ISSN 1220-8841 (Print)
ISSN 2344-4959 (Online)

ROMANIAN NEUROSURGERY

Vol. XXXIV | No. 1 March 2020

The Official Journal of
"Romanian Society of Neurosurgery"
- since 1982 -

LONDON ACADEMIC PUBLISHING

Editorial and Advisory Board

ROMANIAN

NEUROSURGERY

Vol. XXXIV | No. 1 March 2020

London
Academic Publishing

ISSN 1220-8841 (Print)
ISSN 2344-4959 (Online)

First Printing: March 2020
London Academic Publishing Ltd.
27, Old Gloucester Street
WC1N 3AX
London, United Kingdom
Email: contact@lapub.co.uk

www.lapub.co.uk
www.journals.lapub.co.uk
www.journals.lapub.co.uk/index.php/roneurosurgery

Company Reg. No. 10941794
Registered in England and Wales

Contents

Romanian Neurosurgery (2020) XXXIV (1): pp. 9-16
DOI: 10.33962/roneuro-2020-001
www.journals.lapub.co.uk/index.php/roneurosurgery

Overview of patients with vascular pathology and cost analysis of medical care – research market for an entrepreneurial project

Aurelia Mihaela Sandu[1,2], Adrian Mircea Fürtös[1], Radu Mircea Gorgan[1,2]

[1] Department of Neurosurgery, Emergency Clinical Hospital Bagdasar-Arseni, Bucharest, ROMANIA
[2] University of Medicine and Pharmacy "Carol Davila", Bucharest, ROMANIA

Keywords
aneurysm,
arteriovenous malformation,
cavernoma,
dural arteriovenous fistula,
health care costs

Corresponding author:
Aurelia Mihaela Sandu

University of Medicine and Pharmacy "Carol Davila", Bucharest, Romania

aurasandu@gmail.com

ABSTRACT

Introduction: Brain vascular pathology, aneurysms, arteriovenous malformations (AVMs), cavernomas, dural arteriovenous fistulas (DAVF), venous angiomas and capillary telangiectasia, represents a serious health problem worldwide.

Aim: The aim of this article is to perform an analysis of patients with brain vascular pathology and to analyze costs of health services, a research market for an entrepreneurial project, in order to design guidelines for patients' selection and treatment.

Material and methods: We performed an observational, descriptive study of patients with vascular pathology, from 2018 to 2019.

Results: A total of 153 patients with brain vascular pathology were admitted in our department. Mean age was 49.53 ± 13.997 years. Sex ratio was 0.86. Mean hospital stay was 11.33 ± 13.724 days. Seventy-four patients (48.37%) underwent surgery. Seventy-eight patients (50.98%) had cerebral aneurysms. Complications were seen in 24 patients (30.77%) and vasospasm in 32 cases (41.03%). Thirty-six patients underwent surgery. Outcome was favorable, according to mRS(p=0.001) and Karnofsky score(p=0.006). Thirty-three patients (21.57%) had brain AVMs. Twenty patients underwent surgery. Complications were seen in 4 cases (12.12%). The outcome was favourable, according to mRS(p=0.001) and Karnofsky score (p=0.002). Thirty-nine patients (25.49%) had cavernomas. Surgery was performed in 18 cases. The outcome was favourable, according to Engel Epilepsy Surgery Outcome Scale, mRS (p=0.000) and Karnofsky score (p =0.000). Costs of health services were correlated with longer hospitalization, higher mRS, lower Karnosfsky score, presence of complications and presence of vasospasm.

Conclusions: Proper treatment of brain vascular pathology ensures a favourable outcome. Adequate patients' selection and choosing the best treatment can reduce costs. Surgery is the treatment of choice in ruptured aneurysms, AVMs and cavernomas. Early surgery, with specific treatment of the vascular lesion and removal of intracranial blood ensures a better outcome, with lower medical costs. Prevention of complications, aggressive treatment of vasospasm reduces medical costs.

Scan to access the online version

Introduction

Brain vascular pathology is represented by aneurysms and vascular malformations. Vascular malformations are arteriovenous malformations (AVMs), cavernomas, duralarteriovenousfistulas (DAVF), venous angiomas and capillary telangiectasia.[1]

Brain vascular pathology is a serious health problem world-wide. The incidence of cerebral aneurysms is 0.2-7.9% and the prevalence is 5%.[2] The incidence of brain AVMs is 0.15-3%[3,4] and the prevalence is 0.14%[5,6]. The incidence of cavernomas is 0.15-0.56% and the prevalence is 0.17-0.9%.[7] DAVF, venous angiomas and capillary telangiectasia are rare.

Unfortunately brain vascular pathology occurs in young, active people and more than half of patients present with intracerebral hemorrhage, with significant morbidity and mortality. Peak incidence of aneurysms is 55-60 years[2], AVMs become symptomatic in younger people, mean age is 33 years[5,6] and in cavernomas is 42 years[8]. The most common form of presentation is rupture with subsequent brain hemorrhage. Pattern of hemorrhage depends on type of vascular disease. Aneurysms usually rupture in the subarachnoid space, while AVMs and cavernomas cause intraparenchymatal hematoma. Regardless the type of intracerebral hemorrhage, all are major aggressive factors to the brain and carry important risks of death or development of serious neurological complications. Other forms of presentation are seizures, cranial nerves palsies, motor or sensory deficits, intracranial hyperpressure, ischemia, hydrocephalus, etc.

Surgery is the treatment of choice for aneurysms, AVMs and cavernomas.[9-11]Even though a benign pathology, surgery for brain vascular pathology carries significant risks. For cerebral aneurysms embolization can also be tried.[11] Besides surgery, AVMs also benefit from stereotactic radiosurgery and embolization. DAVF is usually treated through embolization and venous angiomas and capillary telangiectasia do not require treatment.

The aim of this article is to perform an analysis of patients with brain vascular pathology and to analyze the costs of health services, a research market for an entrepreneurial project, in order to design guidelines for patients' selection and treatment.

Material and methods

We performed an observational, descriptive study in which we included patients with vascular pathology admitted in the IVth Department of Neurosurgery, Emergency Clinical Hospital Bagdasar-Arseni, from January 2018 to December 2019. Data were collected in a retrospective manner from January 2018 to August 2019 and prospectively from September 2019 to December 2019. We collected data from hospital medical records and electronic health record Hipocrate. Statistic analysis of data was done using SPSS®.

Results

A total of 153 patients with brain vascular pathology were admitted over a period of time of two years in the IVth Department of Neurosurgery, Emergency Clinical Hospital Bagdasar-Arseni from Bucharest, Romania.Patients with brain vascular pathology represented 2.09% from the total number of admissions in our department.

Vascular brain pathology was represented by aneurysms, AVMs, cavernomas and DAVF. (Figure 1) Mean age of all patients with vascular pathology was 49.53 ± 13.997 years. Sex ratio (M/F) was 0.86. Mean hospital stay was 11.33 ± 13.724 days. A total of 74 patients (48.37%) underwent surgery. The rest of 73 patients either had no indications for surgery, refused surgery or benefit from other nonoperative treatments.

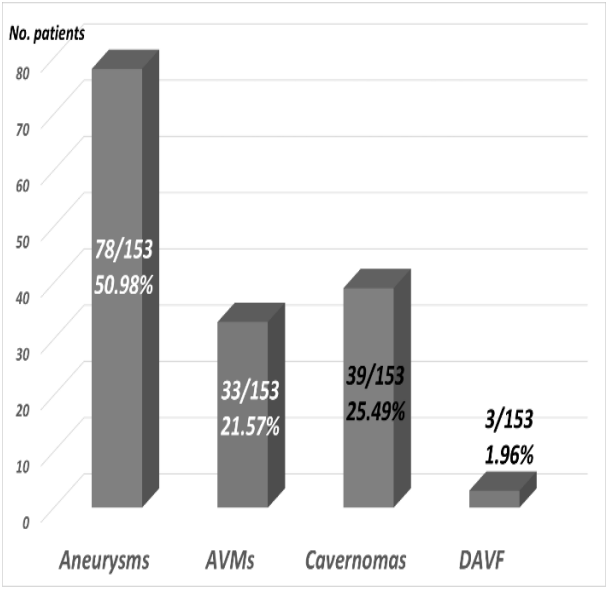


Figure 1. Types of brain vascular pathology.

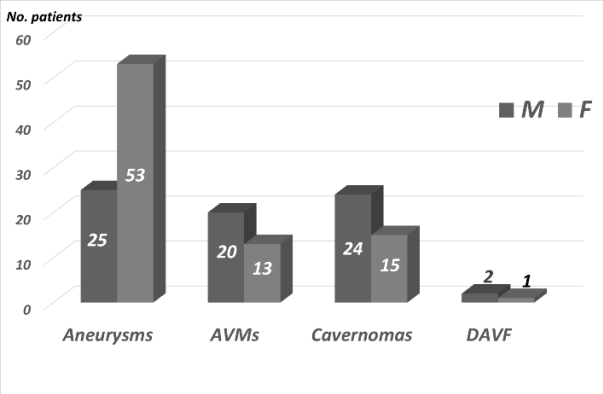


Figure 2. Sex ratio per each type of vascular pathology.

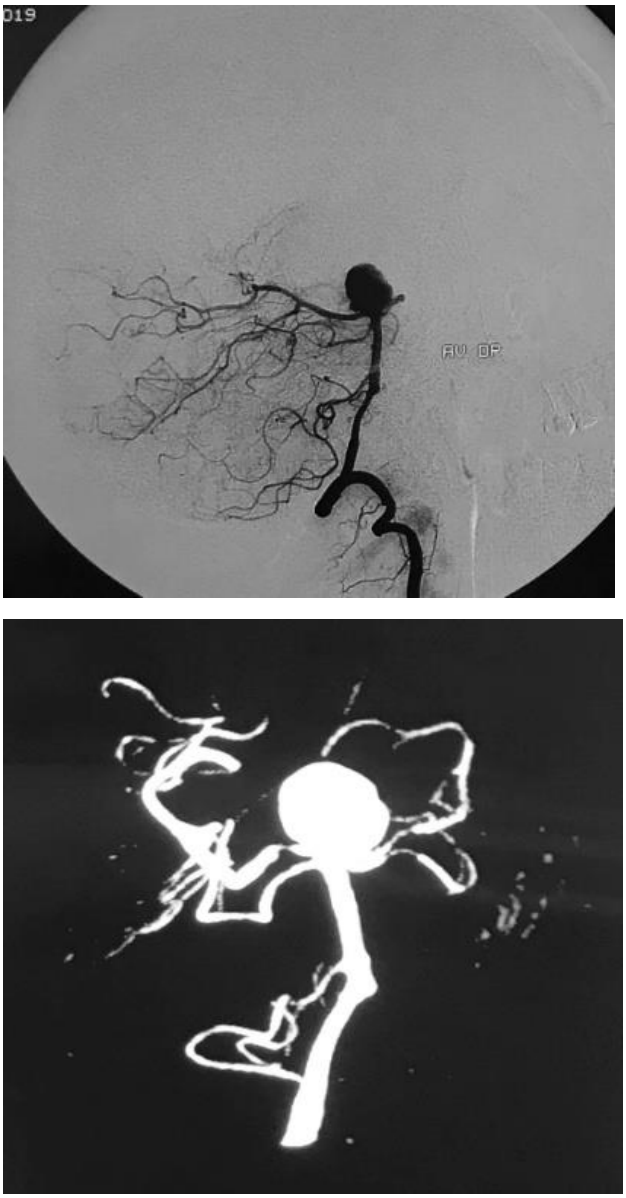


Figure 3. Basilar tip aneurysm.

Aneurysms

Seventy-eight patients (50.98%) were admitted with cerebral aneurysms. Mean age of these patients was 53.73 ± 11.146 years. There were 53 women (67.95%) and 25 men (32.05%). Sixty-two patients (79.49%) had single aneurysm and 16 patients (20.51%) had multiple aneurysms. The latter 16 patients harbor a total number of 35 aneurysms, varying from 2 to 6 lesions per patient. Aneurysms' locations are shown in table 1. Forty-eight (61.54%) had ruptured aneurysms and only 30 people (38.46%) had unruptured ones. From the total number of patients with ruptured aneurysms, according to Hunt and Hess scale, 22 patients were grade I, 4 grade II, 3 grade III, 8 grade IV and 11 grade V. According to Fisher scale, 2 patients had grade I, 17 grade II, 8 grade III and 21 grade IV. Patients with ruptured aneurysms had the following pattern of brain hemorrhage: 45 had subarachnoid hemorrhage, 9 had intraparenchymatal hematoma, 19 intra-ventricular hemorrhage and 2 had subdural hematoma.

Complications were seen in 24 patients (30.77%). Vasospasm, a fearful complication was seen in 32 patients (41.03%). Vasospasmwas more commonly encountered in patients with higher grades on Hunt and Hess and Fisher scales (p=0.000, p=0.000).

Thirty-six patients underwent surgery (34 patients had their aneurysms clipped and in 2 wrapping was done). Outcome was favorable, mRS at discharge were lower than mRSat admission (Z=-3.426, p=0.001) and final Karnofsky scores were higher than initial scores (Z=-2.754, p=0.006).

AVMs

Thirty-three patients (21.57%) were admitted with brain AVMs. Mean age was 40.82± 14.501 years. There were 20 males (60.61%) and 13 females (39.39%). Sixteen patients (48.48%) presented with ruptured AVMs and 13 cases (39.39%) had history of seizures. According to brain hemorrhage pattern 12 patients had intraparenchymatal hematoma, 4 had intraventricular hemorrhage, 3 had subarachnoid hemorrhage and only one had subdural hematoma. Motor deficit was encountered in 11 cases. According to Spetzler-Martin scale, 9 cases (27.27%) were grade I, 8 patients (24.24%) were grade II, 8 (24.24%) grade III, 3 patients (9.09%) were grade IV, 3 cases (9.09%) were grade V and 2 cases (6.06%) had grade VI AVMs. Three patients had flow-related associated

aneurysms. No patient presented cerebral vasospasm.

Twenty patients underwent surgery, 5 cases underwent stereotactic radiosurgery Gamma Knife and 2 patients were embolized. Five patients refused surgery and 2 cases had grade VI Spetzler-Martin AVMs and did not benefit from therapy. Complications were seen in 4 cases (12.12%).

Outcome was favorable, discharge mRSwere lower than admission mRS (Z=-3.332, p=0.001) and finalKarnofsky scores were higher than initial scores (Z=-3.047, p=0.002).

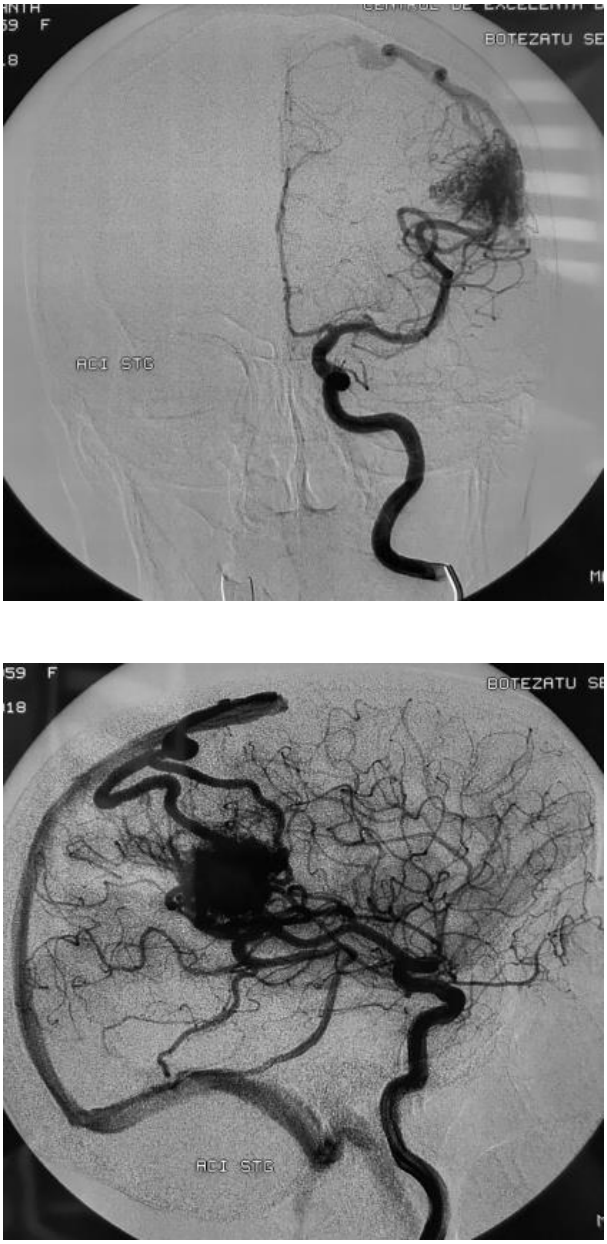


Figure 4. Grade III Spetzler-Martin AVM.

Cavernomas

Thirty-nine patients (25.49%) were admitted with brain cavernomas. Mean age for patients with cerebral cavernomas was 48.28 ± 15.788. Sex ratio (M/F) was 1.6. Thirty-three patients (84.62%) had single cavernomas, but 6 (15.38%) had multiple lesions, the number of cavernomas varying from 3 to 8. Ruptured cavernomas were found in 16 patients. According to the pattern of brain hemorrhage, all 16 patients had intraparenchymatal hematoma, one associated intraventricular hemorrhage and one subdural hematoma. Nine patients had motor deficit and 10 cases were admitted with seizures. Positive diagnosis was done by MRI, T2 echo-gradient and FLAIR. Only 6 patients underwent DSA, and the angiography was negative in all cases. Surgery was performed in 18 cases. Deep cavernomas required intraoperative usage of ultrasound-based neuronavigation system Sonowand®. From the total of 10 patients with epilepsy, 6 underwent surgery, and 3 were free of disabling seizures (Engel IA) postoperative, 2 had worthwhile improvement (Engel IIIB) and one had no worthwhile improvement (Engel IVB). Outcome was favorable, according to mRS (Z=-4.274, p=0.000) and Karnofsky score (Z=-4.283, p =0.000).

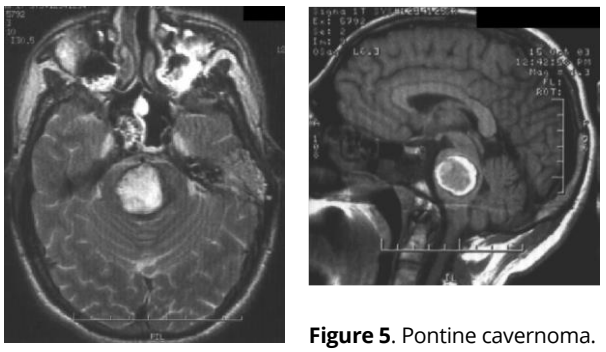


Figure 5. Pontine cavernoma.

Dural AV fistulas

There were only 3 patients (1.96%) with DAVF. Mean age was 52.33 ± 4.933.

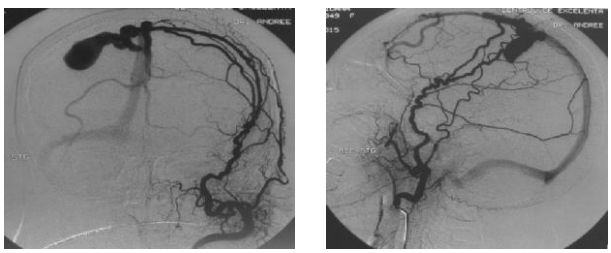


Figure 6. DAVF type IV Merland-Cognard, injected from middle meningeal artery.

Total health care costs accounted for 3215558.4658 lei, and they were composed by 2574452.06 lei hospitalization costs, 14067.4 lei food costs, 15993.085 lei medication costs, 18641.038 lei blood work costs, 105797 lei imaging examinations costs. Hospital staywas positively correlated with hospitalization costs (p=0.000), food costs (p=0.000), medication costs (p=0.000), medical products costs (p=0.000), blood work costs (p=0.000), imaging examinations costs (0=0.002) and total cost of health care(p=0.000). Patients who underwent surgery have higher costs (p=0.000). Patients who presented complications and vasospasm have also higher costs (p=0.000, p=0.000). Higher mRS and lower Karnofsky score at admission were associated with higher costs (p=0.000, p=0.000). Another 575 days for postoperative controls were gathered with additional total costs of 638406.916 lei.

Discussions

Brain vascular pathology was encountered in young, active patients, meanage for the entire group being under 50 years old, with differences for every pathology.

Surgery for vascular lesions is challenging. Only 50% of patients diagnosed with vascular pathology benefit from surgery. This low rate of patients with surgery is caused by several factors. Surgery is indicated when benefits are higher than risks, and we must not forget that we are dealing with high risk surgeries, so there are patients with grade VI AVMs, deep seated unruptured, asymptomatic cavernomas or unruptured ectatic vessels, in which surgery is not required. More, there are patients, or families who refused surgery after they were informed of the possible risks. And there is another group of patients who are not candidate for surgery.

In case of brain aneurysms clipping is the curative treatment.[11,12] Surgical technique depends on location, type and size of aneurysm and angioarchitecture of parental and efferent vessels.[9]One or more clips of different sizes can be used to secure the aneurysm (simple clipping, multiple clipping, tandem clipping, fenestration tubes, clip reconstruction).[9]Other surgical techniques are wrapping and trapping. Surgery for aneurysm must be performed early, to hinder the risk of rebleeding.

Found almost exclusively in cerebral aneurysms, vasospasm is causing important morbidity. Vasospasm occurs between days 3 and 14 after subarachnoid hemorrhage onset, and it is due to presence of blood in the subarachnoid space, which induces arterial vasoconstriction. Vasospasm is associated with higher Fisher and Hunt and Hess grades. Many surgeons do not recommend surgery in this period of time because it increases the risk of vasospasm. Early intensive treatment for vasospasm, such as triple H therapy (hypertension, hypervolemia and hemodilution), intraoperative washing of all subarachnoid spaces from blood clots, calcium channel blockers (nimodipine) or angioplasty may reduce the incidence of vasospasm and its devastating consequences. So, ideally surgery is done early after rupture, before installation of vasospasm and postoperative medical treatment is initiated. But if patient is not admitted in a specialized center before day 3, timing for surgery is debatable. We recommend operating the patient even in the delicate period from day 3 to 14, because clipping the aneurysm allows intraoperative removal of blood clots and postoperative triple H therapy. In our opinion removal of subarachnoid blood, triple H therapy and nimodipine are the most effective treatments. Costs are influences by occurrence of complications, mainly vasospasm, so preventionof vasospasm reduces costs. Vasospasm is not found in patients with AVMs or cavernomas, because they do not bleed in the subarachnoid space, so for this group of patients, specific treatment for vasospasm is not indicated.

Surgery is the treatment of choice in AVMs.[13,14] Complete resection ensures cure of the lesion. Stereotactic radiosurgery causes progressive occlusion of vessels, but unfortunately the effect occurs in time, and until 1-2 years the nidus is still active. Embolization is recommended only as adjuvant before surgery or in grade VI AVMs.[14]The effect of embolization is limited in time, surgery must be performed in the first week after occlusion procedure. After one month the nidus recruits new blood vessels. Principles of surgery for AVMs depend on location, Spetzler-Martin score and angioarchitecture of the lesion. We recommend surgery for all ruptured AVMs and in all lesions located in non-eloquent areas.[15] Conservative treatment and stereotactic radiosurgery can be tried in patients with small, unruptured, located in eloquent areas AVMs.[13] Surgical principles are represented by early coagulation of feeding arteries,

circumferential dissection of the nidus progressing deep and, after complete arterial disconnection of the nidus occlusion of draining veins.[10]We do not perform preoperative embolization of AVMs because it carries the risk of intraprocedural nidus rupture and developing normal perfusion pressure breakthrough. But other authors consider it helpful before surgery.

The angiography was negative in all cases with cavernomas. They are considered to be occult angiographic lesions, because they have very low flow and do not enhance with contrast. We do not recommend angiography in cases suggestive for cavernomas. The imaging of choice is T2 echo-gradient and FLAIR MRI. We recommend surgery in patients with ruptured cavernomas, with intraparenchymatal hematoma, in symptomatic cavernomas and in superficial accessible lesions. Deep, unruptured and asymptomatic cavernomas are kept under surveillance. For brainstem cavernomas, we strongly recommend surgery after the first bleeding, because the second has devastation consequences. In supratentorial cavernomas we recommend resection of hemosiderin ring surrounding the cavernoma. Brain surrounding the lesion impregnated with hemosiderin can be responsible for seizure persistence. So resection of hemosiderin ring ensures good outcome, evaluated through Engel Epilepsy Surgery Outcome Scale. In lesions located in the brainstem we recommend resection of the lesion leaving the hemosiderin ring in place. Infratentorial cavernomas do not produce seizure, and aggressive resection in this delicate area full with eloquent structures induces severe neurological deficits.

We recommend embolization of DAVF.

Total health care costs of patients is composed by hospitalization costs, food costs, medication costs, medical products costs, blood work costs and imaging exams costs.

Longer hospitalization increases costs. Higher costs in the group of patients who underwent surgery compare with patients belonging to the conservative group, even if it is highly significant, it is not relevant because we are a neurosurgical department and we only keep patients for surgery. If a patient has no surgical indication or refuses surgery, we discharge him and he will be treated further in the territorial neurological department. So costs for nonsurgical patients are not adequately evaluated. So, we can conclude that our study illustrates best the surgical group. Food costs are bound to hospitalization cost and comorbidities (e.g. diabetes, renal failure, etc.). Usually blood work costs are not so high, we only take the minimum set before surgery. Vascular pathology does not require special, expensive blood test before surgery. It may increase if patients require repetitive blood analysis (e.g. postoperative anemia, patients in ICU, etc.). Unfortunately, other costs, such as costs with the medical personnel, cost of surgery or costs for days in the ICU, which are included in a bill from the private hospitals, are not included in a state hospital.

Complications also increased costs. So, adequate patients' selection and choosing the best treatment for lowering the rate of possible complications can reduce costs.

Cost analysis is useful as a market research to predict resources needed for patients' investigation and treatment. Such analysis is a useful tool in developing guidelines. The principles of guidelines choosing the best treatment, while optimizing health care services.

A limit of the study is represented by the fact that it precisely illustrates the surgical group, but it is not so reliable in characterizing the conservative group.

Conclusions

Proper treatment of brain vascular pathology ensures a favorable outcome. Adequate patients' selection and choosing the best treatment to lower the rate of possible complications can reduce costs. Surgery is the treatment of choice in ruptured aneurysms, AVMs or cavernomas. Early surgery, with specific treatment of the vascular lesion and removal of intracranial blood ensures a better outcome, with lower medical costs. Prevention of complications, aggressive treatment of vasospasm reduces medical costs. Further analysis is needed to perfect guidelines for treatment of patients with brain vascular pathology.

Table 1. Aneurysms' location.

Aneurysm location	Single aneurysm	Multiple aneurysms	Total no.	Total %
ICA segm C4	3	1	4	4.12
ICA segm C5	4	4	8	8.25
ICA segm C6	10	10	20	20.62
ICA segm C7	2	1	3	3.09
ACoA	18	1	19	19.59
ACA	5	3	8	8.25
MCA	9	11	20	20.62
ACoP	4	3	7	7.22
SCA	1	0	1	1.03
Basilar artery	4	1	5	5.15
PICA	1	0	1	1.03
Vertebral artery	1	0	1	1.03
Total	**62**	**35**	**97**	**100**

Table 2. AVMs' location.

AVM location	Total no.	Total %
F	7	21.21
T	4	12.12
P	4	12.12
O	5	15.15
FP	3	9.09
FT	1	3.03
Sylvian fissure	1	3.03
TP	3	9.09
TPO	2	6.06
Intraventricular	1	3.03
Posterior fossa	2	6.06
Total	**33**	**100**

ACKNOWLEDGEMENT

This paper was co-financed from the Human Capital Operational Program 2014-2020, project number POCU / 380/6/13/125245 no. 36482 / 23.05.2019 "Excellence in interdisciplinary PhD and post-PhD research, career alternatives through entrepreneurial initiative (EXCIA)", coordinator The Bucharest University of Economic Studies".

CONFLICT OF INTERESTS

The authors declare no conflict of interests.

ABBREVIATIONS

ACA anterior cerebral artery
ACoA anterior communicating artery
ACoP anterior posterior artery
AVM arteriovenous malformation
DAVF dural arteriovenous fistula
ICA internal carotid artery
MCA middle cerebral artery
mRS modified Rankin score
PICA posteroinferior cerebellar artery
SCA superior cerebellar artery

REFERENCES

1. McCormick WF. The pathology of vascular ("arteriovenous") malformations. J Neurosurg 24, 807-816. 1966.
2. Greenberg MS. SAH and aneurysms. In Greenberg MS (ed). Handbook of neurosurgery. New York: Thieme Medical Publishers; 2010:1055-1097.
3. Misra M, Aletich V, Charbel FT, Debrun GM, Ausman JI. Multidisciplinary approach to arteriovenous malformations. In Kaye AH, Black PM (eds). Operative neurosurgery. London: Harcourt Publishers Limited; 2000:1137-1151.
4. Stapf C, Mast H, Sciacca RR, Berenstein A, Nelson PK, Gobin YP, Pile-Spellman J, Mohr JP. The New York islands AVM study: design, study progress, and initial results. Stroke 34, e29-34. 2003.

5. Altschul D, Smith ML, Sinson GP. Intracranial arteriovenous malformation. Medscape eMedicine[Surgery/Neurosurgery/Vascular]. 2011.
6. Greenberg MS. Vascular malformations. In Greenberg MS (ed). Handbook of neurosurgery. New York: Thieme Medical Publisher; 2010:1098-1142.
7. Goldstein HE, Solomon RA. Epidemiology of cavernous malformations. Handb Clin Neurol 2017; 143:241-247.
8. Gross BA, Du R. Hemorrhage from cerebral cavernous malformations: a systematic pooled analysis. J Neurosurg 2017; 126:1079-1087.
9. Lawton MT. Seven aneurysms: tenets and techniques for clipping. New York: Thieme Medical Publishers; 2010.
10. Lawton MT. Seven AVMs - tenets and techniques for resection. New York: Thieme Medical Publishers; 2014.
11. Hulsbergen AFC, Mirzaei L, van der Boog ATJ, Smith TR, Muskens IS, Broekman MLD, Mekary RA, Moojen WA. Long-term durability of open surgical versus endovascular repair of intracranial aneurysms: a systematic review and meta-analysis. World Neurosurg 2019; 132:e820-e833.
12. Lindgren A, Vergouwen MD, van der Schaaf I, Algra A, Wermer M, Clarke MJ, Rinkel GJ. Endovascular coiling versus neurosurgical clipping for people with aneurysmal subarachnoid haemorrhage.
13. Kato Y, Dong VH, Chaddad F, Takizawa K, Izumo T, Fukuda H, Hara T, Kikuta K, Nkai Y, Endo T, Kurita H, Xu B, Benes V, Christian R, Pavesi G, Hodaie M, Sharma RK, Agrawal H, Mohan K, Liew BS. Expert consensus on the management of brain arteriovenous malformations. Asian J Neurosurg 2019; 14:1074-1081.
14. Rutledge C, Nelson J, Lu A, Nisson P, Jonzzon S, Winkler EA, Cooke D, Abla AA, Lawton MT, Kim H. Cost determinants in management of brain arteriovenous malformations. Acta Neurochir (Wien) 2020; 162:169-173.
15. Gorgan RM. Malformaţiile arteriovenoasecerebrale. In Ciurea AV (ed). Tratat de neurochirurgie. Bucuresti: Editura Medicală; 2011:117-129.

Romanian Neurosurgery (2020) XXXIV (1): pp. 17-19
DOI: 10.33962/roneuro-2020-002
www.journals.lapub.co.uk/index.php/roneurosurgery

Management of a patient with acute internal hydrocephalus, ventriculitis and bronchopneumonia. Case report

Balasa Daniel, MD, PhD; Tunas Alexandru, MD

Department of Neurosurgery, Clinical County Hospital, Constanta, ROMANIA

ABSTRACT

A 69-year-old patient, with a long history of lung tuberculosis, with lymphopenia was emergently admitted in our hospital for bronchopneumonia, ventriculitis, acute internal hydrocephalic. He was aggressively treated with iv Meropenem and Vancomycin, intraventricular high doses of Vancomycin, aerosols, Dexametazone with healing of internal hydrocephalus, ventriculitis and improvement of bronchopneumonia.

Keywords
bronchopneumonia, intraventricular vancomycin, limphopenia, ventriculitis

Corresponding author:
Balasa Daniel

University of Medicine and Pharmacy "Carol Davila", Bucharest, Romania

balasadaniel100@yahoo.com

OBJECT

The international data in the literature regarding the treatment of ventriculitis is limited. The authors aimed to share their experience in the treatment of ventriculitis and using intraventricular (IVT) and intravenous antibiotherapy.

CASE REPORT

The patient was a 69 year-old man, with a previous history of lung tuberculosis. He was adressed to the emergency department for 2 days of fever (39 degrees C), 4 days of productive cough with purulent expectoration, dyspnea with tachypnea, respiratory failure (SPO2 87%), altered mental status (stupor, GCS 10), neck stifness. The patient was treated by his family doctor with oral cefuroxyme , 4 grams daily . The patient was imunodepressed (lymphocytes 470/microliter)

Chest X-rays and CT scan revealed bronchopneumonia (Fig. 1)

Head CT Scann: Acute hidrocephalus, ventriculitis, parafluid purulent deposits in occipital horns, bilateral. (Fig 2 - blue arrows)

The lumbar puncture revealed a purulent CSF with 4690 white cells/mm^3, 92% of neutrophils.

The patient underwent emergency surgery, extraventricular drainage (EVD) with double purpose: treatment of acute hydrocephalus and treatment of ventriculitis. The ventricular CSF samples were purulent. For this reason we considered unnecesarry a

Scan to access the online version

complementary head MRI scan. Bacteriologic examination of the CSF revealed: Streptococcus pneumoniae. Intravenous antibiotherapy with Meropenem 6 grams/day and Vancomycin 2 grams /day, intraventricular antibiotics (Vancomycin 50 mg/day in the first postoperative day and 100 mg/day in the following days), iv Dexametasone (6 mg q 6 hours), aerosols with salbutamol 5mg/ml, 1 ml. After 22 days of treatment, control CT Scann revealed disappearance of the acute hydrocephalus and the occipital purulent debris (Fig 3).

Figure 1. Bronchopneumonia. Multiple perihilar opacities, bilateral.

Figure 2.

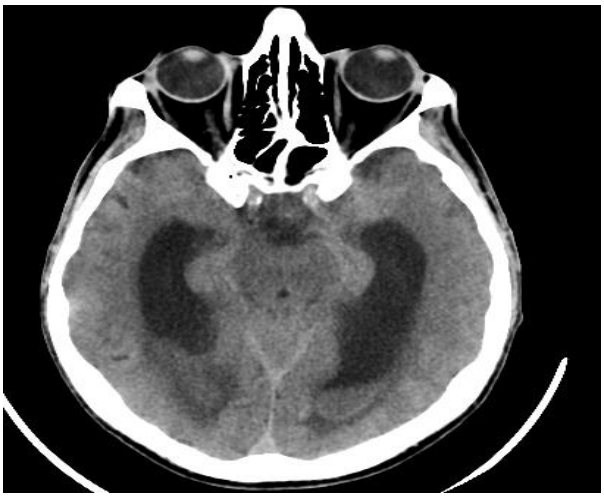


Figure 3. Postoperative contrast CT Scann. No internal hidrocephalus, no ventriculitis. Moderate ventriculomegaly.

The neurological status of the patient progressively improved and the neck stiffness dissapeared, also the respiratory status improved.
Follow time period: 5 months

Discussion

Ventriculitis is defined by high fever, clinical signs of meningitis (nuchal rigidity, photophobia, decreased mental status, seizures), a positive CSF culture, positive Gram stain, decreased CSF glucose, increased CSF proteins, CSF pleocytosis (at least 11 leukocytisis/mL with 50% or more polymorphonuclear neutrophils) [1,2] In the pre-antibiotic era, the great majority of patients died from bacterial meningitis/ventriculitis at the end of the first week of ventriculitis[3]. Nowadays, the incidence of this complication lowered. Central nervous system infections requiring treatment with intraventricular (IVT) vancomycin are becoming increasingly common with advent of intracranial devices and increasing prevalence of multi-drug resistant and nosocomial organisms[4]. Administering vancomycin via IVT route bypasses the blood-brain barrier and allows controlled delivery directly to the desired site of action, achieving higher concentrations for a more reliable bactericidal action[4]. Indications for IVT vancomycin include meningitis unresponsive to intravenous antibiotics, ventriculitis, and intracranial device infections[4]. Recommended dosages of antimicrobial agents

administered by the intraventricular route are vancomycin (5–20 mg/d). Dosages reported in literature ranged from 0.075-50 mg/day, with the most evidence for dosages of 5 to 20 mg/day. Duration of therapy most commonly ranged from 7 to 21 days[4]. For this patient, considering the imunodepression, bronchopneumonia, ventriculitis our option was to administer a higher dose of IVT Vancomycine.The patient received aditional dexamethasone according to current guidelines, as soon as possible when the lumbar puncture reveals a purulent CSF [6]. Such a treatment has been proven to be beneficial in preventing hearing loss and neurological sequelae in adult purulent bacterial meningitis, particularly those due to Streptococcus pneumoniae[6,7]

The mean time to obtain CSF sterilisation in medical literature was 24hours. This time was achieved in this case too. The fever disapeared in the first postoperative day.

There were no adverse effect of antibiotics administered IVT.

Conclusion

Intraventricular and intravenous antibiotics lead very quickly to CSF sterilisation in this critical ill patient.

THE IVT vancomicyne administration appears to be safe and have high efficacy together with IV administration of Vancomicyn and Meropenem.

The IVT Vancomycin dose , higher than the literature data, dramaticaly hastened the healing of ventriculitis and shortened the hospitalisation period of the patient.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest

References

1. Sundbarg G, Kjallquest A, Lundberg N, et al. Complications due to prolonged ventricular fluid pressure recording in clinical practice. In: Brock M, Dietz H, eds. Intracranial Pressure I: Experimental and Clinical Aspects—International Symposium on Intracranial Pressure, Hannover, 1972. Berlin: Springer-Verlag:348–35, 1972.
2. Sundbarg G, Nordstrom CH, Soderstrom S. Complications due to prolonged ventricular fluid pressure recording.Br J Neurosurg;2:485–495, 1988.
3. Adams RD, Kubik CS, Bonner FJ. The clinical and pathological aspects ofinfluenzal meningitis. Arch Pediatr;65:354–76, 1948.
4. Ng K, Mabasa VH, Chow I, Ensom MH. Systematic review of efficacy, pharmacokinetics, and administration of intraventricular vancomycin in adults. Neurocrit Care. Feb;20(1):158-71, 2014.
5. Bijlsma MW, Brouwer MC, Kasanmoentalib ES, Kloek AT, Lucas MJ, TanckMW, et al. Community-acquired bacterial meningitis in adults in theNetherlands, 2006-14: a prospective cohort study. Lancet Infect Dis.2016;16:339–447.
6. Brouwer MC, Mcintyre P, Prasad K, van de Beek D. In: van de Beek D, editor.Corticosteroids for acute bacterial meningitis: Cochrane Database Syst. Rev;2015.https://doi.org/10.1002/14651858.CD004405.pub5.
7. Tunkel AR, Hartman BJ, Kaplan SL, Kaufman BA, Roos KL, Scheld WM, et al.Practice guidelines for the Management of Bacterial Meningitis. Clin InfectDis.;39(9):1267–84, 2004.
8. W Pfisterer, M Mühlbauer, T Czech, A Reinprecht Early diagnosis of external ventricular drainage infection:results of a prospective study . J Neurol Neurosurg Psychiatry;74:929–932, 2003.
9. Ricard JD, Wolff M, Lacherade JC, Mourvillier B, Hidri N, Barnaud G, et al. Levels of vancomycin in cerebrospinal fluid of adult patients receiving adjunctive corticosteroids to treat pneumococcal meningitis: A prospective multicenter observational study. Clin Infect Dis;44:250-5, 2007.
10. Nagl M, Neher C, Hager J, Pfausler B, Schmutzhard E, Allerberger F. Bactericidal activity of vancomycin in cerebrospinal fluid. Antimicrob Agents Chemother;43:1932-4, 1999.
11. Rybak MJ, Lomaestro BM, Rotschafer JC, Moellering RC, Craig WA, Billeter M, et al. Vancomycin therapeutic guidelines: A summary of consensus recommendations from the infectious diseases society of America, the American Society of Health-System Pharmacists, and the Society of Infectious Diseases Pharmacists. Clin Infect Dis;49:325-7, 2009.
12. Wen DY, Bottini AG, Hall WA, Haines SJ. Infections in neurologic surgery. The intraventricular use of antibiotics. Neurosurg Clin N Am;3:343-54, 1992.
13. Fukui MB, Williams RL, Mudigonda S. CT and MR imaging features ofpyogenic ventriculitis. AJNR Am J Neuroradiol.22(8):1510–6, 2001.

Romanian Neurosurgery (2020) XXXIV (1): pp. 20-25
DOI: 10.33962/roneuro-2020-003
www.journals.lapub.co.uk/index.php/roneurosurgery

Accuracy of 2D fluoroscopy with preoperative CT fused neuronavigation in thoracic and lumbar pedicle screw insertion

Adrian Bălașa[1,2], Corina-Ionela Hurghiș[2], Flaviu Tămaș[2], Ioan-Alexandru Florian[3,4], Levente Peter[2], Rareș Chinezu[1,2]

[1] University of Medicine, Pharmacy, Science and Technology "George Emil Palade", Tirgu Mures, ROMANIA
[2] Neurosurgery Clinic, Clinical Emergency Hospital, Tirgu Mures, ROMANIA
[3] University of Medicine and Pharmacy "Iuliu Hatieganu", Cluj Napoca, ROMANIA
[4] Neurosurgery Clinic, Clinical Emergency Hospital, Cluj Napoca, ROMANIA

Keywords
2D fluoroscopy, CT-fluoro matching, image guided surgery, screw accuracy, transpedicular screw fixation

Corresponding author:
Adrian Bălașa

University of Medicine, Pharmacy, Science and Technology "George Emil Palade", Tirgu Mures, Romania

adrian.balasa@umfst.ro

ABSTRACT

Aim: Pedicle screw fixation is an established technique in the lumbar and thoracic area. Fluoroscopy-guided screw placement and subsequently navigation have decreased the rate of misplaced screws, but no technique has wholly eliminated this risk. This paper aims to study the difference between the accuracy of the fluoroscopic guided screw placement to that of the 2D fluoroscopy- preop CT fused neuronavigation guided technique, a lesser-used navigation technique.

Material and Methods: This retrospective study reflects our results using both techniques between March 2018 and March 2019 in both degenerative or traumatic spinal pathology for thoracic and lumbar regions. The accuracy of the screw placement was measured using Mirza grading system on postoperative CT images.

Results: A total number of 56 patients underwent spinal instrumentation surgery. A total of 274 screws were placed with a mean number of 4.89 screws per patient; 199 screws were implanted using neuronavigation and 75 using the freehand-2D fluoroscopy-guided technique. The accuracy rate of pedicle screw placement in the freehand technique guided by 2D fluoroscopy was 88,00%. With the use of neuronavigation, the accuracy increased to 89,96%.

Conclusion: Pedicle screw placement accuracy is higher when guided by CT-fluoro matching neuronavigation compared to freehand fluoroscopy-guided technique and can be used in departments where there is no intraoperative O-arm or 3D fluoroscopy available.

Scan to access the online version

INTRODUCTION

In the past 30 years, spinal surgery has seen a significant increase in the

development of surgical techniques and instrumentation.

Pedicle screw fixation is an established technique in the lumbar and thoracic area (3). There are many techniques of pedicle screw placement starting from the "free hand technique" described by Kim and Lenke (5,17) to modern techniques that use intraoperative image guidance: 2D fluoroscopy, 2D/3D fluoroscopy navigation, cone beam intraoperative CT navigation or intraoperative MRI navigation (11).

Fluoroscopy-guided screw placement has decreased the rate of misplaced screws from 55% to 21% in the thoracic region and from 40% to 12% in the lumbar region (14). Neuronavigation in spinal surgery further decreased the rate of misplaced screws, but no intraoperative navigation technique has wholly eliminated this risk (4,7,13).

Neuronavigation using 2D fluoroscopy- CT fusion is a technique used for pedicle screw placement that was described by Sakai (12). This technique uses a preoperative CT thin cut slice scan that is linked via neuronavigation to a set of intraoperative fluoroscopic images and allows for navigation even in the absence of 3D fluoroscopic C-arms or intraoperative CT machines (12). Despite being introduced more than ten years ago, this technique has not seen wide adoption, and no relevant studies are available to assess its efficacy.

This paper aims to study the difference between the accuracy of the fluoroscopic guided screw placement to that of the 2D fluoroscopy-CT fused neuronavigation guided technique.

Figure 1: Fusion between the intraoperative fluoroscopy and a 3D model of a vertebra (based on the CT scan) and subsequently registration of the defined vertebra.

MATERIALS AND METHODS

This retrospective study was performed between March 2018 and March 2019 at the Department of Neurosurgery of the Tîrgu Mureș Clinical Emergency

Hospital, Romania. Patients were operated by a team of multiple neurosurgeons, all with proper levels of spinal instrumentation expertise. The study has included traumatic and degenerative cases in the thoracic and lumbar spine; redo surgeries or repositioning of misplaced screws were not included.

All patients were operated in a prone position using a Bertchtold translucent table (Stryker, Michigan, USA). Intraoperative fluoroscopy was performed in all cases with a Siemens Siremobil Compact L 200 machine (Siemens, Munich, Germany). For cases in which the neuronavigation guided technique was used, a preoperative CT thin cut slice (under 3 mm slices) scan of the operated area was obtained before surgery. A Curve BrainLab (BrainLab, Munich, Germany) neuronavigation system was used in conjunction with a Spine and Trauma software (BrainLab, Munich, Germany) (Figure 1). A fusion between the intraoperative fluoroscopy and a 3D model of a vertebra (based on the CT scan) and subsequently registration of the defined vertebra was performed using Sakai's (12) previously described technique (Figure 1).

Medtronic polyaxially titanium screws (Medtronic, Minnesota, USA) or Stryker (Stryker, Michigan, USA) monoaxial and polyaxially titanium screws with diameters ranging from 4 to 6 mm, and lengths between 40 and 60 mm were used, depending on the spinal level and pedicle width.

There are two commonly used grading systems used for measuring screw placement accuracy: Zdichavsky (18), (9) and Mirza (10) which use postoperative CT images to analyze the pedicle screw placement accuracy. We have chosen to use in our study the Mirza scoring system (Table 2).

Table 1. Demographic distribution.

No. Patients	**56**	
Sex		
Male	36 (64, 28%)	
Female	20 (35, 72%)	
Age (years)		
Mean	52,76	
Range	20-75 years	
Pathology/Level	**Traumatic**	**Degenerative conditions**
Thoracic	15 patients (26,78%)	0 patients (0%)
Thoraco-Lumbar	1 patients (1,78%)	0 patients (0%)
Lumbar	18 patients (32,14%)	17 patients (30,35%)
Lumbo-Sacral	0 patients (0%)	5 patients (8,92%)
Number of screws implanted	**CT-fluoro-matching neuronavigation**	**2D fluoroscopy Freehand**
THORACAL	46 (23.11%)	32 (42.66%)
LUMBAR	153 (76.88%)	43 (57.33%)

Table 2. Mirza et al. 2 mm increment grading system.

Classification	**Borders**
Grade 0 (optimal)	the screw correctly fits the pedicle
Grade 1 (minor)	under 2 mm of displacement
Grade 2 (moderate)	between 2 to 4 mm of displacement
Grade 3 (severe)	over 4 mm of displacement

Postoperative imaging was analyzed by the senior author (AB) and an independent radiologist. Statistical analyses included descriptive (frequency, mean, standard deviation) and inferential statistics. The Shapiro-Wilk test was applied to determine the distribution of the analyzed data series. For analyzing the quantitative variables the t-Student test was applied for unpaired data and for analyzing the qualitative variables the Fisher test was applied. The significance threshold chosen for the p value was 0.05. Statistical analysis was performed using the GraphPad Prism trial variant.

RESULTS

Between March 2018 and March 2019, a total number of 69 patients underwent spinal instrumentation surgery in the thoracolumbar regions for degenerative or traumatic pathology. Out of these, 56 patients (81.16%) had complete documentation and were included in our study. Second surgery for repositioning of screws was necessary in 2 screws (2 cases).

A total of 274 screws were placed with a mean number of 4.89 screws per patient, ranging from 4 screws to a maximum of 10 screws. In our study, traumatic pathology was represented by 60.71% (n=34) of cases, and the lumbar spine was the most frequent region involved with 39.29% of cases (n=35). Table 1 summarizes the clinical data and demographic distribution of the patients.

The CT fluoro matching neuronavigation method was used in 72.63% of cases, which resulted in a total

of 199 screws implanted; the rest of 75 (27.37%) screws were implanted using the" freehand" technique under fluoroscopy guidance. The distribution of screws by region and the type of technique used is seen in Table 1.

The results of the Mirza 2 mm increment staging system are presented in Table 3 for medial and lateral displacement and Table 4 for the correlation between the severity of displacement and anatomic region.

From 20 mispositioned screws implanted under neuronavigation guidance, 11 screws were in the lumbar area and 9 screws in the thoracic area. The misplaced screws in the thoracic area represented 19.57% of the total number of 46 screws implanted, whilst in the lumbar area, the misplaced screws represented 7.19% of the total of 153 screws implanted. All severly placed screws were in the thoracic area (Table 4). There is a statistically significant association between the region of screw implantation and the malposition rate (p = 0.023, Fischer's test).

Table 3. Grade of screw misplacement using the 2 mm increment (Mirza score) classification and screws direction tendency of cortex perforation.

Severity	Minor (%)	Moderate (%)	Severe (%)	Total (%)	P
Lateral misplacement with Neuronavigation	6 (3.01%)	4 (2,01%)	1 (0,50%)	11(5,52%)	0.55
Lateral misplacement with Fluoroscopy	1 (1,33%)	1 (1,33%)	1 (1,33%)	3 (4%)	
Medial misplacement with Neuronavigation	8 (4.02%)	1 (0,50%)	0 (0%)	9 (4,52%)	0.99
Medial misplacement with Fluoroscopy	5 (6,66%)	1 (1,33%)	0 (0%)	6 (8%)	

Table 4. Grade of screw misplacement using the 2mm increment (Mirza score) classification relative to the vertebral region.

2D Fluoroscopy Freehand	Number of screws (%)	Misplacement rate (%)			p value
		Minor	Moderate	Severe	
Thoracal	32 (42.66)	4 (8.69%)	4 (8.69%)	1 (2.17%)	0.1585
Lumbar	43 (57.33)	10 (6.53%)	1 (0.65%)	0 (0.00%)	
Total	75	20			
CT- fluoro- Matching Neurnavigation					
Thoracal	46 (23.11%)	4 (12.50%)	1 (3.12%)	1 (3.12%)	0.0231
Lumbar	153 (76.88%)	2 (4.65%)	1 (2.32%)	0 (0.00%)	
Total	199	9			

Discussions

This study tries to reflect our experience in transpedicular screw implantation. We focused on the accuracy of transpedicular screw implantation using the two implantation procedures that are used at our institution: freehand technique under fluoroscopy guidance and neuronavigation with CT-2D fluoro-matching.

There are few articles in the literature about pedicle screw implantation that include CT-fluoro-matching neuronavigation or compare this technique with the freehand technique under fluoroscopic guidance (15), and to our knowledge this is the first series presented. This might be because intraoperative 3D fluoroscopy or intraoperative CT has become widely available.

Scoring the screw misplacement is still a difficult task as there are reported more than 35 classifications that analyze pedicle screw misplacement and, in most of them, there is no clear description of the assessment methods used to determine the accuracy of the pedicle screw positioning (2,6). Evermore there are publications showing that moderate lateral or medial displacement of the screws with violation of the pedicle cortex does not commonly relate with neurologic, vascular, and/or visceral complications (8).

We have chosen Mirza scoring system(10) because this seems to be the most widely accepted and one of the most precise scale for scoring pedicle screw placement. (1)

Our overall accuracy rate of pedicle screw placement using the freehand technique guided by 2D fluoroscopy was 88.00%. With the use of the neuronavigation, the accuracy increased to 89.96%. Nevertheless, our results are comparable to different other papers:

- The screw misplacement rate in our study was of about 12% in the fluoroscopy technique group, and pedicle cortex perforation over 4 mm (severely misplaced screw) was seen in 1.33%, comparable to the results previously published by Guedes and Verma (2),(16).
- In our CT-fluoro navigation group, a slight increase in the overall accuracy was noted. Even if this increase in accuracy is not statistically significant, these results are similar to the ones of Kosmopoulous (6), showing over 90% accuracy rate for both techniques, and might be partially explained by good fluoro screw positioning technique.
- Gelalis (3) concluded that neuronavigation increases the accuracy of pedicle screw placement and when using freehand technique there is an increased tendency of medial perforation of pedicle cortex as opposed to neuronavigation where the tendency is to perforate the cortex laterally which decreases the risk of neurological complication even in case of inaccuracy. Our results seem to reconfirm this as in the CT-fluoro group we noted a slight increase in the lateral displacement (5,52% vs. 4%) but also a 50% reduction of medial misplacement (4.52% vs. 8%) (Table 3.)

There are also inherent limitations to CT-fluoro matching neuronavigation technique: due to the acquisition of the preoperative spine CT in the supine position and the prone position in the operating room, a spine displacement most likely occurs and interferes with the accuracy(8). Scanning patients in prone position or spine curvature detection algorithms might further improve this technique. Inaccuracies are also given by the need to fuse a 3D vertebral body model to a 2D intraoperative fluoroscopy, and a less than perfect thoracic imaging allows for navigation errors and screw misplacement that is higher than the rate we obtained in lumbar spine, but still lower than freehand fluoroscopy technique.

Conclusion

Despite its shortcomings, the CT-fluoro matching technique has similar or slightly better results than freehand fluoroscopy and can be used in departments where there is no intraoperative O-arm or 3D fluoroscopy available and a more affordable neuronavigation solution is required.

Conflicts of interest

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

Abbreviations

MRI: magnetic resonance imaging
CT: computer tomography
2D: two-dimensional
3D: three-dimensional

References

1. Aoude AA, Fortin M, Figueiredo R, Jarzem P, Ouellet J, Weber MH: Methods to determine pedicle screw placement accuracy in spine surgery: a systematic review. Eur. Spine J. 24: 990–1004, 2015.
2. De Paula Guedes V, Manffra EF, Aguiar LR: Image-guided surgery in the spine: Neuronavigation vs. fluoroscopy. Coluna/ Columna 14: 181–185, 2015.
3. Gelalis ID, Paschos NK, Pakos EE, Politis AN, Arnaoutoglou CM, Karageorgos AC, Ploumis A, Xenakis TA: Accuracy of pedicle screw placement: A systematic review of prospective in vivo studies comparing free hand,fluoroscopy guidance and navigation techniques. Eur. Spine J. 21: 247–255, 2012.
4. Karhade A, Vasudeva, Pompeu, Lu: Image Guided Spine Surgery: Available Technology and Future Potential. Austin Neurosurg Open Access 3: 1043–1, 2016.
5. Kim YJ, Lenke LG, Bridwell KH, Cho YS, Riew KD: Free Hand Pedicle Screw Placement in the Thoracic Spine: Is it Safe. Spine (Phila. Pa. 1976). 29: 333–342, 2004.
6. Kosmopoulos V, Schizas C: Pedicle Screw Placement Accuracy. Spine (Phila. Pa. 1976). 32: E111–E120, 2007.
7. Laine T, Mäkitalo K, Schlenzka D, Tallroth K, Poussa M, Alho A: Accuracy of pedicle screw insertion: A prospective CT study in 30 low back patients. Eur. Spine J. 6: 402–405, 1997.
8. Liu H, Chen W, Liu T, Meng B, Yang H: Accuracy of pedicle screw placement based on preoperative computed tomography versus intraoperative data set acquisition for spinal navigation system. J. Orthop. Surg. 25: 1–8, 2017.
9. Marios Theologou1 2, , Theologos Theologou1 , Dimitrios

Zevgaridis1 , Nikolaos Skoulios2 , Slavisa Matejic3 CT: Pedicle screw placement accuracy impact and comparison between grading systems Marios. Surg. Neurol. Int. 8: 1–7, 2017.

10. Mirza SK, Wiggins GC, Kuntz IV C, York JE, Bellabarba C, Knonodi MA, Chapman JR, Shaffrey CI: Accuracy of thoracic vertebral body screw placement using standard fluoroscopy, fluoroscopic image guidance, and computed tomographic image guidance: A cadaver study. Spine (Phila. Pa. 1976). 28: 402–413, 2003.
11. Moses ZB, Mayer RR, Strickland BA, Kretzer RM, Wolinsky J-P, Gokaslan ZL, Baaj AA: Neuronavigation in minimally invasive spine surgery. Neurosurg. Focus 35: E12, 2013.
12. Sakai Y, Matsuyama Y, Yoshihara H, Nakamura H, Nakashima S, Ishiguro N: Simultaneous registration with ct-fluoro matching for spinal navigation surgery. A case report. Nagoya J Med Sci 68: 45–52, 2006.
13. Shin MH, Hur JW, Ryu KS, Park CK: Prospective Comparison Study between the Fluoroscopy-guided and Navigation Coupled with O-arm - Guided Pedicle Screw Placement in the Thoracic and Lumbosacral Spines. J. Spinal Disord. Tech. 28: E347–E351, 2015.
14. Tehli O, Harman F, Temiz C, Kacar Y, Kazanci B, Daneyemez M, Solmaz I, Kural C: The Use of Neuronavigation and Intraoperative Imaging in Spinal Stabilization Surgery. Turk. Neurosurg. 26: 771–776, 2016.
15. Tjardes T, Shafizadeh S, Rixen D, Paffrath T, Bouillon B, Steinhausen ES, Baethis H: Image-guided spine surgery: state of the art and future directions. Eur. Spine J. 19: 25, 2010.
16. Verma SK, Singh PK, Agrawal D, Sinha S, Gupta D, Satyarthee GD, Sharma BS: O-arm with navigation versus C-arm: a review of screw placement over 3 years at a major trauma center. Br. J. Neurosurg. 30: 658–661, 2016.
17. Wagner SC, Morrissey PB, Kaye ID, Sebastian A, Butler JS, Kepler CK: Intraoperative pedicle screw navigation does not significantly affect complication rates after spine surgery. J. Clin. Neurosci. 47: 198–201, 2018.
18. Zdichavsky M, Blauth M, Knop C, Graessner M, Herrmann H, Krettek C, Bastian L: Accuracy of pedicle screw placement in thoracic spine fractures - Part I: Inter- and intraobserver reliability of the scoring system. Eur. J. Trauma 30: 234–240, 2004.

Romanian Neurosurgery (2020) XXXIV (1): pp. 26-30
DOI: 10.33962/roneuro-2020-004
www.journals.lapub.co.uk/index.php/roneurosurgery

A rare case of pituitary macroadenoma with synchronous suprasellar meningioma. Case report, surgical strategy and review of literature

Bogdan Ionut David[1,3], Ligia Gabriela Tataranu[2,3], Vasile Gheorghe Ciubotaru[3], Aurelia Mihaela Sandu[3], Radu Mircea Gorgan[2,3]

[1] PhD Student, University of Medicine and Pharmacy "Carol Davila", Bucharest, ROMANIA
[2] University of Medicine and Pharmacy "Carol Davila", Bucharest, ROMANIA
[3] Department of Neurusurgery, Emergency Clinical Hospital Bagdasar-Arseni, Bucharest, ROMANIA

Keywords
pituitary macroadenoma,
synchronous,
staged surgery,
suprasellar meningioma

Corresponding author:
Bogdan Ionut David

University of Medicine and Pharmacy "Carol Davila", Bucharest, Romania

david.bogdan.med@gmail.com

Scan to access the online version

ABSTRACT

Synchronous tumours can be found all along the entire neuraxis, however, some lesions are far less likely to coexist. One of these extremely rare associations is between GH-pituitary adenomas and suprasellar meningiomas. A wide spectrum of transcranial and transsphenoidal approaches were described in the literature for either sellar, suprasellar and parasellar lesions, but no agreement has been reached for the cases of simultaneous occurring lesions.

We present a rare case of a woman with GH-secreting pituitary adenoma and concomitant suprasellar meningioma. The strategy chosen was sequential transsphenoidal surgeries. However, after the first surgery, the remaining tumour mass did not mobilize as expected due to gravity, hence we decided to perform a transcranial subfrontal unilateral approach. Surprisingly, the second surgery revealed a different histopathological result.

Association of a GH-pituitary adenoma and suprasellar meningioma is very rare, only 17 cases being reported in the relevant literature so far. Different authors prefer different strategies, ranging from only transsphenoidal to simultaneous transsphenoidal and transcranial approaches, but no general consensus was established.

In conclusion, the existence of synchronous tumours of the sellar region should be taken into account when imaging studies reveal an intracranial mass developing both sellar and suprasellar. The surgical strategy should be tailored to every specific patient and experience of the neurosurgeon.

INTRODUCTION

Synchronous tumors are found along the entire neuraxis, however some lesions are far less likely to coexist (1). One of these unlikely associations is between pituitary adenomas and suprasellar menin-

giomas. Few cases have been reported in literature, most of the pituitary tumors being GH secreting adenomas. (1) However, a syndrome has yet to be defined consisting of this association, and, with the exception of the previously radio-treated pituitary adenomas that develop subsequent meningiomas, no pathophysiological connection exists.

Even though each type of tumor has distinct radiological findings, there are cases in which they can easily be misdiagnosed or even confused for one single lesion. This rare occurrence must be taken into account by neurosurgeons planning tumor resection in such cases. A wide spectrum of transcranial and transsphenoidal approaches have been described in literature for either sellar, suprasellar and parasellar lesions, but no agreement has been reached for the cases of simultaneous occurring lesions. (2)

The purpose of this article is to present a rare case of a simultaneous GH secreting pituitary adenoma and a supradiafragmatic meningioma, the strategy behind the surgical approach and review of literature.

CASE PRESENTATION

A 57 years old woman, with history of stage II hypertension, type II diabetes mellitus, NYHA II heart failure was admitted with intense headache, visual field disturbances and hormonal disturbances (elevated GH levels: IGF-1 = 98 ng/ml, *N = 10 ng/ml*). The cerebral CT scan showed a contrast enhancing sellar mass which extended suprasellar, slightly eccentric to the left. (Figure 1)

The presumed diagnosis was GH-secreting pituitary macroadenomma with suprasellar extension. We decided to perform surgery via transsfenoidal approach. Due to large size of the tumor and high risk of rupturing the sellar diaphragm, only partial resection was performed and a second transsfenoidal surgery was scheduled for the upcoming three months. The rationale was that the suprasellar part of a tumor originating in the sellar region will gravitationally fall in the sella over the next three months, thus enabling a second minimally invasive surgery.

The first histopathological exam was acidophilic pituitary adenoma with capsular invasion. (Figure 2) Initial postoperative outcome was uneventful, without any additional neurological deficits.

After three months, the cerebral CT scan showed a surprising stationary suprasellar tumor. (Figure 3) We changed our initial strategy, and we decided to operate the tumor via the subfrontal approach on the nondominant side (right). Intraoperative we found a suprasellar meningioma. We achieved tumor complete resection, with no intraoperative incidents.

The second histopathological exam revealed meningothelial meningioma with transitional components. (Figure 4) After removal of the suprasellar tumor, visual field impairment improved. The patient required substitute hormonal therapy for transient diabetes insipidus for the first three months, which was gradually lowered in dosage. At three months postop, CT showed complete tumor removal and the patient was hormonally stable. Even if standard post-surgical strategy is to administer oral corticoids, these were interrupted after only one month, and no further hormonal therapy was required.

The particularity of the case was simultaneous development of a GH-secreting pituitary macroadenoma and suprasellar meningioma, a rare association of two types of histological tumors, with no apparent link between them.

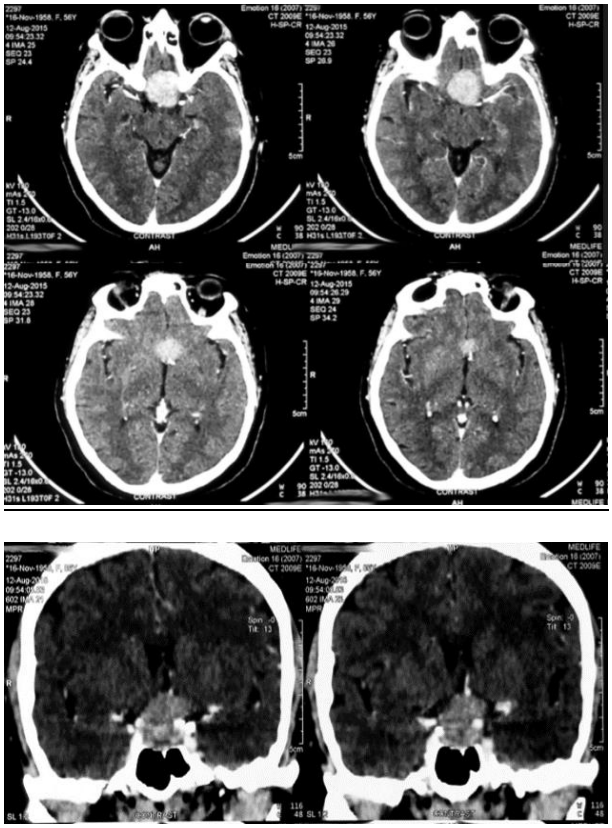


Figure 1. Cerebral CT scan showing a sellar and suprasellar contrast enhanced tumor.

