The follow-up period ranged between six months and six years (6 months-6 years). The success of ETV was defined by a combination of clinical and radiological criteria and ultimately by "shunt-independence". The clinical criteria revolved around the resolution of symptoms and signs caused by the high intracranial pressure by the 3-month follow-up visit. The radiological criteria were defined as the decrease in ventricular size or the arrest of ventriculomegaly as determined by Ultrasonography (US) and/or Computed Tomography (CT), using the Evans Index or the fronto-Occipital horn ratio. All data were obtained from the hospital health records at the NTH and statistics were performed using SPSS Version 20.

OPERATIVE TECHNIQUE

All operations were performed under general anesthesia. During each surgery, the following steps were followed:

- The patient was placed in a supine position with the head resting on a doughnut-shaped head rest.
- The head and neck were flexed at (15°).
- A Longitudinal skin incision was made just in front of coronal suture to the right of the mid-pupillary

line (3 cm off mid line & 1cm anterior to coronal suture).

- The skin and periosteum were opened as a single layer with visual identification of coronal suture.
- A burr hole was drilled abutting the anterior border of coronal suture.
- The dura was coagulated and retracted in a cruciate fashion using a bipolar.
- The pia and arachnoid were coagulated and opened sharply in a cruciate fashion.
- The articulated arm of the endoscope was installed as close as possible to the patient's head.
- Warm ringer lactate solution was connected to the sheath at a height of 15 cm from the patient's ear.
- A Rigid sheath (8mm) with its trocar inside was advanced slowly into the brain tissue aiming the tip of the sheath at the inner canthus in mediolateral plane and towards the tragus in antero-posterior plane.
- A sudden give up was used as an indication that the ventricle has been punctured. The trochar was then removed.
- A telescope with an attached light source and camera (3S) was introduced inside the sheath and locked in the zero position.
- The Foramen of Monro was identified, the arm was released and the sheath was introduced through foramen and placed just above the mammillary body.
- The thinned floor of the 3rd ventricle at the region of the tuber cinereum was identified and punctured using the fenestration forceps.
- The Ringer solutions was allowed to circulate in and out to verify the stoma opening.
- The endoscope was then withdrawn slowly while visualising the entry track.
- Cylindrical pieces of Gelfoam were placed in the track and on the burr hole followed by subcutaneous tissue and skin closure.

RESULTS

Our sample contained a heterogeneous group of patients in relation to the national geographic distribution, age range, and hydrocephalus etiology. The mean age was 4.4 years (range 40 days -38 years). The male: female ratio was 1.23:1 (55% males and 45% females). The most common cause of obstructive hydrocephalus in patients undergoing ETV was aqueductal stenosis (62%; N=29). The

second most common cause was posterior fossa tumors (23%; N=11). Other aetiologies included third ventricle arachnoid cyst (11%; N=5), choroid plexus tumor of the lateral ventricle (2%; N=1) and thalamic tumors (2%; N=1). The overall success rate for ETV was 68%. Subgroup analysis showed that success rates were 62% and 78% for patients with aqueductal stenosis and posterior fossa tumors, respectively. Regarding the age group, the success rate for the younger population (below 10 years) was (62-65%). In the subset of patients with unclear cerebrospinal fluid (CSF), the success rate was 22%. No intraoperative complications were encountered. The net post-operative complication rate was 13% (N=6) and spontaneous closure was charted in 32% (N=15). CSF leak and seizure were the only two recorded postoperative complications at the rates of 9%(N=4) and 4% (N=2), accordingly. The mortality rate was 0% (N=0). None of the patients needed redo ETV or shunt insertion as per their corresponding last follow-up visits.

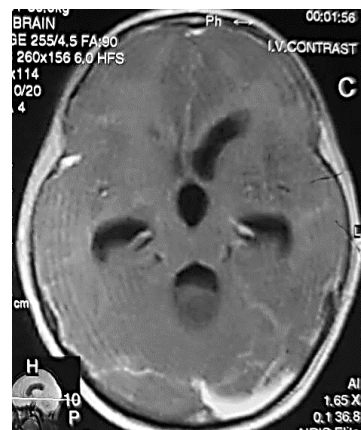


Figure 1.
A. nine-year-old female with obstructive hydrocephalus due to 4th ventricular tumor (histopathologically proved WHO grade 4 Ependymoma). A: Pre-ETV axial T1-weighted brain MRI with contrast showing hydrocephalus with ballooning of the 3rd ventricle and dilatation of both the frontal and temporal horns of lateral ventricles.



B: Early post-ETV brain CT scan (axial section) showing improvement of ventricular dilation with normal shape and size of the 3rd ventricle and the frontal horn of lateral ventricles.

DISCUSSION

ETV is an important treatment option in the management of hydrocephalus. In our cohort of 47 patients with obstructive hydrocephalus, the two most common indications for ETV were aqueductal stenosis and posterior fossa tumors; this corresponds to the results published by Bouras *et al.* in their systematic review and meta-analysis (T. Bouras & S. Sgouros, 2012).

Recently, ETV has replaced extra-cranial shunts as the first-line treatment for occlusive hydrocephalus caused by aqueductal stenosis. Although the efficacy of ETV has been most established with the congenital forms of aqueductal stenosis, it is still a viable treatment option for other acquired aetiologies including those attributed to inflammatory and neoplastic processes (Hellwig *et al.*, 2005). This finding was also reflected in our results, wherein patients with aqueductal stenosis had the highest success rates.

ETV has also proven effective in the management of occlusive hydrocephalus in patients with posterior fossa tumors (Amano *et al.*, 2002) (Ruggiero *et al.*, 2004) (Cinalli *et al.*, 1999) (Schijman, Peter, ReKate, Sgouros, & Wong, 2004) (Sainte-Rose *et al.*, 2001). The incidence of pre-operative hydrocephalus in these patients is estimated at 80%, with 25-30% progressing to persistent post-operative hydrocephalus necessitating surgical intervention (Hellwig *et al.*, 2005). In their series of 206, Sainte-Rose *et al.* concluded that when performed prior to craniotomy, ETV reduced the incidence and progression of post-operative hydrocephalus (Sainte-Rose *et al.*, 2001). However, there exists no consensus regarding the timing and the definitive treatment modality to be used in the management of these patients (Hellwig *et al.*, 2005).

Since its advent in the late 1980s and early 1990s, much experience has been gained with ETV as a management tool in patients with obstructive hydrocephalus. On the other hand, the efficacy of ETV in communicating hydrocephalus has not been scientifically established and is currently under investigation, with some authors reporting encouraging results (Hailong *et al.*, 2008) (Meier, Zeilinger, & Schönherr, 2000) (Michelangelo Gangemi, Maiuri, Buonamassa, Colella, & de Divitiis, 2004). At our institution, patients with communicating hydrocephalus are not currently considered candidates for ETV.

To date, patient selection criteria for ETV is a debatable issue in the neurosurgical community; *i.e.* there is no clear definition of "ETV-eligible" patients and patient selection is currently based on local institutional policies, individual surgeon's experience and patient preference. (M. Gangemi *et al.*, 1999) (Triantafyllos Bouras & Spyros Sgouros, 2012).

Evaluation of ETV outcome has been mainly based on a clinico-radiological criteria. Clinical criteria focus on symptomatic improvement and shunt-independence while the radiological criteria centers on the reduction of the ventricular size and/or cessation of its growth (Buxton, Turner, Ramli, & Vloeberghs, 2002) (Deopujari *et al.*, 2017). However, some recent evidence tells us that the change in ventricular size is an unreliable indicator, especially in the early postoperative period. (Schwartz, Yoon, Cutruzzola, & Goodman, 1996) (Feng *et al.*, 2004). Success rates with ETV have been reported to be between 50-90% (Hellwig *et al.*, 2005). Beams and Grotenhuis have reported the largest series thus far on patients with ETV (339 cases). In the aforementioned series, shunt independence was charted in 258 patients, with a complication rate of 7.7%. (Beems & Grotenhuis, 2004).

Regarding factors that predict poor outcomes after ETV, a history of shunt infection and post-operative meningitis have been identified as independent risk factors for ETV failure (Fukuhara, Vorster, & Luciano, 2000). None of our patients harbored either risk factor.

Both the immediate and long-term complications associated with ETV are currently insufficiently reported in the literature to draw solid conclusions. There exist many inconsistencies in the reported intra-operative, immediate and delayed postoperative complications. In one systematic review, the mortality rate was reported as 0.22% and the permanent morbidity rate was 2.1%. The most common charted complications were CSF infections (1.8%) and CSF leak (1.7%) (T. Bouras & S. Sgouros, 2012). Further studies have also reported mortality rates below 1% (Cohen, 1994; M. Gangemi *et al.*, 1999; Jones, Kwok, Stening, & Vonau, 1994; Teo & Jones, 1996). In our study, no deaths were recorded as per the last follow-up visit for each patient.

The obvious shortcomings in our study are 1) the short follow-up period and 2) the small cohort size. In fact, these two limitations are shared by the majority of the related reports in the literature, an

observation that highlights the need for multi-center, prospective, long-term studies to elucidate the long-term outcomes of ETV.

CONCLUSION

ETV is a promising treatment strategy for patients with obstructive hydrocephalus. However, many questions remain unanswered in relation to its long-term efficacy and safety, calling for further research in this area.

REFERENCES

- Amano, T., Inamura, T., Nakamizo, A., Inoha, S., Wu, C.-M., & Ikezaki, K. (2002). Case management of hydrocephalus associated with the progression of childhood brain stem gliomas. *Child's nervous system : ChNS : official journal of the International Society for Pediatric Neurosurgery*, 18(11), 599-604. doi:10.1007/s00381-002-0637-5.
- Beems, T., & Grotenhuis, J. A. (2004). Long-term complications and definition of failure of neuroendoscopic procedures. *Child's nervous system : ChNS : official journal of the International Society for Pediatric Neurosurgery*, 20(11-12), 868-877. doi:10.1007/s00381-004-0945-z.
- Bouras, T., & Sgouros, S. (2012). Complications of endoscopic third ventriculostomy: a systematic review. *Acta neurochirurgica. Supplement*, 113, 149-153. doi:10.1007/978-3-7091-0923-6_30.
- Bouras, T., & Sgouros, S. (2012). Complications of endoscopic third ventriculostomy: a systematic review. *Acta Neurochir Suppl*, 113, 149-153. doi:10.1007/978-3-7091-0923-6_30.
- Brockmeyer, D., Abtin, K., Carey, L., & Walker, M. L. (1998). Endoscopic third ventriculostomy: an outcome analysis. *Pediatric neurosurgery*, 28(5), 236-240. doi:10.1159/000028657.
- Buxton, N., Turner, B., Ramli, N., & Vloeberghs, M. (2002). Changes in third ventricular size with neuroendoscopic third ventriculostomy: a blinded study. *Journal of neurology, neurosurgery, and psychiatry*, 72(3), 385-387. doi:10.1136/jnnp.72.3.385.
- Choi, J. U., Kim, D. S., & Kim, S. H. (1999). Endoscopic surgery for obstructive hydrocephalus. *Yonsei medical journal*, 40(6), 600-607. doi:10.3349/ymj.1999.40.6.600.
- Cinalli, G., Sainte-Rose, C., Chumas, P., Zerah, M., Brunelle, F., Lot, G., Renier, D. (1999). Failure of third ventriculostomy in the treatment of aqueductal stenosis in children. *Journal of neurosurgery*, 90(3), 448-454. doi:10.3171/jns.1999.90.3.0448.
- Cohen, A. R. (1994). Ventriculoscopy. *Clinical neurosurgery*, 41, 546-562.
- Deopujari, C. E., Karmarkar, V. S., & Shaikh, S. T. (2017). Endoscopic Third Ventriculostomy: Success and Failure. *Journal of Korean Neurosurgical Society*, 60(3), 306-314. doi:10.3340/jkns.2017.0202.013.
- Feng, H., Huang, G., Liao, X., Fu, K., Tan, H., Pu, H., ... Zhao, D. (2004). Endoscopic third ventriculostomy in the management of obstructive hydrocephalus: an outcome analysis. *Journal of neurosurgery*, 100(4), 626-633. doi:10.3171/jns.2004.100.4.0626.
- Fukuhara, T., Vorster, S. J., & Luciano, M. G. (2000). Risk factors for failure of endoscopic third ventriculostomy for obstructive hydrocephalus. *Neurosurgery*, 46(5), 1100-1111. doi:10.1097/00006123-200005000-00015.
- Gangemi, M., Donati, P., Maiuri, F., Longatti, P., Godano, U., & Mascari, C. (1999). Endoscopic third ventriculostomy for hydrocephalus. Minimally invasive neurosurgery: *MIN*, 42(3), 128-132. doi:10.1055/s-2008-1053384.
- Gangemi, M., Maiuri, F., Buonamassa, S., Colella, G., & de Divitiis, E. (2004). Endoscopic third ventriculostomy in idiopathic normal pressure hydrocephalus. *Neurosurgery*, 55(1), 129-134. doi:10.1227/01.neu.0000126938.12817.dc.
- Grunert, P., Charalampaki, P., Hopf, N., & Filippi, R. (2003). The role of third ventriculostomy in the management of obstructive hydrocephalus. Minimally invasive neurosurgery: *MIN*, 46(1), 16-21. doi:10.1055/s-2003-37957.
- Hailong, F., Guangfu, H., Haibin, T., Hong, P., Yong, C., Weidong, L., & Dongdong, Z. (2008). Endoscopic third ventriculostomy in the management of communicating hydrocephalus: a preliminary study. *J Neurosurg*, 109(5), 923-930. doi:10.3171/JNS/2008/109/11/0923.
- Hellwig, D., Grotenhuis, J. A., Tirakotai, W., Riegel, T., Schulte, D. M., Bauer, B. L., & Bertalanffy, H. (2005). Endoscopic third ventriculostomy for obstructive hydrocephalus. *Neurosurgical review*, 28(1), 1-38. doi:10.1007/s10143-004-0365-2.
- Jones, R. F., Kwok, B. C., Stening, W. A., & Vonau, M. (1994). Neuroendoscopic third ventriculostomy. A practical alternative to extracranial shunts in non-communicating hydrocephalus. *Acta neurochirurgica. Supplement*, 61, 79-83. doi:10.1007/978-3-7091-6908-7_14.
- Lu, L., Chen, H., Weng, S., & Xu, Y. (2019). Endoscopic Third Ventriculostomy versus Ventriculoperitoneal Shunt in Patients with Obstructive Hydrocephalus: Meta-Analysis of Randomized Controlled Trials. *World neurosurgery*, 129, 334-340. doi:10.1016/j.wneu.2019.04.255.
- Meier, U., Zeilinger, F. S., & Schönherr, B. (2000). Endoscopic ventriculostomy versus shunt operation in normal pressure hydrocephalus: diagnostics and indication. Minimally invasive neurosurgery: *MIN*, 43(2), 87-90. doi:10.1055/s-2000-8325.
- Mixter, W. J. (1923). *Ventriculoscopy and Puncture of the Floor of the Third Ventricle*. *The Boston Medical and Surgical Journal*, 188(9), 277-278. doi:10.1056/N EJM192303011880909.
- Mugamba, J., & Stagno, V. (2013). Indication for endoscopic third ventriculostomy. *World neurosurgery*, 79(2 Suppl), S20.e19-S20.e23. doi:10.1016/j.wneu.2012.02.016.

23. Rasul, F. T., Marcus, H. J., Toma, A. K., Thorne, L., & Watkins, L. D. (2013). Is endoscopic third ventriculostomy superior to shunts in patients with non-communicating hydrocephalus? A systematic review and meta-analysis of the evidence. *Acta neurochirurgica*, 155(5), 883-889. doi:10.1007/s00701-013-1657-5.
24. Ruggiero, C., Cinalli, G., Spennato, P., Aliberti, F., Cianciulli, E., Trischitta, V., & Maggi, G. (2004). Endoscopic third ventriculostomy in the treatment of hydrocephalus in posterior fossa tumors in children. *Child's nervous system : ChNS : official journal of the International Society for Pediatric Neurosurgery*, 20(11-12), 828-833. doi:10.1007/s00381-004-0938-y.
25. Sainte-Rose, C., Cinalli, G., Roux, F. E., Maixner, R., Chumas, P. D., Mansour, M., . . . Renier, D. (2001). Management of hydrocephalus in pediatric patients with posterior fossa tumors: the role of endoscopic third ventriculostomy. *Journal of neurosurgery*, 95(5), 791-797. doi:10.3171/jns.2001.95.5.0791.
26. Scarrow, A. M., Levy, E. I., Pascucci, L., & Albright, A. L. (2000). Outcome analysis of endoscopic III ventriculostomy. *Child's nervous system : ChNS : official journal of the International Society for Pediatric Neurosurgery*, 16(7), 442-445. doi:10.1007/s00381000307.
27. Schijman, E., Peter, J. C., Rekate, H. L., Sgouros, S., & Wong, T. T. (2004). Management of hydrocephalus in posterior fossa tumors: how, what, when? *Child's nervous system : ChNS : official journal of the International Society for Pediatric Neurosurgery*, 20(3), 192-194. doi:10.1007/s00381-003-0900-4.
28. Schroeder, H. W. S., Niendorf, W.-R., & Gaab, M. R. (2002). Complications of endoscopic third ventriculostomy. *Journal of neurosurgery*, 96(6), 1032-1040. doi:10.3171/jns.2002.96.6.1032.
29. Schwartz, T. H., Yoon, S. S., Cutruzzola, F. W., & Goodman, R. R. (1996). Third ventriculostomy: post-operative ventricular size and outcome. *Minimally invasive neurosurgery: MIN*, 39(4), 122-129. doi:10.1055/s-2008-1052231.
30. Teo, C., & Jones, R. (1996). Management of hydrocephalus by endoscopic third ventriculostomy in patients with myelomeningocele. *Pediatric neurosurgery*, 25(2), 57-63. doi:10.1159/000121098.
31. Tisell, M., Almström, O., Stephensen, H., Tullberg, M., & Wikkelsö, C. (2000). How effective is endoscopic third ventriculostomy in treating adult hydrocephalus caused by primary aqueductal stenosis? *Neurosurgery*, 46(1), 104-111.



Intraneural synovial sarcoma of median nerve. A rare case report with review of literature

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ABSTRACT

Synovial sarcomas are highly aggressive soft tissue tumour with a poor and dismal prognosis. These tumours have a high propensity for distant metastasis and local recurrence. Although originally believed to arise from synovium, these tumours have been found to occur anywhere in body [1],[2]. We report here, a case of median nerve sarcoma in a 15-year female. This is a rare tumour, which is diagnosed only after histopathological examination with only a few cases reported in the literature (Table 1). Although preoperatively tumour was thought to be a nerve sheath tumour, on histopathology analysis was found to be synovial sarcoma. Despite aggressive behaviour, wide local excision is recommended even in smaller lesions. So, the diagnosis should always be kept in differentials of nerve sheath tumour, as what may be a synovial sarcoma.

INTRODUCTION

Synovial sarcomas may arise from different and unusual sites with distinctive morphological genetic features [3]. They are mostly seen in extremities in young adolescents with male preponderance [4]. It has been found in unusual locations in heart, lung, small intestine, soft palate and peripheral nerves. Only a few cases have been reported in peripheral nerve. Prognosis is poor despite radical surgery, radiation and chemotherapy with 50-60 % survival [5].

These sarcomas have origin in synovium because of periarticular location, but less than 5% are continuous with synovium⁴. Sarcomas have their origin from primitive mesenchymal undifferentiated cells⁶. Synovial sarcoma is diagnosed on immunohistochemical basis because most of them present as lump or swelling with no clinical or diagnostic features [7]. Translocation (X;18) is diagnostic in 90% of cases [8]. The case presented here is a rare sarcoma arising from median nerve in upper arm. We have described here clinical, radiological features and its management.

CASE PRESENTATION

A 15 years old female presented with swelling in left arm for last 6

Keywords

intraneural synovial sarcoma,
median nerve,
arm,
soft tissue tumour



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months with swelling of size of 5*4 cm in left arm which has gradually increased in size. She also complained of pain in left arm radiating along medial aspect of forearm up to left hand. The pain was sharp, and more during night. She also complained of tingling sensation in left upper limb, palm, index and thumb and had history of dropping objects from left hand. On examination, there was decreased sensation along radial three digits with no neurovascular deficit.

The patient was evaluated with Magnetic resonance imaging (Figure 1) which revealed a well encapsulated oval lesion in left upper arm medially burrowing in left biceps and coracobrachialis muscle. The lesion minimally indented the left brachial artery. The lesion was in continuity with median nerve which showed mild enhancement in early and late arterial phase with heterogenous enhancement with non-enhancing / cystic areas in venous and delayed phase. Overall findings were in favour of neurogenic tumour.

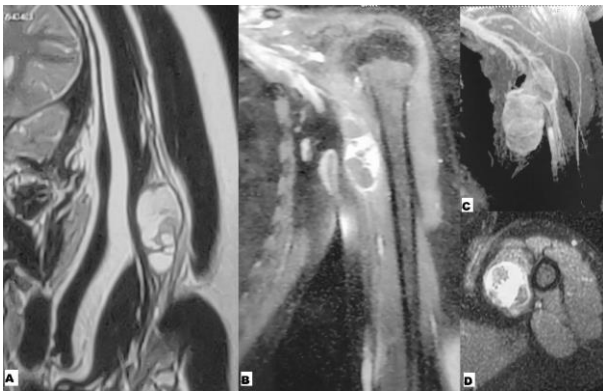


Figure 1.

Electrodiagnostic studies and nerve conduction studies of median nerve were within normal limits. Ultrasound colour Doppler peripheral venous single upper limb was suggestive of hetero-echoic lesion with internal cystic components measuring 43*23 mm causing compression of underlying axillary artery medially at its lower margin. The patient was taken up for surgical resection with preservation of nerve fascicles. The mass found to be intimately associated with nerve, soft in consistency and moderately vascular in nature. Microsacral excision of mass was done with preservation of nerve fascicles (Figure 2).

On histopathological analysis, tumour was greyish tan made up of spindle cells with sheets of

cells with vesicular nuclei and ill-defined cell margins suggestive of mild epithelioid morphology (Figure 3). Focal areas showed perivascular arrangement with cells separated by blood vessels with occasional mitosis and areas of haemorrhage. Collagen and osseous tissue was seen adjacent to tumour. On immunohistochemistry CD 99 was positive, CK negative, Synaptophysin negative, S-100 negative, HMB-45 negative, BCL2 positive and Ki-67 was 15-20 % in cellular areas (Figure 4). The histopathological and immunohistochemical analysis were in favour of synovial sarcoma. Post operatively, whole body PET/CECT scan was done for any metastasis and restaging. This was suggestive of small minimally metabolic active solid soft tissue thickening in proximal left biceps muscle indenting left brachial artery probably? residual lesion. Post excision, patient received radiotherapy.

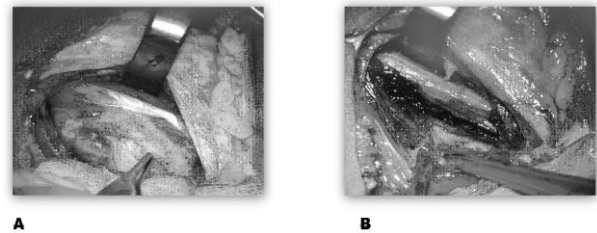


Figure 2.

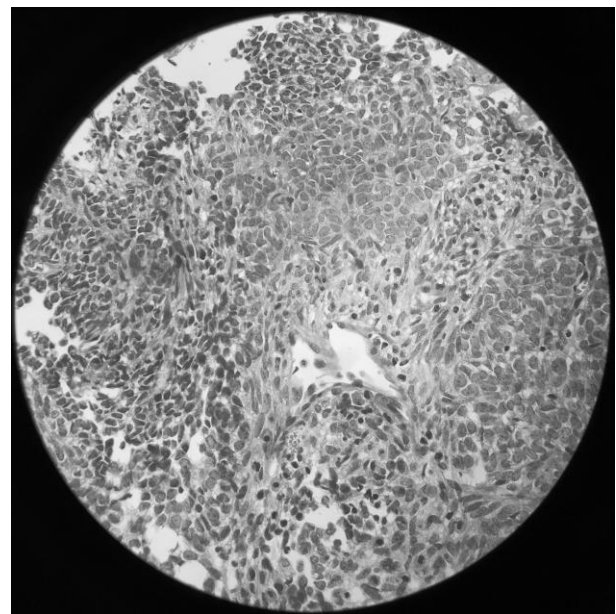


Figure 3.

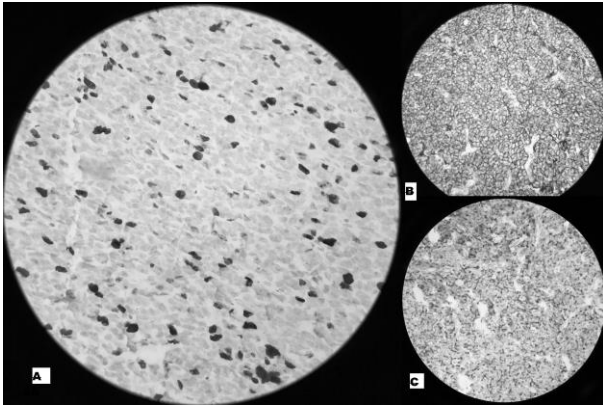


Figure 4.

DISCUSSION

Synovial sarcomas arising from median nerve are very rare and rarest tumours published in literature. Other sarcomas arising from various nerves reported in literature are from radial, common digital, posterior tibialis, peroneal, facial nerve, S1 root, C7 root and brachial plexus. There are three basic histology features of synovial sarcoma. The most common pattern is biphasic followed by monophasic and poorly differentiated synovial sarcomas. Synovial sarcoma with biphasic type consisted of epithelial cells with fibroblast like spindle cell in various proportions. Monophasic type consists of spindle cells only and are difficult to differentiate from other soft tissue and nerve tumours but now can be differentiated on the basis of immunohistochemical analysis⁴. Third pattern of poorly differentiated synovial sarcomas that has worse prognosis and has also been described⁶. Prognosis of biphasic versus monophasic is still under debate^{9,10,11,12}.

The present case in our study was biphasic synovial sarcoma having both epithelial and spindle cells in various proportions. It was immunohistochemical positive for CD99 and bcl2. The neoplastic proliferation with characteristic SS18-SSX1 with chromosomal fusion at molecular level is a feature consistent with diagnosis with synovial sarcoma⁴. Patient was advised for chromosomal analysis, but due to financial constraints patient refused for same. Published cases of median nerve synovial sarcoma with review of literature is summarised in table 1 and table 2 respectively.

Chromosomal translocation is the most efficient way to establish the diagnosis of synovial sarcoma. A gene translocation between chromosomes 18 and X

t(x;18) (p11.2; q11.2) occurs in over 90% of synovial sarcomas⁴. This leads to fusion of one of two variants of the SSX gene with the SYT gene, resulting in either the SYT/SSX1 or SYT/SSX2 chimeric fusion proteins^{4,6}. Nearly all biphasic tumours express SYT/SSX1, while monophasic tumours express SYT/SSX1 in approximately half of the cases and SYT/SSX2 in the remainder¹³. Patients with SYT/SSX2 expressing tumours have a significantly better prognosis when compared to those with SYT/SSX1 tumours in terms of rates of metastasis and overall survival^{13,14,15,16}.

Factors associated with poor prognosis includes old age, tumour size more than 5 cm, bone, nerve and vascular invasion and poor differentiation with higher mitotic index^{12,17,18,19}. Management of synovial sarcoma is wide surgical excision, radiation therapy, and adjuvant chemotherapy which has been found to be correlated to be associated with better prognosis^{2,20}

		thorax muscles	tumor					Additional	
Present series	15F		Median	MRI - Heterogeneous mild enhancing mass involving left arm	Intraneural tumor	4.3*2.3	Gross total excision	yes/no	Excision At present Undergoing Radiotherapy Good Functional recovery

M- Male, F- Female, CT - Computerized tomography, MRI - Magnetic resonance imaging

Table 2

LITERATURE REVIEW

SYNOVIAL SARCOMA OF NERVE- HISTOLOGY, IMMUNOHISTOCHEMISTRY, GENETICS

Published cases	Histologic type	EMA	Keratin	S100 protein	NFP showing nerve association	Miscellaneous	Genetics/PCR
Cugola and pisa	Biphasic	NA	NA	NA	NA		SYT-SSX1
Riechardt et al	Monophasic	Focal +	Focal +	Focal	Axons	Desmin focal	SYT-SSX2
O'connell et al	Biphasic	< 5 %	15-20% +	Neg	Axons	SMA,CD34-	X:18
Tacconi et al	Monophasic	+	+	Focal +	NA	-	NA
Speilmann et al	Biphasic	Focal	Neg	Neg	Axons	CD99+ MSA, Desmin	SYT-SSX2
Chesser et al	Biphasic	NA	+	NA	NA		SYT-SSX1
Zenmyo et al	Monophasic	+	+	NA	NA	bcl2+	T(x:18)(p11;q11) SYT-SSX-? Later
Lestou et al	Monophasic	Minor +	Minor+	Minor+	NA	Vimentin, CD99	Cryptic t(x:18), Ins (6:18) and SYT-SSX2 gene fusion
Chu et al Case 1	Biphasic	+ tumor and perineurium	+	Neg	Axons		t(x:18)(SYT-SSX)
Chu et al case 2	Monophasic	Neg	Neg	Neg nerve fibers +	Axons		t(x:18)(SYT-SSX)
Weinreb et al	Biphasic	+	+	Neg	Neg	Desmin,actin, CD 34	SYT-SSX1
Uehara et al	Monophasic	+	+	+	+		SYT-SSX1
Present series	Biphasic	NA	NA	Neg	-	CD 99 was positive, CK negative, Synaptophysin negative, S-100 negative,HMB-45 negative, bcl2 positive, Ki-67 was 15-20 %	NA

NA- Not assessed, NEG- negative, NFP-neurofilament protein,EMA-Epithelial membrane antigen,PCR-Polymerase chain reaction,+ Positive

Surgical management followed by radiation and chemotherapy is associated with good prognosis^{2,20}. 5-year survival rates of synovial sarcomas have been around 50-60%, in spite of advances in treatment^{2,21,22}. Mortality in synovial sarcoma results

from distant metastasis, most frequently to lung²³. Radical surgical incision followed by radiation allowed for potential decrease in local recurrence but systemic metastasis remained high even with adjunct chemotherapy²⁴.

CONCLUSION

Synovial sarcoma involving median nerve is a rare and aggressive tumour and is one of the few cases already published in literature. Synovial sarcoma can occur anywhere in our body and should be kept in differentials involving peripheral nerves as in our case. As diagnosis is always made post operative on histo-pathological analysis with immunohistochemistry, resulting in change in treatment strategy and final outcome of patient. As in our patient, preoperative diagnosis was a benign neurofibroma/schwannoma, we must be aware of aggressive tumour which overall changes the complete treatment and should be always be kept in differentials and managed accordingly.

REFERENCES

- Sandberg AA, Bridge JA. Updates on the cytogenetics and molecular genetics of bone and soft tissue tumors. Synovial sarcoma. *Cancer Genet Cytogenet.* 2002;133:1-23.
- Guadagnolo BA, Zagars GK, Ballo MT, et al. long term outcomes for synovial sarcoma treated with conservation surgery and radiotherapy . *Int J Radiat Oncol Biol Phys.* 2007;69:1173-80.
- Weiss SW, Goldblum JR: *Enzinger's and Weiss's soft tissue tumors*. 5th ed., Philadelphia, Mosby Elsevier, 2008, 1161-1182.
- Fischer C. Synovial sarcoma .*Ann Diagn Pathol.* 1998;2(2)401-21.
- Zeren H, Moran CA, Suster S, Fishback NF, Koss MN: Primary pulmonary sarcomas with features of monophasic synovial sarcomas. A clinicopathological, immunohistochemical and ultrastructural study of 25 cases. *Hum Pathol*; 26:474-480.
- Enzinger FM, Weiss SW. *Soft tissue tumors*. St. Louis: Mosby;1993. synovial sarcoma; pp.757-86.
- Kransdorf MJ. Malignant soft tissue tumors in a large referral population: distribution of diagnosis by age, sex and location. *Am J Roentgenol.* 1995;164:129-34.
- Palmerini E, Staals EL, Alberghini M, Zanella L, Ferrari C, Benassi MS, et al. Synovial sarcoma: retrospective analysis of 250 patients treated at a single institution. *Cancer.* 2009;115:2988-98.
- Chu PG, Benhattar J, Weiss LM, et al. Intraneural synovial sarcoma: two cases. *Mod Pathol.* 2004;17:258-63.
- Lewis JJ, Antonescu CR, Leung DH, et al. Synovial sarcoma: a multivariate analysis of prognostic factors in 112 patients with primary localized tumors of the extremity. *J Clin Oncol.* 2000;18:2087-94.
- Kawai A, Woodruff J, Healey JH, et al. SYT-SSX gene fusion as a determinant of morphology and prognosis in synovial sarcoma. *N Engl J Med.* 1998;338:153-60.
- Trassard M, Le Doussal V, Hacène K, et al. Prognostic factors in localized primary synovial sarcoma: a multicenter study of 128 adult patients. *J Clin Oncol.* 2001;19:525-34.
- Kawai A, Woodruff J, Healey JH, et al. SYT-SSX gene fusion as a determinant of morphology and prognosis in synovial sarcoma. *N Engl J Med.* 1998;338:153-60.
- Ladanyi M, Antonescu CR, Leung DH, et al. Impact of SYT-SSX fusion type on the clinical behavior of synovial sarcoma: a multiinstitutional retrospective study of 243 patients. *Cancer Res.* 2002;62:135-40.
- Inagaki H, Nagasaka T, Otsuka T, et al. Association of SYT-SSX fusion types with proliferative activity and prognosis in synovial sarcoma. *Mod Pathol.* 2000;13:482-8.
- Nilsson G, Skytting B, Xie Y, et al. The SYT-SSX1 variant of synovial sarcoma is associated with a high rate of tumor cell proliferation and poor clinical outcome. *Cancer Res.* 1999;59:3180-4.
- O'Connell JX, Browne WL, Gropper PT, et al. Intraneural biphasic synovial sarcoma: an alternative "glandular" tumor of peripheral nerve. *Mod Pathol.* 1996;9:738-41.
- Lewis JJ, Antonescu CR, Leung DH, et al. Synovial sarcoma: a multivariate analysis of prognostic factors in 112 patients with primary localized tumors of the extremity. *J Clin Oncol.* 2000;18:2087-94.
- Bergh P, Meis-Kindblom JM, Gherlinzoni F, et al. Synovial sarcoma: identification of low and high risk groups. *Cancer.* 1999;85:2596-607.
- Choong PFM, Pritchard DJ, Sim FH, et al. Long-term survival in high grade soft tissue sarcoma: prognostic factors in synovial sarcoma. *Int J Oncol.* 1995;7:161-9.
- Spillane AJ, A'Hern R, Judson IR, et al. Synovial sarcoma: a clinicopathologic, staging, and prognostic assessment. *J Clin Oncol.* 2000;18:3794-803.
- Krsková L, Kalinová M, Brízová H, et al. Molecular and immunohistochemical analyses of BCL2, KI-67, and cyclin D1 expression in synovial sarcoma. *Cancer Genet Cytogenet.* 2009;193:1-8.
- Enzinger FM, Weiss SW. *Soft tissue tumors*. St. Louis: Mosby; 1993. Synovial sarcoma; pp. 757-86.
- Lewis JJ, Antonescu CR, Leung DH, et al. Synovial sarcoma: a multivariate analysis of prognostic factors in 112 patients with primary localized tumors of the extremity. *J Clin Oncol.* 2000;18:2087-94.
- Cugola L, Pisa R. Synovial sarcoma: with radial nerve involvement. *J Hand Surg (Br)* 1985;10:243-4.
- Rinehart GC, Mustoe TA, Weeks PM. Management of synovial sarcoma of the median nerve at the elbow. *Plast Reconstr Surg* 1989;528-32.
- Tacconi L, Thom M, Thomas DG. Primary monophasic synovial sarcoma of the brachial plexus: report of case

- and review of literature. *Clin Neurol Neurosurg* 1996;98:249-52.
28. Spielmann A, Janzen DL, O'Connell JX, Munk PL. Intraneural synovial sarcoma. *Skeletal Radiol* 1997;26:677-81.
 29. Chesser TJ, Geraghty JM, Clarke AM. Intraneural synovial sarcoma of median nerve. *J Hand Surg(Br)* 1999;24:373-5.
 30. Zenmyo M, Komiya S, Hamada T, et al. Intraneural monophasic synovial sarcoma: a case report. *Spine* 2001;26:310-3.
 31. Lestou VS, O'Connell JX, Robinchaud M, et al. Cryptic t(X;18), ins(6;18), and SYT-SSX2 gene fusion in a case of intraneural monophasic synovial sarcoma. *Cancer Genet Cytogenet* 2002;138:153-6.
 32. Weinreb I, Perez-Ordóñez B, Guha A, Kiehl TR. Mucinous gland predominant synovial sarcoma of a large peripheral nerve : a rare case closely mimicking metastatic mucinous carcinoma. *J Clinical Pathol* 2008;61:672-6.
 33. Uehara H, Yamasaki K, Fukushima T, et al. Intraneural synovial sarcoma originating from median nerve. *Neurol Med Chir* 2008;48:77-82.



Preoperative evaluation of superficial cortical venous drainage*

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FULL TITLE: Preoperative evaluation of superficial cortical venous drainage around the lesion and localization of tumour boundaries preoperatively by applying cod liver capsule over the scalp with the help of MRI and MRV brain and comparison between 2-dimensional time-of-flight (2DTOF) and 3-dimensional contrast-enhanced magnetic resonance venography (3DCEMRV) brain in capsule marked cortical lesion and assessment of post-operative neurological outcomes.

ABSTRACT

Objectives and methodology: The preoperative exact localization of superficial intracranial lesions and superficial cortical veins is often necessary for making craniotomy and evaluation of cortical veins. We developed a simple and cheap method for such localization using cod liver oil capsule during the preoperative MRI and MRV brain examination. With the help of MRV brain, 3DCEMRV and 2DTOF images were taken and superficial cortical veins studied in the marked area for comparison between both modalities of MRV and planning of surgery for avoiding venous injury.

Results: Most of the cases were in the age group 16-60 years (91.6%). The most common clinical manifestation was headache (85.4%) and meningioma (60.4%) was found to be the most common pathology. Clear visualization (Grade 3) of the individual superficial cortical vein was observed in 48 cases (100%) in 3DCEMRV as compared to 2DTOF 22 cases (45.8%) $P < 0.0015$. Clear visualization (Grade3) of superior sagittal sinus was observed in 48 cases (100%) in 3DCEMRV as compared to 2DTOF 33 cases (68.6%) $P < 0.0015$. In post-operative CT Head, we found 4 (8.3%) cases were having venous infarction. 5 patients (10.4%) developed motor weakness postoperatively. In 3 cases, postoperative MRV were done and found no venous injury.

Conclusion: This study showed that preoperative localization and evaluation of the tumoral area and cortical veins with the help of cod liver oil in MRI and MRV brain was very helpful in planning the surgery, making craniotomy and to avoid injury of the veins. This technique is easy to perform and the capsule is easily constructed and inexpensive. 3DCEMRV was found to be better modality than 2DTOF for delineation of veins. Final neurosurgical outcomes were better.