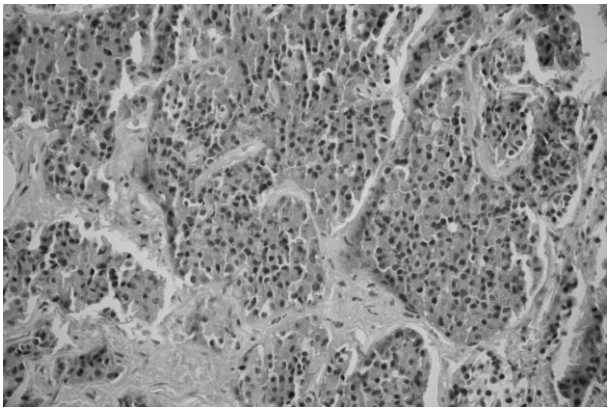


Figure 2. Acidophilic pituitary adenoma; HE; 40x.

Figure 3. Cerebral CT scan, performed 3 months after the first surgery, showing suprasellar contrast enhanced tumor with no sellar tumor.

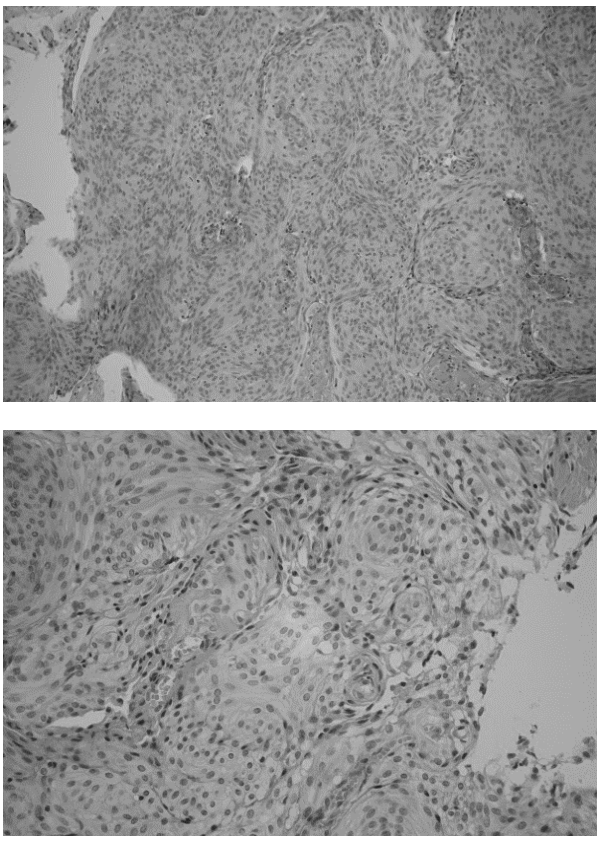


Figure 4. Meningothelial meningioma with transitional components; HE; a. 20x; b. 40x.

Review of literature

MESH terminology consists of two different situations which describe simultaneous development of brain tumors. Thus, one can encounter either collision tumors, in which case one tumor infiltrates the other one, or coincidental tumors, tumors being synchronous, but with different histogenesis, being situated one next to the other or at a distance. (3)

We performed a search in PubMed and Google Scholar for simultaneous occurring brain tumors out of which one would be a pituitary adenoma and we found 43 publications and a total of 63 patients. (1) Furthermore, a search of similar cases to ours found that the rare situation of simultaneous development of a pituitary adenoma and suprasellar or sphenoid wing meningioma has been reported in 32 cases. (4) Moreover, only 17 cases of strictly suprasellar meningiomas were described in the literature, our case being the 18th. (1)

Discussions

Tumors of the sellar region have mesenchymal, neural or epithelial origin, along with cystic and inflammatory processes. Out of all these tumors, pituitary adenomas are the most common, accounting for about 10-15% of all intracranial tumors. (5) Most of the patients treated in our clinic present with both endocrine symptoms, but also intracranial hypertension and optic chiasm symptoms due to large size of the tumor. Thus, most of the patients suffer from macroadenomas (> 10 mm) at the time of diagnosis, and most of the tumors extending beyond the sella turcica, either parasellar or suprasellar.

The existence of simultaneous pituitary adenoma and supradiafragmatic meningioma is a very rare situation. Search of relevant literature performed in PubMed and Google Scholar revealed only 17 other cases, ours being the 18th. (1)

Even though a plethora of possible mechanisms have been forwarded in relation to multiple brain tumors occurring simultaneously and having different histology origins, none have been widely recognized by the neurosurgery community.

While some authors suggest a common receptor activator that triggers both lesions, others believe that one of the tumors might secrete a growth factor responsible for the initiation of aberrant cell growth in the second tumor. Although growth hormone

secreting adenomas have been suggested to induce arachnoid cell transformation, this hypothesis has yet to be confirmed. (6)

Other authors consider favorable factors for synchronously developing adenoma and meningioma variables such as genetic factors, prior trauma or surgery and even exposure to offending biochemical substances. (7)

It is essential to distinguish between cases of pituitary adenoma with a suprasellar component, and cases of a co-existing sellar adenoma and suprasellar meningioma, because different surgical strategies might be adequate for these two different situations. (2) This is particularly difficult, because in most of the times the two lesions are indistinguishable from a radio-imaging point of view, our case did not show any pathognomonic imaging characteristic.

Most authors prefer to perform both tumor resections in either one surgical procedure, or combined transsphenoidal and transcranial approach under the same single general anesthesia. However, sometimes this might not be possible, and a decision must be made on which lesion to perform resection on first. Surgical strategy is tailored according to the symptomatic lesion. So, in such cases it must be seen to whom the symptoms are due. Both tumors were located in the same area, sellar and suprasellar region and both can cause slightly similar neurological complains. Headache is a nonspecific symptom, visual fields deficits are common findings in both pituitary macroadenomas and suprasellar meningiomas, but hormonal imbalances are specific for a pituitary adenoma. So, in cases when lesions cannot be resected using the same approach, the surgeon must first operate the symptomatic tumor.

Regarding the existence of an agreement as to the best surgical approach to be used, we could only find one: use the best approach tailored to the specific case of the patient. Therefore, some authors prefer to perform a transsphenoidal approach for both the lesions (3), others use a combined approach under the same anesthesia, others prefer a two timed transsphenoidal (4), transcranial, and so on.

Two-stage surgery is not an uncommon practice for large pituitary adenomas. In our case, both sellar and suprasellar components were considered to be part of the same pituitary macroadenoma. The treatment of choice for pituitary adenomas is tumor resection via transphenoidal route. During transsphenoidal surgery the sellar part of the tumor is resected, and the soft suprasellar component descends, with the aid of gravity and CSF pulsation and develop itself to the surgeon. When the tumor is more firm this cannot happen during surgery. Vigorous attempts to pull the suprasellar tumor into the sella can be dangerous, due to possibility of adherence to neurovascular structures. More sellar diaphragm can be teared, leading to postoperative CSF fistula. In such cases a two-stage transsphenoidal surgery can be performed, because after a period of time of a few months the suprasellar will descend and through planned elective surgery the tumor can be complety resected via same surgical approach with minimal risks. Even more, first histopathological examination confirmed the nature of the tumor as being GH-secreting pituitary adenoma. That was the rationale for our decision not to extend the resection to the suprasellar component, but rather consider a second transsphenoidal approach 3 months later, in order to allow the rest of the tumor to gravitationally descend in the sella. However, the CT scan at 3 months showed a stationary suprasellar component. The decision was made to perform subfrontal approach in order remove this component. This second lesion turned out to be a meningioma.

This case perfectly illustrates the need for approaching every sellar tumor through the path that offers the most advantages. Thus, a transsfenoidal approach would not have allowed the surgeon to resect the suprasellar component without major risks, whilst a subfrontal approach would not have been possible to resect the entire sellar component.

Overall, the patient had favorable outcome, because he harbored two benign, slow-growing tumors.

Conclusions

Synchronous GH-secreting macroadenoma and suprasellar meningioma developing simultaneously in the sellar and suprasellar region are very rare findings. In selected cases, both minimally invasive transsfenoidal approach and transcranial microsurgical approach must be considered simultaneously in order to achieve complete resection. Moreover, the existence of two different lesions developing in the same region should be

considered in all the cases of pituitary adenomas with a suprasellar component. Further research is needed with genetic profile of patients with synchronous tumors.

Conflicts of interest

The authors declare no conflict of interests.

Founding

None

References

1. Amirjamshidi A, Mortazavi SA, Shirani M, Saeedinia S, HAnif H. Coexisting pituitary adenoma and suprasellar meningioma - a coincidence or causation effect report of two cases and review of the literature. J Surg Case Rep, 2017 (5):rjx039. doi: 10.1093/jscr/rjx039.
2. Lim KZ, Goldschlager T, Chandra RV, Hall J, Uren B, Pullar M. Co-occurrence of pituitary adenoma with suprasellar and olfactory groove meningiomas . Basic Clin Neurosci, 2016. 7:361-365.
3. Prevedello DM, Thomas A, Gardner P, Snyderman CH, Carrau RL, Kassam AB. Endoscopic endonasal resection of a synchronous pituitary adenoma and a tuberculum sellae meningioma: technical case report. Neurosurgery, 2007. 60(4 Suppl 2):E401.
4. Guaraldi F, Prencipe N, di Giacomo V, Scanarini M, Gasco V. Association of craniopharyngioma and pituitary adenoma. Endocrine, 2013. 44:59-65.
5. Cannav S, Curt L, Fazio R, Paterniti S, Blandino A, Marafioti T, Trimarchi F. Coexistence of growth hormone-secreting pituitary adenoma and intracranial meningioma: A case report and review of the literature. J Endocrinol, 1993. 16:703-8.
6. Mathuriya SN, Vasishta RK, Dash RJ, Kak VK. Pituitary adenoma and parasagittal meningioma: an unusual association. Neurol India, 2000. 48:72-4.
7. Goldberg MB, Sheline GE, Malamud N. Malignant intracranial neoplasms, following radiation therapy for acromegaly. Radiology, 1963. 80:465-470.

Romanian Neurosurgery (2020) XXXIV (1): pp. 31-40
DOI: 10.33962/roneuro-2020-005
www.journals.lapub.co.uk/index.php/roneurosurgery

Microsurgical clipping of paraclinoid aneurysms. A single centre experience

Mugurel Radoi[1,2], Ram Vakilnejad[2], Suditu Stefan[2], Florin Stefanescu[2]

[1] Faculty of General Medicine, "Carol Davila" University of Medicine and Pharmacy, Bucharest, ROMANIA
[2] Neurosurgical Department of the National Institute of Neurology and Neurovascular Diseases, Bucharest, ROMANIA

ABSTRACT

Background. Paraclinoid aneurysm is a nonspecific term that includes ophthalmic segment aneurysms and distal cavernous internal carotid artery (ICA) aneurysms. The literature mostly described the frequency to be in the range of 1.3-5%. and a high incidence of being multiple or having a large size.

Methods. A retrospective review of 18 consecutive patients surgically treated for paraclinoid aneurysm was performed. The data of all our consecutive patients were searched to obtain patient and aneurysm characteristics, treatment details, complications and follow up. Clinical outcome was graded according to the modified Rankin scale. The follow-up period varied widely from 3 to 62 months (mean 26 months).

Results. Surgical clipping was performed for 15 ruptured paraclinoid aneurysms; only in 3 cases the aneurysm was unruptured. Post-operative control angiography was performed in 10 patients (55.56%), from which we reported a full occlusion of the aneurysm in 9 patients (90%). Best results were obtained in patients who preoperatively were included in 1st and 2nd grade of Hunt & Hess scale. Two months postoperative follow-up was complete for all but one patient who died 12 days after surgery, from cerebral ischemia resulting from severe cerebral vasospasm. There were excellent and good results (mRS 0–2) in 88% of the cases (15 out of 17 patients) at two months follow-up, and 94% (16 out of 17 patients) at six months follow-up. Three patients with slight disabilities, ranked mRS 1-2 at two months follow-up, improved to mRS 0, with no symptoms at all, at 6 months postoperative control. All 3 patients with a surgically treated non-ruptured paraclinoid aneurysm had an excellent outcome (mRS 0).

Conclusions. Most appropriate treatment is to occlude aneurysms without compromising patency of the parent artery. Anterior clinoidectomy and microsurgical clipping can still be a standard treatment despite of recent development of endovascular coiling procedures.

Keywords
paraclinoid aneurysm, surgical clipping, endovascular treatment, postoperative results

Corresponding author:
Mugurel Radoi

University of Medicine and Pharmacy "Carol Davila", Bucharest, Romania

muguradoi@yahoo.com

Scan to access the online version

INTRODUCTION

Paraclinoid aneurysms is a nonspecific term that includes ophthalmic segment aneurysms and distal cavernous internal carotid artery (ICA)

aneurysms. These aneurysms arise near the anterior clinoid process and represent a considerable surgical challenge owing to their anatomic features, their proximity to the optic nerves and chiasm, and their relationship to complicate dural structures.

Paraclinoid aneurysms are classified according to the ICA segments from which they arise. There are clinoidal segment aneurysms and ophthalmic segment aneurysms. Each variant can be differentiated according to the site of origin, the direction of projection, and relationship with arterial branches, cranial nerves and adjacent dural and osseous structures within the segment (4).

Clinoidal segment aneurysms have two variants: (a) anterolateral variant and (b) medial variant. Ophthalmic segment aneurysms are intradural and include: (a) ophthalmic artery aneurysms, (b) superior hypophyseal artery aneurysms and (c) rare variants of dorsal ICA aneurysms (21).

This article summarizes the results of microsurgical treatment of 18 paraclinoid aneurysms operated in our department, by two senior neurosurgeons, and discusses treatment strategy and potential complication, comparing them with the results reported in the neurosurgical literature.

Methods

This article presents the results of a retrospective study of paraclinoid aneurysms, surgically treated in the Department of Neurosurgery of Institute of Neurology and Neurovascular Diseases, Bucharest. In the period from January 2014 until June 2019, 18 consecutive patients with paraclinoid aneurysms, from a total of 296 patients with carotid system aneurysms, were operated on by two senior neurosurgeons. Admission data, operative reports and imaging studies were reviewed to obtain information on patient's age, gender, aneurysm size and orientation, treatment details, complication and follow-up. From this series of 18 patients, two harboured another middle cerebral aneurysm, both on ipsilateral side.

Most of the patients (15 patients - 83.34%) presented with acute subarachnoid haemorrhage and only 3 presented an unruptured paraclinoid aneurysm. However, in a patient with multiple aneurysms, the source of the haemorrhage was clearly defined to be a middle cerebral artery aneurysm. Digital subtraction angiography with 3D reconstruction or computerized tomography (CT) angiography were used to image the intracranial circulation and to achieve proper orientation and visualization of the paraclinoid aneurysm. At admission, the clinical condition of all patients was classified according to the Hunt and Hess scale. Clinical outcome was assessed according the modified Rankin scale.

An ipsilateral pterional approach was used in all patients and medial sphenoid wing was resected extradurally to the level of the lateral clinoid process. Proximal control was achieved by neck dissection and temporary clipping of either internal carotid artery (ICA) or common carotid artery (CCA). After a standard dural opening, the anterior limb of the Sylvian fissure is split to gain access to the ICA, anterior clinoid process, and optic nerve. The arachnoid layers were cut form distal to proximal, identifying the distal ICA and posterior communicating artery. We continued the microdissection proximally until the neck of the aneurysm on the ICA was identified. The optic nerve partially blocked the origin of most ophthalmic artery aneurysms, and it was untethered and gently mobilized by sectioning the falciform ligament. Visualization of the entire aneurysm neck during clip application was often impossible, so clip deployment proceeded with visualization of one blade and inspection of the other only after the clip was applied.

Distribution of carotid system aneurysms	
Aneurysm location	**Number of patients**
Anterior communicating artery aneurysm	126 (42.5%)
Posterior communicating artery aneurysm	55 (18.5%)
Medial cerebral artery aneurysms	86 (29,33%)
Anterior cerebral artery aneurysm	2 (0.67%)
Anterior choroidal artery aneurysm	3 (1%)
Pericallosal artery aneurysm	6 (2%)
Paraclinoid aneurysms	18 (6%)
Total	**296 (100%)**

Table 1. Distribution of carotid system aneurysms in a series of 296 patients.

Characteristic	No of patients (%)
Female	12 (66.67%)
Male	6 (33.33%)
Hunt &Hess scale	
Grade 0	3 (16.67%)
Grade 1 and 2	10 (55.55%)
Grade 3	4 (22.33%)
Grade 4	1 (5.45%)
Grade 5	0
Modified Fischer Grading Scale	
No SAH present	3 (16.67%)
Focal or diffuse thin SAH	12 (66.67%)
Focal or diffuse thick SAH	2 (11.21%)
Intraventricular haemorrhage	1 (5.45%)

Table 2. Characteristics of patient with paraclinoid aneurysms in our series.

Charactersitic	No of patients (%)
Left paraclinoid aneurysms	10 (55.55%)
Right paraclinoid aneurysms	8 (44.45%)
Location	
Ophthalmic artery aneurysms	16 (94%)
Superior hypophyseal artery aneurysms	1 (4.3%)
Clinoidal segment aneurysms (medial variant)	1 (1.7%)
Size	
≤ 5 mm	3 (16.67%)
5-10 mm	10 (55.55%)
≥ 10 mm	5 (27.78%)

Table 3. Aneurysm characteristics.

RESULTS

Eighteen patients with paraclinoid aneurysms were operated on in our department between January 2014 and June 2019. Of these, two were asymptomatic, two presented with visual symptoms of decreased visual acuity of the relevant eye, and there were 14 ruptured aneurysms without eye involvement. The patient characteristics at admission are presented in Table 2.

Most of the patients were women (66.67%). At the time of surgery, 13 patients were with no subarachnoid haemorrhage or in grade 1 or 2 on Hunt&Hess scale. The aneurysms characteristics are summarized in Table 3. Paraclinoid aneurysms had usually larger diameter than aneurysms in other location. In our series, there were 5 (27.78%) aneurysms with a maximum size more than 10 mm. The patients with ruptured aneurysms, except one, who was in poor neurological status, were operated on in the first 5 days following the symptoms onset.

In only one case we delayed the surgical treatment of the aneurysm, due to severe vasospasm, and important secondary neurological deterioration of the patient. Two patients harboured a coincidental non-ruptured middle cerebral artery aneurysm, which was clipped in the same surgical procedure. Post-operative control angiography was performed in 10 patients (55.56%), from which we reported a full occlusion of the aneurysm in 9 patients (90%).

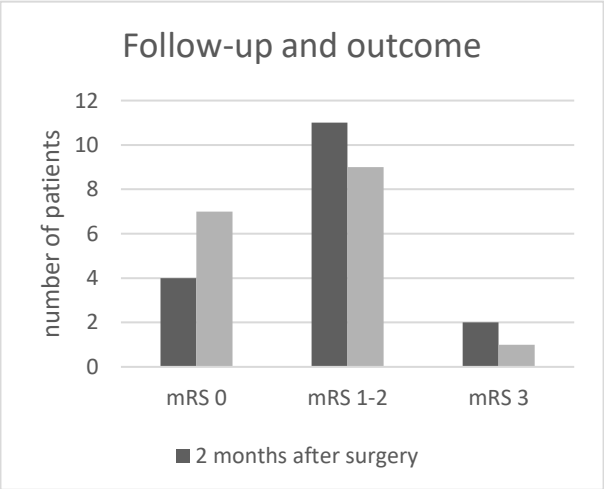
Preoperative vasospasm was demonstrated on cerebral angiography in 3 patients (16.67%) and clinically was manifested in 1 patient (5.56%). On control angiography, cerebral vasospasm was detected on near half of the patients (40%), but, as postoperative event, 2 patients, including the one with preoperative neurological signs, presented clinical symptoms of vasospasm, with decreased level of consciousness, affected speech and motor deficits.

The follow-up period varied widely from 3 to 62 months (mean 26 months). Best results were obtained in patients who preoperatively were included in 1st and 2nd grade of Hunt & Hess scale. Two months postoperative follow-up was complete for all but one patient who died 12 days after surgery, from cerebral ischemia resulting from severe cerebral vasospasm. The mean follow-up was 26 months, obtained in 89% of the patients. The outcome was evaluated using modified Rankin Scale. Overall outcome was assessed at first (two months after surgery) and respectively, second (six months after surgery) postoperative controls. There were excellent and good results (mRS 0–2) in 88% of the cases (15 out of 17 patients) at two months follow-up, and 94% (16 out of 17 patients) at six months follow-up. The most important improvement was recorded for patients graded mRS 1-2 at the first postoperative follow-up. Three patients with slight disabilities, ranked mRS 1-2 at two months follow-up, improved to mRS 0, with no symptoms at all, at 6 months postoperative control. All 3 patients with a surgically treated non-ruptured paraclinoid aneurysm had an excellent outcome (mRS 0).

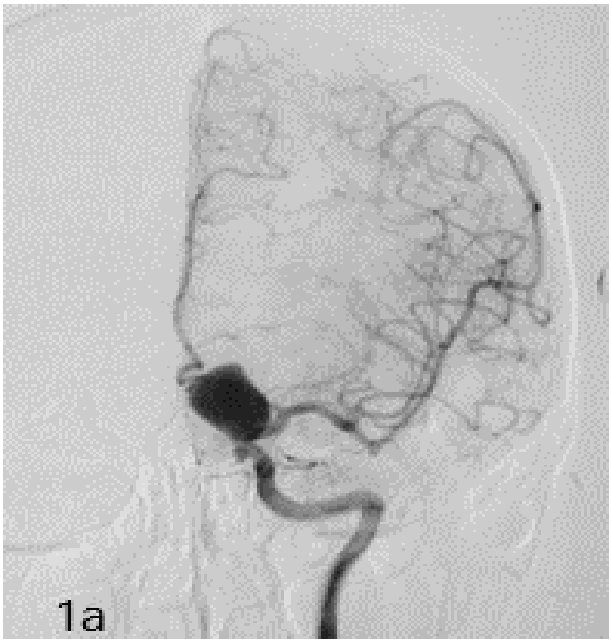
Complications	No of cases
Intraoperative rupture	2 (11.12%)
Re-ruptured before surgery (waiting)	0

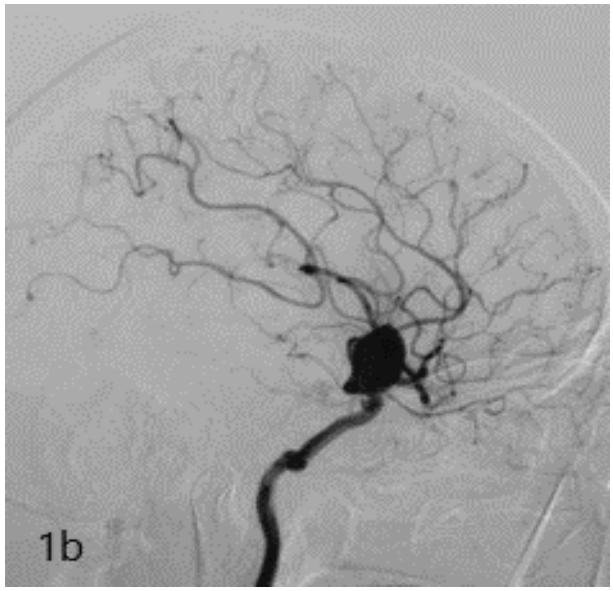
Post-operative rupture		0
Pre-operative vasospasm	angiography	3 (16.67%)
	clinic	1 (5.56%)
Post-operative vasospasm	angiography	**4/10 (40%)**
	clinic	2 (11.12%)
Post-operative subdural hematoma		0
Meningitis		0
Hydrocephalus		1 (5.56%)
VP shunt (within 30 days from surgery)		1 (5,56%)
Complications		**No of cases**
Intraoperative rupture		2 (11.12%)
Re-ruptured before surgery (waiting)		0
Post-operative rupture		0
Pre-operative vasospasm	angiography	3 (16.67%)
	clinic	1 (5.56%)
Post-operative vasospasm	angiography	**4/10 (40%)**
	clinic	2 (11.12%)
Post-operative subdural hematoma		0
Meningitis		0
Hydrocephalus		1 (5.56%)
VP shunt (within 30 days from surgery)		1 (5,56%)

Table 4. Procedural and perioperative complications.



Outcome of the surgically treated paraclinoid aneurysms after 2, and respectively, 6 months follow-up (17 patients; one died at 12 days after surgery). Excellent= mRankin 0, good=mRankin 1–2, fair= mRankin 3.





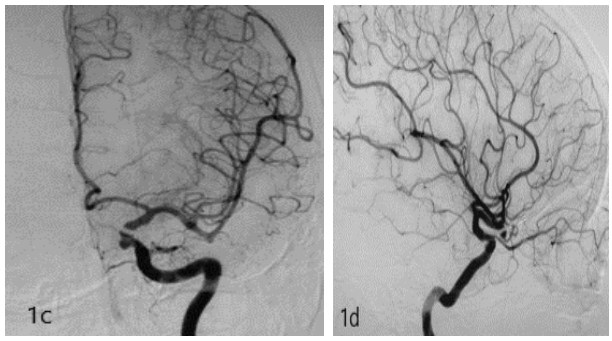
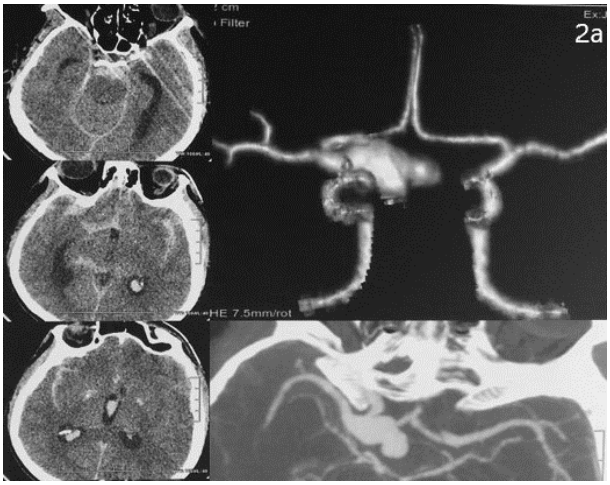
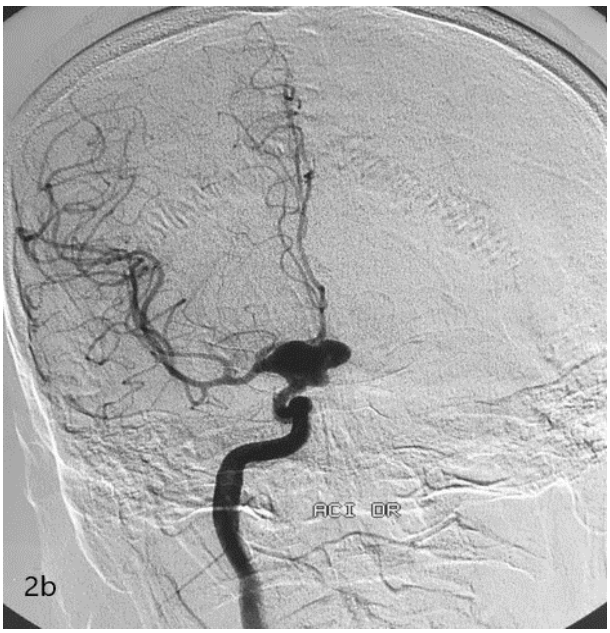
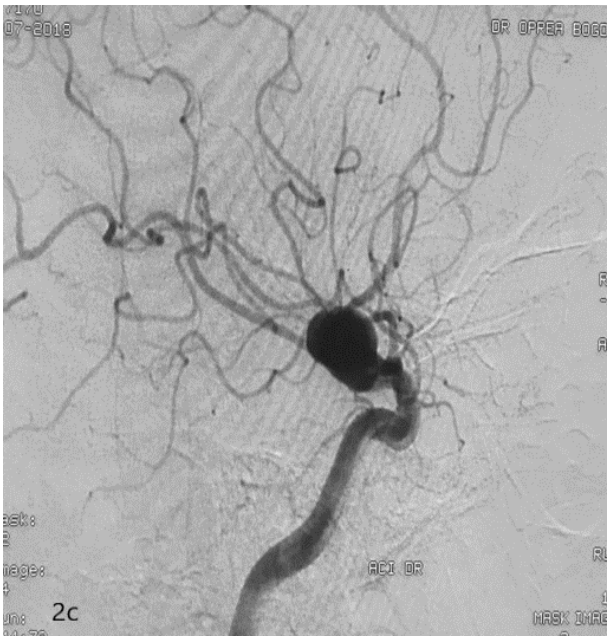
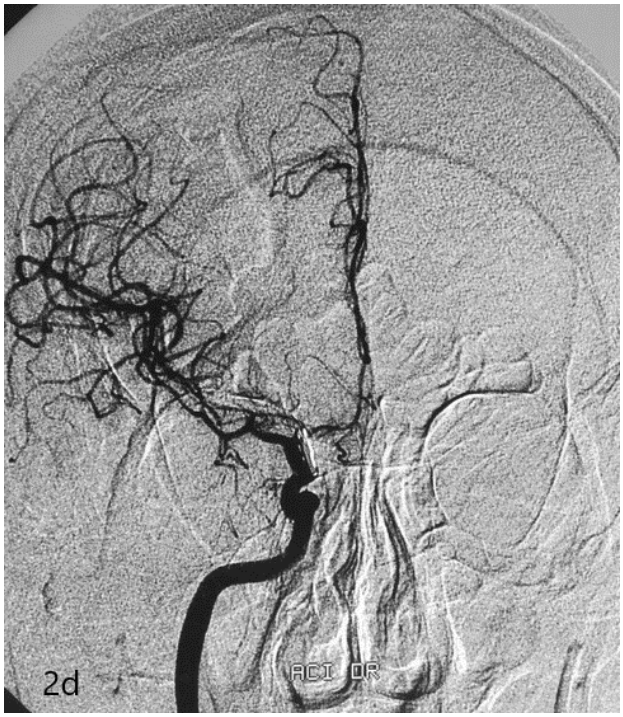


Figure 1: (1a,1b) Preoperative four vessels cerebral angiography showed a large, 14 mm length, ruptured, left ophthalmic artery aneurysm; (1c,1d) Postoperative control cerebral angiography showed the correct clipping of the aneurysm.









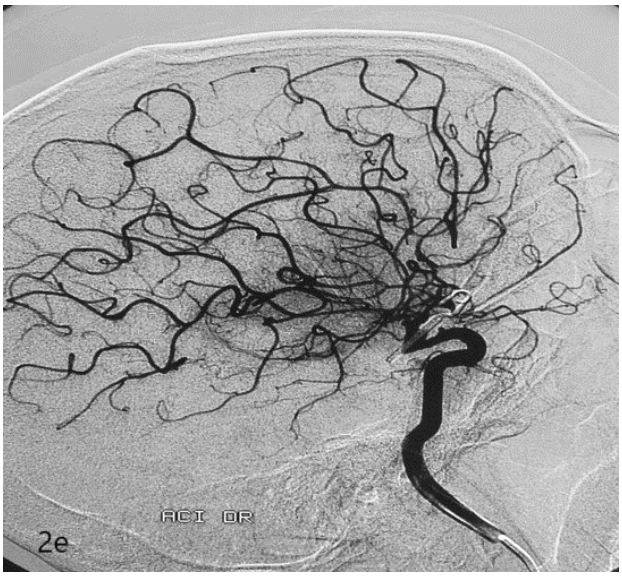


Figure 2: (2a) Preoperative cerebral computed tomography (CT) showed subarachnoid haemorrhage; computed tomography angiography (CTA) demonstrated a large right-sided paraclinoid aneurysm and the relation with the anterior clinoid process; (2b,2c) Preoperative four vessels cerebral angiography showed a 16 mm length right superior hypophyseal artery aneurysm; (2d,2e) Postoperative control cerebral angiography showed the correct obliteration of the aneurysm with a straight 11 Yasargil clip.

Discussion

Paraclinoid aneurysms are classified in clinoidal segment (C5) aneurysms and ophthalmic segment

(C6) aneurysms. This classification system offers prognostic information about lesion's propensity for subarachnoid haemorrhage (SAH) or cranial nerve deficits as well as the anatomic knowledge required for successful low-risk operative management of these lesions (28).

Paraclinoid aneurysms have a female preponderance (female:male ratio 9:1) and a high incidence of being multiple (4,12). Small (<5 mm) asymptomatic clinoidal segment aneurysms carry a very low risk of SAH, and isolated lesions should generally be treated with a conservative management plan including periodic follow-up imaging. Small symptomatic lesions (visual deficits or focal, unrelenting headache) and a lesion, whose protective anterior clinoidal process roof has been removed for treatment of another pathology in the same region, should be treated.

The most frequent paraclinoid aneurysms are ophthalmic aneurysms. The literature mostly described the frequency to be in the range of 1.3-5% (8,30). Although ruptured ophthalmic aneurysms require treatment, unruptured small asymptomatic aneurysms can undergo observation with serial imaging. Up to half of patients with ophthalmic segment aneurysms have additional intracranial aneurysms elsewhere (6,22). Most small aneurysms in the ophthalmic segment have a lower rupture risk than those at other intracranial locations. In this situation and for middle age or older adults, observation is a very reasonable option.

In the past, unless symptomatic, these aneurysms were usually not treated due to higher mortality and morbidity rates compared with other intracranial aneurysms (3). With the development of microsurgical technique, most of these aneurysms became operable, with lower mortality and morbidity (12). Potential management options for paraclinoid aneurysms include observation, endovascular treatment, microsurgical clip ligation, and carotid occlusion with or without bypass. Treatment is indicated for virtually all symptomatic aneurysms and for those larger than 1 cm. The patient presenting with visual loss should be treated urgently, ideally with surgery if the patient's risk factors and the experience of the operating team are reasonable. Endovascular technique is a viable option for the treatment of many aneurysms. The development of endoluminal flow-diverting devices applied to large or giant lesions has become a very good alternative to surgery for very complex paraclinoid lesions.

We considered that with all paraclinoid aneurysms, the patient's neck should be sterilely prepared and marked for dissection if proximal control becomes necessary. Some authors consider proximal control (4,6,27) is often unnecessary for small unruptured aneurysms, but often prudent for aneurysms that are ruptured or large. Proximal control can be achieved by temporary clipping of either the internal carotid artery (ICA) or common carotid artery (CCA). Clamping the CCA avoids potential injury to the ICA. Despite proximal ICA occlusion in the neck, back bleeding from the posterior communicating and ophthalmic arteries can be brisk if intraoperative rupture occurs.

The classic pterional craniotomy is used for adequate exposure of almost all ophthalmic artery aneurysms. An orbital osteotomy can be performed to provide additional exposure for larger aneurysms. The lesser wing is removed extradurally down to the base of the anterior clinoid process. The supraorbital craniotomy through the eyebrow incision is reasonable route for uncomplicated aneurysms.

The patient's head position during surgery for these medially situated aneurysms demands less neck rotation (15-20 degrees) to allow the surgeon to look under the optic nerve after the clinoidectomy. Slightly less head extension lessens the steep viewing trajectory under the anterior clinoid process.

Anterior clinoidectomy has great value in surgical exposure and treatment for most paraclinoid aneurysms. It can be completed both extradurally (Dolenc approach) or intradurally (8). In our opinion, intradural removal is preferred because it allows simultaneous visualization of the optic nerve and aneurysm during the entire dissection and enables immediate bleeding control if the aneurysm ruptures prematurely.

Extradural removal of the anterior clinoidal process is performed by extension of the medial dissection of lesser sphenoid wing (8). It uses of a high-speed diamond drill to hollow out the process until it is disconnected at its points of bony fixation. It is then extracted from its dural attachments, and cavernous sinus bleeding can be controlled with packing. This procedure should be avoided if a clinoidal segment aneurysm is suspected, because such aneurysm can

erode into and through the anterior clinoidal process (4,21).

With **intradural** anterior clinoid process removal, the dura is opened in a curvilinear fashion based on the sphenoid ridge, and the sylvian fissure is widely split, allowing the aneurysm, ICA, and visual system to be partially visualized (9). Two incision are made: first, a 3- to 4-cm longitudinal incision along the lesser sphenoid wing, starting from the tip of the anterior clinoid process and a second dural incision made perpendicular to the first, extending to and including sectioning of the falciform ligament (20). The dura is stripped free from the underlying bone and clinoid process is removed as in extradural way but with better visualization of the optic apparatus and the aneurysm. After anterior clinoidectomy is complete, the optic canal is unroofed, and the optic strut is drilled down to the base of the sphenoid bone. The optic nerve sheath is then sectioned laterally to allow further access to the medial portion of the ICA and mobilization of the optic nerve.

Aneurysm dissection and clip application

Small ophthalmic artery aneurysms are technically easy to clip. After extradural or intradural removal of the anterior clinoid process, opening of the falciform ligament and mobilization of the ICA, the origin of the ophthalmic artery and the neck of the small aneurysm should be readily visible. At the moment of clip application, it is advisable to gently retract the ICA laterally rather the optic nerve medially (2). We consider that temporary clipping of the cervical ICA greatly aids in the dissection maneuver by softening the aneurysmal sac.

Ophthalmic artery aneurysms can be ligated with a strait, curved or side-angled clip, depending on their size, complexity and surgeon preference. Cohen-Gadol considered that side-angled clips orient the blades parallel to the long axis of the parent ICA and allow efficient neck closure without causing accordion-like shortening of the carotid trunk. Especially in broad neck aneurysms he underlined that a perpendicular clipping technique across the ICA leads to partial neck closure, hemodynamic turbulence within the sac, and potentially intraoperative rupture (4).

There is a distinct difference in the technical complexity of ligation for superiorly projecting and anteromedially projecting ophthalmic aneurysms. Although the former is relatively straightforward, the latter are hidden under the optic nerve and often require a tandem clipping technique: a fenestrated clip around the ICA can often be used to close a remnant (18).

Whereas ophthalmic artery aneurysms are often well visualized, superior hypophyseal aneurysms project medially away from the surgeon, with the ICA blocking any substantial view of the neck (14,16). These aneurysms usually require an angled fenestrated clip with the ICA within the fenestration and the clip blades pointing toward the distal dural ring. Because the superior hypophyseal arteries are often very proximal, the tips of the clip blades must extend up to or past the distal dural ring to completely close the neck. If the ring is not circumferentially dissected, the clip blades will remain partially splayed open and the aneurysm sac will continue to fill (14,27).

In paraclinoid aneurysms, usually in large ones, visualization of the entire aneurysm neck during clip application is often impossible, and clip deployment proceeds with visualization of one blade and often inspection of the other, only after the clip is applied. Inspection should reveal no perforator injury. Depending on the anatomy, one or several angled or right angled fenestrated clips may be necessary (14).

Large and giant aneurysms present technical challenges, especially if their neck is extending into the carotid cave (4,14). They require the use of complete flow arrest (aneurysm trapping) or suction-decompression technique for their decompression, manipulation and clip placement. The purpose is not only to prevent intraoperative rupture, but also to obtain adequate neck visualization and reliable neck closure. With large to giant aneurysms, aneurysmal decompression using the retrograde suction-decompression technique can be lifesaving. With this technique, endovascular inflation of a balloon in the cervical ICA is followed by temporary clip occlusion of the distal ICA within the operative field. Retrograde suction of the blood using a balloon catheter in the neck provides dramatic deflation and clip reconstruction of the patent ICA (4).

In our cohort, surgical clipping of 18 paraclinoid aneurysms was performed. Overall, one patient got worse and died after surgery (5.5%; cerebral ischemia resulting from severe cerebral vasospasm), one patient (5.5%) did not improve and the

remainder (88.8%) improved in terms of mRS, at six months follow-up. No new visual field deficits or eye movement defects were recorded postoperatively.

Comparing the results in the literature, Kothandaram et al (19) in 1971, reported two deaths (20%) and two hemiplegic patients (20%) out of ten operated on with ophthalmic aneurysms. Fox (11), reported in 1988, that two of eight patients (25%) with paraclinoid ophthalmic aneurysms showed a transient reduction in visual acuity or ipsilateral blindness. Kobayashi et al (17), in 1989, operated on seven patients with the carotid cave subtype of ophthalmic aneurysms, two (29%) of which presented with visual disturbance postoperatively. In 1990, on a series of 54 patients operated for paraclinoid aneurysms, Day et al (6) reported a morbidity rate of 7% and a mortality rate of 6%. In a very large study by Hoh et al (13), presented in 2000, on 238 patients with carotid-ophthalmic aneurysms from which 180 where clipped, he reported a 6% morbidity rate and only a 0.4% mortality. On a series of 81 patients operated for paraclinoid aneurysms, published in 2005, Yonekawa et al (15), reported a surgery-related permanent morbidity in 6 patients (13%), and a surgery-related mortality rate of 0%. The more recent publication of Sames et al. (2014), on a series of 37 carotid-ophthalmic clipped aneurysms, reported two patients got worse after surgery (5.4%, one unruptured), three patients (8.1%) did not improve and the remainder (86.5%) improved in the terms of GOS (25).

Microsurgery with clip ligation, including skull base approaches, is a proven effective and durable means to permanently securing paraclinoid aneurysms with good outcomes (1,6,26). Complications generally revolve around the anatomic structures encountered within the region—the ICA, arterial perforators, and neighboring cranial nerves. Even though intraoperative angiography may demonstrate initial ICA patency, delayed stenosis or thrombosis can still occur. Any evidence of focal neurological deficits after surgery should be immediately addressed with CT and angiography, and the patient returned to the operating room for emergency re-exploration and clip adjustment if obstruction is identified.

Postoperative visual deterioration, a potential complication in all paraclinoid aneurysm operations, is usually attributable to excessive optic nerve manipulation or perforator compromise during the exposure or clip placement. The clip should not compress or rotate the optic nerve that is especially intolerant of torsion. Delayed visual worsening indicates a need for re-exploration to assure the clip is not displaced, causing compression (4). The first series to define the microsurgical anatomy of the ophthalmic segment and meticulously analyze operative results reported excellent outcomes, defined as no postoperative neurological deficit, in 87% of the patients (6). Ischemic injury and/or increased visual deficits occurred in 11% of patients, almost all of whom had had very large or giant aneurysms of the superior hypophyseal artery (6).

In a later study, Nanda and Javalkar (23) observed an 8.7% rate of visual deterioration after surgery. No statistically significant difference in outcome was noted between giant and non-giant aneurysms (23). De Oliveira and colleagues (24) reported that most patients had improved visual function after surgery. Dehdashti and colleagues (7) found an overall surgery-related visual complication rate of 14%.

A far as the recovery of visual dysfunction is concerned, the results reported in the literature suggest first few months (10,29) or three months (5) for an operation to be done from the onset of symptoms. Furthermore, results in the literature suggest that symptomatic unruptured C6 aneurysms should preferably be treated surgically as the actual cause of the symptoms is eliminated in that way. Coiling or flow diverter implantation can exclude the aneurysm from the circulation by inducing thrombosis in the sac, but will not eliminate its expansive behaviour.

Postoperative oculomotor, trochlear, and abducens palsies, as well as ptosis and miosis secondary to sympathetic fiber disruption, are generally the result of surgical trauma during ACP removal, clip blade advancement, excessive cranial nerve manipulation, or cavernous sinus packing. These deficits are usually partial and transient in nature and are best avoided through careful dissection of the cranial nerves and their blood supply.

Conclusion

Paraclinoid aneurysms are known to demand special therapeutic considerations due to their special location of close vicinity to bony structures and important neural and vascular structures. Size and

location of the aneurysm, and the fact that it is ruptured or not, are main determinants in the decision to perform surgery. Large and giant aneurysms are daunting the handle without generating new visual deficits. Even it is a small series, the mortality and morbidity of paraclinoid aneurysm surgically treated in our department, both ruptured and unruptured, are relatively low and comparable with the results presented in the literature. For an experienced neurovascular team, microsurgical aneurysm clipping can currently be a good therapeutic option for paraclinoid aneurysms.

References

1. Baccin CE, Krings T, Alvarez H, et al.: Multiple mirror-like intracranial aneurysms. Report of a case and review of the literature. *Acta Neurochir (Wien).* 148:1091,2006
2. Batjer HH, Kopitnik TA, Giller CA, Samson DS. Surgery for paraclinoidal carotid artery aneurysms. J Neurosurg.;80(4): 650–658,1994
3. Bohm E, Hugosson R, Wolgast M. Carotid ligation for the treatment of carotid artery aneurysms: Where we are? J Neuroradiol ;38:40-6, 2011
4. Cohen-Gadol A. Paraclinoid Aneurysm in The Neurosurgical Atlas. DOI: https://doi.org/10.18791/nsatlas.v3.ch01.9
5. Date I, Asari S, Ohmoto T. Cerebral aneurysms causing visual symptoms: their features and surgical outcome. Clin Neurol Neurosurg;100: 259-267, 1998.
6. Day AL. Aneurysm of the ophthalmic segment. A clinical and anatomical analysis. J Neurosurg; 72: 677–691, 1990.
7. Dehdashti AR, Le Roux A, Bacigaluppi S, et al.: Long-term visual outcome and aneurysm obliteration rate for very large and giant ophthalmic segment aneurysms: assessment of surgical treatment. *Acta Neurochir (Wien)*, 154:43, 2011.
8. Dolenc VV. A combined epi- and subdural direct approach to carotid-ophthalmic artery aneurysms. J Neurosurg ;62:667-72, 1995
9. Du R, AL Day: Microsurgery of paraclinoid aneurysms. 6th ed JR Youmans Neurological Surgery, Saunders Philadelphia ,Vol. 4:3824, 2011
10. Ferguson CG, Drake CG. Carotid-ophthalmic aneurysms: the surgical management of those cases presenting with compression of the optic nerves and chiasm alone. Clin Neurosurg ;27:263-307, 1980
11. Fox JL. Microsurgical treatment of ventral (paraclinoid) internal carotid artery aneurysms. Neurosurgery 22:32-9, 1988
12. Guidetti B, La Torre E. Carotid-ophthalmic aneurysms. A series of 16 cases treated by direct approach. Acta Neurochir (Wien) 22:289-304,1970
13. Hoh BL, Carter BS, Budzik RF, Putman CM, Ogilvy CS. Results after surgical and endovascular treatment of paraclinoid aneurysms by a combined neurovascular team. Neurosurgery 48: 78–90,2001
14. Joo W, Funaki T, Yoshioka F, Rhoton AL. Microsurgical anatomy of the carotid cave. Neurosurgery.; 70(2 Suppl Operative):300–311; discussion 311–312, 2012
15. Khan N, Yoshimura S, Roth P, Cesnulis E, Imhof H, Yonekawa Y. Conventional microsurgical treatment of paraclinoid aneurysms: state of art with the use of the selective extradural anterior clinoidectomy SEAC. Acta Neurochir, (Suppl) 94:23-29, 2005
16. Kim JM, Romano A, Sanan A, van Loveren HR, Keller JT. Microsurgical anatomic features and nomenclature of the paraclinoid region. Neurosurgery; 46(3): 670–680; discussion 680–682, 2000
17. Kobayashi S, Kyoshima K, Gibo H et al. Carotid cave aneurysms of the internal carotid artery. J Neurosurg ;70:216-21, 1989
18. Korosue K, Heros RC. "Subclinoid" carotid aneurysm with erosion of the anterior clinoid process and fatal intraoperative rupture. Neurosurgery 31(2): 356-359; discussion 359-360, 1992
19. Kothandaram P, Dawson BH, Kruyt RC. Carotid-ophthalmic aneurysms. A study of 19 patients. J Neurosurg 34:544-8, 1971
20. K Kyoshima, S Oikawa, S Kobayashi: Interdural origin of the ophthalmic artery at the dural ring of the internal carotid artery. Report of two cases. *J Neurosurg* 92:488, 2000
21. Lawton MT. Ophthalmic artery aneurysms in *Seven Aneurysms: Tenets and techniques for clipping*. Ed Thieme, 2011.
22. N al-Rodhan: Aneurysms within the cavernous sinus and transitional cavernous aneurysms. R Wilkins S Rengachary *Neurosurgery.* 3rd.ed 1996 McGraw-Hill New York 2283, 1996
23. Nanda A, Javalkar V: Microneurosurgical management of ophthalmic segment of the internal carotid artery aneurysms: single-surgeon operative experience from Louisiana State University, Shreveport. *Neurosurgery* pp 68:355, 2011
24. de Oliveira JG, Borba LA, Rassi-Neto A, et al.: Intracranial aneurysms presenting with mass effect over the anterior optic pathway:neurosurgical management and outcomes. *Neurosurg Focus* 26: E3, 2009
25. Orlicky M,Sames M, Hejcl A, Vachata P. Carotid-ophthalmic aneurysms – Our results and treatment strategy. British Journal of Neurosurgery, early online 1-6, 2014
26. Raco A, Frati A,Santoro A, et al.: Long-term surgical results with aneurysms involving the ophthalmic segment of the carotid artery. *J Neurosurg.* pp 108:1200, 2008
27. Samson DS, Batjer HH. Aneurysms of the anterior carotid wall (ophthalmic). Chapter 4 in: Intracranial Aneurysm Surgery:Techniques. Futura Publishing 1990.

28. Turkmani HA, Day AL. Microsurgery of paraclinoid aneurysms. In Youmans&Winn Neurological Surgery 7th edition vol 4, pp.3298-3306, 2017
29. Vargas ME, Kupersmith MJ, Setton A, Nelson K, Berenstein A. Endovascular treatment of giant aneurysms which cause visual loss. Ophthalmology 101:1091-8, 1994
30. Yasargil MG. Microneurosurgery: In 4 Volumes. New York:Thieme Medical Pub, 1984.

Romanian Neurosurgery (2020) XXXIV (1): pp. 41-45
DOI: 10.33962/roneuro-2020-006
www.journals.lapub.co.uk/index.php/roneurosurgery

Management of facial paralysis following treatment of neurosurgical tumours

Costan V.V.[1], Ciofu M.L.[1], Sava F.[1], Boisteanu O.[2], Dabija M.[3]

[1] Faculty of Dental Medicine, Department of Surgery, Oral and Maxillofacial Surgery, "Grigore T. Popa" University of Medicine and Pharmacy, Iasi, ROMANIA
[2] Faculty of Dental Medicine, Department of Surgery, Anaesthesiology, "Grigore T. Popa" University of Medicine and Pharmacy, Iasi, ROMANIA
[3] Faculty of Medicine, Department of Surgery II, Neurosurgery, "Grigore T. Popa" University of Medicine and Pharmacy, Iasi, ROMANIA

Keywords
facial nerve,
paralysis,
static reanimation,
lagophthalmos,
intracranial tumour

ABSTRACT
The purpose of this study is to present our experience on improving the quality of life of patients with facial paralysis due to an operated intracranial tumour, by performing minimally invasive static reanimation procedures. We reviewed the clinical information pertaining to neurosurgical patients with facial paralysis that underwent static reanimation. The study included 11 patients with complete facial nerve paralysis of all nerve branches, that reported different primary complaints upon presentation. The performed procedures consisted of gold plate insertion into the superior eyelid, inferior eyelid ectropion correction or suture suspension. The functional results were favourable in all cases and the resulting appearance was acceptable. The choice of the different techniques used is discussed. Good outcomes are possible using static reanimation with an adequate adaptation of the techniques to the main patient complaint.

Corresponding author:
Ciofu M.L.

Faculty of Dental Medicine, Department of Surgery, Oral and Maxillofacial Surgery, Grigore T. Popa" University of Medicine and Pharmacy, Iasi, Romania

ciofu_mihai@yahoo.com

Scan to access the online version

INTRODUCTION

Definitive facial nerve injury during neurosurgical procedures for benign or malignant tumors of the posterior and middle cranial fossae, cerebellopontine angle and lateral skull base, is often unavoidable, either due to existing preoperative tumor invasion or compression, or due to the proximity of the nerve, since the most important desiderate is avoiding tumor recurrence (1, 2).

Sequelae associated with facial nerve paralysis lead to important functional impairment as well as aesthetic changes comprising of a ptotic appearance of the involved hemiface with consequences regarding the emotional wellbeing of the patient and social integration. These factors cause a significant decline in the life quality of the patient (3, 4). Static reanimation procedures aim to restore facial symmetry, improve deglutition, mastication and phonation disturbances, while also decreasing complications that endanger vision (5, 6).

The particularity of patients with operated tumors involving the neurosurgical territory is that the nerve is injured along its intracranial route, making functional rehabilitation by nerve graft interposition practically impossible (1). Additionally, using the contralateral facial nerve branches carries the risk for bilateral injury. Therefore, the minimally invasive static reanimation procedures are highly indicated and can provide an increase in the quality of life of those patients without the need to perform major surgery.

In this manuscript we aim to present the benefits of different static facial reanimation procedures that we used in patients operated for neurosurgical tumors, in relation to the main complaints of the patients.

Material and methods

We reviewed the medical charts of patients that presented for facial nerve paralysis following surgery for the removal of tumors in the neurosurgical territory. Only patients that underwent static reanimation procedures were included in the study. The minimum follow-up period was 6 months. The general information, the main complaints of the patients, the time passed from the onset of facial paralysis, the type of static reanimation procedure and the postoperative outcomes were all documented.

Results

In total, 11 patients were included in the study, consisting of 6 men and 5 women, aged 46 to 72 years old. All patients had complete nerve paralysis of all facial nerve branches, showing all known signs of nerve paralysis in different degrees, including facial asymmetry, lagophthalmos, lower eyelid ectropion, brow ptosis, ptosis of the oral commissure with sialorrhea, deglutition, mastication and phonation disturbances. Still, the most bothersome complaint upon presentation was lagophthalmos in 6 patients, sialorrhea and difficulties eating in 3 patients, and facial asymmetry in 2 patients.

The time interval between the onset of facial paralysis and performing the static reanimation procedure was minimum 3 months and maximum 1 year. Surgery was performed under local anesthesia in all cases. 7 patients had multiple procedures, performed either at the same time or in separate sessions. One patient had gold plate insertion into the superior eyelid, lower eyelid ectropion correction and suture suspension of the affected hemiface. Another patient had gold plate insertion into the superior eyelid and inferior eyelid ectropion correction. Five patients had gold plate insertion into the superior eyelid and suture suspension. Two patients had only gold plate insertion into the superior eyelid and another two patients had only suture suspension.

For the patients in which multiple staged procedures were performed, it was considered that the functional results could be improved after the initial facial reanimation surgery, therefore scheduling for another procedure to complete the previous results. After the completion of all procedures, the functional outcomes were optimal, and the aesthetic results were acceptable. Facial asymmetry was ameliorated for all patients that underwent facial suture suspension in all facial segments, while only the upper face was improved in the three patients undergoing only periorbital surgery. Lagophthalmos was improved in all patients with a decrease in the subjective complaints. Sialorrhea and eating difficulties were improved in all patients undergoing suture suspension. There were no infectious complications or hematoma formation postoperatively. Minor ecchymosis and postoperative edema were frequent encounters.

The favorable outcomes are presented through images of a facial nerve paralysis patient that underwent static reanimation by simultaneous gold plate insertion into the superior eyelid and suture suspension (Fig. 1-5).

Figure 1. Preoperative aspect of the patient during eye closure, demonstrating left lagophthalmos and facial asymmetry.

Figure 2. Preoperative aspect of the patient during cheek inflation, showing decreased tonus in the left cheek.

Figure 3. Immediate postoperative aspect following insertion of a gold plate into the left superior eyelid and suture suspension of the left hemiface.

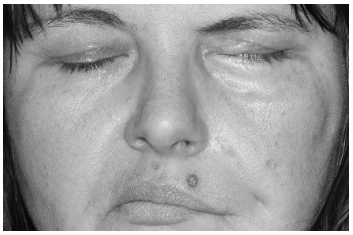


Figure 4. Post-operative aspect 10 days after surgery during eye closure, demonstrating complete left eye closure and ameliorated facial asymmetry with lifting of the left oral commissure, definition of the nasogenian sulcus and lifting of the cheek with improved position of the inferior eyelid.

Figure 5. Postoperative aspect 10 days after surgery showing increased tonus of the left cheek and lifted position of the left oral commissure during cheek inflation.

Discussions

Dynamic facial reanimation involves complex, time consuming procedures performed under general anesthesia, that are not without risks. In patients operated for intracranial tumors in which the injured proximal facial nerve cannot be used for primary reconstruction using interposition nerve grafts, a dynamic reanimation may necessitate the scarification of another cranial nerve, including the trigeminal, accessory, hypoglossal or contralateral facial nerve (1, 2). The indication for such an invasive procedure should carefully consider the general condition of the patient, the existing associated disorders as well as the long-term prognosis. For older patients with multiple comorbidities, oncologic patients, for patients reluctant to undergo another major procedure or for patients with long-standing facial paralysis, static reanimation offers good outcomes with minimal risks, since it involves local anesthesia and short duration of surgery (7, 8).

The goal of static reanimation procedures is to improve the functional and esthetic disturbances associated with facial nerve paralysis. Multiple types of procedures exist, targeting different sequelae of facial paralysis. Often, several procedures are necessary in order to address all issues and achieve best outcomes (9). The good results achieved in our study were due to the careful tailoring of the procedures performed, considering the most bothersome complaint reported by each patient. By addressing the main expressed issue, all patients perceived an improvement after the static reanimation intervention.

The periorbital changes resulting from incomplete eyelid closure and inferior eyelid ectropion are frequently the main focus of reanimation surgery due to the distressing complaints of patients including foreign body sensation and pain, but also due to the serious complications regarding vision that can eventually develop (10, 11). In our study, improving eyelid closure was the main goal in the patients primarily having this complaint. In this regard, we achieved optimal outcomes by inserting a gold plate into the superior eyelid, for added weight. Plate positioning was important in order to achieve maximal eyelid closure and avoid plate exposure over time, similarly to the reports of other authors (12). Other existing procedures that can improve lagophthalmos are levator lengthening and palpebral spring insertion (13). However, superior eyelid loading is the preferred method for many surgeons (14, 15).

Lagophthalmos is known as the condition that can cause the most functional damage in facial nerve paralysis patients, due to the increased corneal exposure and drying with subsequent ulcerations and infections that can eventually cause blindness or even the loss of the affected eye (13, 16). It is frequently the first facial nerve sequelae that is addressed during reanimation procedures. In our study it was also the most prevalent complaint, but in some cases, issues related to difficulties during eating and sialorrhea were reported as more distressing, mainly due to the struggling to eat adequately, but also due to social concerns.

Eating difficulties for facial paralysis patients are mostly caused by the food remaining trapped during mastication in the inferior vestibule that resembles a bag, due to the lack of tonus of the cheek. The associated sialorrhea only worsens the symptoms

due to mucosa dryness, difficulty in forming the food bolus, but also causing frustration during social encounters. Suspension of the oral commissure and genian region using usual sutures or barbed threads, lifts the tissues, preventing saliva leakage, but also improves the tonus of the cheek, therefore decreasing food retaining (17, 18). In addition, lagophthalmos is also improved, since the heavy genian region that pulls down on the inferior eyelid, aggravating ectropion and lagophthalmos, is repositioned superiorly, rendering support to the inferior eyelid (6, 18). In our study, in the patients with this main complaint, suture suspension was first performed, since it allowed the initial alleviation of the most distressing symptoms. Ectropion correction was mainly indicated in addition to gold plate insertion, for the patients exhibiting significant laxity and length of the inferior eyelid.

For the patients that were most bothered by the facial asymmetry, suture suspension was also the best initial choice of static reanimation, since it allowed addressing the upper, middle and lower face simultaneously, and therefore ensured better cosmetic results. Good outcomes regarding the restoration of facial symmetry using suspension techniques have also been reported by other studies (19, 20). Refinements of particular areas can always be made using barbed sutures, due to the minimally invasive character of the procedure, even when plain suture suspension was initially performed (18).

Conclusion

Static reanimation procedures render favorable results when the type of intervention is tailored to fit the most pressing patient complaint. Particularly for neurosurgical patients with intracranial injury of the facial nerve, these procedures ensure a noninvasive modality of increasing the quality of life of the patients with minimal complications.

References

1. Rozen SM, Harrison BL, Isaacson B, Kutz JW Jr, Roland PS, Blau PA, Barnett SL, Mickey BE. Intracranial Facial Nerve Grafting in the Setting of Skull Base Tumors: Global and Regional Facial Function Analysis and Possible Implications for Facial Reanimation Surgery. Plast Reconstr Surg. 2016; 137(1):267-78.
2. Panossian A. Facial paralysis reconstruction in children and adolescents with central nervous system tumors. J Pediatr Rehabil Med. 2014; 7(4):295-305.
3. Nellis JC, Ishii M, Byrne PJ, Boahene KDO, Dey JK, Ishii LE. Association Among Facial Paralysis, Depression, and Quality of Life in Facial Plastic Surgery Patients. JAMA Facial Plast Surg. 2017; 19(3):190-196.
4. Ishii LE, Nellis JC, Boahene KD, Byrne P, Ishii M. The Importance and Psychology of Facial Expression. Otolaryngol Clin North Am. 2018; 51(6):1011-1017.
5. Robinson MW, Baiungo J. Facial Rehabilitation: Evaluation and Treatment Strategies for the Patient with Facial Palsy. Otolaryngol Clin North Am. 2018; 51(6):1151-1167.
6. Costan VV. Management of Extended Parotid Tumors. Editura Springer, 2016; ISBN 978-3-319-26543-8.
7. White H, Rosenthal E. Static and dynamic repairs of facial nerve injuries. Oral Maxillofac Surg Clin North Am. 2013; 25(2):303-12.
8. Kim L, Byrne PJ. Controversies in Contemporary Facial Reanimation. Facial Plast Surg Clin North Am. 2016; 24(3):275-97.
9. Lafer MP, O TM. Management of Long-Standing Flaccid Facial Palsy: Static Approaches to the Brow, Midface, and Lower Lip. Otolaryngol Clin North Am. 2018; 51(6):1141-1150.
10. Joseph SS, Joseph AW, Douglas RS, Massry GG. Periocular Reconstruction in Patients with Facial Paralysis. Otolaryngol Clin North Am. 2016; 49(2):475-87.
11. Homer N, Fay A. Management of Long-Standing Flaccid Facial Palsy: Periocular Considerations. Otolaryngol Clin North Am. 2018; 51(6):1107-1118.
12. Oh TS, Min K, Song SY, Choi JW, Koh KS. Upper eyelid platinum weight placement for the treatment of paralytic lagophthalmos: A new plane between the inner septum and the levator aponeurosis. Arch Plast Surg. 2018; 45(3):222-228.
13. Leckenby JI, Ghali S, Butler DP, Grobbelaar AO. Reanimation of the brow and eye in facial paralysis: Review of the literature and personal algorithmic approach. J Plast Reconstr Aesthet Surg. 2015; 68(5):603-14.
14. Rofagha S, Seiff SR. Long-term results for the use of gold eyelid load weights in the management of facial paralysis. Plast Reconstr Surg 2010;125(1):142e9.
15. Rozen S, Lehrman C. Upper eyelid postseptal weight placement for treatment of paralytic lagophthalmos. Plast Reconstr Surg 2013;131(6):1253e65.
16. Costan VV, Doscas A, Ciofu ML, Boişteanu O, Popescu E. On the management of paralytic lagophthalmos. International Journal of Medical Dentistry 2018, 22(4):433-436.
17. Biglioli F. Facial reanimations: part II--long-standing paralyses. Br J Oral Maxillofac Surg. 2015; 53(10):907-12.
18. Costan VV, Popescu E, Sulea D, Stratulat IS. A New Indication for Barbed Threads: Static Reanimation of the Paralyzed Face. J Oral Maxillofac Surg. 2018; 76(3):639-645.
19. Langille M, Singh P. Static Facial Slings: Approaches to

Rehabilitation of the Paralyzed Face. Facial Plast Surg Clin North Am. 2016; 24(1):29-35.
20. Leach G, Kurnik N, Joganic J, Joganic E. Multivectored Superficial Muscular Aponeurotic System Suspension for Facial Paralysis. J Craniofac Surg. 2017; 28(4):882-887.

Romanian Neurosurgery (2020) XXXIV (1): pp. 46-51
DOI: 10.33962/roneuro-2020-007
www.journals.lapub.co.uk/index.php/roneurosurgery

Mechanical thrombectomy techniques for acute ischemic stroke

A. Chiriac, Georgiana Ion[1], N. Dobrin[1], Dana Turliuc, I. Poeata

[1] "Gr. T. Popa" University of Medicine and Pharmacy, Iasi, ROMANIA
[2] "Prof. Dr. N. Oblu" Clinic Emergency Hospital, Iasi, ROMANIA

ABSTRACT
Mechanical thrombectomy technique was introduced as an effective and secure method in acute ischemic stroke patients suffering from intracranial large vessel occlusion (LVO). In this article, we will review the main mechanical thrombectomy techniques and current trends in this type of treatment for acute ischemic stroke.

Keywords
mechanical thrombectomy, acute ischemic stroke

Corresponding author:
A. Chiriac

"Prof. Dr. N. Oblu" Clinic Emergency Hospital, Iasi, Romania

chiriac_a@hotmail.com

INTRODUCTION

The large global studies have shown that stroke is the second most common cause of death and third commonest cause of disability in the world. The proven limitations of intravenous and intra-arterial thrombolysis for performing large vascular occlusion recanalization resulted in the introduction of mechanical thrombectomy (MT). This was also favoured by technological advances in endovascular neurosurgery with better catheters, to allow more distal access and more effective devices for efficient intravascular thrombus penetration and capture. Starting with 2015, the role of mechanical thrombectomy for the occlusion of large vessels in acute ischemic stroke has been scientifically proven with the publication of seven randomized controlled trials that demonstrated better outcomes compared to medical management alone. Mechanical thrombectomy evolved from the first generation of thrombus penetration devices, to the direct thrombus-aspiration systems and then to the second generation of devices represented by stent retrievers. In this article we will review the main mechanical thrombectomy techniques and current trends in this type of treatment for acute ischemic stroke [1,2].