INTRODUCTION

Imaging of intracranial cortical venous system anatomy is important in planning neurosurgical operations of midline masses such as colloid cyst, DACA Aneurysm, Corpus callosal gliomas, parasagittal and

Keywords

cod liver oil capsule,
2DTOF,
3DCEMRV,
MR venography,
midline lesions,
sagittal sinus,
superficial cortical vein



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parafalcine masses and other midline brain tumors. Parafalcine masses may not be as closely related to the SSS as parasagittal tumors, but because of their close relation, their growth may also contribute to anatomical changes to the nearby cortical veins. During parasagittal tumors resection, saving the cortical veins is important as it offers normal venous drainage of the brain, as well as important collateral drainage. Disruption in venous outflow may result in venous infarction which leads to swelling, hemorrhage, neuronal death and may have catastrophic result [1,2]. MR Venography is very helpful to know the anatomic changes in the cortical veins and its relation with respect to tumor position before surgery so that it can be saved intraoperatively. Assessment of the patency of sagittal venous sinuses is very important to avoid major sinus injury, air embolism and catastrophic bleeding intraoperatively. Moreover, cortical vessels especially the veins are important landmarks in craniotomy. With the help of 3DCEMRV, it has become possible to see the intracranial venous structures noninvasively. MR imaging technique with MIP method also presents more detailed information of brain surface structure by showing cortical veins [3,4]. The purpose of this study is to assess the usefulness of 3DCEMRV for the evaluation of intracranial venous system in preoperative surgical planning of brain tumors. In this study, we have compared the visibility of the intracranial venous system on 3DCEMRV with respect to those of 2DTOFMRV in surgical planning and compared it with intraoperative findings and also evaluated neurological outcomes postoperatively. In few cases post-operative MRV were done to look for any venous injury.

MATERIAL AND METHODS

Patient population

From May 2017 to Feb 2019, 48 patients were included which was admitted in department of neurosurgery Sawai Man Singh Medical College and Hospital, Jaipur. All patients underwent 3DCEMRV, as well as 2DTOFMRV in axial and sagittal planes.

Method

All the patients with midline supratentorial masses were selected on MRI brain images. After this cod liver oil capsules were placed on anterior and posterior ends of medial and lateral border of the

tumour at scalp according to location of tumor on MRI images and thus, the tumour boundaries were marked with the help of permanent marker. Then MRV 2-DTOF image were taken in axial and sagittal view. After that, 0.4ml/kg of gadolinium contrast agent was administered in cubital vein over 4 seconds and 3DCEMRV images were taken in axial and sagittal plane. All MR examinations were performed with 3-Tesla unit. All MR venographic source images were postprocessed with a maximum-intensity projection (MIP) algorithm to create projection venograms for both the 2DTOF MRV and the 3DCEMRV. All images were saved in DICOM software. The marked area concerned, superficial cortical veins such as frontopolar vein, anterior frontal vein, middle frontal vein, posterior frontal vein, precentral vein, central vein, post central vein, anterior parietal vein, posterior parietal vein and occipital vein and bridging veins joining the superior sagittal sinus were studied preoperatively. Sagittal sinus compression were also evaluated, which was defined as a narrowing and filling defect of the lumen. On the basis of these informations and surgeons preference the surgical corridor was decided. Intraoperative findings were compared with that of 3DCEMRV images and intraoperative photographs of cortical veins were taken. In few cases post operative MRV were done to look for any venous injury. All patients neurological status were assessed.

Inclusion Criteria

Conscious, co-operative and stable patients with supratentorial midline brain tumors.

Exclusion Criteria

Unconscious, uncooperative and unstable
Allergic to gadolinium
Ferromagnetic cardiac pacemakers, aneurismal clip and defibrillators.
Claustrophobia

Image analysis

Degree of visualization and patency of the intracranial venous system, superficial as well as superior sagittal sinus on 3DCEMRV was compared with those of 2DTOF in capsule marked tumoral area. The image quality of 8 predefined venous structures was graded as follows: intense and continuous = 3, faint and continuous = 2, noncontinuous = 1, and invisible = 0. In post

operative period, in few cases MRV were done to look for any venous injury.

Ethical consideration

The study protocol was approved by ethical Committee. All patient gave written informed consent to participate after having received full written information about the study objective and conducts. Investigations were done using aseptic precautions. They had right to withdraw from study. Protection was given from any kind of harm. Full confidentiality of data was maintained. No religious issues involved. All religious customs were respected. Study was conducted under supervision.

Statistical analyses

Statistical analyses were done using computer software (SPSS Trial version 23 and primer). The qualitative data were expressed in proportion and percentages and the quantitative data expressed as mean and standard deviations. The difference in proportion was analyzed by using chi square test. Significance level for tests were determined as 95% ($P < 0.05$).

RESULT

In our study, a total of 48 patients were included, in which 25 were males and 23 were females. All patients were conscious, oriented and Glasgow coma scale was 15/15 at the time of admission. Most of the cases were 16 -60 years (91.6%) of age, followed by elderly (>60 years) (4.4%) (Table no.1). Chi-square = 0.008 with 2 degrees of freedom; $P = 0.996$ NS. No significant difference was observed according to age groups and gender. No significant difference was observed in age among the male and females ($P=0.42$ NS). Among 48 cases, 20 were parafalcine and 28 were parasagittal . Majority of the cases were located in frontal lobe. Most common clinical manifestation were found to be headache (85.4%) followed by seizure (14.5%) and limb weakness (12.5%). We found 2 patients with diminution of vision and 2 cases of bladder disturbances (Table no.2). Most common histological diagnosis was found to be meningioma (60.4%) followed by glioma(29.1%) and epidermoid cyst (6.25%) with 1 case of colloid cyst and 1 case of AVM (Table no.3). All lesions (100%) were circumscribed precisely through small craniotomy.

Number of veins visualized in capsule marked area were as follows : Anterior frontal vein (12), Middle frontal vein (20), Posterior frontal vein (26), Central vein (11), Anterior parietal Vein (7), Posterior parietal vein (2), Occipital vein (1) and Superior Sagittal Sinus (48). The Mean grading score of imaging of individual superficial cortical veins were superior in 3DCEMRV as compared to 2DTOF (Table no. 4).

Poor depiction (Grade 2, 1, 0) of superficial cortical vein was observed in 26 cases (54.16%) in 2DTOF (N=48) as compared to 3DCEMRV (N=48)0 (0%). Whereas Clear depiction (Grade 3) of superficial cortical vein was observed in 48 cases (100%) in 3DCEMRV (N=48) as compared to 2DTOF (N=48) 22 case (45.8%) $P < 0.001$ S. (100%) showed in 3DCEMRV. In addition to this, we also found extra other small cortical vein drain the superior sagittal group in 7(14.5%) cases in 3DCEMRV as compared to 2DTOF. Poor depiction (Grade 2, 1, 0) of superficial sagittal sinus was observed 15 (31.2%) in 2DTOF (N=48) as compared to 3DCEMRV(N=48)0 (0%).whereas Clear depiction (Grade 3) of superior sagittal sinus was observed 48 (100%) in 3DCEMRV(N=48) as compared to 2DTOF (N=48)33 (68.6%) $P < 0.001$ S (Table no.5). 14 case (29.1%) showed sagittal sinus compression in both study groups. No new vein encountered during surgery. In post operative CT Head, we found 4 (8.3%) cases of infarction, 1 (2.0%) post operative site hematoma, and 3 (6.2%) cases of pneumocephalus. No residual mass was present in any cases. In the present study 5 patients (10.4%) developed neurological manifestations post operatively in the form of newly developed motor weakness, 4 patients (8.3%) developed altered sensorium and 1 patient died in post operative period, no patient developed seizure. In 3 cases, post operative MRV were done and these were analysed with respective preoperative images and no venous injury were identified.

Age group (years)	Total no. of patient	Male	Female
≤ 18	2(4.1%)	1(2.0%)	1 (2.0%)
18-60	44(91.6%)	23(47.9%)	21(43.7%)

≥ 60	2(4.1%)	1(2.0%)	1 (2.0%)
total	48(100%)	25(52%)	23(47.91%)
Mean±S	42.08±12.1	40.72±11.7	43.57±12.1
D	5	3	5

Table 1. Age and sex distribution.

Clinical symptoms	No. of cases
Headache	41 (85.4%)
Motor deficit	6 (12.5%)
seizure	7 (14.5%)
Diminution of vision	2 (4.1%)
Bladder disturbances	2 (4.1%)

Table 2. Clinical manifestations of patients.

Histological diagnosis	No. of cases
Meningioma	29 (60.4%)
Glioma	14 (29.1%)
Epidermoid cyst	3 (6.25%)
Colloid cyst	1 (2.0%)
AVM	1 (2.0%)
Total	48 (100%)

Table 3. Distribution of patients based on histological diagnosis.

Superficial cortical veins	Total number veins visualisation in capsule marked area in all	Mean grading score (2DTOF /3DCEMRV)	2D TOF /3DCEMRV (SUPERIOR)

	patients (N=48)		
Anterior frontal vein	12	2.29/3.0	3DCEMRV
Middle frontal vein	20	2.4/3.0	3DCEMRV
Posterior frontal vein	26	2.3/3.0	3DCEMRV
Central vein	11	2.36/3.0	3DCEMRV
Anterior parietal vein	7	2.51/3.0	3DCEMRV
Posterior parietal Vein	2	2.8/3.0	3DCEMRV
Occipital vein	1	3.0/3.0	NONE
Superior Sagittal Sinus	48	(2.6/3.0)	3DCEMRV

Table 4. Comparative study of image quality of individual superficial cortical veins in 2DTOF and 3DCEMRV according to mean grading score.

Veins	Poor image (Grade 2,1,0) 2DTOF	Clear image (Grade 3) 2D TOF	Poor image (Grade 2,1,0) 3DCEMRV	Clear image (Grade 3)

				3DCEMR v
Superficial cortical veins	26 (54.16%)	22 (45.8%)	0 (0%)	48 (100%)
Superior sagittal sinus	15(31.2%)	33 (68.7%)	0(0.0%)	48(100%)
Other small cortical vein seen	NONE	NONE	2(4.1%)	5(10.4%)
P Value (2D TOF / 3DCE MRV)	<0.001S	<0.001S	<0.001S	<0.001S

Table 5. Comparison of the image quality of superficial cortical veins and superior sagittal sinus in 2D TOF, 3DCEMRV.

Post operative clinical status of patients	No. of patients
Altered sensorium	4 (8.3%)
Newly developed motor deficit	5(10.4%)
Seizure	0(0%)
Death	1(2.0%)

Table 6. Post-operative neurological manifestations.

Post-operative CT Head	No. of patient
Venous infarction	4 (8.3%)
Post operative site hematoma	1 (2.0%)
Residual mass	0(0.0)
pneumocephalus	3(6.2%)

Table 7. Post-operative CT head.



Figure 1. Image showing preoperative tumour marking with help of cod liver oil capsule.

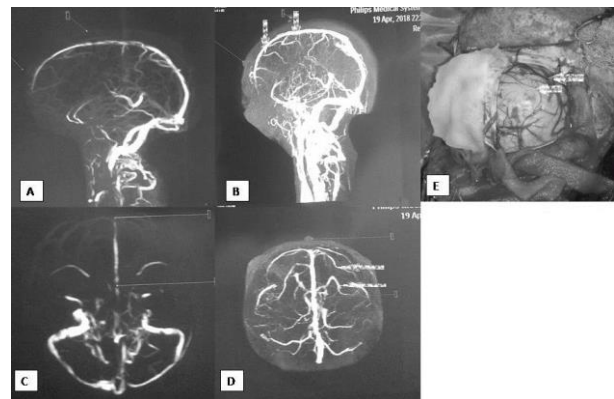


Figure 2. Comparison of 2D TOF and MR 3D CE MRV venography. Thick white arrow denotes superficial cortical veins (anterior and middle frontal vein) and thin arrow denote capsule in the anterior and posterior border of tumoral area. (A: Sagittal view; C: Axial view) 2D TOF MRV, (B: Sagittal view; D: Axial view) 3D CE MRV, showing clear visualization of anterior and middle frontal vein and entire superior sagittal sinus in 3DCEMRV. (E) Intraoperative photograph arrow showing

superficial cortical vein (anterior and middle frontal vein) in defined marked area.

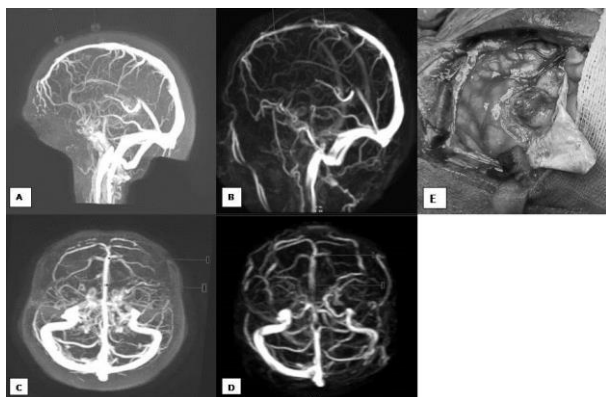


Figure 3. Comparison of preoperative and postoperative superficial cortical vein on MR venography. Thin arrow denote capsule in the anterior and posterior border of tumoral area. **(A: sagittal view; C: Axial view)** showing preoperative MRV and **(B: sagittal view; D: Axial view)** showing postoperative MRV, suggests that no superficial cortical venous injury in defined tumoral and peritumoral area. **(E);** Intraoperative photograph showing superficial cortical vein in defined marked area.

DISCUSSION

Exact localization of superficial cortical venous system anatomy with respect to midline masses is very important in planning neurosurgical operations. On the basis of the cortical area that they drain, the superficial cortical veins are divided into four groups of bridging veins: 1. superior sagittal group, which drains into the SSS; 2. sphenoidal group, which drains into the sphenoparietal and cavernous sinuses; 3. tentorial group, which converges on the sinuses in the tentorium; and 4. falcine group, which empties into the inferior sagittal sinus or straight sinus [5]. Surgery for midline masses such as colloid cyst, DACA Aneurysm, Corpus callosal gliomas, parasagittal and parafalcine masses are synonymous with dissection of the veins surrounding the tumour, specifically the cortical parasagittal and bridging veins, superior sagittal sinus (SSS) and collateral venous channels [6]. This information on intracranial venous system anatomy can be obtained by TOF MRV and CE MRV which are noninvasive techniques. Generally, MRV is performed without using contrast agent through 2D TOF MR venographic techniques [7]. 2D TOF MRV has been widely accepted for the imaging of intracranial venous system despite the well known technique associated pitfalls. A major pitfall of TOF MRV is the

artificial intravascular signal loss that occurs at predictable points in the intracranial venous anatomy [8]. Images of higher spatial resolution are obtained in 3DCE MRV with less scanning time than 2D TOF MRV [9]. Common flow related artifact seen on 2D TOF MRV can be avoided with the use of 3D CE MRV as it is flow insensitive [10]. False-negative results can also occur in 3D CE MRV in patients with enhancing intracranial lesions, such as neoplasms adjacent to dural sinus or veins. [11]. However, various literatures have reported that administration of contrast agent help to highlight these vessels especially small veins and also improve the vascular visualisation [12,13,14,15]. In our study 54.16% and 45.8% cases were having poor (grade 2, 1, 0) and clear (grade 3) depiction of superficial cortical vein respectively on 2D TOF whereas all cases (100%) showed clear (grade 3) depiction of studied vein in 3DCEMRV. Also, 68.7% and 100% cases showed clear depiction (grade 3) of superior sagittal sinus in 2D TOF and 3DCEMRV respectively. The mean grading score of imaging of individual superficial cortical veins were superior in 3DCEMRV as compared to 2D TOF. Thus the results of present study indicate better efficacy of 3DCEMRV over 2D TOF MRV. Study done by Leach et al; [16] also showed gadolinium-enhanced MRV to be superior than TOF MRV and also suggested the best evaluation using MRI. In the present study, the area over the tumor was marked by putting cod liver oil capsules, overlying veins were identified on 2D TOF MRV and 3DCEMRV image after that the surgical corridor was decided. Thus preoperative decision of surgical corridor allowed us to preserve the cortical veins maximally. In the study done by Burtscher et al. [17] have shown that 3-D image technique revealed additional information compared with conventional 2-D images and had an influence on neurosurgical planning and strategy, improving neurosurgical performance and patient outcome. In the present study also, additional other small cortical vein were found in 3DCEMRV in 7 (14.5%) cases as compared to 2D TOF. Information about tumour and overlying superficial cortical veins and sagittal sinus in the marked area are obviating the need for the surgeon to mentally reconstruct the surgical anatomy from 2-D image. Thus, orientation would be faster and more comprehensible [18,19,20]. With this information, the surgeon can plan the best approach for surgery. In the study by Khu et al; also concluded that

knowing the exact location of cortical vein with respect to tumor helps in preserving them during surgery. In our study, 2DTOF and 3DCEMRV images were taken and found that 2DTOF images showed less depiction of superficial cortical veins in the marked area as compared to 3DCEMRV, thus suggesting 3DCEMRV as preferred modality for locating veins. Abnormal radiological findings of intracranial venous structures were confirmed with intraoperative findings. In the study done by R.Klingeblia et al.[22];observed that Image quality was superior (4.3 ± 0.8 ; $P < 0.001$) for 3DCEMRV as compared with 2D TOF MRV (3.1 ± 0.7). In our study assessment of the evaluated sinus and veins was significantly improved by using 3DCEMRV ($P < 0.05$) as compared with 2D TOF MRV. Superior depiction of the cerebral venous anatomy on maximum intensity projection images from 3DCEMRV [22]. So 3DCEMRV is more informative than 2DTOF in delineation of superficial cortical and superior sagittal vein for surgical planning. and to avoid the venous injury while operating. All the patients were followed up and 5 (10.4%) patients developed neurological manifestations post operatively in the form of newly developed motor weakness, 4 (8.3%) patients developed altered sensorium and 1 patient died in post operative period. In post operative CT Head, we found 4 (8.3%) cases of infarction ,1 (2.0%) post op site hematoma, and 3 (6.2%) cases of pneumocephalus. No residual mass was present in any cases. In 3 cases, post operative MRV were done and these were compared with respective preoperative images and no venous injury were identified. Evaluation of preoperative and postoperative MRV can be useful in assessment of venous injury. Our study had the following limitations: There was low statistical impact because of a small number of patients. Furthermore, cerebral venous thrombosis could not be detected in any of the participants; thus, the drawing of conclusions from these data about the performance of CE MRV and MPRAGE sequences in the detection of venous thrombosis remains difficult

CONCLUSIONS

This study showed that preoperative localization and evaluation of tumoral area and cortical veins with the help of cod liver oil in MRI and MRV brain was very helpful in planning the surgery, making craniotomy and to avoid injury of the veins. This technique is

easy to perform and the capsule is easily constructed and inexpensive. 3DCEMRV was found to be better modality than 2DTOF for delineation of veins. Final neurosurgical outcomes were better.

ABBREVIATIONS

3dcemrv: 3-dimensional contrast enhanced magnetic resonance venography;
 2dtof: 2-dimensional time of flight;
 daca: distal anterior cerebral artery;
 mrv: magnetic resonance venography;
 ct: computed tomography;
 sss: superior sagittal sinus;
 mip: maximum-intensity projection;
 sd: standard deviationism: arteriovenous malformation.

REFERENCES

1. Sekhar LN, Chanda A, Morita A (2002) The preservation and reconstruction of cerebral veins and sinuses. *J Clin Neurosci* 91(4):391–399. doi:10.1054/jocn.2001.1008
2. Andrews BT, Dujovny M, Mirchandani HG, Ausman JI (1989) Microsurgical anatomy of the venous drainage into the superior sagittal sinus. *Neurosurgery* 24:514–520.
3. Katada K, Anno H, Takeshita G. MR images of brain surface anatomy scanning (SAS). *Jpn J Magn Reson Med* 1990;9:215- 25.
4. Katada K. MR imaging of brain surface structures: surface anatomy scanning (SAS). *Neuroradiology* 1990;32:439-48.
5. Oka K, Rhoton AL, Barry M, Rodriguez R (1985) Microsurgical anatomy of the superficial veins of the cerebrum. *Neurosurgery* 17 (5):711–748. doi:10.1097/00006123-198511000-00003
6. Hancq S, Baleriaux D, Brotchi J (2003) Surgical treatment of parasagittal meningiomas. *Semin Neurosurg* 14:203–210. doi:10.1055/s-2004-828923
7. Chakeres DW, Schmalbrock P, Brogan M, Yuan C, Cohen L. Normal venous anatomy of the brain: demonstration with gadopentetate dimeglumine in enhanced three-dimensional MR angiography. *AJR Am J Roentgenol* 1991;156:161 - 72.
8. Farb RI, Scott JN, Montanera WJ, Wright GA, Terbrugge KG. Intracranial venous system: Gadolinium-enhanced three-dimensional MR venography with auto-triggered elliptic centric-ordered sequence initial experience. *Radiology* 2002;226:203 – 9
9. Ayanzen RH, Bird CR, Keller PJ, McCully FJ, Theobald MR, Heiserman JE. Cerebral MR venography: normal anatomy and potential diagnostic pitfalls. *AJNR Am J Neuroradiol* 2000; 21: 74–78
10. Farb RI, Scott JN, Montanera WJ, Wright GA, Terbrugge KG. Intracranial venous system: Gadolinium-enhanced three-dimensional MR venography with auto-triggered

- elliptic centric-ordered sequence initial experience. *Radiology* 2002;226:203 - 9.
11. Lewin S, Masaryk TJ, Smith AS, Ruggieri PM, Ross JS. Time-offlight intracranial MR venography: evaluation of the sequential oblique section technique. *AJNR Am J Neuroradiol* 1990;15:1657 - 64.
 12. Chakeres DW, Schmalbrock P, Brogan M, Yuan C, Cohen L. Normal venous anatomy of the brain: demonstration with gadopentetate dimeglumine in enhanced three-dimensional MR angiography. *AJR Am J Roentgenol* 1991;156:161 - 72.
 13. Creasy JL, Price RR, Presbrey T, Goins D, Partain CL, Kessler RM. Gadolinium-enhanced MR angiography. *Radiology* 1990;175:280 - 3
 14. Ikawa F, Sumida M, Uozumi T, Kiya K, Kurisu K, Arita K, Satoh H. Demonstration of venous systems with gadolinium-enhanced three-dimensional phase-contrast MR venography. *Neurosurg Rev* 1995;18:101 - 7.
 15. Stevenson J, Knopp EA, Litt AW. MP-RAGE subtraction venography. A new technique. *J Magn Reson Image* 1995;5:239 - 41.
 16. Leach JL, Strub WM, Gaskill-Shibley MF. Cerebral venous thrombus signal intensity and susceptibility effects on gradient recalled-echo MR imaging. *Am J Neuroradiol*. 2007;28:940-5.
 17. Burtscher J, Kremser C, Seiwald M, Obwegeser A, Wagner M, Aichner F et al (1998) Three-dimensional computer assisted magnetic resonance imaging for neurosurgical planning in parasagittal and parafalcine central region tumors. *Comput Aided Surg* 3(1):27-32
 18. Oka K, Rhoton AL, Barry M, Rodriguez R (1985) Microsurgical anatomy of the superficial veins of the cerebrum. *Neurosurgery* 17 (5):711-748. doi:10.1097/00006123-198511000-00003
 19. Rosahl SK, Gharabaghi A, Shahidi UH, Samii M (2006) Virtual reality augmentation in skull base surgery. *Skull Base* 16(2):59- 66. doi:10.1055/s-2006-931620
 20. Chua GG, Serra L, Kockro RA, Hern N, Nowinski WL, Chan C (1998) Volume-based tumor neurosurgery planning in the Virtual Workbench. *Virtual Reality Annual International Symposium*, 1998. *Proceedings IEEE*: 167-173
 21. Kathleen Joy Khu & Ivan Ng & Wai Hoe Ng; The relationship between parasagittal and falcine meningiomas and the superficial cortical veins: a virtual reality study; *Acta Neurochir* (2009) 151:1459-1464 DOI 10.1007/s00701-009-0379.
 22. R. Klingebiel, H.C. Bauknecht, G. Bohnera, R. Kirschb, J. Bergera and F. Masuhr; Comparative evaluation of 2D time-of-flight and 3D elliptic centric contrast enhanced MR venography in patients with presumptive cerebral venous and sinus thrombosis, 2007 EFNS European Journal of Neurology 14, 139-143.



Long term study on the effects of microsurgical DREZotomy for chronic pain control

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ABSTRACT

The DREZotomy (Dorsal Root Entry Zone tomy) is an analgesic procedure. The analgesic effect is evaluated on 30 patients with chronic pain resulting from respectively: brachial plexus avulsion (66.6%), postherpetic pain (10%), hyperspastic states (6.6%), phantom pain (6.6%), the pain in the stump (6.6%), and spinal cord injuries (3.3%). Pain intensity was evaluated using a visual analogue scale (VAS). At last evaluation, between 12 and 60 months, after DREZotomy, 93% had a good or excellent global pain relief after surgery. According to the component types of pain, 9.6% of patients had good or excellent control of the paroxysmal pain, and 84% of the continuous pain. Kaplan-Meier prediction of lasting global pain control at 60 months of follow-up was calculated at 75.5%. Comparison of the 2 corresponding Kaplan-Meier curves at long term, namely, pain control in 82.8% for the paroxysmal component and in 51.7% for the continuous component, showed a statistically significant difference ($P < 0.0001$). Functional effects are improved by more than 70% according to patients.

INTRODUCTION

Chronic pain is a major public health problem, causing disability and of considerable human suffering. Depressive states are frequently associated with it; without counting the health costs it generates. There are, however, situations where conservative methods such as conventional drugs and psychotherapies are not effective and where the use of "lesional" neurosurgical management can provide an effective solution. This is the case for certain microsurgical techniques including DREZotomy (DREZ: Dorsal Root Entry Zone). This technique consists in interrupting the so-called nociceptive fibers and destroying by coagulation the hyperactive neurons located in the dorsal horn, corresponding to the area of pain, respecting the other fibers. It has been shown to be effective in some cases of chronic pain [2-5, 8, 9].

Keywords

neuropathic pain,
chronic pain,
brachial plexus avulsion,
neurosurgery of pain,
microsurgical DREZotomy



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METHODS

We conducted a prospective study of 30 patients who underwent microsurgical DREZotomy for chronic pain resistant to medical treatment. This work was carried out between July 2007 and January 2015. Our series included 2 women and 28 men, who had an average age of 46 years with extreme ages of 25 and 79 years. All patients had a detailed clinical assessment including the characteristics of their pain, its intensity using the Visual Analog Scale (VAS), as well as an assessment of the psychological impact of the pain. Imaging, represented by CT scan and / or spinal MRI, was performed before the surgery to assess the extent of the lesions.

ETIOLOGIES

The chronic pain that affected our patients was caused by sequelae of brachial plexus avulsion (BPA) in 20 cases (66.6%) (figure 1), postherpetic neuralgia in 3 cases (10%), stump pain in 2 cases (6.6%), the phantom limb in 2 cases (6.6%), spasticity in 2 cases (6.6%) and one case (3.3%) of spinal cord injury.

PAIN DESCRIPTION

In our study, more than half of the patients (56.7%) experienced pain initially, and 33.3% two months after the onset of the lesions. The time between onset of pain and surgery is on average 5 ± 1 year. Three patients had a DN4 (Douleur Neuropathique en 4 Questions, French for neuropathic pain in 4 questions), lower than 3, including 2 with stump pain and one patient with phantom limb pain. In fact, during the interrogation, we noted in 25 patients (83.3%) the presence of tingling. These latter involved 18 patients with BPA (90%), all patients with postherpetic neuralgia, spinal cord injury and a patient with phantom limb pain (50%). Numbness was reported in 3 patients (10%) including two patients with BPA (10%) and a patient with spasticity (50%). Dysesthesia was present in one patient who has BPA. Analogous visual scale (VAS) of our patients was initially on average 9.20 ± 1.06 with extreme values of 7 and 10. Two categories of pain were noted, in particular electrical shooting-like paroxysmal pain, and the continuous pain realizing a painful background most often a burning type. The two components combined were present in 83% of patients. The paroxysmal and background components alone interested 6.7% and 10% of the patients, respectively. In the BPA, the pain concerned

C6 and C7 dermatomes in all cases, and C8 and T1 dermatomes, in 95% and 90% of cases, respectively. For stump pain, the dermatomes affected by the pain range from C6 to T1. For the phantom limb, the pain areas extend from C5 to T1. In the three patients with postherpetic neuralgia, the dermatomes concerned range from C7 to T1 and from T3 to T8 at a frequency of 33.33% of cases for each one, associated in cases with allodynia. In the patient who suffered from chronic pain due to spinal cord injury, the dermatomes involved range from L2 to S2. As for patients suffering from spastic pain, the dermatomes concerned are L1 and L2 but also L4, L5 and S1. Whatever the causal etiology of chronic pain, there is a noticeable impact on several aspects of life, whether individual, social or professional.

MEDICAL TREATMENT

All of our patients received simultaneous treatment with WHO class I and II anticonvulsants, tricyclics and analgesics. However, we note that 5 (16.6%) of our patients took at least occasional WHO class III analgesics during their previous management. Two of our three patients with postherpetic neuralgia used topical lidocaine as a patch in the allodynic areas. Although the duration of treatment was 5 years ± 1 year, there was no reported improvement.

SURGICAL MANAGEMENT

Microsurgical DREZotomy was performed under general anesthesia. The patients were placed in the prone position. The laminectomy was homolateral to the corresponding painful dermatomes and bilateral when the symptomatology concerned both sides. After opening the dura mater, micro-incisions under an operating microscope were performed at the spinal cord dorsolateral sulcus at the entrance of the posterior roots. The incision was of 2 mm deep at an angle of 35° anteriorly and medially when it was the cervical and thoracic levels and 45° when it was the lumbar region. These incisions were followed by low-intensity micro-coagulations (figures 2 and 3).

RESULTS

The effects of DREZotomy on pain are evaluated when the patient is discharged from the hospital, in average, 15 days after surgery, but also evaluated at 3 months and at long term (beyond 6 months). Whatever the etiology, the results of DREZotomy are mainly excellent for 26 patients (86.6%) at discharge,

for 25 patients (86.2%) at 3 months, and for 20 patients (68.9%) in the long term; the good results interested 4 patients (13.3%) at discharge, 4 patients (13.7%) at 3 months and 7 patients (24.1%) in the long term; poor results are rare and concerned only 2 patients (6.8%) in the long term (Figure 4). The long-term overall pain control defined by excellent to good results is studied by the KAPLAN-MEIER curve; this one shows 75.7% pain control over a 60 month follow-up period (Figure 5). In our study, the results are excellent on allodynia, especially in patients with postherpetic neuralgia. The correlation between the number of painful dermatomes and the results of microsurgical DREZotomy is statistically significant; the fewer the number of dermatomes, the more excellent the results of the DREZotomy ($p = 0.0093$). The control of the pain components defined by excellent and good results is analyzed using the KAPLAN-MEIER curve; this one shows that the control of the paroxysmal and continuous components is estimated respectively at 82.8% and 51.7% over a period of 60 months (Figures 6 and 7). This analysis demonstrates the effectiveness of the technique on

the various components of pain with a predilection on the paroxysmal component compared to the continuous component ($p < 0.0001$). Surgical DREZotomy improves the quality of life in all its aspects and whatever the etiology. This improvement is statistically significant in all cases ($p = 0.00000$). There is an average improvement of $83 \pm 15\%$ in daily activity, $79 \pm 14\%$ in walking, $85 \pm 14\%$ in the mood, $77 \pm 15\%$ in social relationships, and $85 \pm 14\%$ in sleep quality. The reduction in the doses of the drugs was performed gradually until stopping, in 3-4 weeks. Analgesic treatment at low doses was continued in 6 patients. Complications are generally few and quickly resolve; limited essentially to neck pain in 15.4% of cases, and in much rarer cases there was: CSF fistula in 3.3% of cases, ataxia, tactile hypoesthesia, transient or permanent arthrokinesthesia, transient dysthesia, and mild motor disorder in 3.7% of cases for each of these complications. We regret a death of one patient which occurred during hospitalization caused by a pulmonary embolism.

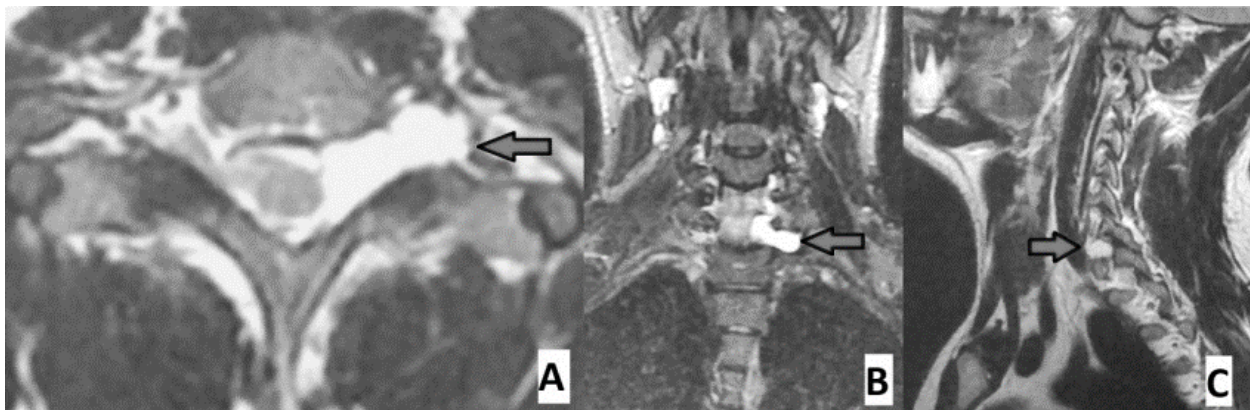


Figure 1. Preoperative cervical spine MRI in T2 weighted images, in patient suffering from brachial plexus avulsion; A: axial slide, B: coronal slide, C: sagittal slide; the arrow shows a pseudomeningocele characteristic lesion of brachial plexus avulsion.

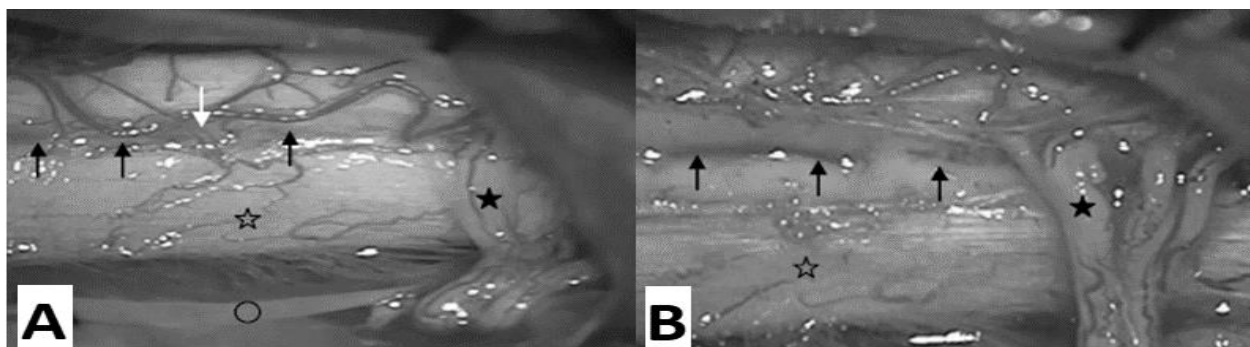


Figure 2. Per operative pictures in patient suffering from brachial plexus avulsion; A: before DREZotomy, B: after DREZotomy. The

black arrows show the DREZotomy zone for an avulsed C6 root, the white arrows show micro blood vessels entering the DREZ zone, the full star shows the C5 root, the empty star shows the spinal cord, and the ring the denticulate ligaments.

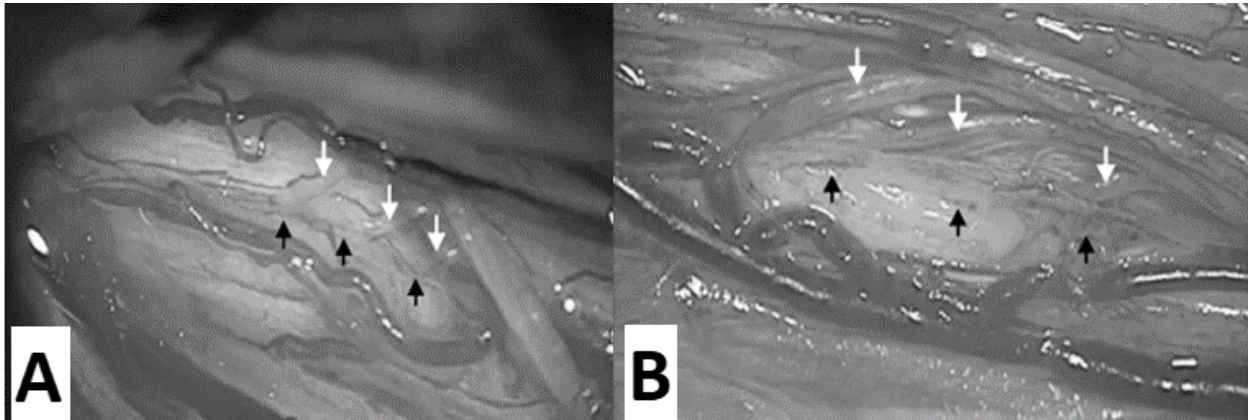


Figure 3. Per operative pictures in a patient suffering from postherpetic neuralgia; A: before DREZotomy, B: after DREZotomy. The black arrows show the DREZotomy zone, the white arrows show atrophied C7 root.

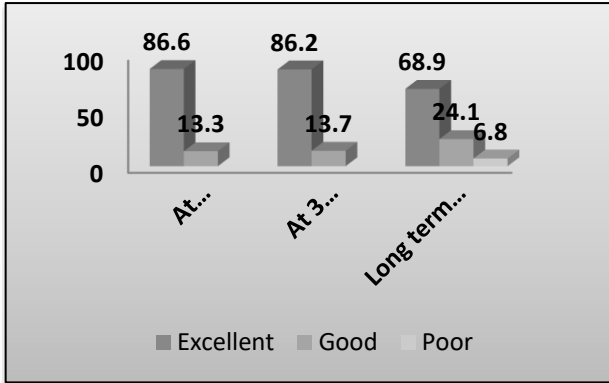


Figure 4. Good to excellent results of DREZotomy on overall pain at discharge, at 3 months and in the long term, all etiologies taken together (Excellent: Improvement greater than 75%; good: Improvement between 75% and 50%, and poor improvement less than 50%).

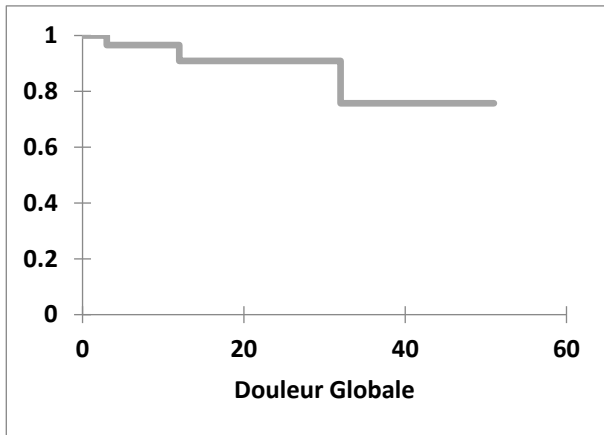


Figure 5. Kaplan Meier curve describing in percentage the long-term overall pain control (excellent to good). Pain control is

estimated at 75.7% over a 60-month follow-up period. Excellent: Improvement greater than 75%; Good: Improvement between 75% and 50%.

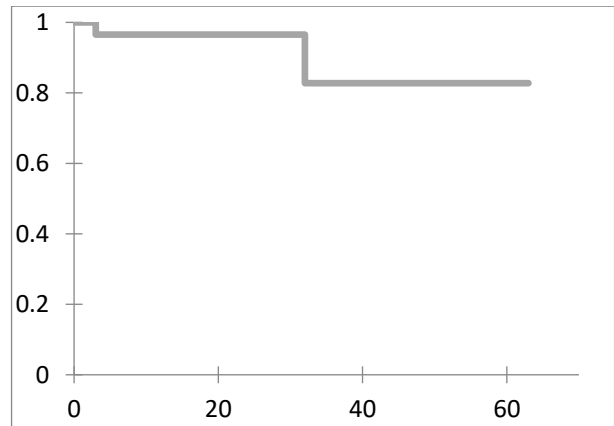


Figure 6. The Kaplan Meier curve describing in percentage the control of the paroxysmal component of pain (excellent to good) in the long term. Pain control is estimated at 82.8% over a period of 60 months. Excellent: Improvement greater than 75%; Good: Improvement between 75% and 50%.

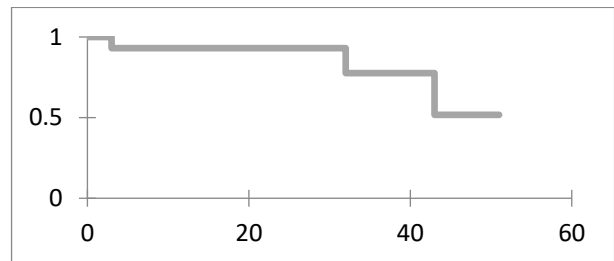


Figure 7. The Kaplan Meier curve describing in percentage the control of the continuous component of pain (excellent to good) in the long term. Pain control is estimated at 51.7% over

a period of 60 months. Excellent: Improvement greater than 75%; Good: Improvement between 75% and 50%.

DISCUSSION

We believe that the effectiveness of DREZotomy on pain reinforces the hypothesis that the dorsal horn plays a main role in the genesis of pain [4]. Guenot et al report the appearance of hyperactive neurons after the experimental rhizotomy performed in animals [13]. DREZotomy significantly suppresses autotomy behavior in animals that have undergone rhizotomy. Neuron hyperactivity has also been recorded using microelectrodes implanted in the dorsal horn during the DREZotomy procedure for BPA pain [12, 14, 16]. The fact that the paroxysmal component is often reduced by DREZotomy, suggests that its origin predominates in the dorsal horn. Central sensitisation by the phenomenon of denervation cannot in itself explain the mechanism generating pain within the dorsal horn. Like other authors, we have frequently observed intraoperatively, the presence of hemosiderin deposits, microcavitations and gliotic tissue within the gray matter of the dorsal horn in the cases of BPA and spinal cord injury [4,10,17]. These lesions can cause a change in normal activity within this region and consequently cause dysfunction of the pain pathways at the spinal or cerebral level [16]. However, it is quite clear that not all pain-generating centers are located at the dorsal horn. As for the mechanism of the continuous component, dominated by the burning sensation which is less influenced by DREZotomy than the paroxysmal component, it remains less clear; however, several hypotheses can be put forward. BPA causes more or less extensive necrosis of cells belonging to the dorsal horn that participate in the spinothalamic tract ascending pathway, as well as in the case of spinal cord trauma. This results in some degree of spino-reticulo-thalamic pain responsible of the continuous component [4]. The mechanisms of postherpetic neuralgia are similar to the above-mentioned mechanisms; the presence of the varicella zoster virus in the sensory neurons of the dorsal ganglion [1] causes inflammation and cell destruction within it [7,15, 23]. The inflammatory process extends along the sensitive fibers to the dorsal horn, causing lesions similar to those observed in the BPA [3], which explains the beneficial effect of DREZotomy on the two components, in particular the component paroxysmal as well as

allodynia. DREZotomy acts by suppressing the collaterals of the A β fibers which are oriented towards the nociceptive layers of the dorsal horn. Several hypotheses are suggested in the stump pain and the phantom limb pain. In fact, the loss of the afferent influx from the periphery leads to irritation of the neurons of the dorsal horn and their hyperexcitability and reduction of inhibitory processes [11,18, 25]. This gives the DREZ area a therapeutic interest. According to the series, paraplegic pain is mostly secondary to the conus medullaris lesion which often accompanies spinal trauma [21], probably in relation with the site corresponding to the thoraco-lumbar junction which is exposed to frequently to fractures. The thoracolumbar junction corresponds to numerous spinal segments condensed on a small poorly vascularized (Adamkiewicz artery) explaining their concomitant involvement during the trauma. The mechanism of spastic pain may be linked to painful spasms, painful contracture and neuropathic pain [21]; which occurs in multiple sclerosis [8,9], and spinal cord injury [10]. This neuropathic disorder is secondary to a demyelization of the spino-thalamic tract [6,24] and the posterior horn where the paroxysmal and continuous components are usually present. The beneficial effect of DREZotomy, on the various components of pain in particular the paroxysmal component was obtained by interrupting the myotatic fibers (monosynaptics), and the nociceptive fibers (polysynaptics), thus depriving the somatosensitive relays of the dorsal horn of all excitatory afferents [22].

CONCLUSIONS

DREZotomy has a satisfactory operating risk / benefit ratio with few side effects. It is suitable for chronic refractory pain especially BPA and spinal cord injury. It significantly attenuates chronic pain in its paroxysmal but also continuous components with supremacy for paroxysmal pain, including in allodynic phenomena.

CONFLICT OF INTEREST

None

REFERENCES

1. Ada Delaney, Lesley A. Colvin, Marie T. Fallon, Robert G. Dalziel, Rory Mitchell, and Susan M. Fleetwood-Walker. Postherpetic Neuralgia: From Preclinical Models to the

- Clinic. Neurotherapeutics: The Journal of the American Society for Experimental NeuroTherapeutics. October 2009. Vol. 6, 630-637.
2. Adam F. et le Bars D. Voies de la douleur. In: Martin C., Riou B. et Vallet B. (2006). Physiologie humaine appliquée. Arnette, Rueil-Malmaison, 845-857.
 3. Allan H. Friedman, Blaine S. Nashold, AND Janice Ovelmen-LevitT. Dorsal root entry zone lesions for the treatment of post-herpetic neuralgia. *J Neurosurg* 60:1258-1262, 1984.
 4. Aichaoui F, Mertens P, Sindou M. Dorsal root entry zone lesioning for pain after brachial plexus avulsion: Results with special emphasis on differential effects on the paroxysmal versus the continuous components. A prospective study in a 29-patient consecutive series. *PAIN_ 152* (2011) 1923-1930
 5. Han ZS, Zhang ET, Craig AD. Nociceptive and thermoreceptive lamina I neurons are anatomically distinct. *Nat Neurosci* 1998;1 :218-25.
 6. Dahm.P.O, Nitescu.P.V, Appelgren.L.K and Curelaru.L. Long-term intrathecal (i.t.) infusion of bupivacaine relieved intractable pain and spasticity in a patient with multiple sclerosis. *European Journal of Pain* (1998) 2: 81-85.
 7. D'Hardemare Vincent, Margot-Dclot Anne, Bruxelles Jean, Bachelart Maximilien, Thiébaud Jean-Baptiste. *Chirurgie de la douleur de la lésion à la neuromodulation*. Springer 2014; 33 :374-387.
 8. Devor M. Sodium channels and mechanisms of neuropathic pain. *J Pain* 2006;7(Suppl. 1): 3-12.
 9. Dickenson A. The inhibitory effects of thalamic stimulation of the spinal transmission of nociceptive information on the rat) (Gerhart KD, Yezierski RP, Fang ZR, Willis WD. Inhibition of primate spinothalamic tract neurons by stimulation in ventral posterior lateral (VPLc) thalamic nucleus: possible mechanisms. *J Neurophysiol* 1983; 49 :406-23.
 10. Dreval ON. Ultrasonic DREZ-operations for treatment of pain due to brachial plexus avulsion. *Acta Neurochir* 1993;122:76-81.
 11. Flor H. Phantom-limb pain: characteristics, causes, and treatment. *Lancet Neurol* 2002;1:182 9.
 12. Guenot M, Bullier J, Rospars JP, Lansky P, Mertens P, Sindou M. Single-unit analysis of the spinal dorsal horn in patients with neuropathic pain. *J Clin Neurophysiol* 2003;20:143-50.
 13. Guenot M, Bullier J, Sindou M. Clinical and electrophysiological expression of deafferentation pain alleviated by dorsal root entry zone lesions in rats. *J Neurosurg* 2002;97:1402-9.
 14. Guenot M, Hupe JM, Mertens P, Ainsworth A, Bullier J, Sindou M. A new type of microelectrode for obtaining unitary recordings in the human spinal cord. *J Neurosurg Spine* 1999;91:25-32.
 15. Hökfelt T, Zhang X, Wiesenfeld-Hallin Z. Messenger plasticity in primary sensory neurons following axotomy and its functional implications. *Trends Neurosci* 1994;17:22-30.
 16. Jeanmonod D, Sindou M, Magnin M, Boudet M. Intra-operative unit recordings in the human dorsal horn with a simplified floating microelectrode. *Electroencephalogr Clin Neurophysiol* 1989;72:450-4.
 17. Kandel EI, Ogleznev KIA, Dreval ON. Destruktsiia vkhodnoi zony zadnykh koreshkov kak metod lecheniia khronicheskoi boli pri traumacheskikh povrezhdeniakh plechevnogo spleteniia. *Zh Vopr Neurokhir Im N N Burdenko* 1987;6:20-7.
 18. Kerr, 1975a. Snyder, 1977 ; livre ablative.
 19. Lazorthes Y, Sallerin-Caute B, Verdie JC, et al. Chronic intrathecal Baclofen administration for control of severe spasticity. *J Neurosurg*. 1990; 72:393-402.
 20. Penn RD, Kroin JS. Continuous intrathecal baclofen for severe spasticity. *Lancet* 2:125-127, 1985) (Lazorthes Y, Sallerin-Caute B, Verdie JC, et al: Chronic intrathecal Baclofen administration for control of severe spasticity. *J Neurosurg* 72:393-402, 1990.
 21. Sindou Marc P., and Mertens Patrick. *Surgery in the Dorsal Root Entry Zone for Spasticity in Adults. Operative Technique in Neurosurgery*. 2005. Elsevier. Inc.
 22. Sindou.M, E Blondet, E Emery, and P Mertens. Microsurgical lesioning in the dorsal root entry zone for pain due to brachial plexus avulsion : a prospective series of 55 patients. *J Neurosurg* 102 : 1018-1028. 2005.
 23. Smith MT, Edwards RR, McCann UD, Haythornthwaite JA. The effects of sleep deprivation on pain inhibition and spontaneous pain in women. *Sleep* 2007;30:494-505.
 24. Thomas RJ. Excitatory amino acids in health and disease. *J Am Geriatr Soc* 1995; 43:1279-89.
 25. Woolf CJ, Ma Q. Nociceptors- Noxious stimulus detectors. *Neuron* 200 ; 55 : 353-64.



Post-traumatic arachnoid cyst without neurological sequels. A case report

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ABSTRACT

An eight-year-old male patient was admitted to the hospital with a history of left median paramedian frontal craniectomy due to car trauma at six months of age. Axial computed tomography of the skull with reconstruction in three dimensions revealed an arachnoid cyst with slight herniation of the brain in the frontal lobe, leading to protrusion against the skullcap, causing dilation of the ex-vacuum of the anterior extension of the homolateral lateral ventricle. He presented asymmetrical lateral ventricles, a reduced base cistern, and a slightly ectatic IV centred ventricle. After the physical examination, no neurological deficit was found, despite the changes identified in the images. It is believed that such conditions may progressively worsen with the development and maturation of nervous tissue over the age of the assessed child. To confirm this, specialized monitoring is of fundamental importance.

INTRODUCTION

The arachnoid cyst is a meningeal expansion, in the form of a bag, filled with liquid with characteristics identical to the liquor, and it can develop in any place where there is an arachnoid. They are congenital and are formed due to valve defects in the arachnoid membranes that facilitate the passage of the cerebrospinal fluid into the cyst and, in the same way, hinder the exit(1). Although many may constitute incidental findings, others cause symptoms due to compression of the brain parenchyma or increased intracranial pressure (2). Due to the complexity and rarity of the case, as it is a post-traumatic arachnoid cyst of the skull in the frontal lobe region in a young patient, aged eight, without any apparent neurological impairment, it was considered relevant for the scientific literature describing this case.

Keywords

encephalocele,
tomography,
X-Ray computed,
craniocerebral trauma,
diagnostic imaging



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CASE REPORT

Male patient, eight years old, enters the surgical center with a history of left median paramedian frontal craniectomy due to car trauma at 6 months of age. A computed tomography scan of the skull was performed with 3D reconstruction, showing an arachnoid cyst in the frontal lobe protruding into the

skull cap determining dilation of the ex-vacuum of the anterior extension of the homolateral lateral ventricle. It also has asymmetrical lateral ventricles, a reduced base cistern and a slightly ectatic centralized ventricle (Figures 1,2 and 3). On physical examination, the patient did not show a neurological deficit.

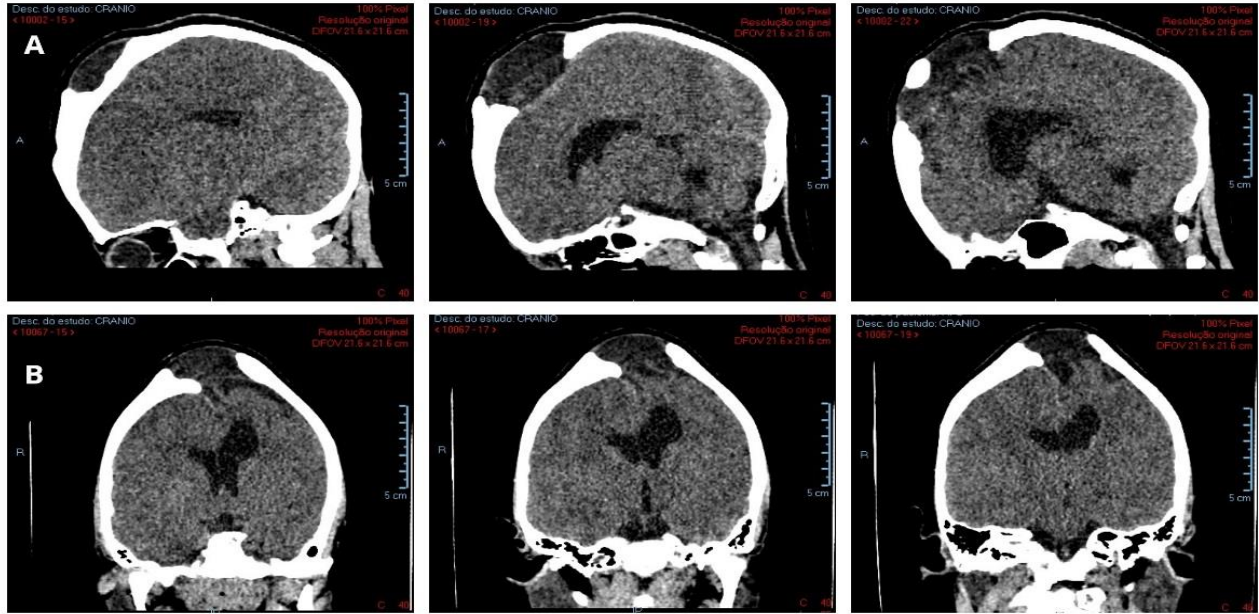


Figure 1: Sagittal (A) and coronal (B) skull computed tomography showing an arachnoid cyst in the frontal lobe protruding into the skullcap, reduced volume cistern and slightly ectasied centered ventricle.

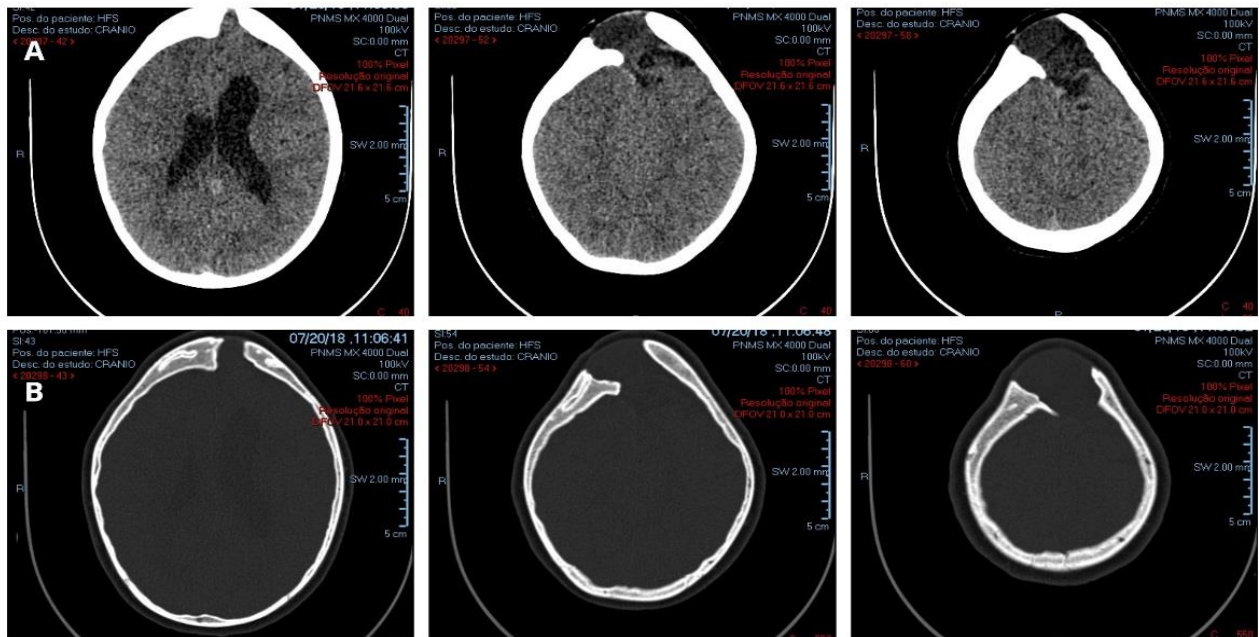


Figure 2: Sequence A of computed tomography of the skull, axial section showing an arachnoid cyst in the frontal lobe protruding

in the skullcap. Sequence B of computed tomography shows dilation of the ex-vacuum of the anterior extension of the homolateral lateral ventricle. Asymmetric lateral ventricles are observed.

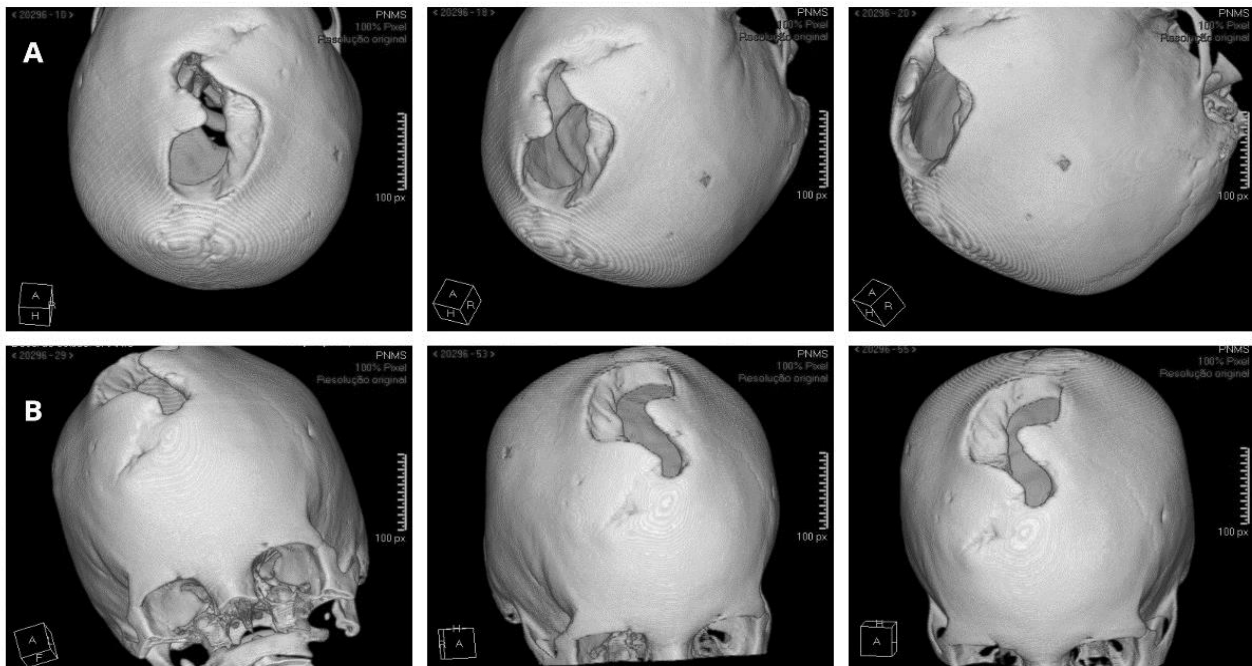


Figure 3: Sequences A and B are computed tomography scans of the skull with 3D reconstruction showing the absence of bone fusion of the anterior fontanelle.

DISCUSSION

Intracranial cysts represent 1% of lesions with a mass effect. Most have a congenital origin and an intra-arachnoid situation, presenting or not communicating with the subarachnoid space (1,2-6). The arachnoid cyst is a meningeal expansion, in the form of a bag, filled with fluid with characteristics identical to cerebrospinal fluid or CSF, and maybe develop in any location where arachnoid exists. Most have a congenital origin and an intra-arachnoid situation, with or without communication with the subarachnoid space (1,2-6).

The development of the cyst can occur due to a primary malformation of the arachnoid, as a consequence of a duplication of the membrane, at an early stage of embryogenesis, however, some cysts originate from the rupture or fragility of the arachnoid membrane in the presence of trauma, tumor or infection (1). Arachnoid cysts occur more frequently in children and young adults, most of them under the age of 20 years (3,5,6), the reasons for this fact are still not well understood. There is no direct relationship between the volume of the cyst and the clinical findings. Large cysts may be

accompanied by moderate symptoms and this may occur due to their location and the brain's adaptability. Among the most frequent clinical manifestations are macrocephaly, headache, focal signs and seizures.

CONCLUSIONS

The therapeutic strategies currently used are essentially surgical, depending on the symptoms, however, as the patient did not present clinical symptoms, the literature has reported the indication for performing craniotomy with membranectomy (1,5,6) or drainage utilizing cystoperitoneal shunt (7-9).

REFERENCES

1. Grollmus JM,, Wilson CB, Newton TH. Paramesencephalic arachnoid cysts. *Neurology* 1976;26:128-134.
2. Koga H, Mukawaa J, Miyagi K, Kinjo T, Okuyama K. Symptomatic intraventricular arachnoid cysts in an elderly man. *Acta Neurochir (Wien)* 1995;137:113-117.
3. Galassi E, Tognetti F, Gaist G, Fagioli L, Frank F, Frank G. CT scan and metrizamide CT cisternography in arachnoid cysts of

the middle cranial fossa: classification and pathophysiological aspects. *Neurology* 1982;7:363-369.

4. Handa J, Okamoto K, Sato M. Arachnoid cyst of the middle cranial fossa: report of bilateral cysts in siblings. *Surg Neurol* 1981;16:127-130.

5. Hayashi T, Aneqawa S, Honda E, et al.. Clinical analysis of arachnoid cysts in the middle fossa. *Neurochirurgia* 1979;22:127-130.

6. Jallo GI, Woo HH, Meshkii CH, Epstein FJ, Wisoff JH. Arachnoid cysts of the cerebellopontine angle: diagnosis and surgery. *Neurosurgery* 1997;40:31-38.

7. Schroeder HHWS, Gaab MR. Endoscopic observation of a slit-valve mechanism in a suprasellar prepontine arachnoid cyst: case report. *Neurosurgery* 1997;40:198-200.

8. Arai H, Sato K, Wachi A, Okuda O, Takeda N. Arachnoid cysts of the middle cranial fossa: experience with 77 patients who were treated with cystoperitoneal shunting. *Neurosurgery* 1996;39:1108-1113. [Links]

9. Borges G, Fernandes YB, Gallani NR. Hemorragia do tronco cerebral após remoção cirúrgica de cisto aracnóide da fissura silviana. *Arq Neuropsiquiatr* 1995;53:825-830



Hypotension intracrânienne spontanée (HIS): A propos d'un cas

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ABSTRACT

L'HIS est une céphalée chronique, liée à une fuite de LCR. Son diagnostic est facilité par l'IRM et sa prise est essentiellement médicale. Nous rapportons le cas d'une jeune femme ayant présenté un tableau clinique et signes IRM compatibles avec ce diagnostic et ayant bien évolué sous traitement.

INTRODUCTION

L'hypotension intracrânienne spontanée (HIS) fût décrite par le Neurologue Allemand Schaltenbrand en 1938 (1). En effet, il définit sous le terme d'aliqorrhée une pathologie d'apparition spontanée associant une très basse pression d'ouverture du liquide céphalorachidien (LCR) (inférieure à 60 mm d'eau) et des céphalées orthostatiques (2).

Le diagnostic de l'HIS a été considérablement facilité par le développement de l'imagerie par résonance magnétique cérébrale, avec une connaissance plus précise des anomalies caractéristiques de cette affection lors de cette exploration (3,4).

OBSERVATION

Histoire de la maladie

Madame N, 38 ans, fonctionnaire, sans antécédents pathologiques notables ni de facteurs de risque cardiovasculaires, a consulté pour céphalées diffuses à type de constriction, fluctuantes, d'aggravation progressive depuis 1 mois et résistantes aux antalgiques du premier palier.

Les céphalées apparaissaient rapidement lors du passage de la position allongée à la position assise ou debout et s'accompagnait de nausées et de deux épisodes de vomissements. On note la présence d'une phonophobie, d'une hypoacousie avec une sensation d'oreille bouchée et des acouphènes à gauche, mais pas de diplopie ni de flou visuel.

Ces céphalées s'associaient également à des douleurs de la nuque, tout en augmentant d'intensité au fur et à mesure du maintien de la position debout ou assise et s'amélioraient rapidement lors du passage

Keywords

céphalée chronique,
fuite du LCR,
hypotension du LCR



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à la position allongée, avant de disparaître complètement par la suite.

Examen Clinique

L'examen neurologique ne retrouvait aucune anomalie. Il n'y avait pas de signes d'hypertension intracrânienne ou de syndrome méningé. L'examen général était normal avec une tension artérielle est de 130/85.

Examens complémentaires

Scanner cérébral avec injection

Réalisé en urgence, lors des premiers accès de céphalées et n'avait pas montré d'anomalies.

IRM cérébrale

L'IRM cérébrale montrait en séquence FLAIR un épaissement diffus des méninges encéphaliques avec un aspect de petits ventricules (Fig. 1).

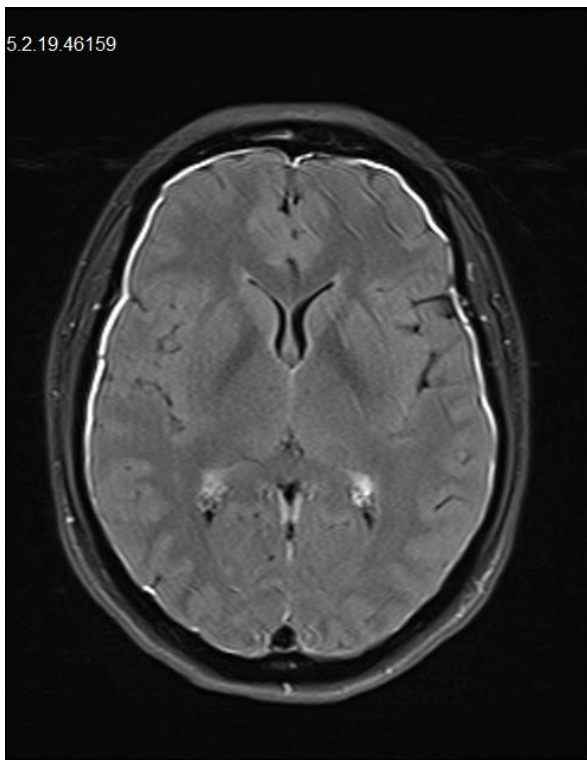


Figure 1.

Il s'y associait de chaque côté une fine lame d'épanchement sous dural fronto-pariétale bilatérale (Fig. 2).

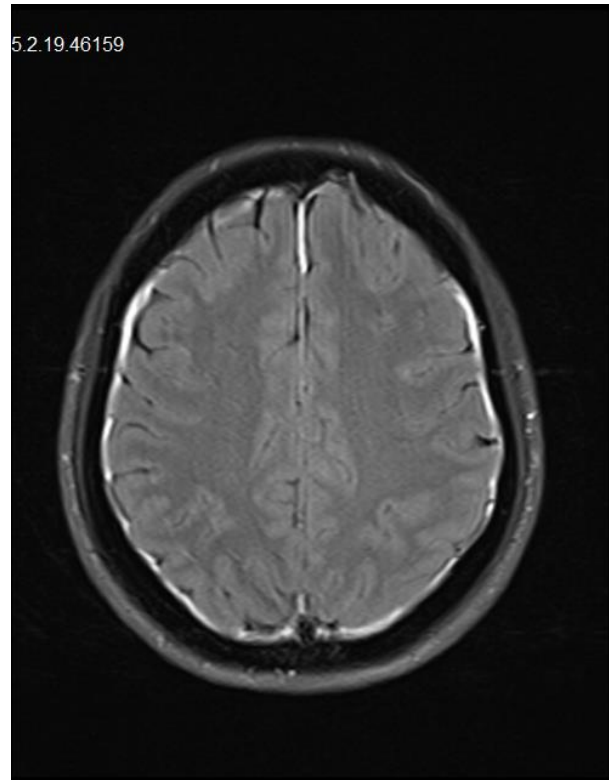


Figure 2.

En T1 après injection de gadolinium, on observait un rehaussement de la dure mère au niveau de la convexité sus et sous tentorielle, de la faux et de la tente du cervelet (Fig. 3).



Figure 3.

On notait enfin une hypertrophie hypophysaire (Fig. 4).



Figure 4.

Il n'y avait pas d'anomalie du parenchyme cérébral.

IRM médullaire

Collection sous durale d'allure liquidienne, antérieure entre C7 et D3, en hypersignal T2 (Fig. 5).



Figure 5.

On observait sur les coupes sagittales cervicales hautes une ptose de l'encéphale et notamment des

amygdales cérébelleuses sans anomalies morphologiques associées (Fig. 4).

PRISE EN CHARGE ET ÉVOLUTION

Devant les céphalées d'aggravation orthostatique, l'absence de notion de traumatisme crânien, de ponction lombaire ou d'intervention neurochirurgicale et les images en IRM, le diagnostic d'hypotension intracrânienne spontanée a été retenu. Le décubitus prolongé au-delà de 24 h avec hyperhydratation a permis l'amélioration de la symptomatologie.

DISCUSSION

L'hypotension intracrânienne spontanée (HIS) est une pathologie qui reste rare, dont la prévalence se situe entre 1/50 000 et 5/100 000, avec une prépondérance féminine (sex-ratio à 2/1) et un pic d'incidence vers 40 ans [5]. Elle est plus fréquente au cours des maladies du tissu conjonctif (Marfan, Ehler-Danlos) [6].

Sa forme classique associe des céphalées postérieures et des cervicalgies à type de constriction, d'intensité modérée à sévère, s'installant dans les 15 minutes suivant l'orthostatisme, soulagée par le décubitus, en l'absence de traumatisme sévère ou de gestes invasifs (chirurgie ou ponction lombaire). L'association à des signes de nature ORL est classique, en lien avec des modifications de pression de l'oreille interne. Une atteinte de nerfs crâniens, notamment du VI, est plus rarement décrite. Toutefois les céphalées deviennent moins caractéristiques avec la chronicisation ; le tableau peut alors évoluer vers des céphalées chroniques quotidiennes, parfois associées à un abus médicamenteux.

La physiopathologie supposée repose sur une hypovolémie du liquide céphalo-rachidien (LCR) secondaire à des brèches dure mériennes le plus souvent médullaires, responsable de la traction sur les différentes structures du cerveau et à l'origine des signes IRM [7]. Ces brèches seraient favorisées par l'association d'un traumatisme parfois mineur et d'une fragilité du tissu méningé spinal.

L'hypertrophie hypophysaire, les collections sous-durales à l'étage cérébral et rachidien, l'expansion veineuse au niveau intracrânien et rachidien et l'épaississement de la dure mère sont

liés à la baisse de pression du LCR qu'ils permettent de compenser (Loi de Monro-Kellie).

Le rehaussement diffus de la dure mère au niveau de la convexité sus et sous tentorielle, de la faux et de la tente du cervelet est un important signe pour le diagnostic différentiel avec une méningite infectieuse ou carcinomateuse.

L'IRM cérébrale peut être normale dans environ 20 % des cas [5, 7,8] et n'exclut donc pas le diagnostic en cas de forte présomption clinique.

L'IRM médullaire avec notamment des séquences T2 et STIR doit être toujours réalisée lorsque l'on suspecte une HIS. En effet, Il s'agit de l'examen le plus performant [9] avec une sensibilité proche de 94 % [3], notamment à la phase précoce, montrant dans 88 % des cas des collections épidurales, dans 78 à 94 % des cas une dilatation des veines épidurales cervicales [6]. Il a également été observé des prises de contraste au contact de ces collections en lien avec la réaction leptoméningée [11,12].

Enfin, il faut penser à une étiologie de l'HIC pouvant expliquer la fuite chronique de LCR (brèche durale) qui est rarement retrouvée à l'IRM médullaire (pseudoméningocèle, ectasie durale, kystes radiculaires, protrusions disco-ostéophytiques, etc).

L'évolution sans traitement peut se faire vers la rémission spontanée, la chronicisation ou des complications (hématome sous-dural, thrombose veineuse cérébrale). Ces dernières modifient les caractéristiques de la céphalée, avec notamment l'apparition rapide de tableaux d'hypertension intracrânienne et un engagement amygdalien. Toute modification de la céphalée chez un patient souffrant d'une HIS justifie donc une réévaluation clinique et l'IRM en urgence.

La prise en charge thérapeutique repose sur le traitement conservateur (repos en décubitus, hyperhydratation) et le blood-patch (plusieurs tentatives pouvant être nécessaires).

Lorsque le patient ne s'améliore pas malgré le traitement médical (voir le blood patch), il y a nécessité d'essayer de trouver la fuite, avec un myéloscanner ou une cisternographie, pour apporter un diagnostic complet et guider ainsi la conduite thérapeutique (blood patch au bon endroit et éventuelle chirurgie).

Les indications chirurgicales sont exceptionnelles et concernent, avant tout, les lésions sévères ou développementales, résistantes au traitement médical [5,13]. Les anomalies de signal méningées

en IRM régressent généralement suite au traitement, avec un décalage par rapport à l'amélioration clinique [5].

CONCLUSION

L'HIS est une céphalée chronique, liée à une fuite de LCR.

Les conséquences de cette hypovolémie du LCR sont essentiellement la ptôse cérébrale et la compensation veineuse. Ces deux conséquences expliquent les manifestations cliniques et l'IRM de cette pathologie.

Enfin, il faut souligner l'intérêt de l'imagerie médullaire, notamment, le myéloscanner pour rechercher une brèche durale; ce qui est recommandé en l'absence d'amélioration au traitement médical et au blood patch.

REFERENCES

- Schaltenbrand G. Neuere anschauungen zur pathophysiologie der liquorzirkulation. Zentrabl Neurochir 193 8;3 :290-300.
- Mokri B. Spontaneous intracranial hypotension Spontaneous CSF leaks. Headache Currents 2005;2(1): 1 1-22.
- Fishrnan RA, Dillon WP. Dura1 enhancement and cerebral displacement secondary to intracranial hypotension. Neurology 1993;43(3 Pt 1):609-6 1 1.
- Sable SG RN. Meningial enhancement and low cerebrospinal fluid pressure headache. An MRI study. Cephalalgia 199 1 ; 1 1 :275-276.
- Schievink WI. Spontaneous spinal cerebrospinal fluid leaks and intracranial hypotension. JAMA 2006;295:2286-96.
- Schievink WI, Gordon OK, Tourje J. Connective tissue disorders with spontaneous spinal cerebrospinal fluid leaks and intracranial hypotension: a prospective study. Neurosurgery 2004;54:65-70.
- Watanabe A, Horikoshi T, Uchida M, Koizumi H, Yagishita T, Kinouchi H. Diagnostic value of spinal MR imaging in spontaneous intracranial hypotension syndrome. AJNR Am Neuroradiol 2009;30(1):147-51.
- Schoffer KL, Benstead TJ, Grant I. Spontaneous intracranial hypotension in the absence of magnetic resonance imaging abnormalities. Can J Neurol Sci 2002;29(3):253-7.
- Schievink WI, Maya MM, Louy C, Moser FG, Tourje J. Diagnostic criteria for spontaneous spinal CSF leaks and intracranial hypotension. AJNR Am J Neuroradiol 2008;29(5):853-6.
- Farb RI, Forghani R, Lee SK, Mikulis DJ, Agid R. The venous distension sign: a diagnostic sign of intracranial

- hypotension atMR Imaging of the brain. *AJNR Am J Neuroradiol* 2007;28(8):1489-93.
11. Good DC, Ghobrial M. Pathologic changes associated with intracranial hypotension and meningeal enhancement on MRI. *Neurology* 1993;43:2698-700.
 12. Mokri B, Parisi JE, Scheithauer BW, et al. Meningeal biopsy in intracranial hypotension: meningeal enhancement on MRI. *Neurology* 1995;45:1801-6.
 13. Schievink WI, Morreale VM, Atkinson JLD, et al. Surgical treatment of spontaneous spinal cerebrospinal fluid leaks. *J Neurosurg* 1998;88:243-6.



Predictors of severe head injury in tertiary centre

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ABSTRACT

Introduction: Severe head injury (SHI) is associated with a high mortality and morbidity rate and is one of the leading causes of death in intensive care units. The aim of this study was to identify predictors of hospital outcome and mortality in ICU admitted SHI patients and to estimate their impact.

Methods: A retrospective analysis was carried on patients (n=321) with a severe head injury, defined as Glasgow Coma Scale (GCS) ≤ 8 who were admitted to the ICU neurosurgical department of National Trauma Center from 2017 to 2018. Both clinical and radiological predictors of hospitalized patients were identified.