The limitations of intravenous thrombolysis have led to the exploration of alternative or complementary treatment approaches for acute ischemic stroke (AIS). Endovascular mechanical thrombectomy has developed over the past years as a safe and efficient intervention for AIS treatment. The fast advancement in catheter and endovascular device technology has led to an increasing number of techniques used in mechanical thrombectomy in patients suffering from an emergent large vessel occlusion.

Scan to access the online version

Clot retriever technique

The need to improve the vascular recanalization technique correlated with reducing the risk of cerebral haemorrhage by limiting thrombolytic administration has led to the development of the first mechanical thrombectomy devices. The first steps in the appearance of mechanical thrombectomy devices were performed by Dr. Y. Pierre Gobin of the University of California, Los Angeles, who patented an endovascular device similar to a crib for the recovery of an accidentally released foreign body (usually coils) in the cerebral vasculature. Due to its limited use the device was subsequently taken over and licensed by Concentric Medical, Inc. and called the Mechanical Embolus Removal in Cerebral Ischemia (MERCI) retriever. The Merci retriever is a corkscrew shape device with helical Nitinol loops specifically designed and tested for distal placement and enbloc removal of the thrombus for first time approved by FDA in 2004. This technique involves physical extraction of the thrombus through a catheter (Figure 1). Even though this device has benefited from considerable redesigning in time, all versions were preferably used in conjunction with an 8- or 9-F balloon guiding catheter to reduce the risk of distal emboli [4,10]. The target vessels were the proximal segments of major cerebral arteries, predominantly M1 segments of the middle cerebral and vertebrobasilar arteries. MERCI and multi-MERCI trials were prospective, nonrandomized, multicenter, and single-arm trials that reported the safety and efficacy of MERCI device in patients presenting with moderate to severe stroke (National Institutes of Health Stroke Scale NIHA score ≥8) from a large-vessel occlusion and treated within 8 hours of symptom onset[2,4,10].

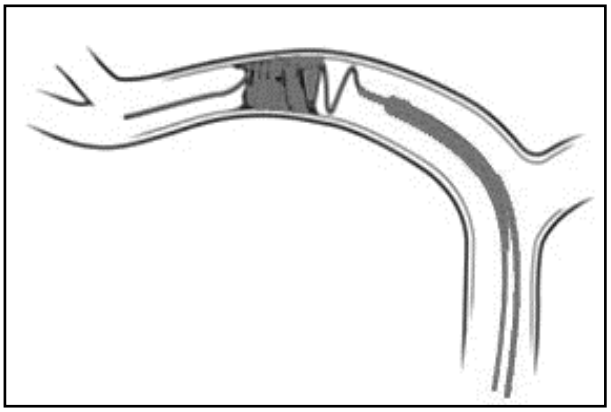


Figure 1: Clot retriever technique

Self-expanding stents technique

Based of development of the retrieval clot technique, attempts to use self-expanding stents for the purpose of assisting and restoring cerebral revascularization have been reported. Intracranial angioplasty with self-expanding stents was described for the treatment of acute ischemic stroke due to intraarterial atherosclerotic lesions. Even if high rates of stent placement success were achieved immediate and delayed complications were brought into discussion (Figure 2).

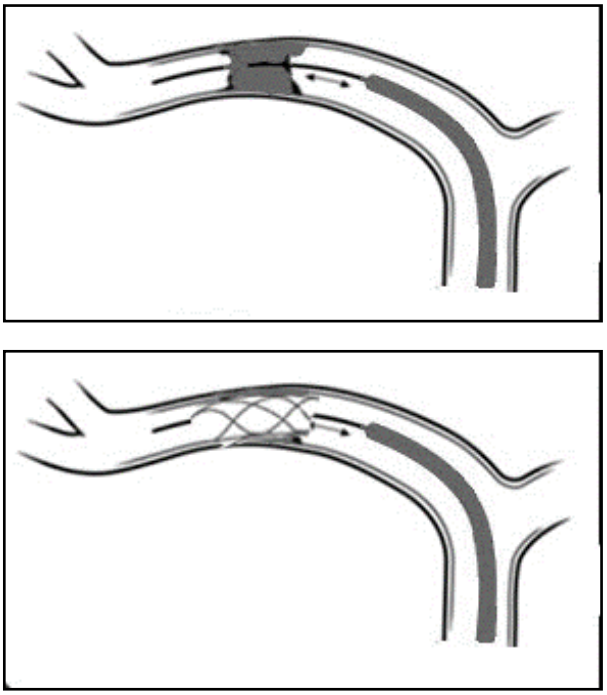


Figure 2: Self-expanding stents technique

Distal emboli due to stent detachment, intra-stent thrombosis or re-stenosis as well as potential haemorrhagic risk of concurrent prophylactic antiplatelet and anticoagulation therapy are just a few of the limitations of the frequent use of this technique. Initial evaluation of the Wingspan stent stent in the SARIS pilot study (ischemic stroke assisted recanalization) in patients with contraindicated or not available intravenous thrombolysis showed a 100% recanalization rate and a 45% favorable clinic evolution at 90 days. A 10% stent related complications was also reported [3,5].

Thrombus aspiration technique

The first attempts of thrombectomy by manual suction were described by the use of catheters

placed in front of the thrombus and by the manual generation of suction through syringes connected to the hub of the catheter, with excellent results [5,11].
In December 2007, the Penumbra (PS) system for aspiration of clots (Penumbra, Inc., Alameda, CA) was the second FDA approved device in the treatment of acute ischemic stroke. This technique involves the use of a large lumen catheter device that is advanced through a guide catheter to a point near occlusion, in contact with the clot. In the case of non-adherent clots, with easy mobilization, after capturing them at the top of the suction catheter, it is withdrawn slowly, maintaining the continuous aspiration at a vacuum pump. Simultaneous aspiration is also applied to the lateral port of the intracranial access system (sheath or guide catheter) to prevent thrombus displacement from the tip of the suction catheter, because it is retracted into the sheath. In the case of adherent clots, a microwire called separator is repeatedly passed through the thrombus to disconnect / fragment the clot, and the constant suction is applied to the PS catheter to aspirate the thrombus fragments.

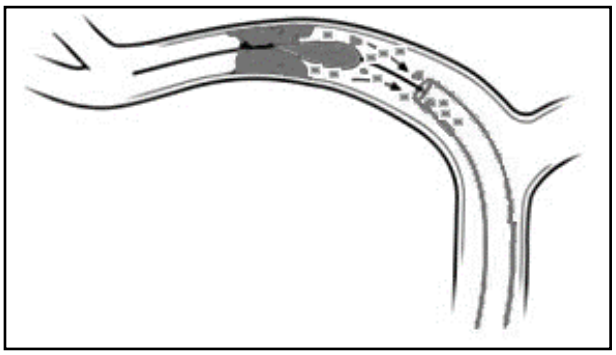


Figure 3: Direct aspiration first pass technique

The introduction of large and flexible suction catheters has led to the development of a new technique called a direct aspiration first pass technique (ADAPT), which can be used as the primary way for revascularization. Kang et al. first reported the use of direct aspiration thrombectomy [2,5,11]. In their report, TICI 2b-3 revascularization was performed in 81.9% of cases. In this technique, the largest possible aspiration catheter is advanced over a microcatheter and microwire (which are used to pass the thrombus and provide stable support for catheter advancement) and positioned immediately adjacent to the occlusion site (Figure 3). Once the suction catheter is in contact with the thrombus, the microcatheter and microwire are removed, and the suction catheter with the clot is withdrawn into the guiding catheter, while continuous negative pressure is applied. Particular attention is given to the moment when the clot is captured, the aspiration forces in the lumen of the vessel are lost and the flow around the intermediate catheter can lead to fragmentation and distal migration during retrieval. This shortcoming can usually be eliminated by using alternating and intermittent aspiration by connecting the continuous aspiration pump tubing into one end of a three-way valve and a 60 cc vacuum syringe. If an optimal control of the catheter advancement exists the aspiration turned on upon contact will be allowed. Thus, the suction force on the thrombus becomes appreciable when the catheter is very close to the thrombus (below 1 mm) and may increase until it comes into contact with the thrombus [2,3,4,5].

Another important element that needs to be carefully analyzed is the direction of the suction catheter in relation to the vessel / thrombus to minimize the number of passes and several procedures. In situation of unfavorable angles, a microcatheter over microwire can be used to direct the tip of the aspiration catheter when positioned across the occlusion. The microcatheter/microwire can be kept inside the system during aspiration.

The continuous technological advances allowed the manufacture of last generation of aspiration catheter systems with larger inner-size, excellent navigability, efficacy, and safety profile. Reperfusion catheters such as ACE68, ACE64 and, more recently, JET7 with a suction lumen of 0.072 " (Penumbra) are the best examples. A prospective study initiated by the European Registry on the ACE Reperfusion Catheters and the Penumbra System in the Treatment of Acute Ischemic Stroke (PROMISE, NCT02678169) showed a mTICI 2b-3 revascularization rate of 93.1%, 90-day mRS 0–2 rate of 61% and a 2.9% of sICH complication, with 7.5% mortality at 90-days[13].

Heit JJ, et al. described in 2018 a so called “SNAKE” technique witch is a technical variation of ADAPT. A SOFIA catheter (Microvention) is advanced into the intracranial circulation without using a guiding wire, microcatheter, or microwire. Because the distal end of the Sofia catheter is extremely soft and pliable it allows a safe “push” of the catheter into different parts of the circle of Willis. When the tip of the Sofia

is successfully reaches the proximal end of the clot, a manual aspiration with a large syringe can be performed, and the SOFIA catheter is slowly withdrawn into the guide catheter. [1,2,5]. Also, the use of smaller thrombo-aspiration catheters such as the Penumbra 3 Max has been reported to be safe and leading to good reperfusion rates especially for distal locations.

STENT RETRIEVER TECHNIQUE

The stent retrievers represent the second generation of mechanical thrombectomy devices with has derived from nondetachable neurovascular stents used for aneurism coiling support. The introduction of these devices was an important advantage for endovascular treatment of AIS, offering the advantage of navigability and rapid recanalisation of a stent without the potential long-term complications. The detachable Solitaire intracranial stent (Covidien/EV3; Plymouth, MN) turned into a primary clot retriever.

The tehchnique utilize a retrievable stent that is advanced within a microcatheter through the thrombus until a few millimeters distal to the clot. Thus, the stent is deployed capturing the thrombus into the stent struts and displacing it peripherally to the vessel wall thus restoring blood flow. After 3-5 minutes, the microcatheter and stent retriever are simultaneous removed under continuous proximal aspiration with a syringe (Figure 4).

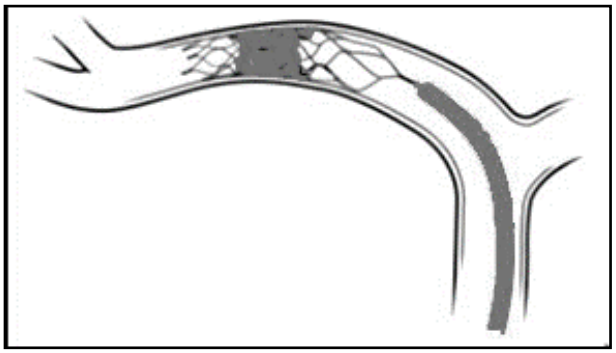


Figure 4: Stent retriever technique

Some improved variants of this technique have also been described in the literature. Haussen et all described an alternative to the conventional unsheathing technique of the stent retriever called **Push and Fluff Technique** or "active deployment". This particular deployment maneuver consists in positioning the stent retriever across the clot and unsheathing the distal end of the device until good wall apposition (anchoring) is achieved (brief unsheathing step). Then, forward force is applied to the device delivery wire, leading to spontaneous retraction of the microcatheter (pushing step). At the main clot area, additional forward tension is applied to the microcatheter while the delivery wire continues to be pushed to maximize the device struts expansion into embolus (fluffing step). The major concern related to this technique was the potential association with adverse events related to the addition of radial force or forward movement of the device especially in curved vessels. The reports a higher first-pass reperfusion success (modified TICI 2b-3 with the first pass), 58% of final full reperfusion and a lower number of overall passes compared to conventional unsheting technique [6].

Dual Stent Retriever Technique was proposed as alternative thrombectomy technique in case of "saddle" bifurcation occlusion. This technique involves the use of two different or identical types of stents retriever that are inserted in parallel or in a Y configuration, with both tips ending in separate bifurcation limbs. The both stent delivery microcatheters require to be placed across the two targeted branches, and then one after another the stent retrievers are deployed at one point. Both stents are gradually retrieved out of the guiding catheter to optimize clot dislodgement. Klisch and colleagues, reports an increased median total recanalization time of 60 minutes (interquartile range 45 to 87 minutes) compared with standard mechanical thrombectomy using single-stent retrieval (range 20 to 48 minutes). Complete recanalization (TICI 2b/3) was achieved in 8 out of 10 cases, with good clinical outcomes (m Rankin Scale score of 2 or less) in 5 patients [2,7,8].

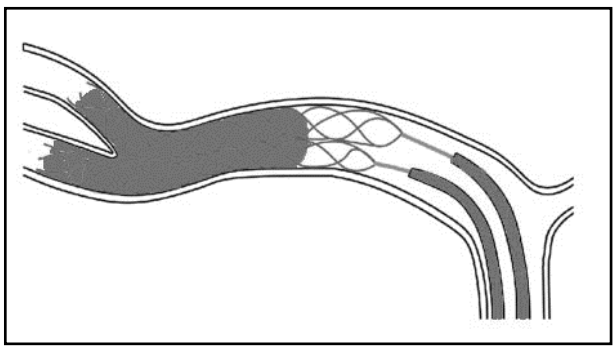


Figure 5: Dual stent retriever technique

COMBINED TREATMENT

Aspiration-Retriever Technique also sometime called "**Solumbra**" due to combination of solitaire stent retrieval and a penumbra aspiration catheter. The technique use a triaxial system which includes an aspiration catheter, a microcatheter and s microwire. The microcatheter over microwire is advanced through the thrombus and the stent retrieval is deployed across the embolus. Then, the aspiration distal access catheter is advanced close to the proximal end of the stent retrieval in close contact with the thrombus. If necessary the microcatheter can be retracted further to clear the tip of the aspiration catheter optimizing the integration with the thrombus. At the end, the stent retrieval is retracted while continuous negative suction is applied through a pump or manually with an aspiration syringe (Figure 6) [8,10].

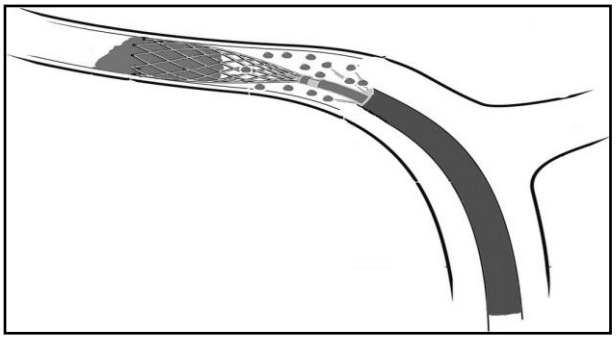


Figure 6: Aspiration-retriever technique

The **PROSPECT Technique** (proximal balloon occlusion together with direct thrombus aspiration during stent retriever thrombectomy) is combining the use of BGC with large bore distal aspiration catheters that are navigated to the face of the thrombus before retrieving the clot with a stent retriever device. The series of cases published in the literature regarding the use of the PROSPECT technique in the treatment of acute ischemic stroke have shown that the rate of thrombus fragmentations can even be further reduced and the rate of complete and particularly complete first-pass reperfusions can be further increased (Figure 7) [5,9].

The late version of this technique, called *PROTECT^{PLUS}* is characterized by the fact that the stent retriever loaded with the clot is not primarily retracted into the aspiration catheter but drawn into the BGC as a unit while aspiration is applied to both the aspiration catheter and the BGC. Maegerlein et all have published in 2018 a study showing that using the PROTECT^{PLUS} resulted in a higher rate of first pass complete reperfusions (59.4% vs. 27.7%, $p< 0.001$) as compared with PROTECT. The PROTECT^{PLUS} also led to shorter procedure times (21min vs. 37min, $p= 0.001$) and higher rates of overall complete reperfusion (73.5% vs. 49.5%, $p= 0.014$)[5,9,12].

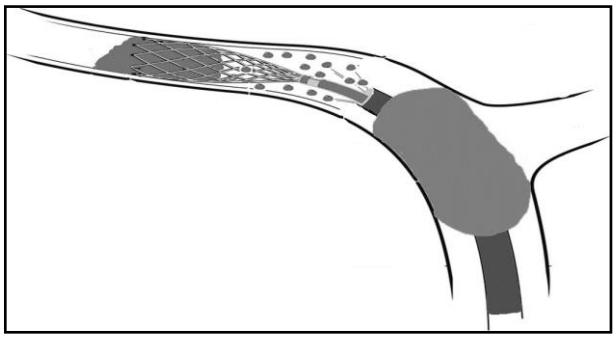


Figure 7: PROSPECT Technique

CONCLUSIONS

Mechanical thrombectomy approach in large vessel occlusion appears to be a promising method of endovascular stroke treatment, due to a significant increase in the rate of complete first-pass reperfusions and good clinical recovering. Mechanical thrombectomy using the combined techniques seems to be effective, fast and safe with high rates of near-complete and complete reperfusion.

REFERENCES

16. Alvarez MC., McCarthy DJ., Samir Sur, Snelling BM., Starke M. (2017) Comparing Mechanical Thrombectomy Techniques in the Treatment of Large Vessel Occlusion for Acute Ischemic Stroke, World Neurosurg. Apr;100:681-682.
17. Al-Bayati, A. R., Nogueira, R. G., Samaniego, E. A., & Haussen, D. C. (2019). Mechanical Thrombectomy: Techniques and Hybrid Approaches for Recanalization. In Acute Stroke Management in the Era of Thrombectomy (pp. 87-103). Springer, Cham.
18. Asadi H, Dowling R, Yan B, Wong S, Mitchell P. Advances in endovascular treatment of acute ischaemic stroke. Intern Med J. 2015;45:798–805.
19. Becker, K. J., & Brott, T. G. (2005). Approval of the MERCI clot retriever: a critical view. Stroke, 36(2), 400-403.
20. Chueh, J. Y., Kang, D. H., Kim, B. M., & Gounis, M. J. (2020). Role of Balloon Guide Catheter in Modern Endovascular Thrombectomy. Journal of Korean Neurosurgical Society, 63(1), 14–25. https://doi.org/10.3340/jkns.2019.0114

21. Haussen, D. C., Rebello, L. C., & Nogueira, R. G. (2015). Optimizating clot retrieval in acute stroke: the push and fluff technique for closed-cell stentrievers. Stroke, 46(10), 2838-2842.
22. Klisch J, Sychra V, Strasilla C, et al. Double Solitaire mechanical thrombectomy in acute stroke: Effective rescue strategy for refractory artery occlusions? AJNR Am J Neuroradiol 2015;36:552–6.
23. Lapergue, B., Labreuche, J., Blanc, R., Marnat, G., Consoli, A., Rodesch, G., ... & Duhamel, A. (2020). Combined use of contact aspiration and the stent retriever technique versus stent retriever alone for recanalization in acute cerebral infarction: the randomized ASTER 2 study protocol. Journal of NeuroInterventional Surgery.
24. Maegerlein, C., Berndt, M. T., Mönch, S., Kreiser, K., Boeckh-Behrens, T., Lehm, M., ... & Friedrich, B. (2018). Further development of combined techniques using stent retrievers, aspiration catheters and BGC. Clinical neuroradiology, 1-7.
25. Massari, F., Henninger, N., Lozano, J. D., Patel, A., Kuhn, A. L., Howk, M., ... & Wakhloo, A. K. (2016). ARTS (aspiration–retriever technique for stroke): initial clinical experience. Interventional Neuroradiology, 22(3), 325-332.
26. Munich, S. A., Vakharia, K., & Levy, E. I. (2019). Overview of mechanical thrombectomy techniques. Neurosurgery, 85(suppl_1), S60-S67.
27. Puri, A. S., & Parthasarathy, R. (2019). Aspiration Retriever Technique in Stroke (ARTS). In 100 Interesting Case Studies in Neurointervention: Tips and Tricks (pp. 359-362). Springer, Singapore.
28. Schramm, P., Navia, P., Papa, R., Zamarro, J., Tomasello, A., Weber, W., ... & Gralla, J. (2019). ADAPT technique with ACE68 and ACE64 reperfusion catheters in ischemic stroke treatment: results from the PROMISE study. Journal of neurointerventional surgery, 11(3), 226-231.

Romanian Neurosurgery (2020) XXXIV (1): pp. 52-57
DOI: 10.33962/roneuro-2020-027
www.journals.lapub.co.uk/index.php/roneurosurgery

The tumour volume influence on tumour recurrence and progression-free survival in the case of atypical meningiomas. Our experience on a series of 81 cases

A.I. Cucu[1], Claudia Florida Costea[1,2], Mihaela Dana Turliuc[1,2], Cristina Gena Dascalu[2], Ioana Jitaru[1], R. Dinu[1], Gabriela Dumitrescu[1], Anca Sava[1,2], B. Dobrovat[1,2], Camelia Bogdanici[2], T. Andrei[1], I. Stirban[1], I. Poeata[1,2]

[1] "Prof. Dr. N. Oblu" Emergency Clinical Hospital Iasi, ROMANIA
[2] "Grigore T. Popa" University of Medicine and Pharmacy, Iasi, ROMANIA

Keywords
atypical meningiomas,
tumour volume,
progression-free survival

Corresponding author:
Claudia Florida Costea

"Grigore T. Popa" University of Medicine and Pharmacy, Iasi, Romania

costea10@yahoo.com

ABSTRACT

Objective: The objective of our study was to evaluate a possible relation between the volume of atypical meningiomas (AMs) and the risk of tumour recurrence, as well as progression-free survival (PFS).

Material and methods: We evaluated 81 patients diagnosed with AMs (WHO grade II meningioma) who have undergone surgery at the "Prof. Dr. N. Oblu" Emergency Clinical Hospital Iasi between January 1, 2010, and December 31, 2019. The recorded data were demographic and imagistic (MRI, contrast-enhanced T1WI). We calculated the tumour volume prior to the surgery and evaluated the tumour recurrence using MRI at 12, 24, 36, 48 and 60 months after the surgery.

Results: 50.6% of patients had meningioma volume < 26.4 cm3. Women had larger tumour volumes than men (52.6%). Patients of age ≤ 60 years old, had tumour volumes > 26.4 cm3 in 58.5% of cases and meningiomas with volumes > 26.4 cm3 recurred earlier (p=0.010). Also, patients who had tumour volumes > 26.4 cm3, had a shorter PFS (40.976 months), compared to patients with tumour volumes < 26.4 cm3, who had better PFS (53.4 months).

Conclusions: the tumour volume of AMs > 26.4 cm3 represents a negative prognostic factor for both early tumour recurrence and reduced PFS.

Scan to access the online version

INTRODUCTION

Meningiomas are the most common primary intracranial tumors in adults and represent about one third of them (26). Out of the histopathological grades of meningiomas, AMs (WHO grade II meningiomas) represent approximately 20-30% of them (27, 28, 33), and their incidence has increased in the last years (10, 33).

Regarding the multiple prognostic factors of tumor recurrence in case of AMs (8, 12, 13, 27, 30, 31), some studies have reported that the size and volume of the tumor would also represent a prognostic factor (16, 17, 22). Thus, some authors have proved that larger size of AMs (for example, over 4.5 cm) are associated with early tumor recurrence (16). Other authors have reached the conclusion that the size of the meningioma is not only a prognostic factor of tumor recurrence, but also of the survival of patients, both in the case of AMs, as well as in the case of anaplastic meningiomas (17).

This study is aimed to evaluate the influence of the tumor volume on early tumor recurrence, as well as on the survival period up to the tumor recurrence.

Material and methods

We evaluated the tumor volume of 81 patients diagnosed with atypical meningioma (AM), who had undergone surgery at the Neurosurgery Department, "Prof. Dr. N. Oblu" Emergency Clinical Hospital Iasi, followed between January 1, 2010 and December 31, 2019. Each patient had the following recorded: demographic data regarding age and sex, and imaging studies (MRI). The tumor volume was calculated using the formula: volume = π /6 x legnth x wirdth x height (5, 18, 21, 29), and was analysed on magnetic resonance images prior to the surgery (contrast-enhanced T1WI). The mean tumor volume calculated was of 26.4 cm3, and depending on it, patients were grouped into two samples: (1) patients whose volume was < 24 cm3 and (2) patients whose volume was > 26.4 cm3. We also performed a qualitative evaluation of the AMs volume and evaluated the relation between the tumor volume, the rate of recurrence and PFS. The patients had an annual imaging examination (MRI), for a period of 5 years, and the tumor reccurence/tumor progression was defined as any contrast-enhancement at the level of the remaining tumor bed, or the increase in size of the the remnant tumor. In the cases of subtotal tumor resections, we named and classified the tumor progression as a tumor recurrence. Depending on its location in the intracranian space, the meningiomas were classified as: (1) skull base meningiomas, (2), convexity meningiomas, (3) parasagittal/falcine meningiomas, (4) posterior fossa meningiomas, and (5) intraventricular meningiomas. In the group of skull base meningiomas were included only those located at the level of the anterior and middle fossa. Posterior fossa meningiomas included all infratentorial meningiomas, including tentorium meningiomas or those located on the cerebellopontine angle or the petroclival junction. The statistical data processing was made in SPSS 24.0 (SPSS Inc., Chicago, IL). The data were characterized through descriptive statistics and frequency distributions. The data normality was checked using the Kolmogorov-Smironov fitting test; after this, we used t-Student and ANOVA tests to compare the samples of normally distributed data and Mann-Whitney and Kruskal-Wallis tests to compare the other samples. The qualitative data were characterized through frequency distributions and contingency tables, and the comparisons were made using the Chi-squared test. All p values were 2-tailed; a p value of 0.05 was considered significant. The actuarial data were represented with Kaplan-Meier plots, and the cumulative incidence curves were compared using the log-rank test. The study was approved by the Reasearch Ethics Committee of the "Grigore T. Popa" University of Medicine and Pharmacy of and by the Ethical Committee of the "Prof. Dr. N. Oblu" Emergency Clinical Hospital of Iași.

Results

Demography (age, sex)

The study group included 81 patients, of which most cases of AMs were in men, in a percentage of 53.1% (n = 43). The age of the patients in the total group ranged from 37 to 87 years, with a mean age of 61 years. When we evaluated the age of patients by sex, we noticed that women have a mean age of onset younger than men (58.42 years), compared to those who have a mean age of disease onset of 63.47 years (p=0.0052). 50.6% of patients had ages ≤ 60 years old (Figure 1). There were no statistically significant differences between the sexes in terms of age distribution. All patient characteristics can be seen in Table I.

Characteristics		n (%)
Gender	male	43 (53.1%)
	female	38 (46.9%)
Age	≤ 60 years	41 (50.6%)
	> 60 years	40 (49.4%)
Tumor localization	convexity	34 (42%)
	parasagittal/falcine	21 (25.9%)
	skull base	17 (21%)
	posterior fossa	6 (7.4%)

	intraventricular	3 (3.7%)
Tumor volume	< 26.4 cm^3	41 (50.6%)
	≥ 26.4 cm^3	40 (49.4%)

Table I. Characteristics of 81 patients with atypical meningiomas.

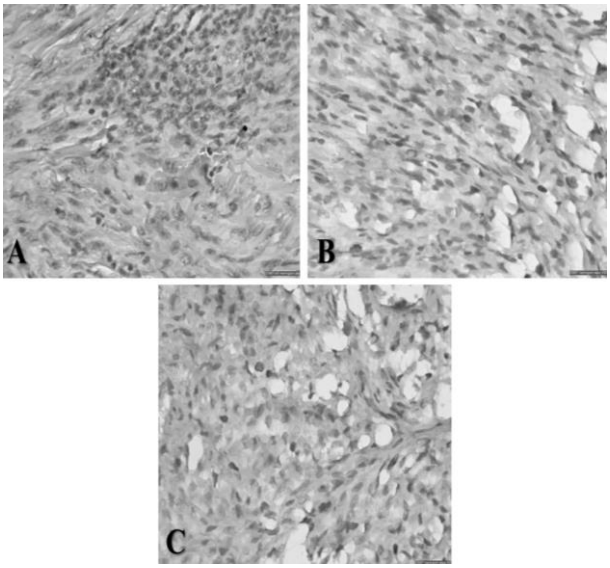


Figure 1. Female, 60 years-old with atypical meningioma. A. Well-cellularized tumor, infiltrative into adjacent dura mater, consisting of meningothelial cells arranged in syncytial pattern, with oval nuclei and fine granular chromatin (3 mitoses/10 high-power fields) (HE, x 400). B and C. two different fields of the same tumor showing high expression of Ki-67 LI (mean 12%) (immunohistochemical staining, x400).

Tumor localization

Regarding the localization of meningiomas at the skull level, most were located on the convexity level (42%, n=34), followed by parasagittal/falcine localization (25.9%) and at the level of the skull base (21%). Smaller percentages were located at the level of the posterior fossa (7.4%) or at the intraventricular level (3.7%) (Table I).

Following the qualitative analysis of the tumor volume, although there was no statistically significant difference between the tumor volume and the location of the meningioma at the level of the intracranial space, we found, however, that the largest tumors were located at the base of the skull, with a mean of 53.724 cm3 (ranging between 3.444-149.094 cm3). These were followed by intraventricular meningiomas (mean of 47.927 cm3), convexity meningiomas, (mean of 41.396 cm3), posterior fossa meningiomas (mean of 39.172 cm3) and those with parasagittal/falcine localization (mean of 36.596 cm3) (Figure 2).

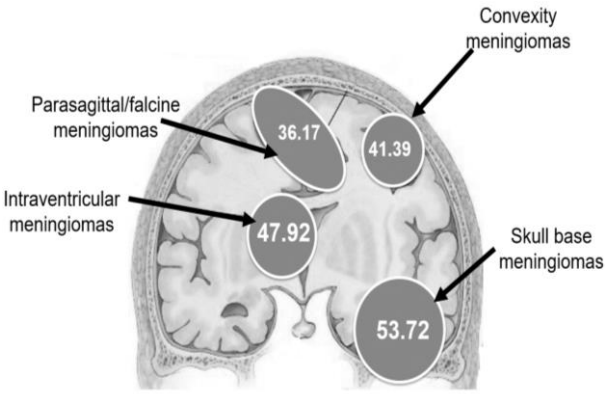


Figure 2. The mean tumor volume depending on location (personal collection of authors, public domain).

Tumor volume

The mean tumor volume was 26.4 cm3 and 50.6% of patients had meningioma volumes of < 26.4 cm3. Analyzing the differences between genres, although we did not identify any statistically significant values, we found that women had larger meningioma volumes than men (52.6%). Also, patients with ages ≤ 60 years old, had tumor volumes > 26.4 cm3 in 58.5% of cases.

We identified a statistically significant relation between the meningioma volume and the tumor recurrence (p=0.010). Tumors with volumes > 26.4 cm3 recurred earlier, and within this group the tumor recurrence rate was 17.1% at 12 months, 19.5% at 24 months and 41.5% at 60 months. On the other hand, tumors with volumes < 26.4 cm3 had no recurrence in the first 12 months, and the recurrence rate at 24 months was 5%. Moreover, 65% (n=26) of meningiomas with volumes < 26.4 cm3 had a slow recurrence, at 60 months. Also, recurrent meningiomas had a larger mean volume (49.438 ± 41.771) compared to meningiomas that did not recur (35.323 ± 35.524).

In regards to PFS, we identified a statistically significant relation between it and the tumor volume (p=0.030). Patients who had tumor volumes > 26.4 cm3 had shorter PFS (40.976 months). Patients who had tumor volumes < 26.4 cm3, had better PFS (53.4 months) (Figure 3).

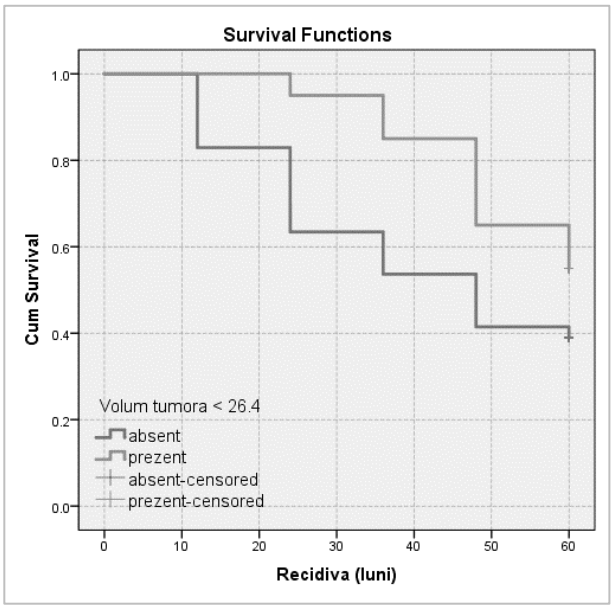


Figure 3. PFS depending on the tumor volume. It is notable that patients with tumor volume < 26.4 cm3 had better PFS.

Discussions

The influence of tumor volume on the tumor recurrence risk

In our study we observed a predominance of convexity meningiomas, in agreement with our previous studies (9, 11). We also noticed that a higher tumor volume has a negative influence on the rate of recurrence. Thus, in cases of meningiomas with tumor volumes > 26.4 cm3, the recurrence rate was higher than in the case of tumors with volumes below this value. This correlation between a larger size of the meningioma and the existence of the risk of tumor recurrence has also been observed by other authors in previous studies (16, 17, 19).

Fernandez et al. reported that the size of over 4.5 cm of AM is associated with a risk of early recurrence (16). Moreover, Garzon-Muvdi et al. also observed in his study that the size of the tumor can be considered an important factor not only for PFS, but also for overall survival. Nakasu et al. also reported the mean size of 4.4 ± 1.4 cm to influence tumor recurrence compared to non recurrent tumors which had a diameter of 3.5 ± 1.5 cm (25). Moreover, various authors reported that smaller sized AMs can represent a protective factor against tumor recurrence (4, 14, 16).

This relation between the larger tumor volume and the risk of tumor recurrence may be explained by the fact that a larger size meningioma makes a complete tumor resection more difficult due to the potential invasion of adjacent structures (3, 15, 20, 23).

Another interesting aspect was observed by Magill et al., who proved in a study conducted on 1113 meningiomas (905 grade I meningiomas and 208 grade II meningiomas) that the larger the size of the tumor, the higher the risks that it would be a grade II meningioma (24). In order to explain this, Magill et al. considered that there would be two possibilities: one would be that the grade II meningiomas grow faster than grade I meningiomas, and a second that once the slow growth tumor reaches a larger size, it develops a microenvironment due to hypoxia, which leads to the phenotype of this tumor becoming more aggressive (24).

In recent years, progress has been made in establishing the genetic factors that govern the growth of meningiomas or leading to their transformation into a more malignant histological grade, and in this sense, in addition to NF2, mutations in SMO, PI3K, TRAF7, KLF4 and AKTI have been identified (1, 6, 7). In the case of recurrent meningiomas, mutations in POLR2A have also been identified (7).

The influence of tumor volume on PFS

We found that the tumor volume of the meningioma influences the survival period until the tumor recurrence (p=0.030). Thus, tumor volumes > 26.4 cm3 had a shorter PFS. This can be explained by the fact that larger sized tumors invade more neuro-vascular structures, which make complete tumor resection more difficult, leaving a remnant tumor sometimes. In this sense, Wang et al. also found that tumors with dimensions > 41.5 mm are associated with a higher risk of tumor recurrence (32). Similarly, Nakasu et al. 1999 also reported that meningiomas with a mean diameter > 44 ± 14 mm have a significantly shorter PFS, consistent with other authors who consider that the tumor size is significantly associated with tumor recurrence in the case of patients diagnosed with AMs (2, 14, 17).

Conclusions

Our patients' series demonstrated that the tumor volume of AMs > 26.4 cm3 increases the tumor recurrence rate and decreases PFS. Moreover, mean tumor volumes had bigger values in case of recurrent AMs. We consider the tumor volume

represents a prognostic factor not only for tumor volume, but also for PFS.

References

1. Abedalthagafi M, Bi WL, Aizer AA, et al. Oncogenic PI3K mutations are as common as AKT1 and SMO mutations in meningioma. Neuro Oncol 2016; 18:649-55.
2. Aizer AA, Arvold ND, Catalano P, et al. Adjuvant radiation therapy, local recurrence, and the need for salvage therapy in atypical meningioma. Neuro Oncol 2014; 16(11):1547-53.
3. Buttrick S, Shah AH, Komotar RJ, Ivan ME. Management of atypical and anaplastic Meningiomas. Neurosurg Clin N Am 2016; 27(2):239-47.
4. Champeaux C, Houston D, Dunn L. Atypical meningioma. A study on recurrence and disease-specific survival. Neurochirurgie 2017; 63(4):273-81.
5. Char DH, Kroll S, Phillips TL. Uveal melanoma. Growth rate and prognosis. Arch Ophthalmol 1997; 115:1014-18
6. Clark VE, Erson-omay EZ, Serin A, et al. Genomic analysis of non-NF2 meningiomas reveals mutations in TRAF7, KLF4, AKT1, and SMO. Science 2016; 339:1077-80.
7. Clark VE, Harmancı AS, Bai H, et al. Recurrent somatic mutations in POLR2A define a distinct subset of meningiomas. Nat Genet 2016; 48:1253-59.
8. Cucu AI, Costea CF, Turliuc MD, et al. Are there any correlations between demographic characteristics, tumor location, and Ki-67 labeling index in intracranial atypical meningiomas (WHO grade II)? Rom J Morphol Embryol 2019; 60(2):567-72.
9. Cucu AI, Costea CF, Turliuc MD, et al. Anatomical localization of intracranial grade II meningiomas in North-Eastern Romania. Our 25-years experience. Rom Neurosurg 2019; 33(3):232-38.
10. Cucu AI, Costea CF, Carauleanu A, et al. Meningiomas related to the Chernobyl irradiation disaster in North-Eastern Romania between 1990 and 2015. Rev Chim (Bucharest). 2018; 69:1562-65.
11. Cucu AI, Costea CF, Poeată I, et al. Anatomical localization of atypical meningiomas: our experience on 81 patients. Med Surg J - Rev Med Chir Soc Med Nat Iaşi 2018; 122(4):744-52.
12. Cucu AI, Turliuc MD, Carauleanu A, et al. Chemical aspects of peritumoral cerebral edema in atypical meningiomas. Rev Chim (Bucharest), 2018; 69(10):2804-07.
13. Cucu AI, Costea CF, Poeata I, et al. Prognostic factors in atypical meningioma. Rom Neurosurg. 2017; 31(2):165-71.
14. Detti B, Scoccianti S, Di Cataldo V, et al. Atypical and malignant meningioma: outcome and prognostic factors in 68 irradiated patients. J Neurooncol. 2013; 115(3):421-27.
15. Durand A, Labrousse F, Jouvet A, et al. WHO grade II and III meningiomas: a study of prognostic factors. J Neurooncol 2009; 95(3):367-75.
16. Fernandez C, Nicholas MK, Engelhard HH, Slavin KV, Koshy M. An analysis of prognostic factors associated with recurrence in the treatment of atypical meningiomas Adv Radiat Oncol 2016; 1(2):89-93.
17. Garzon-Muvdi T, Yang W, Lim M, Brem H, Huang J. Atypical and anaplastic meningioma: outcomes in a population based study. J Neurooncol 2017; 133(2):321-30.
18. Gass JD. Comparison of uveal melanoma growth rates with mitotic index and mortality. Arch Ophthalmol 1985; 103:924-31.
19. Go KG, Kamman RL, Wilmink JT, Mooyaart EL. A study on peritumoural brain oedema around meningiomas by CT and MRI scanning. Acta Neurochir (Wien) 1993; 125:41-46.
20. Goyal LK, Suh JH, Mohan DS, et al. Local control and overall survival in atypical meningioma: a retrospective study. Int J Radiat Oncol Biol Phys 2000; 46(1):57-61.
21. Guthoff R. Modellmessungen zur Volumenbestimmung des malignen Aderhautmelanoms. Graefe Arch Klin Ophthalmol 1980; 214:139-46.
22. Hale AT, Wang L, Strother MK, Chambless LB. Differentiating meningioma grade by imaging features on magnetic resonance imaging. J Clin Neurosci 2018; 8:71-75.
23. Hanft S, Canoll P, Bruce JN. A review of malignant meningiomas: diagnosis, characteristics, and treatment. J Neurooncol 2010; 99(3):433-43.
24. Magill ST, Young JS, Chae R, et al. Relationship between tumor location, size, and WHO grade in meningioma. Neurosurg Focus 2018; 44(4):E4.
25. Nakasu S, Nakasu Y, Nakajima M, Matsuda M, Handa J. Preoperative identification of meningiomas that are highly likely to recur. J Neurosurg 1999; 90(3):455-62.
26. Ostrom QT, Gittleman H, Liao P, et al. CBTRUS Statistical Report: Primary brain and other central nervous system tumors diagnosed in the United States in 2010-2014. Neuro Oncol 2017; 19(suppl 5):v1-v88.
27. Pasquier D, Bijmolt S, Veninga T, Rezvoy N, Villa S et al. Atypical and malignant meningioma: outcome and prognostic factors in 119 irradiated patients. A multicenter, retrospective study of the Rare Cancer Network. Int J Radiat Oncol Biol Phys 2008; 71(5):1388-93.
28. Pearson BE, Markert JM, Fisher WS, et al. Hitting a moving target: evolution of a treatment paradigm for atypical meningiomas amid changing diagnostic criteria. Neurosurg Focus 2008; 24(5):E3.
29. Richtig E, Langmann G, Müllner K, Richtig G, Smolle J. Calculated tumour volume as a prognostic parameter for survival in choroidal melanomas. Eye (Lond) 2004; 18(6):619-23.
30. Ros-Sanjuan A, Iglesias-Moroño S, Carrasco-Brenes A, Bautista-Ojeda D, Arraez-Sanchez MA. Atypical Meningiomas: Histologic and Clinical Factors Associated With Recurrence. World Neurosurg 2019; 125:e248-e256.

31. Roser F, Nakamura M, Bellinzona M, et al. The prognostic value of progesterone receptor status in meningiomas. J Clin Pathol 2004; 57(10):1033-37.
32. Wang YC, Chuang CC, Wei KC, Hsu YH, Hsu PW et al. Skull base atypical meningioma: long term surgical outcome and prognostic factors. Clin Neurol Neurosurg 2015; 128:112-26.
33. Willis J, Smith C, Ironside JW, Erridge S, Whittle IR et al. The accuracy of meningioma grading: a 10-year retrospective audit. Neuropathol Appl Neurobiol 2005; 31(2):141-49.

Romanian Neurosurgery (2020) XXXIV (1): pp. 58-65
DOI: 10.33962/roneuro-2020-008
www.journals.lapub.co.uk/index.php/roneurosurgery

Puncture site bleeding complications in patients with Clopidogrel hyper-response. Three case reports

Nedeljković Žarko[1], Vukasinović Ivan[2], Majstorović Branisalva[3], Milosević Medenica Svetlana[2], Milićević Mihailo[1], Ilić Rosanda[1], Aleksić Vuk[1], Paunović Aleksandra[1], Stanimirovic Aleksandar[1], Scepanović Vuk[1], Milić Marina[1], Todorović Marko[1], Milisavljević Filip[1], Grujičić Danica[1]

[1] Clinic for Neurosurgery, Clinical Center of Serbia
[2] Center for Radiology and Magnetic Resonance, Clinical Center of Serbia
[3] Center for Anesthesiology, Clinical Center of Serbia

Keywords
intracranial aneurysms, dual antiplatelet therapy, Clopidogrel hyper-response

Corresponding author:
Vuk Aleksic

Clinic for Neurosurgery, Clinical Center of Serbia

aleksicvuk@hotmail.com

Abstract

Dual antiplatelet therapy (clopidogrel and acetylsalicylic acid) is a standard for the embolization of planned intracranial aneurysms with CNS stent due to the possibility of stent thrombus formation. All anti-aggregation drugs, including those listed, have bleeding as a side effect. Three patients with aneurysm had an elevated response to antiplatelet therapy with clopidogrel, which was confirmed by a multiplate test on the "VerifyNow" system. After reducing the dose of clopidogrel or after interrupting it, with the introduction of low molecular weight heparin for the duration of five days, aneurysms were successfully resolved by intracranial implantation of the stent. Perioperative angiograms and postoperative CT angiograms have verified hematomas at the place of punction of the femoral artery. Bleeding was resolved by the femoral artery suture by a vascular surgeon. All patients were discharged home without further complications and with dual antiplatelet therapy. By measuring the platelet function in vitro, the degree of inhibition of platelet activity achieved by the action of the drug can be assessed. A specific test can identify those patients who are highly responsive to the drug with increased platelet reactivity and the possibility of increased risk of bleeding. Our suggestion is to reduce the dosage of clopidogrel or to leave it out for 24 hours with preventive doses of low molecular weight heparin or to change the strategy of treatment of intracranial aneurysm, i.e. avoiding implantation of CNS stent.

Scan to access the online version

Introduction

According to statistics, about 5% of the population has intracranial aneurysm, out of which 1 to 2% suffer from aneurysm rupture with a dramatic clinical manifestations and consequences [1,2,3].Today, it is

believed that endovascular treatment is a better, more modern and less aggressive treatment associated with less complications and lower mortality, shorter length of hospital stay and lower costs of treatment compared to conventional neurosurgical treatment, i.e. microsurgical clipping of aneurysms [4,5,6,7,8]. The current standard of preoperative preparation for stent placement in aneurysms involves dual antiplatelet therapy with acetylsalicylic acid and clopidogrel.Antiplatelet drugs inhibit platelet aggregation and thus stop the formation of thrombus. Thrombus formation is a complex mechanism of responses to vascular changes, injuries, atherosclerosis and other diseases.The data indicate that in addition to the effect of these drugs, other local and systemic factors (diabetes mellitus, left ventricular hypertrophy, atrial fibrillation) may be the cause of unwanted ischemic events [9,10]. Other antiplatelet drugs: prasugrel, dipyridamole, cilostazol, cangrelor and ticagrelor have not been analyzed so far, andtherefore are the subject of recent researches that are yet to define the benefits of their application in therapy.

CASE 1

Pre-procedural angiograms showed the condition of 59-year-old patient, after the coiling of ACI aneurysms on both sides. From the medical documentation, it is learned that the patient's embolization of aneurysms was done on both internal carotid arteries in 2016. Hematological parameters on admision: RBC 4,75x10 ^{12} /L, Hgb 138 g/L, Hct 0,45 L/L. Due to the impaction of the coilling to the right internal carotid artery, placement of the stent was planned, therefore, three days before the intervention, the patient was administered clopidogrel75 mg twice daily and acetylsalicylic acid 100 mg once a day. Drug-resistence testing on the „VerifyNow" device showed an increased sensitivity of platelet activity to the action of clopidogrel. Clopidogrel was discontinued for fivedays and acetylsalicylic acid was continued. During this period, a low molecular weight heparin is administered at a dose of 0,3 ml twice a day, and then it was proceeded to the procedure. By accessing the left femoral artery, the Flow diverting Silc 3x20mm stent is placed over the aneurysm`s neck on the right internal carotid artery, which causes the pathway obstruction of the contrast agent in aneurysm. During puncture there was perforation of the left femoral artery and extravasation of the contrast medium with the expansion pathway caudally. The intervention was interrupted, the puncture site waspressed with long-term compression in groin. The patient complained of pain in the left side of abdomen. Physical examination found palpatory pain sensation of the abdomen and drop in blood pressure.CT angiography of the leg and abdomen showed the extravasation of the contrast agent at the level of the left femoral artery with expansion towards the retroperitoneum.[Figure 1, Figure 2].

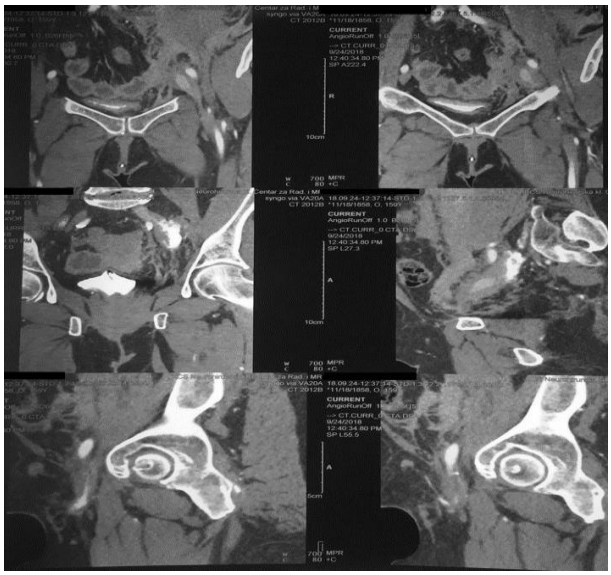


Figure 1. Angiographic presentation of aneurysms and CT angiographic exploration of bleeding at the puncture site.

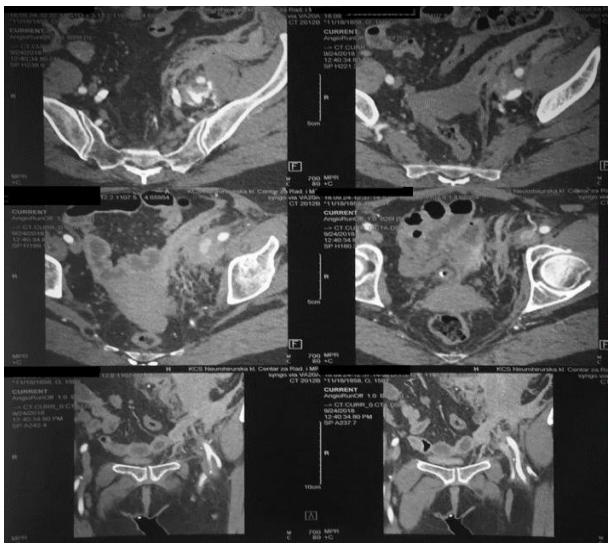


Figure 2. Angiographic presentation of aneurysms and CT angiographic exploration of bleeding at the puncture site.

The vascular surgeon was called for consultation. The patient was transferred and operated in the Clinic for vascular and endovascular surgery of Clincal center of Serbia where hematoma was evacuated and suture of left femoral artery was performed. Hematological parameters two days after femoral artery puncture: RBC 2,56x10 ^{12} /L, Hgb 80 g/L, Hct 0,25 L/L. In the postoperative course, the patient received fraxiparine at a dose of 0,3 ml twice daily for the next eight days. After leaving the hospital, dual antiplatelet therapy (clopidogrel and acetylsalicylic acid) was continued for the next three months once a day, followed by acetylsalicylic acid onwards.

Case 2

A 45-year-old female patient was previously planned for the placementof an intracranial stent due to an aneurysm on anterior communicating artery. She was taking clopidogrel 75 mg twice a day and acetylsalicylic acid 100 mg once a day for three days. Hematological parameters on admision: RBC 4,13x10 ^{12} /L, Hgb 128 g/L, Hct 0,409 L/L. Drug-resistence testing on the „VerifyNow" device showed an increased sensitivity of platelets to the action of clopidogrel. Administration of clopidogrel was stopped for the next five days, it was continued with the administration of acetylsalicylic acid 100 mg once a day. Also, 0,3 ml of fraxiparin was administered twice a day, and then it was proceeded with procedure. Through the right femoral artery, the guiding catheter was placed in the left internal carotid artery, the microcatheter was positioned in the aneurysm which was excluded from the circulation by placing the coil. As aneurysm was resolved by a coil, the dual antiaggregation therapy was interrupted in the postoperative course. In the early postprocedural period, hematoma formation in the groin region occured, and therefore changes in the complete blood count: RBC2,87x10^{12}/L, Hgb 91 g/L, Hct 0,264 L/L. The patient complained of pain in the lower right quadrant of the abdomen in the area above the right inguinal ligament. A vascular surgeon was consulted, after which a patient was admitted to the Clinic for vascular and endovascular surgeryof Clinical center of Serbia. CT angiography of the leg and abdomen was performed, which showed the presence of a lesion in the area of the transition of the external iliac to the right common femoral artery with extravasation of contrast.[Figure3, Figure4, Figure5] Due to active retroperitoneal bleeding, conciliar decision was to undergo emergency surgical treatment. The patient was postoperative hemodynamically stable with palpable pedal pulses.

Figure 3. CT angiography of the leg and abdomen showing the presence of a lesion in the area of the transition of the external iliac to the right common femoral artery with extravasation of contrast.

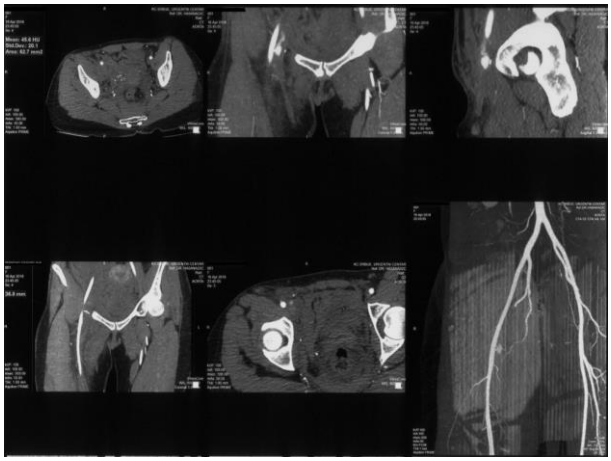


Figure 4. CT angiography of the leg and abdomen showing the presence of a lesion in the area of the transition of the external iliac to the right common femoral artery with extravasation of contrast.

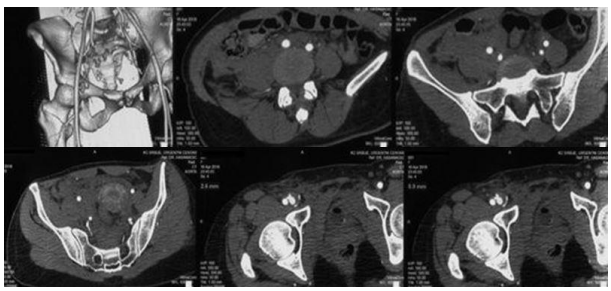


Figure 5. CT angiography of the leg and abdomen showing the presence of a lesion in the area of the transition of the external

iliac to the right common femoral artery with extravasation of contrast.

CASE 3

A 47-year-old female patient was admitted to perform an endovascular treatment due to the presence of aneurysm on the supraclinoid segment of the left internal carotid artery, which was randomly detected as part of the examination of pituitary microadenoma. The width of aneurysm neck was 6,5 mm. The patient was planned for placing of the flow diverting stent over the aneurysm neck. In the preprocedural period, the patient received dual antiplatelet therapy (clopidogrel 75 mg twice a day and acetylsalicylic acid 100 mg once a day) for three days. Drug-resistence testing on the „VerifyNow" device showed an increased platelet sensitivity to clopidogrel. An aneurysm embolization was attempted on two occasions. During the second intervention, placement of microcatheter was attempted distal from the aneurysm neck in order to place a "flow-diverting" stent, which failed even after several attempts, and as a result, the procedure was interrupted. In the early postprocedural course, the condition of the patient worsened in terms of confusion, paleness, hypotension, and the presence of a larger subcutaneous hematoma in the right groin. CT angiography of the leg and abdomen was performed, which showed extravasation of contrast at the puncture site [Figure 6, Figure 7, Figure 8,Figure 9, Figure 10]. The patient was transferred to the Clinic for vascular and endovascular surgery of Clinical center of Serbia where the suture of right external iliac artery and evacuation of hematoma were performed. During the third hospitalization, the patient was again prepared for stent placement with dual antiplatelet therapy, and the response to clopidogrel was again too strong. This time, it was also attempted to place the stent through the aneurysm neck, but while the microwirewas passing, further distal from the branch of the left middle cerebral artery, extravasation of contrast occured. Protamine sulfate was administered by venous route, which is an antidote for intravenous administration of heparin and the contrast extravasation was no longer shown. It was attempted again to go through with a microcateter distally from the neck of aneurysm, but even after additional attempts, it failed due to extremely unfavorable angioarchitecture.Subsequently, the microcatheter was used to access the fundus of aneurysm and to place the "pCONUS" stent, and then, after placement of several coils, aneurysm was excluded from the circulation with less residual filling and preserved passage of all blood vessels. Postprocedurally, the patient was somnolent, slowed down, intubated andwith progressive deterioration of consciousness. There was an acute hydrocephalus occurrence that was taken cared by the external drainage of the cerebrospinal fluid [Figure10]. On this occasion, a subdural hematoma was formed, which gradually spontaneously absorbed. Ten days after embolization, a permanent drainage system, ventriculoperitoneal shunt, was placed. It was continued with the administration of clopidogrel 75mg and acetylsalicylic acid 100mg once a day for three months, and then only acetylsalicylic acid 100mg going forward. There has been a recovery in the general and neurological condition of the patient with a residual right-sided weakness [Figure 11].

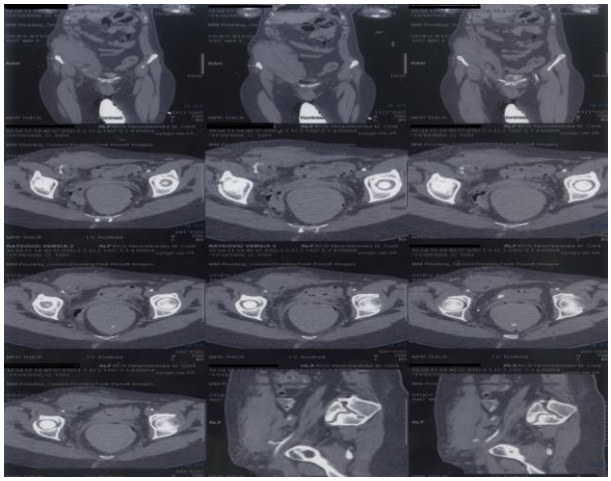


Figure 6. CT angiography of lower extremities et pelvis showing extravasation of contrast at the puncture site.

Figure 7. CT angiography of lower extremities et pelvis showing extravasation of contrast at the puncture site.

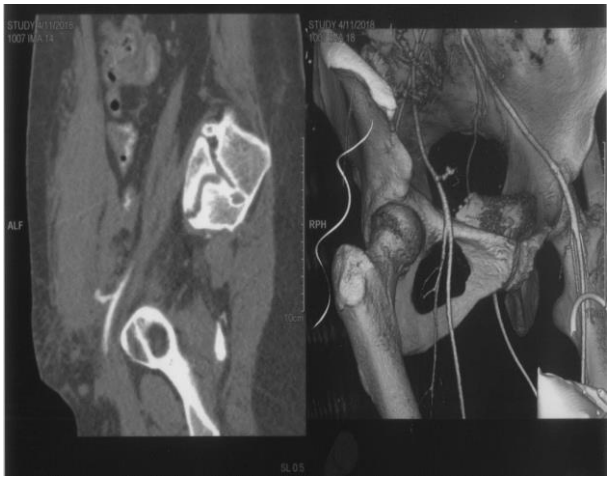


Figure 8. CT angiography of lower extremities et pelvis showing extravasation of contrast at the puncture site.

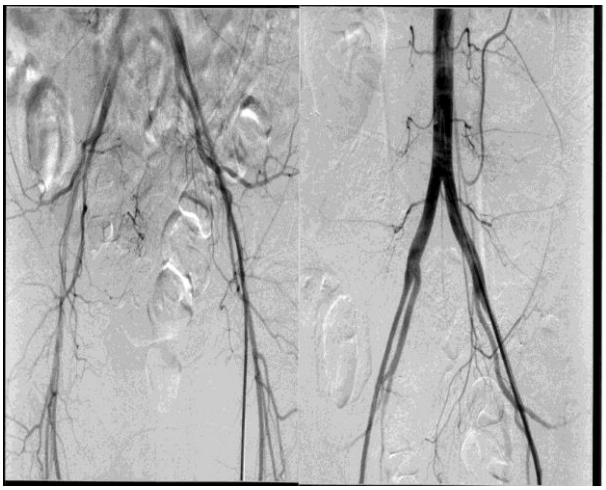


Figure 9. CT angiography of lower extremities et pelvis showing extravasation of contrast at the puncture site.

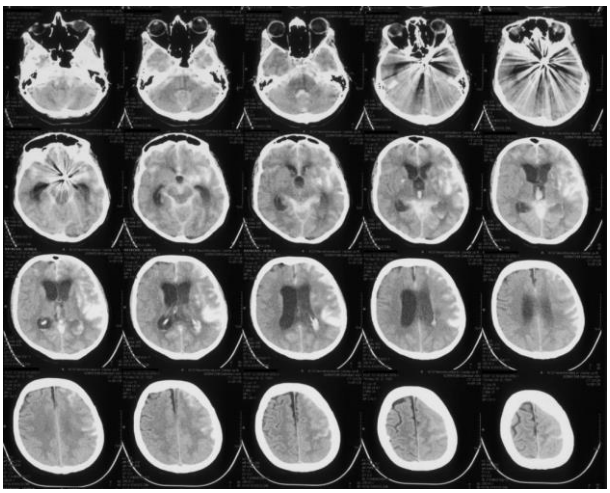


Figure 10. Head CT scan showing the occurance of acute hydrocephalus.

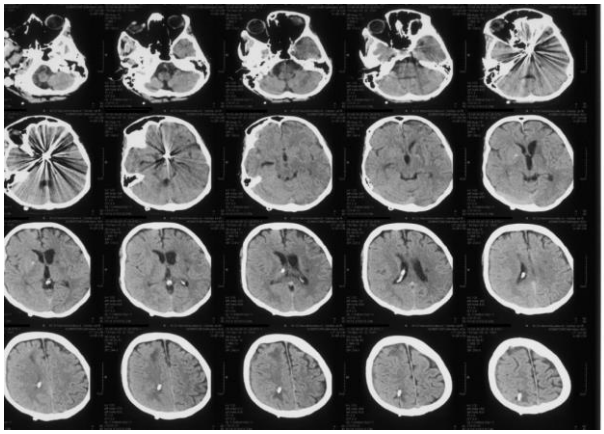


Figure 11. Control head CT scan 1 month after the embolisation of aneurysm and VP shunt placement.

Discussion

Neuroradiological procedures, such as embolization of intracranial aneurysms, have been routinely applied since their implementation in 1990 to the present day. They are performed in general anesthesia for better control of pain and immobility of patients, control of local blood flow at the site of vascular changes via mean arterial pressure, with the use of a continuous or bolus dose of heparin by the venous line. Dual antiplatelet therapy is used pre- and post-procedually in cases of stent implatantion.In principle, anticoagulant or antiplatelet therapy is widely used in various areas of medicine to prevent the development of occlusive thrombosis. When it comes to drugs, the prevention is achieved by changing the coagulation of the blood. Coagulation of the blood, composed of a series of proteolytic reactions, is a complex cascade processof which the ultimate result is the creation of insoluble fibrin.

The platelet function is regulated by three types of compounds. The first group consists of compounds that are formed outside of the platelet and act on receptors located on the platelet membrane: catecholamines, collagen, thrombin, prostacyclin. The second group of compounds consists of compounds that form in the platelet itself but act on membrane receptors: ADP, prostaglandins D2 and E2, and serotonin. The third group of compounds are produced byplatelet, but they act in the platelet itself: thromboxane A2, cyclic AMP and GMP.

In clinical practice, acetylsalicylic acid and clopidogrel are generally available. Because of the high resistance to clopidogrel antiplatelet activity

(16%) measured by the same in vitro platelet function tests, a full range of drugs is available. Current studies evaluate the efficacy of P2Y12 receptor inhibitors: prasugrel, ticagrelor, cangrelor, elinogrel; then prothrombin antagonists, glycoprotein inhibitors, phosphodiesterases, and others.

A response to antiplatelet therapy with clopidogrel confirmed by drug-resistence testing on the „VerifyNow" device. Verifynow System (Accumetrics Inc., San Diego, California, USA) monitors the platelet aggregation by agglutination of human-fibrinogen-coated granules, which is triggered by activated platelets stimulated by agonists in citrate full blood. This system measures induced platelet aggregation by ADP and prostaglandin E1, following an increase in light transmission (P2Y12) test. It uses an iso-thrombin receptor activating peptide (iso-TRAP) that activates platelets regardless of the action of clopidogrel. The results are expressed in the PRU (platelet reactivity unit) units showing the degree of aggregation mediated by P2Y12 receptors. [11,12,13]

Acetylsalicylic acid irreversibly inhibits the COX-1 enzyme, then the synthesis of cyclic endoperoxides and all prostaglandins that result therefrom. The COX-1 inhibition inhibits the synthesis of thromboxane, and then prostacyclin and prostaglandin E2. Due to irreversible enzyme inhibition, once daily dosing of the drug is sufficient. There are data indicating that at low doses of 100 mg per day, the biosynthesis of thromboxane can be selectively inhibited without any influence on prostacyclyin synthesis. As a result, small doses of acetylsalicylic acid restrict the therapeutic effect of prostaglandins that have a significant role in a large number of physiological and pathophysiological processes in the body (regulation of arterial blood pressure, renal function or interaction with antihypertensive effect of diuretic, and angiotensin-converting enzyme, etc.) [14,15,16]

Simultaneous administration of analgesic drugs from a group of non-selective COX1 inhibitors, such as ibuprofen and naproxen, can lead to a reduction in acetylsalicylic acid by competing for the same receptor [17,18] in the preoperative and postoperative period.

Clopidogrel is a drug whose metabolite is a platelet aggregation inhibitor. To switch to an active metabolite that inhibits platelet aggregation, clopidogrel is metabolized by the CYP450 enzyme. The active metabolite of clopidogrel selectively inhibits adenosine-diphosphate adhesion (ADP) for its P2Y12 receptors on platelets and in this way, via ADP-mediated activation of the IIb/IIIa glycoprotein complex, inhibits platelet aggregation. ADP binds to delta granulomas in platelets. ADP achieves its effect by binding to its platelet receptors and leads to: inhibition of adenylate cyclase by stimulating Gi protein and decreasing the concentration of intracellular cAMP, the formation of inositol triphosphate (IP3) leading to the mobilization of calcium from cell depots, the release of arachidonic acid leading to the creation of TXA2 and activation phospholipase A2. It is believed that ADP exhibits its effect by binding to three purinergic receptors: P2Y1, P2Y12, and P2X1. It is believed that P2Y12 plays a key role in the pathogenesis of arterial thrombus, mediating in ADP-induced platelet aggregation, potentiating the secretion of granules under the action of agonists and potentiating the inhibition of the antithrombotic effect of natural regulators of the platelet function, prostacyclin. P2Y12 is associated with Gi2 protein whose stimulation leads to the inhibition of adenylate cyclase and the activation of phosphatidylinositol 3 kinase (PI3K). Since this receptor is considered to have a central role in the formation of a coagulum, it is a target molecule for antithrombotic therapy. Due to irreversible binding, the effect of inhibition on the affected platelets lasts for the entire duration of their plasma life (about 7-10 days), and re-establishment of platelet function occurs after plasma exchange time [19]. This drug was approved in 1997 and after showing similar antithrombotic efficacy, but also as a significantly safer medication for patients, it completely replaced ticlopidine. For those patients with a history of stroke, it is known that antithrombotic therapy reduces incidence of re-ischemic events up to 22%. Clopidogrel proved superior to aspirin in reducing risk and was recommended for secondary prevention of stroke.

Due to irreversible inhibition of P2Y12 receptor, the effect of clopidogrel is present during the life of platelets (7-10 days). Cell recovery is expected in the period of 3 to 5 days, that is, the platelets are fully functional on average for 7 days after taking the last dose of the drug. The maximum inhibition of the P2Y12 receptor was achieved after 4-5 days of daily administration of 75 mg of clopidogrel. This interval

can be reduced to 3-5 hours by taking aloading dose of 300-600mg. Thus, in addition to the proven antithrombotic effect, clopidogrel has its disadvantages: its antithrombotic effect is delayed due to the necessary metabolism and activation of the prolapse, as well as interindividual variability in the therapeutic response to the drug.

The CYP enzyme system is responsible for 40-80% metabolism of almost all drugs in the active metabolite. Since clopidogrel and lipophilic statins are metabolised via the CYP3A4/3A5 enzyme system, the inhibition of the formation of the active metabolite of clopidogrel and unwanted thrombosis can occur [14,15]. Observations and randomized clinical studies have shown that the concomitant use of clopidogrel and proton pump inhibitors for pre-operative purposes, and due to gastric irritation, can lead to a reduction in the anti-aggregation effect and an increase in the number of repeated thromboses or ischemic events [20]. Dual anti-aggregation therapy is used to prevent the formation of thrombus or stent thrombosis. However, antiaggregation drugs can cause bleeding as an adverse drug reaction [21,22]. In the literature there are few studies that investigate increased sensitivity of platelets to the action of clopidogrel as opposed to postprocedural bleeding aneurysms [23]. For this reason, we listed 3 clinical cases with various causes of bleeding at the puncture site.

We believe that preprocedural management procedures should be acceptable in order to solve the resulting complication and complete the procedure. We summarized the mechanism of action, the current clinical assessment, and the standards for the use of antiagregating drugs in the pre-procedural, procedural and postprocedural period for the treatment of unruptured aneurysms.

N	Sex	Age	Aneurysm
N[1]	F	59	ACI bill.
N[2]	F	45	ACoA
N[3]	F	47	ACI l.sin.

Table 1. Anthropological characteristics of the patients.

N	Clopidogrel bleeding<95-208<thrombosis	Acetylsalicylic acidbleeding<350-549<thrombosis
N[1]	3	359
N[2]	39	525
N[3]	14	370

Table 2. Results of the resistance test for clopidogrel and acetylsalicylic acid.

References

1. Samardžić M, Grujičić D, Prstojević B. Spontanaintrakranijalnakrvarenja, U: Samardžić M, Ur. Hitnastanja u neurohirurgiji. Beograd, I.P. Obeležja;2010.
2. Maud A, Lakshminarayan K, Suri MF, Vazguez G, Lanzino G, Qureshi A. Cost-effectiveness analysis of endovascular versus neurosurgical treatment for ruptured intracranial aneurysms in the United States. J Neurosurg 2009;110(5):880 – 6.
3. Greving JP, Rinkel GJ, Buskens E, Algra A. Cost-effectiveness of preventive treatment of intracranial aneurysms: new data and uncertainties. Neurology 2009;73(4):258 – 65.
4. Wolstenholme J, Rivero-Arias O, Gray A. Treatmant Pathways, Resource Use, and Cost of Endovascular Coiling Versus Surgical Clipping After SAH. Stroke 2008;39:111 – 9.
5. Higashida RT, Lahue BJ, Torbey MT, Hopkings LN, Leip E, Hanley DF. Treatment of unruptured intracranial aneurysms: a nationwide assessment of effectiveness. AJNR Am J Neuroradiol 2007;28(1):146 – 51.
6. Varma MK, Price K, Jayakrishnan V, Manickam B, Kessell G. Anaesthetic considerations for interventational neuroradiology. BJA 2007;99(1):75 – 85.
7. Labalette C, Houdart E, David S. Embolization of intracranial aneurysms: reimbursement and perspectives. J Radiol 2010;91:895 – 900.
8. Majstorović BM, Milošević-Medenica SB, MilakovićBD,Naumović TS. New Interventional Neuroradiological Procedures in Serbia – The Direct Costs of Classical Neurosurgical Intervention in Relation to the Endovascular Treatment of Intracranial Aneurysms. Hospital Pharmacology. 2015;2(2):272-278.
9. JenningsLK.Role of platelets in atherothrombosis. Am J Cardiol2009;103:4A–10A.
10. Bhatt DL. Role of antiplatelet therapy across the spectrum of patients with coronary artery disease. Am J Cardiol 2009;103:11A–9A.
11. Prinz V, Finger T, Bayerl S, Rosenthal C, Wolf S, Liman T, VajkoczyP.High prevalence of pharmacologically induced platelet dysfunction in the acute setting of brain injury. Acta Neurochir (Wien) 2016;158(1):117-23.
12. Flechtenmacher N, Kämmerer F, Dittmer R, Budde U, Michels P, Röther J, Eckert B. Clopidogrel Resistance in Neurovascular Stenting: Correlations between Light Transmission Aggregometry, VerifyNow, and the Multiplate. AJNR Am J Neuroradiol. 2015;36(10):1953-8.
13. Jastrzębska M, Chełstowski K, Wódecka A, Siennicka A, Clark J, NowackiP.Factorsinfluencingmultiplate whole blood impedance platelet aggregometry measurements, during aspirin treatment in acute ischemic stroke: a pilot study. BloodCoagul Fibrinolysis 2013;24(8):830-8
14. Hall R, Mazer CD. Antiplatelet drugs: a review of their

pharmacology and management in the perioperative period.AnesthAnalg 2011;112(2):292-318.
15. Benedek IH, Joshi AS, Pieniaszek HJ, King SY, Kornhauser DM. Variability in the pharmacokinetics and pharmacodynamics of low dose aspirin in healthy male volunteers. J ClinPharmacol 1995;35:1181–6.
16. Teo KK, Yusuf S, Pfeffer M, Torp-Pedersen C, Kober L, Hall A, Pogue J, Latini R, Collins R; ACE Inhibitors Collaborative Group. Effects of long-term treatment with angiotensin-converting-enzyme inhibitors in the presence or absence of aspirin: a systematic review. Lancet. 2002;360(9339):1037-43.
17. Václavík J, TáborskýM. Antiplatelet therapy in the perioperative period. Eur J Intern Med. 2011 Feb;22(1):26-31.
18. Catella-Lawson F, ReillyMP, Kapoor SC, Cucchiara AJ, DeMarco S, Tournier B et al. Cyclooxygenase inhibitors and the antiplatelet effects of aspirin. N Engl J Med 2001;345:1809-15.
19. Matula JM, Lesiak M, Michalak M, Kowal J, Mączyński M, Siniawski A, Mularek-Kubzdela T, Wiśniewski S, Grajek S. Association between high on-treatment platelet reactivity and occurrence of cerebral ischemic events in patients undergoing percutaneous coronary intervention. Thromb Res. 2016;138:49-54.
20. Saw J, Brennan DM, Steinhubl SR, Bhatt DL, Mak KH, Fox K. Lack of evidence of a clopidogrel-statin interaction in the CHARISMA trial. J Am Coll Cardiol 2007;50:291–5.
21. Trenk D, Hochholzer W, Frundi D, Stratz C, Valina CM et al. Impact of cytochrome P450 3A4-metabolized statins on the antiplatelet effect of a 600-mg loading dose clopidogrel and on clinical outcome in patients undergoing elective coronary stent placement. Thromb Haemost 2008;99:174–81.
22. Ho PM, Maddox TM, Wang L, Fihn SD, Jesse RL, Peterson ED et al. Risk of adverse outcomes associated with concomitant use of clopidogrel and proton pump inhibitors following acute coronary syndrome. JAMA 2009;301:937–44.
23. Kim MS, Park ES, Park JB, Lyo IU, Sim HB, Kwon SC. Clopidogrel Response Variability in Unruptured Intracranial Aneurysm Patients Treated with Stent-Assisted Endovascular Coil Embolization : Is Follow-Up Clopidogrel Response Test Necessary? J Korean Neurosurg Soc. 2018 Mar;61(2):201-211.

Romanian Neurosurgery (2020) XXXIV (1): pp. 66-76
DOI: 10.33962/roneuro-2020-009
www.journals.lapub.co.uk/index.php/roneurosurgery

Risk factors and clinical and neurological consequences of intraoperative rupture of brain aneurysms in microsurgical operations

Bindu A. V., Orlov M. Yu., Litvak S. O., M. V. Yeleynik

Department of neurosurgical pathology of vessels of the head and neck SI "Academician A.P." Romodanov Institute of neurosurgery of NAMS of Ukraine", Kyiv, UKRAINE

Keywords
AA-arterial aneurysm, IOC-intraoperative complications, SAH-subarachnoid haemorrhage, IORA-intraoperative rupture of aneurysm

Corresponding author:
Arthur Vasylovych Bindu

"Academician A.P. Romodanov" Institute of neurosurgery of NAMS of Ukraine

artur.bindiu@gmail.com

ABSTRACT

Objective. to assess the frequency, risk factors and clinical and neurological consequences of intraoperative rupture of arterial aneurysm (AA) of the brain (B) in clipping operations of the B AA.

Materials and approaches. A retrospective analysis of microsurgical operations clipping of cerebral aneurysms in 1449 (100%) patients for the period from 2011 to 2018 was performed, of which 141 (9.73%) cases had intraoperative rupture of the aneurysm (IORA). Preoperative examination: clinical and neurological examination, CT of the brain, cerebral angiography (CAG), duplex scanning of the main vessels of the head and neck. The analyzed criteria are risk factors of IORA: AA size, localization, shape, duration of surgery after the primary rupture of AA, the presence of hypertension and the patient's condition before surgery.

Results. The frequency of IORA in clipping operations of B AA was 9.73% (141 patients) in a series of observations 1441 (100%). Most often IORA-141 (100%) was registered in clipping operations of AA of complex ACA-AcomA (86 (61%) cases out of 141 (100%)). IORA is possible at all stages of the operation with the maximum frequency of contact breaks – 135 (95.74%); the rarest-6 (4.26%) - non - contact IORA (at the stage of craniotomy) was recorded. At the preoperative stage, the vast majority of patients with subsequent IORA were diagnosed with cerebral edema, AA of large size, atherosclerotic changes in the aneurysm-affected segment of the artery and cervical areas of the aneurysm, high blood pressure during surgery, adhesive arachnoid changes. At the time of discharge from the hospital, according to the Glasgow results scale: 69 (48.94%) full or partial restoration of labor activity, 18 (12.77%) had limited daily activities without the need for outside assistance, 37 (26 24%) deep disability) Deaths were in the group of "contact" IORA - 17 (12.06%). At 6 (4.26%) of "non-contact" IORA, a deepening of initial neurological symptoms was recorded with a suppression of the level of consciousness, the addition of pyramidal insufficiency, speech impairment and psycho-organic syndrome, and a deepening of the phenomena of initial cerebral arterial vasospasm.

Conclusions. IORA is predominantly in contact with a frequency of occurrence-9.73 %. The most common risk factors for IORA were: cerebral edema, large AA, atherosclerotic changes in the aneurysm-affected artery segment and cervical

Scan to access the online version

aneurysm sites, high blood pressure during surgery, adhesions arachnoid changes. IORA leads to deepening of initial neurological symptoms, phenomena of initial vasospasm of cerebral arteries with the level of total mortality-17 (12.06%).

INTRODUCTION. ESSAYS ON THE HISTORY OF BRAIN ANEURYSM SURGERY

The first description of cerebral aneurysm belongs to Weissmann in May 1676. It took two centuries until Hodson (1815) and Bramwell (1886) established that the extravasation of blood into the subarachnoid space is a consequence of the rupture of an intracranial aneurysm and called this form of "subarachnoid" hemorrhages (HSA)[22].

The start of brain aneurysm surgery is provided by the Scottish neurosurgeon Dot, who in 1933 published two cases of microsurgical exclusion of a cerebral aneurysm from the bloodstream by enveloping the aneurysm with a piece of muscle [1]. On March 23, 1937, Walter Dandy first excluded a SAC-shaped aneurysm of the right internal carotid artery from the bloodstream by clipping the neck of the aneurysm with a metal clip to a 43-year-old patient. In 1944 Walter Dandy published the first monograph devoted to the surgical treatment of brain aneurysms, in which the author publishes the results of surgical treatment of 107 patients with various localisations of aneurysms, thereby demonstrating that patients with such a complex pathology necessarily require surgery [3].

Nowadays microsurgical and endovascular approaches are used in aneurysm surgery (Konovalov A. N., 1973; Oleshkevich F. V., 1973; Zozulya Yu. A., 1986; Romodanov A. P. et al., 1990; Shcheglov V. I., 1998; Serbinenko F. A., 1971; Suzuki J., 1979; Yasargil GM, 1984).

The first method of surgical treatment of brain aneurysms was carotid artery ligation, which was used by A. Pare (1510-1590) as a method of stopping arterial bleeding in victims with neck wounds. For decades, it was the only possible method of surgical treatment of AA, which at the end of the XIX century. widely used by the founders of neurosurgery (Horsley, 1891; Shing, 1911). The latter described AA as random finds of " cysts that pulsate."

In 1960, removable clips, which are widely used in AA surgery were proposed. In the 1970s, neurosurgery began to use the operating microscope. Given the rapid development of cerebrovascular neurosurgery, in particular microsurgical clipping of brain aneurysms, in the 1980s and 1990s, a large number of publications appeared on the problem of intraoperative complications (IOC), including intraoperative ruptures of aneurysms (IORA), with clipping of cerebral aneurysms [1].

But despite the historical development and achievements of modern neurosurgery, cerebral IORA remains the most frequent IOC [2,3,4] . There are contact and non-contact IORA. Contact include IORA, which occur during direct mechanical action on the aneurysm: during the retraction of the medulla with a spatula, during arachnoid dissection, clipping the neck of a sacculated aneurysm. IORA, which occurs in the early stages of an operation, is considered non-contact, prior to the dissection of the dura mater (DM), when there is no mechanical effect on the aneurysm (when shifting the patient on the operating table, induction of anesthesia, osteoplastic trepanation). According to the literature [9,10] contact IORA occur in 91-94% of cases, non-contact IORA-2.5-9% of cases. The main criteria and risk factors, which affect the frequency of IORA, consider the size of the aneurysm, its location, shape, duration of surgery after the primary rupture of the aneurysm, the presence of hypertension and the severity of the patient's condition before surgery [11,12,13,14,15,16,17,18]

The size of the aneurysm. Most authors believe that non-contact IORAs are more likely to be expected in small aneurysms (5-6 mm), and contact IORAs in large aneurysms (more than 15 mm), as they are more difficult to isolate and exclude from the bloodstream [6,14].

Localization of AA. The greatest risk for IORA is represented by aneurysms of ACA-AcomA and ICA complex [19] through hemodynamic prerequisites and a high frequency of such aneurysms (23.2-40.3%) [20]

The severity of the patient's condition before surgery. To date, a clear relationship between IORA and the severity of the patient before surgery has not been established. Some authors indicate that the incidence of IORA is higher in patients with severity of IV-V degree according to the Hunt–Hess scale [18,21]. However, J. Schramm and C. Cedzich (1993), P. D. Le Roux et al. (1996), T. Inagawa (1999) have not established a clear dependence of the frequency of IORA on the severity of patients according to the Hunt-Hess scale.

Time of surgery after a primary AA rupture. Surgery in the early stages after the primary rupture of the aneurysm, in the presence of cerebral edema, with difficulty in accessing AA, the need to perform sufficient traction of the cerebral substance is often accompanied by IORA [5,7,11,12,15,11].

METHODS AND MATERIALS

A retrospective analysis of the surgical treatment of 141 patients with intraoperative ruptures in the course of clipping of brain aneurysms, out of the total number of 1449 operated patients in the period from 2011 to 2018, was performed. The age of the patients ranged from 19 to 78 years (average age-46.7 + 11.6 years). Males-71 (50.35%), females-70 (49.65%). After primary rupture 121 (85,82%) patients were operated, repeated rupture was observed in 20 (14,18%) patients.

Perioperative examination of patients included clinical and neurological examination, brain CT scan, cerebral angiography (CAG), ultrasound (US) duplex scanning with a color Doppler mapping of the head and neck vessels mode and transcranial Doppler (TCD) was performed in all patients according to the standard methods at admission and in dynamics of perioperative period every 2 days, and in severe cases, daily. The severity of vasospasm (BC) was established by the complex of data of CAG and TCD, the following US criteria were used: the severity of distinct degree-systolic linear velocity (syst. linear velocity of blood flow) > 240 cm/s, critical degree-syst. linear velocity of blood flow>300 cm/s.

Subarachnoid (SAH) hemorrhage was detected in 67 (47.52%) patients, subarachnoid parenchymal hemorrhage – in 49 (34.75%) patients; subarachnoid ventricular – in 6 (4.26%) patients; subarachnoid parenchymal-ventricular – in 19 (13.46%) patients; SAH complicated by the formation of intracerebral hematoma – in 50 (35.46%) patients.

In the acute period of AA rupture (the first 21 days) 127 patients were operated (73 patients-the first 3 days; 35 patients-4-7 days after the rupture; 15 patients-8-14 days; 4 patients-15-21 days, 22 days and later-14 patients (Tab.1).

The period from the moment of AA rupture prior the operation	Number of patients	
	N	P, %
1-3 days	73	51.77
4-7 days	35	24.82
8-14 days	15	10.64
15-21 days	4	2.84
After 21 days	14	9.93
Total	141	100

Table 1. Term of operation after the last AA rupture

Before the operation, the severity of the patients' condition was assessed according to the Hunt-Hess scale (Tab.2), the level of consciousness according to the Glasgow coma scale.

Among the patients operated in early terms patients with severity of a condition of III-IV degree on H-H prevailed. With the increase in the duration of the operation, the proportion of patients in a compensated state (I-II degree according to H-H) increased.

Severity of the condition according to the scale of the Hunt-Hess	Number of patients	
	N	P, %
I	34	24.11
II	61	43.26
III	37	26.24
IV	5	3.55
V	4	2.84
Total number	141	100.00

Table 2.

Consciousness before the operation was clear in 86 (62.41%) patients, moderate stunning-in 32 (22.70%) patients, deep stunning – in 13 (9.22%), SOPOR-in 3 (2.13%), moderate coma (according to GCS 6-7 points) – in 1 (0.71%) patient, deep coma (according to GCS 4-5 points) – in 4 (2.84%) patients.

Focal neurological disorders before surgery were in 40 (28.37%) patients. In 13 (9.42%) patients there were violations of motor functions of the Central type, 17 (12.32%) - oculomotor disorders, 6 (4.35%) - a combination of paresis and oculomotor disorders, 1 – a combination of motor and mental disorders, 2 (1.45%) – a combination of motor and afatic disorders.

Computer tomography. All patients underwent brain CT before a surgery. Intracranial haemorrhage was assessed by CT scan (Fig.3) According to the scale of C. M. Fisher et al. (1980) (Tab. 4). The presence of cerebral edema or cerebral ischemia,

lateral or axial displacement were also taken into account.

The nature of the hemorrhage	Number of patients	
	Ab.	%
Subarachnoid	67	47,52%
Subarachnoid parenchymal	49	34,75%
Subarachnoid ventricular	6	4,26%
Subarachnoid parenchymal ventricular	19	13,46%
Total	**141**	**100**

Table 3. The nature of intracranial hemorrhage.

Degree of SAH		Number of patients	
		Ab.	%
I	Blood in the liquor spaces is not visualized	5	3.62
II	Diffuse hemorrhage or vertical size in the cistern less than 1 mm	73	52.90
III	The apparent convolution in the cistern or the vertical size of the blood signal in the cistern is more than 1 mm	54	39.13
IV	Intracerebral or intraventricular hemorrhage	6	4.35
Total		**138**	**100**

Table 4. Degree of SAH according to the Fisher scale.

Cerebral angiography. All patients underwent cerebral angiography at the planning stage of the operation. According to the results of CAG, 135 (95.74%) patients had single aneurysms, 6 (4.26%) had multiple aneurysms (Table. 5).

Localization of aneurysm (rupture)	Number of patients	
	Ab.	%
ACA-AcomA on the left	47	33.33
ACA-AcomA on the right	36	25.53
MCA on the left	9	6.38
MCA on the right	17	12.06
ICA on the left	11	7.80
ICS on the right	14	9.93
PICA on the left	0	0
PICA on the right	1	0.71
Multiple AA	ACA-AcomA AA rupture	6	3	4.26	2.13
	MCA AA rupture		1		0.71
	ICA AA rupture		1		0.71
	PICA AA rupture		1		0.71

Table 5.

Transcranial Doppler. TCD in the perioperative period was carried out in all cases: before surgery 25 (17.73%) patients, after surgery - in all cases (100 %). With an increase in the linear velocity of blood flow through the arteries of the base of the brain more than 120 cm/s and the value of the Lindegaard index more than 3, vascular spasm was considered moderate, with a blood flow rate exceeding 200 cm/s and the value of the Lindegaard index more than 6 - expressed. Angiospasm was assessed as non-widespread if it covers 1-2 arteries of the arterial circle of the brain, and was considered common with spasm of 3 or more arteries. Among the examined patients before surgery angiospasm was detected in 11 (7.80%) patients (Tab.6).

The degree of vasospasm	Number of patients (out of 141)		Number of patients (out of 11)	
	Ab.	%	Ab.	%
I	10	7.09	10	90.91
II	1	0.71	1	9.09
III-IV	0	0	0	0

Table 6.

Surgical interventions. All patients underwent microsurgical interventions. In 107 (75.89%) patients clipping of aneurysm with blood washing from basal cisterns was performed, in 22 (15.60%) patients – clipping of aneurysm was combined with removal of intracerebral hematoma, in 3 (2.13%) patients – clipping of aneurysm of was combined with external ventriculostomy, in 6 (4.26%) patients operated on vital indications due to dislocation syndrome and

volume intracranial hemorrhage - clipping of the aneurysm, removal of intracerebral hematoma and decompression trepanation of the skull were performed. Intraoperative assessment of the blood flow in the aneurysm-affected arterial segments of cerebral arteries and radicality of clipping was assessed using intraoperative contact dopplerography with a 20 Hz sensor. In the postoperative period, MSCT-angiography and CAG were performed.

RESULTS AND DISCUSSION

Treatment results were assessed at hospital discharge according to the Glasgow results scale (Jennett B., Bond M., 1975). This scale includes 5 stages (Tab.7).

Value	Number of points
Recovery of the patient without neurological disorders, or with minimal deficiency. Complete or partial recovery of labour activities.	5
Moderate disability. Neurological disorders that limit daily activities, but the patient does not need help.	4
Deep disability. The patient needs outside care	3
Vegetative state or a deep psycho-organic disorders	2
Death	1

Table 7.

Satisfactory results were recorded in most cases – 81 (61.71%) at the time of discharge from the hospital: complete or partial resumption of work – 69 (48.94%), recovery of labour activities without the need for assistance was 18 (12.77%) according to the Glasgow results scale (Tab.8).

Number of points (Glasgow results scale))		Description	Number of patients	
			Ab.	%
5	Complete recovery	Recovery of the patient without neurological disorders, or with minimal deficiency. Complete or partial recovery of labour activities.	69	48.94
4	Moderate disability	Neurological disorders that limit daily activities, but the patient does not need help.	18	12.77
3	Deep disability.	The patient needs outside care	37	26.24
2	Vegetative state	-	0	0
1	Death	-	17	12.05

Table 8. Glasgow results scale

Recorded IORA dominated on the stages of the isolation and clipping of AA – 126 (89.36%) (Tab. 9).

The stage of the operation at which the AA rupture occurred	Number of patients	
	Ab.	%
Non-contact (at the stage of craniotomy)	6	4.26
Early arachnoid dissection	3	2.13
When the affected AA artery is isolated	6	4.26
When an aneurysm is isolated	119	84.40
At the stage of aneurysm clipping	7	4.96

Table 9. The frequency of IORA at different stages of surgical interventions.

Non-contact AA rupture was recorded in 6 (4.26%) patients out of 141 (Tab.10), men 4 (2.84%), women 2 (1.42%), age from 40 to 52 years; 5 (3.55%) patients underwent clipping of single aneurysms, in 1 (0.71%) – multiple (ACA-AcomA on the right and MCA on the right); in all cases, the aneurysms were of medium size (from 4.0 mm to 14 mm); 3 (2.13%) patients were operated after primary hemorrhage, 3 (2.13%) – after repeated AA ruptures; 2 (1.42%) patients had SAH and 4 (2.84%) - subarachnoid parenchymal hemorrhage with the formation of intracerebral hematoma.

The frequency of non-contact rupture in our observations was 4.26%. Features of the manifestation of non-contact rupture of AA during surgery were: signs of arterial bleeding with basal spread and signs of edema-swelling of the brain, which were associated with spontaneous short-term (up to several minutes) fluctuation of blood pressure (not significant in relation to the systemic hemodynamics). Two (1.42%) patients were operated on the first 3 days after SAH, 2 (1.42%) – on the 4-6th day after rupture, 1 (0.71%) - on the 9th day after the last AA rupture, 1 (0.71%) – on the 16th day.

In all cases of non-contact rupture of AA, all further stages of surgery: isolation of the arterial segment affected by the aneurysm, isolation of the initial sections of the perforant arteries, neck and body of AA itself, clipping of the aneurysm, were performed using the technique of gradual temporary blocking of blood flow in the proximal segments of the cerebral arteries in relation to the location of the aneurysm. Thus, the gradual blocking of segments of cerebral arteries on the affected side from the proximal segment of the internal carotid artery (ICA) aneurysm to the affected segment (A1 segment of the ACA or the M1 segment of MCA) with a stages offset of clips for temporary locking in the distal direction to the arterial segment affected by the aneurysm and reperfuse of pre-blocked (proximal) segment was performed to 3 (2,13%) patients, in 3 (2,13%) cases a temporary blockage of blood flow to highlight the AA and its clipping of an arterial segment directly affected by the aneurysm (A1 segment of the ACA or the M1 segment of MCA) was technically possible . In the postoperative period, the secondary cerebral ischemia as a result of strengthening of the phenomena of initial cerebral angiospasm in conjunction with the duration of temporary blocking of blood flow, which exceeded 5 minutes, was observed in 2 (1.42%) patients with non - contact intraoperative breaks AA MCA, according to MSCT of the brain.

Operation	AA localization	AA size (mm)	Number of chambers	The period after a rupture	Number of patients			
					Ab.	%	Ab.	%
AA clipping. Rehabilitation of basal cisterns	ACA-AcomA on the right	4-14	Single-chamber	After 21 days	1	0.71	2	1.45
	MCA on the right	4-14	Single-chamber	On the 8-14th day	1	0.71		
AA clipping. Sanitation of the basal cisterns. Removal of I/M stroke-hematoma.	MCA on the right	4-14	Single-chamber	First 3 days	1	0.71	2	1.45
				4-7 days	1	0.71		
AA clipping. Sanitation of the basal cisterns. Removal of I/M stroke-hematoma. Decompressive craniotomy.	ACA-AcomA on the right	9	Single-chamber	First 3 days	1	0.71	1	0.72
AA clipping. Blood washing from the basal cisterns. Decompressive craniotomy.	ACA-AcomA on the right	4-14	Single-chamber	5th day	1	0.71	1	0.72

Table 10. Operation for a non-contact rupture of B AA.

Angiospasm in the early postoperative period was observed in the vast majority-5 (83, 33%) out of 6 (100%) patients with non-contact IORA (Tab.11).

Angiospasm	Number of patients (out of 141)		Number of cases (out of non-contact ruptures -6)	
	Ab.	%	Ab.	%
I degree	0	0	0	0
II degree	3	2.13	3	50.00
III degree	2	1.42	2	33.33
IV degree	0	0	0	0
The absence of vasospasm	1	0.71	1	16.67

Table 11. Angiospasm after surgery.

In all patients with angiospasm in the postoperative period, there was a deepening of the initial neurological symptoms in the form of the

appearance or deepening of pyramidal insufficiency, motor, sensory aphasia or mental disorders. In 1 patient operated on for ACA-AcomA AA on the left, in the postoperative period, right-sided hemiparesis joined; in 1 patient operated on for rupture of SA MCA on the right with the formation of intracerebral hematoma in the postoperative period, there were signs of psychoorganic syndrome (Walter-Buehl triad) according to the explosive type with psychotic complications; in 3 patients, the symptoms combined paresis (sensorimotor, motor aphasia) and organic PSYCHOSYNDROME.

Contact intraoperative AA rupture was observed in 135 (100%): men – 67 (49.6%), women – 68 (50.4%), which was 95.74% of 141 cases (100%). Age of patients - from 19 to 68 years. The vast majority of patients with contact IORA were operated for ACA-AcomA AA (Tab. 14). In 4 (2.96%) cases there were multiple AA: ACA-AcomA (3 cases) and ICA.

Localization of aneurysm		Number of patients	
		Ab.	%
ACA-AcomA	On the left	47	34.81
	On the right	38	28.15
MCA	On the left	9	6.67
	On the right	14	10.37
ICA	On the left	12	8.89
	On the right	14	10.37
PICA	On the left	0	0
	On the right	1	0.74
Total		135	100

Table 12. Localization of B AA complicated by contact IORA.

The degree of consciousness according to the GCS before surgery in 84 (62.22%) patients was clear, in 30 (22.22 %) was impaired by the type of moderate stunning; in 13 (9.63%) - deep stunning; in 3 (2.22%) – sopor; in 1 (0.74%) – moderate coma (according to the GCS 6-7 points); in 4 (2.97%) - deep coma (according to the GCS 4-5 points).

Out of 71 (52.59%) patients operated on in the first 3 days after AA rupture, 32 (23.7%) patients had SAH; 22 (16.29%)-subarachnoid parenchymal hemorrhage; 3 (2.22%)-subarachnoid ventricular hemorrhage; 14 (10.37%) - subarachnoid parenchymal-ventricular hemorrhage.

The overall mortality rate after IORA was 17 (12.06%) cases out of 141 (100%) cases, whereat deaths in the contact IORA group – 17 (12.6% out of 135 (100%)). Although according to the literature, a significant increase in the number of fatal cases is observed after early IORA, which occurred at the stages of craniotomy before the opening of TMO and the beginning of arachnodal dissection, when there is no possibility of direct control of bleeding from the aneurysm by temporary or permanent blocking of the neck or body of the aneurysm, or the imposition of a temporary clip on the affected segment of the artery, there was no such trend in the analyzed series of observations. However, there was a significant increase in the number of cases of deepening of the initial neurological symptoms in the group of patients with non-contact IORA, which probably, given a larger sample, could be associated with an increase in the number of deaths.

Table 13. Operations on B AA complicated by contact IORA.

Surgical interventions in the first 3 days after AA rupture (81)								
Operation	AA localization	AA size (mm)	Number of chambers		Number of patients			
					Ab.	%	Ab.	%
AA clipping. Rehabilitation of basal cisterns	ACA-AcomA	On the left	4-14	single-chamber	37	26.24	14	9.93
		On the right	15-24	multichamber			3	2.13
			4-14	single-chamber			8	5.67
			up to 3	single-chamber			1	0.71
	MCA on the right	4-14	single-chamber				3	2.13
		15-24	multichamber				1	0.71
	ICA	On the left	4-14	single-chamber			1	0.71
			5-24	single-chamber			1	0.71
		On the right	4-14	single-chamber			5	3.55

AA clipping. Sanitation of the basal cisterns. Removal of I/M stroke-hematoma.	ACA-AcomA	On the left	15-24	double chamber	36	25.53	1	0.71
			4-14	single-chamber			2	1.42
				double chamber			1	0.71
				three-chamber			1	0.71
			up to 3	single-chamber			2	1.42
		On the right	15-24	multichamber			1	0.71
			4-14	single-chamber			5	3.55
				multichamber			1	0.71
			up to 3	single-chamber			1	0.71
	MCA	On the left	4-14	single-chamber			3	2.13
			Up to 3	single-chamber			1	0.71
		On the right	15-24	single-chamber			1	0.71
				multichamber			1	0.71
			4-14	single-chamber			5	3.55
			Up to 3	single-chamber			1	0.71
	ICA	On the left	15-24	single-chamber			1	0.71
			4-14	single-chamber			1	0.71
		On the right	giant	double chamber			1	0.71
			4-14	single-chamber			6	4.26
AA clipping. Drainage of the ventricular system.	PICA on the right	7	single-chamber		1	0.71	1	0.71
AA clipping. Removal of I/M hematoma. Decompressive craniotomy.	ACA-AcomA	On the left	4-14	single-chamber	6	4.26	4	4.26
		On the right	15-24	multichamber			1	0.71
	MCA on the left	4-14	single-chamber				1	0.71
AA clipping. Blood washing from the basal cisterns. Decompressive craniotomy.	ACA-AcomA on the right	4-14	single-chamber		1	0.71	1	0.71

Surgical interventions for 4-7 days after AA rupture - 34

Operation	AA localization	AA size (mm)	The presence of diverticula, q-ty of chambers		Number of patients			
					Ab.	%	Ab.	%
AA clipping. Rehabilitation of basal cisterns	ACA-AcomA	On the left	15-24	single-chamber	24	17.02	1	0.71
				multichamber			1	0.71
			4-14	single-chamber			5	3.55
			up to 3	single-chamber			1	0.71
		On the right	15-24	multichamber			2	1.42
			4-14	multichamber			1	0.71
				single-chamber			4	2.84
			up to 3	single-chamber			1	0.71
	MCA	On the left	15-24	multichamber			1	0.71
		On the right	4-14	single-chamber			1	0.71
	ICA	On the left	giant	multichamber			1	0.71
			4-14	single-chamber			2	1.42
		On the right	15-24	multichamber			1	0.71
			4-14	single-chamber			2	1.42
AA clipping. Sanitation of the basal cisterns. Removal of I/M stroke-hematoma.	ACA-AcomA	On the left	4-14	single-chamber	9	6.38	2	3.62
		On the right	Up to 3	single-chamber			1	0.71
			4-14	single-chamber			1	0.71
	MCA	On the right	4-14	single-chamber			2	1.42
	ICA	On the left	4-14	single-chamber			2	1.42
		On the right	Up to 3	single-chamber			1	0.71

AA clipping. Drainage of the ventricular system.	MCA on the left	4-14	single-chamber	1	0.71	1	0.71	

Surgical interventions on the 8-14th day -14 (9,93%)

Operation	AA localization	AA size (mm)	The presence of diverticula, q-ty of chambers		Number of patients			
					Ab.	%	Ab.	%
AA clipping. Rehabilitation of basal cisterns	ACA-AcomA	On the left	4-14	single-chamber	12	8.51	1	0.71
				multichamber			1	0.71
		On the right	4-14	single-chamber			2	1.42
			up to 3	double chamber			1	0.71
	MCA	On the left	4-14	single-chamber			1	0.71
		On the right	15-24	multichamber			1	0.71
			4-14	multichamber			1	0.71
	ICA	On the left	4-14	multichamber			1	0.71
				single-chamber			1	0.71
		On the right	Up to 3	single-chamber			2	1.42
AA clipping. Sanitation of the basal cisterns. Removal of I/M stroke-hematoma.	ACA-AcomA	On the left	4-14	double chamber	2	1.42	1	0.71
		On the right	15-24	multichamber			1	0.71

Surgical interventions on the 15-21 day -11 (7,8%)

Operation	AA localization	AA size (mm)	The presence of diverticula, q-ty of chambers		Number of patients			
					Ab.	%	Ab.	%
AA clipping. Rehabilitation of basal cisterns	ACA-AcomA	On the left	15-24	multichamber	10	7.09	1	0.71
			4-14	single-chamber			2	1.42
				multichamber			1	0.71
			up to 3	single-chamber			1	0.71
		On the right	4-14	Single-chamber			1	0.71
	MCA on the right	4-14	Single-chamber				1	0.71
	ICA	On the left	4-14	multichamber			1	0.71
		On the right	4-14	single-chamber			2	1.42
AA clipping. Blood washing from the basal cisterns. Decompressive craniotomy.	ICA on the left	4-14	single-chamber	1	0.71		1	0.71

Surgical interventions after 21 days-13 (9,22%)

Operation	AA localization	AA size (mm)	The presence of diverticula, q-ty of chambers		Number of patients			
					Ab.	%	Ab.	%
AA clipping. Rehabilitation of basal cisterns	ACA-AcomA	On the left	15-24	multichamber	13	9.22	1	0.71
			4-14	single-chamber			3	2.17
			Up to 3	single-chamber			1	0.71
		On the right	4-14	single-chamber			1	0.71
				multichamber			1	0.71
	MCA	On the left	4-14	multichamber			1	0.71
		On the right	4-14	single-chamber			1	0.71
	ICA	On the left	4-14	multichamber			1	0.71
				single-chamber			1	0.71
		On the right	4-14	single-chamber			2	1.42

Summary

1. The frequency of IORA in B AA clipping operations was 9.73% (141 patients) in a series of observations 1441 (100%). According to our observations, most often IORA-141 (100%) was registered in operations pf clipping of AA complex ofACA-AcomA (86 (61%) cases out of 141 (100%)).
2. IORA is possible at different stages of the aneurysm clipping operation with a maximum frequency of contact breaks -135 (95.74%): at the stage of AA isolation (119 cases-84.40%), with the imposition of a clip on the neck of AA - 7 (5.07%), with the isolation of the artery affected by aneurysm 6 (4.26%) and with arachnoid dissection – 3 (2.17%). Non-contact IORA (at the stage of craniotomy) was recorded less often-6 (4.26%).
3. The most common risk factors for IORA were: cerebral edema, large AA, atherosclerotic changes in the aneurysm-affected artery segment and cervical aneurysm sites, high blood pressure during surgery, adhesions arachnoid changes.
4. At the time of discharge from the hospital, according to the Glasgow results scale: 69 (48.94%) full or partial restoration of labor activity, 18 (12.77%) had limited daily activities without the need for outside assistance, 37 (26 24%) deep disability) Deaths were in the group of "contact" IORA - 17 (12.06%). At 6 (4.26%) of "non-contact" IORA, a deepening of initial neurological symptoms was recorded with a suppression of the level of consciousness, the addition of pyramidal insufficiency, speech impairment and psycho-organic syndrome, and a deepening of the phenomena of initial cerebral arterial vasospasm.

Abbreviations

AA – arterial aneurysm; SA– sacculated aneurysm; IOC – intraoperative complications; SAH – subarachnoid hemorrhage; BP – blood pressure; IORA– intraoperative aneurysm rupture; TC – temporary clipping; ACA-AcomA– anterior cerebral - anterior communicating artery; MCA– middle cerebral artery; ICA – internal carotid artery; PICA– posterior inferior cerebellar artery; IAH – induced arterial hypotension; H-H – Hunt-Hess scale; GCS – Glasgow coma scale; CAG – cerebral angiography; MSCT – multispiral computed tomography.

References

1. Lai LT, O'Neill AH. History, Evolution, and Continuing Innovations of Intracranial Aneurysm Surgery. World Neurosurg. 2017 Jun; 102:673-681. doi: 10.1016/j.wneu.2017.02.006. PubMed PMID: 28189863.
2. Acciarri N, Toniato G, Raabe A, Lanzino G. Clipping techniques in cerebral aneurysm surgery. J Neurosurg Sci. 2016 Mar;60(1):83-94. PubMed PMID: 26657306.
3. Lawton MT, Du R. Effect of the neurosurgeon's surgical experience on outcomes from intraoperative aneurysmal rupture. Neurosurgery. 2005 Jul;57(1):9-15; discussion 9-15. doi: 10.1227/01.neu.0000163082.20941.ef. PubMed PMID: 15987535.
4. Liu Q, Jiang P, Wu J, Gao B, Wang S. The Morphological and Hemodynamic Characteristics of the Intraoperative Ruptured Aneurysm. Front Neurosci. 2019 Mar 26;13:233. doi: 10.3389/fnins.2019.00233. PubMed PMID: 30971874; PubMed Central PMCID: PMC6443834.
5. Tian Z, Zhang Y, Jing L, Liu J, Zhang Y, Yang X. Rupture Risk Assessment for Mirror Aneurysms with Different Outcomes in the Same Patient. Front Neurol. 2016 Dec 5;7:219. doi: 10.3389/fneur.2016.00219. PubMed PMID: 27994571; PubMed Central PMCID: PMC5136536.
6. Choque-Velasquez J, Hernesniemi J. Microsurgical clipping of a large ruptured anterior communicating artery aneurysm. Surg Neurol Int. 2018 Nov 28;9:233. doi: 10.4103/sni.sni_345_18. PubMed PMID: 30595954; PubMed Central PMCID: PMC6287331.
7. Дзяк Л.А., Зорин Н.А., Голик В.А., Скребец Ю.Ю. Артериальные аневризмы и артериовенозные мальформации головного мозга. Днепропетровск: Пороги; 2003.
8. Dzyak LA, Zorin NA, Golik VA, Skrabets Yu. [Arterial aneurysms and arteriovenous malformations of the brain]. Dnepropetrovsk: Porogi, 2003. Russian.
9. Крылов В.В., Природов А.В. Факторы риска хирургического лечения аневризм средней мозговой артерии в остром периоде кровоизлияния. Нейрохирургия. 2011;(1):31-41.
10. Krylov V.V., Prirodov A.V. Risk factors of surgical treatment for middle cerebral artery aneurysms in acute period of subarachnoid hemorrhage. The Russian Journal of Neurosurgery. 2011;(1):31-41. Russian. Available from: https://elibrary.ru/item.asp?id=16449822
11. Хейреддин А.С., Филатов Ю.М., Белоусова О.Б., Пилипенко Ю.В., Золотухин С.П., Сазонов И.А., Зарзур Х.Х. Интраоперационный разрыв церебральных аневризм-частота и факторы риска. Журнал Вопросы нейрохирургии им. Н.Н. Бурденко. 2007;(4):33-8.
12. Kheireddin AS, Filatov IuM, Belousova OB, Pilipenko IuV, Zolotukhin SP, Sazonov IA, Zarzur KhKh. [Intraoperative rupture of cerebral aneurysm--incidence and risk factors]. Zh Vopr Neirokhir Im N N Burdenko. 2007 Oct-Dec;(4):33-8; discussion 38. Russian. PubMed PMID: 18274133.
13. Lakićević N, Vujotić L, Radulović D, Cvrkota I, Samardžić M. Factors Influencing Intraoperative Rupture of Intracranial Aneurysms. Turk Neurosurg. 2015;25(6):858-85. doi: 10.5137/1019-5149.JTN.12966-14.2. PubMed PMID: 26617133.
14. Leipzig TJ, Morgan J, Horner TG, Payner T, Redelman K,

Johnson CS. Analysis of intraoperative rupture in the surgical treatment of 1694 saccular aneurysms. Neurosurgery. 2005 Mar;56(3):455-68; discussion 455-68. doi: 10.1227/01.neu.0000154697.75300.c2. PubMed PMID: 15730570.

15. Nanda A, Vannemreddy P. Management of intracranial aneurysms: factors that influence clinical grade and surgical outcome. South Med J. 2003 Mar;96(3):259-63. doi: 10.1097/01.SMJ.0000051906.95830.1F. PubMed PMID: 12659357.
16. Lin TK, Hsieh TC, Tsai HC, Lu YJ, Lin CL, Huang YC. Factors associated with poor outcome in patients with major intraoperative rupture of intracranial aneurysm. Acta Neurol Taiwan. 2013 Sep;22(3):106-11. PubMed PMID: 24030089.
17. Lawton MT, Du R. Effect of the neurosurgeon's surgical experience on outcomes from intraoperative aneurysmal rupture. Neurosurgery. 2005 Jul;57(1):9-15; discussion 9-15. doi: 10.1227/01.neu.0000163082.20941.ef. PubMed PMID: 15987535.
18. Liu Q, Jiang P, Wu J, Gao B, Wang S. The Morphological and Hemodynamic Characteristics of the Intraoperative Ruptured Aneurysm. Front Neurosci. 2019 Mar 26;13:233. doi: 10.3389/fnins.2019.00233. PubMed PMID: 30971874; PubMed Central PMCID: PMC6443834.
19. Forget TR Jr, Benitez R, Veznedaroglu E, Sharan A, Mitchell W, Silva M, Rosenwasser RH. A review of size and location of ruptured intracranial aneurysms. Neurosurgery. 2001 Dec;49(6):1322-5; discussion 1325-6. doi: 10.1097/00006123-200112000-00006. PubMed PMID: 11846931
20. Kopitnik TA, Horowitz MB, Samson DS. Surgical management of intraoperative aneurysm rupture. In: Schmidek HH, Sweet WH, editors. Schmidek & Sweet's Operative Neurosurgical Techniques: Indications, Methods, and Results. 4th ed. Philadelphia: WB Saunders Co; 2000. p. 1275–1281.
21. Chen SF, Kato Y, Kumar A, Tan GW, Oguri D, Oda J, Watabe T, Imizu S, Sano H, Wang ZX. Intraoperative rupture in the surgical treatment of patients with intracranial aneurysms. J Clin Neurosci. 2016 Dec;34:63-69. doi: 10.1016/j.jocn.2016.01.045. PubMed PMID: 27692502.
22. Lawton MT, Du R. Effect of the neurosurgeon's surgical experience on outcomes from intraoperative aneurysmal rupture. Neurosurgery. 2005 Jul;57(1):9-15; discussion 9-15. doi: 10.1227/01.neu.0000163082.20941.ef. PubMed PMID: 15987535.
23. Weir B, Disney L, Karrison T. Sizes of ruptured and unruptured aneurysms in relation to their sites and the ages of patients. J Neurosurg. 2002 Jan;96(1):64-70. doi: 10.3171/jns.2002.96.1.0064. PubMed PMID: 11794606.
24. Della Puppa A, Rossetto M, Volpin F, Rustemi O, Grego A, Gerardi A, Ortolan R, Causin F, Munari M, Scienza R. Microsurgical Clipping of Intracranial Aneurysms Assisted by Neurophysiological Monitoring, Microvascular Flow Probe, and ICG-VA: Outcomes and Intraoperative Data on a Multimodal Strategy. World Neurosurg. 2018 May;113:e336-e344. doi: 10.1016/j.wneu.2018.02.029. PubMed PMID: 29452324.
25. de Rooij N. K., Linn F. H., van der Plas J A, et all. Incidence of subarachnoid haemorrhage: a systematic review with emphasis on region, age, gender and time trends. In: J. Neurol. Neurosurg. Psychiatry 2007,78, p.1365–1372.

Romanian Neurosurgery (2020) XXXIV (1): pp. 77-84
DOI: 10.33962/roneuro-2020-010
www.journals.lapub.co.uk/index.php/roneurosurgery

Surgical challenges and outcome of endoscopic endonasal approaches in the management of recurrent pituitary adenomas

Ahmed Nageeb Mohamed Taha, Hosam Shata Mohamed, Mahmoud Saad Mohamed

Neurosurgery Department, Mansoura College of Medicine, Mansoura University, EGYPT

ABSTRACT

Background: Various surgical approaches for the management of midline anterior skull base meningiomas exist in the literature. The main surgeon target is proper selection the appropriate approach that achieves total removal of the lesion without causing morbidity or mortality and facilitates safe effective removal of the tumor.

Objectives: To evaluate the role of the extended pterional approach for excision of midline anterior skull base meningiomas as regard the effectiveness, extent of resection and surgical outcome.

Patients and methods: This retrospective study involved 23 cases with midline anterior skull base meningiomas resected through the extended pterional approach. Patients' clinical data, operative notes, imaging studies and clinical follow-up data were analyzed and evaluated.

Results: Tumors studied were 9 olfactory groove meningiomas, 8 tuberculum Sellae meningiomas, 4 planum sphenoidale meningiomas and 2 diaphragma sellae meningiomas. Gross total resection tumor excision in 15 cases (64.5%), subtotal excision in 5 cases (21.5%) and partial excision in 3 cases (14%). Complications were diabetes insipidus (2 cases 8.6%), CSF rhinorrhea (3 cases 12.9%) and visual deterioration (3 cases 12.9%). We had two cases of mortality.

Conclusion: The extended pterional approach allows safe and effective removal of midline anterior skull base meningiomas. It expands the exposure offered by the classic pterional approach and minimizing the necessity for applying fixed brain retraction with good cosmetic outcome and less approach related morbidities in comparison with the extensive skull base approaches.

Keywords
extended pterional,
meningioma,
skull base,
extent of resection

Corresponding author:
Mahmoud Saad Mohamed

Neurosurgery Department,
Mansoura Collage of Medicine,
Mansoura University, Egypt

dr_mhmodsaad@yahoo.com

Scan to access the online version

INTRODUCTION

Pituitary adenomas are relatively common brain tumor with benign features, and, in fact, they are found in 10%-17% of the general population. Surgical resection continues to be the preferred treatment except for prolactin-secreting tumors. The nature of the pituitary adenoma itself suggests the possibility of tumor recurrence, regardless of its endocrinological characteristics. Recurrence rate

of pituitary adenoma after surgical resection has been reported up to 30%, and regrowth after incomplete tumor removal was reported in up to 75% of cases [2,5,10,12,16].

Recurrent tumors can be managed with observation, medical therapy, radiotherapy, radiosurgery, or revision surgery. Additionally, the combined treatment paradigm is essential in some cases because all treatment modalities have advantages and disadvantages. Surgery for a recurrent lesion is burdened by increased risk of mortality and morbidity, and it often results in incomplete resection compared with initial surgery. Radiosurgery and stereotactic radiotherapy can be recommended as adjuvant treatments to obtain long-term control with low procedure-related morbidity in selective cases. However, the repeated surgical resection is crucial in cases of sizable adenomas compressing the optic apparatus, or hormone-secreting adenomas [1,2,3,4,7,9,14].

Nearly a century ago, Harvey Cushing realized the limitations of microsurgical approaches for treating pituitary adenomas. Cushing and his colleagues used a radium bomb to deliver a single-session, focused radiation to treat pituitary adenomas. Henceforth, neurosurgeons and radiation oncologists have employed repeat resection or ionizing radiation to treat selected patients with recurrent or residual pituitary adenomas[1,6,22,24].

During the last century, pituitary surgery has been developed with various technical modifications and instrumental advances, and the outcome has been generally excellent, with high rates of clinical improvement and endocrinological remission and minimal rates of morbidity and mortality. The introduction of endoscopy in the transsphenoidal approach (TSA) has tremendously advanced the midline skull base surgery. In the literatures, the efficacy and safety of endoscopic TSA in the management of pituitary adenomas have been proven and the complication rates of endoscopic TSA are at least comparable with those of microscopic series. However, the reports on endoscopic TSA as a revision surgery have been less commonly published [6,8,11,13,15,19,26,28].

We retrospectively evaluated the efficacy and safety of the endoscopic TSA for recurrent pituitary adenomas as regard extent of resection, ICA injury and CSF leakage and related reconstruction obstacles.

Material and methods

A retrospective study is conducted on 30 patients with recurrent pituitary adenomas that were re-operated up on via endoscopic endonasal transsphenoidal approach at the Neurosurgery Department, Mansoura University over the period from 2013 to 2019. Patients who did a previous transsphenoid either via microscopic or endoscopic technique are included in the study. The indications to do another endoscopic endonasal transsphenoidal approach were big residual or recurrent tumor compressing the chiasm, persistent endocrinological problem due to residual adenoma. Patient demographic and previous surgery data were collected and analyzed including any previous medical treatment, radiation treatment and any previous surgery related morbidities. Routine pre-operative complete ophthalmological assessment was done for all cases and was repeated after surgery to assess the outcome. Pre- and postoperative endocrinological assessment were done for all cases including serum prolactin, free cortisol, ACTH, free thyroxine, TSH, GH and insulin-like growth factor 1 (IGF-1). Serum electrolytes and fluid chart were done for all cases with diabetes insipidus that present either prior or after the surgery. Pre-operative contrast-enhanced MRI were done for all the patients and then 3 months after surgery and then yearly for follow up. The tumors were assessed regarding its size and extension. Tumor volumes were assessed by the ellipsoid model "(ABC)/2" equation. The degree of cavernous sinus invasion was evaluated based on Knosp criteria. The extent of tumor resection (EOR) was assessed based on a post-contrast MRI study done 3 months after surgery.

Surgical technique

We utilize the standard endoscopic approach for pituitary adenoma surgery which is widely described in the literature, and we will stress on our steps in relation to surgery for recurrent cases. Lumbar drain was utilized for cases with history of previous CSF rhinorrhea with the initial surgery and for cases we have encountered intra-operative CSF leakage and was left for 1 to 3 postoperative days for postoperative drainage. The important steps to ensure efficient safe surgery for recurrent cases were binostril four handed endoscopic technique with wide sphenoidotomy and obtaining adequate

sellar exposure to have the full panoramic view ofthe carotid prominences, medial and lateral optico-carotid recesses, planum and clivus. Our biggest challenge was the way to achieve adequate reconstruction after tumor resection. Options were reusing prior flap from the previous surgery, obtaining a contralateral nasoseptal flap but in many cases with extensive adhesions and disturbed normal anatomyespecially those previously irradiated; abdominal fat graft or fascia lata graft was utilized to achieve sellar reconstruction.

Results

Thirty patients involved in this study were operated up on via endoscopic endonasal transsphenoidal surgery for recurrent or residual pituitary adenomas. 18 were females and 12 were males. Age ranged from 29 to 61 years (mean 50 years). Twenty-four patients (80%) had single prior endoscopic endonasal transsphenoidal surgery. The remaining 6 patients (20%) had 2 previous transsphenoidal surgeries. In 23 patients; the initial surgery was done at Neurosurgery Department, Mansoura University Hospitals while the remaining seven cases were previously operated up on in other hospitals. The initial surgery was done microscopically in 13 cases while the other 17 cases were operated up on via the endoscopic approach (table 1).

The mean interval between the previous surgery and the redo endoscopic endonasal surgery was 30 months (ranged from 19 months to 42 months). 19 patients (63.3%) presented with tumor growth on follow up MRI brain before the onset of clinical symptoms andwere re-operated uponbecause of growth of the residual tumor or persistent symptoms related to a persistent hormone hypersecretion. In 11 patients (36.6%); the tumor recurred after previous total resection documented on postoperativeMRI brain. 21 patients (70%) had macro-adenomasat recurrence while the remaining nine patients (30%) had giant adenomas.

The clinical presentation was visual affection in 9 patients (30%), headache in 3 patients (10%), amenorrhea galactorrhea in 5 patients(16.6%)and persistent acromegalic features in 8 patients (26.6%).Only 4 patients received adjuvant radiotherapy following first surgery (table 1).17 patients (56.6%) had nonfunctional pituitary adenomas. 8 patients (26.6%) had GH adenomas and 5 patients (16.6%) had a prolactinoma. Tumor locations varies at time of presentation and determined radiologically with contrast enhanced MRI brain and distributed into: confined to sella in 7 cases (23.3%), sellarwithsuprasellar extension in 12 cases (40%), sellar with parasellarextension in 6cases (20%) and sellar, suprasellar and parasellar extension in 5 cases (16.6%) (table 2).

Overall Gross total resection was achieved in 17 patients (56.6%) while subtotal and partial resection in 13 patients (43.4%) confirmed by Gadolinium enhanced magnetic resonance imaging of the brain done 3 months after surgery (table 3). Total resection was achieved in 11 cases of non-functioning adenoma, 3 cases of growth hormone secreting adenomas and 3 cases of prolactinomas. Total resection was achieved in 9 (47.3%) residual adenomas and in 8 (72.7%) recurrent adenomas.According to the tumor extension; total resection was feasible in 5 out of 6(83.3%) cases where the tumor is confined to sellae, in 10 out of 12 (83.3%) cases where is tumor is sellar and suprasellar extension and in 1 out of 11 (9.09%) of cases that had parasellar extension into the cavernous sinus. Subtotal resection was done in 6 cases of non-functioning adenoma, 5 cases of growth hormone secreting adenomas and 2 cases of prolactinomas. Subtotal resection was achieved in 10 (52.6%) residual adenomas and in 3 (27.2%) recurrent adenomas. According to the tumor extension; subtotal resection was done in 1 out of 6 (16.6%) cases where the tumor is confined to sellae, in 2 out of 12 (16.6%) cases where is tumor is sellar and suprasellar extension and in 10 out of 11 (90.9%) of cases that had parasellar extension into the cavernous sinus (table 3).

No further treatment was required for the 17 cases with total tumor resection. Among 6 cases of non-functioning adenoma with incomplete resection; 4 cases were done via microscopic transcranial approach and 2 of them offered 3-dimensional radiotherapy after the transcranial surgery while the other 2 cases were treated with gamma knife radiosurgery. We have 5 cases of acromegaly with incomplete resection; one was offered additional microscopic transcranial approach followed by conformal radiotherapy, 3 offered Gamma knife radiosurgery and one died from ruptured carotid pseudoaneurysm from intra-operative vascular injury. 2 cases of residual prolactin secreting adenomas were treated

medically with dopamine agonists and one of them received conformal radiotherapy after failed trail of medical treatment.

We have a case of mortality from carotid injury in a case of recurrent growth hormone secreting adenoma. 3 cases had post-operative CSF rhinorrhea; 2 passed conservatively and one required another endoscopic repair. 4 cases developed post-operative diabetes insipidus (2 required permanent treatments). 3 cases developed post-operative hypopituitarism requiring long term hormonal replacement therapy. Improvement of the visual manifestations occurred in 5 (55.5%) out of the 9 cases presented with visual affection regarding visual acuity and visual field disturbances (table 4). Regression of acromegalic features and normalization of growth hormone and insulin-like growth factor occurred in 3 (37.5%) out of 8 acromegalic cases while the other 5 (62.5%) cases failed to improve necessitating further treatment to control endocrinological problem. Restoration of normal menstrual cycle and cessation of galactorrhea occurred in 3 cases after total tumor removal while the other 2 cases with residual tumor were kept on medical treatment and one case received radiotherapy.

Table 1. Clinical and demographic distribution of recurrent adenoma patients

Feature	Number (%)
Gender:	
Male	12 (40%)
Female	18 (60%)
Pathology:	
Nonfunctioning	17 (56.6%)
GH producing	8(26.6%)
Prolactinoma	5 (16.6%)
Number of previous surgeries:	
1	24 (80%)
2	6 (20%)
Previous resection:	
Recurrent tumor	11 (36.6%)
Residual tumor	19 (63.3%)
Clinical presentation:	
Tumor regrowth (MRI)	14 (46.6%)
Visual	9(30%)
Acromegaly	8 (26.6%)
Headache	3 (10%)
Amenorrhea galactorhea	5 (16.6%)

ILLUSTRATED CASES

Case 1: Forty-two years old male patient with recurrent non-functioning pituitary adenoma (fig. 1 a & b) with visual compression totally resected (fig. 1 c & d) with marvelous visual improvement.

Case 2: Fifty-four years old female patient with recurrent growth hormone secreting adenoma with visual compression (fig. 2 a & b); subtotal resection due to cavernous sinus invasion with good optic apparatus decompression (fig. 2 c & d) and followed by Gamma knife radiosurgery.

DISCUSSION

Recurrent or residual pituitary adenomas after transsphenoid approach whether done microscopically or endoscopically represent a potential challenge in deciding the best appropriate treatment. Many options of treatment are available including just follow up, medical treatment for some functioning adenoma, radiotherapy including gamma knife, microscopic transcranial resection and repeat the transsphenoid approach. The decision is based up many factors including size of the residual or recurrent adenomas, pattern of the tumor extension, nature and function of the adenoma, clinical presentation and data regarding previous operation, surgery related morbidities and if any adjuvant treatment was given after the initial surgery **(1,3,8,21,23,25,29).**

Tumor extension	Total	Residual adenoma	Recurrent adenoma
Sellar	7	2	5
Sellar& suprasellar	12	6	6
Sellar&parasellar	6	6	0
Sellar&suprasellar& parasellar	5	5	0

Table 2. Pattern of tumor extension

Extent of previous resection	Number of cases	Gross total resetion	Tumor extension
Residual tumor	19	9 (30%)	2 sellar, 6 sellar & supra sellar, 1 sellar &p arasellar
Recurrent tumor	11	8 (26.6%)	4 sellar, 4 sellar & suprasellar

Table 3. Pattern of tumor extension Extent of tumor resection correlated to pattern of tumor extension

In this study we evaluated our results of endoscopic transsphenoid approach for recurrent and residual pituitary adenomas. Among the 30 cases involved in this study; 19 cases were residual tumors and 11 cases were recurrent after initial total resection. Among the 19 cases with residual tumors; 11 (57.9%) cases had tumor extension to the cavernous sinus and this reflects that the extent of tumor resection was more related to the pattern of tumor extension rather than the nature of the adenoma or the technique of surgery whether microscopic or endoscopic. In 8 patients with residual adenomas; the tumor was confined to sella in 2 cases and sellar with suprasellar extension in 6 cases and the reason for incomplete tumor removal was inadequate opening all the sphenoid septationswith limited delineation of the sellar floor with less room for manipulating the endoscope and the instrument.

Table 4. Outcome of surgery of recurrent pituitary adenomas correlated to literature reports

series	No. of cases	EOR (No. of cases, %)		No. of cases, % of cases				Other complications
		GTR	STR	Complications	Hypopit.	DI	CSF leak	
Curent study	30	17, 56.6%	13, 43.4%	8, 26.6%	0	4, 13.3%	3, 10%	Mortality: 1 (carotid injury)
Negm et al. 2017	41	24, 55.5%	2, 4.9%	8, 19.5%	4, 9.7%	2, 4.9%	1, 2.4%	Hematoma: 1 (2.4%)
Wang et al. 2015	29	16, 55%	11, 38%	5, 17%	0	1, 3.4%	1, 3.4%	2 (6.9%) cases of deerioration of vision due to hemorrhage in tumor bed & intracranially; 6th nerve palsy: 1 (3.4%)
Tajudeen et al. 2015	27	17, 63%	7, 26%	6, 22%	1, 3.7%	2, 7.4%	0	Abducent palsy: 1 (3.7%)
Cavallo et al. 2012	59	37, 63%	9, 15%	8, 13.5%	4, 6.7%	3 (5%)	1 (1.7%)†	Hematoma: 1 (1.7%)
Rudnik et al. 2006	20	8, 40%	0	4, 20%	3, 15%	0	1, 5%	
Hwang et al. 2013	30	15, 50%	7, 23%	5, 16%	1, 3.3%	4 (13.3%)	0	Mengitis: 2 (6.7%)
Yamada et al. 2010	53	31, 58.5%	0	5, 9%	1, 2%	1, 2%	1, 2%	Epistaxis: 1 (2%), pituitary abscess: 1 (2%)
Alahmadi et al. 2011	39	18, 46%	21, 54%	6, 13%	0	0	1 (2.6%)†	1 MI, 1 HCP, 1 sinusitis, 1 crushing

Mattozo et al. 2006	30	17, 57%	0	5, 17%	Hypoth: 1, 3.3%	1, 3.3%	1, 3.3%	Sinusitis: 1 (3.3%), hyponatremia: 1 (3.3%)
Benveniste et al. 2005	96	0	0	29, 30%	15, 15.6%	5, 5.2%	1, 1%	CN palsy: 1, graft site infection: 1, sinusitis: 5, epistaxis: 1

CN = cranial nerve; HCP = hydrocephalus; Hypopit = hypopituitarism; Hypoth = hypothyroidism; MI = myocardial infarction; PE = pulmonaryembolism.

* Represents a case of hypocortisolemia.

† Patient also had meningitis.

The importantsurgical step to facilitate efficient tumor resection was the use of binostril four handed endoscopic technique with wide sphenoidotomy and obtaining adequate sellar exposure to have the full panoramic view ofthe carotid prominences, medial and lateral optico-carotid recesses, planum and clivus. With this technique we achieved total resection of the all 8 cases with residual adenomas and 8 out of 11 recurrent cases involving the sellar and suprasellar area [2, 4, 9,18,20,24,27,29].