Results: Total mortality rate was 5.9%. 243 (75.7%) of the patients were male and 78 (24.3%) were female. 55% of cases were due to traffic accidents. Patients Middle Ages group was 60%. Coexisting injuries, found in 25% of the patients aggravated the prognosis. Blood grouping pattern in SHI were B+, A+ and O+ 36.1, 28 and 24.3% respectively. 45% of the patients had Tattoo which was aggravating factors. The outcome is highly correlated with GCS' values. CT scan findings revealed that patients with subdural hygroma after few days of admission CT scan which was very important prognostic factors in SHI.

Conclusions: SHI has high mortality and morbidity in today world as it has a high negative impact on young people, especially men with blood group B+. The age of the patient, presence of Tattoo, GCS at admission, the CT scanning at admission and CT scanning after a week of admission were significant predictors of outcome.

INTRODUCTION

Head injury (HI) is injuries to the scalp, skull, or brain (Traumatic Brain injury) produced by trauma, possibly leading to permanent or temporary impairment of cognitive, physical, and psychosocial functions, with diminished or altered state of consciousness. Severe traumatic brain injury (TBI) is associated with a high mortality and morbidity rate and is one of the leading causes of death in the intensive care units (3, 16). This is a retrospective study to identify predictors of hospital outcome/ mortality in ICU admitted severe TBI patients and to estimate their impact. Moreover, this study also intends to find the recent trends of head injury.

MATERIALS AND METHODS

A retrospective analysis was done on patients with severe head injury,

Keywords

Glasgow Coma Scale,
CT scan,
severe head injury,
subdural hygroma,
tattoo



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defined as Glasgow Coma Scale (GCS) ≤ 8 , who were admitted to the ICU neurosurgical department of National Trauma Center from June 2017 to July 2018. CT scan brain plain was done as a part of routine evaluation for all patients when presented in emergency department (ER). Patient's inpatient files, ER and police records were used to retrieve the data. The consciousness level was assessed by the Glasgow Coma Scale (GCS) and the outcome was assessed by the Glasgow Outcome Score (GOS). Patient characteristics like age, gender distribution, mode of injury, alcohol influence, GCS on admission, pupil reactivity, presence of extra-cranial associated injuries, laboratory tests and CT scan results were evaluated.

Outcome assessment for survivors was based on data from the medical records of patients during their hospitalization, and that obtained within a 6 month-period after discharge. Unfavorable outcome (death or severe disability) at six months was defined with the GOS. The scale comprises five categories: death, vegetative state, severe disability, moderate disability, and good recovery. Our results are statistically analyzed and discussed. Inclusion criteria: All the severe head injury patients who were admitted in intensive care unit under the Neurosurgery care during the study period. Exclusion criteria: The severe head-injured patients who had polytrauma with cervical injury.

RESULTS

In this study, there were 321 patients with severe head injury. 243 (75.7%) of the patients were male and 78 (24.3%) were female (Figure 1). Patients Middle Ages group was 60% (Figure 2). The number of patients with severe head injury admitted to the hospital was 321, out of which 174 (54%) patients had a head injury following Road traffic accident (RTA), 114 (36%) sustained head injury following fall from height, 20 (6%) secondary to physical assault, and 13 (4%) attributed to other modes of injury (Figure 3). Most of the cases admitted in ICU (96%) were intubated and mechanically ventilated. The radiological findings on head injury patients showed contusion in most cases around 29% (Figure 4). Non-surgical (conservative) treatment was provided to 225 (70%) patients while 96 (30%) patients required surgical intervention (Figure 5). Coexisting injuries, found in 25% of the patients, affected the prognosis. Two third of the patients admitted with SHI were

under the influence of alcohol (Figure 6). Blood grouping pattern in SHI were B+, A+ and O+ 36.1, 28 and 24.3% respectively (Figure 7). 45% of the RTA patients had Tattoo which was aggravating factors. CT scan findings revealed that patients with subdural hygroma after few days of admission CT scan which was very important prognostic factors in TBI. Outcome highly correlated with GCS score.

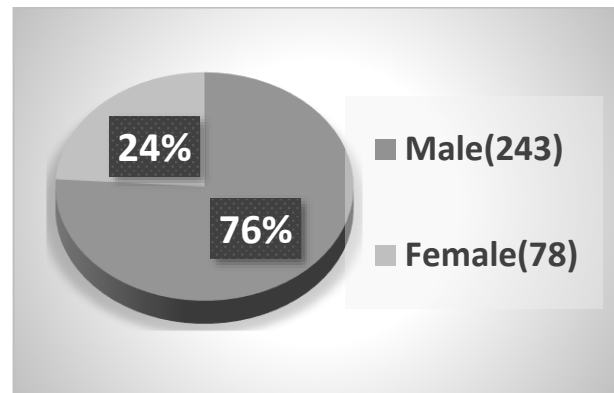


Figure 1. Gender distribution of admitted SHI.

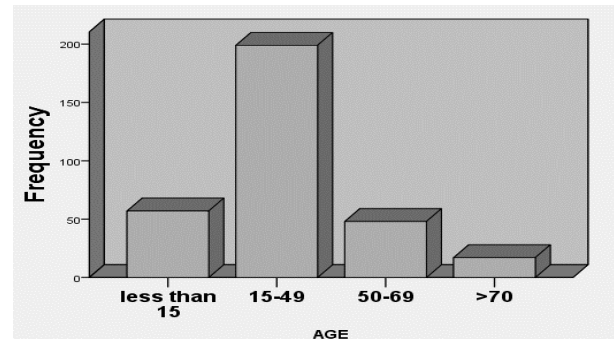


Figure 2. Age distribution of admitted SHI.

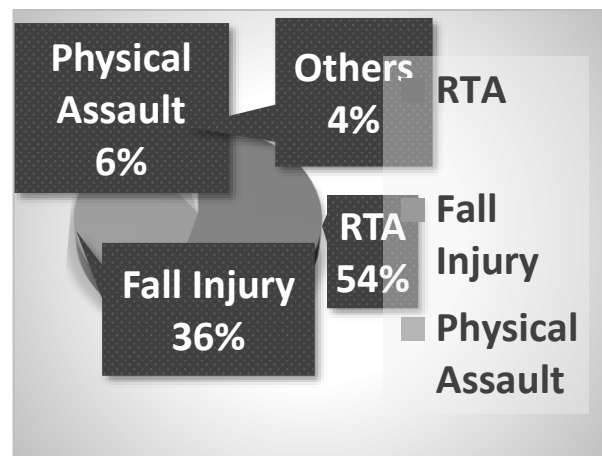


Figure 3. The distribution of causes of injury in admitted SHI (n=321).

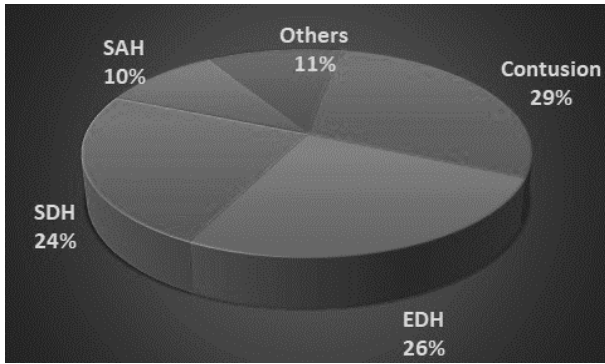


Figure 4. Radiological Finding of admitted SHI patients.

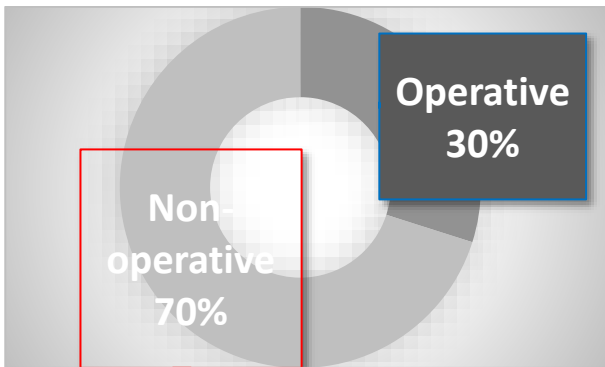


Figure 5. Management of admitted SHI patients.

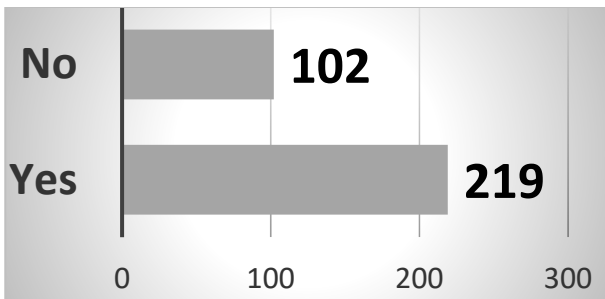


Figure 6. Numbers of patients under alcohol influences.

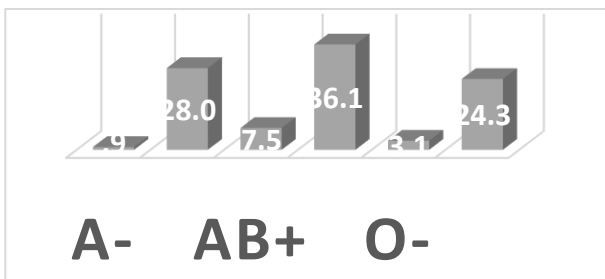


Figure 7. Blood grouping pattern in Severe Head injuries.

DISCUSSION

Severe head injury (SHI) is the most common global cause of morbidity and mortality in people under 45

years of age and possess major public health and socioeconomic challenges, which can negatively impact the daily life activities and possess high risk of readmission to hospital and subsequent death. Despite advancements in diagnosis and treatment, there are many queries regarding the diagnosis, prognosis and best possible treatment of SHI. There are no acceptable clinical and pathological predictive factors have been recognized for developing clinical trials and promoting effective therapeutic strategies to improve the prognosis. There were a lot of different prognostic factors related to outcome. The most important prognostic factors included: age, Glasgow Coma Scale, pupil size, coexistence of other injuries, a history of previous head injury, alcohol abuse and lower socioeconomic, educational status, hypotension, hypoxia, glucose, coagulopathy, haemoglobin, Marshall CT classification and traumatic subarachnoid haemorrhage (1, 2, 9, and 11). This article focuses mainly on the clinically significant aspects of potential interrelationship between potential diagnostic and prognostics tools in severe brain injury. Assessing prognosis after traumatic brain injury was both very important and difficult.

In this study, the majority of patients were male 76% and only 24% were female (Figure 1). Patients of both sexes primarily belong to an age group of 15-50 years in the age-distribution curve of SHI (Figure 2). This could be credited to this group of people predominantly being more active and vulnerable in the community and country. In the present study, RTA was established as the most common cause of SHI. It contributed to almost 55% of the head injury cases leading to admission (Figure 3). This finding can be attributed to more men being drivers and more prone to be involved in RTA (5, 6). Noticeably, cases of head injuries due to the physical assault appear to be on the rise, compared to zero percent in the study of Gongal and Devkota (1979) to 6% in the present series (4).

In Nepal, the zero-tolerance policy against drinking and driving significantly reduced the number of RTA in past years. However, two third of the patients with SHI, in this study were under the influence of alcohol. Patients with SHI with GCS \leq 8 were intubated and mechanically ventilated in ICU. Nearly all patients in the present study underwent CT imaging of the head immediately after emergency admission. Neurosurgical procedures were

performed between admission to the emergency room and transfer to intensive care unit.

Initial CT examination demonstrated abnormalities in approximately 90% of patients with severe head injury. CT scan plays a vital role in early assessment of patients with severe head injury. Repeat CT scan was done within 48-72 hours to exclude conversion of contusion into increase hematoma. Our study showed, patients with SHI developed subdural hygroma after a week, as seen on their CT scans. Subdural hygroma is defined as acute or chronic cerebrospinal fluid accumulation in the subdural space, which is a virtual space between the arachnoid and subdural membranes (17). In our series, bilateral subdural hygroma were located mainly frontoparietal region representing more than 77% of the cases. In the literature, this sort of lesion is usually related with contusions, cerebral atrophy, subdural and epidural hematomas or post operative complications (12, 15, and 17). Although etiology is well documented, pathogenesis of this condition is not well explained. Some author proposed that the formation of a one-way valve allowing the passage of CSF into the subdural space, caused by a traumatic tear of the arachnoidal mater or rupture of incidental arachnoids cyst (7, 13). In our series, we noticed prognosis of patients could be good once bilateral subdural hygroma in the CT scan.

The study also revealed a correlation between tattoo and SHI. 45% of RTA cases with SHI had tattoo. However, present research does not indicate any particular group, age, gender, occupation, personality or a particular level of social status that get tattoos. There are many reasons to why people get tattoo; describing personal tales, expressing individuality, spiritual meaning, fashion, scar hiding, marking new beginnings like birth, adulthood, marriage, divorce, death, etc. and even marking memories as milestone for love or loss (14).

Another interesting finding of our study, Blood grouping pattern in SHI were B+, A+ and O+ 36.1, 28 and 24.3% respectively (Figure 7). In our country, normal blood grouping distribution are A, O, B and AB at 55.05%, 21.64%, 14.72%, and 8.6% respectively (8). But O, A, B and AB were 46%, 41%, 9% and 4% in the USA respectively (10). These findings can be incidental or crucial. Further study needs to be conducted as our study had its limitation; small study group, single center findings and limited follow up to understand its outcome. We strongly recommended

that prediction factors should be kept in mind for the best management and outcome of severe head injury patients. Head trauma should highly be prevented in order to reduce incidence of TBI related mortality. Thus, awareness of the impact of head trauma should be raised through information campaigns.

CONCLUSION

Severe head injury has a high negative impact on young people, especially men and also B+ blood group. The age of the patient, presence of Tattoo, GCS at admission, CT scanning at admission and CT scanning after week of admission with hygroma were significant predictors of outcome.

REFERENCES

1. Baguley IJ, Nott MT, Howle AA, Simpson GK, Browne S, et al. Late mortality after severe traumatic brain injury in New South Wales: a multi center study. *Med J Aust*, 2012; 196: 40-45.
2. Brazinova A, Rehorcikova V, Taylor MS, Buckova V, Majdan M, et al. Epidemiology of Traumatic Brain Injury in Europe: A Living Systematic Review. *Journal of Neurotrauma* 2016; 33:1-30.
3. Cesar Reis et al. What's New in Traumatic Brain Injury: Update on Tracking, Monitoring and Treatment; *International Journal of Molecular Science*, 2015; 16: 11903-11965
4. Gongol DN, Devkota UP. An analysis of head injuries in children of Nepal. *Journal of Nepal Medical Association (JNMA Souvenir)*: 1979;181-9.
5. Husson EC, Ribbers GM, Willemse-van Son AH, Verhagen AP, Stam HJ, et al. Prognosis of six-month functioning after moderate to severe traumatic brain injury: a systematic review of prospective cohort studies. *J Rehabil Med*, 2010; 42: 425-436.
6. Koliass AG, Guilfoyle MR, Helmy A, Allanson J, Hutchinson PJ: Traumatic brain injury in adults. *Pract Neurol*, 2013; 13: 228-235.
7. Kusuno, K., Yoshida, Y., Takahashi, A., & Ishii, S.: Chronic subdural hygroma caused by rupture of arachnoid cyst. As a probable course of chronic subdural hematoma--case report. *Neurologia Medico-Chirurgica*, 1984; 24(5), 349-354. <http://dx.doi.org/10.2176/nmc.24.349>.
8. Lava Shrestha, uzwali malla: ABO and Rh Blood Groups and their Ethnic Distribution in a Teaching Hospital of Kathmandu, Nepal, 2013; Vol 52 190. DOI: <https://doi.org/10.31729/jnma.2112>
9. Maas AI, Steyerberg EW, Butcher I, Dammers R, Lu J, Marmarou A, et al. Prognostic Value of Computerized Tomography Scan Characteristics in Traumatic Brain Injury: Results from the IMPACT Study. *J Neurotrauma*. 2007; 24(2): 303-314.

10. Mollison PL, Engelfriet CP, Conteras M. The Rh blood Group system. In *Blood Transfusion in Clinical Medicine*, 9th Edition. Oxford: Black well Scientific Publication. 1993; 2008-09.
11. Murray GD, Butcher I, McHugh GS, Lu J, Mushkudiani NA, Maas AI, et al. Multivariable prognostic analysis in traumatic brain injury: results from the IMPACT study. *J Neurotrauma* 2007; 24(2): 329-337.
12. Park, J., Cho, J.-H., Goh, D.-H, et al. Postoperative Subdural Hygroma and Chronic Subdural Hematoma after Unruptured Aneurysm Surgery: Age, Sex, and Aneurysm Location as Independent Risk Factors. *Journal of Neurosurgery*, 2015; 1-8.
13. Pillai, P., Menon, S. K., Manjooran, R. P., Kariyattil, R., Pillai, A. B., & Panikar, D. Temporal Fossa Arachnoid Cyst Presenting with Bilateral Subdural Hematoma Following Trauma: Two Case Reports. *Journal of Medical Case Reports*, 2009; 3(1), 53. <http://dx.doi.org/10.1186/1752-1947-3-53>.
14. Shannon Bell; Tattooed: A Participant Observer's Exploration of Meaning, *The Journal of American Culture*, 2004; 22(2):53-58: DOI: 10.1111/j.1542-734X.1999.2202_53.x.
15. Sun, H.-L., Chang, C.-J., & Hsieh, C.-T. Contralateral Acute Subdural Hematoma Occurring after Evacuation of Subdural Hematoma with Coexistent Contralateral Subdural Hygroma. *Neurosciences (Riyadh, Saudi Arabia)*, 2014;19(3), 229-232.
16. Traumatic Brain Injury-Definition, Epidemiology, Pathophysiology: Segun Toyin Dawodu, Updated Aug 16, 2017; <https://emedicine.medscape.com/article/326510-overview>.
17. Zanini, M. A., Resende, L. A. D. L., Faleiros, A. T. D. S., & Gabarra, R. C. Traumatic Subdural Hygromas: Proposed Pathogenesis Based Classification. *The Journal of Trauma*, 2008; 64 (3), 705-713. <http://dx.doi.org/10.1097/TA.0b013e3180485cfc>.



Cerebral revascularization by EC-IC bypass in ischemic conditions of different etiologies

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ABSTRACT

Background: Even after the failure of EC-IC bypass trial, EC-IC bypass can help many patients in preventing future stroke. Here we present a case series of patients with cerebral ischemia from different etiological modes who underwent EC-IC bypass with positive end results.

Methods: Patients in these cases series with TIA/stroke/recurrent stroke were evaluated clinically for the history of TIA or recurrent/hemodynamic TIA (in rest or during work) or progressive hemiparesis/aphasia/visual disturbances or sudden hemiplegia/hemiparesis/aphasia with subsequent significant (days to a week) recovery. MRI of the brain was done in ischemic protocol in all cases. To see the arterial pathology dynamic CTA was also done in all cases except one case. DSA was done in 03 cases. When clinical features, cerebral ischemia on MRI and arterial stenosis/occlusion on angiogram were concordant with each other, only then cerebral revascularization was done. After bypass, all patients were followed up regularly. All recorded data were reviewed retrospectively.

Results: Total no. of cases were 08. The most common presentation was hemiparesis. Etiologies were infective thrombosis of ICA, orbital cellulitis, thrombosed giant ICA aneurysm, single & multiple vessel occlusion and MCA stenosis. High flow EC-IC bypass was done in one case. STA-MCA bypass was done in rest of the cases. All patient were ambulant with static neuro-status without new stroke till last follow up. All bypasses were patent and functioning till last follow up (clinical, Doppler/Imaging).

Conclusion: In carefully selected cases cerebral revascularization in ischemic conditions can result positive outcome.

INTRODUCTION

After introduction by Yasargil in 1967, cerebral revascularization by EC-IC bypass has become an indispensable tool for managing patients with hemodynamic ischemia, or for managing patients with complex aneurysms or skull base tumors; that are not amenable to radical resection; because of major vascular involvement. [2,14] Patients with

Keywords

cerebral revascularization,
EC-IC bypass,
ischemic conditions,
different etiologies



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hemodynamic ischemia have an annual stroke rate of 25%, which increases by 2% every year.[14] They can develop a fatal stroke. This category also includes Moya moya disease. [14,18] After failure of EC-IC bypass trial, neurosurgeons were in search of cases where EC- IC bypass would help the patients in preventing future stroke and neurological improvement. Here we present a case series of patients with cerebral ischemia from different etiological modes underwent EC-IC bypass with positive end results.

METHODS

Patients in these cases series with TIA/stroke/recurrent stroke were evaluated clinically for history of TIA or recurrent/hemodynamic TIA (in rest or during work) or progressive hemiparesis / aphasia / visual disturbances or sudden hemiplegia / hemiparesis / aphasia with subsequent significant (days to week) recovery. Permanent hemiplegia cases were not included in this case series. Then the cases were evaluated neuro-radiologically for cerebral ischemia with or without infarct/s and possible cerebral revascularization by EC-IC bypass. CT scan of head was done to exclude hemorrhage and other pathology such as tumor. MRI of brain was done in ischemic protocol (All images including DW, ADC, PW, DTI and MRA&MRV including neck vessels) to see cerebral ischemic zone/s (DW& PW mismatch), cortico-spinal tract & other major tracts and intracranial or extracranial arterial stenosis. To see the arterial pathology dynamic CTA was also done in all cases except one case. DSA was done in 03 cases. When clinical features, cerebral ischemia on MRI and arterial stenosis/occlusion on angiogram were concordant with each other, only then cerebral revascularization by EC-IC bypass was done. After bypass all patient were followed up regularly (clinically and radiologically). All recorded data were reviewed retrospectively. All bypass operation was done from January 2015 to March 2018

REPRESENTATIVE CASES

Case 1 (Table 1, 2; Figure 1 - a, b, c)

A 38 years old male presented with headache, vomiting, dropping of left eyelid and visual disturbance. Left sided visual acuity reduced to finger count with left sided complete ophthalmoplegia. CT scan of head showed pan-sinusitis with skull base osteitis. MRI of head showed

pan-sinusitis with both cavernous sinus involvement and right parietal infarct (Figure 1a-A,B&C). He underwent diagnostic endoscopic rhino-sinuscopy, which showed rhino-sinusitis involving all sinuses & nose with pseudo-membranes. Biopsy reported inflammatory sinusitis. Fungal and tubercular cultures were negative. Routine bacterial culture-revealed methicillin resistances *Staphylococcus aureus* (MRSA). Then he was put on high dose injectable antibiotic for three week that resulted significant radiological improvement (Figure 1a-D,E&F) and then patient was discharged with oral antibiotic for three month.

Three weeks after discharge he again presented with altered level of consciousness and right sided hemiplegia (UE>LE, Muscle power in right UE MRC grade 1/5 and in lower limb 2/5) and motor aphasia. MRI of head showed resolving rhino-sinusitis with patchy infarcts and ischemic zone involving fronto-parieto-occipital zones especially on left side and old infarct on right parietal area (Figure 1b). MRA showed bilateral complete occlusion of ICA and whole brain was perfused by basilar artery (Figure 1c-A&B). On urgent basis the patient underwent left-sided STA-MCA bypass.

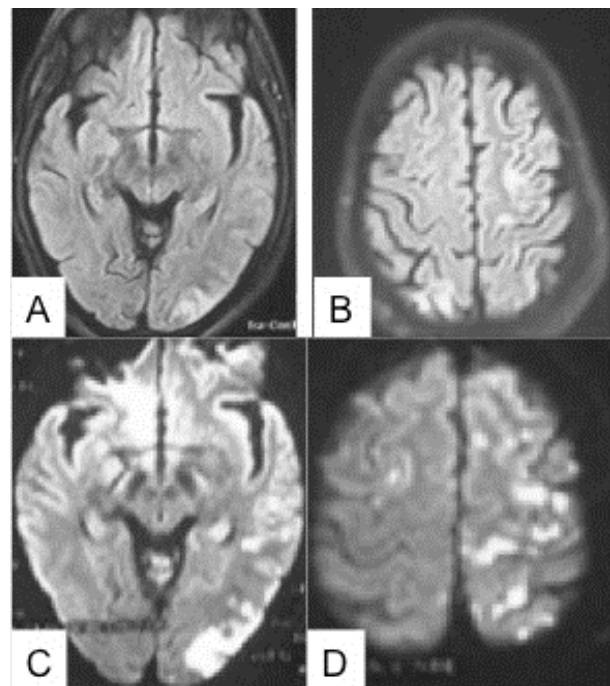


Figure 1.

Operation

Under GA the patient was positioned typically supine

with the head turned more than 60°. At this point, we used digital palpation technique and a handheld Doppler probe to map out the course of the STA both frontal and parietal branches.

The incision was started at the level of the zygoma and carried up to near midline behind the hairline. Both branches were procured up to superior temporal line very carefully to avoid thermal damage or avulsion injury for branches. Papaverine solution and plain local anesthetic agent (2% lidocaine) was used to irrigate the STA for prevention of vasospasm. A mini pterional craniotomy was done very carefully (not to damage the procured STA). After durotomy a small posterior Sylvian split was done to find out a suitable M3 as a recipient vessel for bypass. Among the frontal and parietal branch the suitable and larger frontal branch was used to make a STA-MCA anastomosis. After bypass patency was checked clinically and with micro-doppler. Dura was loosely closed around the STA (not water tight). Along the temporal margin of bone flap a portion was removed so that the STA would not be kinked or compressed by the bone. Mini plates and screws were used to fix the bone flap. Rest of the wound was closed accordingly without drain.

Postoperative course

Patient recovered well from anesthesia. In postoperative days the patient recovered hemiparesis quickly. By the end of 7th POD patient became ambulant. By the end of 04 weeks after operation he returned to his professional work and muscle power on right side body improved at the level of MRC grade 4+/5. Post-operative CT scan showed no hematoma or new infarct. CTA showed patent STA-MCA bypass on left side (Figure 1c-C&D).

Case 2 (Table 1, 2; Figure 2 - a, b, c)

A 55 years old policeman presented with recurrent occasional episode of fall during walking without loss of consciousness or convulsion and unable to move right side of body with aphasia lasting for 7-10 minutes. The frequency of events increases last few months and reached 3-5 times/day for last few days before presented to us. He was on adequate anti platelet therapy. He was a smoker but non-diabetic and non-hypertensive. CT scan of head showed multiple infarcts especially in left cerebral hemisphere. CTA showed absent both VA and left ICA (Figure 1a) with scarcity of left. MCA, PCA and

posterior fossa vessels. MRI of brain showed multiple old infarcts with ischemic areas especially in left hemisphere and ischemic zone in left PICA area. He was advised for urgent cerebral revascularization. But the patient developed left PICA infarct 06 hours before the scheduled 'urgent' operation (Figure 2b-A&B). In this situation we proceeded for cerebral revascularization by left-sided CCA-RA-MCA high flow bypass by keeping in mind that posterior fossa decompression might be needed at any time.

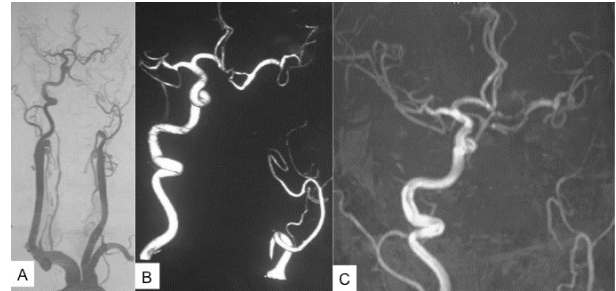


Figure 2a.

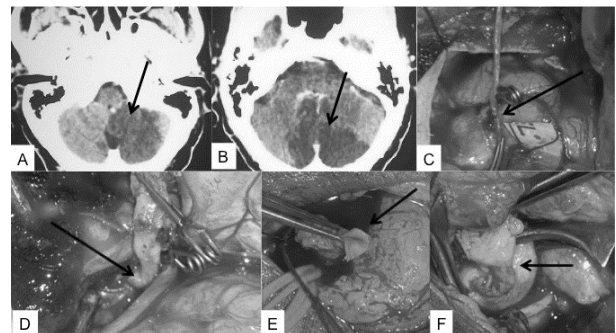


Figure 2b.

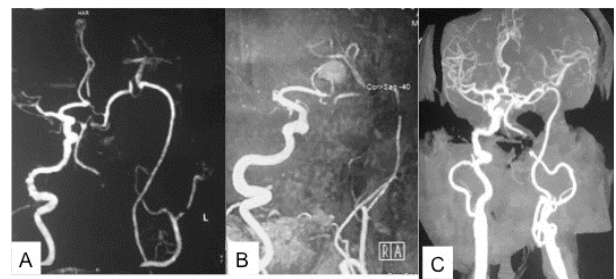


Figure 2c.

Operation

Under general anesthesia with endotracheal intubation patient was placed in supine position. Head was fixed with 3-pin head holder with neck extension and head turning to the opposite (right) side (30°). Head end of the table was elevated (20°).

Eye, ear, pressure points and nerves areas were protected. Left upper limb placed on a side 'limb rest' in extended elbow, 30° abducted from the trunk in supine for radial artery procurement. After preparation, left front of the forearm, right side of the neck and left pterional areas were draped properly.

With longitudinal incision, radial artery was harvested from brachial bifurcation at elbow to wrist (20 cm). The artery was distended with intraluminal injection of heparin and papaverine mixed normal saline. Then the artery was kept in heparin and papaverine mixed normal saline. The forearm wound was closed with a drain.

A curve incision on the left side of the neck was made from the tip of mastoid and extended downward & medially 2cm posterior to the angle of mandible to the mid line. After cutting platysma and investing deep fascia sternocleidomastoid muscle was retracted laterally. With further dissection posterior belly of digastric muscle, hypoglossal nerve, internal jugular vein, common carotid, internal carotid and external carotid artery with its branches were identified.

A left-sided pre-coronal post hairline curvilinear incision was made and superficial temporal artery (STA) and its parietal branch was procured and prepared for STA-MCA insurance bypass as donor artery. A temporally extended pterional craniotomy was done. Temporal bone was removed down to the middle fossa floor. In the cervical wound, a blunt index finger dissection was made in between the digastric muscle and hypoglossal nerve upward and superiorly to styloid process and then finger dissection was continued upward, medially & anteriorly to lateral pterigoid plate. A curved medium sized artery forceps was passed from middle fossa floor to the fingertip and with finger guidance the arterial tip was brought out into the cervical wound and then A 26Fr thoracostomy tube was passed from cervical wound to the middle fossa floor. Radial artery (RA) graft was passed from middle fossa floor to cervical wound through the tube. With stabilization of both ends of RA graft thoracostomy tube removed. RA graft was made twist free by injecting heparinized solution into the lumen.

After durotomy a STA -MCA (Temporal M4) 'insurance bypass' was done with 10/0 nylon and checked for patency and function with micro Doppler (Figure 2b-C). After Sylvian dissection temporal M2

was identified and prepared for bypass. Cranial end of RA graft was also prepared for bypass and the RA graft and temporal M2 bypass was made after systemic heparinization with 3000 unit of injection heparin (Figure 2b-D). The patency of anastomosis was checked by retrograde flow of blood through the caudal end of RA graft in cervical wound.

With the control of common carotid artery (CCA) an anastomosis was made between caudal end of RA graft and CCA (Figure 2b-E&F). The patency and flow through the anastomoses and RA graft were checked with micro doppler. Cervical wound and craniotomy wound were closed with drains.

Postoperative course

Postoperatively, he was on tab. Aspirin and inj. Heparin. CT scan on 1st postoperative day (POD) showed no infarct or any gross hematoma (Figure). CT angiogram on second POD showed left ECA-RAG-M2 bypass with pre-operative PICA infarct without further swelling. Post operatively he became non-communicable and he used shout with inappropriate & slang words especially at night and did not want to take food. Gradually these symptoms improved within three weeks and became communicable but improvement of intellectual functions was slow and incomplete. He became continent by three weeks. Within one week he could stand and walk though there was some instability. He became fully ambulant without any cerebellar dysfunction by three months. By the end of one year after operation his intellectual and cognitive function returned to near preoperative level and he returned to his job without any further hemodynamic TIA. CT scan and CTA after one year showed patent bypass without any new infarct.

RESULTS

Total no. of cases were 08. Six were male and 02 were female. Age range was 25-57 (average 38 years). Follow up period was 12-48 months. Details of all cases were shown in Table 1 & 2 (Figure 1-7).

Most common presentation was hemiparesis. Infection was the etiology of thrombosis of cavernous ICA in two cases. In one case orbital cellulitis spread in CS and ICA with aneurysm was thrombosed (Figure 6-A,B&C). In the other cases pan rhino-sinusitis (of MRSA) spread to both cavernous sinus and both ICA was occluded. In one interesting case giant partially thrombosed ICA bifurcation

aneurysm thrombosed totally with distal ICA, A1 and M1 (Figure 7). Acute thrombosis of ICA with aneurysm in CS occurred in two female cases where both developed hemiparesis and one was 3 months pregnant. In one case there was intractable TIA with impending major stroke where whole brain was supplied by only right sided ICA and he developed PICA infarct 12 hours before the 'scheduled urgent' revascularization operation; Only high flow EC-IC bypass was done in this case (Figure 2a,b&c). Post operatively he developed 'behavioral, intellectual and? psychogenic' symptoms that recovered slowly.

STA- MCA bypass was done in rest of the cases. MCA stenosis was the etiology in two cases; one was of 27 years of age (Figure 3). Average ischemic time was 28 minutes (range 25-35 minutes). There was no clamp related infarction. In one case patient developed postoperative insulo-frontal infarct that not related to temporary clamping (Figure 6-C, D & F). All patient improved neurologically. All patient were ambulant with static neuro-status without new stroke/TIA till last follow up. All bypass were patent till last follow up (clinical, Doppler/Imaging).

Table 1. Particulars of cases (age, sex, clinical presentations, investigations and image finding/s).

No	Age/ sex	Presentation/s	Investigation/ s	Image finding/s
1. (Figure1)	38/M	Headache, vomiting, dropping of left eyelid, visual disturbance. Left sided visual acuity reduced to finger count with left sided complete Ophthalmoplegia, Three weeks later (on antibiotics): presented with altered level of consciousness and right sided hemiplegia (UE>LE, Muscle power in right UE MRC grade1/5 and in lower limb 2/5) and motor aphasia	CT MRI Diagnostic sino-nasal endoscopy MRI MRA	Pansinusitis with skull base osteitis pansinusitis with both cavernous Sinus involvement and right parietal infarct Rhino-sinusitis involving all sinuses with pseudomembranes *Biopsy-Inflammatory sinusitis *Culture: <i>Fungal-Negative, Tubercular-Negative, Routine bacterial culture- Multiple antibiotic resistance Staphylococcus Aureus</i> Resolving rhino-sinusitis with patchy infarcts and ischemic zone involving fronto-parieto-occipital zones especially on left side. Old infarct on rt. parietal area. Bilateral complete ICA block (whole brain is perfused by basilar artery)
2. (Figure2)	55/M	Recurrent episode of fall during walking and unable to move lt. side of body with aphasia,5-8 times/day before presentation even with	CT scan CTA	Multiple infarcts specially in lt. hemisphere Absent both VA and lt. ICA. Scarcity of lt. MCA, PCA and posterior fossa vessels

		adequate anti platelet therapy	MRI MRA	Multiple old infarcts with ischemic areas especially in Lt hemisphere. Ischemic Lt PICA area. The pt. developed Lt. PICA infarct 12 hours before the scheduled 'urgent' operation.
3. (Figure3)	27/M	H/O TIA (recurrent hemiparesis and aphasia), sudden rt. Hemiplegia (MRC grade-2/5), and global aphasia	MRI CT & CTA DSA	Cerebral infarcts and ischemic zones in left hemisphere especially Parieto-occipital lobe. CT-Cerebral infarcts and ischemic zones in left hemisphere. CTA- left M1 bifurcation stenosis. Scarcity of Lt. MCA vessels Left M1 bifurcation stenosis/occlusion with delayed filling of MCA territory.
4. (Figure4)	45/M	Headache, left hemiparesis (UE>LE, Grade1/5) and motor aphasia. H/O TIA. One-week later motor power on left side 2/5.	CT CTA MRI MRA	Right parieto-occipital and right periventricular (corona radiata) infarcts. Complete occlusion of right-sided ICA from neck to ICA bifurcation with decrease vasculatures on right MCA zone. Right parieto-occipital and right periventricular (corona radiata) infarcts/ischemia with diffusion -perfusion mismatch. Tractography showed intact major tracts. Complete occlusion of right sided ICA from neck to ICA bifurcation with decrease vasculatures on right MCA zone
5. (Figure 5)	27/F	Sudden headache, eyeache, vomiting, rt. ptosis with complete ophthalmoplegia, Lt. hemiparesis, 2 nd trimester pregnancy	MRI & MRA DSA	MRI-right cavernous sinus thrombosis with 'target' sign. MRA-occlusion of rt. ICA with giant CS ICA aneurysm (thrombosed). Scarcity of rt. MCA vessels Rt. ICA occlusion with scarcity of blood flow in rt. MCA
6. (Figure 6)	25/F	Fever, Lt. proptosis, headache, eyeache, vomiting, Lt. ptosis with complete ophthalmoplegia, rt. hemiparesis	CT scan MRI & MRA	CT-Lt eye proptosed otherwise normal MRI-Lt cavernous sinus thrombosis with 'target' sign. Lt eye was proptosed.

				MRA-occlusion of lt. ICA with CS ICA aneurysm (thrombosed). Scarcity of lt. MCA vessels with M1 narrowing
7.	57/M	H/O TIA with motor dysphasia and hemiparesis. Rt. Hemiparesis (3/5) and global aphasia	CT scan & CTA MRI MRA	CT-left ganglio- capsular, periventricular, parietal and frontal small infarct/ischemias. CTA- left M2 (upper trunk occlusion) and M2(lower trunk stenosis) Left ganglio- capsular, periventricular, parietal and frontal small infarct/ischemias. Left M2 (upper trunk occlusion) and M2(lower trunk stenosis)
8. (Figure7)	35/M	Initially headache, visual disturbance. Later visual problem improved with progressive left. Spasticity and hemiparesis (3/5)	CT & CTA MRI CT & CTA (4month later) DSA	Right ICA bifurcation Giant partially thrombosed aneurysm Right sided giant partially thrombosed aneurysm Aneurysm size decreased to less than half. Anterior corona radiata infarct. No aneurysm on CTA with occlusion of rt. ICA bifurcation. Scarcity of rt. MCA vessels Non-visualization of right ICA bifurcation, A1 and M1 with aneurysm. Delayed filling of right MCA territory through cortical anastomoses. Right ICA only supplied the right PCA through PCom

[M-male,F-female, H/O-history of, Rt/rt-right, Lt/lt-left, UE-upper extremity, LE-lower extremity, TIA-transient ischemic attack, MRC-medical research council, CT-computed tomography, CTA-CT angiogram, MRI-magnetic resonance imaging, MRA- MR angiogram, DSA-digital subtraction angiogram, ICA-internal carotid artery, MCA-middle cerebral artery, PCA-posterior cerebral artery, Pcom-posterior communicating artery, CS-cavernous sinus, PICA-posterior inferior cerebellar artery, VA-vertebral artery]

Table 2. Etiology of infarct/ischemia, indication of operation, surgical treatment, complication/s and outcome of bypass.

No	Etiology	Indication/s	Surgical treatment	Complication	Outcome
1. (Figure 1)	Bilateral complete occlusion of ICA due infective inflammatory involvement of cavernous sinus	Acute on chronic hemodynamic ischemia of whole brain specially ICAs territory.	Left STA-MCA bypass	None	Ophthalmoplegia, hemiparesis and aphasia recovered completely. Rhinopansinusitis resolved completely

2. (Figure 2)	Chronic occlusion of three out of 04 intracranial arteries caused hemodynamic stroke and ischemia	Chronic and hemodynamic ischemia specially left hemisphere and cerebellum	Radial graft high flow EC-IC (CCA-RA-M1) bypass with insurance STA-MCA bypass	Early post operative period-behavioral changes	Behavioral changes recovered. No hemodynamic TIA.
3. (Figure 3)	Acute occlusion of M1 bifurcation on Chronic MCA stenosis	Acute on chronic ischemia	Left STA-MCA bypass (parietal)	None	Almost near total recovery of neuro-deficit.
4. (Figure 4)	Complete occlusion of Right ICA from neck.	Ischemic right cerebral hemisphere (with infarcts) specially MCA territory	Right STA- MCA bypass	None	Hemiparesis improved (MRC grade-4/5). Aphasia improved to dysphasia
5. (Figure 5)	Sudden ICA occlusion with occlusion of giant aneurysm in CS ICA	Ischemic hemiparesis (with CS thrombosis)	STA-MCA bypass with decompression of CS	None	Hemiparesis and ophthalmoplegia recovered completely. Normal delivery of healthy baby
6. (Figure 6)	Sudden ICA occlusion with occlusion of aneurysm in CS ICA due to orbital cellulitis spread in to lt. CS	Ischemic hemiparesis (with infective CS thrombosis)	STA-MCA bypass with decompression of CS. C/S of CS purulent content Injectable antibiotic (long term)	Anterior insular cortex and Broca's area infarct-motor dysphasia	Hemiparesis, dysphasia, ophthalmoplegia recovered completely
7.	Acute occlusion of M2 on chronic stenosis	Acute on chronic ischemia	Left double STA -MCA bypass (parietal and temporal)	None	Motor recovery-near normal, Global aphasia converted to sensory aphasia
8. (Figure 7)	Thrombosis of giant aneurysm with progressive occlusion of ICA bifurcation	Right MCA territory ischemia	Right STA- MCA bypass	None	Hemiparesis improved (MRC grade-4/5).

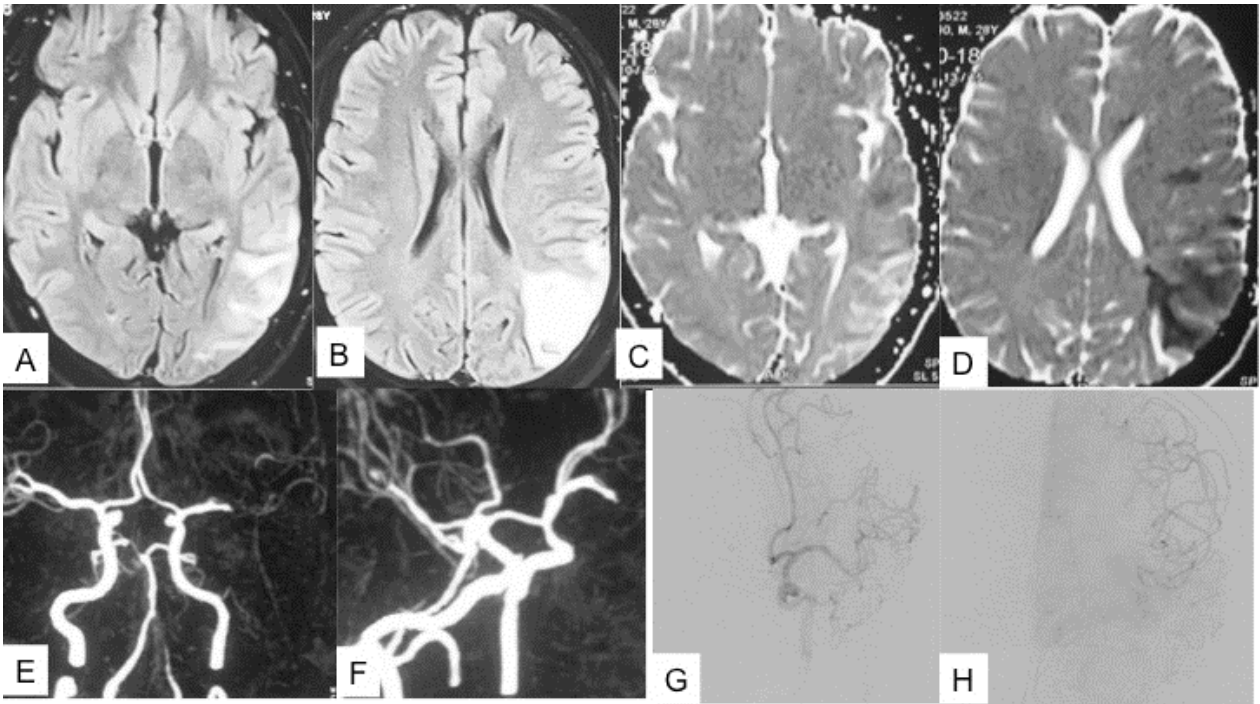


Figure 3a.

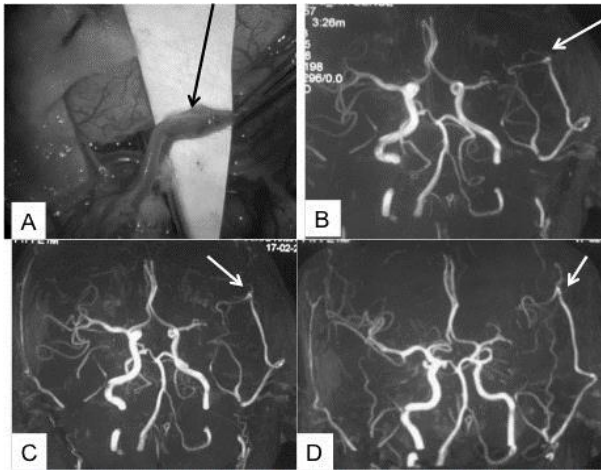


Figure 3b.

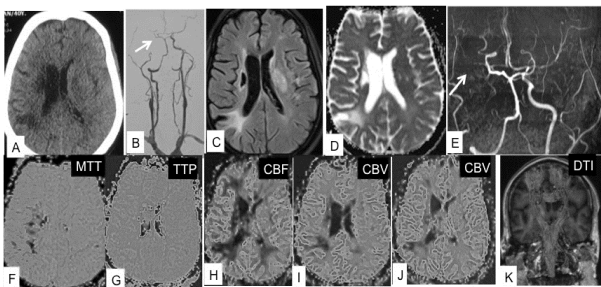


Figure 4a.

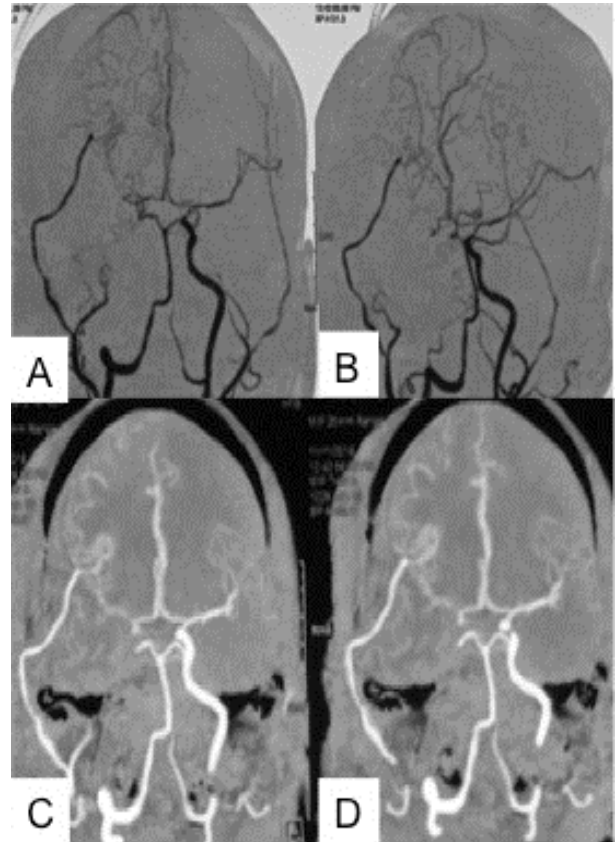


Figure 4b.

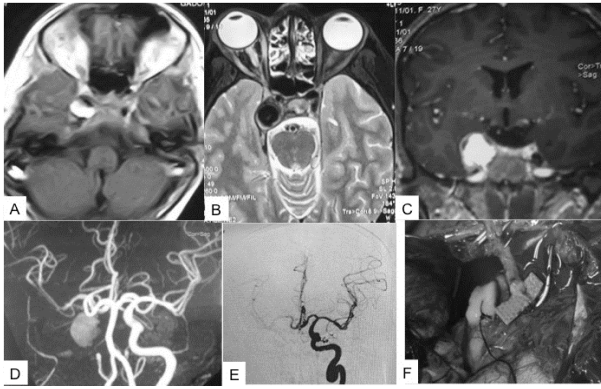


Figure 5a.

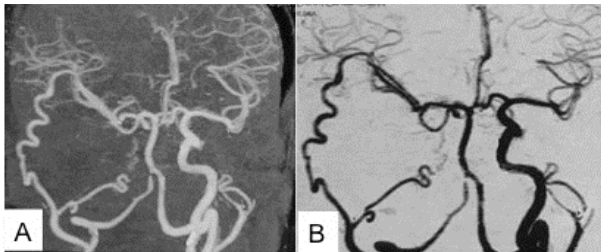


Figure 5b.

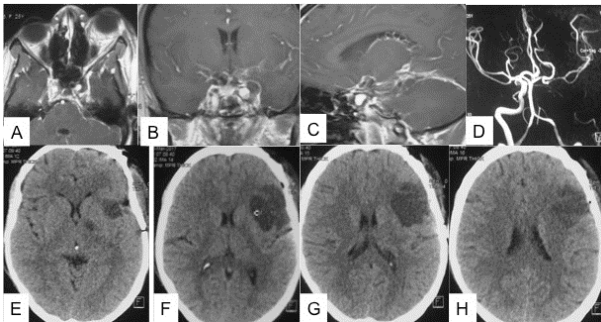


Figure 6.

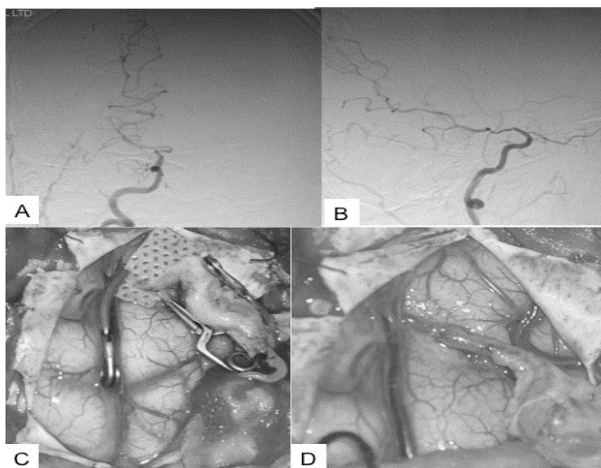


Figure 7.

DISCUSSION

In 1961, Pool and Potts [2,9] first attempted cerebral revascularization with a synthetic graft using a plastic tube to create a superficial temporal artery (STA) to anterior cerebral artery shunt but angiography showed thrombosed tube though patient recovered and survived. In 1963, Woringer and Kunlin [11] performed the first extracranial to intracranial (EC-IC) bypass of the common carotid artery (CCA) – intracranial (IC) internal carotid artery (ICA) using a saphenous vein (SV) graft but patient did not survive, while the graft was patent on autopsy.

In 1967, Yaşargil performed the first EC-IC bypass in a patient with an occluded ICA.[10] In 1972 Yaşargil also started STA-middle cerebral artery (MCA) bypass for moyamoya disease. In 1971 Lougheed did the first EC-IC bypass using an SV graft, while in 1978 Ausman performed EC-IC bypass using a radial artery graft. In the 1970s, Sundt et al.[3] and others performed posterior circulation revascularization to treat steno-occlusive disease, vertebrobasilar insufficiency, and unclippable complex aneurysms.

EC-IC bypass study (1977-1985) [15] evaluated the result of EC-IC bypass as a means to decrease the subsequent stroke rate for the treatment of “symptomatic atherosclerotic lesions of the ICA and/or MCA.” But the study failed to show significant difference between the EC-IC bypass group and medically treated group. The study identified two important subgroups that appeared to do EC-IC bypass; 1. Patients with severe MCA stenosis and 2.those with the persistence of ischemic symptoms in known ICA occlusion.

IC or MCA atherosclerosis results to an ischemic cerebrovascular event through: (1) Hypo-perfusion, (2) thrombosis at the site of stenosis, (3) thromboembolism, and (4) direct occlusion of small perforating vessels. [9,16]

The clinical presentation may vary from an acute ischemic deficit to intermittent neurological symptoms. The pathophysiologic changes of chronic hypo-perfusion/ischemia have been categorized into three stages-

Stage 0: Normal hemodynamics; Stage 1: Reflex vasodilation in response to inadequate collaterals and a falling perfusion pressure with resultant increases in cerebral blood volume and prolongation of mean transit time, but with preservation of cerebral blood flow (CBF) and normal oxygen extraction fraction (OEF); Stage 2: Misery perfusion in

response to cerebral perfusion pressure falling below the range of auto-regulatory capability exemplified by falling CBF and increasing OEF and maintenance of the cerebral metabolic rate of oxygen (CMRO₂). [9]

Progressive MCA stenosis, severe stenosis (>70% stenosis), female gender, National Institutes of Health Stroke Scale score >1, concurrent diabetes, borderline body mass index values, hyperlipidemia, white ethnicity, and the presence of hemodynamic stenosis increases the risk for stroke. [9]

The beginning assessment of transient ischemic attack (TIA) or ischemic stroke should include computed tomography (CT) of head to exclude the possibility of IC hemorrhage (and rarely other pathologies such as cavernous sinus mass, giant thrombotic aneurysm etc.) and estimate the extent of ischemic change in consideration of thrombolysis. Diffusion-weighted (DW) magnetic resonance imaging (MRI) with magnetic resonance angiography (MRA) including neck vessels, and perfusion studies can be subsequently obtained to better delineate the extent of infarct and/ ischemia as well as provide more information for further management strategies. [16] TCD, angiography, MRA, and computed tomographic angiography [CTA] or parenchymal perfusion (CT perfusion, positron emission tomography [PET], single-photon emission computed tomography [SPECT], and magnetic resonance perfusion studies are the main neuro-radiological evaluations to see the intra cranial flow related pathology. MRA and CTA both provide excellent detail regarding the caliber of vessels; although, MRA has been shown to overestimate the degree of stenosis in some cases. These noninvasive techniques were sufficiently accurate to exclude more than 50% stenosis, but further confirmatory studies were needed to characterize the stenosis and digital subtraction angiography (DSA) is the gold standard. Perfusion studies [16] (include PET scans, SPECT scans, xenon CT perfusion studies, CT perfusion, and perfusion MRIs) permit the extrapolation of CMRO₂, OEF, and CBF and provide information on the perfusion of the brain. The Carotid Occlusion Surgery Study [17] employs measuring OEF by PET.

Since the first STA-MCA procedure was described by Yasargil, [2,18] many variations have been reported but STA -MCA bypass remains the main workhorse of a vascular neurosurgeon. Many of

these variations have been developed in dealing with complex intracranial aneurysms and skull base tumors. These variations include anastomoses between the bilateral anterior cerebral arteries; occipital artery-to-posterior, inferior cerebral artery (PICA), anterior and inferior cerebral artery (AICA). Others includes PICA to PICA, vertebral artery to PICA, STA to SCA or PCA, subclavian artery to PCA, PCA to SCA, and even a tandem occipital artery to AICA and PICA anastomoses. [9]

Complications in STA-MCA bypass are limited and include early postoperative TIA, delayed stroke, development of a pseudo-aneurysm, and wound dehiscence. High-flow bypass grafts is more prone to develop complications than low-flow STA-MCA bypass. Radial artery grafts may suffer vasospasm or intimal hyperplasia and eventually occlude. Pro-atherogenic changes can occur in SV grafts, which eventually leads to occlusion. After parent vessel occlusion thromboembolic complications are common after high flow bypass mainly due to the change in intracranial hemodynamics. Preoperative antiplatelet medications, as well as intraoperative anticoagulation, can prevent these thromboembolic events. In patients without vascular reserve, prolonged temporary occlusion times can lead to territory infarcts without changes in the neuro-monitoring. So it is important to minimize occlusion times in these patients. In longstanding perfusion deficiency, reperfusion hemorrhage may be problematic after revascularization, though the incidence is low. Other complications involve the site of graft harvests such as infection, ischemic hand, or hematoma. [9]

Here in representative case 1 (high flow bypass), there were postoperative behavioral, intellectual and cognitive dysfunctions that recovered slowly. There may be hyper-perfusion of chronically ischemic brain tissue but postoperative radiology did not show any hyper-perfusion signs. Retrogradely we thought, could we avoid such complication by giving low flow bypass.

Most of the ICA and other intracranial arterial stenosis or occlusion (acute or chronic) leading to cerebral infarct/ischemia is caused by atherosclerosis predisposing and precipitating by many factors. Other causes include dissection, vasculitis, vasospasm and Moyamoya disease. [5,13]

In this series most of the cases are unique from of etiological point. Spontaneous occlusion of

cavernous ICA was reported in the literature but such occlusion associated with cerebral ischemia was not reported.[8] Infection from paranasal sinuses or orbit usually spread into cavernous sinus but usually does not occlude the ICA; in case of fungal infection, fungus may erode the arterial wall and can cause mycotic aneurysm with distal embolism or occlusion of ICA.[4,12]

Giant partially thrombosed ICA bifurcation aneurysm case, at initial presentation patient refused operation for aneurysm then he returned with left hemiparesis with complete occlusion aneurysm as well as occlusion of parent ICA with A1 and M1 (proximally up to PCom). ICA was only supplying right PCA through PCom. Young patient with MCA stenosis recovered near to normal neurology.

In our series, in a few cases, we did revascularization on urgent basis. Nussbaum ES et al [6] did emergency EC-IC bypass in patients with acute ischemic injury, which was seemed to be safe and effective where patients were relatively young. They found bypass was successful in arresting progression of stroke, and in some cases resulted in rapid neurological improvement. Although the study on EC-IC bypass failed to show a benefit from the bypass procedure Nussbaum ES et al [7] continued to perform the operation in selected cases. Carefully selected individuals with occlusive cerebrovascular disease and persistent ischemic symptoms, despite maximal medical therapy, seem to obtain demonstrable and durable benefit from cerebral revascularization.

CONCLUSION

In this small series, cases with concordant clinical features, MRI findings and angiographic finding underwent bypass surgical procedure and we found such concordances ended up with positive result (though short term) with cerebral revascularization.

ABBREVIATIONS

ADC-afferent diffusion co-efficient
AICA-anterior inferior cerebellar artery
CBF-cerebral blood flow
CCA-common carotid artery
CT- computed tomography
CTA- CT angiography
CS -cavernous sinus
DW-diffusion weighted

DSA-digital subtraction angiography
ECA- external carotid artery
EC-IC extracranial-intracranial
GA-general anesthesia ICA-internal carotid artery
IC-intracranial hemorrhage
LE-lower extremity
MCA-middle cerebral artery
MRI-magnetic resonance imaging
MRA-magnetic resonance angiography
MRSA-mithicillin resistance staphylococcus aureus
OEF-oxygen extraction fraction
PCA-posterior cerebral artery
Pcom-posterior communicating
POD-post operative day
PICA-posterior inferior cerebellar artery
PW-perfusion weighted
PET-positron emission tomography
RA-radial artery
RAG-radial artery graft
SCA-superior cerebellar artery
SPECT-single photon emission computed tomography
STA-Superficial temporal artery
SV-shapenous vein
TCD-transcranial doppler
TIA-transient ischemic attack
UE-upper extremity
VA-vertebral artery

DECLARATION

Ethics approval and consent to participate – Not applicable (NA)
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Acknowledgements - None

DISCLOSURE

There is no conflict of interest and nothing to disclose.

REFERENCES

1. Baldawa SS, Pendharkar H, Menon GR, and Nair SR. Thrombosed giant cavernous carotid artery aneurysm secondary to cervical internal carotid artery dissection: An unusual entity. *Indian J Radiol Imaging*. 2011 Jul-Sep; 21(3): 225–227. doi: 10.4103/0971-3026.85373.
2. Biswas A, Samadoni A E, Elbassiouny A, Sobh K, and Hegazy A. Extracranial to intracranial by-pass anastomosis: Review of our preliminary experience from a low volume center in Egypt. *Asian J Neurosurg*. 2015 Oct-Dec; 10(4): 303–309. doi: 10.4103/1793-5482.162711.
3. Eric S. Nussbaum, M.D. Donald L. Erickson, M.D. Extracranial-Intracranial Bypass for Ischemic

- Cerebrovascular Disease Refractory to Maximal Medical Therapy Neurosurgery, Volume 46, Issue 1, 1 January 2000, Pages 37–43, <https://doi.org/10.1093/neurosurgery/46.1.37>.
4. Failure of extracranial-intracranial arterial bypass to reduce the risk of ischemic stroke. Results of an international randomized trial. The EC/IC Bypass Study Group. *N Engl J Med.* 1985;313:1191–200. [PubMed: 2865674].
 5. Fiedler J, Pribán V, Skoda O, Schenk I, Schenková V, Poláková S. Cognitive outcome after EC-IC bypass surgery in hemodynamic cerebral ischemia. *Acta Neurochir (Wien)* 2011;153:1303–11.
 6. Grubb RL, Jr, Powers WJ, Clarke WR, Videen TO, Adams HP, Jr, Derdeyn CP Carotid Occlusion Surgery Study Investigators. Surgical results of the Carotid Occlusion Surgery Study. *J Neurosurg.* 2013;118:25–33. [PMCID: PMC4246998] [PubMed: 23101451].
 7. Hwang SW, Lee SK, David CA. Youmans Neurological Surgery. 6th ed. Ch. 355. Vol. 4. Philadelphia, PA: Saunders, An Imprint of Elsevier Inc; 2011. Intracranial occlusive disease.
 8. Kalani MY, Zabramski JM, Hu YC, Spetzler RF. Extracranial-intracranial bypass and vessel occlusion for the treatment of unclippable giant middle cerebral artery aneurysms. *Neurosurgery.* 2013;72:428–35.
 9. Lee B, Kim C, Carrasco J. Intracranial Infectious Aneurysm in Orbital Cellulitis. *Orbit.* 2015; 34(4):175–8. doi: 10.3109/01676830.2015.1014515. Epub 2015 May 8.
 10. Neil JA, r. Orlandi RR, and Couldwell WT (2016) Malignant fungal infection of the cavernous sinus: case report. *J Neurosurg* 124:861–865. DOI:10.3171/2015.2.JNS142668].
 11. Nussbaum ES, Janjua TM, Defillo A, Lowary JL, Nussbaum LA. Emergency extracranial-intracranial bypass surgery for acute ischemic stroke. *J Neurosurg.* 2010 Mar;112(3):666–73. doi: 10.3171/2009.5.JNS081556].
 12. Obusez EC, Hui F, Hajj-Ali RA, Cerejo R, Calabrese LH, Hammad T, et al.. High-resolution MRI vessel wall imaging: spatial and temporal patterns of reversible cerebral vasoconstriction syndrome and central nervous system vasculitis. *AJNR Am J Neuroradiol.* 2014; 35:1527–1532. doi: 10.3174/ajnr.A3909.
 13. Pool DP, Potts DG. Aneurysms and Arteriovenous Anomalies of the Brain: Diagnosis and Treatment. New York: Harper & Row; 1965.
 14. Sundt TM, Jr, Whisnant JP, Piepgras DG, Campbell JK, Holman CB. Intracranial bypass grafts for vertebral-basilar ischemia. *Mayo Clin Proc.* 1978;53:12–8. [PubMed: 625139].
 15. Swartz RH, Bhuta SS, Farb RI, Agid R, Willinsky RA, Terbrugge KG, et al.. Intracranial arterial wall imaging using high-resolution 3-tesla contrast-enhanced MRI. *Neurology.* 2009; 72:627–634. doi: 10.1212/01.wnl.0000342470.69739.b3.CrossrefMedline Google Scholar.
 16. Thanapal S, Duvuru S, Sae-Ngow T, Kato Y, and Takizawa K. Direct Cerebral Revascularization: Extracranial-intracranial Bypass. *Asian J Neurosurg.* 2018 Jan-Mar; 13(1): 9–17. doi: 10.4103/ajns.AJNS_76_17.
 17. Woringer E, Kunlin J. Anastomosis between the common carotid and the intracranial carotid or the sylvian artery by a graft, using the suspended suture technic. *Neurochirurgie.* 1963;9:181–8. [PubMed: 14076765].
 18. Yaşargil MG. Microsurgery Applied to Neurosurgery. Stuttgart: Georg Thieme Verlag, Academic Press; 1969. Diagnosis and indications for operations in cerebrovascular occlusive disease; pp. 95–118.



A technical note. The role of lilliequist membrane fenestration during the pterional approach for anterior circulation aneurysm clipping

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ABSTRACT

The Lilliequist membrane is a critical membrane located at the base of the brain separating the supratentorial from the infratentorial cisterns. The advantages of Lilliequist membrane fenestration as a critical part of the pterional trans-Sylvian approach for ruptured anterior circulation aneurysm clipping is not well established. We demonstrated that the fundamental role of Lilliequist membrane fenestration is brain relaxation through the egress of CSF that is not usually gained from other modalities (e.g., placement of a lumbar drain, fenestration of lamina terminalis) in this specific setting.

BACKGROUND

Dissection through the arachnoid planes and fenestration of the accessible cisterns are the cornerstones of modern microsurgery while dealing with intracranial lesions.

The Lilliequist membrane is a horizontal arachnoid membrane attached to the dorsum sellae anteriorly, the mammillary body superiorly and the basilar artery posteriorly. It is bounded by the oculomotor nerve on each side. Embryologically, the Lilliequist membrane is considered a remnant of the primary tentorium (1).

The first recognition of the Lilliequist membrane was in 1875 by Key and Retzius (2). Eight decades later, it was rediscovered by an eminent Swedish radiologist named Bengt Lilliequist (1923-1972) as part of his doctoral thesis titled "The subarachnoid cisterns. An anatomical and roentgenologic study" (3). The Lilliequist membrane became a universally accepted medical eponym and officially settled through

Keywords

lilliequist membrane
fenestration,
pterional approach,
anterior circulation aneurysm



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Yasargil's paper "Anatomical observations of the subarachnoid cisterns of the brain during surgery" in 1976 (34). Since that time, the Lilliequist membrane underwent detailed descriptions in literature based on cadaveric and radiological studies (4-8).

In this article, we describe the technique and benefits of Lilliequist membrane fenestration to achieve brain relaxation during the pterional trans-Sylvian approach for ruptured anterior circulation aneurysm clipping.

PERTINENT ANATOMY OF THE LILLIEQUIST MEMBRANE

Intracranially, the arachnoid membranes can be either outer or inner membranes. The outer membranes include both the convexity and the basal arachnoid membranes, while the inner membranes include the trabecular arachnoid membranes which encase the neurovascular bundle within the cisterns (8). The Lilliequist membrane is an inner rather than outer arachnoid membrane (9).

There is some controversy regarding the number of layers of the Lilliequist membrane. The early literature published by Lilliequist and Yasargil described a single-layer membrane (5,10-12). Later studies by Matsuno and Rhoton (13,14) assumed the two-layer model (the diencephalic membrane and the mesencephalic membrane). Recently, the existence of a third layer was described, but it was questionable as to whether this third layer represents a distinct layer or sparse arachnoid trabeculae (8).

From the dorsum sellae, the diencephalic membrane extends superoposteriorly to the mammillary bodies between the chiasmatic and the interpeduncular cisterns and can sometimes be identified radiologically in the sagittal MRI view. The mesencephalic membrane is a thin sheet that extends inferoposteriorly to the pontomesencephalic junction, between the interpeduncular and the prepontine cisterns. Laterally, the Lilliequist membrane is attached to the oculomotor nerve and/or the uncus and tentorium. The Lilliequist membrane is closely related and variably penetrated by the posterior communicating and basilar arteries and their perforating branches (4-8,10-14).

THE GENERAL INDICATIONS FOR LILLIEQUIST MEMBRANE FENESTRATION

Lilliequist Membrane fenestration plays an important

role in multiple procedures related to microscopic or endoscopic cranial surgery:

1. In surgery for tumors in the sellar and parasellar regions such as craniopharyngiomas, meningiomas, basilar tip aneurysms, and arachnoid cysts, Lilliequist membrane fenestration provides an effective surgical corridor and promotes brain relaxation through CSF egress. (1,15,17).
2. Lilliequist membrane fenestration is one of the most critical steps for endoscopic third ventriculostomy success.
3. Some reports consider the microscopic fenestration of the basal subarachnoid cisterns as an effective method to decrease the intracranial pressure in acute intra-operative brain swelling and also in severe traumatic brain injury. These reports advocate the inclusion of Lilliequist membrane fenestration as a mandatory procedural step to achieve a good surgical outcome (16,18)

THE TECHNIQUE FOR LILLIEQUIST MEMBRANE FENESTRATION DURING THE PTERIONAL APPROACH FOR ANTERIOR CIRCULATION ANEURYSM CLIPPING

The pterional approach is the preferred approach for the clipping of most anterior circulation aneurysms except for the distal anterior cerebral artery aneurysm. Using the trans-Sylvian route, the Lilliequist membrane can be approached laterally. Opening the Lilliequist membrane can be achieved through different corridors; namely, the carotid-oculomotor triangle, the optico-carotid triangle and even through the interoptic space. Fenestration through the carotid-oculomotor space is the most convenient, relatively wide and safe technique.

After dural opening, the Sylvian fissure is widely opened from inside to outside on the frontal side of the Sylvian veins and the proximal segment of the middle cerebral artery until the internal carotid artery bifurcation. Frontal and temporal retractors are gently applied; these retractors should "hold" the brain tissue with intermittent retraction to avoid vascular compromise. The frontal lobe is elevated, and the temporal lobe is retracted gently downward and posteriorly. The carotid cistern is opened and the thickened arachnoid fibers over the origins of the anterior and middle cerebral arteries are released. Once the 3rd nerve is identified, the carotid-oculomotor triangle is dissected and subsequently

the deeper membrane of Lilliequist can be identified. As the exposure is deepened, the surgeon will extend the arachnoid opening laterally over the third nerve. Sectioning of the Lilliequist membrane should be started from its anterior part as there is a large space behind the membrane. Then, the interpeduncular cistern is opened through caudal dissection of the Lilliequist membrane between the oculomotor nerve and the internal carotid artery to release the cerebrospinal fluid. At this point, the posterior communicating and the anterior choroidal arteries may be visualized as they arise from the posterior surface of the internal carotid artery (the posterior communicating artery courses perpendicular to the Lilliequist membrane whereas the anterior choroidal artery courses obliquely into the crural cistern) (12).

The egress of the CSF and the identification of the basilar artery within the inter-peduncular cistern is the critical step that ensures the completion of the fenestration process. Additional steps were suggested by some reports including irrigation and mechanical cleaning of blood clots beyond the Lilliequist membrane to minimize the possibility of postoperative vasospasm and hydrocephaly (15,16).

Sharp dissection is always advised to minimize the risk of uncontrolled bleeding from tearing of the vessel wall. Also, the blunt and vigorous dissection may cause disastrous consequences by injuring the hypothalamus either directly or indirectly, by affecting its blood supply. Structures at risk during this step include the posterior communicating artery or its perforators, the oculomotor nerve or its blood supply and the proximal part of the superior cerebellar artery.

In the case of SAH due to a ruptured aneurysm, several studies have described the effectiveness of Lilliequist membrane fenestration, either alone or in tandem with lamina terminalis fenestration, in reducing the incidence of post-SAH hydrocephalus, (9, 12, 15, 19-21). However, None of these studies focused on the effect of Lilliequist membrane fenestration on brain relaxation specifically, which is crucial in such complex procedures regardless of the presence of hydrocephaly.

Lilliequist membrane fenestration will create a free connection between the supratentorial and infratentorial compartments which represents the crucial advantage of Lilliequist membrane fenestration over the opening of the cisterns or

lamina terminalis; this fact was supported by Yaşargil and Winkler-Lawton et al reports through the "Fifth ventricle theory" (12, 19). The fifth ventricle theory can be described as the following; in cases of aneurysmal SAH, the Lilliequist membrane will entrap blood clots and it will be inflamed and thickened, thus the interpeduncular and prepontine cisterns will be isolated and dilated with CSF creating a "fifth ventricle". At this point, Lilliequist membrane fenestration will result in dramatic CSF egress with striking brain relaxation which renders further surgical steps safer and more effective. Castro-Flores et al have suggested that in several patients, even after the opening of the supratentorial cisterns, the CSF pathway will remain constricted until the Lilliequist membrane is opened (15).

AN ILLUSTRATIVE CASE

A 59-year male presented to the emergency department with an altered level of consciousness (Glasgow Coma Scale:9) of sudden onset. Initial resuscitation was done and the patient was transferred to the intensive care unit. A brain CT scan showed left gyrus rectus hemorrhage. Contrasted brain CT scan and CT angiography revealed complex ruptured anterior communicating artery aneurysm with a wide neck, and hypoplastic left Proximal anterior cerebral artery. The anterior communicating artery was vertically oriented, and the anterior cerebral arteries were connected distally to the anterior communicating artery, with two Murphy's teats (Figure 1A).

Surgical clipping through the standard right pterional trans-Sylvian approach was chosen. After dural opening, around 20cc of CSF was drained through a lumbar drain. A microsurgical arachnoid dissection was performed using an operative microscope; starting with the opening the proximal Sylvian fissure and the carotid cisterns; there was minimal egress of CSF.

The Lilliequist membrane was identified and opened lateral to the supraclinoid internal carotid artery through the carotid-oculomotor space. At this point, there was a striking CSF egress that filled the surgical field despite repetitive suctioning. This CSF egress had a profound effect on brain relaxation, provided us a wide operative corridor, and rendered the subsequent steps of aneurysm dissection and clipping more feasible. (Figure 2).

The postoperative course was uneventful and the patient gradually improved and was discharged with no neurological deficits (Figure 1B and C).

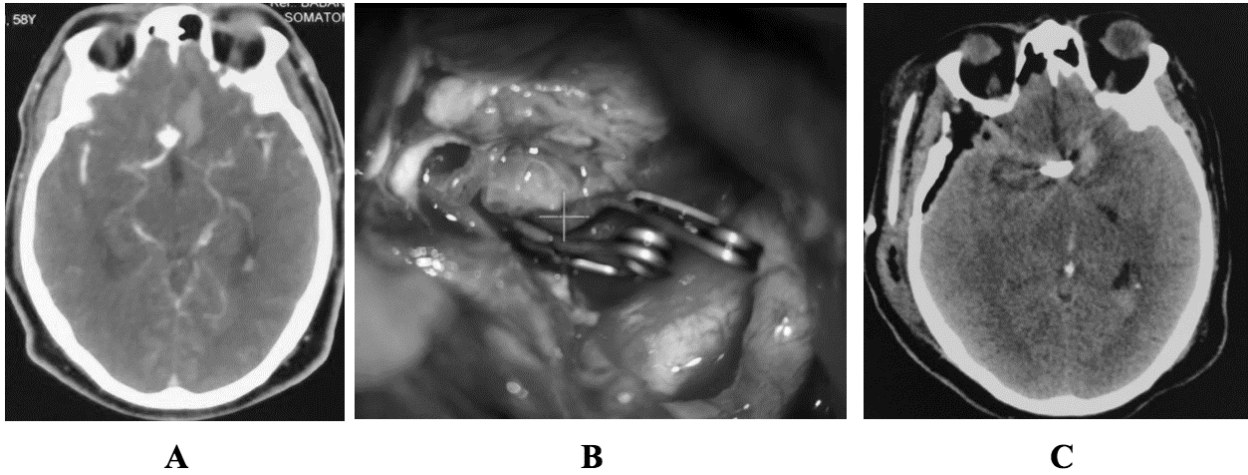


Figure 1. Illustrative case of a ruptured anterior communicating artery aneurysm. **A:** contrasted CT scan of the brain showing the left gyrus rectus due to a ruptured anterior communicating artery aneurysm. **B:** An intraoperative microscopic view showing a two-clip reconstruction for the aneurysmal neck through the right pterional trans-Sylvian approach. **C:** An early postoperative brain CT scan showing the clipping result.

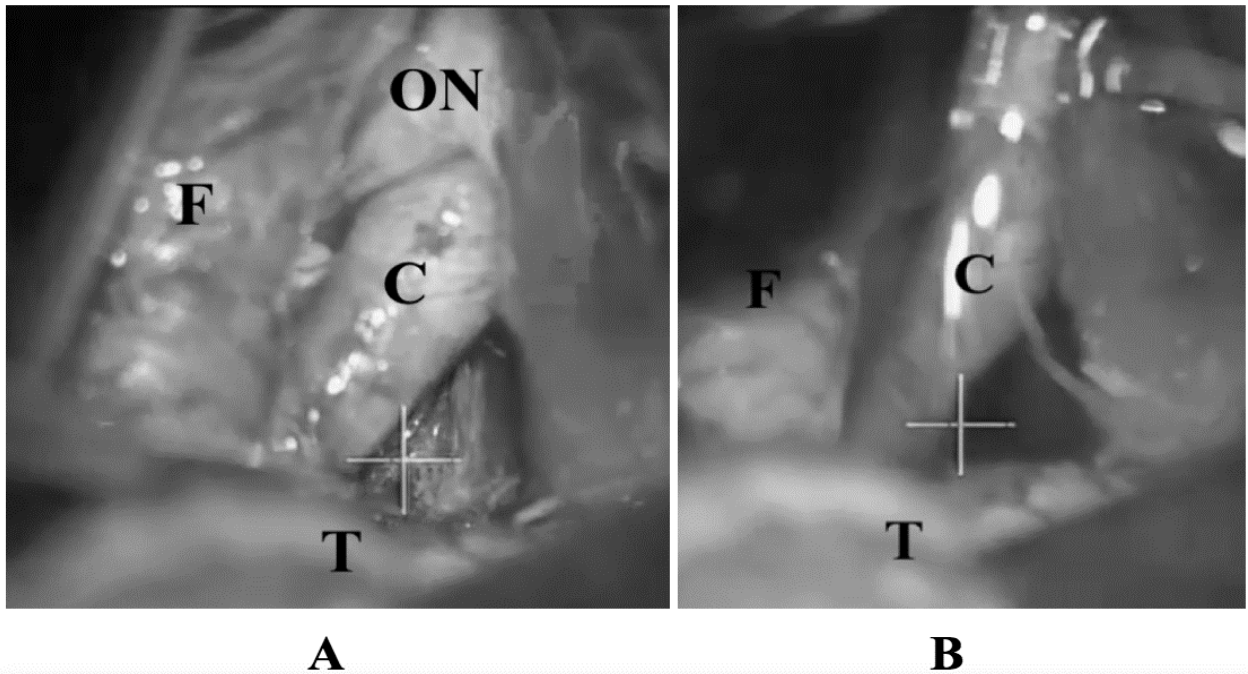


Figure 2. Stages of Lilliequist membrane fenestration through the right pterional trans-Sylvian approach (Intraoperative microscopic view). The Lilliequist membrane identified as a whitish layer lateral to the supraclinoid internal carotid artery through the carotid-oculomotor space. **(A)** Opening the Lilliequist membrane resulted in striking CSF egress that filled the surgical field several times **(B)**. (+): pointer on the Lilliequist membrane, F: Frontal lobe, T: Temporal lobe, C: internal carotid artery, ON: optic nerve.