Extensive tumor extension into the cavernous sinus is a barrier against total resection of pituitary adenoma disregarding the nature of the adenoma and whether it is de novo or recurrent. Some pituitary adenomas like growth hormone secreting adenomas had aggressive inherent biology enabling them to more easily invade surrounding dural and/or bony structures with early parasellar extension into cavernous sinus. The challenge of surgery inside the cavernous sinus increase markedly in recurrent cases particularly if the tumor is fibrous or if the patient was given radiation after the initial surgery. Aggressive attempts of tumor resection inside the cavernous sinus is hazardous and increase the potential surgical risks of vascular injuries and ocular cranial nerves palsy. Among the 19 cases with residual tumors; 11 (57.9%) cases had tumor extension to the cavernous sinus and from the 11 cases with residual tumor in the cavernous sinus; we were able to achieve total tumor resection in one case at the 2nd surgery and in 10 cases; a residual tumor was left in the cavernous sinus for further adjuvant treatment and the reason for doing the redo surgery was that the tumor growth after the initial surgery and reduction of the tumor size has the advantage of reducing the mass effect to improve the visual manifestation, better achieving hormonal control in functioning adenomas and would improve the response and reduce the associated morbidities of subsequent adjuvant treatment[1,2,4,5,7,8,17,21,28].

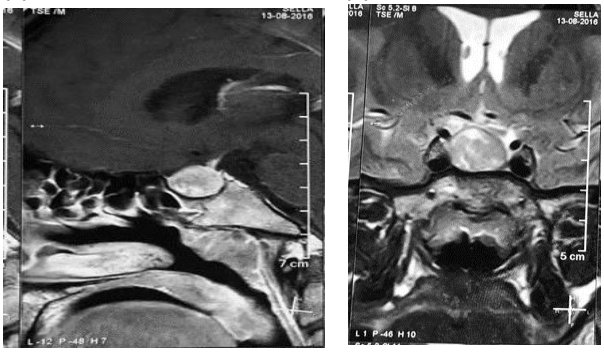
The extent of tumor resection for recurrent adenomas is addressed in many studies in the literature and the growth total resection could be achieved endoscopically in 40% to 63%. The studies of microscopic resection of recurrent pituitary adenomas had similar results with chance of total adenoma resection up to 60%. Although the similarity of the results in the literature; we did all the recurrent cases endoscopically as the we had 19 cases (63.3%) of residual tumor that kept growing after the initial surgery and in 11 of them (57.9%); the residual tumor was in the parasellar region which is a hidden location for the microscopic view. Moreover, the extended endoscopic trans-sphenoidal approach allowed better visualization and hence removal of the sellar and suprasellar component of the tumor than what could be done microscopically. Regardless of the better visualization and tumor manipulation, cavernous sinus invasion remained our greatest limiting factor. The overall Gross total resection in our study was achieved in 17 patients (56.6%) while subtotal and partial resection in 13 patients (43.4%) which is similar to what achieved in other reports in the literature [3,4,6,11,13,14,15,18,20,22,26,27].

Reoperations on any recurrent tumor had a higher incidence of complication in comparison for operating a newly developed tumor. Rates of complications for recurrent pituitary tumors in many studies the literature ranged from 9.4% to 31.3%. Presence of scars and adhesions interfere with natural dissection plans adding difficulties for surgery. The challenge is valid for reoperation on recurrent adenomas with higher incidence of morbidities. One critical complication is the occurrence of CSF rhinorrhea after surgery. Intraoperative CSF leakage could be encountered easily because of scaring of the initial surgery and easily injuring the arachnoid during tumor

dissection. Multilayer reconstruction is crucial and if vascularized flaps could be obtained during the redo surgery; it was the best to prevent post-operative CSF leakage. In cases we were not able to harvest vascularized flap; we obtained fascialata and fat graft with fibrin glue to do solid reconstruction when we faced intra-operative CSF leakage. Then lumbar drain was left up to 3 days after surgery. Despite our aggressive reconstruction plan; 3 cases had post-operative CSF rhinorrhea; 2 passed conservatively and one required another endoscopic repair. The incidence of post-operative pituitary hormonal insufficiency was slightly higher with surgery for recurrent adenomas. A meta-analysis of the result endoscopic surgery for recurrent adenomas reported transient DI in 7%, permanent DI in 2.5%, and anterior pituitary hormonal insufficiency in 1.5%. We had 2 cases (6.6%) of transient DI, 2 cases (6.6%) of permanent DI, 3 cases (10%) developed post-operative hypopituitarism requiring long term hormonal replacement therapy. We had a case of mortality from vascular injury during doing a case of recurrent growth hormone secreting adenomas **(1,7,8,10,16,17,19,22,23,29).**

Conclusions

Endoscopic endonasal approach for recurrent or residual pituitary adenomas allows optimum visualization and better delineation of all hidden corridors to achieve the safest maximum resection. Although cavernous sinus involvement still prevents total resection but aggressive safe tumor debulking is essential to reduce the compression manifestation, improve hormonal control and optimize the result of any further adjuvant treatment including radiotherapy or radiosurgery. Adequate endoscopic exposure is crucial to improve the extent of resection and minimize the potential morbidities. Solid reconstruction plan is very important to minimize post-operative CSF leakage.



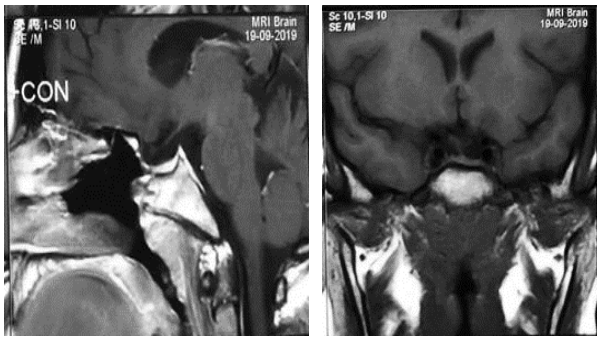


Figure 1. Preoperative MRI image of recurrent nonfunctioning pituitary adenoma sagittal view (a), axial view (b), postoperative follow-up MRI images sagittal view (c), axial view (d).

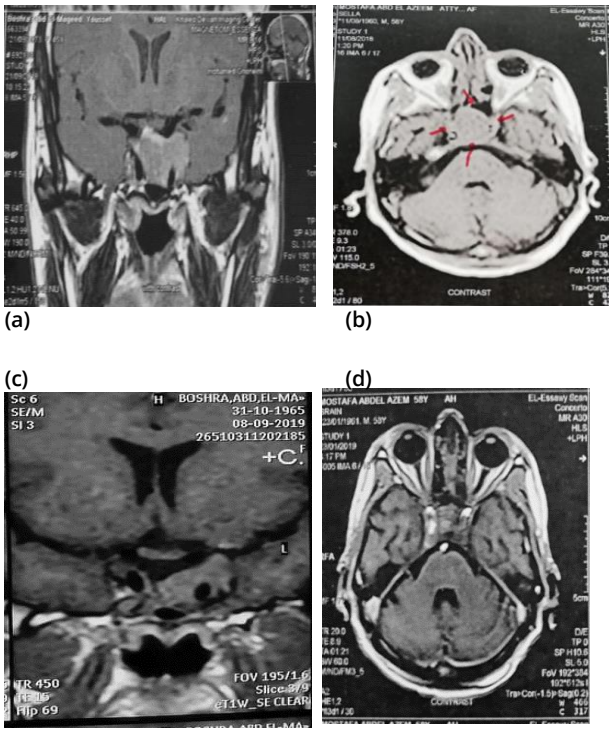


Figure 2. Preoperative MRI image of recurrent growth hormone secreating adenoma sagittal view (a), axial view (b), postoperative follow-up MRI images sagittal view (c), axial view (d).

Abbreviations

EOR=extent of resection, COZ= cranio-orbito-zygomatic, ICT= intracranial tension, CSF= cerebrospinal fluid, OGM= olfactory groove meningioma, GTR= gross total resection, STR= subtotal resection, TSM= tuberculum sellae meningioma, PSM= planum sphenoidale meningioma.

References

1. Yamada S, Fukuhara N, Oyama K et al. Repeat transsphenoidal surgery for the treatment of remaining

or recurring pituitary tumors in acromegaly. Neurosurgery 67:949–956, 2010

2. Tajudeen BA, Mundi J, Suh JD, Bergsneider M et al. Endoscopic endonasal surgery for recurrent pituitary tumors: technical challenges to the surgical approach. J NeurolSurg B Skull Base 76:50–56, 2015
3. Wang S, Xiao D, Wang R et al. Repeated transsphenoidal surgery for resection of pituitary adenoma. J CraniofacSurg 26:452–455, 2015
4. McLaughlin N, Eisenberg AA, Cohan P et al. Value of endoscopy for maximizing tumor removal in endonasaltranssphenoidal pituitary adenoma surgery. J Neurosurg 118:613–620, 2013
5. Krings JG, Kallogjeri D, Wineland A et al. Complications following primary and revision transsphenoidal surgeries for pituitary tumors. Laryngoscope 125:311–317, 2015
6. Hwang JM, Kim YH, Kim JW et al. Feasibility of endoscopic endonasal approach for recurrent pituitary adenomas after microscopic transsphenoidal approach. J Korean NeurosurgSoc 54:317–322, 2013
7. Jahangiri A, Wagner J, Han SW et al. Morbidity of repeat transsphenoidal surgery assessed in more than 1000 operations. J Neurosurg 121:67– 74, 2014
8. DhandapaniS, Singh H, Negm HMet al. Endonasal endoscopic reoperation for residual or recurrent craniopharyngiomas. J Neurosurg2016.
9. FerreliF, Turri-Zanoni M, CanevariFRet al. Endoscopic endonasal management of non-functioning pituitary adenomas with cavernous sinus invasion: a 10- year experience. Rhinology 53:308–316,2015
10. HofstetterCP, Nanaszko MJ, Mubita LLet al.Volumetric classification of pituitary macroadenomas predicts outcome and morbidity following endoscopic endonasaltranssphenoidal surgery. Pituitary 15:450–463, 2012
11. Cavallo LM, SolariD, TasiouAet al. Endoscopic endonasaltranssphenoidal removal of recurrent and regrowing pituitary adenomas: experience on a 59-patient series. World Neurosurg 80:342–350, 2013
12. Dehdashti AR, Ganna A, Karabatsou K et al.Pureendoscopic endonasal approach for pituitary adenomas: early surgical results in 200 patients and comparison with previous microsurgical series. Neurosurgery 62:1006–1017,2008
13. Alahmadi H, Dehdashti AR, GentiliF: Endoscopic endonasal surgery in recurrent and residual pituitary adenomas after microscopic resection. World Neurosurg 77:540–547,2012
14. CappabiancaPandSolari D: The endoscopic endonasal approach for the treatment of recurrent or residual pituitary adenomas: widening what to see expands what to do? World Neurosurg 77:455–456,2012
15. Rudnik A, Zawadzki T, Galuszka-Ignasiak B et al. Endoscopic transsphenoidal treatment in recurrent and residual pituitary adenomas-first experience. Minim InvasNeurosurg 49:10-14, 2006
16. Santoro A, Minniti G, Ruggeri A et al. Biochemical remission and recurrence rate of secreting pituitary adenomas after transsphenoidaladenomectomy: long-term endocrinologic follow-up results. SurgNeurol 68:513- 518, 2007
17. Karabatsou K, O'Kelly C, Ganna A et al. Outcomes and quality of life assessment in patients undergoing endoscopic surgery for pituitary adenomas. Br J Neurosurg 22:630-635, 2008.
18. Kelley RT, Smith JL, Rodzewicz GM.Transnasal endoscopic surgery of the pituitary: modifications and results over 10 years. Laryngoscope 16:1573-1576, 2006.
19. Kitano M, Taneda M, Shimono T et al. Extended transsphenoidal approach for surgical management of pituitary adenomas invading the cavernous sinus. J Neurosurg 108:26-36, 2008.
20. Knosp E, Steiner E, Kitz K et al. Pituitary adenomas with invasion of the cavernous sinus space: a magnetic resonance imaging classification compared with surgical findings. Neurosurgery 33:610- 617, 1993.
21. Kurosaki M, Luedecke DK, Abe T: Effectiveness of secondary transnasal surgery in GH-secreting pituitary adenomas. Nedcor J 50:635-642, 2003.
22. Laws ERJr, Foda NC, Redmond MJ: Transsphenoidal surgery following unsuccessful prior therapy. An assessment of benefits and risks in 158 patients. J Neurosurg 63:823-829, 1985.
23. Losa M, Mortini P, Barzaghi R et al. Early results of surgery in patients with nonfunctioning pituitary adenoma and analysis of the risk of tumor recurrence. J Neurosurg 108:525-532, 2008.
24. Mattozo CA, Dusick JR, Esposito F et al. Suboptimal sphenoid and sellar exposure: a consistent finding in patients treated with repeat transsphenoidal surgery for residual endocrine-inactive macroadenomas. Neurosurgery 58:857-865, 2006.
25. Benveniste RJ, King WA, Walsh J et al. Repeated transsphenoidal surgery to treat recurrent or residual pituitary adenoma. J Neurosurg 102:1004-1012, 2005.
26. Negm HM, Al-Mahfoudh R, Pai M et al. Reoperative endoscopic endonasal surgery for residual or recurrent pituitary adenomas. J Neurosurg 127:397–408, 2017
27. Cappabianca P, Alfieri A, Colao A et al. Endoscopic endonasal transsphenoidal surgery in recurrent and residual pituitary adenomas: technical note. Minim InvasNeurosurg 43:38-43, 2000.
28. Cappabianca P, Cavallo LM, Colao AM et al. Surgical complications associated with endoscopic endonasaltranssphenoidal approach for pituitary adenomas. J Neurosurg 97:293-298, 2002.
29. Chang EF, Zada G, Kim S et al. Long-term recurrence and mortality after surgery and adjuvant radiotherapy for nonfunctional pituitary adenomas. J Neurosurg 108:736-745, 2008.

Romanian Neurosurgery (2020) XXXIV (1): pp. 85-88
10.33962/roneuro-2019-081
www.journals.lapub.co.uk/index.php/roneurosurgery

A lateral meningocele in a 48 years lady revealed by a CSF fistula. Exceptional case

Ibrahim Assoumane[1,2], Nadhim Benmedakhene[1], Adakal Ousseini[3], Bachir Sabrina[1], Nadia Lagha[1], Sidi Said Abderahmanne[1], Abdelhalim Morsli[1]

[1] Department of Neurosurgery CHU Bab El Oued, Algiers, ALGERIA
[2] Department of Neurosurgery Maradi Reference Hospital, NIGER
[3] Department of general surgery, Faculty of Health Sciences, University of Maradi, NIGER

ABSTRACT

Background: Lateral meningocele is defined by the presence of protrusions of the arachnoid and the dura matter extending laterally through inter- or intravertebral foramina. It is an extremely rare condition; to the best of our knowledge, only a few cases are reported in the literature and most of them in childhood.

Case presentation: Authors reported a case of a 48 years old lady who consulted for a lombo-sacral mass right-sided with a CSF fistula. The Spinal MRI objectified a meningocele lateralized in the right side associated with multiples malformations. The patient underwent surgery and the meningocele was closed after excision of the associated subcutaneous lipoma. The long-term outcome was favourable and the follow up was assured by clinical examination monthly in the first year.

Conclusion: Lateral meningocele is very rarely reported, it is usually associated with multiples malformations. Surgical treatment is a good option for treatment for avoiding complications. The prognostic depends on the preoperative status and the associated malformations.

Keywords
lateral,
meningocele,
spinal sacral

✉
Corresponding author:
Ibrahim Assoumane

Department of Neurosurgery CHU Bab El Oued, Algiers, Algeria

assoubrahim18@gmail.com

INTRODUCTION

Lateral meningocele is defined by the presence of protrusions of the arachnoid and the dura matter extending laterally through inter- or intravertebral foramina.

These often occur in the setting of Marfan syndrome, neurofibromatosis type 1or lateral meningocele syndrome but may also be seen as isolated anomalies.

It is extremely rare condition, and only few cases are reported in the literature and most of them in childhood. The most reported cases are in thoracic and cervical regions. The localization at the sacral spine is very infrequent [1]

Here we report the management of a fistulized lumbo sacral lateral meningocele in a 48 years old lady in the department of neurosurgery of Bab El Oued teaching hospital of Algiers Algeria.

Scan to access the online version

CASE PRESENTATION

Madame B N aged of 48 years with past medical history of surgical intervention for a right foot deformation consulted in the emergency unit of our department for Cerebro Spinal Fluid (CSF) leaking in the lumbosacral region through a small mass. The patient reported that she was born with a small mass in the lumbo sacral region but any investigation was performed and she never consulted for. One year before, she was victim of a traffic accident and since date the volume of the mass grows progressively with the extension toward the right side.

At the admission the clinical exam found conscious patient in good general health, temperature 37 degre celcius, complaining of back pain and a paraparesia coted 4/5, a right clubfoot with stigmas of previous surgical interventions. There was no sensory or sphincter disturbance and no <<café au lait>> lesions. In the lumbo sacral region right sided there was a mass of 15 cm of size, well epidermised with large implantation base (Figure 1). Through the masse there was a fistula of Cerebro Spinal Fluid (CSF).

The biological investigations are normal; Spinal MRI (Figure 2) objectified a spinal cord extension up to S1 and a subcutaneous liquid mass extending to the right sacral region as a meningocele. This investigation revealed other malformations; a tethered spinal cord at S1 level, a subcutaneous lipoma, a malformative L2-L3 fusion.

We operated the patient under general anesthesia, on prone position. We performed an "ogive fashion" skin incision, after the excision of the subcutaneous lipoma, we dissected the meningocele and found a fibrous stalk attached in the wall of the meningocele, this fibrous element was detached before the closure of the meningocele using silk 3/0. (Figure 3)

The post-operative outcome was marqued by the relief of back pain and improvement of paraparesia. The patient exited from the hospital seven days after surgery with appointment in three days for total removal of sutures. After 3 days the patient came back for the appointment but we discover a liquid collection, we use a needle gauge to carry out a puncture of 125 cc of serum bloody. The bacteriological study revealed a staphylococcus a coagulase negative. We put her under Acetazolamide 250mg twice daily, oxacillin 1g bid daily.

The evolution was uneventful and the patient was discharged from the hospital after 7 days of treatment with appointment in one month. She was followed up monthly and after 2 years there is no complaint. She recovered the motor disturbance with the help of physiotherapy.

Figure 1. Pre-operative image of the Mass in the lumbo sacral region right sided.

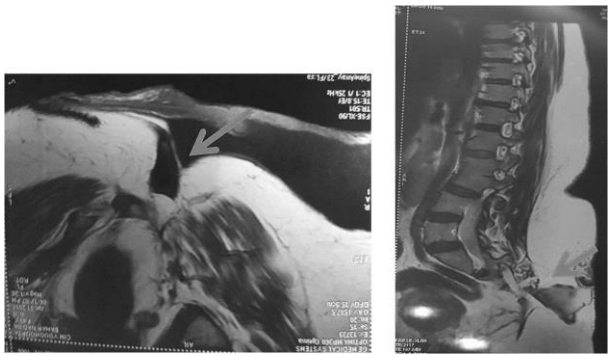


Figure 2. Spine MRI showed a spinal cord extension up to S1 and a subcutaneous liquid mass extending to the right sacral region as a meningocele.

Figure 3. Per operative image showing the walls of the meningocele.

Discussion

Meningomyelocele are the most common forms of neural tube defects. Lateral presentations are extremely rare [2, 5], the incidence of lateral meningoceles was reported to be 0.3% [4]

These lateral presentations often occur in the setting of Marfan syndrome or neurofibromatosis type 1but our patient did not present any café au lait lesions.

Lateral meningocele is also seen in Lateral meningocele syndrome.

This syndrome is a rare disorder, originally described by Lehman et al. in 1977 [3]; Patients with lateral meningocele syndrome present widened spinal canal with scalloping of the posterior surfaces of the vertebral bodies and multiple lateral meningoceles, frequently associated with distinctive craniofacial features such as downslanting palpebral fissures, ptosis, mandibular hypoplasia, a high palate, and skeletal abnormalities such as hypoplasia of the posterior arch of the atlas, short stature, scoliosis, and kyphosis [3], our patient did not present any distinctive craniofacial features.

To the best of our knowledge lateral meningocele in adult not associated with Marfan syndrome or neurofibromatosis type 1or Lateral meningocele syndrome has not been published earlier.

Many meningomyelocele often are associated with tethered cord [2].

Some authors reported that Lateral meningoceles are usually associated with vertebral defects such as hemivertebrae, scoliosis, absence of neural arches on the affected side, and widening of the spinal canal and intervertebral foramina. Scalloping of the pedicles, laminae and vertebral bodies that are adjacent to the meningocele result in an enlarged spinal canal. Butterfly vertebra and segmental anomalies of the vertebral bodies may be found in as many as 43% of affected patients. Sacral anomalies, such as confluent sacral foramina and partial sacral agenesis, occur in up to 50% of reported cases [6,7].

Our patient presented many associated malformations, a tethered spinal cord at S1 level, a subcutaneous lipoma, and a malformative L2-L3 fusion.

A patient with lateral meningoceles may remain asymptomatic or may suffer from paraparesis or back pain [8]. Chronic symptoms in lateral meningoceles usually arise when meningeal protrusions compress against or deform the adjacent structures such as vertebral bodies, nerves, and viscera.

The index patient presented a mass since birth but the clinical sign were back pain and paraparesia. The consultation was motivated by the CSF fistula upon the right sacral mass. Patient leaving in some regions did not come to consultation early because of ignorance or poverty.

Surgical closure is the treatment of choice, because the lateral meningocele does not have spontaneous regression and generally progresses its enlargement with a corresponding increase in the risk of complications like bladder or bowel dysfunction or neurological deficits. Surgical excision may be indicated in cases where giant and symptomatic cysts are present and they are causing bladder or bowel dysfunction or neurological deficits [9].

Conclusion

Lateral meningocele is a very rarely reported entity and only few published cases are reported in the literature. Surgical treatment is a good option of treatment avoiding complications. The prognostic depends on pre-operative status and the associated malformations.

Conflict of interests

None

References

1. Erkulvrawtr S, El Gammal T, Hawkins J: Intrathoracic meningoceles and neurofibromatosis. Arch Neurol 1979, 36:557-559.
2. G Raghavendra Prasad, TM Rashmi, Lateral Meningomyelocele in a Neonate: A Case Report, Journal of Neonatal Surgery 2016; 5(1):4.
3. Alves D, Sampaio M, Figueiredo R, Lea˜o M. 2013. Lateral meningocele syndrome: Additional report and further evidence supporting a connective tissue basis. Am J Med Genet Part A 161A:1768–1772.
4. Gripp KW, Scott CI Jr, Hughes HE, Wallerstein R, Nicholson L, States L, Bason LD, Kaplan P, Zderic SA, Duhaime AC, Miller F, Magnusson MR, Zackai EH: Lateral meningocele syndrome: three new patients and review of the literature. Am J Med Genet 1997, 70:229-239.
5. Shetty DS, Lakhkar BN: Lateral sacral lipomeningomyelocele: a rare anomaly. Neurol India 2002, 50:204-206.
6. Philip N, Andrac L, Moncla A, Sigaudy S, Zanon N, Lena G, Choux M: Multiple lateral meningoceles, distinctive facies and skeletal anomalies: a new case of Lehman syndrome.

Clin Dysmorph 1995, 4:347-351.
7. Heckly A, Carsin B, Poulain P: Diagnosis-related pitfall of a lateral sacral cyst. J Neurosurg Spine 2005, 2:72-74.
8. Mizuno, J, Nakagawa H, Yamada T, Watabe T, 2002. Intrathoracic giant meningocele developing hydrothorax: a case report. J. Spinal Disord. Tech. 15, 529–532.
9. Gohar Mushtaq a,b, Iqbal Hussain c, Jalaluddin A, Khan a, Mohammad A, Kamal. Lateral meningocele with asymmetric canal stenosis: A case study. Saudi Journal of Biological Sciences (2015) 22, 102–105.

Romanian Neurosurgery (2020) XXXIV (1): pp. 89-91
DOI: 10.33962/roneuro-2020-011
www.journals.lapub.co.uk/index.php/roneurosurgery

Spinal intramedullary cysticercosis mimicking spinal tumour

Ahmed Ansari[1], Sheshank Agrawal[2], Sadaf Riyaz[3]

[1] Assistant Professor, Department of Neurosurgery, UPUMS, Saifai, UP, INDIA
[2] Resident Neurosurgery, UPUMS, Saifai, UP, INDIA
[3] Department of Microbiology, UPUMS, Saifai, UP, Etawah, UP, INDIA

ABSTRACT
Neurocysticercosis is a relatively uncommon entity with even more rare spinal intramedullary variety. We present a case of cervico- dorsal intramedullary NCC mimicking spinal tumour with per operative finding mimicking abscess.

Keywords
neurocysticercosis,
intramedullary,
spinal tumour

Corresponding author:
Ahmed Ansari

Department of Neurosurgery,
UPUMS, Saifai, UP, India

ahmed.ansari2@gmail.com

INTRODUCTION

Cysticercosis is caused by larval stage of tape worm Taenia Solium and is the single most common parasitic cause of epilepsy in resourse-poor endemic region.

Prevalence of NCC may reach up to 4% in endemic population with about 1.5-3% prevalence of all NCC as spinal cysticercosis.

Most common site for NCC as described is subarachnoidal space with intramedullary involment, and extremely rare in spinal NCC scenario.

CASE REPORT

A 17years old female presented with lower back ache for last 8 months with bilateral lower limb weakness for last 1 months which was gradual in onset and progressive with bowel disturbance and urinary complaints.

On neurological examination, there was motor weakness with 0/5 power in bilateral lower limb with exaggerated deep tendon reflexes in lower limbs.

The MRI reveled a solitory focal well defined rounded intramedullary mass lesion at C7-D1 level. The lesion showed T2 hyperintens rim with central T2 hyperintesity and was isointense on T1 images (Figure 1). Mild perifocal hyperintensity suggestive of edema but no other focal lesion observed.

She underwent C7 to D1 laminectomy with mid-line myelotomy with drainage of yellow coloured pus material with biopsy of contained margins (Figure 2). Afterwards irrigation was done and dura was repaired.

Scan to access the online version

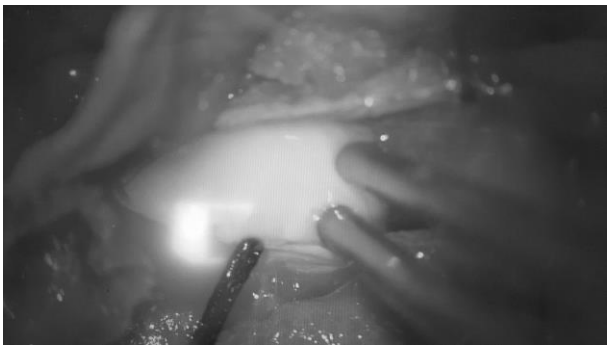
Neuropathological examination revealed examined biopsy specimen with invaginated scolex of cysticercosis with hooklets and surrounding tissue with fibrocollagenous wall with an area of inflammatory reactions comprising of modified histiocytes, few plasma cells and neutrophils.

In early post-operative analysis, no foci of other sources of cysticercosis were found. Her neurological status remained stable and no additional deficits occurred.

Patient was discharged with relief in pain symptoms with bilateral lower limb power improvement to 1/5 with elevation of bladder and bowel symptoms. At two months follow up, the patient developed power of 4/5 in bilateral lower limbs.

Figure 1. MRI T1 WI showing hyperintense lesion at C7-D1 level.

Figure 2. Yellowish coloured fluid draining out of the intramedullary component of the lesion.



Discussion

Worldwide, cysticercosis is the most common parasitic infection affecting the CNS. NCC typically involves the brain parenchyma, intracranial subarachnoid space, or ventricular system and is often self-limited unless hydrocephalus requires surgical intervention. Spinal NCC is rare even in endemic regions, and may require more aggressive management because of the natural confines of the spinal canal. The location and the size of the lesion, and the inflammatory response generated by cyst breakdown are the important factors in the management of spinal NCC.

Spinal cysticercosis can be leptomeningeal, intramedullary or epidural. Leptomeningeal is the most frequent, intramedullary is quite rare and epidural is an extremely rare form. Spinal involvement is quite rare and the migration of the cysticercus through the ventriculo-ependymal pathway and hematogenous dissemination has been hypothesized to be the possible mechanism. However, Queiroz *et al.* did not find any evidence for ependymal route of spread of intramedullary cysticercosis. Rokitansky, in 1856, firstly described intramedullary cysticercosis. Because of limited space in the spinal canal, mass effect of these lesions is poorly tolerated necessitating for aggressive management.

Blood flow to the brain is approximately 100-fold greater than that of the spine explaining the lower incidence of spinal cysticercosis. In the spine, thoracic cord has higher incidence due to high blood flow in this segment. Queiroz et al. estimated the location of cysticerci in spine as: cervical-34%, thoracic- 44.5%, lumbar-15.5% and sacral-6% .

MRI is the investigation of choice. Mathuriya et al. described MRI findings for various stages of intramedullary cysticercosis. Usually, MRI is described as hypointense rim with hyperintense core on T2WI and hypointense or isointense lesion on T1WI as is our case. However, these are not specific and the same changes can also be present in neoplastic, inflammatory, demyelinating, vascular, and granulomatous diseases. The entire neuraxis should be evaluated to find additional lesions.

In the present case, an isolated intramedullary cystic lesion was demonstrated at C7-D1 with absence of cranial cysticercosis.This is in contrast with the previous hypothesis that concomitant intracranial lesions are present in all patients with

spinal cysticercosis. Our finding is supported by Parmar et al. who found only 2 patients with brain neurocysticercosis among 6 patients with intramedullary cysticercosis. Perifocal edema was present in all of their 6 patients as in ours.

Surgical treatment is indicated in spinal NCC in which patients had severe and progressive neurological dysfunction regardless of whether medical therapy has been attempted. The inflammatory process may be so severe that some cysts cannot be readily or completely resected. Excision of intramedullary NCC lesions has been described as being possible after myelotomy or requiring microsurgical dissection from the parenchyma prior to removal. We performed a 3-level laminectomy plus midline myelotomy to reach the lesion and removed it subtotally in order to preserve the neural tissue.

Albendazole or praziquantel, with or without steroids are used. Albendazole is preferred because its blood levels are improved by corticosteroids, whereas those of praziquantel are diminished.

Finally, we conclude that spinal intramedullary cysticercosis represents a diagnostic challenge and surgery is required to decompress the cord, confirm the diagnosis and provide a route for definitive therapy. Patient recovery may be variable. Despite promising reports, the safety and efficacy of medical treatment remains unproved.

References

1. Alsina GA, Johnson JP, McBride DQ, et al. Spinal neurocysticercosis. Neurosurg Focus 2002; 12: e8.
2. Sheehan JP, Sheehan J, Lopes MB, et al. Intramedullary spinal cysticercosis. Case report and review of the literature. Neurosurg Focus 2002; 12: e10.
3. Singh P, Sahai K. Intramedullary cysticercosis. Neurol India 2004; 52: 264-5.
4. Mathuriya SN, Khosla VK, Vasishta RK et al. Intramedullary cysticercosis: MRI diagnosis. Neurol India 2001; 49: 71-4.
5. Homans J, Khoo L, Chen T, et al. Spinal intramedullary cysticercosis in a five-year-old child: case report and review of literature. Pediatr Infect Dis J 2001; 20: 904-8.
6. De Souza Queiroz L, Filho AP, Callegaro D, et al. Intramedullary cysticercosis. Case report, literature review and comments on pathogenesis. J Neurol Sci 1975; 26: 61- 70.
7. Torabi AM,Quiceno M, Mendelsohn DB, et al. Multilevel intramedullary neurocysticercosis with eosinophilic meningitis. Arch Neurol 2004; 61: 770-2.
8. Parmar H, Shah J, Patwardhan V, et al. MR imaging in intramedullary cysticercosis. Neuroradiology 2001; 43: 961-7.
9. Mohanty A,Venkatrama SK, Das S. Spinal intramedullary cysticercosis. Neurosurgery 1997; 40: 82-7.
10. Sharma BS, Banerjee AK, Kak VK. Intramedullary spinal cysticercosis. Case report and review of literature. Clin Neurol Neurosurg 1987; 89: 111-6.

Romanian Neurosurgery (2020) XXXIV (1): pp. 92-100
DOI: 10.33962/roneuro-2020-012
www.journals.lapub.co.uk/index.php/roneurosurgery

The extended pterional approach for midline anterior skull base meningiomas. Technical considerations and clinical outcome

Ahmed Nageeb Mohamed Taha, Hosam Shata Mohamed, Mahmoud Saad Mohamed

Neurosurgery Department, Mansoura Collage of Medicine, Mansoura University, EGYPT

ABSTRACT

Background: Various surgical approaches for the management of midline anterior skull base meningiomas exist in the literature. The main surgeon target is proper selection the appropriate approach that achieves total removal of the lesion without causing morbidity or mortality and facilitates safe effective removal of the tumour.

Objectives: To evaluate the role of the extended pterional approach for excision of midline anterior skull base meningiomas as regarding the effectiveness, extent of resection and surgical outcome.

Patients and methods: This retrospective study involved 23 cases with midline anterior skull base meningiomas resected through the extended pterional approach. Patients' clinical data, operative notes, imaging studies and clinical follow-up data were analyzed and evaluated.

Results: Tumors studied were 9 olfactory groove meningiomas, 8 tuberculum Sellae meningiomas, 4 planum sphenoidale meningiomas and 2 diaphragma sellae meningiomas. Gross total resection tumour excision in 15 cases (64.5%), subtotal excision in 5 cases (21.5%) and partial excision in 3 cases (14%). Complications were diabetes insipidus (2 cases 8.6%), CSF rhinorrhea (3 cases 12.9%) and visual deterioration (3 cases 12.9%). We had two cases of mortality.

Conclusion: The extended pterional approach allows safe and effective removal of midline anterior skull base meningiomas. It expands the exposure offered by the classic pterional approach and minimizing the necessity for applying fixed brain retraction with good cosmetic outcome and less approach-related morbidities in comparison with the extensive skull base approaches.

Keywords
extended pterional,
meningioma,
skull base,
extent of resection

Corresponding author:
Mahmoud Saad Mohamed

Neurosurgery Department,
Mansoura Collage of Medicine,
Mansoura University, Egypt

dr_mhmodsaad@yahoo.com

Scan to access the online version

INTRODUCTION

Meningiomas are benign slowly growing tumors originating from arachnoid cap cells and represent almost 20% of primary intracranial tumors. Although it's benign nature; the existence of meningiomas in certain location is challenging for neurosurgeons. Meningiomas occurring in the midline anterior skull base are among those challenging cases. Depending on the site of origin; midlineanterior skull base meningiomas are classified into: Olfactory groove menin-

giomas planum sphenoidale meningiomas, tuberculum sellae meningiomas, diaphragma sellae meningiomas and dorsum sellae meningiomas. The clinical presentation is variable and varies according to site or origin and size of the tumor but commonly present with frontal manifestations, headache, visual impairment and occasionally manifestation of hypothalamic dysfunction. The surgical challenge of these tumors is how to achieve radical tumor resection without endangering the important neurovascular structures in the vicinity of these tumors. One important step to achieve such goal is choosing the proper approach to achieve adequate tumor resection with minimal morbidities including the approach related complications [2,5,7,9,13,18,22,26].

The surgical approaches utilized to remove midline anterior skull base meningiomas include the pterional, the cranio-orbito-zygomatic, the sub-frontal, and the anterior interhemispheric approaches. Each approach has its advantage and its limitations. The appropriate approach should allow adequate tumor exposure, easily dissection from the surrounding important structures without the need for applying excessive brain retraction. The goal is usually achieved with approaches including the cranio-orbito-zygomatic approach and despite it provide the surgeon with adequate exposure he needs but the approach has its potential functional and cosmetic morbidities [1,4,7,10, 14,18,20,28].

In this study; we reported our experience with extended pterional approach as it allow safe and adequate exposure to most of the midline anterior skull base meningiomas and discussing our surgical results for such challenging meningiomas.

Patients and methods

Retrospective study including twenty-three patients with midline anterior fossa meningiomas were operated up on via the extended pterional approach in the Neurosurgery Department, Mansoura University during the period from February 2015 till July 2019. Patients' demographic, clinical, radiological and operative data are collected and retrospectively analyzed. Duration of clinical presentation varied from 6 to 72 months. Origin of the meningiomas was assessed from preoperative magnetic resonance imaging studies and confirmed from the surgeon operative data. The extent of tumor resection was evaluated via the operative notes and postoperative magnetic resonance imaging studies done 3 months after surgery.

Patients were operated up on via the extended pterional approach that include the Yasergil standard pterional approach with modifications including extension of the craniotomy to the frontal bone to allow access via the sub-frontal corridor and adding osteotomy along the lateral sphenoid wing toexpose the superior orbital fissure and drilling the orbital roof to flatten its surface and expand the exposure via the sub-frontal corridor. The dura is opened in a curvilinear fashion over the sylvian fissure and the incision is directed toward the falciform ligamentproviding unobstructed working angles for the para-sellar and sub-frontal corridor.

We routinely do a post contrast computed tomography scans in the first day after surgery to check for any approach related problems. All patients were then followed up by doing Magnetic Resonance Imaging study 3 months after surgery and then yearly. The mean follow-up period was 26 months, range (6-50 months).

Results

Retrospective analysis of 23 patients with midline anterior fossa meningiomas underwent surgery through the extended pterional trans-sylvian approach. The range of patients' age in our study from22 years to 63 years (the mean age was 51.96±10.81 standard deviation. There was a significant female predominance (n=18, 78.3%) females and males were (n= 5, 21.7%).

Duration of symptoms ranged from 5 to 108 months (mean: 10 months). Visual diminution was the most common clinical presentation in our patients in 18 cases (78.2%), followed by headache in 16 cases (69.5%) then frontal manifestations in 8 cases (34.8%) and anosmia occurred in 4 cases (17.4%), fundoscopic examination: bilateral papilledema was detected in 15 cases (65.2%). Unilateral optic atrophy was in 4 cases (17.4%), bilateral optic atrophyin 3 cases (13.1%). Unilateral papilledema with optic atrophy in the other side (Foster-Kennedy syndrome) occurred in 2 cases (8.7%) (Table 1).

The origin of the meningioma was the olfactory groove in 9 cases, thetuberculum sellae in 8 cases, the planum sphenoidale in 4 cases and the diaphragma sellae in 2 cases. Histopathological diagnosis of the meningiomas were; meningothelial

type in 10 cases, transitional type in 6 cases, psammomatous type in 4 cases, fibrous type in 2 cases and atypical type in only one case. We had 12 cases of medium sized meningiomas, 9 cases of large sized meningiomas and 2 cases of giant meningiomas (Table 2).

Gross total resection (Simpson grades I) wasachieved in 15 patients (65.3%). Subtotal resection (Simpson grades II &III) was achieved in 5 cases (21.7%). Partial resection (Simpson grades IV) was achieved in 3 cases (13.1%). Table (3) demonstrate the extent of resection in correlation to meningioma location and tumor size. Total resection was more feasible for olfactory groove meningiomas (in eight of the nine cases in our study). Also, the extent of total resection was achieved more in small sized meningiomas compared to medium and large sized ones. Regarding the visual outcome (table 7); 13 patients (56.5%) out of the 18 cases presented with visual disturbances showed post-operative visual improvement, 5 patients (21.7%) remained stable while 2 patients (8.6%) had post-operative visual deterioration which was transient in one of them.

No major post-operative morbidities occurred in our operated cases (Table 4). No post-operative morbidities happened in 12 cases (52.2%). Frontal manifestations were the most common morbidities and occurred in 4 cases (17.4%). 3 cases experienced postoperative diabetes insipidus (12.9%) (transient in 2 cases). Post-operative seizures occurred in 2 cases. 2 cases (8.6%) had post-operative visual deterioration that was transient in one of them. One case (4.3%) of tuberculum sellae meningioma developed transient CSF rhinorrhea that was managed conservatively with transient lumbar drain.

The average duration for follow-up in our study was 25 months, ranged from 6 to 50 monthswith no tumor recurrences reported in the follow-up period. Four cases were offered post-operative adjuvant treatment (17.3%) in the form of Gamma knife radiosurgery in three cases and conformal 3-dimensional radiotherapy in one patient. We had two mortalities (8.6%) in our study (table 4). One mortality happened from intraoperative vascular injury; resulted in massive infarction of both frontal lobes and the 2nd case died from severe hypothalamic dysfunction.

ILLUSTRATED CASES

Case 1: 64 years old female patient with suprasellar meningioma (Fig. 1 a & b); subtotal resection was achieved leaving a small part of tumor attached to the pituitary stalk (Fig. 1 c & d).

Case 2: 39 years old male patient with large suprasellar meningioma extending superiorly to 3rd ventricle (Fig. 2 a & b) that totally removed via extended pterional approach (Fig. 2 c&d).

Case 3: 50 years old female patient with suprasellar meningioma (Fig. 3 a & b) that totally excised (Fig. 3 c & d).

Fig. 1 (a)

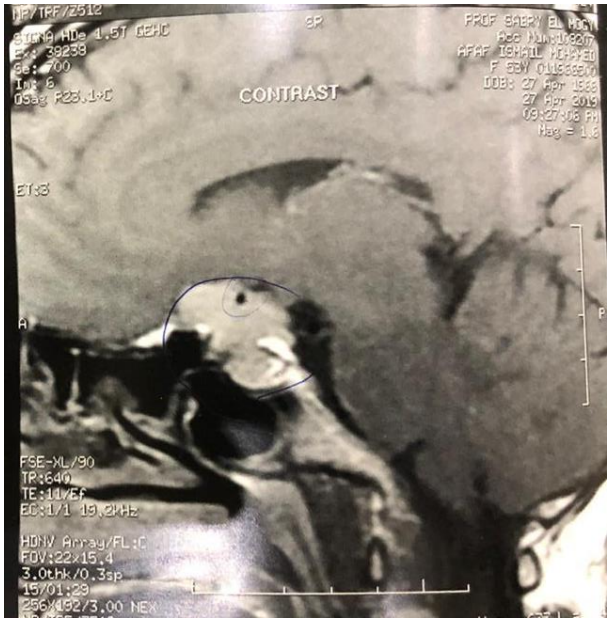


Fig. 1 (b)

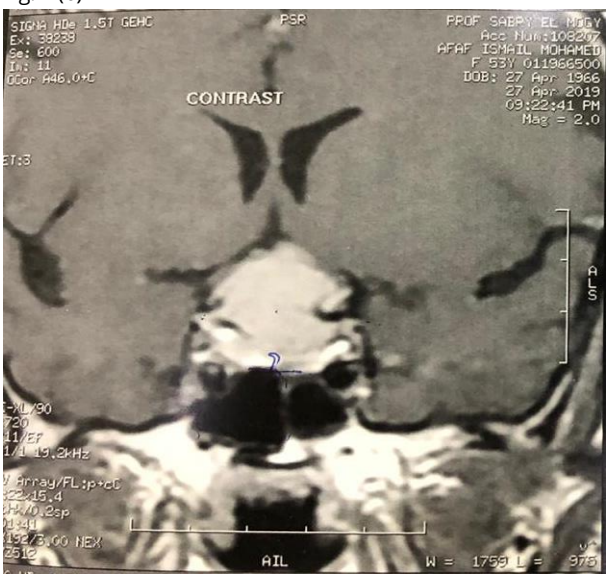


Fig. 1 (c)

Fig. 1 (d)

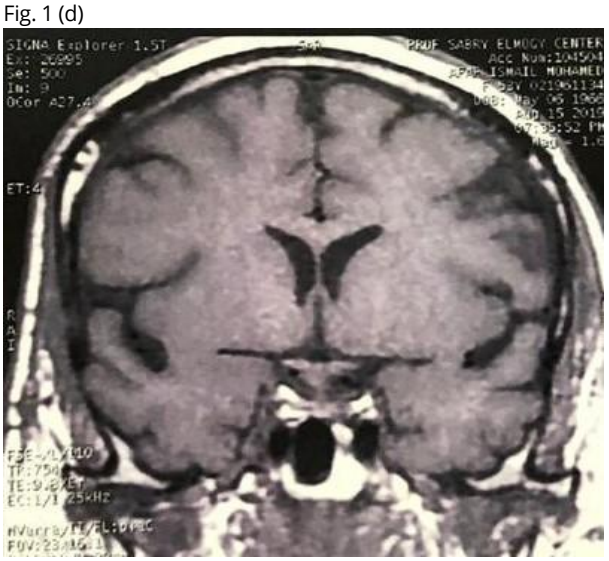


Figure 1. Preoperative MRI image of suprasellar tumor sagittal view (a), coronal view (b), postoperative follow-up MRI images sagittal view (c), coronal view (d).

Fig. 2 (a)

Fig. 2 (b)

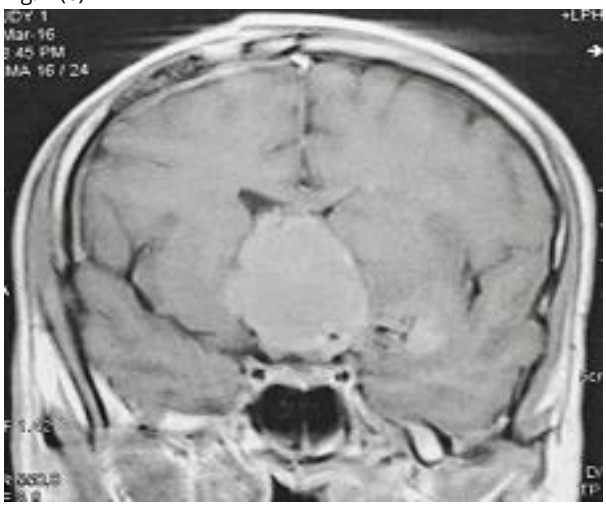


Fig. 2 (c)

Fig. 2 (d)

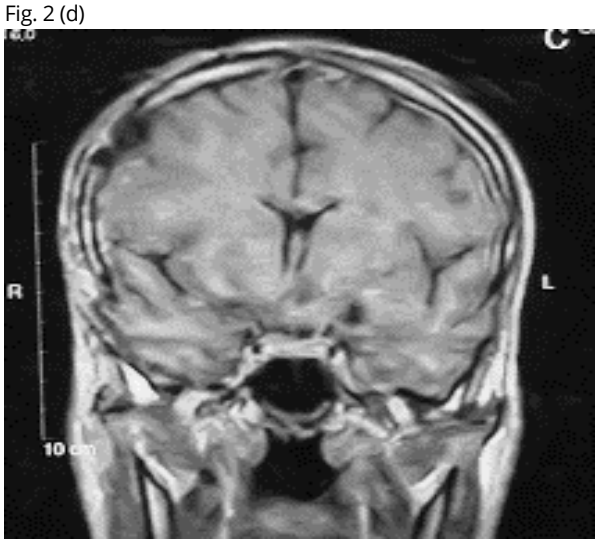


Figure 2. Preoperative MRI image of suprasellar tumor sagittal view (a), coronal view (b), postoperative follow-up MRI images sagittal view (c), coronal view (d).

Fig. 3 (a)

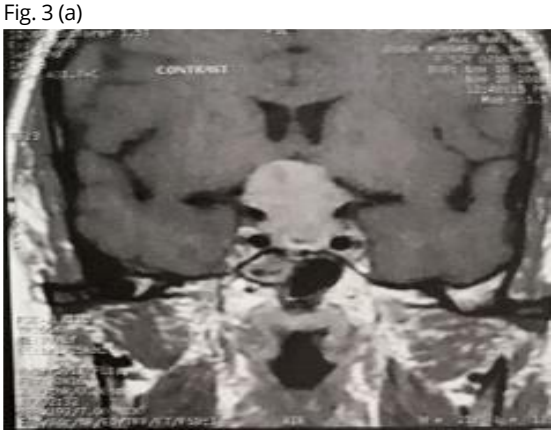


Fig. 3 (b)

Fig. 3 (c)

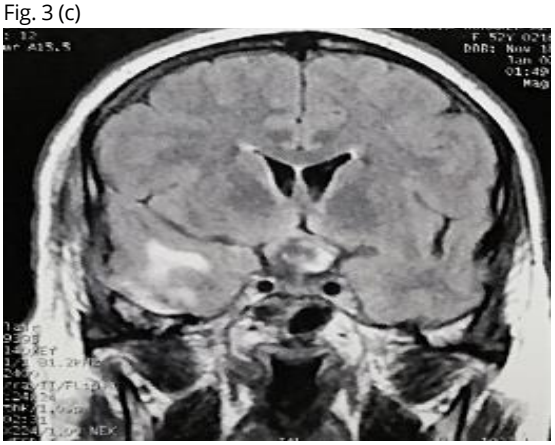


Fig. 3 (d)

Figure 3. Preoperative MRI image of suprasellar tumorcoronal view (a), sagittal view (b), postoperative follow-up MRI images coronal view (c), s agittal view (d).

Table 1. Clinical presentation of our case series.

Clinical	Number	Percentage
Visual manifestations	18	78.2%
Headache	16	69.6%
Behavior changes	8	34.8%
Anosmia	4	17.4%
Seizure	3	13.1%
Hormonal disturbance	1	4.3%
Fundoscopy • Bilateral Papilledema • Unilateral optic atrophy • Bilateral optic atrophy • Foster-Kennedy syndrome	 15 4 3 2	 65.2% 17.4% 13.1 8.7

Table 2. Classification of tumor types according to origin, histopathological examinationand tumor size (distribution, average).

Tumor origin	Number	Percentage	Average Size in cm^3 (range of size)
Olfactory Groove	9	(39.1%)	5.14 (3.7 – 6.5)
Tuberculum sellae	8	(34.8%),	3.95 (3.6 - 4.2)
Planum sphenoidale	4	(17.4%)	3.80 (3.5 – 4.3)
Diaphragma sellae	2	(8.7%)	3.65 (3.5 – 3.8)
Histopathological types			
Meningothelial type	10	(43.5%)	
Transitional type	6	(26.1%)	
Psammomatous type	4	(17.4%)	
Fibrous type	2	(8.7%)	
Atypical meningioma (WHO grade II)	1	(4.3%)	
Tumor size			

Medium2-4cm3	12	52.2%
Large 4-6	9	39.2%
Giant >6	2	8.6%

Table 3. Correlation between EOR and location of meningioma and tumor size.

	GTR Grade I	**STR Grade II & III**	**Partial Grade IV**	**Total**
Meningioma location				
Olfactory groove	8 (34.4%)	1(4.3%)	0	9
Planum sphenoidale	4 (17.2%)	2 (8.6%)	2 (8.6%)	8
Tuberculum sellae	2 (8.6%)	1 (4.3%)	1 (4.3%)	4
Diaphragma sellae	1 (4.3%)	1 (4.3%)	0	2
Meningioma size				
2cm - 3.9 cm.	9 (39%)	2 (8.6%)	1 (4.3%)	12
4cm - 5.9 cm.	4 (17.2%)	3 (13%)	2 (8.6%)	9
>6cm	2 (8.6%)	0	0	2

Table 4. Visual and surgical outcome and complication of extended pterional approach in anterior skull base meningiomas.

	Number	***Percentage***
Surgical outcome		
GTR	15	64.5%
STR	5	21.5%
Partial	3	14%
Mortality	2	8.6%
Complications		
Frontal manifestations	4	17.4%
Seizures	2	8.6%
Diabetes insipidus	3	12.9%
Transient CSF rhinorrhea	1	4.3%
Visual deterioration	2	8.6%
Visual outcome		
Improved vision	14	60.7%
Stable vision	5	21.7%
Visual deterioration (one is transient)	2	8.6%

Table 5. Review of literature of case series of microsurgical management of anterior skull base meningiomas

Case series	**Year**	**No. Patients**	**GTR (%)**	**Visual Improvement (%)**	**Recurrence (%)**	**Mortality (%)**	**Years of F/U**
Recent series	2019	23	64.5	78.1	N/A	8.6	4.6
Lynch et al.	2015	38	86	89.4	5.2	2.6	5.7
Romani et al.	2009	65	91	21.4	9	0	3.7
Bassioni et al.	2007	55	100	83.3	8.9	0	N/A
Colli et al.	2007	17	94.1	N/A	0	11.8	4.2
Hentschels and Demonte	2003	13	85	92.3	0	0	2
Goel et al.	2002	70	84	N/A	1.4	2.8	N/A
Jallo&Benjamim	2002	23	86.9	55	4.5	8.6	93
Fahlbusch& Schott	2002	47	98	80	4.2	0	N/A
Zeugaridis et al.	2001	62	N/A	65	N/A	3.2	5.2

Turazzi et al.	1999	37	100	100	0	2.7	4
Al-Mefty	1993	35	91	25	N/A	8.6	N/A
Ojemann	1991	14	71	73	N/A	0	N/A
Solero et al.	1983	55	78	60	3	2.3	N/A
Symon &Rosentein	1977	33	82	N/A	3.1	3	N/A

Discussion

Midline anterior skull base meningiomas have only and exclusively treated through direct microsurgical excision. The main neurosurgical challenge is to achieve total surgical excision with no or minimal postoperative morbidities. However, the surgical difficulties are facilitated by improved microsurgical facilities, more understanding of the microsurgical anatomy through adequate neurosurgical training and progressive learning curve and feasibility of variable surgical approaches. Effective reaching the surgical target with minimal normal anatomical disruption is the cornerstone of the surgeon focus [1,3,4,6,8,11,14,16,25].

A wide range of surgical approaches have been described in the literature totreat midline anterior skull base meningiomas, including the pterional approach with its modifications and the sub-frontal approach with unilateral or bilateral sub-frontal exposure. Each of these approaches has its advantages as well as its limitations. Approach selection is dependent on the tumor size, location, and pattern of extension, tumor relation to the important neurovascular structures and the surgeon's experience and familiarity with the approach [2,3,9,12,15,17,19,25,27].

Many authors advocated the bilateral sub-frontal craniotomy for large symmetrical midline lesions with its advantage of wide exposure of the anterior cranial base and excellent view for dissection of the both anterior cerebral arteries and the optic pathways. Other authors prefer to use the unilateral sub-frontal corridor which can allow tumor resection without the increased risk encountered in the bilateral sub-frontal exposure. In many other cases series; the fronto-lateral approaches including the pterional approach, the cranio-orbito-zygomatic approach and the extended pterional approach were preferred by the neurosurgeons to remove midline anterior skull base meningiomas [1, 3,7,15,20,21,23,24,25,27].

The extended pterional approach provides certain advantages compared to the bi-frontal craniotomy. It provides the shortest distance to the tuberculum sellae. It allows early exposure to the basal cisterns for CSF release to have good brain relaxation to minimize frontal lobe retraction. Also, sylvian fissure dissection provided by the approach allow untethering of the frontal lobe from temporal lobe facilitating full exposure of the neurovascular structures with minimal or no frontal lobe retraction which is difficult to achieve from the bilateral or unilateral sub-frontal exposure. Moreover, the extended pterional approach provides the surgeon with more working angles for tumor exposure and dissection that cannot be achieved from the sub-frontal approach. The extended pterional approach can replace bi-frontal craniotomy for resection of giant midline intradural anterior skull base tumors except for extradural skull base tumors extending to the intradural space [3.4,6,10,15,20,24,25,26,27].

Pterional craniotomy is a highly flexible skull base approach that gives excellent exposure of the anterior cranial fossa, the circle of Willis, and the interpeduncular region. Because of its simplicity, flexibility, efficiency; this approach is most utilized for pathologies along the anterior skull base. The major limitation for this exposure is the need for more frontal lobe retraction for lesion with more superior extension reaching the third ventricle and when the tumor extends inferolateral in skull base. The cranio-orbito-zygomatic approach can expand the exposure for the hidden areas for the classic pterional approach involving the orbital apex, the paraclinoid and parasellar areas, the cavernous sinus and the interpeduncular fossa. Tumors with significant superior extension can be addressed well with expanded inferior-to-superior and medial to lateral operative working angles provided by the cranio-orbito-zygomatic approach. The cranio-orbito-zygomatic approach is a more complex approach and technically demanding with the concern regarding the cosmetic problems due to the

extensive osteotomes necessary for the exposure [2,3,8,9,13,15,22,24,25,26,28].

The extended pterional approach is a modification of the classic pterional approach that obviates the limitation of the classic approach for skull base exposure and minimizes the necessity for the more extensive cranio-orbito-zygomatic approach. The additional osteotomes at the sphenoid wing and orbital roof expands the sub-frontal trajectory to the midline anterior skull base without necessity for more frontal lobe retraction. The osteotomy of the orbital roof gives most of the advantages of that provided by the cranio-orbito-zygomatic approach with less chance for cosmetic deformities [2,3,15,24,25,26].

In our study; the mean age of the cases in our study was 51.96 years. Most of our cases were in their 5th and 6th decades and 78.3% of our cases were females. Visual manifestations were the most presenting symptoms followed by headache, frontal manifestation then anosmia and behavioral changes. The demographic data and the clinical manifestations of our cases were like other reports in the literatures discussing this pathological entity [1,5,6,11,12,21,27].

The onset of clinical presentation for midline anterior skull base meningiomas corelated the origin of the meningiomas. More anteriorly located tumors like olfactory groove meningiomas may reach large size before being symptomatic. Earlier clinical presentation in smaller sized meningiomas originating in a close proximity to the optic nerves and the chiasm. In our study, the average size for the OGMs was 5.14 cm for the TSMs was 3.95 cm while the average size of the PSMs was 3.80 cm [2,4,5,9,11,15,27].

Gross total resection (GTR) was achieved in 15 (65.3%) patients and subtotal resection in 5 (21.7%). The highest percentage of gross total tumor excision was in OGM cases (8 tumors out of 9), followed by PSMs (4 tumors out of 8), and then TSMs (2 tumors out of 4) .The extent of tumor resection of midline anterior skull base meningiomas was addressed in many case series with variation in results depending on what is defined as total or near total resection in each series. Several studies have advocated that attempting gross total resection should not be with the price of endangering the vision or the hypothalamic function. The extent of gross total tumor resection of such meningiomas varied from 35% to 100% (table 5). The appropriate approach selection is crucial to improve the extent of resection as reported in many series. Skull base approaches including the cranio-orbito-zygomatic and cranio-orbital approach facilitated adequate tumor exposure and better achievement of tumor resection but with increasing cosmetic morbidities. The extended pterional approach used in our case series facilitated more adequate exposure of the tumor with minimal cosmetic sequalae [2,4,5,8,9,13,15,18, 19,23,24,25,26,27].

The preservation of vision is one of the cardinaltargets of surgical management. Some case series concluded that better visual outcomes are associated with tumors smaller than 3 cm than tumors larger than 3 cm in diameter[2,8,16]. In our series, improvement of vision occurred in 60.7% of patients and preserved vision with no further deterioration in 21.7%.

Surgery for midline anterior skull base tumors still carry the risk for postoperative morbidities despite the improvement achieved in the modern neurosurgical facilities. In our study, the highest complication rate was frontal manifestations; behavior changes (n=4, 17.4%), diabetes insipidus (n=3, 12.9%), CSF rhinorrhea (n=1, 4.3%) then visual deterioration (n=1, 4.3%). Cushing reported an operative mortality of 27.5%. As a result of the refinements of microsurgical techniques, death rates had declined in subsequent series. Al-Mefty[15] and Solero et al.[27] observed higher mortality rates in patients with tumors exceeding 3 cm in diameter, compared with mortality rates in patients with smaller tumors. In our study there were only 2 mortalities (8.6%). In our series, 11patients (47.8%) harbored large or giant tumors, but we noticed that there was no increase in mortality in this group of patients [2,7,9,15,18,19,22,25].

Several series have been published on midline anterior skull base tumors (table 5) with long term follow up for recurrences beyond 10 years following Simpson 1 or 2 grade resections. In current study, no tumor recurrences recorded during the follow up period which extended up to 54 months. A longer follow up period is crucial for better assessment of the actual recurrence rate [3,6,7,8,15,23,27].

Conclusion

The extended pterional approach allows safe and effective removal of midline anterior skull base meningiomas. It expands the exposure offered by

the classic pterional approach and minimizing the necessity for applying brain retraction with good cosmetic outcome and less approach related morbidities related to the more extensive skull base approaches.

Abbreviations

EOR= extent of resection, COZ= cranio-orbito-zygomatic, ICT= intracranial tension, CSF= cerebrospinal fluid, OGM= olfactory groove meningioma, GTR= gross total resection, STR= subtotal resection, TSM= tuberculum sellae meningioma, PSM= planum sphenoidale
meningioma.

References

1. Colli BO, Carlotti CG Jr, Assirati JA Jr et al. Olfactory groove meningiomas: Surgical technique and follow-up review. Arq Neuropsquiatr 65 (3B):795-9, 2007.
2. Fahlbusch R and Schott W. Pterional surgery of meningiomas of the tuberculum sellae and planum sphenoidale: surgical results with special consideration of ophthalmological and endocrinological outcomes. J Neurosurg 96 (2):235-43, 2002.
3. Yasargil MG. General operative techniques. In: Yasargil MG, editor.Microneurosurgery, Vol. 1: Microsurgical Anatomy of the basal cisterns and vessels of the brain, diagnostic studies, general operative techniques and pathological considerations of the intracranial aneurysms. New York: Georg Thieme/Thieme-Stratton p. 208-33, 1984.
4. Romani R, Laakso A, Kangasniemi M et al. Lateral supraorbital approach applied to tuberculum sellae meningiomas: experience with 52 consecutive patients. Neurosurgery 70 (6):1504-19, 2012.
5. Zivgaridis D, Medele RJ, Muller A et al. Meningiomas of the sellar region presenting with visual impairment: Impact of various prognostic factors on surgical outcome in 62 patients. Acta Neurochir 143: 471-476, 2001.
6. Jallo GI and Benjamin V. Tuberculum sellae meningiomas: microsurgical anatomy and surgical technique. Neurosurgery 51 (6):1432-40, 2002.
7. McDermott MW, Rootman J, Durity FA. Subperiosteal, subperiorbital dissection and division of the anterior and posterior ethmoid arteries for meningiomas of the cribriform plate and planum sphenoidale: technical note. Neurosurgery 36 (6):1215-9, 1995.
8. Nakamura M, Struck M, Roser F et al. Olfactory groove meningiomas: clinical outcome and recurrence rates after tumor removal through the frontolateral and bifrontal approach. Neurosurgery 62 (6 Suppl 3):1224-32, 2008.
9. Aguiar PH, Tahara A, Almeida AN et al. Olfactory groove meningiomas: approaches and complications. J Clin Neurosci 16 (9):1168-73, 2009.
10. Symon L. Olfactory groove and suprasellar meningiomas. In: Krayonbühl H, editor. Advances and technical standards in neurosurgery. Springer p. 67-91, 1977.
11. Goel A, Muzumdar D, Desai KI. Tuberculum sellae meningioma: a report on management on the basis of a surgical experience with 70 patients. Neurosurgery 51 (6):1358-64, 2002.
12. Goel A andMuzumdar D. Surgical strategy for tuberculum sellae meningiomas. Neurosurg Q 15: 25-32, 2005.
13. Simpson D. The recurrence of intracranial meningiomas after surgical treatment. J Neurol Neurosurg Psychiatry 20:22-39, 1957.
14. Spektor S, Valarezo J, Fliss DM, Gil Z, Cohen J, Goldman J et al. Olfactory groove meningiomas from neurosurgical and ear, nose, and throat perspectives: approaches, techniques, and outcomes. Neurosurgery 57 (4 Suppl): 268-80. 2005.
15. Al-Mefty O. Tuberculum sellae and olfactory groove meningiomas. In: Sekhar LN, Janecka IP, editors. Surgery of cranial base tumors. New York: Raven; 1993. p. 507-19.
16. Bassiouni H, Asgari S, Stolke D. Olfactory groove meningiomas: functional outcome in a series treated microsurgically. Acta Neurochir 149 (2):109-21, 2007.
17. Bassiouni H, Asgari S, Stolke D. Tuberculum sellae meningiomas: Functional outcome in a consecutive series treated microsurgically. Surg Neurol 66: 37-45, 2006.
18. De Monte F. Surgical treatment of anterior basal meningiomas. J Neuro-Oncol 29:239-248, 1996.
19. El Gindi S. Olfactory groove meningioma: Surgical techniques and pitfalls. Surg Neurol 54:415–417, 2000.
20. Park CK, Jung HW, Yang SY et al. Surgically treated tuberculum sellae and diaphragm sellae meningiomas: the importance of short-term visual outcome. Neurosurgery 59 (2):238-43, 2006.
21. Refaat MI, Eissa EM, Ali MH. Surgical management of midline anterior skull base meningiomas: experience of 30 cases. Turk Neurosurg 25 (3):432-7, 2015.
22. Mathiesen T and Kihlstrom L. Visual outcome of tuberculum sellae meningiomas after extradural optic nerve decompression. Neurosurgery 59:570-576, 2006.
23. Obeid F and Al-Mefty O: Recurrence of olfactory groove meningiomas. Neurosurgery 53: 534-543, 2003.
24. Turazzi S, Cristofori L, Gambin R. The pterional approach for the microsurgical removal of olfactory groove meningiomas. Neurosurgery 45:821-826, 1999.
25. Lynch, Jose Carlos, Gonçalves et al. The extended pterional approach allows excellent results for removal of anterior cranial fossa meningiomas. Arquivos de Neuro-Psiquiatria 74 (5): 382-387, 2016.
26. Hendricks BK and Cohen GA.The Extended Pterional Craniotomy: A Contemporary and Balanced Approach Operative Neurosurgery opz117, vol.0, 2019.
27. Solero CL, Giombini S, Morello G. Suprasellar and olfactory meningiomas: report on a series of 153 personal cases. Acta Neurochir 67 (3-4):181-94, 1983.
28. Romani R, Lehecka M, Gaal E, et al. Lateral supraorbital approach applied to olfactory groove meningiomas: experience with 66 consecutive patients. Neurosurgery 65 (1): 39-53, 2009.