CONCLUSION

Fenestration of the Lilliequist membrane has a fundamental role during the pterional trans-Sylvian

approach for ruptured anterior circulation aneurysm clipping as it allows brain relaxation through egress of CSF that is not usually achieved through other

modalities (e.g, placement of a lumbar drain, fenestration of lamina terminalis). Further studies are needed to verify this observation towards making the fenestration of the Lilliequist membrane a recommended surgical step rather than an optional one.

ABBREVIATIONS

MRI: magnetic resonance imaging;

CSF: cerebrospinal fluid, SAH: subarachnoid hemorrhage.

DECLARATION

Consent for publication: Our local institutional consent was obtained from the patient.

Competing interests: The authors declare that they have no competing interests.

Funding: Nothing to declare.

REFERENCES

1. Froelich SC, Aziz KM, Cohen PD, Loveren HR, Keller JT. Microsurgical and endoscopic anatomy of Lilliequist's membrane: a complex and variable structure of the basal cisterns. *Operative Neurosurgery*. 2008 Jul 1;63(suppl_1):ONS1-9.
2. Retzius MG, Key A. Studien in der Anatomie des Nervensystems und des Bindegewebes, von Axel Key und Gustaf Retzius,... Samson und Wallin; 1875.
3. Connor Jr DE, Nanda A. Bengt Lilliequist: life and accomplishments of a true renaissance man. *Journal of neurosurgery*. 2017 Feb;126(2):645-9.
4. Matsuno H, Rhoton Jr AL, Peace D. Microsurgical anatomy of the posterior fossa cisterns. *Neurosurgery*. 1988 Jul 1;23(1):58-80.
5. Brasil AV, Schneider FL. Anatomy of Lilliequist membrane. *Neurosurgery* 1993; 32:956 -960.
6. Zhang M, An PC. Lilliequist's membrane is a fold of the arachnoid mater: study using sheet plastination and scanning electron microscopy. *Neurosurgery*. 2000 Oct 1;47(4):902-9.
7. Vinas FC, Panigrahi M. Microsurgical anatomy of the Lilliequist's membrane and surrounding neurovascular territories. *min-Minimally Invasive Neurosurgery*. 2001 Jun;44(02):104-9.
8. Lü J, Zhu XI. Microsurgical anatomy of Lilliequist's membrane. *min-Minimally Invasive Neurosurgery*. 2003 Jun;46(03):149-54.
9. Zhang XA, Qi ST, Huang GL, Long H, Fan J, Peng JX. Anatomical and histological study of Lilliequist's membrane: with emphasis on its nature and lateral attachments. *Child's Nervous System*. 2012 Jan 1;28(1):65-72.
10. Lilliequist B. The anatomy of the subarachnoid cisterns. *Acta radiologica*. 1956 Jul(1-2):61-71.
11. Epstein BS. The role of a transverse arachnoid membrane within the interpeduncular cistern in the passage of pantopaque into the cranial cavity. *Radiology*. 1965 Nov;85(5):914-20.
12. Yasargil MG. *Microneurosurgery*. New York: Thieme Verlag, 1984; 27: 15±53
13. Matsuno H, Rhoton Jr AL, Peace D. Microsurgical anatomy of the posterior fossa cisterns. *Neurosurgery*. 1988 Jul 1;23(1):58-80.
14. Rhoton AL. The posterior fossa cisterns. *Neurosurgery* 2000; 47 (Suppl): 297 ± 297
15. Juan Antonio Castro-Flores., et al. "Efficacy of the Lamina Terminalis Fenestration Associated With the Lilliequist Membrane Fenestration in Reducing Shunt-Dependent Hydrocephalus Following Aneurysm Surgery in the Acute Phase of Aneurysmal Subarachnoid Hemorrhage". *EC Neurology* 2.4 (2015): 162-166.
16. Cherian I, Grasso G, Bernardo A, Munakomi S. Anatomy and physiology of cisternostomy. *Chinese Journal of Traumatology*. 2016 Feb 1;19(1):7-10.
17. Miyajima M, Arai H, Okuda O, Hishii M, Nakanishi H, Sato K. Possible origin of suprasellar arachnoid cysts: neuroimaging and neurosurgical observations in nine cases. *Journal of neurosurgery*. 2000 Jul;93(1):62-7.
18. Abdulqader MN, Al-Tameemi AH, Salih H, Hoz SS, Al Ramadan AH, Salazar LR. Acute intra-operative brain swelling managed effectively with emergency basal cisternostomy: A case report. *Journal of Acute Disease*. 2018 Jan 1;7(1):43.
19. Winkler EA, Burkhardt JK, Rutledge WC, Rick JW, Partow CP, Yue JK, Birk H, Bach AM, Raygor KP, Lawton MT. Reduction of shunt dependency rates following aneurysmal subarachnoid hemorrhage by tandem fenestration of the lamina terminalis and membrane of Lilliequist during microsurgical aneurysm repair. *Journal of neurosurgery*. 2017 Dec 15:1-7.
20. Ultrastrüktürel HS. Ultrastructural changes in the Lilliequist membrane in the hydrocephalic process and its implications for the endoscopic third ventriculostomy procedure. *Turkish neurosurgery*. 2011;21(3):359-66.
21. Wang SS, Zheng HP, Zhang FH, Wang RM. Microsurgical anatomy of Lilliequist's membrane demonstrating three-dimensional configuration. *Acta neurochirurgica*. 2011 Jan 1;153(1):191-200.



Intraventricular dissemination of the pilocytic astrocytomas in an adult

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ABSTRACT

Pilocytic astrocytoma (PA) is a subset of gliomas characterized by a benign course with an excellent prognosis and rarely metastasizing or spreading along the neuraxis. We report a case of a 56-year female with intraventricular dissemination of pilocytic astrocytoma in an adult and discuss the clinical significance diagnosis and management including the peculiar pattern of dissemination of the pilocytic astrocytoma. The course of the disseminated disease may not be as good as that of patients with localized recurrence or totally resected primary disease and can vary from rapid progression to prolonged stabilization.

INTRODUCTION

Pilocytic astrocytoma (PA) is subset of gliomas characterized by a benign course with an excellent prognosis and rarely metastasizing or spreading along the neuraxis. 1-14 Dissemination of a pilocytic astrocytoma is a rare event that occurs in 2-4% of cases particularly in children. 6, 8, 11 We report a case of intraventricular dissemination of pilocytic astrocytoma in an adult and discuss the clinical significance diagnosis and management including the peculiar pattern of dissemination of the pilocytic astrocytoma.

CASE REPORT

A 56-year old female patient presented with progressively increasing headache, vomiting and blurring of vision without any focal neurological deficits or seizures. Her general and systemic examination was unremarkable. Neurologically she was conscious, alert and oriented to time, place and person. Higher mental functions were normal. She had bilateral papilloedema and other cranial nerves were normal. Motor and sensory examination was normal. Her general and systemic examination was normal. The patient underwent MRI brain and it showed minimally contrast enhancing bilateral thalamic lesions, with intraventricular spread in third ventricle and the aqueduct causing obstructive hydrocephalus (Figure 1, 2 and 3). The patient underwent

Keywords

pilocytic astrocytoma,
leptomeningeal
dissemination,
cerebrospinal fluid,
metastasis,
hydrocephalus



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endoscopic exploration of the third ventricular lesion that could be excised as well as the lesion in the aqueduct also could be excised. Part of the lesion in the fourth ventricle was left alone as it was densely adherent to the surrounding structures. Intra-operatively external ventricular drainage was (EVD) was inserted and was kept in situ for 5 days till the CSF was acceptably clear. She later underwent right ventriculo-peritoneal shunt surgery.

Histopathological examination of the section and additional made serial deeper section showed few fragments bits of cellular neoplasm of astrocytic origin. The tumour composed admixture of compact cellular and oedematous loose-knit tissue showing scattered protoplasmic astrocytes having fairly uniform round to oval or slightly elongated nuclei with delicate - open chromaWr pattern and barely discernible cytoplasm. The fibrillary background showed few microcytic spaces and vascular proliferation comprising few ectatic congested blood vessels Coupie of foe show doubtful Rosenthal fibers (Figure-4). No granulomas were seen. Histopathological features were consistent with - 'pilocytic astrocytoma'. Section shows a tumor of moderate cellularity with cells amidst fibrillary background. Microcystic change, hyalinized vasculature were seen. No mitosis, necrosis, microvascular proliferation seen. Rosenthal fibres and eosinophilic granular bodies were seen. The tumor cells express GFAP & P 53 (focal) and were Immunonegative for synaptophysin 8i EMA. The Mib-1 labeling index was approximately 1%. Low grade astrocytic neoplasm suggestive of pilocytic astrocytoma. In post-operative made she did well as she was conscious, with no focal neurological deficits.



Figure 2. MRI brain T1W sagittal images and T2W coronal image showing the blockage of aqueduct by tumour.

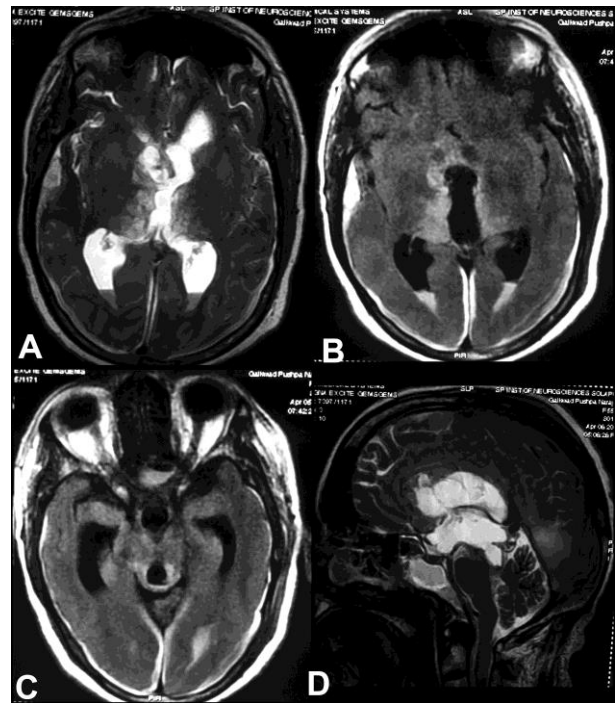


Figure 3. Post-operative MRI T2W and FLAIR axial images showing although the tumor could be removed from upper part of the aqueduct but still the lower part is closed.

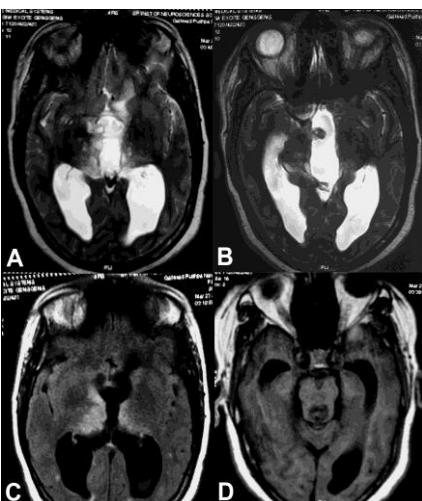


Figure 1. MRI brain T1W, T2W and Flair axial images showing bilateral thalamic tumor with blockage of the aqueduct and associated hydrocephalus.

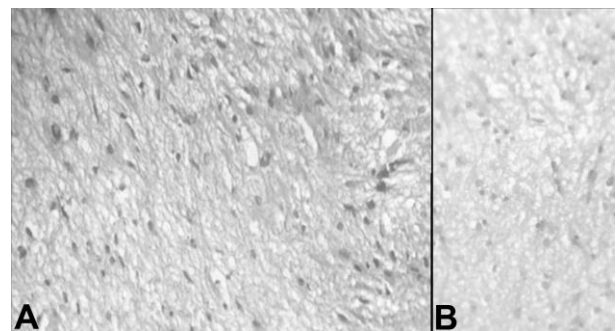


Figure 4. Histopathological examination of the section and additional made serial deeper section showed few fragments bits of cellular neoplasm of astrocytic origin. The tumour composed admixture of compact cellular and oedematous

loose-knit tissue showing scattered protoplasmic astrocytes having fairly uniform round to oval or slightly elongated nuclei with delicate - open chromatin pattern and barely discernible cytoplasm. The fibrillary background showed few microcystic spaces and vascular proliferation comprising few ectatic congested blood vessels. Some show Rosenthal fibers.

DISCUSSION

Dissemination of primary central nervous system neoplasms along the neuraxis is commonly associated with high grade lesions i.e. medulloblastoma, germ cell tumors and malignant gliomas.^{1-5,11,15-18} Dissemination of low-grade gliomas has been documented only in few cases.^{1-14, 19} Usually the site of dissemination is spinal^{1, 6, 8, 11, 16, 19-23} but in rare cases it can be intraventricular metastasis leading to hydrocephalus.²¹ Clinically it can occur after a long postoperative period or may be the first sign of disease or of relapse.^{1, 6, 8, 11, 16, 19-25} The clinical presentation ranges from asymptomatic cases to hydrocephalus, meningismus, worsening of focal deficits, new onset of neurological deficits and onset of seizures.^{8, 11, 19, 24} The tumour spread in these patients is via the CSF and it is accepted that low grade astrocytomas in proximity to ventricles or CSF cisterns are more likely to spread than deeply located tumors.^{21, 26, 27} Several mechanisms have been hypothesized to explain the spread of intracranial tumors by CSF pathways including malignant transformation, cellular anaplasia, surgical manipulation, natural history, multiplicity and presence of cell adhesion molecules (CD44 adhesion molecule as it may play a role in astrocytic invasion and adhesion).^{7, 28} It has been suggested that the tumor mass located in the floor of the third ventricle may breach the ependyma, invading the ventricular cavity and thus resulting in to ependymal or leptomeningeal seeding.^{2, 3, 5, 8, 9}

Pilocytic astrocytoma is a benign tumor that corresponds to histological Grade I^{13, 29-31} and associated with 20-year survival rates of greater than 90% in patients who undergo total excision of the lesion.^{8, 13, 29, 30} Presently there is not much known about the optimum treatment and course of disseminated low-grade astrocytomas.^{1, 2, 14, 16, 32} Probably hydrocephalus, biopsy and partial resection may also be additional favorable factors, although this remains unproven.^{8, 14} It has been suggested that the total resection must be performed as often as possible and no adjuvant therapy should be carried out for low grade gliomas,

^{13, 14} however the treatment of the disseminated tumor, remains controversial.^{14, 20, 25} The course of the disseminated disease may not be as good as that of patients with localized recurrence or totally resected primary disease and can vary from rapid progression to prolonged stabilization.^{1, 2, 14, 16, 25, 32, 11, 20, 21}

REFERENCES

1. Civitello LA, Packer RJ, Rorke LB, Siegel K, Sutton LN, Schut L. Leptomeningeal dissemination of low-grade gliomas in childhood. *Neurology* 1988;38:562-566.
2. Gajjar A, Bhargava R, Jenkins JJ, et al. Low-grade astrocytoma with neuraxis dissemination at diagnosis. *Journal of neurosurgery* 1995;83:67-71.
3. Kellie SJ, Kovnar EH, Kun LE, et al. Neuraxis dissemination in pediatric brain tumors. Response to preirradiation chemotherapy. *Cancer* 1992;69:1061-1066.
4. Packer RJ, Siegel KR, Sutton LN, Litmann P, Bruce DA, Schut L. Leptomeningeal dissemination of primary central nervous system tumors of childhood. *Annals of neurology* 1985;18:217-221.
5. Ushio Y, Arita N, Hayakawa T, et al. Leptomeningeal dissemination of primary brain tumors in children: clinical and experimental studies. *Progress in experimental tumor research* 1987;30:194-205.
6. McCowage G, Tien R, McLendon R, et al. Successful treatment of childhood pilocytic astrocytomas metastatic to the leptomeninges with high-dose Cyclophosphamide. *Medical and Pediatric Oncology* 1996;27:32-39.
7. Morikawa M, Tamaki N, Kokunai T, et al. Cerebellar pilocytic astrocytoma with leptomeningeal dissemination: case report. *Surgical neurology* 1997;48:49-51; discussion 51.
8. Mamelak AN, Prados MD, Obana WG, Cogen PH, Edwards MS. Treatment options and prognosis for multicentric juvenile pilocytic astrocytoma. *Journal of neurosurgery* 1994;81:24-30.
9. Garcia DM, Fulling KH. Juvenile pilocytic astrocytoma of the cerebrum in adults. A distinctive neoplasm with favorable prognosis. *Journal of neurosurgery* 1985;63:382-386.
10. Gjerris F, Klinken L. Long-term prognosis in children with benign cerebellar astrocytoma. *Journal of neurosurgery* 1978;49:179-184.
11. Shapiro K, Shulman K. Spinal cord seeding from cerebellar astrocytomas. *Child's brain* 1976;2:177-186.
12. Haddad SF, Menezes AH, Bell WE, Godersky JC, Afifi AK, Bale JF. Brain tumors occurring before 1 year of age: a retrospective reviews of 22 cases in an 11-year period (1977-1987). *Neurosurgery* 1991;29:8-13.
13. Wallner KE, Gonzales MF, Edwards MS, Wara WM, Sheline GE. Treatment results of juvenile pilocytic astrocytoma. *Journal of neurosurgery* 1988;69:171-176.

14. Figueiredo EG, Matushita H, Machado AGG, Plese JPP, Rosemberg S, Marino R. Leptomeningeal dissemination of pilocytic astrocytoma at diagnosis in childhood: two cases report. *Arquivos de neuro-psiquiatria* 2003;61:842-847.
15. Pezeshkpour GH, Henry JM, Armbrustmacher VW. Spinal metastases. A rare mode of presentation of brain tumors. *Cancer* 1984;54:353-356.
16. Mishima K, Nakamura M, Nakamura H, Nakamura O, Funata N, Shitara N. Leptomeningeal dissemination of cerebellar pilocytic astrocytoma. Case report. *Journal of neurosurgery* 1992;77:788-791.
17. Arseni C, Horvath L, Carp N, Constantinescu A, Ciurea V. Spinal dissemination following operation on cerebral oligodendroglioma. *Acta neurochirurgica* 1977;37:125-137.
18. Bell WO, Packer RJ, Seigel KR, et al. Leptomeningeal spread of intramedullary spinal cord tumors. Report of three cases. *Journal of neurosurgery* 1988;69:295-300.
19. Pollack IF, Hurtt M, Pang D, Albright AL. Dissemination of low grade intracranial astrocytomas in children. *Cancer* 1994;73:2869-2878.
20. Auer RN, Rice GP, Hinton GG, Amacher AL, Gilbert JJ. Cerebellar astrocytoma with benign histology and malignant clinical course. Case report. *Journal of neurosurgery* 1981;54:128-132.
21. Jamjoom AB, Jamjoom ZA, al-Rayess M. Intraventricular and leptomeningeal dissemination of a pilocytic cerebellar astrocytoma in a child with a ventriculoperitoneal shunt: case report. *British journal of neurosurgery* 1998;12:56-58.
22. Tamura M, Zama A, Kurihara H, et al. Management of recurrent pilocytic astrocytoma with leptomeningeal dissemination in childhood. *Child's nervous system : ChNS : official journal of the International Society for Pediatric Neurosurgery* 1998;14:617-622.
23. Versari P, Talamonti G, D'Aliberti G, Fontana R, Colombo N, Casadei G. Leptomeningeal dissemination of juvenile pilocytic astrocytoma: case report. *Surgical neurology* 1994;41:318-321.
24. Patt S, Haberland N, Graupner H, Schreiber D, Kalff R. May 1999--16 year old male with an unexpected MRI finding. *Brain pathology (Zurich, Switzerland)* 1999;9:743-744.
25. Kanda M, Tanaka H, Shinoda S, Masuzawa T. Leptomeningeal dissemination of pilocytic astrocytoma via hematoma in a child. Case report. *Neurosurgical focus* 2002;13:ECP2.
26. Prados M, Mamelak AN. Metastasizing low grade gliomas in children. Redefining an old disease. *Cancer* 1994;73:2671-2673.
27. Obana WG, Cogen PH, Davis RL, Edwards MS. Metastatic juvenile pilocytic astrocytoma. Case report. *Journal of neurosurgery* 1991;75:972-975.
28. Cooper DL, Dougherty GJ. To metastasize or not? Selection of CD44 splice sites. *Nature medicine* 1995;1:635-637.
29. Garcia DM, Latifi HR, Simpson JR, Picker S. Astrocytomas of the cerebellum in children. *Journal of neurosurgery* 1989;71:661-664.
30. Geissinger JD. Astrocytomas of the cerebellum in children. Long-term study. *Archives of neurology* 1971;24:125-135.
31. Kleihues P, Cavenee W. Tumours of the nervous system: pathology and genetics. World Health Organization Classification of Tumours IARC Press, Lyon 2000:198-203.
32. Packer RJ, Lange B, Ater J, et al. Carboplatin and vincristine for recurrent and newly diagnosed low-grade gliomas of childhood. *Journal of clinical oncology : official journal of the American Society of Clinical Oncology* 1993;11:850-856.



Solitary plasmacytoma of occipital bone

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ABSTRACT

We report a case of a 35-year-old male who presented with a pulsatile swelling in the posterior parieto-occipital area. CT and MRI revealed an extra-axial mass. Gross total resection was performed. Histologically it was found to be a plasmacytoma. No recurrence has been noted in the last 48 months of follow up.

INTRODUCTION

Plasma cell neoplasm is characterized by proliferation of single clone of plasma cells. Solitary plasmacytoma of skull bone in a young adult is very rare, it is hence being reported.

CASE REPORT

A 35-year-old male presented with headache, vomiting and decreased vision and swelling in the occipital region. The swelling had appeared about 3 days after a fall sustained 6 months back. It had started as a small swelling but had since then gradually increased in size.

On examination, the patient was conscious, alert and oriented. He had a 6x4 cm sized cystic swelling over the left parieto-occipital region. The swelling had irregular margins and had a smooth surface. Its consistency was solid with areas of cystic change. There was no pulsation or tenderness. CT scan revealed an osteolytic solid mass. MRI showed hyper-intense lesion on T1 with enhancement on contrast. It had mixed intensity on T2. Laboratory investigations showed a hemoglobin of 15.9 g/dL, total WBC count was 8700/microL, differential count P52L35M8E4B1, and platelet count of 2.37 lakhs /dL. There was no evidence of hypercalcemia. No M component was detected in the serum protein electrophoresis.

The patient underwent craniotomy under general anesthesia in Jan 2016. The tumor was purple in color, nodular, vascular and separable from the underlying dura. Margins of the bone were nibbled and cranioplasty with methyl meth-acrylate was done in the same sitting. Histopathology revealed the diagnosis of plasma cell tumor. He was given a course of radiotherapy. The patient was planned for bone

Keywords

plasmacytoma,
skull,
solitary



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marrow aspiration which he refused. Skeletal survey did not reveal any other lesion. There was no evidence of recurrence in the last 45 months.

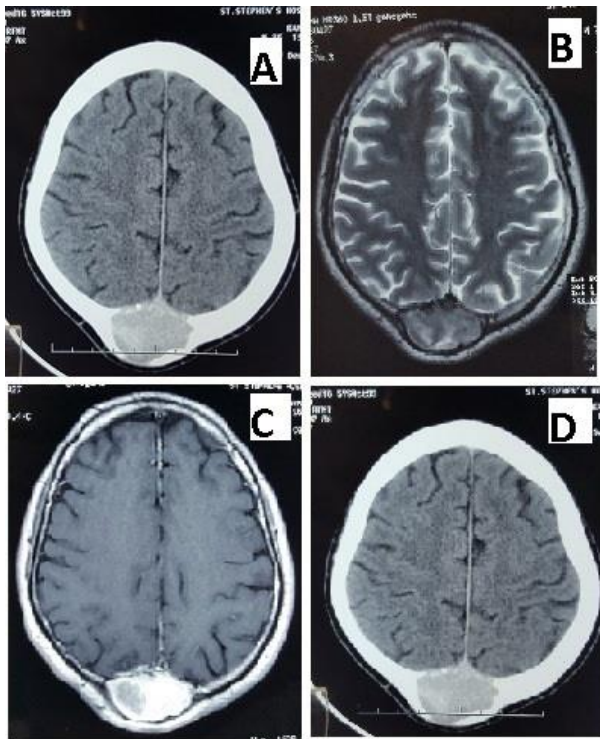


Figure 1.

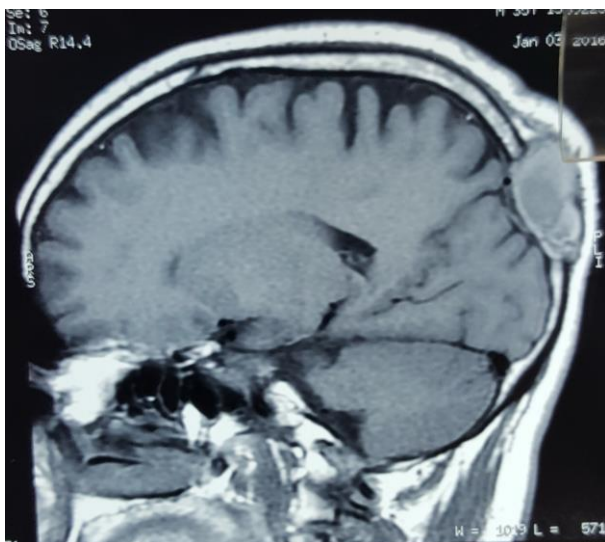


Figure 2.

DISCUSSION

Solitary plasmacytoma of the skull without signs of multiple myeloma is a rare entity and only a few cases have been reported in the literature. ⁽¹⁾ It may

involve cranial vault, skull base or orbit. Symptoms and signs are non-specific and usually lack neurological deficits. The diagnostic characteristics are based on the presence of radiological solitary skull lesion, histological evidence of plasma cells, fewer than 5% plasma cells in the bone marrow aspirate, less than 24mg/dl monoclonal protein in the serum, urine negative for Bence-Jones proteins and no evidence of hypergammaglobulinemia, hypercalcemia or anemia and no recurrence in 2 years of follow up. ⁽²⁾

Usual presentation of the plasmacytoma of skull is as a painless or painful lump without any neurological deficit which depends upon the size and location of the tumor. ⁽³⁾ Cosmetic skull deformity is usual reason for referral to neurosurgeon.

Total surgical resection followed by radiotherapy has been advocated as effective in managing the solitary plasmacytoma. ⁽¹⁾ Arienta et al. report that if total resection has been achieved, then radiotherapy should be reserved for the case of tumor recurrence. ⁽⁴⁾ Furthermore, there are reports of complete cure after biopsy and radiotherapy because plasma cell neoplasms are exquisitely sensitive to radiation. ⁽⁵⁾

The lesion can be highly vascular. ⁽⁶⁾ Preoperative embolization has been resorted to in order to reduce vascularity. ⁽⁷⁾ Craniectomy and cranioplasty is recommended because of high recurrence rate from the residual tumor cells on the involved bone surface. ⁽⁸⁾

Computed tomography reveals destructive well demarcated soft tissue mass with peripheral bony fragments. Usually, the lesion is isointense on T1WI and T2WI and enhances on contrast administration in both CT and MRI. However, in our patient it was slightly hyperdense on CT and slightly hyperintense on T1W1 with mixed attenuation on T2WI. In the absence of early diagnosis of multiple myeloma, the lesion can be misdiagnosed as a meningioma, lymphoma or even myxoma.

DECLARATIONS

Conflict of interest: None

Acknowledgements: None

REFERENCES

1. Singh AD, Chacko AG, Chacko G, Rajshekhar V. Plasma cell tumors of the skull. *Surg Neurol* 2005;64:434-8.

2. Tanaka M, Shibui S, Nomura K, Nakanishi Y. Solitary plasmacytoma of the skull: A case report. *Jpn J Clin Oncol*. 1998;28(10):626-630.
3. Jacquet G, Vuillier J, Viennet A, Godard J, Steimle R. Solitary plasmacytoma simulating pituitary adenoma. *Neurochirurgie* 1991;37:67-71.
4. Arienta C, Caroli M, Ceretti L, Villani R. Solitary plasmacytoma of the calvarium: Two cases treated by operation alone. *Neurosurgery* 1987;21:560-3.
5. Strojan P, Soba E, Lamovec J, Munda A. Extramedullary plasmacytoma: Clinical and histopathologic study. *Int J Radiat Oncol Biol Phys* 2002;53:692-701.
6. Alessandro G, Alfonso C, Marzia D, et al. Plasmacytoma of the skull. *Eur J Haematol* 2011;88:369.
7. Kuo Y, Huang W, Wu J (November 01, 2018) Surgical Treatment for a Giant Solitary Plasmacytoma with Skull Erosion. *Cureus* 10(11): e3535. doi:10.7759/cureus.3535
8. Bindal AK, Bindal RK, van Loveren H, Sawaya R. Management of intracranial plasmacytoma. *J Neurosurg* 1995 83:218-21.



Headache, seizures and loss of consciousness in an elderly male following groin hernia surgery

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ABSTRACT

Headache after lumbar puncture is a common occurrence and have a benign course in the majority. In rare cases, it can be a manifestation of rare but potentially life-threatening intracranial complications. We discuss a case of 65 years male patient who was operated for left inguinal hernia under spinal anaesthesia, had persistent headache partial response to conservative measures developed one episode of seizures and lapsed into altered sensorium. Imaging findings were suggestive of extensive left frontal-temporoparietal acute SDH with mass effect and midline shift. The hematoma was evacuated and the patient recovered well. Prolonged and persistent post-dural puncture headache complicated by atypical neurological deterioration following spinal anaesthesia should prompt the physician to consider the possibility of intracranial complications and to seek immediate radiological investigations.

INTRODUCTION

Headache after lumbar puncture [post-dural puncture headache (PDPH)] is a common occurrence in most cases after lumbar puncture and most of post-LP headaches are not severe and have a benign course (1-4) but in rare cases PDPH can also be a manifestation of a rare but potentially life-threatening complication such as acute subdural hematoma (SDH). (3, 5-16).

CASE REPORT

A 65-years old male patient was operated for left inguinal hernia one week back under spinal anaesthesia. He was complaining of persistent headache and with the diagnosis of PDPH he was given analgesics, oral hydration, and bed-rest without much improvement. On 7th day he had multiple episodes of vomiting, one episode of generalized tonic-clonic seizures and lapsed into altered sensorium. His general and systemic examination was normal. Per abdomen examination was normal. Bowel sounds were normal. Neurologically he was in altered

Keywords
postdural puncture
headache,
subdural hematoma,
headache,
seizure,
lumbar puncture



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sensorium. Glasgow coma scale was 4. Pupils were bilateral 3 mm and reacting to light. Plantars were bilateral extensor. There was no history of hypertension or diabetes. There was no history suggestive of bleeding disorders, using tobacco, or alcohol. Complete blood count including platelets and white blood cells and coagulation tests were within normal range. A plain computed tomography (CT) of brain revealed an extensive left frontal-temporo-parietal acute SDH with mass effect and midline shift (Figure-1). The patient underwent emergency craniotomy and evacuation of hematoma. He was electively ventilated and could be weaned off from ventilator and recovered well.

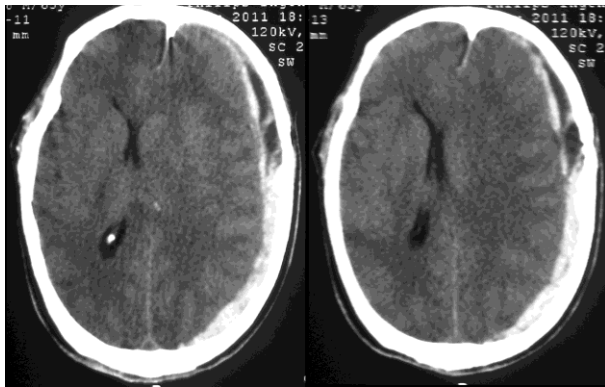


Figure 1. Plain computed tomography scan brain showing large acute left frontal-temporo-parietal subdural hematoma with mass effect and midline shift.

DISCUSSION

Dural puncture with subsequent postdural puncture headache (PDPH) is a recognized complication of spinal anesthesia. (13, 17) The possible mechanism for PDPH is the leakage of cerebrospinal fluid depletes the cushion of CSF supporting the brain and its sensitive meningeal vascular coverings, resulting in gravitational traction on the pain-sensitive intracranial structures causing classical headache. (2, 18) Post-dural puncture headache (PDPH) is classically postural and responds within 48 h to increased fluid intake and bed rest (5, 19, 20) and more frequently seen in the younger patients and in women. (4, 21, 22) Severe and prolonged post-dural puncture headache with or without new neurological signs or deficits should be regarded as a warning sign of an intracranial complications (i.e. subdural haematoma or intracerebral haemorrhage). (3, 5-11, 14, 15, 23-27) In contrast to adults, the dura is adherent to skull and may be less stretchable

duramater due to either atherosclerosis or age-related mechanical changes in the epidural space explaining the lower incidence of PDPH. (28) The mechanism responsible for PDPH i.e. the leakage of CSF and subsequent lowering of the intracranial pressure following dural puncture and caudal movement of the spinal cord and brain stretching and tearing dural veins, results in subdural bleeding in the elderly. (2, 5, 7, 13, 18, 27, 29, 30) The management of acute SDH depends on size of the hematoma as smaller lesions in conscious patients can be managed conservatively under close supervision, (5, 7, 9, 31, 32) however the larger lesions with neurological deficits require neurosurgical intervention and evacuation of hematoma. (7, 8) While considering the patient of PDPH for conservative management, particularly a lumbar autologous extradural blood patch as a possible means to stop the CSF leakage one should be cautious and has low threshold for cranial imaging as the procedure can worsen the patient and neurological deterioration if there is intracranial hematoma. (7)

CONCLUSION

In summary, prolonged and persistent post dural puncture headache complicated by atypical neurological deterioration following spinal anesthesia should prompt the physician to consider the possibility of intracranial complications and to seek immediate radiological investigations to avoid a potential negative outcome.

REFERENCES

1. Evans RW, Armon C, Frohman EM, Goodin DS. Assessment: Prevention of post-lumbar puncture headaches. *Neurology*. 2000;55(7):909-14.
2. Ahmed S, Jayawarna C, Jude E. Post lumbar puncture headache: diagnosis and management. *Postgraduate medical journal*. 2006;82(973):713.
3. Kim HJ, Cho YJ, Cho JY, Lee DH, Hong KS. Acute subdural hematoma following spinal cerebrospinal fluid drainage in a patient with freezing of gait. *Journal of Clinical Neurology (Seoul, Korea)*. 2009;5(2):95.
4. Kuntz K, Stevens J, Offord K, Ho M. Post-lumbar puncture headaches. *Neurology*. 1992;42(10):1884-.
5. Acharya R, Chhabra S, Ratra M, Sehgal A. Cranial subdural haematoma after spinal anaesthesia. *British journal of anaesthesia*. 2001;86(6):893.

6. Macon ME, Armstrong L, Brown E. Subdural hematoma following spinal anesthesia. *Obstetric Anesthesia Digest*. 1990;10(2):108.
7. Verda M, Martnez-Lage J, Alonso B, Snchez-Ortega J, Garcia-Candel A. Non-surgical management of intracranial subdural hematoma complicating spinal anesthesia Manejo no quirrgico de hematoma subdural intracranial tras anestesia espinal complicada. *Neurociruga*. 2007;18(1).
8. Pavlin DJ, McDonald JS, Child B, Rusch V. Acute subdural hematoma-an unusual sequela to lumbar puncture. *Anesthesiology*. 1979;51(4):338.
9. Mantia AM. Clinical report of the occurrence of an intracerebral hemorrhage following post-lumbar puncture headache. *Anesthesiology*. 1981;55(6):684.
10. Duarte WL, Geber DG. Hematoma Subdural aps Puno Inadvertida da Dura-Mter. Relato de Caso. *Revista Brasileira de Anestesiologia*. 2008;58(4):387-90.
11. Chiravuri S, Wasserman R, Chawla A, Haider N. Subdural hematoma following spinal cord stimulator implant. *Pain physician*. 2008;11(1):97-101.
12. Lee ACW, Lau Y, Li CH, Wong YC, Chiang AKS. Intraspinal and intracranial hemorrhage after lumbar puncture. *Pediatric Blood & Cancer*. 2007;48(2):233-7.
13. Cantais E, Behnamou D, Petit D, Palmier B. Acute Subdural Hematoma following Spinal Anesthesia with a Very Small Spinal Needle. *Anesthesiology*. 2000;93(5):1354-5.
14. Komplikasyonu EA, Hematom S. Intracranial Chronic Subdural Haematoma as a Complication of Epidural Anesthesia. *Turkish Neurosurgery*. 2009;19(3):285-7.
15. Suess O, Stendel R, Baur S, Schilling A, Brock M. Intracranial haemorrhage following lumbar myelography: case report and review of the literature. *Neuroradiology*. 2000;42(3):211-4.
16. Newrick P, Read D. Subdural haematoma as a complication of spinal anaesthetic. *British Medical Journal (Clinical research ed)*. 1982;285(6347):1047-8.
17. Berger CW, Crosby ET, Grodecki W. North American survey of the management of dural puncture occurring during labour epidural analgesia. *Canadian Journal of Anesthesia/Journal canadien d'anesthsie*. 1998;45(2):110-4.
18. Hatfalvi BI. Postulated Mechanisms for Postdural Puncture Headache and Review of Laboratory Models: Clinical Experience. *Regional Anesthesia and Pain Medicine*. 1995;20(4):329.
19. Nolte C, Lehmann TN. Postpartum headache resulting from bilateral chronic subdural hematoma after dural puncture. *American Journal of Emergency Medicine, The*. 2004;22(3):241-2.
20. Skoldefors E, Olofsson CI. Intracranial subdural haematoma complicates accidental dural tap during labour. *European Journal of Obstetrics and Gynecology and Reproductive Biology*. 1998;81(1):119-21.
21. Bridenbaugh PO, Greene NM, Brull SJ. Spinal (subarachnoid) neural blockade. Neural blockade in clinical anaesthesia and management of pain 3rd Edition Philadelphia: Lippincott-Raven. 1998:203-42.
22. Leibold RA, Yealy DM, Coppola M, Cantees KK. Post-dural-puncture headache: characteristics, management, and prevention. *Annals of emergency medicine*. 1993;22(12):1863-70.
23. Stocks G, Wooller D, Young J, Fernando R. Postpartum headache after epidural blood patch: investigation and diagnosis. *British journal of anaesthesia*. 2000;84(3):407.
24. Vaughan D, Stirrup C, Robinson P. Cranial subdural haematoma associated with dural puncture in labour. *British journal of anaesthesia*. 2000;84(4):518.
25. Kayacan N, Arc G, Karsl B, Erman M. Acute subdural haematoma after accidental dural puncture during epidural anaesthesia. *International journal of obstetric anaesthesia*. 2004;13(1):47-9.
26. Ozdemir O, Calisaneller T, Yildirim E, Caner H, Altinors N. Acute spontaneous spinal subdural hematoma in a patient with bilateral incarcerated inguinal hernia. *Joint Bone Spine*. 2008 May;75(3):345-7.
27. Hung C, Tan S. Acute-on-chronic subdural haematoma, a rare complication after spinal anaesthesia. *Hong Kong Med J*. 2003;9(5):384-6.
28. Vandam LD, Dripps RD. Long-term follow-up of patients who received 10,098 spinal anesthetics. *Journal of the American Medical Association*. 1956;161(7):586.
29. Thorsn G. Neurological Complications After Spinal Anaesthesia: And Results from 2493 Follow-up Cases: *Kungl. Boktrykeriet PA Norstedt & Soner*; 1947.
30. Lee K. The pathogenesis and clinical significance of traumatic subdural hygroma. *Brain injury*. 1998;12(7):595-603.
31. Scavone BM, Wong CA, Sullivan JT, Yaghmour E, Sherwani SS, McCarthy RJ. Efficacy of a prophylactic epidural blood patch in preventing post dural puncture headache in parturients after inadvertent dural puncture. *Anesthesiology*. 2004;101(6):1422.
32. Blake D, Donnan G, Jensen D. Intracranial subdural haematoma after spinal anaesthesia. *Anaesthesia and intensive care*. 1987;15(3):341-2.