Romanian Neurosurgery (2020) XXXIV (1): pp. 101-105
DOI: 10.33962/roneuro-2020-013
www.journals.lapub.co.uk/index.php/roneurosurgery

Computed tomography-based morphometric measurements of the atlas (C1) posterior arc

Yasar Karatas[1], **Bulent Kaya**[1], **Mehmet Fatih Erdi**[2], **Fatih Keskin**[2], **Densel Arac**[2], **Emir Kaan Izci**[3], **Mehmet Uyar**[2], **Ali Sami Kıvrak**[1], **Erdal Kalkan**[1]

[1] Medova Hospital, TURKEY
[2] Necmettin Erbakan University, Meram Medical Faculty, TURKEY
[3] Meram Training and Research Hospital, Department of Neurosurgery, TURKEY

ABSTRACT

Study design: Single-center retrospective study

Objectives: This study is performed to determine the anatomic feasibility of the C1 posterior arc screw and help select an optimal screw trajectory in treating patients with craniovertebral junction pathologies.

Material and Methods: We reported a single-centre retrospective study. Forty patients (20 male and 20 female) who underwent cervical computed tomography (CT) were chosen from the hospital records. Based on CT images, we measured left laminar length (LLL), right laminar length (RLL), left laminar angle (LLA), right laminar angle (RLA), left laminar axial thickness (LLAT), right laminar axial thickness (RLAT), left laminar coronal thickness (LLCT), right laminar coronal thickness (RLCT), and craniocaudal angle (CCA) of the C1 posterior arc.

Results: The mean values and standard deviations (SD) for nine parameters at the C1 posterior arc were determined. LLL, RLL, LLCT, and RLCT were statistically longer in men than women. RLAT was bigger in men but there was no statistical difference. RLA was statistically wider in women than men. LLA and CCA were wider in women but there was no statistical difference, LLAT was bigger in women but there was no statistical difference. There was no statistical difference in measurements by age.

Conclusion: The results of this study are important to avoid neurovascular injury and pedicle breakage because of choosing large screw while performing C1 laminar screw fixation.

Keywords
computed tomography, morphometric measurements, atlas (C1) posterior arc

Corresponding author:
Yasar Karatas

Medova Hospital,
Turkey

yasarkrts@gmail.com

Scan to access the online version

INTRODUCTION

The first cervical vertebra is also referred to the atlas[1]. Posterior wiring[2,3], transarticular screws[4], and pedicle or lateral mass screws[5] methods largely used for C1 posterior fixation. Recently, the screw fixation has been preferred instead of wiring or hooks because of providing rapid stability and great fusion rate[6]. However, screw fixation is associated with an increased risk of vertebral artery (VA), spinal cord, and root injury. Missing to identify VA anomalies can cause iatrogenic

VA injury and cerebrovascular accidents in cervical spine surgery[7]. The most dangerous level is the C1-2 level for a posterior approach and C7 for an anterior approach. Therefore, an excellent assessment for the variations in the course of the VA is vital for surgeons working from a posterior approach in the upper cervical spine[8]. In this study, we measured C1 posterior arc parameters to create an alternative surgical way to lateral mass screws and pedicular screws.

Materials and methods

Fourty patients (20 male and 20 female) who underwent cervical computed tomography (CT) between 2017 and 2019 in our hospital were chosen from the hospital's picture archiving and communication system (PACS). None of the patients included in the study had cervical spine or craniovertebral junction trauma. Siemens Somatom Perspective 128 slices CT was used to perform CT examinations and measurements on patients. CT scans were performed by the same team. Heads of all patients were fixed in the same position on computed tomography. We assessed axial, sagittal, and coronal CT cuts and measured nine parameters on the C1 posterior arc. Left and right laminar lengths (LLL, RLL) were calculated by measuring the line from posterior tubercle to transverse foramen (Figure 1). Left and right lamina angles (LLA, RLA), which also mean mediolateral angle, were calculated as the angle of the lamina with the line passing through the anterior and posterior tubercle (Figure 2). Left and right axial laminar thicknesses (LLAT, RLAT) were measured from the middle of the lamina (Figure 3). Left and right coronal laminar thicknesses (LLCT, RLCT) were measured from the middle of the lamina (Figure 4). Craniocaudal angle (CCA) was measured as the angle of the lamina with the line that parallel to the earth (Figure 5). We investigated the difference between men and women and patients under fifty and patients over fifty years. Data were analyzed by SPSS (version 24.0, SPSS Inc.) and expressed as mean ± SD. Comparisons were made using the t-test. Differences among the groups were assessed using the independent samples test. A p value < 0.05 was considered statistically significant.

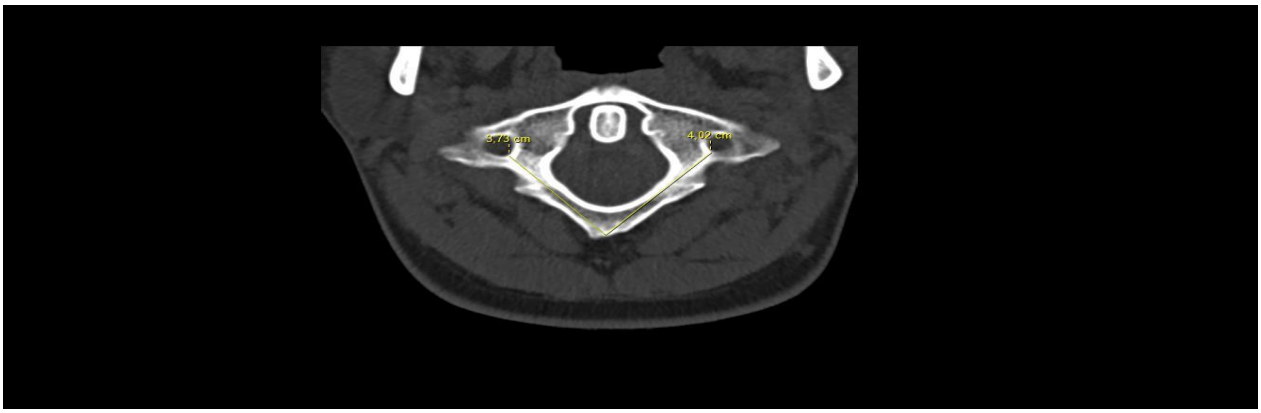
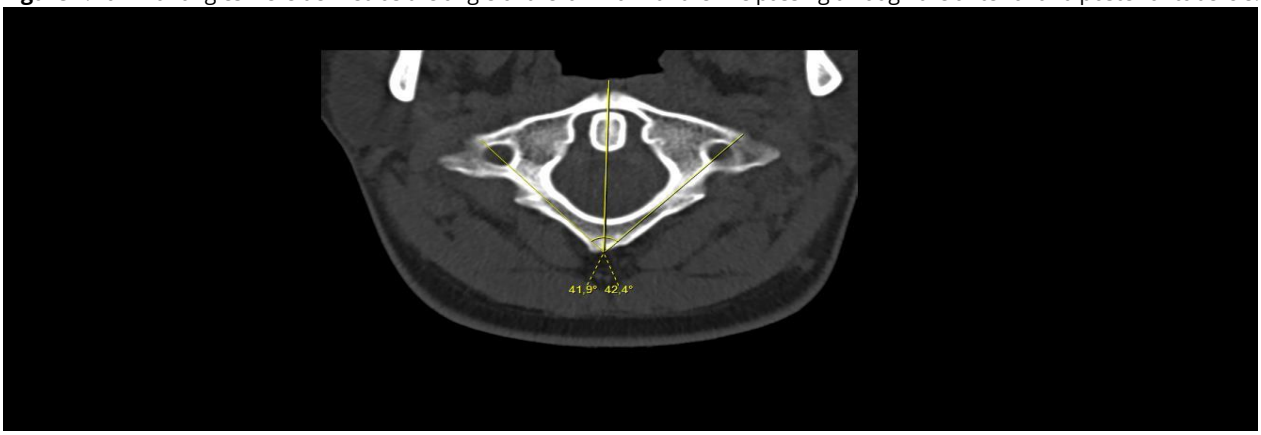


Figure 1. Laminar length was defined as linear measurement from posterior tubercle to transverse foramen both right and left side.

Figure 2. Laminar angles were defined as the angle of the lamina with the line passing through the anterior and posterior tubercle.



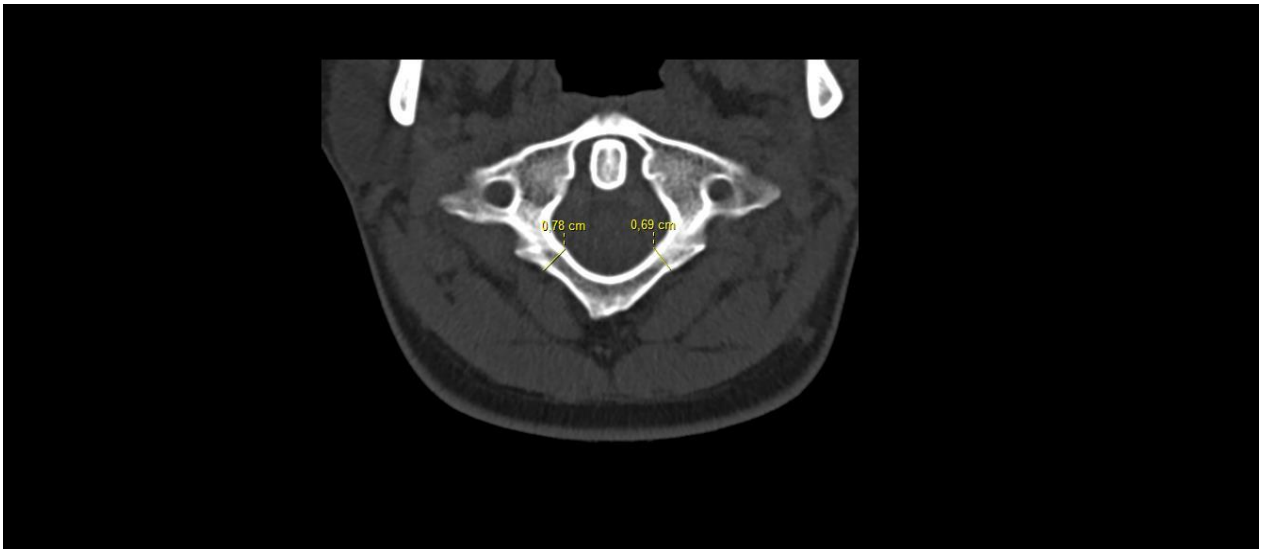
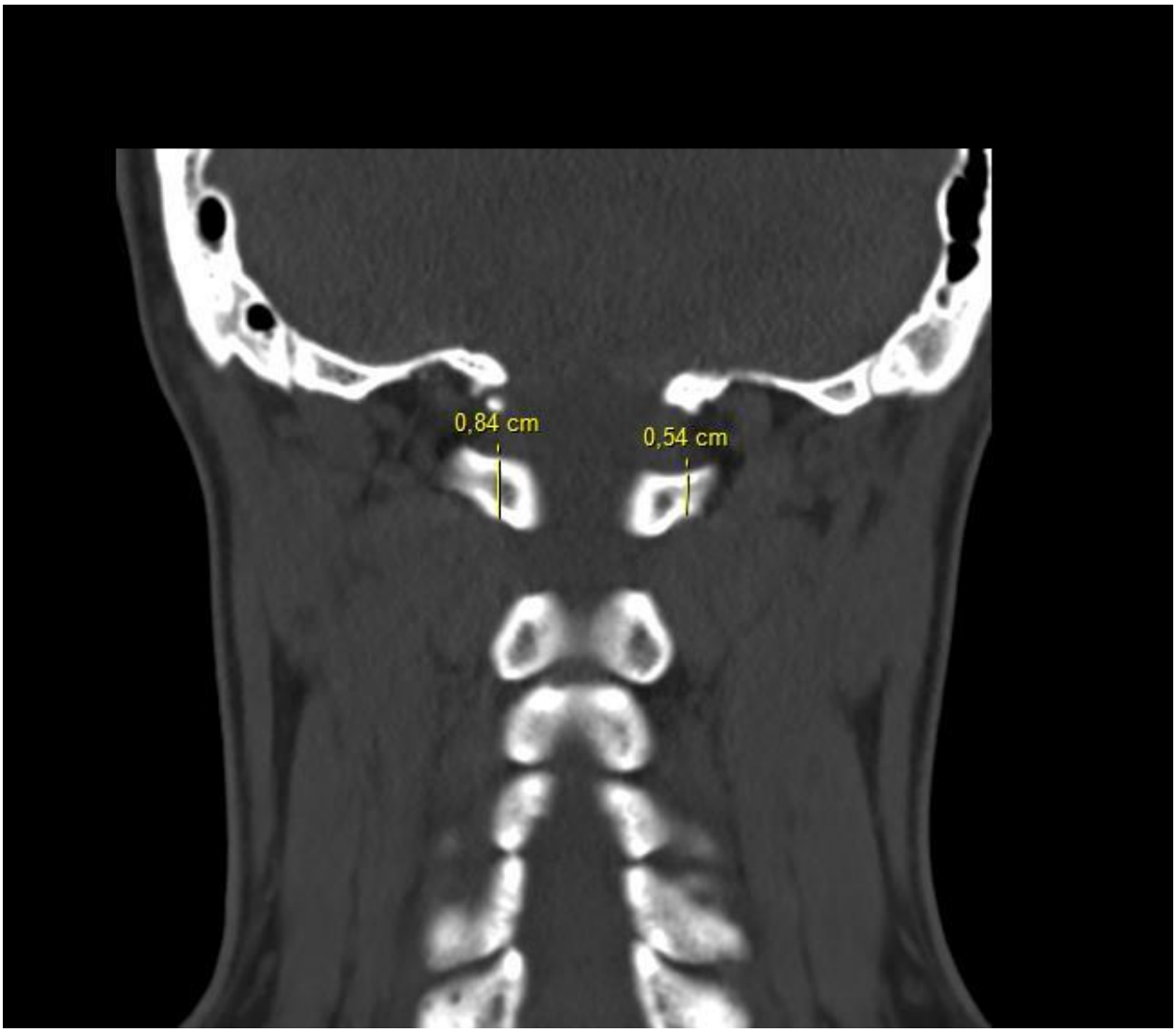


Figure 3. Axial laminar thickness was defined as the mediolateral diameter of the lamina at its middle point.

Figure 4. Coronal laminar thickness was defined superior-inferior diameter of the lamina.



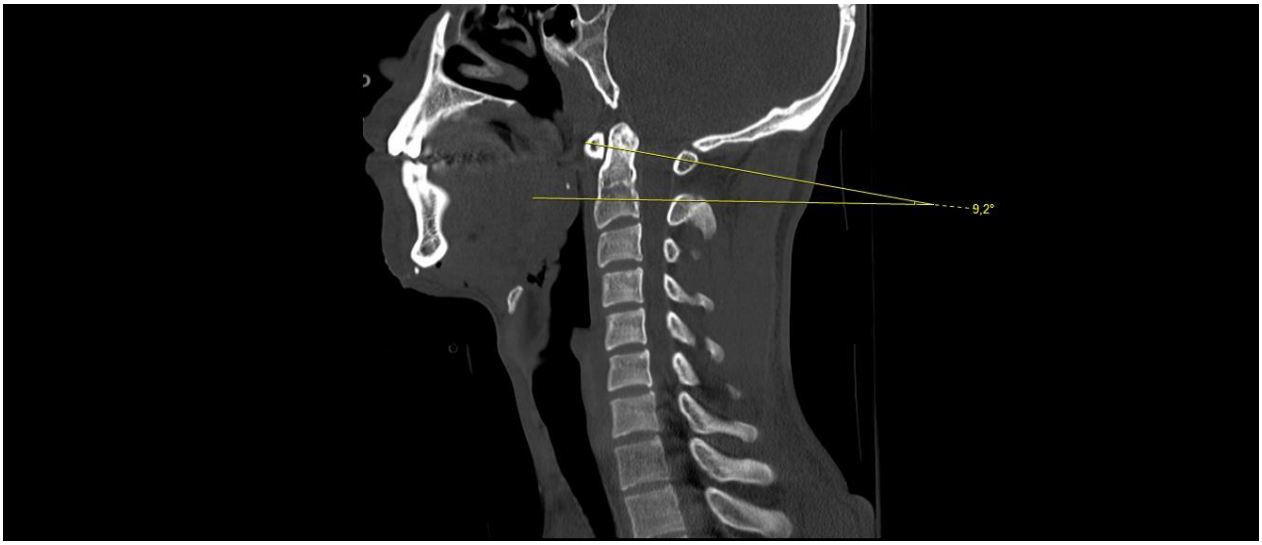


Figure 5. Cranio-caudal angle (CCA) was measured as the angle of the lamina with the line that parallel to the earth.

Results

A total of 40 patients and their lamina were analyzed. Measurements belong to men and women are presented in Table 1. There were 20 men and 20 women in the study. The mean LLL was 350,8500±26,12374 mm in male and 315,0000±26,24380 mm in female. There was a statistical difference between male and female by LLL(p=.000). The average laminar length in right side (RLL) in men was significantly longer (342,0500±22,48854 mm) than that in women (314,5000±24,15411 mm) (p=0.001). RLA was statistically wider in women (48,2650±3,49666 mm) than men (40,5650±15,85308 mm). LLA was measured 45,1800±10,04084 mm in men and 48,3200±2,94379 mm in women. These results revealed that no statistical significance was detected in the LLA along with men and women (p > 0.050). LLAT was measured 60,9000±9,74355 mm in men and 61,1000±10,70121 mm in women. There were no statistical differences between the groups. RLAT was measured 63,6000±10,89858 mm in men and 61,2500±11,77363 mm in women. There were no statistical differences between the groups. RLCT were statistically longer in men (57,1000±11,87611 mm) than women (44,9500±12,06768 mm). LLCT was measured 55,5500±10,56048 mm in men and 46,0000±12,13520 mm in women. There were no statistical differences between the groups. The mean CCA was 10,0250±5,07459 mm in male and 10,3700±5,19535 mm in female. There were no statistical differences between the groups.There were 25 patients (62.5%) under 50 years and 15 patients (37.5%) upper 50 years. Measurements belong to age are presented in Table 2. There was no statistical difference in any measurements by age.

Discussion

Craniovertebral junction (CVJ) is osteoligamentous membranous complex composed between brain and spinal cord. Layers of muscles, ligaments, and membranes promote bony complex of occiput, atlas, and axis from all around which helps in providing motion as well as stability to this field. CVJ can be affected by congenital, developmental, degenerative, traumatic, and neoplastic pathologies[9, 10]. It is very hard to diagnose instability of the CVJ and treat due to their complex anatomical composition and biomechanical characteristics. CVJ injuries may lead to sudden fatality or delayed impairment of neurological function[11]. Especially C1–2 fixation techniques have been improved to overcome those troubles. C1 lateral mass - C2 pedicle screw fixation using has been raised since it was presented in 1994 by Goel and Laheri[5] and modified in 2001 by Harms and Melcher[6]. The patients that have anomalies on the bone or VA anatomy are under injury risk while performing these techniques. The authors reported C1 posterior arch screw to reduce the risk of VA injury[12]. The coexistence of a small pedicle and high riding vertebral artery is a risk factor for vertebral artery injury[13]. Interlaminar screw placement is safe in patient that have unilateral vertebral artery occlusion, patients have large paravertebral venous

plexus, patients with fracture near lateral mass or pedicle screw placement site[14, 15]. Studying near the fracture site can cause much bleeding. While interlaminar screw placement, surgeons don't study around large venous plexus. This situation provides to avoid much bleeding. Zarro et al compared the pullout strength of C1 lateral mass screw with unicortical C1 posterior arc screw. They found that unicortical C1 posterior arc screw is stronger than the C1 lateral mass screw in the axial direction[16]. Jin et al. showed that there is no statistical difference between unilateral C1 posterior arc screw- C2 laminar screw combined with unilateral C1-2 pedicular screw and whole pedicular screw insertion as performing acute stability in cadaver study[17]. As a result, the C1 interlaminar screw placement is safe salvage technique for craniovertebral junction stabilization and provides strong fusion.

Conclusion

The appropriate laminar screw sizes in the left side in men are 350 mm length, 60 mm axial thickness and 55 mm coronal thickness. The appropriate laminar screw trajectories in the left side in men angles were 45' medio-lateral angle and 10' craniocaudal angle. The appropriate laminar screw sizes in right side in men are 342 mm length, 40 mm axial thicknes and 57 mm coronal thickness. The appropriate laminar screw trajectories in the right side in men angles were 40' mediolateral angle and 10' craniocaudal angle. The appropriate laminar screw sizes in left side in women are 315 mm lenght, 61 mm axial thickness and 46 mm coronal thickness. The appropriate laminar screw trajectories in the left side in men angles were 48' mediolateral angle and 10' craniocaudal angle. The appropriate laminar screw sizes in the right side in women are 314 mm length, 61 mm axial thickness and 44 mm coronal thickness. The appropriate laminar screw trajectories in the left side in men angles were 48' mediolateral angle and 10' craniocaudal angle.

References

1. Arslan SG, Dildes N, Kama JD. Cephalometric investigation of first cervical vertebrae morphology and hyoid position in young adults with different sagittal skeletal patterns. Sci World J. 2014;2014:159784.
2. Gallie WE. Fractures and dislocations of the cervical spine. Am J Surg. 1936;46:495-9.
3. Brooks AL, Jenkins EB. Atlanto-axial arthrodesis by the wedge compression method. J Bone Joint Surg Am. 1978;60:279-84.
4. Grob D, Magerl F. Surgical stabilization of C1 and C2 fractures. Orthopade. 1987;16:46-54.
5. Goel A, Laheri V. Plate and screw fixation for atlantoaxial subluxation. Acta Neurochir (Wien). 1994;129: 47-53.
6. Harms J, Melcher RP. Posterior C1-C2 fusion with polyaxial screw and rod fixation. Spine (Phila Pa 1976). 2001;26:2467-71.
7. Burke JP,Gerszten PC,WelchWC. Iatrogenic vertebral artery injury during anterior cervical spine surgery. Spine J. 2005;5(5):508-514.
8. Smith MD, Emery SE, Dudley A, Murray KJ, Leventhal M. Vertebral artery injury during anterior decompression of the cervical spine. A retrospective review of ten patients. J Bone Joint Surg Br. 1993;75-B(3):410-415.
9. Rhoton AL., Jr The foramen magnum. Neurosurgery. 2000;47(Suppl 1):S155–93. [PubMed]
10. Kawashima M, Tanriover N, Rhoton AL, Jr, Ulm AJ, Matsushima T. Comparison of the far lateral and extreme lateral variants of the atlanto-occipital transarticular approach to anterior extradural lesions of the craniovertebral junction. Neurosurgery. 2003;53:662–74.
11. Dickman CA, Papadopoulos SM, Sonntag VK, Spetzler RF, Rekate HL, Drabier J : Traumatic occipitoatlantal dislocations. J Spinal Disor. 1993;6 : 300-313.
12. Jin GX, Wang H, Li L, et al. C1 posterior arch crossing screw fixation for atlantoaxial joint instability. Spine (Phila Pa 1976). 2013; 38(22): E1397–E1404.
13. Yeom JS, Buchowski JM, Kim HJ, Chang BS, Lee CK, Riew KD. Risk of vertebral artery injury: comparison between C1-C2 transarticular and C2 pedicle screws. Spine J. 2013;13:775–785.
14. Matsubara T, Mizutani J, Fukuoka M, et al. Safe atlantoaxial fixation using a laminar screw (intralaminar screw) in a patient with unilateral occlusion of vertebral artery: case report. Spine (Phila Pa 1976). 2007; 32(1): E30–E33.
15. Ma XY, Yin QS, Wu ZH, et al. C1 pedicle screws versus C1 lateral mass screws: comparisons of pullout strengths and biomechanical stabilities. Spine (Phila Pa 1976). 2009; 34(4): 371–377.
16. Zarro CM, Ludwig SC, Hsieh AH, et al. Biomechanical comparison of the pullout strengths of C1 lateral mass screws and C1 posterior arch screws. Spine J. 2013; 13(12): 1892–1896.
17. Jin GX and Huan W. Unilateral C-1 posterior arch screws and C-2 laminar screws combined with a 1-side C1–2 pedicle screw system as salvage fixation for atlantoaxial instability. J Neurosurg Spine. 2015; 24: 315–320.

Romanian Neurosurgery (2020) XXXIV (1): pp. 106-109
DOI: 10.33962/roneuro-2020-014
www.journals.lapub.co.uk/index.php/roneurosurgery

Intraventricular arachnoid cyst of lateral ventricle in an elderly patient

Rajneesh Misra, Sushil Kumar, Sandeep Sharma, Pankaj Bharadva

St. Stephens Hospital, Tis Hazari, Delhi, INDIA

ABSTRACT

Arachnoid cysts are usually located in relation to the arachnoid cisterns. Intra-ventricular location is rare and its embryological emergence in this site is controversial. We report a large intra-ventricular cyst in a 61-year-old female who presented with decreased vision, headache and right hemiparesis. MRI was suggestive of cystic lesion in the lateral ventricle and was excised completely through a craniotomy.

Keywords
intra-ventricular arachnoid cyst,
magnetic resonance imaging,
lateral ventricle

Corresponding author:
Rajneesh Misra

St. Stephens Hospital, Tis Hazari, Delhi, India

misra_rajneesh@yahoo.com

INTRODUCTION

Arachnoid cyst comprises about 1% of intracranial space occupying lesions and are located in relation to arachnoid cisterns, most common locations being sylvian, chiasmatic, suprasellar, quadrigeminal and cerebellopontine cisterns. Symptomatic presentation of an intraventricular arachnoid cyst is very rare especially in the seventh decade of life and hence it being reported.

CASE REPORT

A 61-year-old lady presented with decreased vision and headache of about one-year duration and weakness in the right side of the body for the preceding two and half months.

On examination: She was conscious; visual acuity was limited to perception of light in the right eye and finger counting at a distance of 2ft in left eye. There was increased tone in all limbs and right hemiparesis with a power grade III. A retinal hole had been detected in the right eye in another hospital.

Investigations: routine blood investigations, urinalysis, kidney function, liver function, ECG were reported to be normal. MRI of the brain revealed a large lesion, hypo-intense on T1WI and hyper-intense on T2WI suggestive of cystic mass in the lateral ventricle (Fig 1).

Scan to access the online version

Craniotomy revealed large ventricle containing well defined thin walled cyst which could be easily separated from the ependyma. It was excised in-toto after coagulation of its attachment to the vessels. Histopathology of the cyst wall confirmed it to be arachnoid cyst.

Postoperatively her vision improved to finger counting at 6ft and regained full power in the right sided limbs. Her vision continued to improve over the period of observation to 6/16 and 6/24 which could be improved to 6/9 with glasses.

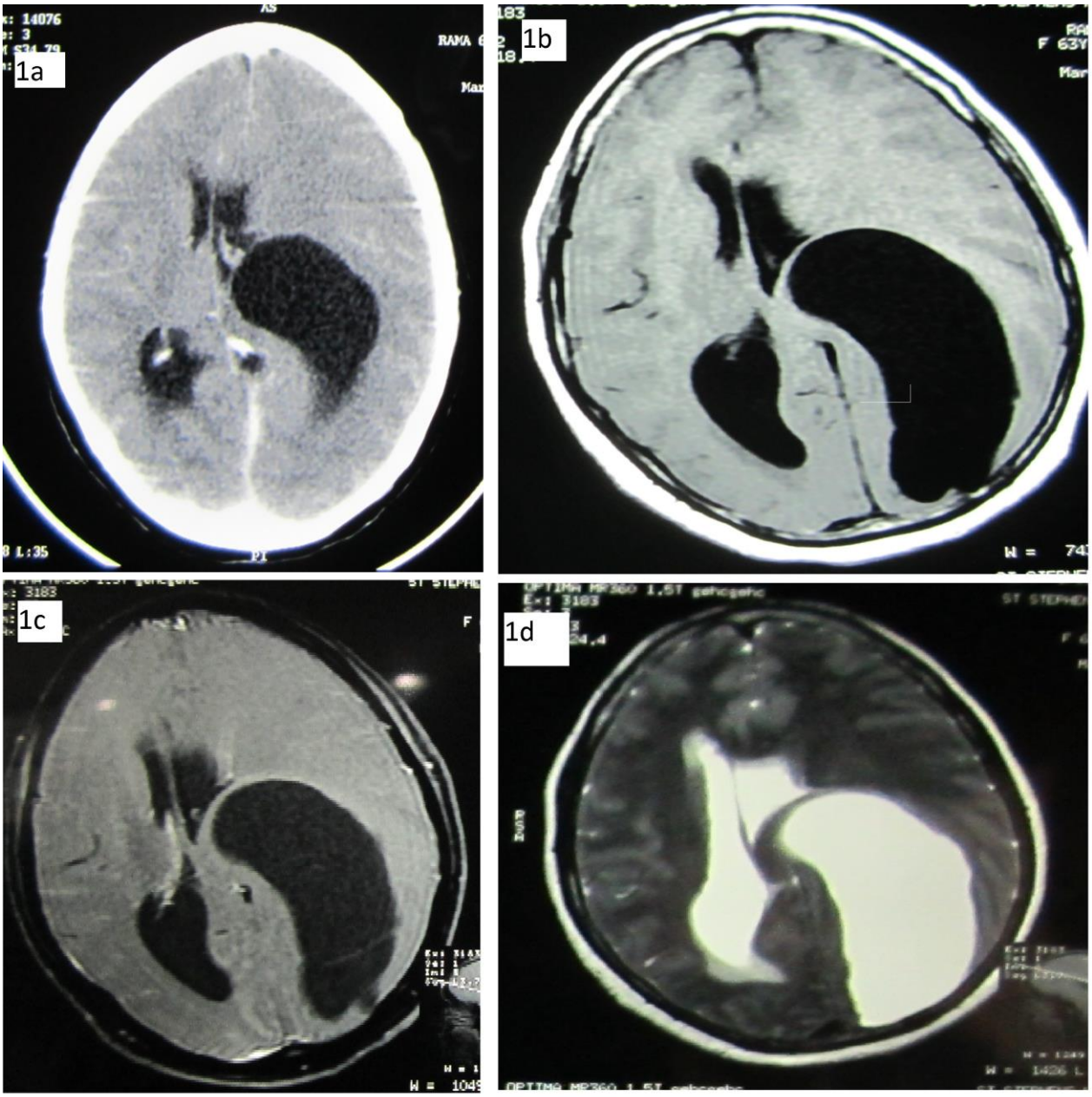


Figure 1. Showing imaging characteristics of the intraventricular arachnoid cyst. On contrast CT (1a) , it shows no enhancement of the cyst wall. The cyst is hypointense on T1 (1b), hyperintense on T2 (1d) with no wall enhancement on T1 contrast (1c).

Discussion

Most of the arachnoid cysts are asymptomatic and are discovered incidentally on imaging for other indications [(1)]. When they are symptomatic, the presenting features most often include headache, vomiting and seizures. Park et al collected 22 cases of arachnoid cyst of lateral ventricle from the literature and added one of their own [(2)]. Of these, 15

were adults and 8 were children. Headache was the most common presenting feature in this series. Wong et al in 1993 reported a case which presented with positional psychosis due to intermittent blockage of temporal horn that occurred when the patient had been recumbent for 2 -3 hours [3]. Focal neurological deficits like hemiparesis as seen in our patient are rare at presentation. These lesions are seen usually in children or in the 4th decade of life. Recently, relatively large series of pediatric patients treated endoscopically have been published [4]. However; our patient seems to have been the oldest in the 7th decade of life. What process had altered the status quo between the cyst and the patient is debatable as she had been apparently living with it for almost all of her life.

There are primarily two theories about the embryological origins of the arachnoid cysts. The first is the 'arachnoid splitting theory' [5]. As the name suggests, it proposes that these cyst s arise as a result of congenital splitting of the arachnoid layer wherein, the CSF accumulates gradually. The other theory is in specific context of middle cranial fossa arachnoid cysts and suggests that these arise as a result of temporal lobe agenesis as the primary event. However, it is named as 'subarachnoid cyst theory' [6].

There are various mechanism proposed for expansion of the arachnoid cysts. These include, but are not limited to, fluid secretion by cyst wall, presence of osmotic gradient and ball valve mechanism [7].

As mentioned previously, arachnoid cysts are purported to arise by a congenital splitting of the arachnoid membrane and subsequent accumulation of CSF in this 'potential space'. However, normally, there is no arachnoid tissue in the ventricle. Thus, the origin of an arachnoid cyst in intraventricular location is controversial. According to Yeates and Enzmann, intraventricular arachnoid cysts arise from the vascular mesenchyme by invagination into adjacent brain, picking up an outer covering of adjacent glial tissue [8]. Nakase et al. postulate that cyst arises from the arachnoid layer brought with vascular mesenchyme which it invaginates via the choroidal fissure [9]. Usually, the cysts are located in the occipital horn and/or trigonal region and cause dilatation of the temporal horn and/ or occipital horn. In our patient, the cyst pathogenesis seems to mirror the mechanism proposed by Nakase et al. as it had an attachment to the choroidal fissure.

Imaging characteristics are those of any CSF containing cavity viz. hypo-intense on T1, hyper-intense on T2 and without any contrast enhancement. Symptomatic cysts require surgical intervention. The options available include endoscopic fenestration, cyst de-roofing, partial or complete removal and cysto-peritoneal shunt. Our patient was treated by complete removal of cyst wall through a craniotomy. Endoscopic fenestration was the safer and less invasive option. In Park's series, there were various combinations and permutations of procedures applied for management of adult as well as pediatric intraventricular arachnoid cysts-open removal, open fenestration or partial cyst removal and endoscopic fenestration and shunt [2] . Kurokawa et al recommended cyst resection rather than shunt because he found that the size of the ventricle was not reduced after the shunt procedure and the shunt malfunctioned due to wrapping of the shunt by the collapse cyst [10]. Park et al had case of large arachnoid cyst of the lateral ventricle extending from the suprasellar cistern. The cyst wall could be easily separated from the ventricular ependymal after coagulation and transection of the attachments.

Cysto-peritoneal shunt or complete removal of the cyst wall are necessary to prevent recurrence, whereas cyst opening alone is insufficient. Our patient was treated by complete removal of the cyst wall through a craniotomy to avoid recurrence.

Conclusions

The embryological origin of intraventricular arachnoid cysts is controversial and the jury is still out. Our intraoperative observation during the cyst excision had shown the cyst to have an attachment to choroid fissure of the left lateral ventricle. This is in agreement with Nakase et al.'s propounded theory that intraventricular arachnoid cyst arises from the arachnoid layer brought with vascular mesenchyme which it invaginates via the choroidal fissure.

References

1. Al-Holou WN, Yew AY, Boomsaad ZE, Garton HJ, Muraszko KM, Maher CO. Prevalence and natural history of arachnoid cysts in children. J Neurosurg Pediatrics 2010; 5:578–585.

2. Park SW, Yoon SH, Cho KH, Shin YS. A large arachnoid cyst of the lateral ventricle extending from the supracerebellar cistern-case report. Surg Neurol. 2006 Jun;65(6):611-4.
3. Cheuk-Wah Wong, Sheung-Fat Ko, Yau-Yau Wai, Arachnoid Cyst of the Lateral Ventricle Manifesting Positional Psychosis, Neurosurgery, Volume 32, Issue 5, May 1993, Pages 841–843.
4. Copley, P., Kirkman, M.A., Thompson, D. James G, Aquilina K. Endoscopic surgery for intraventricular arachnoid cysts in children: clinical presentation, radiological features, management, and outcomes over a 12-year period. Childs Nerv Syst (2018) 34: 257.
5. Starkman SP, Brown TC, Linell EA: Cerebral arachnoid cyst. J Neuropathol Exp Neurol 17:484-500, 1958.
6. Robinson RG: Congenital cysts of the brain: arachnoid malformations. Prog Neurol Surg 4:133-174, 1971.
7. Basaldella L, Orvieto E, Dei Tos AP, Della Barbera M, Valente M, Longatti P. Causes of arachnoid cyst development and expansion. Neurosurg Focus. 2007;22(2): E4.
8. Yates A, Enzman D. An intraventricular arachnoid cyst. J Comp Assist Tomogr 1979; 3:697-700.
9. Nakase H, Hisanaga M, Hashimoto S, Imanishi M, Utsumi S. Intraventricular arachnoid cyst. Report of two cases. J Neurosurg 1988; 68:482-6.
10. Kurokawa Y1, Sohma T, Tsuchita H, Kitami K, Suzuki S, Ishikawa A. A case of intraventricular arachnoid cyst. How should it be treated? Childs Nerv Syst. 1990 Sep;6(6):365-7.

Romanian Neurosurgery (2020) XXXIV (1): pp. 110-115
DOI: 10.33962/roneuro-2020-015
www.journals.lapub.co.uk/index.php/roneurosurgery

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

Sandeep Bhardwaj, Anil Kansal, Rohit Bansal, Rakesh Sharma, Akangsha Sharma

BLK Superspeciality Hospital, New Delhi, INDIA

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 body1,2. 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.

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

Corresponding author:
Sandeep Bhardwaj

BLK Superspeciality Hospital,
New Delhi, India

sandeep74699@gmail.com

Scan to access the online version

INTRODUCTION

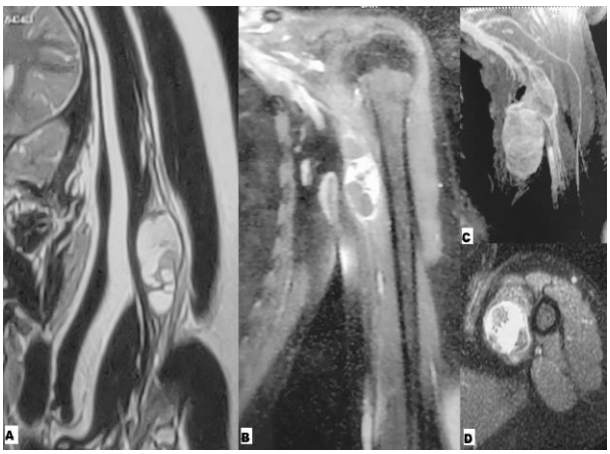
Synovial sarcomas may arise from different and unusual sites with distinctive morphological genetic features[3]. They are mostly seen in extremities in young adolescents with male preponderence[4]. It has been found in unusual locations in heart, lung, small intestine, soft palate and peripheral nerves. 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 synovium[4]. Sarcomas have their origin from primitive mesenchymal undifferentiated cells[6]. 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].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

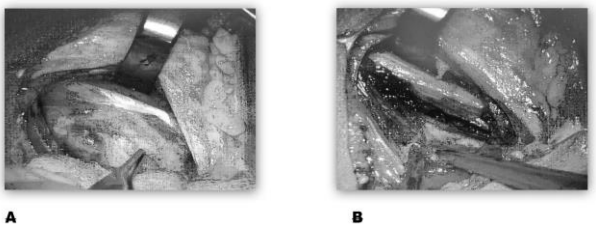
A 15-year female presented with swelling in left arm for last 6 months with swelling of size of 5*4 cm in left arm which has gradually increased in size. She also complained of pain in left arm radiating along medial aspect of forearm up to left hand. The pain was sharp, and more during night. She also 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.

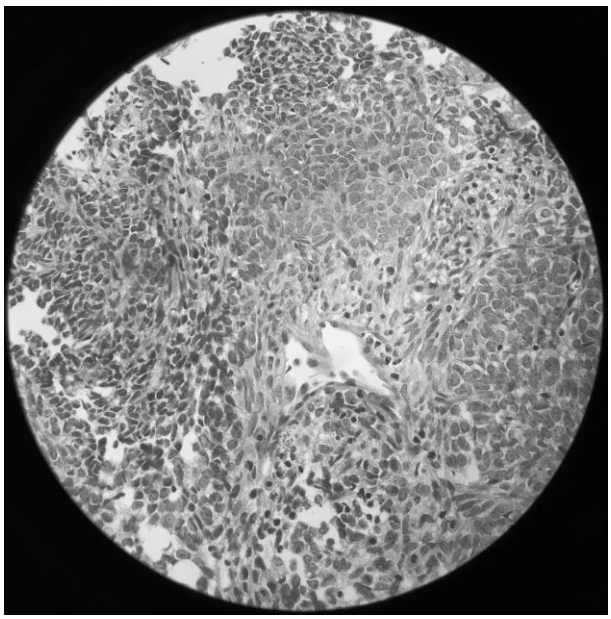
The patient was evaluated with Magnetic resonance imaging (Figure 1) which revealed a well encapsulated oval lesion in left upper arm medially burrowing in left biceps and coracobrachialis muscle. The lesion minimally indented the left brachial artery. The lesion was in continuity with median nerve which 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 tumor.

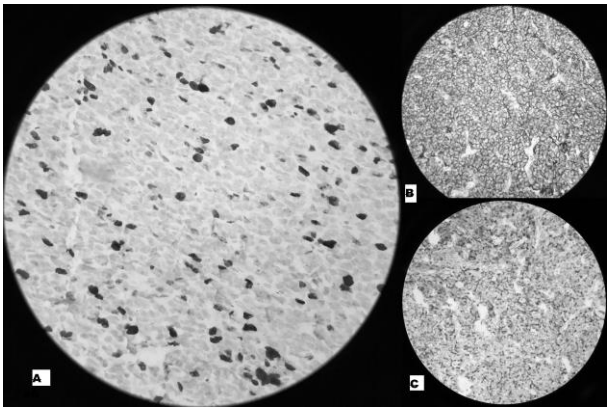


Electrodiagnostic studies and nerve conduction studies of median nerve were within normal limits. Ultrasound color Doppler peripheral venous single upper limb was suggestive of hetero-echoic lesion with internal cystic components measuring 43*23 mm causing compression of underlying axillary artery medially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

On histopathological analysis, tumor was greyish tan made up of spindle cells with sheets of cells with vesicular nuclei and ill-defined cell margins suggestive of mild epitheloid morphology (Figure 3). Focal areas showed perivascular arrangement with cells separated by blood vessels with occasional mitosis and areas of hemorrhage. Collagen and osseous tissue was seen adjacent to tumor. 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.







Discussion

Synovial sarcomas arising from median nerve are very rare and rarest tumors 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 tumors but now can be differentiated on the basis of immunohistochemical analysis[4]. Third pattern of poorly differentiated synovial sarcomas that has worse prognosis and has also been described[6]. Prognosis of biphasic versus monophasic is still under debate[9,10,11,12].

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

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

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 sarcoma[4]. 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 sarcomas[4]. 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 tumors express SYT/SSX1, while monophasic tumors express SYT/SSX1 in approximately half of the cases and SYT/SSX2 in the remainder[13]. Patients with SYT/SSX2 expressing tumors have a significantly better prognosis when compared to those with SYT/SSX1 tumors in terms of rates of metastasis and overall survival[13,14,15,16].

Factors associated with poor prognosis includes old age, tumor 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]

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%, inspite of advances in treatment[2,21,22]. Mortality in synovial sarcoma results from distant metastasis, most frequently to lung[23]. Radical surgical incision followed by radiation allowed for potential decrease in local recurrence but systemic metastasis remained high even with adjunct chemotherapy[24].

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
Rinehart 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
Tacooni 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

CONCLUSION

Synovial sarcoma involving median nerve is a rare and aggressive tumor 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-patholoical analysis with immuno-histochemistry, 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 tumor which overall changes the complete treatment and should be always be kept in differentials and managed accordingly.

References

1. 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.
2. Guadangnolo BA, Zagars GK, Ballo MT, et al. long term outcomes for synovial sarcoma treated with conservation surgery and radiotherapy . Int J Radiat Oncol Biol Phy. 2007;69:1173-80.
3. Weiss SW, Goldblum JR: Enzinger's and Weiss's soft tissue tumors. 5th ed., Philadelphia, Mosby Elsevier, 2008, 1161-1182.
4. Fischer C. Synovial sarcoma .Ann Diagn Pathol. 1998;2(2)401-21.
5. Zeren H, Moran CA, Suster S, Fishback NF, Koss MN: Primary pulmonary sarcomas with features of monophasic synovial sarcomas. A clinicopathological, immunohistochemical and ultrasturctural study of 25 cases. Hum Pathol; 26:474-480.
6. Enzinger FM, Weiss SW. Soft tissue tumors. St. Louis: Mosby;1993.synovial sarcoma;pp.757-86.
7. 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.
8. 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.
9. Chu PG Benhattar J, Weiss LM, et al. Intraneural synovial sarcoma: two cases. Mod Pathol. 2004;17:258-63.
10. 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.
11. 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.
12. 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.
13. 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.
14. 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.
15. 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.
16. 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.
17. 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.
18. 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.
19. Bergh P, Meis-Kindblom JM, Gherlinzoni F, et al. Synovial sarcoma: identification of low and high risk groups. Cancer. 1999;85:2596–607.
20. 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.
21. Spillane AJ, A'Hern R, Judson IR, et al. Synovial sarcoma: a clinicopathologic, staging, and prognostic assessment. J Clin Oncol. 2000;18:3794–803.
22. 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.
23. Enzinger FM, Weiss SW. Soft tissue tumors. St. Louis: Mosby; 1993. Synovial sarcoma; pp. 757–86.
24. 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.
25. Cugola L, Pisa R. Synovial sarcoma: with radial nerve involvement. J Hand Surg (Br) 1985;10:243-4.
26. Rinehart GC, Mustoe TA, Weeks PM. Management of synovial sarcoma of the median nerve at the elbow. Plast Reconstr Surg 1989;528-32.
27. 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. Skeltal 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-Ordonez 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.

Romanian Neurosurgery (2020) XXXIV (1): pp. 116-122
DOI: 10.33962/roneuro-2020-016
www.journals.lapub.co.uk/index.php/roneurosurgery

Microbiota Gut-Brain Axis and Neurodegenerative Disease. A systematic review on Alzheimer's disease, Amyotrophic lateral sclerosis and Parkinson Disease

Poluyi Edward Oluwatobi[1], Imaguezegie Grace Ese[2], Poluyi Abigail Oluwatumininu[3], Morgan Eghosa[4]

[1] Department of Life Science, Clinical Neuroscience, University of Roehampton, London, UNITED KINGDOM
[2] Lagos University Teaching Hospital, Idi-araba, Surulere, Lagos, NIGERIA
[3] College of Medicine, University of Lagos, NIGERIA
[4] Ambrose Alli University, Ekpoma, Edo, NIGERIA

Keywords
gut dysbiosis, central nervous system, gastrointestinal tract, enteric nervous system, neuroimmune system, neuroendocrine system, autonomic nervous system (ANS)

Corresponding author:
Poluyi Edward

Department of Life Science, Clinical Neuroscience,
University of Roehampton, London, United Kingdom

eddiepolz@yahoo.com

ABSTRACT

This review highlights the microbiota gut-brain axis and neurodegenerative diseases excluding studies on animal models. Gut microbiota is capable of modulating some brain activities via the microbiota gut-brain axis. A bidirectional communication exists between the gastrointestinal (GI) tract and the central nervous system (CNS) in the microbiota gut-brain axis. Gut dysbiosis has been linked to neurodegenerative diseases as a result of the imbalance in the composition of its microbiota, which has a damaging effect on the host's health. The association between the role and mechanism of CNS disease and gut microbial is yet to be fully explored. Although some studies have shown a positive relationship between a rich diverse microbial community and the brain of the host, and a negative relationship between microbial dysbiosis, intestinal infection and human brain health, our knowledge, however, is limited due to the inability to identify the major players in this heterogeneous microbial community.

Scan to access the online version

INTRODUCTION

Clinicians and biomedical researchers have shown great interest in the role of gut microbe function and the central nervous system (CNS), mainly in the modulation of cholecystokinin. However, this has been extended to a generalized description; the microbiota gut –brain axis especially in its link to neurodegenerative disease.[1] The human body and its microbial community such as the skin, vaginal mucosa, oral mucosa and most importantly the gut, co-exist in a symbiotic relation-

ship. This relationship plays a vital role in health and neurodegenerative diseases.[2] Microbiome is inherited maternally during vaginal delivery through the vaginal fluid enriched with microbiota. This becomes a major player of immune defense and eventually becomes modified to suit the individual's unique composition.[3,4]

Microbiota concentration is highest in the gut of the human body.[5] This can be a source of energy for cells as they provide essential micro nutrients such as thiamine, vitamins A, B, D and K. They also provide nutrients in form of short chain fatty acids (SCFAs) like acetate, propionate, and butyrate; as the ultimate energy source for colonocytes. When SCFAs like butyrate, acetate, and propionate are stimulated, it could result in an increased production of immunoglobulin (IgG).[6] Gut microbiota also serves as a barricade between humans and their environment especially in the protection of environmental hazards[3] and when it is disease-free, can prolong the lifespan of humans.[6]

Disruptions of gut microbiota barrier can lead to many diseases.[7] It was previously considered stable and unique for each individual but has now been reported to have long lasting changes.[1] This could be secondary to pathophysiological disruption of short intraluminal regulatory loop that leads to major dysfunction of various cells and microflora, thus, affecting the enteric nervous system(ENS) within the submucosal and muscularis layers of the colon.[8]

Gut dysbiosis has been linked to neurodegenerative diseases as a result of the imbalance in its composition which alters and causes damaging effect on the host's health.[9]

The aim of this review is to highlight the microbiota gut-brain axis and functional linkages to neurodegenerative diseases such as Alzheimer's disease (AD), Amyotrophic lateral sclerosis (ALS) and Parkinson Disease (PD), excluding animal models.

Discussion

Communication between gut microbiota and the development of the central nervous system (CNS)

Microbiota which consists of bacteria, viruses, fungi and other microorganisms can alter adult hippocampus neurogenesis (AHN), thus affecting the pathogenesis of symptoms of diseases of CNS.[7,10] It helps in the permeability and maintenance of the blood brain barrier (BBB) and is important in the maturation of glia cells of the CNS. The absence of a complex host microbiota could result in an altered glial cells number, a decrease in permeability and could halt the development of the blood brain barrier, thus, causing an impaired immune response resulting in CNS disease.[11]

Although, invasive pathogen by microbes is not the only route to the aetiology of neurodegenerative diseases, it has a systemic impact on the microbiota community via the enteric nervous system, immune system, blood stream, intercellular signaling and the vagus nerve.[5]

The Microbiome Gut-Brain Axis

The association between the role and mechanism of CNS disease and gut microbial have yet to be fully explored.[11] A bidirectional communication exists between the gastrointestinal (GI) tract and the central nervous system (CNS) in the microbiota gut-brain axis. This is effective under physiological conditions in immune defense, digestive system modulation, perception and sensory response to visceral stimuli, secondary to its incorporation to the CNS, neuroendocrine and neuroimmune systems, autonomic nervous system (ANS) and enteric nervous system (ENS).[3] Studies have reported four key section to the gut-brain axis, these include: activation of the immune defense, neuroendocrine pathways regulation, autonomic sensorimotor connections and lastly, the interaction between the blood brain barrier and gut microbiota metabolites.[2] Microbially derived molecules such as short-chain fatty acids (SCFAs), secondary bile acids, and tryptophan metabolites mediate the communication of microbes and the CNS, although CNS modulation via neuroimmune and neuroendocrine mechanisms interacts with enterochromaffin cells, mucosal immune system and enteroendocrine cell and sometimes enters the systemic circulation and penetrates the blood-brain barrier, it still remains difficult to ascertain if they induce responses only or get to the brain directly via long-distance neural signaling with vagal and/or afferents from the spinal cord.[12] SCFA are the main microbial mediators in the gut–brain axis.[2] SCFA are released from action of microbiota in the gut which are further metabolized by intestinal absorption and finally removed through urination.[11]

The gut microbiome harbours 150 times more genes than the human genome.[2] They are

heterogeneous group of bacterial species, fungi and viruses found in the gastrointestinal tract having a symbiotic relationship with the human host. The microbiome plays an important function in metabolism, blood-brain barrier integrity, immune defense, secretion, maintaining homeostatic balance of barrier integrity and coordinating the transfers of signals through primary afferent pathway of the vagus nerve to the CNS via the gut-brain axis. The vagus nerves serves as the primary afferent pathway for various factors influencing the formation of microbiota, these include: medications, lactation, ingested meal and parturition.[11]

The use of functional imaging, for example, functional magnetic resonance imaging (fMRI) and Positron emission tomography (PET) revealed resting brain function alteration in patients with functional gastrointestinal disorders, via the possibility of gut-brain axis dysfunction.[13]

Intriguingly, the concept that microbiome influences the gut-brain axis may seem debatable but it is used routinely by clinician to treat patients. For instance, the use of oral antibiotics medication and laxatives for treatment of patients with altered mental state secondary to hepatic encephalopathy.[1]

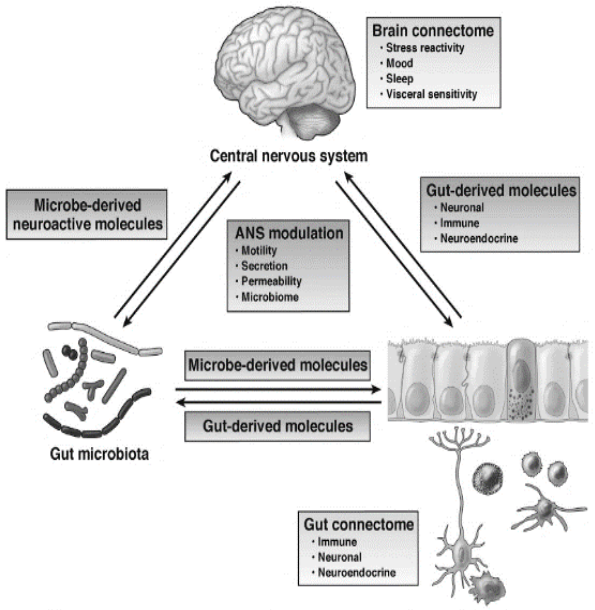


Figure 1. Bidirectional interactions of four key sections in the gut-brain axis[12].

The Gut Microbiota in Neurodegenerative disorders

The aetiology of most neurodegenerative diseases are multifactorial and speculative, there is possibility that the apotheosis of these mechanism of gut microbiome-host symbiotic relationship contribute to a large extent to the disease pathogenesis in the genetically-predisposed.[14] Microbiota research on neurodegenerative diseases is still in the infancy stage, interestingly, recent studies have linked the potential role of the gut-microbiota axis on central nervous system (CNS) related conditions and its influence on the gut brain axis. [5,7,11] Chronic neuroinflammation is a unique feature associated with the pathology of most neurodegenerative diseases (ALS, PD, AD).[6] Although, bacterial toxin has been linked to the aetiology of neurodegenerative disease, systemic inflammation and its infectious burden is suggested to be the prime factor that predisposes to neurodegeneration or immune response of the host to gut dysbiosis rather than an isolated single infectious agent. This inflammation is the initial stimulus to the neuropathological cascade seen in neurodegenerative disease that drifts gradually along the sacral autonomic nerves and dorsal motor nucleus of the vagus nerves, to the midbrain cortex through trans-synaptic spread and hence degeneration of the CNS.[5,14] Gut mucosa lymphocytes secrete pro-inflammatory mediators which causes protein misfolding in neighbouring enteric neurons. This is the main cause of pathogenesis seen in neurodegenerative disease such as AD and PD.[14]

Although, bacteria causes misfold of amyloid protein as seen in Alzheimer's disease (AD) and Parkinson disease (PD), aggravating oxidative stress, neuroinflammation and progressive neurodegeneration, neurodegeneration could also provide a favourable atmosphere for the growth of microbes via the micro-environmental change [5,6] Spirochetes, herpes simplex virus type 1 and *chlamydophila pneumoniae* infections have been associated with the brain of AD patients and fungal infection in both AD and Amyotrophic lateral sclerosis (ALS).[5] Prion-like transmission through molecular mimicry pathway suggests that microbiota is vital in prion-like transmission seen in neurodegenerative disease.[10] Minuit quantities of cyanobacteria which produce β-N-methyl amino-L-alanine (BMAA) are reported to be increased in the brain of patients with AD, PD and amyotrophic lateral sclerosis (ALS). This has been linked to the misfolding of proteins and aggregation seen in neurodegenerative disease.[3]

Constipation is a common symptom in PD, this is likely due to neurodegeneration of autonomic centers of the enteric nervous system (ENS).[9] Urinary tract infection is also a common symptom seen in neurodegenerative disease especially in AD and PD. A variety of microbes have been identified in healthy urinary tracts using novel laboratory culture methods, this could be linked to the immune system modulation**.** Interestingly, microbiome found in the skin have been suggested to be linked to microbiological and amyloid axis between the skin and the brain, but no detailed evidence has been reported.[14]

While research has focused on neural and inflammatory signaling, the future role of circulating metabolites of gut microbe-has been under-explored.[2]

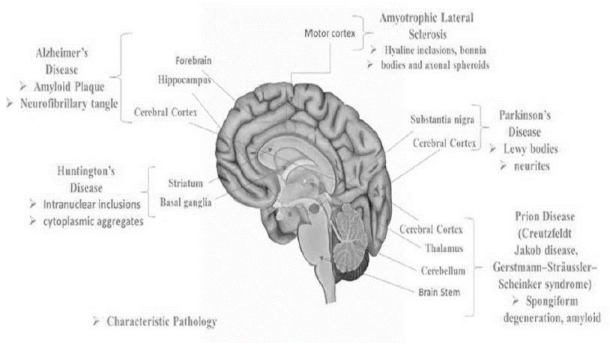


Figure 2. Interaction between gut microbiota and neurodegenerative disorders[15].

Gut Microbiota and Alzheimer's disease (AD)

AD is a progressive degenerative neurological disease with intracellular neurofibrillary tangles, extracellular β-amyloid (Aβ) and senile plaques.[7] No detailed study on the interaction of gut microbiota and alzheimer's disease has been reported[5], however a study reported the gut microbiota imbalance and the development of AD as a result of inflammation and increasing the permeability of the intestine and endothelium.[6,7] High levels of inflammatory markers, total and phosphorylated tau protein have been reported in AD patient with *Helicobacter pylori (H. Pylori)* infection.[6] Studies have also linked the association of serum antibodies and AD and a reduction in brain-derived neutrophilic factor (BDNF).[5,7] High levels of inflammatory markers (as shown in figure 3), total and phosphorylated tau protein have been reported in AD patient with *Helicobacter pylori (H. Pylori)* infection.[6]

Gut microbial diversity is reduced in patient with Alzheimer's disease (AD). Patients with AD show an increase in intestinal bacteria such as *Bacteroides, Bacteroidetes* resulting in an exaggerated translocation of Lipopolysaccharide (LPS) to the CNS from the gut. This would in turn cause neuroinflammation and exacerbate AD, while a decrease in *Bifidobacterium, Firmicutes* seen in AD patients is associated with a decrease in permeability of the intestine.[11] Excessive high fat diet, for example corn oil causes an alteration in the composition of gut microbiota thereby increasing intestinal permeability. This is associated with an increased risk of AD, however diet containing n-3 polyunsaturated fatty acids have not been linked to AD. Intestinal microbial metabolites of dietary fats for example Trimethylamine N-oxide(TMAO) can serve as a biomarker for AD, which supports the association of excessive high fat diet meal seen in western diet with AD.[6]

Intriguingly, although gut microbiota such as *E. coli*, *Bacillus subtilis*, *Mycobacterium tuberculosis, Staphylococcus aureus*, *Salmonella typhimurium*, and *Salmonella enterica*, and fungi are capable of secreting amyloid (resulting in a rise of CNS amyloid levels distorting the dynamic amyloid protein causing aggregation in the brain and a high risk in AD), they are also able to reduce amyloid of patients with AD via an indirect diet-mediated mechanism. For example *Bacillus spp* and *Lactobacillus spp* produces acetylcholine, a deficient neurotransmitter in AD.[6, 7]

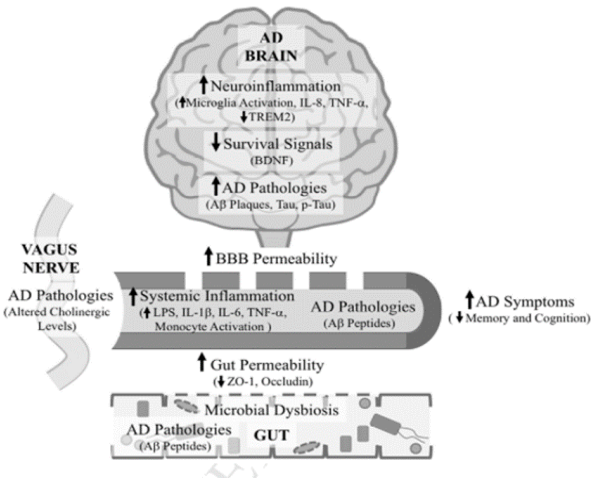


Figure 3. Interaction between gut microbiota and inflammatory markers in the pathogenesis of AD[6]. Tumour necrosis factor (TNF), triggering receptor expressed on myeloid cells-2 (TREM2,), zonula occludens (ZO), interleukins (IL), lipopolysaccharide (LPS).

Gut Microbiota and Amyotrophic Lateral sclerosis (ALS)

ALS is a progressive degenerative disorder that affects the brain and spinal cord neurons, it is a multi-system disorder that affects the gastrointestinal tract which could be as a result of the increased permeability of the intestine that would increase circulating lipopolysaccharide (LPS).[6, 10] Chronic neuroinflammation and microglial activation are features of ALS.[6] The pathogenesis of ALS involves increase circulating LPS (derived from gram-negative bacteria cell walls) and innate immune response, suggesting a concept of gut-derived neurotoxins. Tight junction proteins (occludin, VE-cadherin/CD144) and the junction adhesion molecule (JAM)) in the lumber spine are reduced leading to the disruption of the blood-spine cord barrier (BSCB) and blood brain barrier (BBB) in patients with ALS thus, facilitating permeability and increased exposed of motor neurons to the toxic substance released from the gut (Figure 4).Dysfunction of intestinal barrier can lead to the passage of toxins to the blood from the lumen of the intestine.[10] Constipation is a common symptom in ALS, however details about the effect of changes in the gut microflora on gastrointestinal motility in ALS patients seem skeptical.[6,10]

In a nutshell, research on the dynamics and relationship of gut microbiota and the stages of ALS is limited.[10]

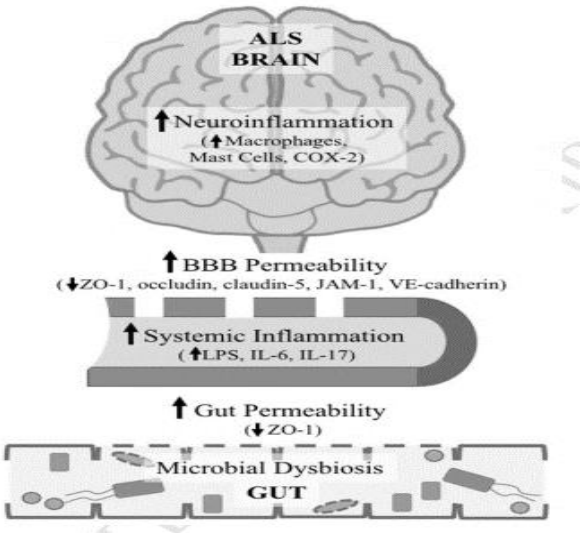


Figure 4. Interaction between gut microbiota and the pathogenesis of ALS[6]. Tumour necrosis factor (TNF), cyclooxygenase (COX), interleukin (IL), vascular endothelial (VE), zonula occludens (ZO).

Gut Microbiota and Parkinson Disease (PD)

The loss of dopamine neurons in the substantia nigra, aggregation of misfolded protein (α-synuclein), calcium overload, mitochondrial dysfunction and oxidative stress account for most of the pathogenesis seen in PD.[11]

Although, there is inadequate information on the microbiota-host relationship in PD[5], recent studies have shown the role of gut microbiota bi-directional communications between gut and brain in the pathogenesis of PD. Studies have reported over expression of protein inclusions (α-synuclein) causing misfolding and aggregation in PD,[3] which reaches the brain via the glossopharyngeal and vagus nerves.[11] Interestingly, misfold and aggregation of α-synuclein is not specific to PD, there is a descending gradient in the frequency of amount of lewy bodies seen in the GI tract. This ranges from submandibular gland with the highest frequency, to lower oesophagus, stomach, small intestine, large intestine and rectum with the lowest frequency. The vagal innervation from the dorsal motor nucleus of the vagus nerve (DMNV) and the enteric nervous system (ENS) dopaminergic neurons distribution coincide with the descending gradient of the amount of lewy body's pathology.[9]

A clinical staging system of PD (Braak stages) proposes that the genesis of neurodegenerative disease is via a dual-hit mechanism in which the neurodegenerative process starts in the olfactory bulb following inhalation and in the ENS of the gut secondary to neurotropic pathogen ingestion. This later advances forward to the temporal lobe from the olfactory bulb and backward to the intermediolateral nucleus (IML) cord or dorsal motor nucleus of the vagus (DMNV) and then to the brain stem from the gut through the sympathetic and parasympathetic nerves respectively as illustrated in Figure 5.