Safety and efficacy of mini doppler in recurrent pituitary tumours. Report of 12 cases

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ABSTRACT

Background: Pituitary surgery is the most common surgery used to remove pituitary tumours. The use of mini doppler in surgical removal of an endonasal pituitary tumour has shown good short-term clinical outcomes and few complications in patients. Cavernous sinus invasion limits the surgical excision and still a challenge of gross total resection.

Objective: The main objective of this study is to evaluate the outcome of surgical removal of an endonasal pituitary tumour using mini doppler.

Method: A total of 12 patients were studied retrospectively from 2012 to 2018 in a single institution (Private hospital) in Dhaka, Bangladesh. The male and female ratio was 7:5.

Results: 92% of cases of the total number of patients had satisfactory removal/ neurological improvement/hormonal improvement. Among 12 cases, 8 cases had transient diabetes insipidus and one patient had CSF leak.

Conclusion: The intraoperative Doppler is a useful tool to localize the carotids, which provides safer resection of endonasal pituitary tumours. Thus, it is very safe and effective for laterosellar resection of recurrent pituitary tumours and for cavernous sinus invasions.

INTRODUCTION

Pituitary tumours are unusual growths that occur in your pituitary gland. Recurring pituitary adenomas can cause visual problems to re-emerge, as well as the loss of pituitary function. It is generally divided into three categories depending upon their biological functioning: benign adenoma, invasive adenoma, and carcinomas. Most adenomas or tumours are benign, about 35% are invasive and just 0.1% to 0.2% are carcinomas.¹ Since the initial description of a transnasal method for pituitary tumour care in 1907, transsphenoidal surgery has undergone continuous development, marked by close collaboration between neurosurgeons and otolaryngologists. Painful excentric muscle training

Keywords
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and ultrasound (US) and doppler-guided sclerosing injections of polidocanol have shown good clinical results, and the traditional surgical approaches have been less necessary.¹⁻³ In 1910, Oskar Hirsch developed a lateral endonasal approach which he initially conducted as a five-step procedure over several weeks before simplifying the procedure with a single step submucosal transseptal approach.⁴ For many years, the conventional surgical treatment of mid-stage Achilles tendinosis consisted of a dorsal approach with central longitudinal tenotomy and tea excision.⁵⁻⁷ A pilot study using an US and dopplerguided scraping technique combined with a short rehabilitation period showed promising results.⁸ The scraping technique is based on the same findings that started the sclerosing polidocanol injection treatment, where gray-scale Us and doppler showed a relationship between vessels and nerves on the ventral side of the Achilles and chronic tendon pain.⁹⁻¹¹ Endoscopic endonasal pituitary surgery for the treatment of recurrent pituitary tumours is becoming increasingly common. The endoscopic endonasal transsphenoidal approach (eTSS) allows for more panoramic viewing and wider access to the base of the skull.¹² Pituitary adenomas are most often classified as functional or non-functional, depending on their pattern of hormonal secretion. We addressed the safety and effectiveness of mini doppler in recurrent pituitary in this study and this doppler is commonly used in neurosurgical practice at present. It is a safe and non-invasive testing tool for cerebrovascular diseases. It is used to examine parameters of the blood flow, to diagnose stenosis, occlusion, and deformity of major head and neck arteries. Doppler ultrasound in carotid and measures both qualitative and quantitative blood flow parameters vertebral arteries, and other forms of care.¹³⁻¹⁴ Nowadays the application of Doppler ultrasonography is becoming increasingly important in endoscopic transsphenoidal surgery. And that is indeed a safe and effective strategy. This study aims to evaluate the outcome of surgical removal of an endonasal pituitary tumour using mini doppler.

METHODS

A retrospective chart review was performed on 12 patients who underwent surgical removal of an endonasal pituitary tumour using mini doppler between 2012 and 2018 in a single institution (Private hospital) Dhaka, Bangladesh. Informed

consent from the patients to archive and process personal data in anonymous form was obtained.

Inclusion criteria were, namely: patients developed recurrence of the tumour with symptoms like headache, vomiting, visual disturbances or hormonal imbalances. Exclusion criteria were, namely: Asymptomatic recurrence of the tumour. Follow up was carried out with routinely MRI of the brain with contrast performed in 1, 6 and 12 months. Then yearly for all patients. Minimum follow up in this study was 2 years. The hormonal study was assessed by an endocrinologist for functional tumours monthly for the first few months and then every 6 months and according to clinical manifestations.

OPERATIVE PROCEDURE

For all 12 patients, the same procedure was carried out as follows. The endonasal transsphenoidal approach was done using the binostril technique. Anatomical landmarks are posterior choana on both sides, from there 1.5 cm above the mucosal flap with scar tissue were separated and in the midline part of the keel of the sphenoid and the sphenoidal bony defects were identified. A high-speed drill to enlarge the defect and to localize the internal carotid arteries mini doppler was used. For recurrent tumours it's difficult to localize particularly in the cavernous sinus and with the help of mini doppler medial opticocarotid recess (MOCR) can be identified and hence the tumour removal became safer. In some cases, where the tumour extended beyond the carotid, the whole of the artery was exposed by drilling and the tumour was removed from the lateral side. Cavernous sinus invasions were removed by localisation of carotid through mini doppler and opening up of cavernous sinus and through two suckers in two hands, along the longitudinal axis of artery tumour was removed to avoid injury to the cranial nerves. Bleeding from cavernous sinus was stopped by using fibrillar surgical. A fascia was taken from the thigh and fibrin glue was used for dural closure.

POSTOPERATIVE COMPLICATIONS AND MANAGEMENT

Diabetes insipidus (DI)

- If the consecutive three hours the urine output \geq 250 ml then injection Vasopressin 5 IU intramuscular was given.

- After 48 hours when the nasal pack was removed Desmopressin nasal spray was used 2 puff in one nostril and according to urine output.
- Half strength normal saline IV was used to reduce the sodium level.
- Avoidance of Hydrocortisone IV to minimize the DI except for those cases where the cortisol level was low.
- In all cases the DI was transient and over one to two weeks the urine output became normal.

For the CSF leak, the patient was managed with lumbar drainage with bed rest which was failed and re-exploration was done through an endoscopic

approach to seal the leak with dural substitute and glue.

Patient with little residual tumour was followed up with contrasted-MRI of the brain every 6 months and found no recurrence of size nor symptoms for the last 3 years.

RESULTS

Characteristics of patients are summarized in Table 1. It was reported that 8.33% of patients had a residual tumour and 91.67% total removal of tumours among all patients. Among 12 cases, 8 cases had transient diabetes insipidus and one had CSF leak (Figure 1).

Table 1. Characteristics of patients.

Pt	Sex, age	Surgery year	Clinical presentation	Type of tumour	Complication	Follow-up
1	Male,45	2012	Headache, visual disturbance	Non-functional adenoma	Transient DI	No residual
2	Male,38	2013	Weight loss, tachycardia, irritability	TSH secreting tumour	Transient DI	No residual Normal level of hormone
3	Male,28	2014	Headache, Vomiting, visual loss	Non-functional tumour apoplexy	Transient DI	No residual
4	Male,32	2015	Abnormal growth of hands and feet	Growth hormone-secreting tumour	Transient DI	No residual Normal level of hormone
5	Male,52	2016	Headache	Non-functional tumour	CSF leak Vlth nerve palsy	No residual
6	Male,36	2017	Headache, visual disturbance	Non-functional tumour	Transient DI	No residual
7	Male,26	2017	Headache, vomiting	Non-functional tumour apoplexy	Transient DI	Small residual

8	Female,25	2012	Amenorrhoea	Prolactinoma	None	No residual Normal level of hormone
9	Female,30	2013	Infertility	Prolactinoma	None	No residual Normal level of hormone
10	Female,42	2017	Headache	Non-functional tumour	Transient DI	No residual
11	Female,24	2018	Headache	Cortisol secreting tumour	None	No residual
12	Female,43	2018	Headache	Non-functional adenoma	Transient DI	No residual

Table 2. Complication rates in male and female patients.

Gender	DI	Postoperative hormonal deficiency	Residual Tumour
Male	6	None	1
Female	2	None	0

Table 3. Tumour resection types.

Tumour type	Cavernous Sinus invasion	Gross total removal	Cranial Nerve Injury
Non-Functional	4	3	1(VIth nerve)
Functional	None	All	None

*Incomplete removal of one non-functional tumour was due to adherence of tumour with cranial nerve inside the cavernous sinus.

COMPLICATIONS

The total number of patients was 12 in this study and among them, 8 patients had Diabetes Insipidus and 1 patient had CSF leak. There were 6 male patients and 2 female patients having DI.

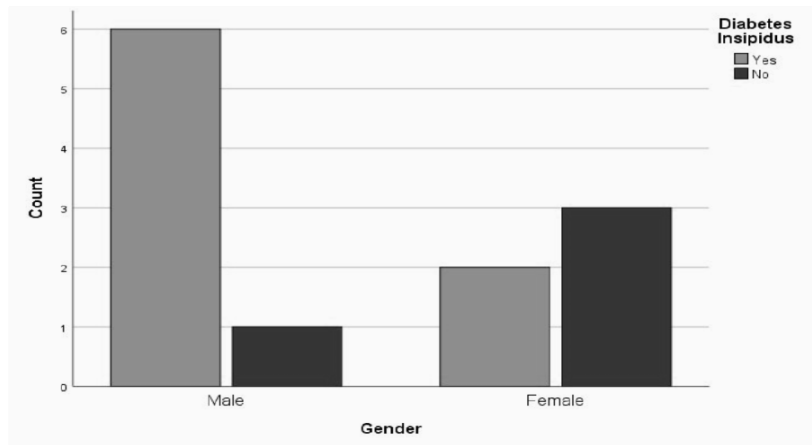


Figure 1. Bar charts showing the number of patients having diabetes insipidus.

In the figure (2A), intraoperative image showing the safe incision line in the midline and the probe, blue arrow touched is the safe incision line. And in figure (2B), contrast MRI brain T1 weighted image in sagittal section is showing recurrent pituitary tumour.

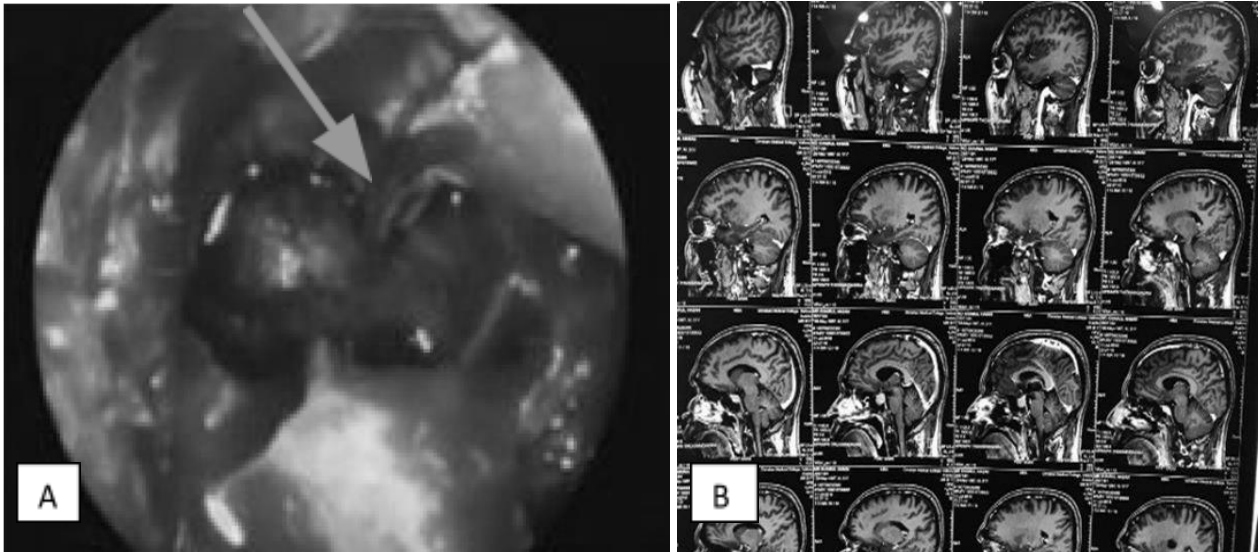


Figure 2 (A, B). Intraoperative image and MRI image of recurrent pituitary tumour.

DISCUSSION

The mini doppler controlled surgical removal of an endonasal pituitary tumour has shown good short-term clinical outcomes and few complications in patients at varying levels of activity. With this treatment method, rapid pain relief and a return to even high-level sporting activity are feasible. The technique provides the greatest degree of freedom and efficiency at specific anatomical goals for sagittal

surgery. This method is one step better for the treatment of recurrent pituitary tumours for improved quality and hospital stay time. More quantitative and qualitative data are required for calculating better results. In reaction to the procedure, athletes back in full pain-free training and competition are found to have a good result. The follow-up period after surgery was also not long, and with time, further failures could occur. Ongoing

research will concentrate on longer-term clinical outcomes and impacts on the thickness and function of the tendons. By using this strategy, since these lesions typically displace the optical apparatus away from the surgeon, tumour removal will begin craniopharyngioma and preexisting growth hormone and gonadal deficiencies experienced a complete failure of anterior pituitary function and DI following surgery immediately after opening the dura mater, resulting in prompt chiasm decompression. The early decompression probably allows the removal of any tumour adherent to the optical apparatus and/or its arachnoidal and vascular connections with less chance of visual degradation. This approach requires minimal nasal mucosal dissection, resulting in fewer sinonasal complications and a quicker and less painful rhinological recovery.¹⁵⁻¹⁶ There are a variety of vascular problems associated with an approach through the nose and sublabial route use. The major drawback to the direct endonasal route is the fairly narrow and slightly off-midline direction of operation.¹⁷ This issue was largely solved by several technical innovations, including the use of low-profile micro-dissection instruments and cutting blades and the use of mini-dopplers, the use of angled endoscopes for more panoramic cephalad and lateral visualization beyond the microscope's tunnel vision, and the use of intraoperative surgical navigation to validate surgical trajectory and main land making.¹⁸ Unfortunately, the complete removal by any method in the three other patients was not a realistic goal given the size and invasiveness of the tumours. Some of the patients in this series encountered new permanent endocrinological abnormalities, with the new DI occurring in 8 cases. Preservation of pituitary function in such patients is challenging particularly those in whom the anterior pituitary function has already failed.¹⁹ In our series of 12 patients, the complication rate was low. A common clinical complication in our series was a postoperative CSF leak that occurs in 1 patient who had neurological deficits, including new cranial nerve palsy. Skin incision or brain retraction is also no longer required. These benefits lead to a decrease in complications, faster patient recovery, minimal postoperative discomfort and a decrease in overall costs. The cavernous sinus extension limits the excision of tumour and adjuvant radiotherapy is the alternate choice of treatment.²⁰

CONCLUSION

The intraoperative Doppler is a useful tool to localize the carotids, which provides safer resection of endonasal pituitary tumours. Thus, it is very safe and effective for laterosellar as well as removal of intracavernous portions of tumour by two hand technique.

CONFLICT OF INTEREST

There is no potential conflict of interest relevant to this research.

FINANCIAL DISCLOSURE

No specific funding was provided for this research.

PATIENT CONSENT

This study obtained patient consent directly from the patient.

ETHICAL APPROVAL

As the authors, we hereby declare that all experiments have been examined and approved by the appropriate ethics committee and have therefore been performed by the ethical standards laid down in the 1964 Declaration of Helsinki.

AUTHOR'S CONTRIBUTIONS

The author's contributions include manuscript preparation and editing. The manuscript has been prepared and approved by all the authors to be submitted and published.

REFERENCES

1. Laws ER. Transsphenoidal tumours surgery for intrasellar pathology. *Clin. Neurosurgery*. 1979; 26: 391-397.
2. Foppiani L, Ruelle A, Cavazzani P, Del Monte P. Hyperthyroidism unmasked several years after the medical and radiosurgical treatment of an invasive macroprolactinoma inducing hypopituitarism: a case report. *Cases J*. 2009; 2:6449. Published 2009 Jul 29. doi: 10.4076/1757-1626-2-6449.
3. Elhadi, Ali M., Douglas A. Hardesty, Hasan A. Zaidi, M. Yashar S. Kalani, Peter Nakaji, William L. White, Mark C. Preul, and Andrew S. Little. "Evaluation of surgical freedom for microscopic and endoscopic transsphenoidal approaches to the sella." *Operative Neurosurgery* 11, no. 1 (2015): 69-79.
4. Lanzino G, Laws ER. Pioneers in the development of transsphenoidal surgery: Theodor Kocher, Oskar Hirsch, and Norman Dott. *J Neurosurg*. 2001 Dec; 95(6):1097-103.
5. Leadbetter WB, Mooar PA, Lane GJ, et al. The surgical treatment of tendinitis. Clinical rationale and biologic basis. *Clin Sports Med* 1992; 11: 679- 712.
6. Nelen G, Martens M, Burssens A. Surgical treatment of chronic Achilles tendinitis. *Am J Sports Med* 1989; 17: 754 - 9.

7. Åström M. On the nature and etiology of chronic achilles tendinopathy [dissertation]. Lund, Sweden: University of Lund, 1997.
8. Alfredson H, Ohberg L, Zeisig E, et al. Treatment of midportion Achilles tendinosis: similar clinical results with US and CD-guided surgery outside the tendon and sclerosing polidocanol injections. *Knee Surg Sports Traumatol Arthrosc* 2007; 15: 1504 – 9.
9. Andersson G, Danielson P, Alfredson H, et al. Nerve-related characteristics of ventral paratendinous tissue in chronic Achilles tendinosis. *Knee Surg Sports Traumatol Arthrosc* 2007; 15: 1272 – 9.
10. Åström M, Gentz CF, Nilsson P, et al. Imaging in chronic achilles tendinopathy: a comparison of ultrasonography, magnetic resonance imaging and surgical findings in 27 histologically verified cases. *Skeletal Radiol* 1996; 25: 615 – 20.
11. Weinberg EP, Adams MJ, Hollenberg GM. Color Doppler sonography of patellar tendinosis. *Am J Roentgenol* 1998; 171: 743 – 4.
12. Rahman M M, Khan R A, et al. Surgical Outcome of Endoscopic Endonasal Surgery for Non- Functional Pituitary Adenoma; *Int J Med Res Prof*. Sep 2018; 4(5):168-72. [Google Scholar]
13. Krylov V, ed. Surgery of cerebral aneurysms. M: Author's issue; 2011; 1: 126165 (In Russ.).
14. Nikitin Yu. Doppler ultrasound in diagnostics of disorders of the major arteries of head and base of brain. *Uchebnoe posobie*. M: Institute of Neurology, RAMS, Spectromed; 1995. (In Russ.).
15. Badie B, Nguyen P, Preston JK: Endoscopic-guided direct endonasal approach for pituitary surgery. *Surg Neurol* 53:168–173, 2000.
16. Zada G, Kelly DF, Cohan P, Wang C, Swerdloff R: Endonasal transsphenoidal approach for pituitary adenomas and other sellar lesions: an assessment of efficacy, safety, and patient impressions. *J Neurosurg* 98:350–358, 2003.
17. Das K, Spencer W, Nwagwu CI, Schaeffer S, Wenk E, Weiss NH, et al. Approaches to the sellar and parasellar region: anatomic comparison of endonasal-transsphenoidal, sublabial-transsphenoidal, and transthemoidal approaches. *Neurol Res* 23:51–54, 2001.
18. Cook SW, Smith Z, Kelly DF: Endonasal transsphenoidal removal of tuberculum sellae meningiomas: technical note. *Neurosurgery* 55:239–246, 2004.
19. König A, Ludecke DK, Herrmann HD: Transnasal surgery in the treatment of craniopharyngiomas. *Acta Neurochir* 83:1–7, 1986.
20. Juyoung Hwang, Ho Jun Seol, Do-Hyun et al: Therapeutic Strategy for Cavernous Sinus-Involving Non-Functioning Pituitary Adenomas Based on the Modified Knosp Grading System. *Brain Tumour Res Treat* 2016; 4(2):63-69.



Surgical management of intramedullary spinal cord metastasis. Report of three cases that revealed unknown malignancies

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ABSTRACT

Intramedullary spinal cord metastasis (IMSCM) is a rare complication of malignancies still studied by case reports; although surgery is related with the best results, the management is still debated considering the risks and the low life expectancy; in fact, many authors prefer conservative management (radiotherapy, chemotherapy), and so less than 200 cases reported in the literature with patients IMSCM were treated surgically; reporting such cases will help to understand the pathology and elaborate a clear management protocol. We report three cases of IMSCM operated at our department; in those patients, the intramedullary lesions revealed the primary tumours.

INTRODUCTION

Face to patients with intramedullary spinal cord metastasis (IMSCM) the management is a challenge, because of the low life expectancy and the bad general condition of those patients. We report three patients with IMSCM managed surgically, in those patients the spinal cord location revealed the primary tumour.

CASE PRESENTATION

Case 1

The first patient is a 57 years old man, with heart disease and smoking history of 30 years. The onset of the troubles dates back to 5 months, with weakness of the left hemibody, and Bravais-Jacksonian convulsions; a month later, there was the appearance of upper back pain with radiation to the limbs as well as functional impotence which confined the patient to bed. The clinical exam at the admission found: cauda equina syndrome with Urinary urgency, Saddle anaesthesia, Abolition of myotatic reflexes in the two lower limbs, there was a

Keywords
intramedullary,
metastases,
spinal cord



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significant muscular atrophy of the two lower limbs, total deficit of the dorsiflexion and plantar flexion of the foot, sensitive deficit (hypoesthesia in the path L5-S1), partial deficit in flexion and extension of the leg on the thigh and the thigh on the trunk, and a Babinski sign. Initially a brain MRI objectified right parietal nodular lesion of 2 cm diameter (figure 1A). The spinal MRI objectified an intramedullary lesion of conus medullaris in heterogeneous hypersignal on T1 WI and T2 WI (figure 1B, C, D). The diagnosis of central nervous metastasis was retained and the investigations for the initial tumour was pushed; the

thoracic CT objectified a right apical pulmonary neoplasia with parietal extension classified T3 N0 Mx (figure 1E). The patient was operated and a gross macroscopic removal of the conus medullaris tumour was performed. In post operative there was a slight recovery on the sensitive and motor functions. The anatomopathological study was in favor of a metastasis of lung adenocarcinoma. The patient was sent then to oncology where a complement of radiotherapy and chemotherapy was performed. There were no complications within 30 days.

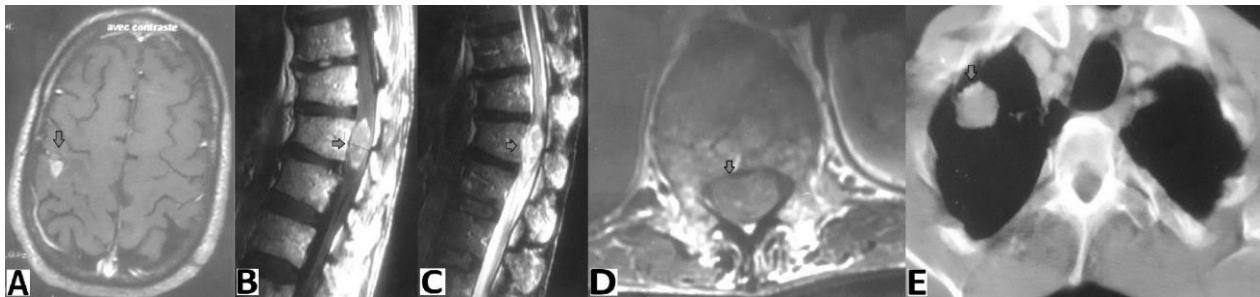


Figure 1. Images of patient 1; (arrows: the tumour in different locations). (A): axial T1 injected brain MRI, objectifying the right parietal metastasis; (B): sagittal T1 WI injected spinal MRI showing the enhancement of conus medullaris nodule; (C): sagittal T2 WI spinal MRI; (D): axial T1 WI spinal MRI; (E): axial thoracic CT, objectifying right apical pulmonary neoplasia with parietal extension.

Case 2

The second patient is a 41 years old man, with past medical history of what it was thought to be lung abscess treated with antibiotics without following. The onset of the disorder dates back to two months before admission, marked by the installation of muscle weakness in the lower limbs of rapid progression. The neurological exam at the admission found a total flasco-spasmodic paraplegia, abolishing of osteotendinous reflexes of the two lower limbs and bilateral Babinski sign. A spinal MRI objectified an expansive intraspinal intramedullary process of the conus medullaris with heterogeneous hypersignal on T1 WI and T2 WI (figure 2A, B). Already in the preoperative assessment, a chest X-rays performed for the anesthesia preoperative assessment objectified a suspect inhomogeneous opacity on the lower left lobe (figure 2C). As part of the investigations thoraco-Abdomino-pelvic CT objectified a lower left lobe pulmonary excavated mass, and multiple hepatic hypodense lesions (figure 2D, E). The patient underwent a CT-guided transparietal puncture of the lung lesion brought 40 cc of pus, the bacteriological study was negative but the

anatomopathological examination was in favor of lung squamous cell carcinoma. The patient was operated; he underwent a partial removal of the tumour of the conus medullaris through Th 11, Th 12 and L1 laminectomy. In post operative the patient was stable. The pathological examination was in favor of a metastasis of a lung squamous cell carcinoma. The patient was sent then to oncology where a complement of radiotherapy and chemotherapy was performed. There were no complications within 30 days.

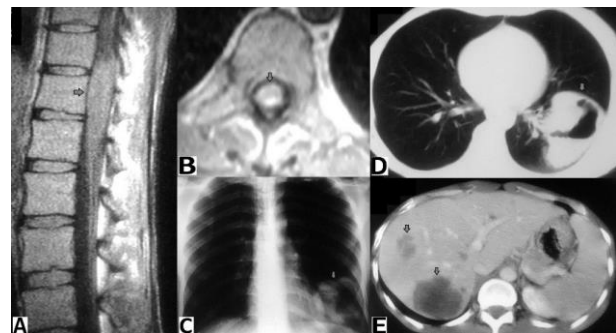


Figure 2. Images of patient 2; (arrows: the tumour in different locations). (A): sagittal T1 WI spinal MRI, showing the conus medullaris tumour; (B): axial T1 WI spinal MRI; (C): chest X-rays

performed in the preoperative assessment showing suspect inhomogeneous opacity on the lower left lobe; (D): axial thoracic CT confirmed the presence of a lower left lobe tumour with necrosis; (E): abdominal CT, showing multiple liver metastasis.

Case 3

The third patient is 26 years old woman without past medical history; the onset of the disorder dates back to one month before admission with the appearance of a weakness in the four limbs, urinary and fecal urgency. The neurological exam at the admission found neck pain, paresthesia in the four limbs, a total flasco-spasmodic tetraplegia, abolishing of osteotendinous reflexes on the four limbs, right Babinski sign, and urinary retention. Spinal MRI objectified a fusiform enlargement of the cervico-dorsal spinal cord with extensive lesions from C4 to Th 2, hyposignal T1WI, hyper signal T2 WI, after injection of gadolinium there was an enhancement in form of small heterogeneous eccentric masses

extended on 8.5 cm from C4 to Th 2, with edematous infiltration of the overlying spinal cord segments up to the bulbomedullary junction as well as underlying segments down to Th 7 (figure 3). The patient was operated and the tumour was removed through a C3 to Th2 laminectomy. The histological exam found a metastasis from a breast carcinoma; in fact a thoraco-abdominal CT was performed and found a suspect left breast mass (figure 3F); the patient was sent then to oncology for further investigations and management (radio and chemotherapy). In post operative immediately the patient was stable, but three weeks later she presented breathing difficulties for what she was admitted; an urgent spinal CT was performed without abnormalities, but the thoracic CT objectified an atelectasis; the patient was put under oxygenotherapy unfortunately 3 days later she presented sudden hemodynamic instabilities followed by a heart arrest and the resuscitation efforts failed.

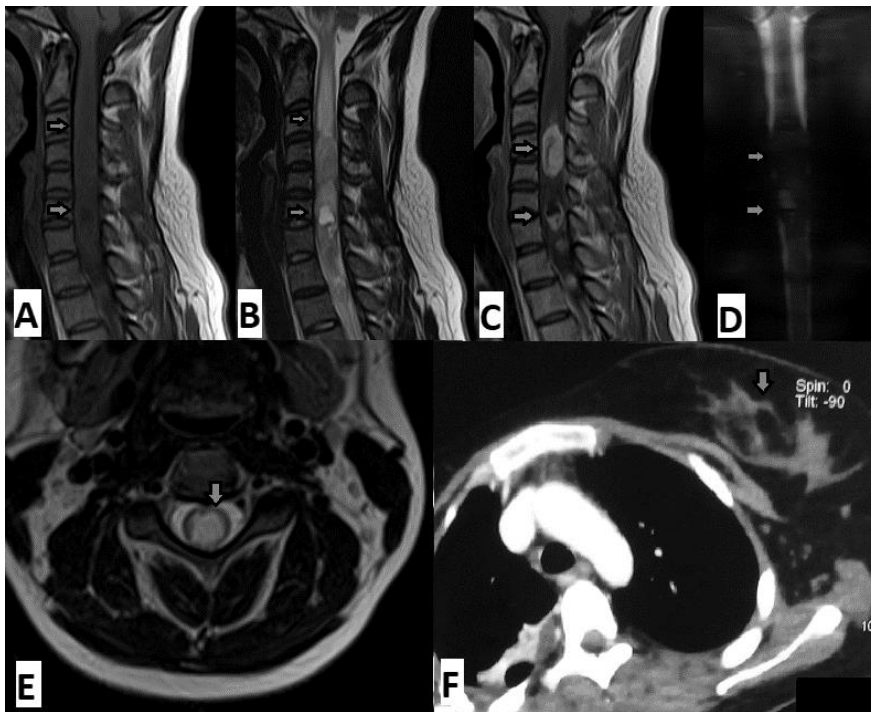


Figure 3. Images of patient 3. (A): sagittal T1 WI spinal MRI, the tumour appears isointense and hypointense with spinal cord swallowing (arrows); (B): sagittal T2 WI spinal MRI, the tumour appears with heterogeneous signal mostly hyperintense with the presence of myelomalacia and a cyst (arrows); (C): sagittal T1 WI injected spinal MRI, heterogeneous enhancement of multiple nodules (arrows); (D): MR-myelography, objectifies the amount of swallowing the tumour was creating; (E): axial T2 WI spinal MRI, showing spinal cord compression by an intramedullary formation (arrows); (F): thoracic CT, showing the left breast suspect nodule (arrow).

Table 1: Summary of our IMSCM cases (*morbidity* and mortalities were evaluated within one month).

Patients	Age Sex	Locations	Primary tumours	Management	Other metastasis	Outcome	Morbidity & Mortalities
Case 1	57 M	Conus	Lung	Surgery	Brain	Improvement	None

Case 2	41	M	Conus	Lung	Surgery	Liver	Stable	None
Case 3	26	F	Cervico-dorsal	Brest	Surgery	None	Stable	Death

DISCUSSION

Metastasis to the spinal cord are rare ^[1,2,3,5], and described mostly by case reports ^[5]. Based mostly on Sung et al review of the English literature ^[3], Jincai et al performed In 2019 a summary of prior studies about IMSCM treatment and outcome, they reported also their own series of 61 patients; in their paper, in sum 519 patients have been reported since 1960 of them only 152 were treated surgically ^[1]. As in our cases, the most common primary locations are the lung and the breast respectively ^[1, 5]; other locations are less common including colon ^[6], melanoma ^[2], and renal ^[2]; in 10 patients (3.3%) of Sung et al series the primary tumour was unknown ^[2]. One of our three patients had brain location, and in the Jincai et al review 206 (40%) of the patients with IMSCM have brain metastasis ^[1], so we find it very reasonable to ask systematically for brain MRI for those patients even in absence of clinical symptoms. There is no clear protocol for the management of IMSCM ^[1, 2, 4, 5]; surgery gave an improvement rate of 77% and a stability of 23 % according to kalayci et al ^[3], for that it seems to be the optimal management tool if the general condition allows such heavy procedure, and also if the life expectancy is acceptable, in fact the prognosis of IMSCM is widely depending on the other locations and is frequently poor. For some authors, surgery could be difficult to propose, and they prefer radiotherapy that might assure an acceptable local control, except for radioresistant tumours such as renal cell carcinoma and melanoma ^[1, 2, 4, 5]. In our cases, the three patients were without past medical history of diagnosed malignancies, and images were not specific; after surgery, the investigations were urged by our pathology laboratory colleagues, even before giving the final histopathological results; so the diagnosis was based on surgical biopsy of the tumours and the invasive management was inevitable ^[5]. The surgeon should not consider radical removal of the tumour, but rather spinal cord decompressing by debulking ^[5], keeping in mind the fact that it is about a low life expectancy patient; the surgeon also is warned to

not in any case expose the patient to complications that could deteriorate the quality of his short life.

CONCLUSION

Intra spinal cord metastasis are rare complications of some malignant tumours, especially lung and breast cancers; surgery is the treatment of chose if the life expectancy and the general condition of the patient are good; otherwise radiotherapy could in some cases assure relief and local control of the tumour.

CONFLICT OF INTEREST

None.

FOUNDING

None.

REFERENCES

1. Jincai Lv, Bailong Liu, Xiaoyue Quan, Cheng Li, Lihua Dong, Min Liu. Intramedullary spinal cord metastasis in malignancies: an institutional analysis and review. *OncoTargets and Therapy* 2019;12 4741–4753.
2. Wen-Shan Sung, Mei-Jo Sung, Jon Ho Chan, Benjamin Manion, Jeeuk Song, Arvind Dubey, Albert Erasmus, Andrew Hunn. Intramedullary Spinal Cord Metastases: A 20-Year Institutional Experience with a Comprehensive Literature Review. *World Neurosurg.* (2013) 79, 3/4:576-584. <http://dx.doi.org/10.1016/j.wneu.2012.04.005>.
3. M. Kalayci, F. Cagavi, S. Gul, S. Yenidunya, and B. Acikgoz. Intramedullary spinal cord metastases: diagnosis and treatment – an illustrated review. *Acta Neurochir (Wien)* (2004) 146: 1347–1354. DOI 10.1007/s00701-004-0386-1.
4. Ondrej Kalita. Current Insights into Surgery for Intramedullary Spinal Cord Metastases: A Literature Review. *International Journal of Surgical Oncology* Volume 2011, Article ID 989506, 5 pages doi:10.1155/2011/989506.
5. Jörg Klekamp, Madjid Samii. *Intramedullary Tumours*. In: Jörg Klekamp, Madjid Samii. *Surgery of Spinal Tumours*, 1st edition; New York: Springer Berlin Heidelberg;2007. P124-127.
6. Tai-Hsin Tsai, I-Cheng Lin, Pei-Chen Lin, Chieh-Hsin Wu, Chih-Lung Lin, and Yu-Feng Su. Intramedullary spinal cord metastasis from colon cancer: analysis of 19 reported cases. *Spinal Cord Series and Cases* (2016) 2, 15026; doi:10.1038/scsandc.2015.26.



Malfunction of a ventriculoperitoneal shunt during pregnancy. Two clinical cases and literature review

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ABSTRACT

Bringing a pregnancy to term is possible for a woman carrying a ventriculoperitoneal bypass valve, however, pregnancy can be a source of malfunction of the bypass system. We report two cases of malfunction of a VPS during the pregnancy's 3rd trimester in two patients aged 25 and 30 years respectively. The valve was examined in both cases and the persistence of the neurological signs required a cesarean section. The diagnostic aspects and management strategies were discussed as regards these two cases and throughout the literature review.

INTRODUCTION

Since the introduction of derivative CSF techniques in the treatment of hydrocephalus, the prognosis for children with this pathology has considerably improved [2,12,15,24,26] Bacterial meningitis is a common cause of morbidity in pediatric wards and constitutes the main aetiology of hydrocephalus occurrence [8.23]. With the development of neurosurgery in the past 20 years, many children are operated on from an early age and most often benefit from the establishment of a VPS. Today, many of these female children are of childbearing age. Malfunction can occur in 50% to 70% of women in labour with VPS, and doesn't necessarily mean anything in the development of future pregnancies. [18]. Treatment may be difficult and requires multidisciplinary collaboration between neurosurgeons, obstetrician-gynecologists-, and anaesthetists.

CLINICAL CASES

Case 1

A 25-year-old pregnant woman, second gesture and primiparous, was admitted to the gynec-obstetrics emergency department for seizures

Keywords
malfunction,
pregnancy,
ventriculoperitoneal shunt



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and impaired alertness. There was a record in the case of a VPS bypass performed at the age of 5 years for obstructive hydrocephalus. A previous pregnancy 5 years ago had been carried to term without incident. The physical examination revealed a Glasgow score of 11 without focal neurological deficit. The diagnosis of a pregnancy toxemia in the 3rd trimester was brought up. However, blood pressure and pulse were normal, blood sugar and proteinuria dosages were also normal. The obstetric examination found a pregnancy of 32 weeks of amenorrhea (WA), a uterine height (UH) at 29 cm in relation to gestational age, and fetal heart sounds (FHS) being normal. The VPS examination noted resistance to depression of the valve reservoir and the Cranio-cerebral CT scan showed dilation of the ventricular cavities with a ventricular drain in place (Figure 1). An evaluation of the VPS has been carried out. Removal of the peritoneal drain did not reveal any obstruction of the distal catheter, and it was reintroduced into the peritoneal cavity. The post-operative course was marked by a noteworthy improvement in the state of consciousness. The patient was kept under intensive care monitoring observation. A week later moderate signs of intracranial hypertension (ICHT) reappeared. A caesarean was performed at 34 WA. Upon extraction of the newborn, the peritoneal drain was found in the vesico-uterine pouch. After verifying its permeability and extensive washing of the operating site, the drain was replaced in the peritoneal cavity. The post-operative course was satisfactory for the mother and for the child throughout a follow-up of 15 months.



Figure 1. Head CT scan showing ventricular dilation.

Case 2

A 30-year-old pregnant woman, first gesture, consulted in neurosurgery for headache and visual disturbances having developed for a week during a pregnancy of 34 weeks. There was a record of VPS for post-meningitis hydrocephalus at the age of 6 years. A valve revision was performed at the age of 16 following a VPS malfunction and the patient had retained neurosensory sequelae with a significant decrease in visual acuity. Upon examination, the blood pressure was normal. The obstetrical examination found an UH at 30cm in relation to the gestational age and the fetal heart sounds were normal. The cranioencephalic scanner had shown dilation of the ventricular cavities. Given the worsening of neurological disorders with impaired alertness, a revision of the VPS was carried out. The peritoneal drain has been repositioned on the other side of the midline. The clinical improvement had been brief and the outcome on day two was marked by the onset of seizures and a new impairment of consciousness. An emergency Cesarean was performed with the extraction of a newborn with a good Apgar score. The development was marked by an improvement in consciousness but with permanent blindness in the mother.

DISCUSSION

Ventriculoperitoneal shunt remains the most widely used method for the treatment of hydrocephalus [18]. Its complications, which are most often mechanical and/or infectious, have been widely reported in the literature [1,3,4,7]. In pregnant women with a VPS, malfunction can occur in 50% to 70% of cases [2,18,20,26]. Furthermore, it is established that VPS has a higher incidence of fault during pregnancy than other types of leads [2,5,10,21,24,25,27]. It usually occurs during the pregnancy's 3rd trimester [2,5,18,26]. A summary of the cases reported in the literature is shown in Table 1. The mechanism for the occurrence of VPS malfunction remains still little understood [11]. The mechanism most often described would be the increase in intra-abdominal pressure due to the presence of the fetus, which would hinder resorption of CSF in the peritoneum [5,11,12,13]. The regression of symptoms observed in most series after childbirth is an argument in favor of this mechanism. The second hypothesis would be the compression of the catheter by a large uterus or a neighboring viscera

such as the stomach, bladder or liver [14,15]. In our 1st case, the peritoneal drain was wound in the vesico-uterine pouch. In our 2nd case, the malfunction is linked to a functional obstruction of the peritoneal catheter, probably related to the increase in intra-abdominal pressure. It has been noted by several authors that there is no correlation between the type of valve and the occurrence of the fault. [17, 20,21].

The classic clinical picture is the occurrence of signs of intracranial hypertension (ICHT) whose intensity is variable. These are usually isolated or associated with persistent headaches to vomiting or visual disturbances [11]. These signs can also be observed in pregnant women without VPS malfunction noted. In the series of Wishoff et al. [27], 59% of patients with VPS had signs of intracranial hypertension. The onset of drowsiness, confusion syndrome, blindness, or altered consciousness is evidence of the severity of intracranial hypertension. The occurrence of inaugural convulsive seizures as in our first case, or intense headache, can mimic the image of pre-eclampsia in the third trimester of pregnancy and mislead the diagnosis of valve malfunction [2,10,15,24]. A malfunction can be suspected through the revision of the VPS which sometimes displays resistance to the depression of the valve reservoir. However, the majority of current VPS systems are no longer equipped with a flexible CSF tank. The scanner and/or MRI, in the event of a malfunction, assesses the dilation of the ventricular cavities associated or not with transependymal resorption and erasure of the cortical grooves with a well-positioned ventricular drain. There is however a risk of irradiation of the fetus by X-rays [6,18]. Isotopic cisternography can diagnose the malfunction, but also risks fetal exposure to radioactive products [5, 6, 18,27]. MRI is the examination of choice because it does not involve any risk of fetal irradiation, and makes it possible to assess the hydrodynamics of the LCS on the flow sequences [2,6,13,18,27]. The problem is the existence of a shunt which is not compatible with MRI, and the deregulation of the drainage pressure by the magnetic field [9]. In our two cases, the scanner made it possible to diagnose the malfunction of the VPS.

The treatment of VPS malfunction during pregnancy requires a multidisciplinary collaboration between obstetrician-gynecologist,

anesthesiologist-resuscitator, and neurosurgeon. It must take into account gestational age, and especially the mother's neurological state. Valve overhaul is sometimes not necessary. Liakos et al [17], in their series, had respectively reported a rate of malfunction and valve revision of 13.7% and 5% among 138 pregnancies followed in 70 patients. When the neurological signs are moderate, a conservative treatment is proposed as a first intervention by several authors. [5,11,13,14,27]. The bed rest sometimes associated with a diuretic treatment such as furosemide or acetazolamide and the daily mechanical pumping of the valve reservoir when possible most often allow a regression of the symptoms of ICHT and allow to carry pregnancy to term. Mechanical pumping can be associated with regular suction of the valve reservoir [5, 11, 12]. However, we believe that this method, even if it can be effective in managing ICHT, carries a high risk of infection. In our 1st case, the change of site of the peritoneal drain allowed us to attain an acceptable period for childbirth. Hawg et al 2010, have advocated for the same approach. Rees et al made the change to the complete system although obstruction of catheter was not proven. Several authors argue for the conversion of VPS to VAS (ventriculoatrial shunt) in the event of malfunction [11,15,21,22,25]. The arguments proposed are the low rate of malfunction and neurological signs observed in the patients with a VAS during pregnancy. Sova et al [26] had proposed the externalisation of the peritoneal catheter until delivery, but this approach in our opinion increases the risk of infection. Endoscopic ventriculocisternostomy can be performed in case of VPS malfunction on obstructive hydrocephalus, and at the same time allows the complete removal of the defective bypass system [9,25]. The obstetric approach also depends on the neurological condition of the patient. In most of the series vaginal delivery is recommended [2,6,9,13,18]. On the other hand, in the event of significant neurological deterioration, a cesarean is necessary. In this case, the revision of the VPS can be carried out simultaneously.

Table 1. Summary of cases from the literature.

Authors	Maternal age (years)	Gestational age (WA)	symptoms	treatment	Type of birth	anaesthesia	outcome	
							mother	new-born
Freo et al [10]	35	36	coma	Shunt revision	Caesarean section	general	good	good
Hwang et al [13]	32	-	Headache and drowsiness	Shunt revision	Caesarean section	general	good	good
Cuisimano et al [5]	21	30	Headache and vomiting	Pumping and suction	Caesarean section	general	good	good
Houston et al [12]	26	33	Headache and nausea	suction	Vaginal delivery	No anaesthesia	good	good
Riffaud et al [25]	33	20	Headache, vomiting and visual disturbance	Ventriculocisternostomy and valve removal	Vaginal delivery	epidural anaesthesia	good	good
	26	15	Headache and visual disturbance	Ventriculocisternostomy and valve removal	Vaginal delivery	epidural anaesthesia	good	unspecified
	27	8	headache	Ventriculocisternostomy and valve removal	Vaginal delivery	epidural anaesthesia	good	good
Hanakita et al [11]	25	32	Headache, visual disturbance and alertness disorder	conversion of VPS to VAS	Vaginal delivery	No anaesthesia	good	good
Fletcher et al. [9]	32	36	Headache, amnesia and urination	Shunt revision	Caesarean section	general	good	good
Rees et al [24]	27	20	Convulsions and alertness disorder	Change of ventriculoperitoneal bypass device	abortion	No anaesthesia	good	bad
Sova et al [26]	27	27	Headache, diplopia and Parinaud	External ventricular drain	Caesarean section	general	good	good

			syndrome					
Murakami et al [21]	20	unspecified	Headache and visual disturbance	conversion of VPS to VAS	Vaginal delivery	epidural anaesthesia	good	good
Kleinman et al. [14]	20	29	Headache and vomiting	pumping	Vaginal delivery	No anaesthesia	good	good
Kurtsoy et al. [15]	34	31	Headache, vomiting and balance disorder	conversion of VPS to VAS	unspecified	unspecified	good	good

CONCLUSION

The treatment of a bypass malfunction during pregnancy is sometimes difficult. It requires close collaboration between neurosurgeons, obstetrician-gynecologists, and anaesthetists. Awareness of these different actors is necessary given the number of female children who have reached the reproductive age. The choice of ventriculocisternostomy in the treatment of hydrocephalus in children when it is indicated may allow avoiding the occurrence of this complication. It is also a treatment of choice when hydrocephalus or VPS malfunction occurs during pregnancy. In all cases, carrying out a pre, peri and post conceptual survey of patients with VPS is necessary.

REFERENCES

- Ahmed A, Sandlas G, Kothari P, Sarda D, Gupta A, Karkera P, Joshi P. Outcome analysis of shunt surgery in hydrocephalus. *J. Indian. Assoc. Pediatr. Surg.* 2009, 14 (3): 98-101.
- Bradley NK, Liakos AM, Mcallister JP, Magram G, Kinsman S, Bradley MK. Maternal shunt dependency: implications for obstetric care, neurosurgical management, and pregnancy outcomes and a review of selected Literature. *Neurosurgery.*1998, 43 (3): 448-60.
- Browd SR, Ragel BT, Gottfried ON, Kestle JR. Failure of cerebrospinal fluid shunts: part I : Obstruction and mechanical failure. *Pediatr. Neurol.* 2006, 34 (2),83-92.
- Browd SR, Ragel BT, Gottfried ON, Kestle JR. Failure of cérebrospinal fluid shunts: part II: over drainage, loculation, and abdominal complications. *Pediatr. Neurol.* 2006,34 (3):171-6.
- Cusimano MD, Meffe FM, Gentili F, Sermer M. Ventriculoperitoneal shunt malfunction during pregnancy. *Neurosurgery.* 1990,27(6):969-71.
- Cusimano MD, Meffe FM, Gentili F, Sermer M. 1991.Management of pregnant woman with cerebrospinal fluid shunt. *Pediatr. Neurosurg.*1991,92 (17):10-3.
- Drake JM, Kestle JR, Milner R, Cinalli G, Boop F, Piatt JJR, Haines S, Schiff SJ, Cochrane DD, Steinbok P, Macneil N.1998 Randomized trial of cerebrospinal fluid shunt valve design in pediatric hydrocephalus. *Neurosurgery.* 1998,43:294-303.
- Eholier Sp, Boni B, Aoussi E, Konan A, Orega M, Koffi A L, Ba Zézé V, Bissagnene E, Kadio A. Complications neurochirurgicales des méningites Purulentes en zone tropicale. *Neurochirurgie.* 1999,45(3):219-24.
- Fletcher H, Crandon lw, Webster D. Maternal Hydrocephalus in Pregnancy and Delivery: A Report of Two Cases. *West. Indian. med. J.*2007,56(6):558-9.
- Freo U, Pitton M, Carron M, Ori C. 2009.Anesthesia for urgent sequential ventriculoperitoneal shunt revision and cesarean delivery. *Int. j. obstet. Anesth.* 2009,18(3): 284-7.
- Hanakita J, Suzuki T, Yamamoto Y, Kinuta Y, Nishihara KJ. Ventriculoperitoneal shunt malfunction during pregnancy. *J Neurosurg.*1985, 63(3):459-60.
- Houston CS, Clein LJ. Ventriculoperitoneal shunt malfunction in a pregnant patient with meningomyelocele. *CMAJ.*1989,141(1):701-2.
- Hwang S.C, Tae-Hee K, Bum-Tae K, Soo-Bin I, Won-Han S. Acute Shunt Malfunction after Cesarean Section Delivery. *J. Korean. Med. Sci.*2010,25(4):647-50.
- Kleinman G, Sutherling W, Martinez M, Tabsh K. Malfunction of ventriculoperitoneal shunts during pregnancy. *Obstet Gynecol.* 1983,61(6):753-4.
- Kurtsoy A, Selçuklu A, Kemal Kr, Pasaoglu A, Kavuncu Al. Management of Maternal Hydrocephalus. *Turkish Neurosurgery.*1994,4:174-5.

16. Landwehr JB, JR, Isada NB, Pryde PG, Johnson MP, Evans MI, Canady AI. Maternal neurosurgical shunts and pregnancy outcome. *Obstet Gynecol.* 83(1):134-7.
17. Liakos AM, Bradley NK, Magram G, Muszynski C. Hydrocephalus and the reproductive health of women: the medical implications of maternal shunt dependency in 70 women and 138 pregnancies. *Neurol Res* .2000,22(1):69-88.
18. Maheut-Lourmière J, Chu Tan S. Hydrocephalus during pregnancy with or without neurosurgical history in childhood. Practical advice for management *Neurochirurgie.*2000,46 (2):117-21.
19. Monafared AH, Kee SK, Apuzzo MLJ, Collea JV. Obstetric management of pregnant woman with extracranial shunts. *Can. Med. Assoc. J.* 1979,120(5):562-3.
20. Mouelhi C, Srasra M, Zhioua F, Ferchiou M, Zine S, Meriah S. Maternal hydrocephaly and pregnancy. *Rev. Fr. Gynecol. Obstet.* 89(2):88-90.
21. Murakami M, Morine M, Iwasa T, Takahashi Y, Miyamoto T, Hon PK, Nakagawa Y. Management of maternal hydrocephalus requires replacement of ventriculoperitoneal shunt with ventriculoatrial shunt: a case report. *Arch. Gynecol.Obstet.* 2010,282 (3):339-42.
22. Okagaki A, Hanzaki H, Moritake K, Mori T.1990. Case report: pregnant woman with a ventriculoperitoneal shunt to treat hydrocephalus. *Asia. Oceania. J.Obstet. Gynaecol.* 1990,16: 111-3.
23. Orega M, Eholier S P, Boni N, Konan A, Koffi A L. Méningites purulentes et complications neuro-chirurgicales chez l'enfant à Abidjan. *Médecine d'Afrique Noire.* 2005,52(4):251-2.
24. Rees GJ, Francis C, Sizer A R. Convulsions in an undiagnosed pregnancy due to blocked ventriculo-peritoneal shunt.*J. Obstet. Gynaecol.* 2008,26(5), 533-4.
25. Riffaud L, Ferre, JC, Carsin-Nicol B, Morandi X. Endoscopic third ventriculostomy for the treatment of obstructive hydrocephalus during pregnancy.*Obstet. Gynecol.* 2006,108:801-4.
26. Sova M, Smrcka, M, Baudysova O, Gogela J Management of a shunt malfunction during pregnancy. *Bratisl Lek Listy.* 2001,102(12):562-3.
27. Wishoff JH, Kratzert KJ, Handwerker SM, Young BK, Epstein F. Pregnancy in patients with cerebrospinal fluid shunts: report of a series and review of the literature. *Neurosurgery.* 1991,29(6):827-31.



Cranium metastasis of HPV positive oropharyngeal squamous cell carcinoma

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ABSTRACT

Head and neck squamous cell carcinomas rarely cause cranial metastases, but HPV related subgroup is known for unusual metastasis sites on the body. Despite the good overall treatment responses in this group, intracranial metastases are always accepted as a sign of rapid deterioration. We present a case of cranium metastasis of HPV related oropharyngeal squamous cell carcinoma, which had been very well responded to the treatment for years but deteriorates in a short period after the cranium metastasis.

INTRODUCTION

Head and neck squamous cell carcinomas (HNSCC) are known as rarely causing intracranial metastases. In the last years, HPV related subgroup of HNSCC has become a distinct subject of studies. They mostly occur at oropharynx. The incidence of HPV-positive HNSCC is increasing. Unlike the HPV-negative one, this subgroup does not show a relationship with tobacco and alcohol, which are prominent risk factors of HNSCC. More favourable results of chemotherapy and radiotherapy have been reported in comparison to the HPV-negative variant. The more successive local control means better morbidity and survival rates. On HPV-positive cases, a better prognosis was reported even on lung metastasis, which is the most frequent site of distant recurrence. Also, a different pattern of metastasis has been mentioned in recent studies (3). On the other hand, the presence of brain metastasis has always been accepted as a bad sign regardless of HPV status.

We present a case of a patient who has one of the most extended survival lengths reported in the literature despite unusual multisite metastases but deteriorated rapidly due to very aggressive cranium metastasis.

CASE REPORT

Sixty-four years old female patient had submitted to hospital with pain on the chest and back in 2013. Examinations revealed metastatic

Keywords

metastasis,
human papilloma virus,
HPV,
cranium,
squamous cell



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lymphadenopathies on mediastinum and metastases on the lung, liver, and vertebra corpus, but the primary tumour had not been found. With consecutive periods of chemotherapy (including carboplatin, capecitabine, and cisplatin-etoposide) and radiotherapy, local control was achieved, and the patient has succeeded five years of survival. In 2018, the patient had multiple enlarged lymph nodes in the cervical area. Following the tru-cut biopsy results, the pathology report pointed out CD117 positivity and low metabolic activity despite the clinical features. As a result, pathological examinations could not suggest a diagnosis. The new PET scan revealed multiple mediastinal and intraabdominal lymphadenopathies and improved uptake in various areas in the gastrointestinal system. With chemotherapy and radiotherapy, the local cure was succeeded. Other sites also responded to the treatment. In November 2018, in an examination for head-ache, extensive cranium metastasis was spotted. The patient was consulted to neurosurgery, and surgery was offered (Fig. 1). During the decision period of patient, right hemiparesis and headache complaints were developed. In concomitant MRIs within three months period, significant enlargement was discovered, and operation was performed (Fig. 2,3).

First, question mark skin incision was performed, and the tumour was revealed, extruding from the calvarium. A large frontoparietal craniotomy flap, which was covering the whole tumour and adherent dura, was elevated. Because of the cohesiveness, the dura was incised and elevated with the tumour. No invasion to parenchyma was observed but cerebrum was edematous. Because of that after bone-tumour-dura complex resection, cranioplasty was planned for a further time.

Pathologic examination revealed that the tumour was HPV positive oropharyngeal squamous cell carcinoma metastasis with strong p16 and neuroendocrine marker positivity. Different from the previously pathological examination, ki-67 index was found to be very high (about %80). Post-op PET scan also revealed an oropharyngeal mass with improved up-take near the base of the tongue.

DISCUSSION

HPV positive HNSCC are characterized by a better therapeutic response and lower recurrence in comparison to HPV negative variant, which means

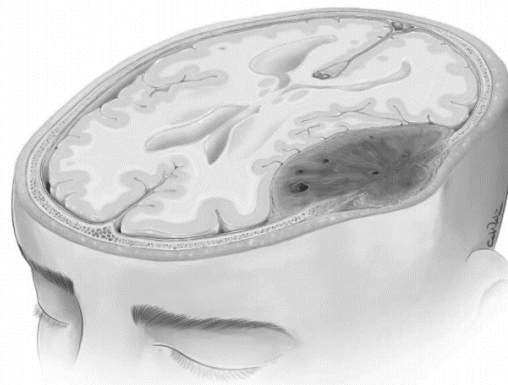


Figure 1.

overall better clinical outcome in terms of survival and morbidity (2). Because of this nature of HPV positive HNSCC, even in the presence of metastases, more direct and curative approaches are preferred instead of palliative treatment options.

Besides the better outcomes, the distant metastasis pattern of HPV positive SCC also differs. The longer survival lengths, thank to better therapeutic results, may contribute to increased numbers of distant metastases on HPV positive SCC. The most frequent metastasis sites for HNSCC are known as lung, liver, and bone (5). In this order, liver and bone metastases were reported as much rarer than the lung. An increased incidence in bone metastases, especially in vertebrae, were reported in patients with p16 positive tumours lately. The recent articles are reporting HPV positive SCC cases with multisite metastasis, including unusual sites such as long bones, brain, dura, pancreas, and skin (3,4). Among the metastasis sites, the brain was accepted to have the worst prognosis and defined as “non-curable”. Brain metastasis frequency on HNSCC was mentioned in a few articles as 0.43 – 5.7%.

The tumour-free episodes in HPV positive SCC are reported to be longer than non-HPV variants in several studies (2). When the recurrence or first metastasis was spotted during follow-ups, patients usually have metastasis on more than one site. In both HPV + and – tumours, metastases have much more aggressive behaviors than their primary tumours. Independent from the response of these metastases to the treatment, different, unexpected new metastases may occur, and the death of patients is usually caused by these aggressive new tumours. The development time of metastases is

approximately two years following the primary tumour treatment (4, 5). In a recent study about brain metastases of HNSCC, mean time of interval between diagnosis of primary and brain metastasis was reported as 26 months. Among the eleven patients, two longest intervals were 137 and 66 months, and the latter patient had 65.7 months of disease-free time, meaning prognosis of the brain metastasis was the decisive factor in patients' life (1).

Besides clinical features, HPV positive SCC's are known to be hard to detect on PET and MRI scans. As in our case, this mass had not been detected previous PET and MRI scans.

CONCLUSION

Extended patient follow-up times are suggested for HPV positive tumours in many papers as a conclusion (2, 4). Under these circumstances, we think that it would not be wrong to define the length of follow-up as "lifelong". Considering positive features of HPV-positivity, with further analysis of metastasis pattern of HPV positive HNSCC and with the developments of neurosurgical oncology, much longer survivability can be achieved in this patient group.

PATIENT CONSENT

The patient has consented to the submission of the case report for submission to the journal.

DECLARATION OF CONFLICT OF INTEREST: None

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REFERENCES

- Ahmed A, Sandlas G, Kothari P, Sarda D, Gupta A, Karkera P, Joshi P. Outcome analysis of shunt surgery in hydrocephalus. *J. Indian. Assoc. Pediatr. Surg.* 2009, 14 (3): 98-101.
- Bradley NK, Liakos AM, Mcallister JP, Magram G, Kinsman S, Bradley MK. Maternal shunt dependency: implications for obstetric care, neurosurgical management, and pregnancy outcomes and a review of selected Literature. *Neurosurgery.*1998, 43 (3): 448-60.
- Browd SR, Ragel BT, Gottfried ON, Kestle JR. Failure of cerebrospinal fluid shunts: part I : Obstruction and mechanical failure. *Pediatr. Neurol.* 2006, 34 (2),83-92.
- Browd SR, Ragel BT, Gottfried ON, Kestle JR. Failure of cérebrospinal fluid shunts: part II: over drainage, loculation, and abdominal complications. *Pediatr. Neurol.* 2006,34 (3):171-6.
- Cusimano MD, Meffe FM, Gentili F, Sermer M. Ventriculoperitoneal shunt malfunction during pregnancy. *Neurosurgery.* 1990,27(6):969-71.
- Cusimano MD, Meffe FM, Gentili F, Sermer M. 1991.Management of pregnant woman with cerebrospinal fluid shunt. *Pediatr. Neurosurg.*1991,92 (17):10-3.
- Drake JM, Kestle JR, Milner R, Cinalli G, Boop F, Piatt JJR, Haines S, Schiff SJ, Cochrane DD, Steinbok P, Macneil N.1998 Randomized trial of cerebrospinal fluid shunt valve design in pediatric hydrocephalus. *Neurosurgery.* 1998,43:294-303.
- Eholier Sp, Boni B, Aoussi E, Konan A, Orega M, Koffi A L, Ba Zézé V, Bissagnene E, Kadio A. Complications neurochirurgicales des méningites Purulentes en zone tropicale. *Neurochirurgie.* 1999,45(3):219-24.
- Fletcher H, Crandon lw, Webster D. Maternal Hydrocephalus in Pregnancy and Delivery: A Report of Two Cases. *West. Indian. med. J.*2007,56(6):558-9.
- Freo U, Pitton M, Carron M, Ori C. 2009.Anesthesia for urgent sequential ventriculoperitoneal shunt revision and cesarean delivery. *Int. j. obstet. Anesth.* 2009,18(3): 284-7.
- Hanakita J, Suzuki T, Yamamoto Y, Kinuta Y, Nishihara KJ. Ventriculoperitoneal shunt malfunction during pregnancy. *J Neurosurg.*1985, 63(3):459-60.
- Houston CS, Clein LJ. Ventriculoperitoneal shunt malfunction in a pregnant patient with meningomyelocele. *CMAJ.*1989,141(1):701-2.
- Hwang S.C, Tae-Hee K, Bum-Tae K, Soo-Bin I, Won-Han S. Acute Shunt Malfunction after Cesarean Section Delivery. *J. Korean. Med. Sci.*2010,25(4):647-50.
- Kleinman G, Sutherling W, Martinez M, Tabsh K. Malfunction of ventriculoperitoneal shunts during pregnancy. *Obstet Gynecol.* 1983,61(6):753-4.
- Kurtsoy A, Selçuklu A, Kemal Kr, Pasaoglu A, Kavuncu Al. Management of Maternal Hydrocephalus. *Turkish Neurosurgery.*1994,4:174-5.
- Landwehr JB, JR, Isada NB, Pryde PG, Johnson MP, Evans MI, Canady Al. Maternal neurosurgical shunts and pregnancy outcome. *Obstet Gynecol.* 83(1):134-7.
- Liakos AM, Bradley NK, Magram G, Muszynski C. Hydrocephalus and the reproductive health of women: the medical implications of maternal shunt dependency in 70 women and 138 pregnancies. *Neurol Res* .2000,22(1):69-88.
- Maheut-Lourmière J, Chu Tan S. Hydrocephalus during pregnancy with or without neurosurgical history in childhood. *Practical advice for management Neurochirurgie.*2000,46 (2):117-21.
- Monafared AH, Kee SK, Apuzzo MLJ, Collea JV. Obstetric management of pregnant woman with extracranial shunts. *Can. Med. Assoc. J.* 1979,120(5):562-3.

20. Mouelhi C, Srasra M, Zhioua F, Ferchiou M, Zine S, Meriah S. Maternal hydrocephaly and pregnancy. *Rev. Fr. Gynecol. Obstet.* 89(2):88-90.
21. Murakami M, Morine M, Iwasa T, Takahashi Y, Miyamoto T, Hon PK, Nakagawa Y. Management of maternal hydrocephalus requires replacement of ventriculoperitoneal shunt with ventriculoatrial shunt: a case report. *Arch. Gynecol.Obstet.* 2010,282 (3):339-42.
22. Okagaki A, Hanzaki H, Moritake K, Mori T.1990. Case report: pregnant woman with a ventriculoperitoneal shunt to treat hydrocephalus. *Asia. Oceania. J.Obstet. Gynaecol.* 1990,16: 111-3.
23. Orega M, Eholier S P, Boni N, Konan A, Koffi A L. Méningites purulentes et complications neuro-chirurgicales chez l'enfant à Abidjan. *Médecine d'Afrique Noire.* 2005,52(4):251-2.
24. Rees GJ, Francis C, Sizer A R. Convulsions in an undiagnosed pregnancy due to blocked ventriculoperitoneal shunt.*J. Obstet. Gynaecol.* 2008,26(5), 533-4.
25. Riffaud L, Ferre, JC, Carsin-Nicol B, Morandi X. Endoscopic third ventriculostomy for the treatment of obstructive hydrocephalus during pregnancy.*Obstet. Gynecol.* 2006,108:801-4.
26. Sova M, Smrcka, M, Baudysova O, Gogela J Management of a shunt malfunction during pregnancy. *Bratisl Lek Listy.* 2001,102(12):562-3.
27. Wishoff JH, Kratzert KJ, Handwerker SM, Young BK, Epstein F. Pregnancy in patients with cerebrospinal fluid shunts: report of a series and review of the literature. *Neurosurgery.* 1991,29(6):827-31.



Contribution of memory evaluation in temporal epilepsy surgery

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ABSTRACT

Memory assessment is a crucial step in the pre-surgical assessment of temporal epilepsy. Indeed, it participates to a certain extent in the process of localization of the epileptogenic zone and also makes it possible to anticipate the possible risks of surgical treatment on the memory.

We propose to specify the contribution of memory evaluation in 60 patients' candidates for temporal epilepsy surgery, in terms of localization of the "epileptogenic zone" and in the appreciation of the risk of postoperative memory decline.

I. INTRODUCTION

During the preoperative assessment, the neuropsychological results, particularly in terms of memory evaluation, participate to a certain extent in the process of localizing the epileptogenic zone. They also make it possible to anticipate the possible risks of a surgical treatment on memory and language.

We propose to specify the contribution of memory evaluation in 60 patients' candidates for temporal lobe epilepsy surgery, in terms of localization of the "epileptogenic zone" and in the appreciation of the risk of postoperative memory decline. We present a case of a patient who has one of the most extended survival lengths reported in the literature despite unusual multisite metastases but deteriorated rapidly due to very aggressive cranium metastasis.

II. PATIENTS AND METHODS

1. Population studied

We included in our study 60 patients on the following criteria:

- Severe, drug-resistant temporal lobe epilepsy (at least 03 major antiepileptics at optimal doses have been tried as monotherapy and in association with an evolutionary decline of at least 02 years);

Keywords

memory,
pre-surgical assessment of
epilepsy,
pharmaco-resistant temporal
lobe,
epilepsy



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- Age less than 50 years;
- Presence of a temporal lesion on brain MRI;
- Patients whose "epileptogenic zone" is clearly defined at the end of the non-invasive pre-surgical exploration.
- Patients operated on for epilepsy surgery with a regulated temporal lobectomy with broad hippocampal resection.

2. Applied methods

- We have studied hemispheric dominance in all of our patients.
- We specified the anatomical lesions found on brain MRI and we separated the patients into two groups (one group with hippocampal sclerosis and another with a lesion of another nature).
- We also specified the lateralization of the "epileptogenic zone" and the extent of the surgical procedure (hippocampus resected or not).
- An overall intellectual efficiency (MMS) and memory (verbal and visual) assessment was performed on all of our patients. The exploration of verbal memory was carried out by the Grober and Buschke test. As for visual memory, it was assessed by the DMS-48 test. The frequency of memory disorders was evaluated pre-operatively in the whole population and was then evaluated according to the associated lesions and the lateralization of the "epileptogenic zone"
- The development of postoperative memory disorders was specified in all patients and was then studied according to the preoperative memory status and the nature of the lesions found on MRI).

3. Place and period of the study

The patients were recruited from the Epileptology consultation of the department of Neurology of Bab El Oued University Hospital over a period from January 2010 to December 2019.

III. RESULTS

Distribution of patients according to manual dominance:

100% of patients are right-handed.

Distribution of patients according to the nature of the lesion found on MRI:

32 patients had hippocampal sclerosis on brain MRI (53%) and 28 patients had a lesion of another nature (Neuroepithelial dysembryoplastic tumours, Gangliogliomas and finally Cavernomas) (47%).

Distribution of patients according to the lateralization of the epileptogenic zone:

The lateralization of the epileptogenic zone was on the left (dominant hemisphere for language) in 27 patients (45%) and on the right (minor hemisphere) in 33 patients (55%).

	Number of patients	Percentage %
Presence of memory deficit	39	65
Absence of memory deficit	21	35
Total	60	100

Table 1. Frequency of preoperative memory disorders in all operated patients.

A memory deficit is found preoperatively in 65% of operated patients.

	Epileptogenic zone on the left (dominant hemisphere)		Epileptogenic zone on the right (minor hemisphere)	
	Number of patients	Percentage %	Number of patients	Percentage %
Deficit of the visual memory	00	00	33	100
Deficit of the verbal memory	27	100	00	00
Total	27	100	33	100

Table 2. Nature of memory deficit according to the lateralization of "the epileptogenic zone".

All the patients with an epileptogenic zone lateralized on the left (dominant hemisphere) have a deficit in verbal memory and all patients with an epileptogenic zone on the right (minor hemisphere) have a deficit in visual memory.

	Group with Hippocampal Sclerosis		Group with another lesion	
	Number of patients	Percentage %	Number of patients	Percentage %
Presence of memory deficit	30	94	09	32

Absence of memory deficit	02	06	19	68
Total	32	100	28	100

Table 3. Frequency of preoperative memory disorders according to the nature of the lesion.

A memory deficit is found in 93% of patients with hippocampal sclerosis and only in 32% of patients with a lesion of another nature.

	Number of patients	Percentage %
Post-operative decline	9	15
No effects	51	85
Improvement	00	00
Occurrence of global amnesia	00	00
Total	60	100

Table 4. Postoperative memory status in all operated patients.

Postoperative memory decline is noted in 15% of operated patients. No cases of global amnesia have been noted.

	Group with pre-operative deficit		Group without pre-operative deficit	
	Number of patients	Percentage %	Number of patients	Percentage %
Post-operative decline	00	00	09	43
No effects	39	100	12	57
Improvement	00	00	00	00
Total	39	100	21	100

Table 5. Postoperative memory status based on the presence or absence of a preoperative memory deficit.

No patient had a memory decline in the preoperative memory deficit group; whereas a decline is noted in 43% of the cases in the group without preoperative deficit.

	Group with Hippocampal Sclerosis	Group with another lesion

	Number of patients	Percentage %	Number of patients	Percentage %
Post-operative decline	02	06	07	25
No effects	30	94	21	65
Improvement	00	00	00	00
Total	32	100	28	100

Table 6. Postoperative memory status depending on the nature of the lesion found on MRI.

No patient had memory decline in the hippocampal sclerosis group; while a decline is noted in 25% of cases in another lesion group.

IV. DISCUSSION

The high frequency of memory disorders found in our series confirms the data in the literature. Indeed, temporal lobe epilepsy is the greatest provider of memory disorders observed in epileptic patients [1]. Patients with hippocampal sclerosis were more likely to have a preoperative memory deficit than patients with other lesions. These results agree with the data in the literature; indeed, the presence of hippocampal sclerosis, whether right or left, appears to be a predictive factor for the presence of memory impairment in a patient with temporal lobe epilepsy [2; 3]. In addition, the severity of the memory deficit appears to be correlated with the degree of neuronal loss of the diseased hippocampus [4]. It has also been shown that when temporal lobe epilepsy is linked to a structural lesion of another nature, disorders of verbal memory are correlated with left hippocampal neuronal loss [5].

In our series, the nature of the memory deficit observed in right-handed patients depended on the side on which the epileptogenic zone was lateralized. Indeed, the deficit concerned in all cases the verbal memory when the epileptogenic zone was lateralized on the left (dominant hemisphere) and the visual memory when the epileptogenic zone was lateralized on the right (minor hemisphere). This observation is consistent with the data in the literature which recognizes a lateralizing value for the memory deficit [6]. Indeed, temporal lobe epilepsies concerning the dominant hemisphere are characterized by a deficit in verbal memory [7; 8; 2; 9] and temporal lobe epilepsies concerning the

minor hemisphere (the right for right-handers) can be associated with a deficit of non-verbal memory [10; 11; 12].

The risk of a memory deficit after an anterior temporal lobectomy is much greater when the preoperative memory efficiency is normal [13; 14; 15; 16; 17]; This could be explained by the fact that the absence of a preoperative memory deficit suggests that the hippocampus concerned by the excision is still functional.

The data from our study confirm this view; indeed, patients who underwent an intervention comprising a large hippocampal resection with a preoperative memory deficit, did not degrade their memory scores postoperatively. This could be explained by the fact that the preoperative deficit suggests a loss of the function of the hippocampus concerned by the resection.

This favourable postoperative outcome in terms of memory is also noted in patients with hippocampal sclerosis with a correct preoperative memory score. This recalls the favourable prognostic value, in terms of memory, of the presence of hippocampal sclerosis (see above). Indeed, the presence of unilateral hippocampal sclerosis (especially when it is severe) with a correct memory score, is compatible with a loss of hippocampal function compensated by the contralateral side.

Finally, no global amnesia was noted due to the absence of signs indicating a dysfunction of the contralateral hippocampus, such as the case of simultaneous impairment of verbal and visual memory. This notion underlines the major interest of memory evaluation in the context of the pre-surgical assessment of temporal lobe epilepsy surgery.

IV. CONCLUSION

During the preoperative assessment of temporal epilepsies, the results of the memory assessment participate to a certain extent in the process of localization and lateralization of the epileptogenic zone.

They also make it possible to anticipate the possible risks of a surgical treatment on the memory.

REFERENCES

1. Rouleau I. La neuropsychologie de l'épilepsie: 50 ans de progrès. *Revue de neuropsychologie* 2010; 2: 2703-271.

2. Hermann BP, Seidenberg M, Schoenfeld J, Davies K. Neuropsychological characteristics of the syndrome of mesial temporal lobe epilepsy. *Arch Neurol* 1997 ; 54:369-76.
3. Marques CM, Caboclo LOSF, da Silva TI et al. Cognitive decline in temporal lobe epilepsy due to unilateral hippocampal sclerosis. *Epilepsy Behav* 2007; 10: 477-85.
4. Sallie B. The impact of epilepsy surgery on cognition and behavior. *Epilepsy Behav* 2008; 12: 592-9.
5. Sass KJ, Spencer DD, Kim JH et al. Verbal memory impairment correlates with hippocampal pyramidal cell density. *Neurology* 1990; 40: 1694-1697.
6. Henry TR, Roman DD. Presurgical epilepsy localization with interictal cerebral dysfunction. *Epilepsy Behav* 2011; 20: 194-208.
7. Delanney R, Rosen A, Mattson R, Novelly R. Memory function in focal epilepsy : a comparison of non surgical, unilateral temporal lobe epilepsy and frontal lobe samples. *Cortex* 2000; 16:103-117.
8. Hermann BP, Wyler A, Richey E, Rea JM. Memory function and verbal learning ability in patients with complex partial seizures of temporal lobe origin. *Epilepsia* 1987; 28: 547-554.
9. Selwa LM, Serent S, Giordani B et al. Serial cognitive testing in temporal lobe epilepsy: longitudinal changes with medical and surgical therapies. *Epilepsia* 1994; 35: 743-749.
10. Alessio A, Damasceno BP, Camargo CH, Kobayashi E, Guerreiro CA, Cendes F. Differences in memory performance and other clinical characteristics in patients with mesial temporal lobe epilepsy with and without hippocampal atrophy. *Epilepsy Behav* 2004; 5.
11. Helmstaedter C, Pohl C, Hufnager A, Elger C. Visual learning in non-resected patients with right lobe epilepsy. *Cortex* 1991; 27:547-555.
12. Jones-Gotman M. Presurgical neuropsychological evaluation for localization and lateralization of seizure focus. In: Lüders H, Ed. *Epilepsy surgery*. New York : Raven Press, 1991: 469-75.
13. Baxendale S, Thompson P, Harkness W, Duncan J. Predicting Memory Decline Following Epilepsy Surgery: A Multivariate Approach. *Epilepsia* 2006; 47: 1887-94.
14. Helmstaedter C, Kurthen M, Lux et al. Chronic epilepsy and cognition : a longitudinal study in temporal lobe epilepsy. *Ann Neurol* 2003; 54: 425-432.
15. Jones-Gotman M, Smith ML, Risse GL, Westerveld M, Swanson SJ, Giovagnoli AR, et al. The contribution of neuropsychology to diagnostic assessment in epilepsy. *Epilepsy Behav* 2010; 18:3-12.
16. Lee TMC, Yip JTH, Jones-Gotman M. Memory Deficits after Resection from Left or Right Anterior Temporal Lobe in Humans: a Meta-Analytic Review. *Epilepsia* 2002; 43: 283-91.
17. Rausch R, Kraemer S, Pietras CJ et al. Early and late cognitive changes following temporal lobe epilepsy. *Neurology* 2003; 60: 951-959.

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