Gut microbiota dysbiosis have been linked to patients with PD and increased incidence of small intestinal bacteria overgrowth (SIBO) compared to control.[3,11]Over-proliferation of gut microbiota alters intestinal motility.[16] Constipation is a common and early symptom of PD, and can be as a result of prolong intestinal transit time secondary to impaired colonic motility.[9] It is 2 times more prevalent in patients with PD compared to control, and a 2 times probability of individuals with constipation having PD within 10 years.[3] Intriguingly, a high prevalence of

Helicobacter pylori (H. Pylori) infection has been reported in patient with PD.[9] Studies have linked changes in intrinsic ratios of gut microbiota fecal specimens with PD.[8] The faecal samples of patients with PD shows an increase in bacteria such as Lactobacillus and a decrease in *Prevotella, Clostridium coccoides* and *Bacteroides fragilis faecal samples*. The decrease *Prevotella* counts is associated with an increase in permeability of the gut and a decrease in synthesis of mucin. Although *Prevotella* counts is not unique to the diagnosis of PD alone, the hydrogen sulfide they secrete is reported to have a protective effect on dopaminergic neurons.[11] Studies have also reported an abundance of *Enterobacteriaceae* count isolated from faecal sample in patients with gait anomaly and postural instability, suggesting a link of microbiota to the PD. Further research on the early communication between gut microbiota and PD would give new useful insights in the intervention for diagnosing and treating patients with PD.[11]

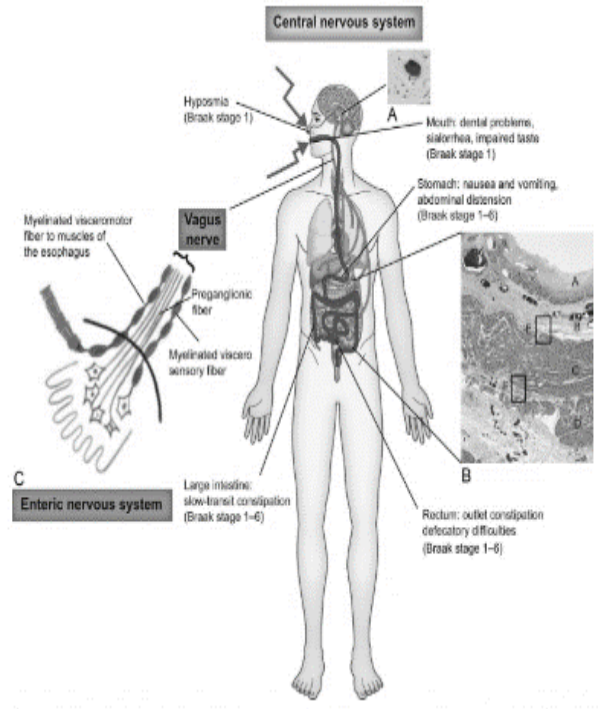


Figure 5. Genesis of neurodegenerative disease via dual-hit mechanism involving the olfactory bulb and ENS[9].

Conclusion

Gut microbiota is capable of modulating some brain activities via the microbiota gut-brain axis. Although, studies have shown a positive relationship between rich diverse microbial community and the brain of the host, and a negative relationship between microbial dysbiosis, intestinal infection and human brain health, our knowledge however, is limited due to the inability to identity the major players in this heterogeneous microbial community. These findings are particularly promising in finding out if microbiota changes precede or succeed the pathogenesis seen in neurodegenerative disease.[6] This review excluded literatures on evidence from animal and pharmacological models which some other previous studies focused. Although animal models can be controlled experimentally, this microbial community can be easily misrepresented or missed when scanning for total composition of the microbiota composition**.** Besides**,** reduced diversity in microbiota population is seen more in animal models compared to humans.

Acknowledgement

My sincere gratitude to God almighty for making it possible to complete this module. My utmost appreciation also goes to my supervisor Dr Costabile for guiding me through this review paper.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial,
or not-for-profit sectors.

Abbreviations

AD: Alzhemier's disease
AHN: Adult hippocampus neurogenesis
ALS: Amyotrophic lateral sclerosis
ANS: Autonomic nervous system
BBB: Blood brain barrier
BDNF: Brain derived neurotrophic factor
BMAA: β-N-methyl amino-L-alanine
BSCB: Blood-spinal cord barrier
CNS: Central nervous system
COX: Cyclooxygenase
ENS: Enteric nervous system
fMRI: Functional magnetic resonance imaging
GI: Gastrointestinal
IL: Interleukin
JAM:Junction adhesion molecule
LPS: Lipopolysaccharide
PD: Parkinson disease
PET: Positron emission tomography
SCFAs: Short chain fatty acids
SIBO: Small intestinal bacteria overgrowth
TMAO: Trimethylamine N-oxide
TNF: Tissue necrosis factor
TREM2: Triggering receptor expressed on myeloid cells-2
ZO: Zonula occludens

References

1. 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.
2. Guadangnolo BA, Zagars GK, Ballo MT, et al. long term outcomes for synovial sarcoma treated with conservation surgery and radiotherapy . Int J Radiat Oncol Biol Phy. 2007;69:1173-80.
3. Weiss SW, Goldblum JR: Enzinger's and Weiss's soft tissue tumors. 5th ed., Philadelphia, Mosby Elsevier, 2008, 1161-1182.
4. Fischer C. Synovial sarcoma .Ann Diagn Pathol. 1998;2(2)401-21.
5. Zeren H, Moran CA, Suster S, Fishback NF, Koss MN: Primary pulmonary sarcomas with features of monophasic synovial sarcomas. A clinicopathological, immunohistochemical and ultrasturctural study of 25 cases. Hum Pathol; 26:474-480.
6. Enzinger FM, Weiss SW. Soft tissue tumors. St. Louis: Mosby;1993.synovial sarcoma;pp.757-86.
7. 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.
8. 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.
9. Chu PG Benhattar J, Weiss LM, et al. Intraneural synovial sarcoma: two cases. Mod Pathol. 2004;17:258-63.
10. 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.
11. 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.
12. 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.
13. 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.
14. 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.
15. 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.
16. 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.
17. 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.
18. 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.
19. Bergh P, Meis-Kindblom JM, Gherlinzoni F, et al. Synovial sarcoma: identification of low and high risk groups. Cancer. 1999;85:2596–607.
20. 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.
21. Spillane AJ, A'Hern R, Judson IR, et al. Synovial sarcoma: a clinicopathologic, staging, and prognostic assessment. J Clin Oncol. 2000;18:3794–803.
22. 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.
23. Enzinger FM, Weiss SW. Soft tissue tumors. St. Louis: Mosby; 1993. Synovial sarcoma; pp. 757–86.
24. 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.
25. Cugola L, Pisa R. Synovial sarcoma: with radial nerve involvement. J Hand Surg (Br) 1985;10:243-4.
26. Rinehart GC, Mustoe TA, Weeks PM. Management of synovial sarcoma of the median nerve at the elbow. Plast Reconstr Surg 1989;528-32.
27. 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. Skeltal 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-Ordonez 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.

Romanian Neurosurgery (2020) XXXIV (1): pp. 123-126
DOI: 10.33962/roneuro-2020-017
www.journals.lapub.co.uk/index.php/roneurosurgery

Association of platelet count and platelet indices with cranial meningioma

Yasar Karatas[1], Fatih Keskin[2], Mehmet Fatih Erdi[2], Bulent Kaya[1], Densel Arac[2], Emir Kaan Izci[3], Erdal Kalkan[1]

[1] Medova Hospital, Department of Neurosurgery, Konya, TURKEY
[2] Necmettin Erbakan University, Meram Medical Faculty, Department of Neurosurgery, Konya, TURKEY
[3] Meram Training and Research Hospital, Department of Neurosurgery, TURKEY

ABSTRACT

Introduction and Objectives: In this study, we aimed to investigate whether platelet count (PLT) and platelet indices included mean platelet volume (MPV), platecrit (PCT), platelet distribution width (PDW) values can be used as diagnostic markers in cranial meningiomas.

Materials and Methods: The study included results of 29 patient and 47 healthy contributors. Based on pathologies, the patients were divided into two groups. The first group included meningioma patients and the second one included healthy individuals. Healthy contributors named control group. Platelet count and platelet indices were determined using Sysmex XN 550 haematology analyzer. The preoperative platelet count (PLT) and platelet indices included mean platelet volume (MPV), platecrit (PCT), platelet distribution width (PDW) values were recorded from the routine laboratory tests.

Results: There was no statistically significant difference in PLT between the meningioma and healthy groups (p = 0.217). There was a statistically significant difference in PCT between the meningioma group and the healthy group (p = 0.002). There was a statistically significant difference in PDW between meningioma group and healthy group (p = 0.001). In terms of MPV, there was a statistically significant difference between meningioma group and the healthy group (p = 0.001)

Conclusion: Platelet count and indices are easily available in the routine blood tests. Despite the retrospective design and small sample size, our findings suggest that altered MPV, PDW and PCT levels might serve as potential biomarkers for the diagnosis of meningiomas.

Keywords
platelet,
platelet indices,
inflammation,
meningioma

Corresponding author:
Yasar Karatas

Medova Hospital,
Department of Neurosurgery,
Konya, Turkey

yasarkrts@gmail.com

Scan to access the online version

INTRODUCTION

Platelets are reproduced from megakaryocytes[1]. We know that platelets play an important role in the coagulation process. Platelets have been studied for many years for their vital role in tumorigenesis. Many different mechanisms of the relationship between cancer cells and platelets have been demonstrated. Newly, platelets were declared to be associated with the development and progression of malignan-

cies[2,3]. The relation between cancer and inflammation was identified in the last century. As known, neutrophils, and lymphocytes are responsible for inflammation. New studies reported that platelets play a major role in inflammation[4,5]. Platelets are the first to accumulate at the site of damage and they secrete proinflammatory and growth factors such as platelet activated factor (PAF), vascular endothelial growth factor (VEGF), platelet derivate growth factor (PDGF), and thromboxane A2 at the injured area. VEGF plays a key role in regulating angiogenesis via increasing vascular permeability and facilitating the branching and formation of new blood vessels in the tumor site [6].Meningioma is a central nervous system tumor arising from the arachnoid cells present in the arachnoid mater. It is the most common benign primary tumor of the brain. Currently, there is still no consensus for biomarkers that can be used for early diagnosis and prognosis for meningiomas. In this study, we used platelet indices for the search of potential biomarkers for meningiomas.

Materials and methods

The study included results of 29 patient and 47 healthy contributors. The patients underwent surgery. Patients' laboratory and pathology results were retrospectively reviewed from the hospital archive. Based on pathologies, the patients were divided into two groups. The first group included meningioma patients and the second one included healthy indivudials. Healthy contributors named control group. Platelet count and platelet indices were determined using Sysmex XN 550 hematology analyzer. The preoperative platelet count (PLT) and platelet indices included mean platelet volume (MPV), platecrit (PCT), platelet distribution width (PDW) values were recorded from the routine laboratory tests.

Statistical analysis

The R 3.2.1 package program was used to analyze data. Descriptive statistics of the quantitative variables included in the study were determined by mean, standard deviation, median, minimum and maximum values; qualitative variables are indicated by frequency and percentage. The suitability of the continuous variables to the normal distribution was examined by the Shapiro Wilk test. Mann Whitney U test was used to compare two groups of continuous variables that do not show normal distribution. The relationships between continuous variables were interpreted with the Pearson correlation coefficient. Pearson chi-square and Yates chi-square tests were used to comparing qualitative variables between groups. In all statistical analyzes in the study, p values less than 0.05 were considered statistically significant.

Results

A total of 76 contributors were included in this study. 29 of them meningioma patients, and 47 of them were healthy contributors**.** There was no statistically significant difference in PLT between the meningioma and healthy groups (p = 0.217)(Figure 1). There was a statistically significant difference in PCT between the meningioma group and the healthy group (p = 0.002)(Figure 2). There was a statistically significant difference in PDW between meningioma group and healthy group (p = 0.001)(Figure 3). In terms of MPV, there was a statistically significant difference between meningioma group and the healthy group (p = 0.001)(Figure 4).

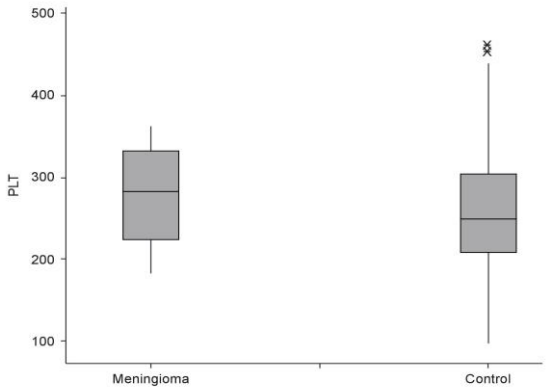


Figure 1. Comparison of PLT levels between the patients and the healthy control group.

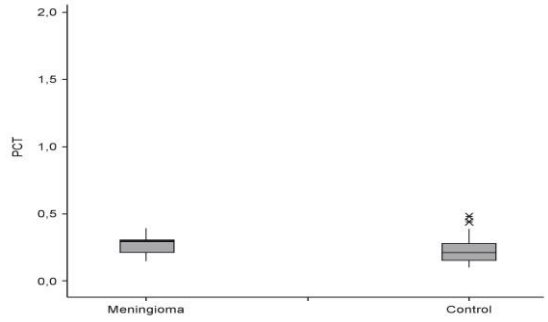


Figure 2. Comparison of PCT levels between the patients and the healthy control group.

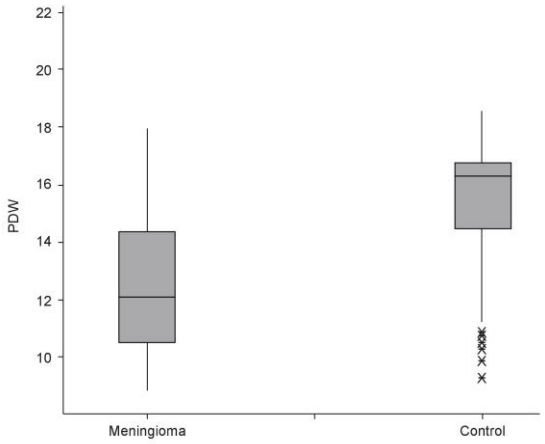


Figure 3. Comparison of PDW levels between the patients and the healthy control group.

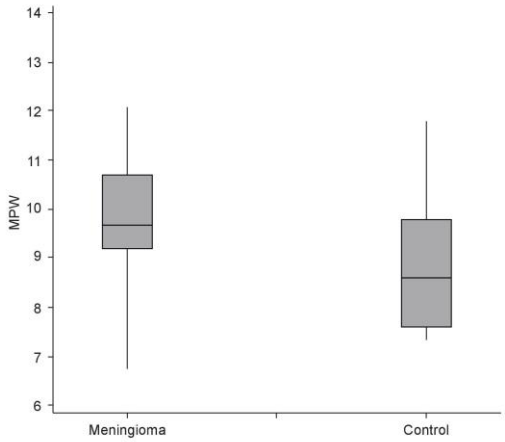


Figure 4. Comparison of MPV levels between the patients and the healthy control group.

DISCUSSION

Although it is known that genetic factors are essential in the development of cancer, the host inflammatory response is reported to be very important in the underlying mechanism of carcinogenesis [7]. Releasing inflammatory cytokines and chemokines in the damaged areas caused platelet activation. Platelets facilitate endothelial adhesion of leukocytes. Recently, researchers suggest that platelets act an important role in neoplastic disease. They put forward that platelets cause encapsulation of neoplastic cells and provide them to get out of recognition by the host immune system [8,9]. Meningiomas are the common benign neoplasms of the central nervous system. They appear approximately 20 percent frequently in all of the brain tumors and risk factors for these neoplasms are still little known [10]. High level of different cytokines which is multiple pathways of carcinogenesis was found in brain tumor specimens [11]. Platelets increase megakaryocytes and pro-inflammatory cytokines released by cancer [12].

In this study, we aimed to measure PLT and it's indices in patient with meningioma, and healthy group. PDW values were found significantly lower in meningioma group compared to the healthy control group but PCT and MPV values were found significantly higher in meningioma group compared to the healthy control group. Numerous study proposes that PLT increases in many cancer types also some others propose that no changes in PLT. Taucher et al showed that platelet count is significantly higher in patients with breast cancer compared with the control group [13]. Inagaki et al found that platelet count is significantly higher in patients with non-small cell lung cancer [14.] Ma, Ozaksit and Okuturlar et al established that there were no statistical changes in PLT while compared the groups similarly to our study [15, 16, 17]. In our study, we found no statistical changes in PLT while comparing the groups. The average platelet volume in the blood mean MPV and heterogeneity in platelet volume mean PDW. MPV is generally used for determining the platelet activity and function in the platelet indices [18]. Theoretically, reduction in platelet counts causes stimulation of megakaryocytes and they are transformed into large platelets [19]. Larger platelets are more metabolically and enzymatically active than smaller platelets [20]. Cho et al. found that patients with hepatocellular carcinoma had higher MPV. In our study, we found statistical higher MPV level in meningioma patients compared to the healthy group. Inagaki N and Kumagai S et al. showed that the patients had non-small cell lung cancer with poor prognosis had low MPV levels [14, 21]. There are different results in PDW in many research. Inagaki et al. found lower PDW levels similarly our study in non-small cell lung cancer [14]. Okuturlar et al. showed no statistical difference between breast cancer patients and the healthy groups in PDW [17]. Ma et al. found that increase PDW levels in epithelial ovarian cancer [15]. Xuegong et al. have established that the level of PCT is higher in patients with epithelial ovarian cancer compared to the healthy group [22]. Ozaksit et al. showed that there is no difference between the malignant and benign ovarian masses and the control group in terms of

PCT values [16]. We found statistical lower levels of PCT in meningioma patients compared with the healthy group.

Conclusion

We suggest that the usability of PLT and indices in the early diagnosis of meningioma patients and should be researched by further studies that have larger populations diagnosed with meningioma and their grades.

References

1. George JN. Platelets. Lancet. 2000;355(9214):1531–1539.
2. Franco, A. T. Corken, A. & Ware, J. Platelets at the interface of thrombosis, inflammation, and cancer. Blood 126, 582–588, https:// doi.org/10.1182/blood-2014-08-531582 (2015).
3. Buergy, D. Wenz, F. Groden, C. & Brockmann, M. A. Tumor-platelet interaction in solid tumors. Int J Cancer 130, 2747–2760, https://doi.org/10.1002/ijc.27441 (2012).
4. Balkwill F. & Mantovani A. Inflammation and cancer: back to Virchow? Lancet. 2001;357:539-45.
5. Tozkoparan E, Deniz O, Ucar E, et al. Changes in platelet count and indices in pulmonary tuberculosis. Clin Chem Lab Med. 2007;45:1009-13.
6. Cai J, Ma H, Huang F, et al (2013). Correlation of bevacizumab induce hypertension and outcomes of metastatic colorectal cancer patients treated with bevacizumab: a systematic review and meta-analysis. World J Surg Oncol, 11, 306.
7. Coupland LA, Parish CR (2014). Platelets, selectins, and the control of tumor metastasis. Semin Oncol, 41, 422-34.
8. M. R. Barnard, L. A. Krueger, A. L. Frelinger III, M. I. Furman, and A. D. Michelson, "Whole blood analysis of leukocyte-platelet aggregates," Current Protocols in Cytometry, 2003.
9. R. Leblanc and O. Peyruchaud, "Metastasis: new functional implications of platelets and megakaryocytes," Blood, vol. 128, no. 1, pp. 24–31, 2016.
10. Sergentanis TN, Tsivgoulis G, Perlepe C, Ntanasis-Stathopoulos I, Tzanninis IG, Sergentanis IN, Psaltopoulou T. Obesity and Risk for Brain/CNS Tumors, Gliomas and Meningiomas: A Meta-Analysis. PLoS One. 2015;10:e0136974. [PMC free article] [PubMed] [Google Scholar]
11. Albulescu, R.; Codrici, E.; Popescu, I.D.; Mihai, S.; Necula, L.G.; Petrescu, D.; Teodoru, M.; Tanase, C.P. Cytokine patterns in brain tumor progression. Mediat. Inflamm. 2013, 2013, 979748. [CrossRef] [PubMed]
12. Mutlu H, Artis TA, Erden A, Akca Z. Alteration in Mean Platelet Volume and Platicrit Values in Patients With Cancer That Developed Thrombosis. Clin Appl Thromb Hemost. 2013;19:331-3.
13. Taucher S, Salat A, Gnant M, Kwasny W, Mlineritsch B, et al. (2003) Impact of pretreatment thrombocytosis on survival in primary breast cancer. Thromb Haemost 89(6): 1098-1106.
14. Inagaki N, Kibata K, Tamaki T, et al (2014). Prognostic impact of the mean platelet volume/platelet count ratio in terms of survival in advanced non-small cell lung cancer. Lung Cancer, 83, 97-101.
15. Ma X, Wang Y, Sheng H, et al (2014). Prognostic significance of thrombocytosis, platelet parameters and aggregation rates in epithelial ovarian cancer. J Obstet Gynaecol Res, 40, 178-83.
16. Ozaksit G, Tokmak A, Kalkan H, et al (2015).Value of the platelet to lymphocyte ratio in the diagnosis of ovarian neoplasms in adolescents. Asian Pac J Cancer Prev, 16, 2037-41.
17. Okuturlar Y, Gunaldi M, Tiken EE, et al (2015). Utility of peripheral blood parameters in predicting breast cancer risk. Asian Pac J Cancer Prev, 16, 2409-12.
18. Gasparyan AY, Ayvazyan L, Mikhailidis DP, Kitas GD. Mean platelet volume: a link between thrombosis and inflammation? Curr Pharm Des. 2011;17(1):47–58.
19. Alsweedan SA, Al-Shurman A, Mahmoud AS (2008). Diagnostic value of platelet indices in children with leukemia. J Pediatr Hematol Oncol, 30, 953-5.
20. Kannar V, Raja V, Suresh TN (2017) Evaluation of platelet indices in oral squamous cell carcinoma. Clin Cancer Investig J 6(1): 40-43.
21. Kumagai S, Tokuno J, Ueda Y, Marumo S, Shoji T, Nishimura T, Fukui M, Huang CL. Prognostic significance of preoperative mean platelet volume in resected non-small cell lung cancer. Mol Clin Oncol. 2015;3:197-201.
22. Xuegong Ma, Yingmei Wang, Hongna Sheng, Wenyan Tian, Zheng Qi, Fei Teng, Fengxia Xue. Prognostic significance of thrombocytosis, platelet parameters and aggregation rates in epithelial ovarian cancer. Journal of Obstetrics and Gynaecology Research 2014, 40 (1): 178-83.

Romanian Neurosurgery (2020) XXXIV (1): pp. 127-132
DOI: 10.33962/roneuro-2020-018
www.journals.lapub.co.uk/index.php/roneurosurgery

Fast growing recurrent grade I meningioma. A rare case report

Prashank Goel[1], Daljit Singh[2]

[1] Senior resident, Department of Neurosurgery, G. B Pant Institute of Post Graduate Medical Education and Research, New Delhi, INDIA
[2] Director, Professor and Head of Department, Department of Neurosurgery, G. B PantInstitute of Post Graduate Medical Education and Research, New Delhi, INDIA

Keywords
grade I meningioma,
recurrence,
fast-growing

ABSTRACT
Meningiomas are mostly regarded as benign tumours, accounting for 13% to 26% of all primary intracranial tumors1. According to 2016 World Health Organization (WHO) classification, meningiomas are classified into grade I (benign), II (atypical), and III (anaplastic).2 Meningioma are neoplasms derived from arachnoidal (meningothelial) cells. These lesions can occur in people of any age but commonly present in middle age. Women are more likely to develop a meningioma, with a female/male ratio of approximately 2:1 intracranially and 10:1 in the spine.3 Even after complete removal, meningiomas have been estimated to recur in 10 to 32% of the cases within 10 years. However, recurrences in grade 1 meningiomas are rare and occur after long duration.4 We herein report a case of grade 1 meningioma that recurs very fast, within the duration of 1 yr and the recurrent size of the tumor was approx three times the size of the primary one. To our knowledge, very few cases in the literature have been reported with such a fast-growing grade 1 meningioma.

Corresponding author:
Prashank Goel

G. B Pant Institute of Post Graduate Medical Education and Research, New Delhi, India

prashankgoel1@gmail.com

CASE REPORT
A 45 years female, case of right cavernous sinus meningioma (figure 1,2) operated 1 year back, was admitted to our department of neurosurgery with altered sensorium for last 20 days. Simpson grade III excision of the tumor was done 1 year back. The histopathology report of first surgery was transitional meningioma grade 1 with MIB index 10-12%. After the first surgery post-operative scan (figure 3) reveals gross total excision of tumor and patient went back with no deficits. Now after 1 year patient present to us in altered sensorium, right lower motor neuron paresis of facial nerve and paresis of lower cranial nerves over the right side. MRI Brain plain with contrast was done which reveals a large recurrent mass lesion extending from right

Scan to access the online version

cavernous sinus to ipsilateral cerebellopontine angle attaching to the petrous bone with gross hydrocephalus (figure 4,5). As the patient presented in altered sensorium, patient was intubated, put on ventilator and emergency ventriculoperitoneal shunt was done. Subsequently right retromastoid suboccipital craniotomy and tumor decompression was done and a part of tumor inside the cavernous sinus was left behind (figure 6). Histopatholgy report again came out to be transitional meningioma. Post operatively patient continued on ventilator, tracheostomised and after few days gradually weaned from tracheostomy. At the time of discharge, patient was conscious, oriented, following commands and walking with support.

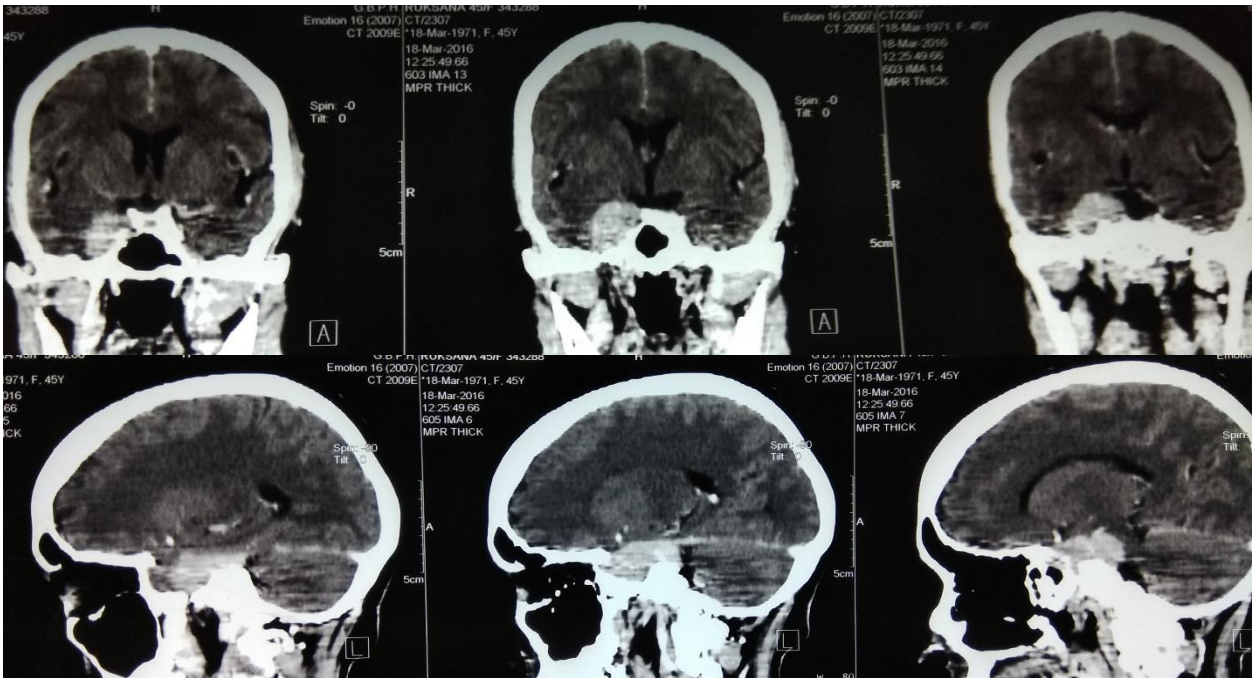
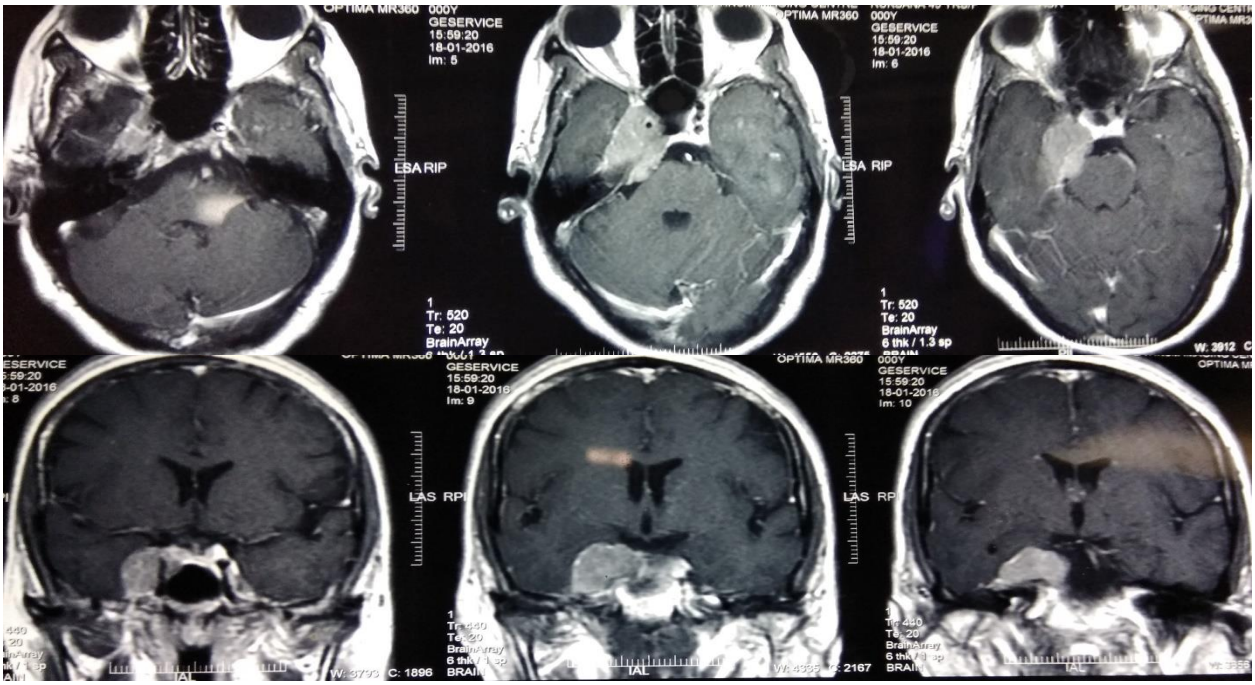


Figure 1. Contrast CT scan (coronal and saggital images) of patient done preoperatively 1 yr back, showing homogenously contrast enhancing lesion in right cavernous sinus.



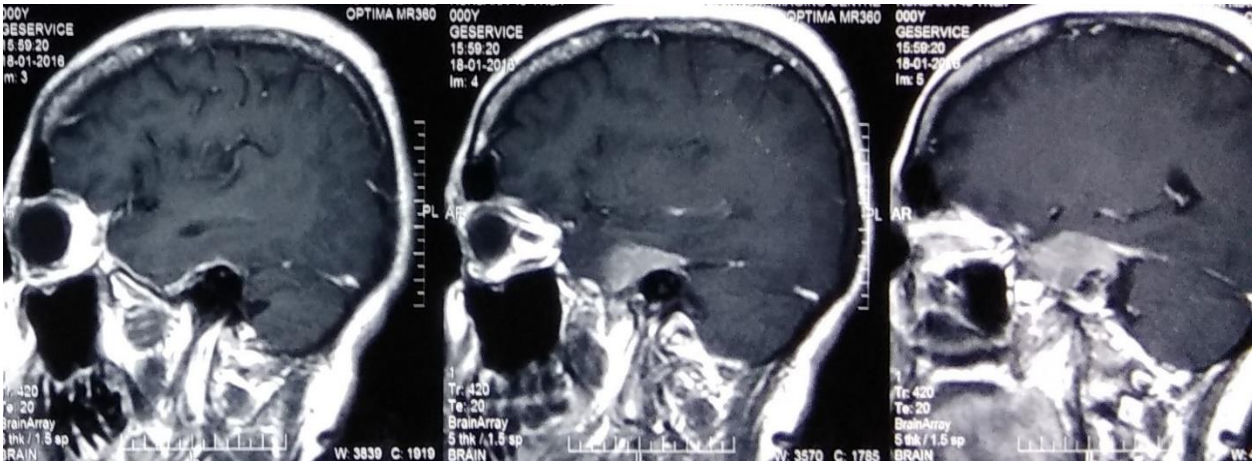


Figure 2. MRI Brain (axial, coronal and saggital images) of patient done preoperatively 1 yr back, showing homogenously contrast enhancing lesion in right cavernous sinus.

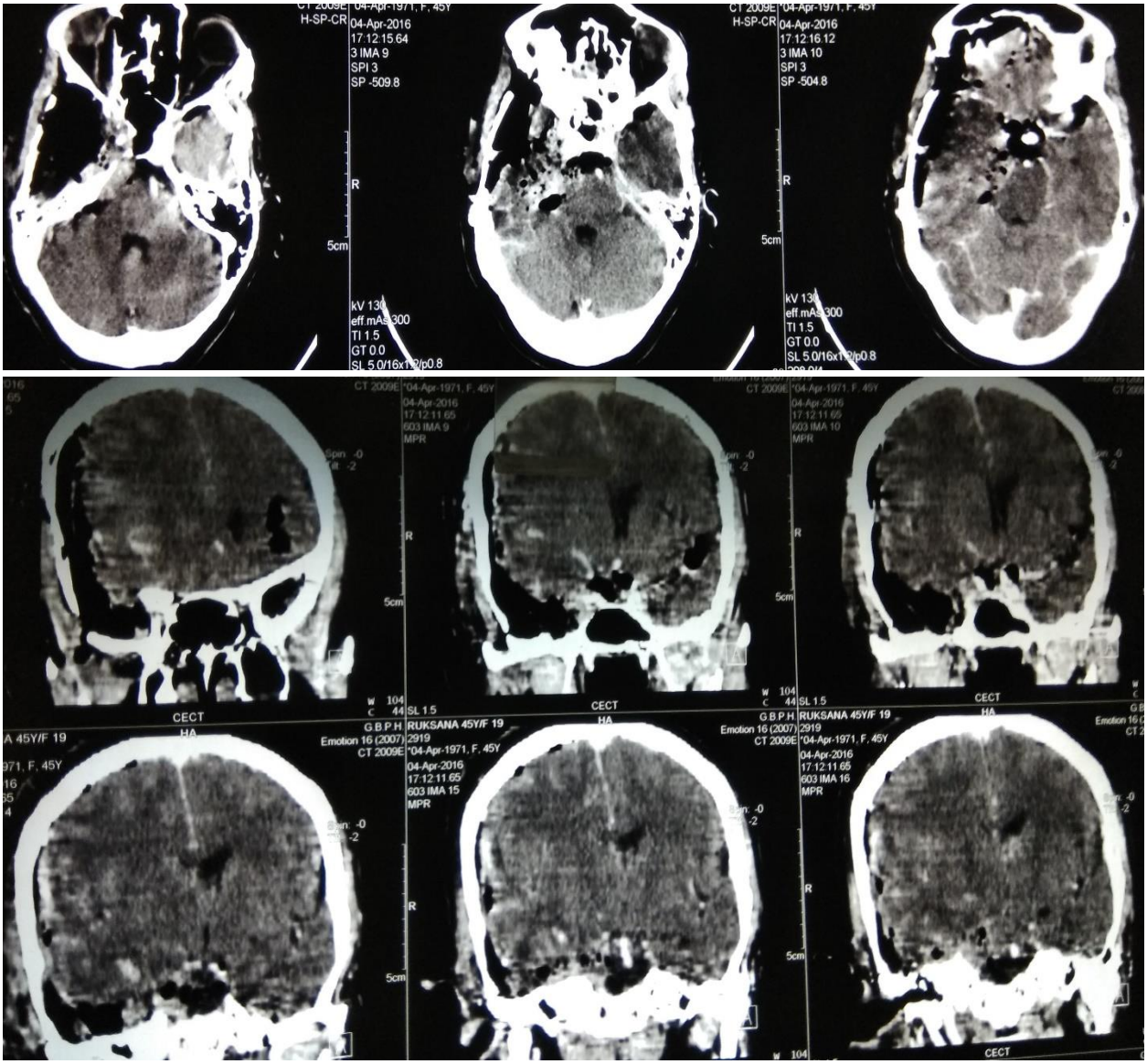


Figure 3. Contrast CT (axial and coronal) post-operative images of the patient done 1 year back, showing gross total resection of meningioma.

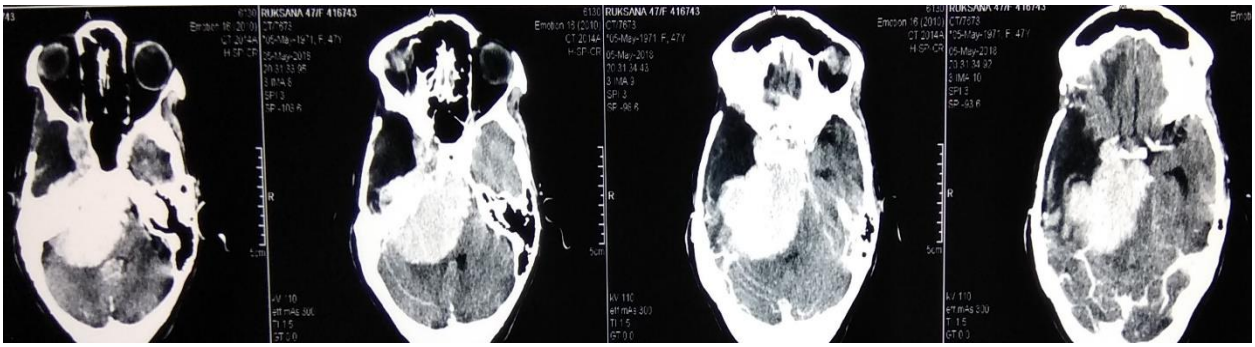


Figure 4. Contrast CT axial images showing brilliantly homogenous contrast enhancing lesion in cavernous sinus with extension to the right cerebellopontine angle.

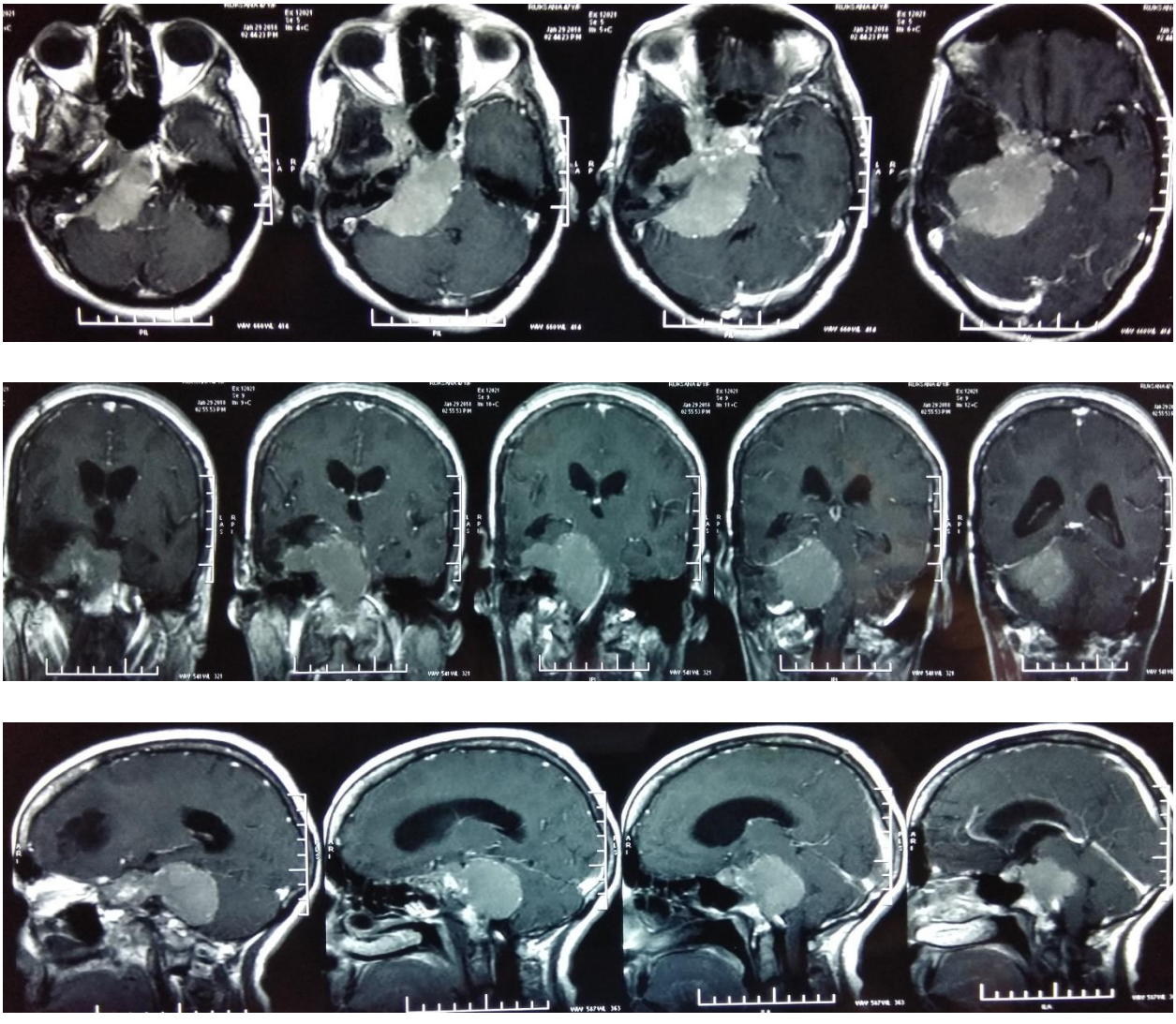


Figure 5. MRI Brain contrast images showing brilliantly homogenous contrast enhancing lesion in cavernous sinus with extension to the right cerebellopontine angle.

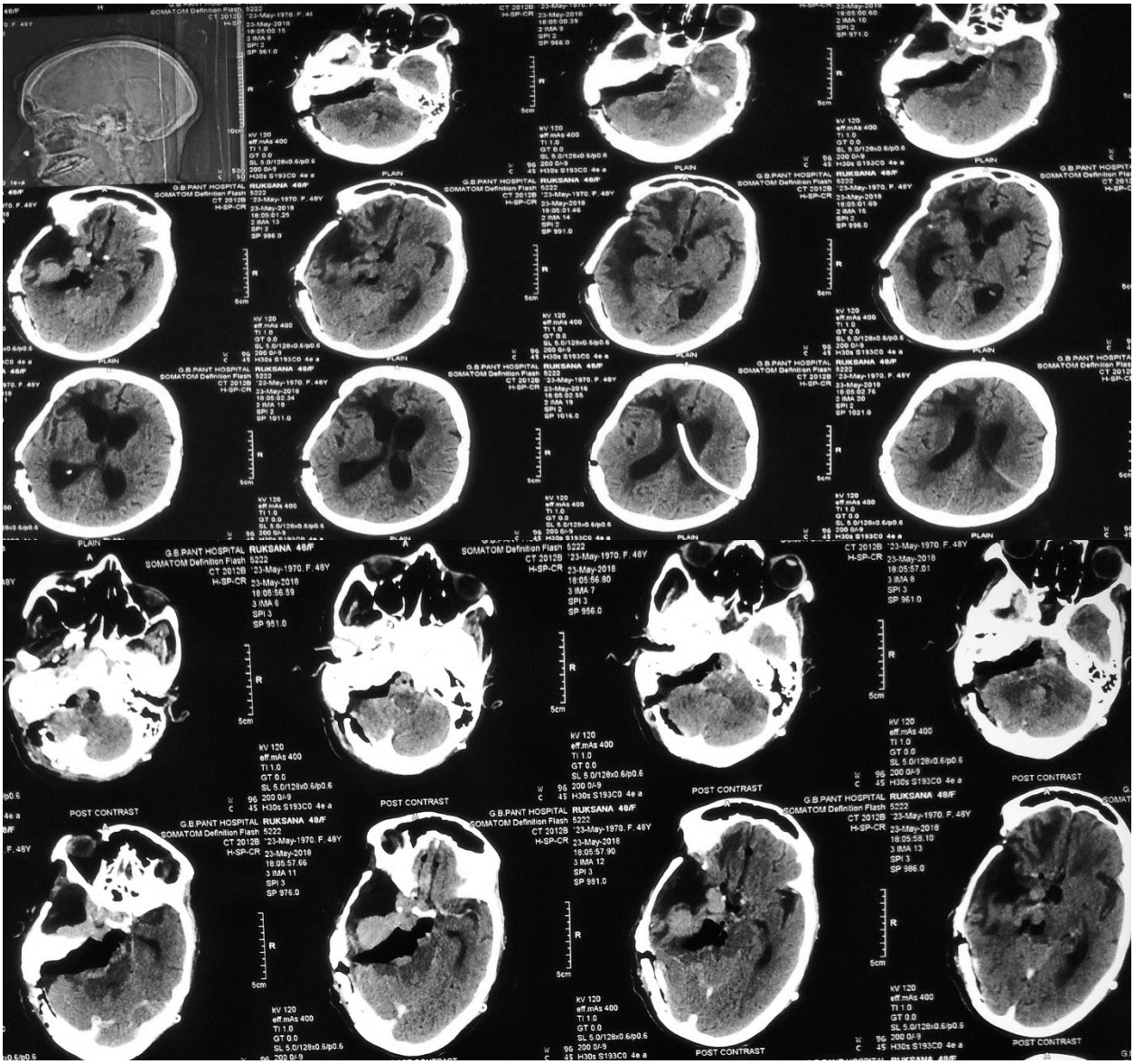


Figure 6. CT brain palin and contrast post-operative images showing total removal of meningioma in C P angle with residual tumor in cavernous sinus with vp shunt in situ.

Discussion

Recurrences in grade 1 meningioma are rare and occur after long duration. In our case patient had recurrent grade I meningioma and that too in just 1 year and now tumor has grown to such a big size that it had extended to right cerebelloopontine angle displacing the midbrain causing mass effect and hydrocephalus. To date very few cases has been reported in literature where the grade 1 meningioma recurs in such a short interval and of such a big size. Recurrences in grade 2 and grade 3 are common.

In the study by Gallagher[5], the median time to recurrence /progression was 60 months (range 6 to 134 months) and another study done by Ildan[4], the mean time to recurrence was 60.5 ± 27.9 months (range, 28 to 114 months) for benign meningiomas and 39 ± 14.5 months (range, 28 to 68 mos) for malignant meningiomas. Despite complete total resection, 7-20% of benign (Grade I), 29-40% of atypical (Grade II), 50-78% of anaplastic (Grade III) meningiomas recur.[6]

The factors involved in tumor recurrence, other than tumor grading are extent of surgery, age, gender, location and brain invasion. Meningiomas in males and children tend to be aggressive and recur more than in females[7,8]. However, Ildan et al[4], and

Adegbite et al[9] reported that age and gender has no influence on proliferative activity. Location was one of the factors implicated in recurrence. Meningiomas of the skull base and tumors close to major sinuses were reported to have a high proliferative index and were associated with recurrence[4,8,12]. Bitzer et al[10] and Ide et al[11] reported that large tumors were more prone for recurrence than smaller tumors, as large sized tumors were associated with a higher incidence of tumor infiltration and adherence to arachnoid membrane and the adjacent brain tissue. Nakasu et al[12] found that lobulated tumors recurred more often than round tumors.

Gallagher et al reported that 5-year recurrence / progression free survival (RPFS) for Simpson grade 1 was 96.8%, 2: 100%, 4: 82.4% and 5: 0%. Simpson grade and gross total/subtotal resection were significant predictors of recurrence with most of the recurrences occurring in the subtotal resection group and only a few occurring in patients who underwent total removal.

The overall mortality among recurrent meningiomas was 9.7%, and recurrence-related mortality constituted 75% of all meningioma related mortalities in the series by Guarnaschelli et al[2].

Conclusion

Recurrence in grade 1 meningioma is rare. Recurrence is common with subtotal resection of meningiomas. The predicting factors for recurrence in this case were subtotal resection, primay tumor location (cavernous sinus) with high MIB index. A regular radiological follow up should be done in these patients, so that recurrence could be identified at an early period without causing more impact on patient's general condition.

References

1. Liu Y, Song DP, Wang T. Meningiomas with different histological grade in the same patient: Case report. Medicine (Baltimore). 2017;96(50):e9086.
2. Guarnaschelli JJ, Stawicki SP. Brief communication: Recurrent brain meningiomas. OPUS 12 Scientist 2008;2(2):32-34.
3. Commins, D. L., Atkinson, R. D., & Burnett, M. E. (2007). Review of meningioma histopathology, Neurosurgical Focus FOC, 23(4), E3.
4. Ildan F, Erman T, Göçer AI, et al. Predicting the probability of meningioma recurrence in the preoperative and early postoperative period: a multivariate analysis in the midterm follow-up. Skull Base. 2007;17(3):157-71.
5. Gallagher MathewJ, Jenkinson Michael D, Brodbelt Andrew R, Mills Samantha J, Chavredakis Emmanuel. WHO Grade 1 meningioma recurrence: Are location and Simpson grade still relevant?. Clinical Neurology and Neurosurgery http://dx.doi.org/10.1016/j.clineuro.2016.01.006
6. Babu S, Uppin SG, Uppin MS, Panigrahi MK, Saradhi V, Bhattacharjee S, Sahu B P, Purohit A K, Challa S. Meningiomas: Correlation of Ki67 with histological grade. Neurol India 2011;59:204-7
7. Darlix A, Zouaoui S, Rigau V , et al. Epidemiology for primary brain tumors: a nationwide population-based study. J Neurooncol 2017;131:525–46.
8. Kasuya H, Kubo O, Tanaka M, Amano K, Kato K, Hori T. Clinical and radiological features related to the growth potential of meningioma. Neurosurg Rev 2006;29:293-7.
9. Adegbite A B, Khan M I, Paine K WE, Tan L K. The recurrence of intracranial meningiomas after surgical treatment. J Neurosurg. 1983;58:51–56.
10. Bitzer M, Wöckel L, Morgalla M, et al. Peritumoral brain edema in intracranial meningiomas: influence of tumor size, location and histology. Acta Neurochir (Wien) 1997;139:1136–1142.
11. Ide M, Jimbo M, Kubo O, Yamamoto M, Takeyama E, Imanaga H. Peritumoral brain edema and cortical damage by meningioma. Acta Neurochir Suppl (Wien) 1994;60:369–372.
12. Nakasu S, Nakasu Y , Nakajima M. Potential doubling time and tumor doubling time in meningiomas and neurinomas. Acta Neurochir (Wien) 1996;138:763–770.

Romanian Neurosurgery (2020) XXXIV (1): pp. 133-136
DOI: 10.33962/roneuro-2020-019
www.journals.lapub.co.uk/index.php/roneurosurgery

A giant occipital encephalocele. A case report

M. Al-Zekri, F. Aichaoui, A. Khelifa, A. Morsli

Department of Neurosurgery, CHU Bab El Oued, Algiers, ALGERIA

ABSTRACT

Occipital encephalocele is a rare congenital anomaly that is characterized by herniation of brain tissue through a defect in the skull. Because of their enormous size, they pose a surgical challenge. Occipital encephalocele is the commonest of all encephalocele; the management of encephalocele defects requires immediate surgical closure. The author reports a case of a five months old female baby who presented a progressively increasing swelling over the occipital region. This swelling was present since birth. Surgery was planned to reduce the size of the swelling as well as its contents. The sac was excised and reduced sufficiently enough to accommodate the healthy-looking brain tissue. This infant did well postoperatively.

Keywords
encephalocele,
dysraphism,
spina bifida

Corresponding author:
Muneer Al-Zekri

Department of Neurosurgery, CHU Bab El Oued, Algiers, Algeria

muneeralzekri30@yahoo.com

Scan to access the online version

INTRODUCTION

An encephalocele is a congenital herniation of intracranial contents throughout a cranial defect. The intracranial contents which extrude to the exterior from the defect, may incorporate cerebrospinal fluid (CSF), meningeal structures, or/and brain tissue (2, 4). Occipital encephaloceles constitute 80 to 90% of all encephaloceles and occurs between the lambda and the foramen magnum, predominantly in the midline (1, 4, 5). In giant occipital encephaloceles the size of the swelling is larger than the size of the head from which they arise, and because of their enormous size they pose a surgical challenge (1, 3, 5). Meningoencephalocele is diagnosed antenatally using sonography; it can achieve diagnostic accuracy in 80% of cases. Other imaging modalities including: CT scan, MRI (6) and the management of encephalocele defects requires immediate surgical closure (5).

CASE PRESENTATION

A five months female baby presented with progressively increasing swelling over the occipital region since birth. The baby was born at term from consanguineous marriage. On neurological examination the patient was conscious; in a general state preserved with a weight of 7 kg. She has a slight psychomotor delay with a closed anterior fontanel. Local examination showed: an occipital encephalocele of large size, well epidermised, measuring 28 cm with relatively wide neck (Fig.1).

Magnetic Resonance Imaging (MRI) of the brain showed a giant encephalocele at the occipital region with cerebral hernia and meninges across a large bone defect (Fig.2 A, B). Surgical treatment consists of the exclusion of the malformation while respecting the healthy brain tissue. The patient was put in lateral position; first, we reduced the volume of the sac by subtracting its cerebrospinal fluid (LCS) content; then, a circumferential incision was placed over the sac, and the neck was dissected out; after that, the sac was opened to expose nervous content. The resection of the sac and non-functional cerebral tissue was performed up to the limit of the healthy tissue visible next to the bone defect. in the end a watertight meningeal closure was done, with cutaneous and subcutaneous closure (Fig. 3 A,B,C,D,E,F). The post-operative course was without complications.

Figure 1. Preoperative picture showing massive occipital encephalocele.

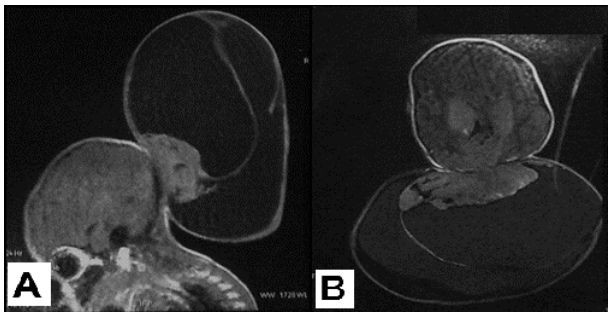


Figure 2. Preoperative T1 WI MRI; A: Sagittal, and B: transversal.

Figure 3. Preoperative views; A: LCS extraction, B: incision over the sac, C: Sac was then opened and exposure of its nervous content, D: resection of the sac, E: meningeal closure, F: cutaneous and subcutaneous closure.

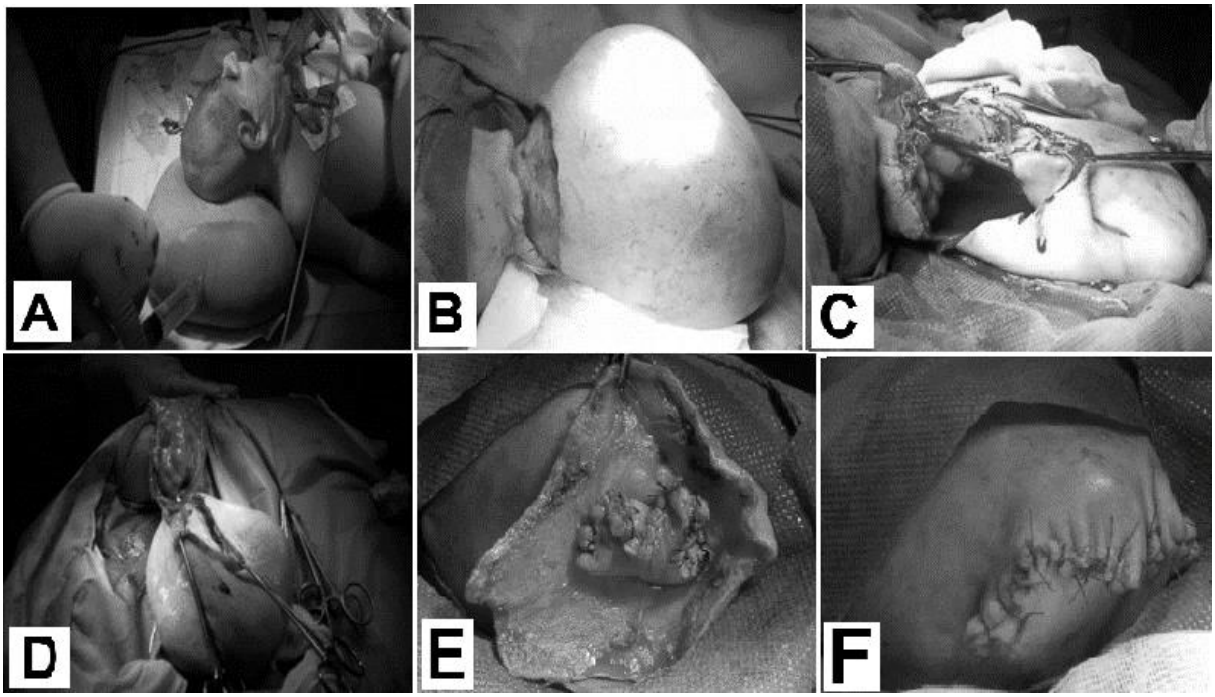


Figure 4. Postoperative picture.

Discussion

Encephaloceles are congenital anomalies of the central nervous system. They are protrusions of brain tissue. The cranial defect lead to the development of a sac with variable contents (2, 4, 7). The content of occipital encephalocele may include meninges, occipital lobes, or ventricles and rarely contain cerebellum, brainstem, or torcula. Typically, bony defect may be only confined to only occipital bone or may descend down to involve defect of the posterior lip of foramen magnum and even up to the posterior arch of the atlas and accordingly divided into supra and infra torcular types (4,5). The size of occipital encephalocele may vary from small, large to giant and could be pedunculated or sessile (1). It may be associated with hydrocephalus; or other organs and extremities malformations. Cortical dysplasia and corpus callosum agenesis are frequently present (4,5). Encephaloceles account for 10 to 20% of all craniospinal dysraphisms and 80% of them are occipital. They are more common in female than male (1, 3, 5). Encephalocele is believed to be multifactorial: genetic and environmental. Genetic background may increase the risk of developing this condition. Also, due to close relationship between encephalocele and neural tube defects, folate deficiency has been believed to play a role in the pathogenesis of encephalocele (6). These lesions are usually covered either with normal skin, dysplastic skin or a thin distorted meningeal membrane (3). The severity of mental retardation is higher and the prognosis is worse in cases neural aberrations asssociation. In a study supported by French et al. 17% of patients with encephaloceles had normal development, whereas severe mental retardation and physical delay were present in 83% of the patients (4). MRI could reveal the exact anatomical description of the encephalocele and displaced brain structures; it also reveals the configuration of the brainstem regions. Moreover, postnatal follow-up MRI confirms the prenatal findings and shows additional morphological information such as vascular anatomy. Furthermore, Magnetic Resonance Angiography is the optimal investigation to visualize the relationship of the sac to the venous sinuses; while CT scans are used to detect the extent of cranial defect (6). Patients with a large encephalocele are considered complicated cases because of CSF leakage or difficulty in repositioning the viable extruded brain tissue inside the cranium without increasing intracranial pressure (4). Further, torcula as a content poses a great challenge as its injury may lead to cerebral deep venous system thrombosis (5). Surgery is indicated for esthetic goal, to avert the risk of rupture, CSF leak, and meningitis. Before incision, aspiration of CSF may aid in the dissection of the sac, especially in large or giant encephalocele (5). The surgical intervention involves excision of the nonfunctional neural tissues hernia with dural repair. In case of a very large cerebral hernia, surgical treatment may be limited only to wide cranioplasty. Intra-operatively, care must be taken to identify the contents of the sac. Rarely, the sagittal sinus, torcula and transverse sinus are in the vicinity of the sac. Major veins should be spared to prevent any venous infarction (8). In the postoperative period, the possibility of aggravation of hydrocephalus should be kept in mind and its occurring may need shunt surgery; about 60% of posterior encephalocele may develop hydrocephalus requiring surgical management (5). The factors of poor prognosis are microcephaly with neurological disorder and the importance of the nervous elements inside the sac including the brain, the cerebellum and the brainstem.

Conclusion

Encephaloceles are rare and complicated defects. It is associated with other congenital anomalies such as hydrocephalus, Dandy–Walker malformation, and microcephaly. Its treatment like excision and repair when done in early age, greatly reduces

complications like CSF leak, reduced IQ level of the patients and other effects of associated anomalies are controlled in time. Closed monitoring post-operatively is essential as the patient may developed hydrocephalus and required ventriculo-peritoneal shunt.

References

1. H. D. Nath, A. K. Mahapatra, S. A. Borkar: A giant occipital encephalocele with spontaneous hemorrhage into the sac: A rare case Asian Journal of Neurosurgery Vol. 9, Issue 3, July-September 2014 DOI:10.4103/1793-5482.142736.
2. Lal Rehman, Ghulam Farooq, Irum Bukhari: Neurosurgical interventions for occipital encephalocele, Rehman L, Farooq G, Bukhari I. Neurosurgical interventions for occipital encephalocele. Asian J Neurosurg 2018; 13:233-7.
3. Amit Agarwal, Aruna Vijay Chandak, Anand Kakani, Shivshankar Reddy: A giant occipital encephalocele Agarwal A, Chandak AV, Kakani A, Reddy S. A giant occipital encephalocele. APSP J Case Rep 2010; 1:16
4. Ashraf El Badrya ,Azza Abdel Azeez , Ahmed AbdelKhalek: Microsurgical management results of occipital Benha Medical Journal, Vol. 35 No. 2, May-August 2018
5. Guru Dutta Satyarthee, Luis Rafael Moscote-Salazar, Nidia Escobar Hernandez, Jose Aquino-Matus, Paulo Cesar Puac-Polanco, Samer S Hoz, Willem Guillermo Calderon-Miranda: A giant occipital encephalocele in neonate with spontaneous hemorrhage into the encephalocele sac: Surgical management. J Pediatr Neurosci 2017; 12:268-70.
6. Ahmed Alwahab, Adnan Kharsa, Alaa Nugud and Shomous Nugud: Occipital meningoencephalocele case report and review of current literature Alwahab et al Chinese Neurosurgical Journal (2017) 3:40, DOI 10.1186/s41016-017-0104-5.
7. Ashfaq Ul Hassan : A giant occipital encephalocele Universal Journal of Applied Science 1(1): 8-10, 2013
8. Sim SK, Theophilus SC, Noor Azman AR :Surgical management of large occipital encephalocoele: A Case Report the International Medical Journal Malaysia Volume 13 Number 2, Dec 2014.

Romanian Neurosurgery (2020) XXXIV (1): pp. 137-140
DOI: 10.33962/roneuro-2020-020
www.journals.lapub.co.uk/index.php/roneurosurgery

Early diagnosis and management of traumatic dura tear with brachial plexus injury without spinal lesions in childhood

A. Khelifa[1], **L. Berchiche**[1], **I. Assoumane**[2], **M. Al-Zekri**[1], **B. Yakoubi**[1], **A. Morsli**[1]

[1] Neurosurgical Department of BEO University Hospital, Algiers, Algeria
[2] National Hospital of Niamey, Niger

Abstract

Although the dura is a thick membrane, it could be severed in spine surgery and less frequently by a traumatism (7,8,10); in this case, it is usually accompanied with bone or ligamentous injury (2). Rare cases are reported of a traumatic dura tear without spinal lesion and would suspect in the first place a brachial plexus injury (2,8). Dura tears are rarely seen in childhood (3). We report a case of early diagnosis of dura tear in brachial plexus injury without bone or ligamentous lesions on a child of 4 years old and we discuss the diagnostic and treatment modalities.

Keywords
dura tear,
paediatric spinal trauma,
brachial plexus injury

Corresponding author:
Khelifa Adel

Neurosurgical Department of BEO University Hospital, Algiers, Algeria

drkhelifaadel@gmail.com

Introduction

Although the dura is thick membrane it could be severed in spine surgery and less frequently by a traumatism (7,8,10), in this case it is usually accompanied with bone or ligamentous injury (2). Rare cases are reported of a traumatic dura tear without spinal lesion and would suspect in the first place a brachial plexus injury (2,8). Dura tears are rarely seen in childhood (3).

Case presentation

The patient is a 04 years old girl without past medical history; victim of traffic collision accident; clinically the patient was conscious presenting a left upper limb monoparesis. Simple X-ray, CT, and then dynamic X-rays found no more than a simple straightening of the cervical spine (figure 1). We complete by a cervical MRI witch objectified a left epidural collection expanding from C2 to C5, hypointense on T1 weighted images hyperintense T2 weighted images causing spinal cord and roots compression (figure 2). We operate the patient approaching the collection through a laminoplasty from C3 to C7. The collection appeared to be a CSF leak through a dura tear which was repaired with a watertight closure. In post-operative the patient was oriented to physical medicine where she recovered totally from her motor deficit in two months.

Scan to access the online version

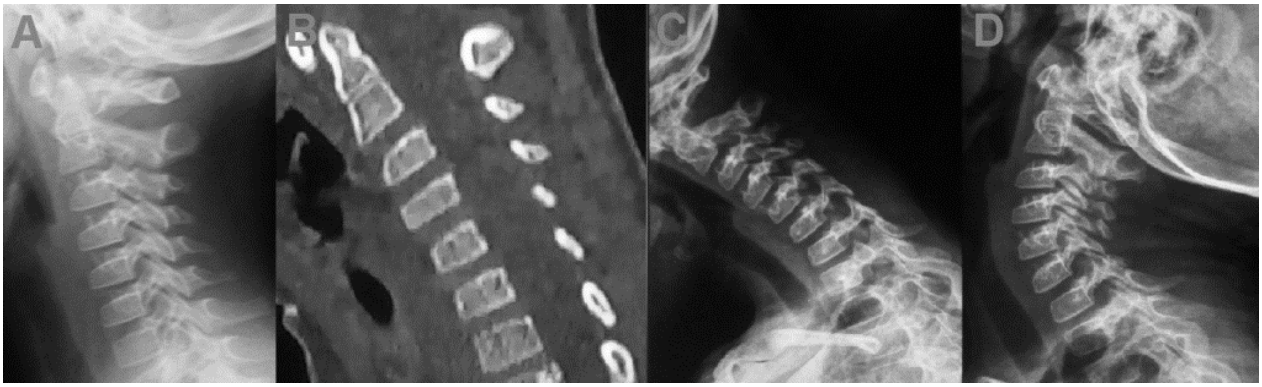


Figure 1. A: simple spine X-Ray. B: cervical spine CT (bone window). C: Dynamic X-Ray on flexion. D: Dynamic X-Ray on extension; those images showed a straightening of the cervical spine without bone lesions or instabilities.

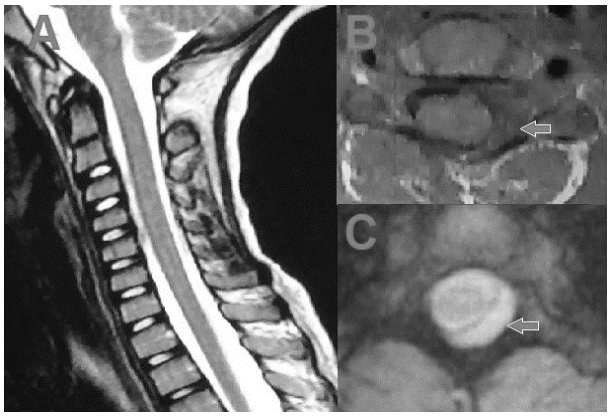


Figure 2. Preoperative MRI showing an epidural collection (the arrow). A: Sagittal T2 weighted image, B: Axial T1 weighted image, C: Axial T2 weighted image.

Discussion

The dura mater is a thick membrane of connective tissue that covers the brain and the spinal cord composed of collagen and elastin fibers, it's biomechanical proprieties was frequently studied especially in the lumbar region, but there is no available study of the difference in those proprieties between adults and children (1).The dura tear (DT) can be in the first place iatrogenic and less frequently traumatic or congenital. In fact DTs have an incidence that could reach 17% of all spine surgeries according to some series (2,4). A large study held by Hiroyuki Yoshihara and Daisuke Yoneoka in 2009 included 190,021 patients who underwent a cervical spine surgery a DT was found during surgery in 855 patients (0.45% of all patients), this sample include 880 patients under 17 years old and no one was diagnosed with DT (3). Traumatic DTs are rarer than iatrogenic (7,8,10), even though they still complicate 18% to 36% of all spine injuries (2). Traumatic DT can be divided into penetrating and blunt traumas. Burst fractures associated with laminar fracture are predictive 100% of sensitivity and 74% of specificity to a DT (6). Luszczyk et al published a study including 1615 spinal traumas, 187 patients of theme was diagnosed with traumatic DT; 36% (67 patients) of these DTs were in the cervical spine; 26.2% are burst fractures (AO Type A3), 16% flexion distraction lesions (AO Type B), and 36.4% are fracture dislocations (AO Type C); other lesions included atlanto axial dislocations, flexion compressions lesions, extension lesions, and sacral fractures; the study did not report DTs without spinal lesion (2). In absence of spine lesion traumatic DT can complicates brachial plexus injuries, in fact DT can be the result of the stretching of spinal roots between two mobile parts (8,10). DT resulting of brachial plexus injuries are rarely reported (8). In those cases the DT is located in the transition zone (11). According to Bonney's classification of the pregonglionar injuries of the brachial plexus, roots lesions distal to the transition zone (type B) could be complicated by an intra or extra spinal DT (13). The DT will allow under hemodynamic pressure the CSF leakage along the nerve course and in the epidural space (11). The DT by itself doesn't present a clinical problem (6) and it could heal spontaneously (11). But in some cases this collections will spread and gain an arachnoid covering forming what most authors call pseudomeningocele (2,3,5,7,8,10,11) causing compression of nerve structures and responsible of neurologic deficit appearing months or years after the traumatism (8,10). Other complications are seen: spinocutaneous fistula (2,3,4 ,6,7,8), meningitis (2,3,4,8), arachnoiditis (2,7,8), epidural abscess (2,7), and intracranial hypotension (2,7,8,9,10). Large part

of DTs could pass asymptomatic (8,10). Neurologic deficit could happen due to the compression or even the entrapment of the roots or nerves by a pseudomeningocele, appearing months or years after the traumatism (6,7,8,10,11); other clinical presentations could include: subcutaneous palpable masse that might be painful, collection or fistula (7,8,10), cervical pain, headache, or sciatalgia (7,8). As in our case, conventional CT is unhelpful to the diagnosis and MRI should be performed in presence of neurologic deficit non explained by the CT. MRI sensitivity could reach 97% and 100% in some studies (7,9), it shows an epidural collection hypointense in T1 weighted images and hyperintense in T2 weighted images. That collection is considered a diminution of dura sac volume rather than an epidural fluid, T.Hosoya et al proposed to call it "the floating dura sign" (9,12). T. YAGI et al classified epidural fluid collections in three types according to their relation with the dural sac: type V, for ventral location; type C, for circularly; and type D for dorsally (9). Fat suppression sequences are very helpful to differentiate the collection from the epidural fat (9,12). When a DT is highly suspected invasive imaging modalities might be performed like CT myelography or Retrograde Radionuclide Myelography (7,8,10,11,12). The management of patients with dura tears differs whether the dura tear was diagnosed in pre, per, or in post-operative. Away from the operating room, in pre- or post-operative; if a CSF leak was found, the bed rest in the Trendelenburg position could be effective; the body is laid supine, or flat on the back with a 15 to 30-degree inclination and the feet elevated (3,8,9,10). Another option that showed to be effective is to put a subarachnoide lumbar drain for 4 days collecting from 200 to 300 ml per 24 hours (2,3,4,7,8). Recently blood patch has been reported as very effective technique, by injecting 20 ml of precubital venous blood in the epidural space and in contact with the CSF it will form a clot that obstructs the DT (7,8,9,10). With an intraoperative DT and in any case a watertight closure should be tented as long as possible and a in addition gelfoam and surgicel are recommended (2,3,4,7,8,10). Hermetic fascia closure is mandatory (7).In case of brachial plexus injury the dura tear could be far lateral and thus a direct closure is difficult, in such cases Mayfield and Kurokawa proposed to open the dura medially and plunge a piece of fat or muscle with witch the lateral tear is obstructed from the intradural space (7). The fibrine glue is an easy effective complement or alternative if the suture is not possible (2,4,5,7,8). A subfascial drain is preferred (7).

Conclusion

Dura tear is an unfortunate accident that could happen in several conditions including spinal traumatism. Although it's not a problem by itself and could heal spontaneously or stay asymptomatic, some complications could appear later, though spinal MRI with adequate sequences should be a part of the diagnosis protocol even if the lesion is well defined by the spinal CT.

Declarations of interest

None.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

References

1. Chauvet D, Carpentier A, Allain JM, Polivka M, Crépin J, George B. Histological and biomechanical study of dura mater applied to the technique of dura splitting decompression in Chiari type I malformation. Neurosurg Rev. 2010 Jul;33(3):287-94; discussion 295. doi: 10.1007/s10143-010-0261-x. Epub 2010 May 4.
2. Myles J. Luszczyk et al. Traumatic dural tears: what do we know and are they a problem? The Spine Journal 14 (2014) 49–56. http://dx.doi.org/10.1016/j.spinee. 2013.03.049
3. Hiroyuki Yoshihara, and Daisuke Yoneoka. Incidental Dural Tear in Cervical Spine Surgery Analysis of a Nationwide Database. J Spinal Disord Tech _ Volume 28, Number 1, February 2015.
4. M. Lopesa, T. Faillot b. Dural tears: Regarding a series of 100 cases. Neurochirurgie 61 (2015) 329–332. http://dx.doi.org/10.1016/j.neuchi.2015.06.005
5. Soo Eon Lee, Chun Kee Chung, Tae-Ahn Jahng, Chi Heon Kim. Dural tear and resultant cerebrospinal fluid leaks after cervical spinal trauma. Eur Spine J. DOI 10.1007/s00586-014-3332-9.
6. By Frank P. Cammisa, Jr, Frank J. Eismoni, And Barth A. Green. Dural Laceration Occurring with Burst Fractures, and Associated Laminar Fractures. The Journal Of Bone And Joint Surgery.1989.
7. Stephen J. Bosacco, Michael J. Gardner, and James T. Guille. Evaluation and Treatment of Dural Tears in Lumbar Spine Surgery A Review. Clinical Orthopaedics And Related Research. Number 389, pp. 238–247.2001.

8. Mark W. Hawk, And Kee D. Kim. Review of spinal pseudomeningoceles and cerebrospinal fluid fistulas. Neurosurg. Focus / Volume 9 / July, 2000.
9. Takashi YAGI, Toru HORIKOSHI, Nobuo SENBOKUYA, Hiroaki MURAYAMA, and Hiroyuki KINOUCHI. Distribution Patterns of Spinal Epidural Fluid in Patients with Spontaneous Intracranial Hypotension Syndrome. Neurol Med Chir (Tokyo) 58, 212–218, 2018. doi: 10.2176/nmc.oa.2017-0227.
10. Samantha Knight. Pseudomeningocele. Encyclopedia of Clinical Neuropsychology. 25 March 2017. DOI: https://doi.org/10.1007/978-3-319-56782-2_9036-2.
11. Lt Col Hariqbal Singh, Maj Vljai Chandran, Col Sk Khanna, Lt Col L Satija. Post Traumatic Pseudomeningocele. Mjafi 1999; 55 : 151-152.
12. Hosoya T, Hatazawa J, Sato S, Kanoto M, Fukao A, Kayama T. Floating dural sac sign is a sensitive magnetic resonance imaging finding of spinal cerebrospinal fluid leakage. Neurol Med Chir (Tokyo). 2013;53(4):207-12.
13. Sophie.J. Camp. Peripheral nerve injury. Neurosurgery: Cases with Expert Commentary. Page 179. E.Book: books.google.

Romanian Neurosurgery (2020) XXXIV (1): pp. 141-146
DOI: 10.33962/roneuro-2020-021
www.journals.lapub.co.uk/index.php/roneurosurgery

Lumbar unilateral laminotomy for bilateral decompression in lumbar spinal stenosis

Amin Sabry, **Wael Zakarya, Amr Farid Khalil**

Neurosurgery Department, Mansoura University, EGYPT

ABSTRACT

Background: Multiple surgical approaches are existing for the management of lumbar canal stenosis.

Objective: This study was conducted to assess the outcomes of unilateral laminotomy with bilateral decompression in such cases.

Patients and methods: This prospective study was conducted at Mansoura University Hospitals, and we included a total of 12 cases with lumbar canal stenosis. All cases underwent unilateral laminotomy with bilateral canal decompression during the period between July 2017 and July 2018. Post-operative outcomes included ODI, and VAS score for both leg and back pain.

Results: The age of the cases ranged between 38 and 62 years. We included 7 males and 5 females. ODI, lower extremity, and back pain showed a significant decrease after the operation ($p < 0.05$).

Conclusion: Unilateral laminotomy with bilateral canal decompression is a safe and feasible approach to managing LSS. Excellent outcomes are expected regarding leg pain and quality of life, while slight improvement is anticipated regarding low back pain.

Keywords
lumbar stenosis,
lumbar decompression,
lumbar laminectomy,
unilateral laminotomy,
bilateral decompression,
lumbar spine,
spinal stenosis

Corresponding author:
Amr Farid Khalil

Neurosurgery Department,
Mansoura University, Egypt

dr_amr77@yahoo.com

Scan to access the online version

INTRODUCTION

Lumbar spine stenosis (LSS) is one of the commonest spinal pathologies, that present with buttock or lower limb pain associated with decreased neurovascular space in the lumbar spine region. Low back pain may be present or not [2].

Degenerative LSS usually starts in the 5th or 6th decades of life. It is characterized by hypertrophy of ligamentum flavum, intervertebral disc bulging, thickening of the facet joint, and arthropathy. These changes lead to inevitable canal narrowing [2, 15]. Cases may express intermittent neurological claudications, and the quality of life is markedly decreased [13].

Although conservative treatment can provide a temporary symptom relief, surgical decompression will be eventually needed. Nowadays, multiple surgical approaches are existing for management of such disorder. There is no definite data favoring one technique over another one [8].

Conventional laminectomy is the commonest approach performed for degenerative LSS [5]. However, the integrity of posterior spine

complex is negatively affected. Furthermore, elevation of paravertebral muscles from the spinous process leads to spinal muscle atrophy and weakness in the trunk extensors [2].

As most of LSS patients are of old age, multiple comorbidities are usually present. Thus, the invasiveness of surgery must be kept into consideration since more invasive procedures are associated with higher morbidity and mortality rates along with increased health costs [13].

Unilateral laminotomy with bilateral decompression provides the advantage of preserving neural arch and facet joint of the other side. Hence, stability is more preserved, and neural tissue are protected against posterior scarring. Besides, it offers shorter operative time, less blood loss, and less post-operative morbidity when compared to the conventional approach [3, 10, 12].

The success rate of unilateral approach in patients with bilateral canal decompression ranges between 68 and 94% [2].

This study was conducted at Mansoura University Hospitals aiming to evaluate the outcome of unilateral laminotomy with bilateral canal dilatation in LSS.

Patient and methods

Study design

This is a prospective study that was conducted during the period between July 2017 and July 2018.

Study cases

A total of 12 cases with degenerative LSS were included in the study. All cases experienced failure of medical treatment or physiotherapy for at least 3 months. Cases with neurological claudication or radiculopathy, and radiological features of LSS were included. Cases with previous spine surgery, spinal tumours, instability, or spondylolisthesis were excluded.

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. Besides, an MRI of the lumbosacral spine was ordered for all cases. In addition, plain X ray was also performed to exclude instability.

Pain was assessed via visual analogue score (VAS), while functional status was evaluated by Oswestry Disability Index (ODI).

Surgical procedure

The operation was performed when the patient was in prone position. A midline incision was created over the stenotic area as localized in pre-operative MRI. By the aid of microscope or vascular loupe, a unilateral laminotomy was carried out, whereas the inferior aspect of cranial hemilamina and the superior aspect of the caudal hemilamina were partially resected. The spinous process base was undercut after ipsilateral decompression. Then, bilateral flavectomy with contralateral neural foramen decompression was done. Following bilateral decompression, the nerve roots were visualized easily and at that point, the operation was ended.

Post-operative care and follow up

All cases were transferred to the recovery room, them to the internal ward. Mobilization was encouraged on the 1st post-operative day. Post-operative VAS and ODI were recorded at 6-and 12-month visits. Post-operative radiological evaluation of stability was not routinely performed unless the patient is still complaining of back pain or claudication is still existing.

Statistical analysis

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

Data were tested for normal distribution using the Shapiro Walk test. Qualitative data were represented as frequencies and relative percentages. Quantitative data were expressed as median (Range). Kruskal Wallis test (KW) was used to test the significance between values at more than two time points (preoperative, at 6 months and at 12 months). Wilcoxon signed rank test was used to test significance between two different time points. For all tests, p value (< 0.05) was considered significant.

Figure 1. Sagittal T2 pre-operative MRI shows severe spinal canal stenosis with multiple disc bulge and ligamentum flavum thickening.

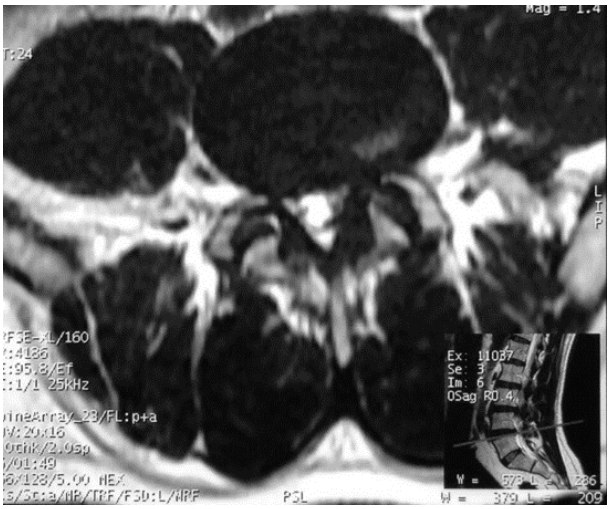


Figure 2. Axial T2 pre-operative MRI shows Severe spinal canal stenosis with a disc bulge and ligamentum flavum thickening.

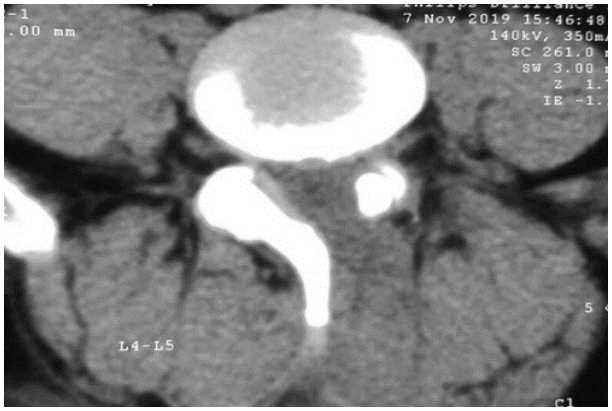


Figure 3. Axial CT scans L4 spine after the patient underwent a left L4 Unilateral laminotomy.

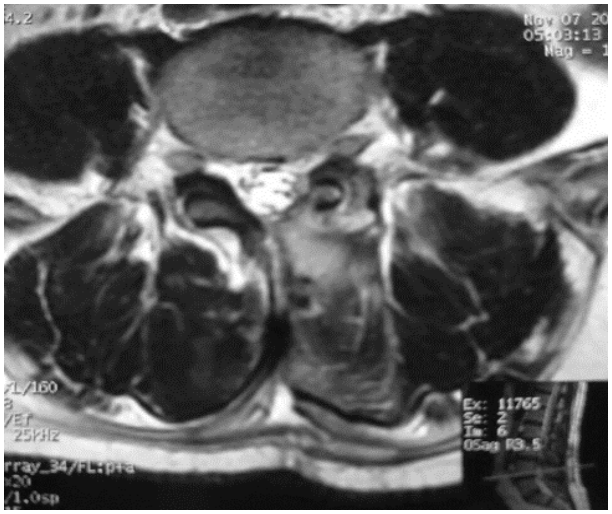


Figure 4. Axial T2 Post-operative MRI shows marked widening of the spinal canal after left Unilateral laminotomy.

Figure 5. Sagittal T2 post-operative MRI after double level (L3-L4 and L4-L5 decompression) via Unilateral laminotomy.

Figure 6. Post-operative CT 3D lumber spine after double level (L3-L4 and L4-5 Unilateral laminotomy).

Results

The age of the included cases ranged between 38 and 62 years (median = 56). We included 7 males (58.33%) and 5 females (41.66%). These data are shown at table (1).

Preoperative ODI ranged between 22 and 36 (median = 28). It decreased significantly after operation down to 7 and 6 scores at 6-month and 1-years follow up visits ($p < 0.001$).

After operation VAS score decreased down to 1 at the scheduled follow up visits. It has a median value of 9 before operation. Table (3) illustrates these data. Back pain also significantly decreased after operation from score 6 preoperatively down to 3 and 2 scores after 6 and 12 months respectively. These data are illustrated at table (4).

Table 1. Patient criteria.

Variable	Data
Age	56 (38 – 62)
Sex -Male -Female	 7 (58.33%) 5 (41.66%)

Table 2. ODI before and after surgery.

	Preoperatively	After 6 months	After 1 year
	28 (22-36)	7 (3-11)	6 (3-10)
P1		< 0.001*	< 0.001*
P2			0.145
P	< 0.001*		

P: significance between different durations
P1: significance in relation to preoperative value.
P2: significance in relation to 6 months value.
*: statistically significant ($p<0.05$)

Table 3. VAS score for limb pain before and after surgery.

	Preoperatively	After 6 months	After 1 year
	9 (7-10)	1 (1-3)	1 (0-2)
P1		0.001*	< 0.001*
P2			0.124
P	< 0.001*		

P: significance between different durations
P1: significance in relation to preoperative value.
P2: significance in relation to 6 months value.
*: statistically significant ($p<0.05$)

Table 4. VAS score for back pain before and after surgery.

	Preoperatively	*After 6 months*	*After 1 year*
	6 (4-7)	3 (2-4)	2(1-4)
P1		0.009*	0.001*
P2			0.108
P	0.005*		

P: significance between different durations
P1: significance in relation to preoperative value.
P2: significance in relation to 6 months value.
*: statistically significant ($p<0.05$)

Discussion

Multiple surgical approaches have been proposed for the surgical management of LSS. The main surgical goal is to decompress the lumbar canal. However, anatomy should be preserved to maintain the biomechanical function of the lumbar spine [2].

On using the conventional decompression technique, some authors reported paraspinal muscle atrophy on CT scan [7], and others showed electromyographic abnormalities following that approach [14]. These changes can lead to spinal instability and increase the need for spinal fusion surgery [6].

Conversely, the unilateral approach decrease the incidence of these complications after surgery [13].

This study was conducted at Mansoura University Hospitals aiming to evaluate the surgical outcomes of unilateral laminotomy with bilateral decompression for LSS.

A total of 12 cases were included with a median age of 56 (range, 38-62). We included 7 males (58.33%) and 5 females (41.66%).

Another Egyptian study handled the same perspective included 21 cases in the unilateral approach group, with a mean age of 47.2 years (range, 33 – 69). The presence of young age groups is due to the presence of cases with disco-ligamentous causes of LSS, which is more common in young age due to sedentary life and overweight (like in Egyptian population), compared to the bony type which is common in the older population [1].

In the current study, the operation successfully decreased ODI score from 28 preoperatively down to 7 and 6 scores 6 and 12 months after operation ($p < 0.001$).

Regarding ODI in other studies, it decreased from 28.7 preoperatively down to 5.55 and 6.5 at 1-month and 1-year follow-up visits respectively. However, that change was not significantly different from the conventional approach group [1].

Another study used both Japanese Orthopedic Association Score (JOA) and Neurogenic Claudication Outcome Score (NCOS) to evaluate the outcomes in unilateral laminectomy patients. There was a significant increase in both parameters form 4.35 and 26.9 up to 10.2 and 61.15 after operation respectively. This improvement was also better than the conventional approach group. That study reported that excellent and good outcomes were achieved in 14 cases in the unilateral hemi-laminectomy group (70%) [13].

On assessment of leg pain in the current study, it decreased from 9 preoperatively (range, 7 – 10), down to 1 at the scheduled follow up visits ($p < 0.001$).

In the previously mentioned Egyptian study, the pre-operative VAS score for lower extremity pain was 9.04, and it decreased significantly down to 1.38 and 1.46 at 1-month and 12-month follow-up visits ($p = 0.001$) [1].

This comes in line with the findings of Çavuşoğlu and his colleagues who stated that most VAS changes occur between operation and early follow up [4].

When it comes to back pain in our study, it was also assessed via VAS score which decreased significantly from 6 (range, 4 – 7) before operation, to 3 (range, 2 – 4) after 6 months ($p = 0.009$), and 2 (range, 1 – 4) after 1 year ($p = 0.001$).

In another study, the mean value of pre-operative VAS score for back pain was 5.42. It decreased slightly down to 2.82 and 1.96 at 1-month and 1-year follow-up visits respectively [1].

Another study has also published that VAS score for back pain has decreased from 7.6 pre-operatively down to 2.95 after operation. This decrease was also more significant when compared to the conventional approach [13].

Another recent study also stated that detailed lower back pain VAS score before surgery was 51.5 in motion, 63.0 while standing, and 37.8 while sitting; and showed LBP while standing was significantly greater than LBP while sitting ($p < 0.01$). After surgery, LBP while standing was significantly improved relative to that while sitting ($p < 0.05$), and levels of LBP in the three postures became almost the same with ODI improvement. Bilateral VAS scores showed significant improvement equally on both sides ($p < 0.01$) [16].

Regarding complications encountered in the current study, durotomy was encountered in only one case (8.33%), who was managed conservatively.

Other authors reported that unintended durotomy occurred in 4.5 % of their cases [11]. Another study reported that that complication occurred in about 5 – 15% of cases [4]. In addition, Ng and his colleagues also reported that the incidence of that complication was 14% [9]. This comes in line with our results.

In the study conducted by Abbas and his associates, early post-operative complications were encountered in 2 cases (18.2%); one had CSF leak, and the other had a wound hematoma [1].

The main disadvantage with our study is the relatively small sample size. So, more studies with larger sample size should be conducted in the near future.

Conclusion

Unilateral laminotomy with bilateral canal decompression is a safe and feasible approach in managing LSS. Excellent outcomes are expected regarding leg pain and quality of life, while slight improvement is anticipated regarding low back pain.

References

1. Abbas M, Elwany A, and Farag R: Efficacy and Safety of Using Unilateral Approach for Bilateral Neural Decompression in Lumbar Spinal Stenosis. Egyptian Spine Journal 2017, 24(1),42-9.
2. Amir S, Qadir M, Pasha I F, and Qureshi M A: To determine the efficacy of unilateral approach for Bilateral Decompression in Lumbar Spinal Stenosis. Journal of Saidu Medical College 2018, 7(2).
3. AYDIN Y: Efficacy of unilateral laminectomy for bilateral decompression in lumbar spinal stenosis. Turkish neurosurgery 2007, 17(2),100-8.
4. Çavuşoğlu H, Kaya R A, Türkmenoglu O N, Tuncer C, Colak I, and Aydın Y: Midterm outcome after unilateral approach for bilateral decompression of lumbar spinal stenosis: 5-year prospective study. European Spine Journal 2007, 16(12),2133-42.
5. Galiano K, Obwegeser A A, Gabl M V, Bauer R, and Twerdy K: Long-term outcome of laminectomy for spinal stenosis in octogenarians. Spine 2005, 30(3),332-5.
6. Lipson S J: Spinal-fusion surgery—advances and concerns. New England Journal of Medicine 2004, 350(7),643-4.
7. Mayer T G, Vanharanta H, Gatchel R J, Mooney V, Barnes D, Judge L, Smith S, and Terry A: Comparison of CT scan muscle measurements and isokinetic trunk strength in postoperative patients. Spine 1989, 14(1),33-6.
8. Moisi M, Fisahn C, Tkachenko L, Tubbs R S, Ginat D, Grunert P, Jeyamohan S, Reintjes S, Ajayi O, and Page J: Unilateral laminotomy with bilateral spinal canal decompression for lumbar stenosis: a technical note. Cureus 2016, 8(5).
9. Ng L C, Tafazal S, and Sell P: The effect of duration of symptoms on standard outcome measures in the surgical treatment of spinal stenosis. European Spine Journal 2007, 16(2),199-206.
10. Oertel M F, Ryang Y-M, Korinth M C, Gilsbach J M, and Rohde V: Long-term results of microsurgical treatment of lumbar spinal stenosis by unilateral laminotomy for bilateral decompression. Neurosurgery 2006, 59(6),1264-70.
11. Podichetty V K, Spears J, Isaacs R E, Booher J, and Biscup R S: Complications associated with minimally invasive decompression for lumbar spinal stenosis. Clinical Spine Surgery 2006, 19(3),161-6.
12. Rahman M, Summers L, Richter B, Mimran R, and Jacob R: Comparison of techniques for decompressive lumbar laminectomy: the minimally invasive versus the “classic” open approach. min-Minimally Invasive Neurosurgery 2008, 51(02),100-5.
13. Ramesh K, and Vimalan G: Comparative study between functional outcome of lumbar canal stenosis treated with surgical decompression by laminectomy and unilateral partial hemi laminectomy approaches. International Journal of Research in Orthopaedics 2018, 4(4),610.
14. See D, and Kraft G: Electromyography in paraspinal muscles following surgery for root compression. Archives of physical medicine and rehabilitation 1975, 56(2),80-3.
15. Steurer J, Nydegger A, Held U, Brunner F, Hodler J, Porchet F, Min K, Mannion A F, Michel B, and Collaboration L R: LumbSten: the lumbar spinal stenosis outcome study. BMC musculoskeletal disorders 2010, 11(1),254.
16. Takahashi H, Aoki Y, Saito J, Nakajima A, Sonobe M, Akatsu Y, Inoue M, Taniguchi S, Yamada M, and Koyama K: Unilateral laminectomy for bilateral decompression improves low back pain while standing equally on both sides in patients with lumbar canal stenosis: analysis using a detailed visual analogue scale. BMC musculoskeletal disorders 2019, 20(1),100.

Romanian Neurosurgery (2020) XXXIV (1): pp. 147-149
DOI: 10.33962/roneuro-2020-022
www.journals.lapub.co.uk/index.php/roneurosurgery

Glioblastoma of septum pellucidum

Rajneesh Misra, **Sushil Kumar, Kundan Kumar**

Dept. of Neurosurgery, St. Stephens Hospital, Tis Hazari, Delhi, INDIA

ABSTRACT

A rare case of glioblastoma multiforme (GBM) of septum pellucidum is being reported. There were symptoms of altered behaviour, memory and changes in sensorium. The MRI was suggestive of a tumour arising from the septum pellucidum. Glioblastomas arising from septum pellucidum are rare. While even a partial endoscopic excision of the more common pathology viz. colloid cyst is acceptable, only, a safe maximal excision of a highly malignant pathology like glioblastoma is an acceptable goal in order to give any benefit of surgical exploration to the patient.

Keywords
glioblastoma multiforme,
septum pellucidum,
transcortical approach

Corresponding author:
Rajneesh Misra

Dept. of Neurosurgery,
St. Stephens Hospital,
Tis Hazari, Delhi, India

misra_rajneesh@yahoo.com

INTRODUCTION

Glioblastoma multiforme (GBM) is a common primary brain tumor accounting for nearly 15-20 % of all intracranial tumors. (1) Lee and Manzano enumerated the most common tumors in the intraventricular tumors and the GBM doesn't find a mention in the first fourteen. (2) The table 1 below summarizes all such tumors in the intraventricular location.

Usually it involves frontal or temporal lobe and involvement of septum pellucidum is very rare. Hence the present case is being reported.

CASE REPORT

A 70-year-old male was admitted with inappropriate behavior and talking of 20 days duration and impaired memory with progressive drowsiness of 4 days duration. On higher mental function testing, he talked irrelevantly, and did not recognize relatives. On physical examination, the tone was increased in all the four limbs and the GCS was E2V3M5. The deep tendon reflexes were brisk.

MRI revealed a mass lesion in relation to septum pellucidum and bulging into right lateral ventricle (Fig 1). A right frontal craniotomy and a trans-cortical entry into the ventricle revealed tumor arising from septum pellucidum postero-superior to the foramen of Monroe. The tumor was soft, suckable and moderately vascular. It was infiltrative in nature. He did not improve neurologically after surgery. Post-operative radiotherapy was suggested but the family members were not willing for the same. Histo-pathology was suggestive of diagnosis of grade IV glioma (glioblastoma multiforme).

Scan to access the online version

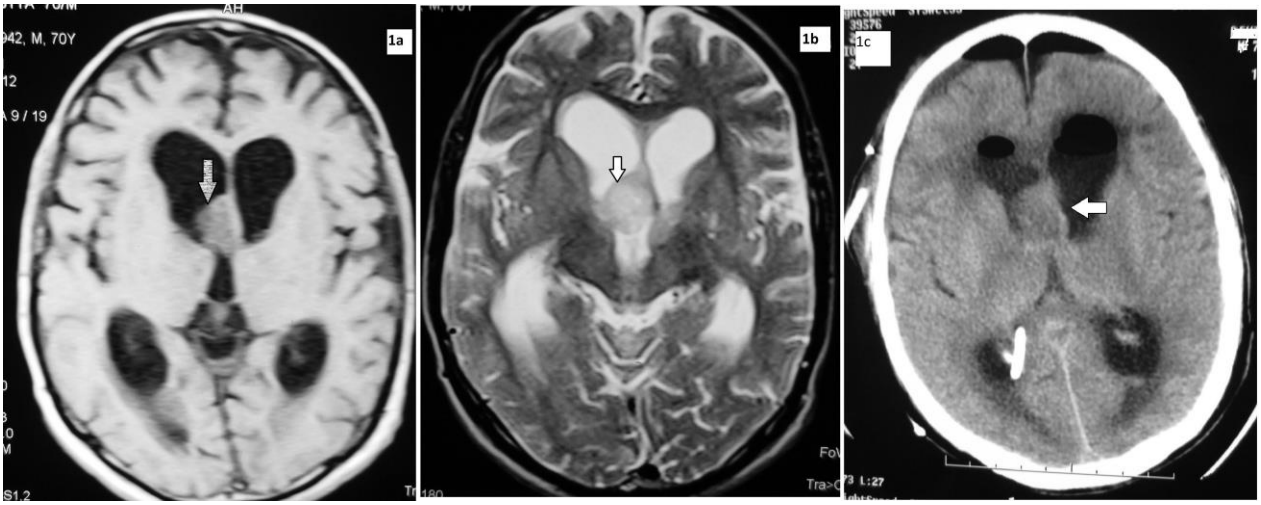


Figure 1. (1a) Axial T1 plain MRI showing iso- to hypo-intense lesion occupying the lower margin of septum pellucidum; (1b) The same lesion on T2 shows hyper-intensity; (1c) Post-operative CT brain showing pneumocephalus and residual lesion.

Discussion

GBM is one of the most common tumors encountered in neurosurgical practice. It usually involves the region of centrum semiovale with preference for frontal and temporal lobes. Involvement of the septum pellucidum by a tumor is a rare occurrence as it usually gets involved by a spread from the surrounding structures in the lateral ventricles, thalamus or fornices. Septum pellucidum is a part of limbic system and forms a relay between corpus callosum and fornix. This is classically known to have functions involving emotions, consciousness and memory. Structurally, it contains glial cells, neurons and veins connecting to choroid plexus. (4) Multiple theories have been proposed regarding the origin of GBM in this region. The most often quoted origin is that from ependyma which separated early during differentiation and later migrated towards ventricular cavity. A less commonly mentioned hypothesis states that the tumor originates elsewhere in the CNS and spreads to septum pellucidum and third ventricle through the CSF. (5) However, in our case, there was no evidence of tumor elsewhere. Galli et al. in their paper in 2004 proposed another interesting mechanism invoking the progenitor neural stem cells from the subventricular zone. (6) They observed that the subventricular progenitor stem cells are known to migrate to fimbrial fornix in injury. (7) A similar mechanism could work for spread of the tumour.

Only two of the nine cases of GBM reported in the lateral ventricles were seen to be arising from the septum pellucidum (8). Majority of the cases of GBM of septum reported have been from autopsy series. (9, 10, 11) Choi et al reported GBM of pellucidum on autopsy in a 41-year-old woman. (11)

GBM of septum pellucidum arises from the neuroglial cells of the septum and lateral ventricles. (2) There is no pathognomonic feature of the GBM of septum pellucidum GBM. Headache, visual deficits, signs of raised intracranial pressure, memory disturbances and personality changes are the features that can occur with any lesion causing slowly progressive rise in ICP. Patient's impaired memory could be explained by involvement of the limbic system pathways. (12) Therefore, diagnosing a high-grade lesion of septum pellucidum on clinical grounds is extremely unlikely.

On imaging, GBM is known exhibit aggressive features including evidence of vasogenic edema, necrosis and mass effect. None of these features were present in our case. It is possible that, due to strategic location of the lesion, it became symptomatic due to obstructive hydrocephalus before it could attain enough size to show evidence of its own mass effect or have undergo necrosis. Computed tomography of such septal lesions shows irregular hypo-density with widening of the septum. MRI usually shows enhancing ring with central hypo-intense area, although we did not find such appearance in our case. (8) Epstein and Epstein reported a widened septum pellucidum on pneumo-ventriculogram and GBM of septum pellucidum and corpus pellucidum on necropsy. (10) MRI can

diagnose the cases with indentation of septum and unilateral or bilateral mass arising from septum as demonstrated in our case. Reasonable differentials to be considered due to location of the lesion include central neurocytoma, subependymal giant cell astrocytoma and intraventricular meningioma.

Surgical de-bulking followed by radiotherapy is the standard treatment. However, attendants of the patient were not keen on radiotherapy when educated about the prognosis of the GBM.

Conclusions

The standard treatment for glioblastoma is safe maximal decompression followed by chemo-radiotherapy. However, glioblastoma arising from septum pellucidum is distinctly rare occurrence. It offers an opportunity for a more thorough decompression. However, this is possible if the clinician holds reasonable suspicion about the nature of the lesion and opts for intraoperative frozen section to guide the surgical decision making.

The authors declare no conflict of interests.
No funding was obtained for producing this work.

References

1. Lopes MBS, VandenBerg SR, Scheithauer BW: Histopathology, immunochemistry and ultrastructure of brain tumors. In Kaye AH, Laws ER Jr, editirs. Brain tumors. 1st ed. NewYork: Churchill Livingstone; 1995.pp 125-162.
2. Lee TT, Manzano GR: Third ventricular glioblastoma multiforme; case report. Neurosurg Rev 1997; 20: 291-294.
3. Wen PY, Kesari S. Malignant gliomas in adults. N Engl J Med 2008; 359(5):492–507.
4. Sarnat HB, NetskyMG. Evolution of the Nervous System. New York,NY: Oxford University Press; 1974.
5. Prieto R, Pascual JM, Roda JM. Third ventricle glioblastoma. Case report and review of literature. Clin Neurol Neurosurg 2006;108(2):199–204.
6. Galli R, Binda E, Orfanelli U, et al. Isolation and characterization oftumorigenic, stem-like neural precursors from human glioblastoma.Cancer Res 2004;64(19):7011–7021.
7. Steffenhagen C, Dechant FX, Oberbauer E, et al. Mesenchymal stem cells prime proliferating adult neural progenitors toward an oligodendrocyte fate. Stem Cells Dev 2012;21(11):1838–1851.
8. Secer HI, Dinc C, Anik I, Duz B, GonulE :Glioblastoma of multiforme of lateral ventricle: report of nine cases. Br J Neurosurg 2008;22(3):398-401.
9. Chusid JG, and Gutierrez-Mahoney C: Glioblastomamultiforme of of septum pellucidum. J Neurosurg 1954;11(3):251-257.
10. Epstein JA and Epstein BS: Glioblastomamultiforme of bith the septum pellucidum and the corpus callosum. J Neurosurg1957;14(6):688-692.
11. ChoiSK,Byun BJ, Lee IS and Chi JG: Glioblastomamultiforme of the septum pellucidum: An autopsy case report. J Korean Neurosurg Soc. 1980;9(2):497-504.
12. McKean-Cowdin R, Razavi P, Barrington-Trimis J, et al. Trends in childhood brain tumor incidence: 1973–2009. J Neurooncol 2013;115(2):153–160.

Romanian Neurosurgery (2020) XXXIV (1): pp. 150-153
DOI: 10.33962/roneuro-2020-023
www.journals.lapub.co.uk/index.php/roneurosurgery

Efficacy of c-arm scopy-guided erector spinae plane block (ESPB) in postoperative pain control and reduction of opioid side effects in spinal instrumentation surgery

Yasar Karatas[1], Fatih Keskin[2], Mehmet Fatih Erdi[2], Bulent Kaya[1], Ibrahim Kosemen[1], Mehmet Kenan[2], Erdal Kalkan[1]

[1] Medova Hospital, Konya, TURKEY
[2] Necmettin Erbakan University, Meram Medical Faculty, Department of Neurosurgery, Konya, TURKEY

Keywords
lumbar,
instrumentation,
surgery,
pain,
erector spinae plane,
block

ABSTRACT

Background: Spinal instrumentation surgery causes significant pain in patients. As a result, patients are exposed to excessive opioid use and the associated side effects, as well as prolonged hospital stay, resulting in economic burden. Local anaesthetics can help both reduce postoperative pain and minimize the side effects associated with systemically administered opioid analgesics.

Objective: The aim of this retrospective study was to investigate the effect of erector spinae plane block on analgesia in spinal instrumentation surgery and to reduce the side effects of excessive opioid use.

Materials and Methods: We reported a retrospective study. Thirty patients, who underwent spinal instrumentation surgery from 2017 to 2018, were chosen from the hospital records. We performed spinal instrumentation and decompression by laminectomy to all patients under general anaesthesia. While 15 of these patients underwent erector spinae plane block, these patients received patient-controlled analgesia postoperative period. The other 15 patients received only patient-controlled analgesia postoperative period. We analyzed patients' data for differences in preoperative and postoperative visual analogue scores, nausea vomiting scores, constipation life quality scale, patient-controlled analgesia shot count and mean opioid consumption of patients.

Results: The data of 30 patients undergoing lumbar spinal instrumentation surgery were retrospectively analyzed. There was no significant difference in the age, preoperative VAS, preoperative ODI and sex between the two groups (p>0,05). In addition, there were statistically significant differences in postoperative VAS, postoperative ODI, Nausea Vomiting Score, Constipation Life Quality Score (CLQS), petidin consumption and PCA shot count (p <0.05). In all variables with significant differences, the values in the block group were lower than the non-block group.

Conclusion: ESPB provides effective analgesia and reduces side effects due to excessive opioid usage.

Corresponding author:
Yasar Karatas

Medova Hospital,
Department of Neurosurgery,
Konya, Turkey

yasarkrts@gmail.com

Scan to access the online version

Introduction

Open lumbar spine instrumentation surgery causes significant soft tissue damage because of large dissection and stripping the muscles. As a result, patients complain of nociceptive, neuropathic and inflammatory pain. [1]. In recent years, pain control after spinal surgery has been targeted with minimal opiate use. The use of gabapentinoids, acetaminophen, neural blockages and long-term local anesthetics is supported by scientific evidence [2,3]. Erector spinae plane block (ESPB) was defined for thoracic and abdominal pain previously [4,5]. There are various articles about postoperative pain control of erector spinae plane block. However, for the first time, we present a study showing that the amount of excessive opioid use is reduced and the associated side effects are reduced.

Materials and methods

We reported a retrospective study. This retrospective study included the data of thirty patients, who underwent spinal instrumentation surgery from 2017 to 2018. Preoperative and postoperative data such as age, sex, VAS, ODI, constipation quality life score, nausea vomiting score, petidine use, number of pca shot were collected. Each patient's outcome was noted by the same surgeon. Postoperative VAS, CLQS, nausea vomiting score measured in postoperative day 1 and postoperative ODI measured in postoperative day 30.

1. Surgery and postoperative protocol

The same surgeon performed all operations. Instrumentation and decompression by laminectomies were performed for all patient through a midline incision followed by stripping of paraspinal musculature. The patients underwent the same level surgical procedure. The participants had access to intravenous patient-controlled analgesia that comprised 1 mg/h continuous pethidine administration and 20 mg pethidine bolus infusion when requested by the participants. The bolus was permitted three times in a hour for all participants.

2. Erector spinae plane block technique

C-arm-scopy guided erector spinae plane block was performed just before surgıcal incision by the same surgeon. ESPB was made bilaterally at the level of instrumentation and decompression. After the patient was placed in prone position and sterile isolation was achieved, transverse process was determined by scopy. With a 20-G needle, the bottom of the transverse process was reached. The bilateral ESPB were performed by injecting 5% bupivacaine (20 mL into each side) for each level into the deep surface of the erector spinae muscle and the transverse processes of the lumbar vertebrae

3.Statistical analysis

Statistical analysis of the study was performed in R 3.1.2 package program. Descriptive measures of the quantitative variables in the study are shown with their median, minimum and maximum values. The suitability of the variables to normal distribution was examined by Shapiro Wilk test. Mann Whitney U test was used to compare two independent variables. Intragroup changes of variables were examined by Wilcoxon test. Fisher exact chi-square test was used for group comparison of qualitative variables. In all statistical analyzes in the study, p values less than 0.05 were considered statistically significant.

Results

There was no statistically significant difference between the groups in terms of age, preop VAS and preop ODI ($p > 0.05$). In addition, there were statistically significant differences in postop VAS, postop ODI, Nausea Vomiting Score, CLQS, petidine consumption and PCA Shot Count ($p < 0.05$). In all variables with significant differences, the values in the non-block group were higher than the block group (Table 1). The change in VAS and ODI values in the block group was statistically significant ($p < 0.001$). Similarly, the change in VAS and ODI values were significant in the non-block group ($p < 0.001$). Both VAS and ODI variables were significantly reduced in both groups (Figure 1). On the other hand, the mean decrease in VAS values in the block group was 79.0%, whereas the mean decrease in VAS values in the non-block group was 67.1% and there was a statistically significant difference between the groups in terms of VAS values ($p < 0.001$). In other words, there has been much more decline in the block group. (Figure 2) The mean decrease in ODI values in the block group was 55.4% and the mean decrease in ODI values in the non-block group was 48.3% and there was no statistically significant difference between the groups in terms of the decrease in ODI values ($p = 0.202$). Decreases in ODI values were similar in both groups (Figure 3). There

was no statistically significant difference between the groups in terms of gender (p = 1,000). In both groups, the ratio of men and women is 80% to 20% (Table 2).

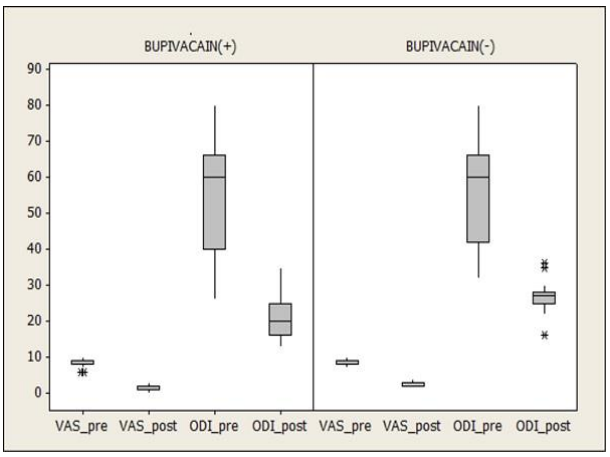


Figure 1. Both preoperative and postoperative VAS and ODI variables in the groups.

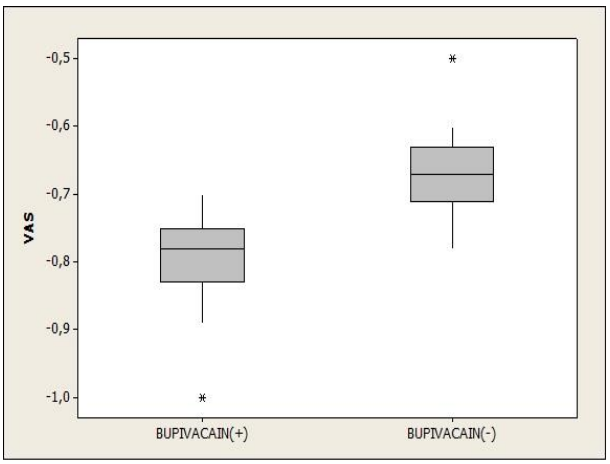


Figure 2. The mean decrease in VAS values in the groups.

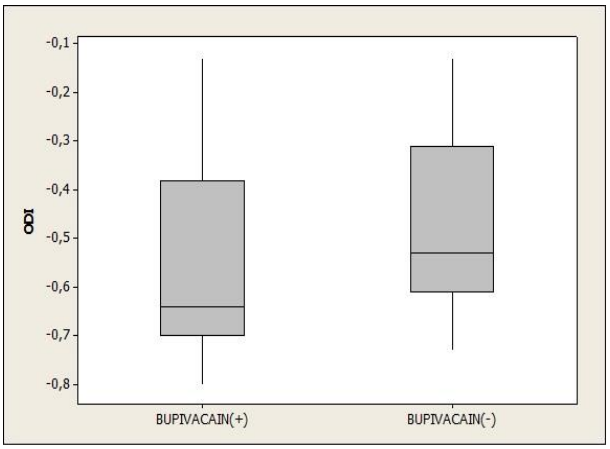


Figure 3. The mean decrease in ODI values in the groups.

Table 1. Mean age of groups, preoperative VAS, postoperative VAS, preoperative ODI, postoperative ODI, Nausea Vomiting Score, CLQS, petidine consumption and PCA Shot Count are shown in table.

	ESPB WITH BUPIVACAIN	ESPB WITHOUT BUPIVACAIN	p
Age	61,13 ± 18,28 66 (14-81)	64,20 ± 13,72 69 (38-85)	0,870
VAS preop	8,20 ± 1,21 8 (6-10)	8,73 ± 0,88 9 (7-10)	0,250
VAS postop	1,73 ± 0,70 2 (0-3)	2,87 ± 0,64 3 (2-4)	**<0,001**
ODI preop	54,47 ± 14,70 60 (26-80)	55,53 ± 14,03 60 (32-80)	0,806
ODI postop	22,13 ± 6,83 20 (13-35)	26,80 ± 4,81 27 (16-36)	**0,023**
Nausea Vomiting Score	1,27 ± 1,22 1 (0-4)	3,07 ± 1,71 3 (1-8)	**0,002**
CLQS	14,47 ± 7,76 15 (4-30)	26,93 ± 8,67 27 (16-42)	**<0,001**
Petidin Comsumption	42,53 ± 22,96 35 (12-91,6)	146,25 ± 29,83 149,3 (85-200)	**<0,001**
PCA Shot Count	1,80 ± 1,37 2 (0-5)	7,40 ± 1,60 8 (5-10)	**<0,001**

#Mann Whitney U test

Table 2. The ratio of men and women in the groups. #Mann Whitney U test

	ESPB WITH BUPIVACAIN	ESPB WITHOUT BUPIVACAIN	p
Male **Female**	3 (%20,0) 12 (%80,0)	3 (%20,0) 12 (%80,0)	1,000

#Fisher Exact Chi-Square test

Discussion

Posterior spine surgery is a very painful procedure for patients on the first postoperative day [6]. Although opioids are an important pillar of postoperative analgesia treatment, high doses given for severe pain control have important side effects such as cognitive impairment, respiratory depression, addiction, constipation, nausea and vomiting [7]. The risk of cardiac and respiratory complications increases in patients without effective pain control. In addition, delay in mobilization due to pain is a separate problem in these patients. Hospital stay is prolonged and chronic pain may occur [8]. The paravertebral muscles and the posterior elements of the spine are innervated by the dorsal roots of the

spinal nerves. Dorsal roots are separated from the root immediately after the exit of the spinal nerve from the foramen and are distributed in the superficial soft tissue. In the erector spinae plane block, the local anesthetic diffuses in the musculo-fascial plane and affects the dorsal roots of the spinal nerve at several levels [9]. In addition to systemic analgesic therapies, regional methods also play an important role in multimodal analgesic therapy. Neuraxial blocks, including patient-controlled bolus or continuous infusion of epidural and spinal blocks, are some of the regional methods. It is an important advantage that the ESPB only blocks sensory fibers compared to other peripheral nerve blocks [10, 11]. ESPB have been successfully used in abdominal surgery, neuropathic pain, pneumothorax and breast implant surgery [4, 12, 13]. In this study we used ESPB for postoperative pain relief after spinal instrumentation surgery. Chin KJ et al. demonstrated that interfacial blocks provide long-term postoperative pain control and reduce opioid use. 20 mL of local anesthetic injection has been shown to extend from the infiltration site to 3 or 4 levels caudal [14]. We used 20 mL bupivacaine injections for each side. Al-Alami et al. conducted a study describing the ultrasound-guided dorsal root block at the T12 and L5 level, but did not present a report containing patient outcome data [15]. We described a c-arm scopy-guided ESPB and reported the outcomes of patients about postoperative pain score, opioid use and its side effects.

Conclusion

In conclusion, on the basis of this retrospective study, it appears that c-arm scopy-guided ESP block, is a safe and effective technique for postoperative pain management, can reduce opioid use and its side effects

References

1. Mathiesen O, Dahl B, Thomsen BA, et al. A comprehensive multimodal pain treatment reduces opioid consumption after multilevel spine surgery. Eur Spine J 2013;22:2089-96.
2. Devin CJ, McGirt MJ. Best evidence in multimodal pain management in spine surgery and means of assessing postoperative pain and functional outcomes. J Clin Neurosci 2015;22:930-8.
3. Bajwa SJ, Haldar R. Pain management following spinal surgeries: An appraisal of the available options. J Craniovertebr Junction Spine 2015;6:105-10.
4. Forero M, Adhikary SD, Lopez H, Tsui C, Chin KJ. The erector spinae plane block: a novel analgesic technique in thoracic neuropathic pain. Reg Anesth Pain Med 2016; 41: 621-7.
5. Chin KJ, Adhikary S, Sarwani N, Forero M. The analgesic efficacy of pre-operative bilateral erector spinae plane (ESP) blocks in patients having ventral hernia repair. Anaesthesia 2017; 72: 452-60.
6. Gerbershagen HJ, Aduckathil S, van Wijck AJ, Peelen LM, Kalkman CJ, Meissner W. Pain intensity on the first day after surgery: a prospective cohort study comparing 179 surgical procedures. Anesthesiology 2013; 118: 934-44.
7. Savarese JJ, Tabler NG Jr. Multimodal analgesia as an alternative to the risks of opioid monotherapy in surgical pain management. J Healthc Risk Manag 2017; 37: 24-30.
8. Thepsoparn M, Sereeyotin J, Pannangpetch P: Effects of combined lower thoracic epidural/general anesthesia on pain control in patients undergoing elective lumbar spine surgery: a randomized controlled trial. Spine (Phila Pa 1976). 2018, 43:1381-85.
9. Saito T, Steinke H, Miyaki T, et al. Analysis of the posterior ramus of the lumbar spinal nerve: the structure of the posterior ramus of the spinal nerve. Anesthesiology 2013; 118: 88-94.
10. Sinatra RS, Torres J, Bustos AM: Pain management after major orthopaedic surgery: current strategies and new concepts. J Am Acad Orthop Surg. 2002, 10:117-29.
11. Andreae MH, Andreae DA. Regional anaesthesia to prevent chronic pain after surgery: a Cochrane systematic review and meta-analysis. Br J Anaesth 2013;111:711-20.
12. Bonvicini D, Giacomazzi A, Pizzirani E. Use of the ultrasound-guided erector spinae plane block in breast surgery. Minerva Anestesiol 2017; 83: 1111-2.
13. Ueshima H, Otake H. Erector spinae plane block provides effective pain management during pneumothorax surgery. J Clin Anesth 2017; 40: 74. [CrossRef]
14. Chin KJ, McDonnell JG, Carvalho B, Sharkey A, Pawa A, Gadsden J: Essentials our current understanding: abdominal wall blocks. Reg Anesth Pain Med. 2017, 42:133-83. 10.1097/AAP.0000000000000545
15. Al-Alami A, Abou El Ezz A, Kassab F. Ultrasound guideddorsal ramus nerve block for reduction of postoperative pain in patients undergoing lumbar spine surgery: a case series imaging study. Middle East J Anaesthesiol 2015;23:251-6.

Romanian Neurosurgery (2020) XXXIV (1): pp. 154-157
DOI: 10.33962/roneuro-2020-024
www.journals.lapub.co.uk/index.php/roneurosurgery

The role of strict patient-positioning during nursing in the management of intracerebral migration of gravitational bullet injury

Samir H. Aldelfi[1], Salima B. Alsaadi[2], Mohammed Maan AbdulAzeez[3], Mukarram N. Musarhad[1], Mohammed D. Khudhair[1], Samer S. Hoz[1]

[1] Neurosurgeon, Department of Neurosurgery, Neurosurgery Teaching Hospital, Baghdad, IRAQ
[2] Radiologist, Department of Neuroradiology, Neurosurgery Teaching Hospital, Baghdad, IRAQ
[3] Medical student, College of Medicine, Baghdad University, Baghdad, IRAQ

Keywords
patient-positioning, intracerebral migration, gravitational bullet injury

ABSTRACT

The intracranial migration of bullet was described in literature since Cushing time and the First World War [1]. The literature is still away from delivering a clear guideline and constitutes more of case reports rather than comprehensive well-designed studies [2-13], this mostly due to the variability and diversity in the presentation and management of such cases. The migration of bullet can be a sequel of any type of penetrating injury to the skull [14]. Intracranial migration after gravitational (falling) bullet injury is a unique type of injury that constitutes of significant human and material losses with differences in biomechanics and structural brain changes after the insult especially regarding the velocity of impact and the degree of yaw for the intracranially settled bullet [15]. The gravitational bullets injuries are considered by the international disease classification system as celebratory firing, that is quite common and is part of the traditional happy (marriage) or funeral event in the middle east in general and in rural areas of Iraq in particular, and also reported in some areas around the world (South America, North Africa, and middle of Asia) [15,16].

Corresponding author:
Mohammed Maan AbdulAzeez

College of Medicine,
Baghdad University,
Baghdad, Iraq

mohammed.wwt@gmail.com

Scan to access the online version

INTRODUCTION

In the available articles [17-28], the accepted management options for the retained intracranial bullet with possible migration, include:

- Observation, activity restriction and follow-up: for bullets with minimal risk of migration, those include small irregular bullet fragment with pure intraparanchymal location. Here, It is critical to know that the migration of bullet is rarely expected three months from the insult as the gliosis (encapsulated within tough scar) will surrounds the bullet and render it extremely resistant for migration [22,28].

- Early surgery: for bullets that retained within the ventrciles that have a high risk for obstructive hydrocephalus and those just close to a hematoma cavity [22-26].
- Delayed surgery: for migrating (moving) bullet that cause new neurological symptoms, large bullets with smooth (intact) contour, resides near the ventricles, moves toward an eloquent area, migrates for a significant distance (2.5 cm), moves freely in the cistern with risk of neurovascular compression or injury, and those resides in accesible and safe location. Still some authors consider surgical removal of the bullet once migrated based on their personal experience [27].

Theories on spontaneous bullet migration

Although the exact mechanism is not always known or being multifactorial [21], theories suggest different explanations for the mechanism, the trajectory and the final destination of the intracranially migrated bullet.

The gravidity, the pulsating intracranial contents and softening of the surrounding tissues are the mechanism that is commonly mentioned [26].

The common expected trajectory is usually posteriorly either toward the sella turcica for bullets in the anterior cranial fossa or toward the torchula herophili for those in the posterior parietal, posterior temporal and occipital lobes. Otherwise, the trajectory will be back along the initial bullet tract (the damaged brain). In rare occasions, the bullet may take out of the above suggested directions, here, surgeons should consider abscess development as a possibility [17-20,27].

Patient positioning after admission

For cases with retained intracranial bullet after gravitational (falling) bullet head injury, where the initial decision is close clinical follow-up (non-surgical), the patient positioning during admission is critical.

Although activity restriction is part of the general practice during this observation period, some surgeons suggest a more strict head positioning during the nursing period and consider it as pivotal part of the management of such instances [14,28].

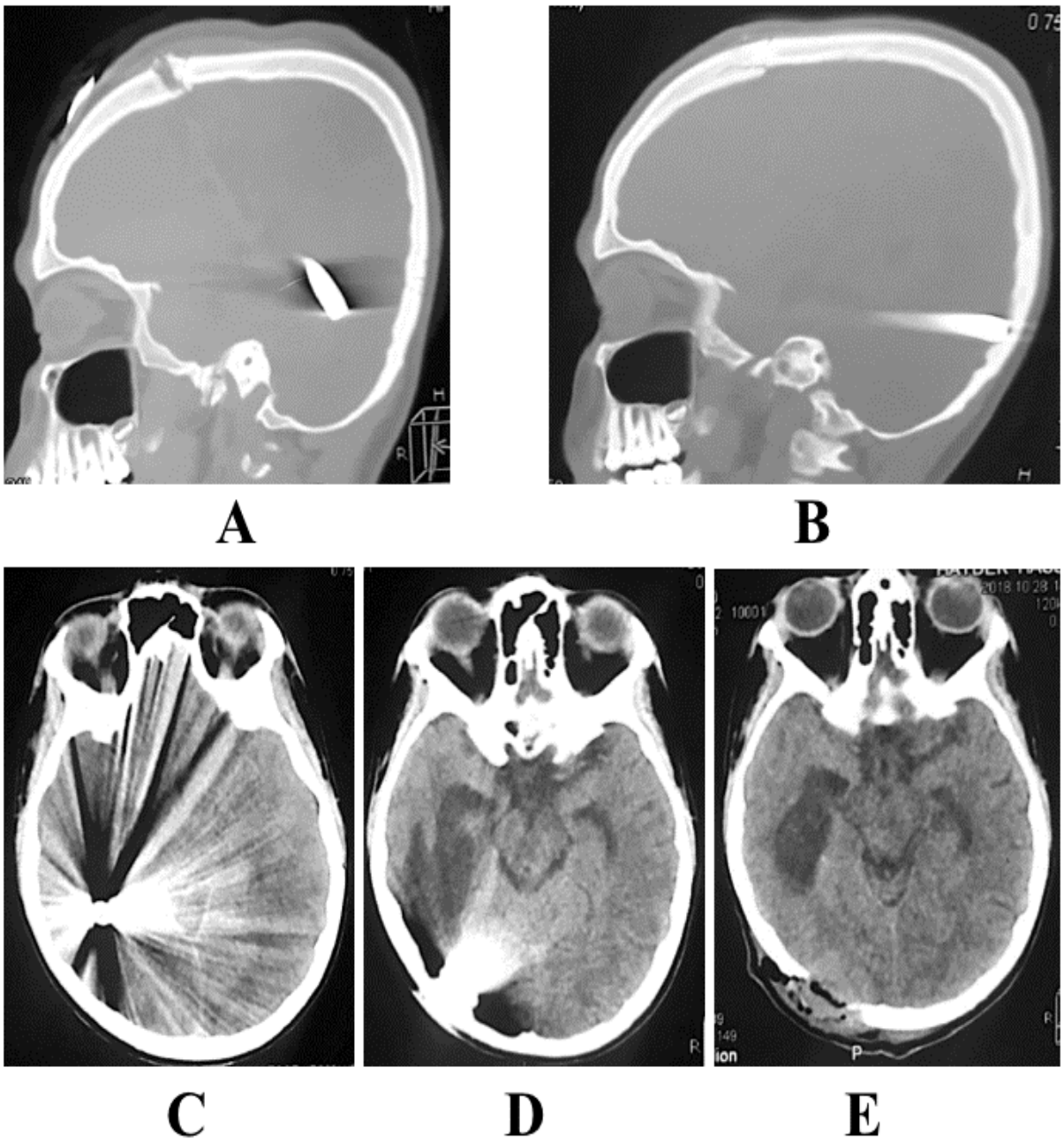
Both Rapp et al. [14] and Taarnhoj [28] describe in their reports that they use the strict patient positioning during nursing in few cases, the aim of this positioning is an attempt to change or control the direction of bullet migration in migration-possible cases. In our practice, we routinely implement this strict-positioning technique to render a deep bullet with possible migration to an accessible and easy to remove one.

Although Rapp et al. [14] and Taarnhoj [28] document using this positioning technique for penetrating bullet injury in general, our clinical experience suggests using this technique in gravitiational (falling) bullet injury will be more fruitful taking in consideration multiple factors that are unique for the falling bullets. The gravitiational (falling) bullets are usually non-fragmented, not very small nor too large, smooth outer contour and with less adhesive surrounding brain tissue (less cavitation and necrosis as compared to other types of penetrating bullet injuries), these factors make this falling bullets are more liable for migration and also more amenable to be affected and guided by the patient positioning during the early observation period (Figure 1 shows an illustrative case scenario for the mentioned technique).

Thus, in cases of gravitational cranial bullet injury with a retained bullet that not mandate early surgery, we recommend a patient-specific activity restrictions and strict head positioning taking in consideration the possible trajectories of migration and well-designed for dragging and controlling the bullet movement into an accessible and a safe to remove destination.

Figure 1. An illustrative case for the use of strict patient-positioning during nursing in the management of intracerebral migration of gravitational bullet injury. A twelve-years-old male, a victim of gravitational bullet injury, presented to the emergency department of the neurosurgery teaching hospital. Baghdad. Iraq with disturbed consciousness (Glasgow Coma Scale 14), left side weakness (Grade 4 both upper and lower limbs), pai in the sky (contralateral upper quadrantanopia) visual field defect, with a small wound (1 cm) in the right frontal area (inlet). Initial lateral skull X-ray showed the frontal inlet and the posterior temporal location of the bullet (A), that is confirmed by an urgent brain CT scan (C). The initial decision was to observe the patient as there were not significant wound or hematoma. The patient was put in strict supine position except for bathroom times with physiotherapy to his left side. Three days later, with the patient still having a severe headache, we did a follow-up skull x-ray then new brain CT scan that showed the migration of the bullet posteriorly (B and D respectively). Here, the decision was to remove the bullet as it moved to a surgically accessible location. the surgery went uneventful and the post-operative brain CT scan showed no

hematoma at the site of surgery (E). The patient discharged 4 days later, fully conscious with only the same initial visual field defect. On next visits his vision improved, he returned back to school and having good marks too.



References

1. Cushing H. A study of a series of wounds involving the brain and its enveloping structures. British journal of Surgery. 1917;5(20):558-684.
2. Duman H, Ziyal IM, Canpolat A: Spontaneous sub- falcial transcallosal migration of a missile to the contralateral hemisphere causing deterioration in neurological status— case report. Neurol Med Chir (Tokyo) 42:332-333, 2002.
3. Fujimoto Y, Cabrera HT, Pahl FH, de Andrade AF, Marino JR: Spontaneous migration of a bullet in the cerebellum—case report. Neurol Med Chir (Tokyo) 41:499-501, 2001.
4. Hughes BD, Vender JR: Delayed lead pulmonary em- boli after a gunshot wound to the head. Case report. J Neurosurg 105(3 Suppl):233-234, 2006.
5. Kamenski RS, Emch TM, Dastur KJ, Altschuler EM: Short interval spontaneous migration of an intra- cranial bullet fragment. J Trauma 65:E13-5, 2008.

6. Karabagli H: Spontaneous movement of bullets in the interhemispheric region. Pediatr Neurosurg 41: 148-150, 2005.
7. Ozkan U, Ozate ̧s M, Kemalog ̆lu S, Gu ̈zel A: Spontaneous migration of a bullet into the brain. Clin Neu- rol Neurosurg 108:573-575, 2006.
8. Hussain, M., Bari, M. (2011). Intracerebral migration of stray bullet leading to sudden and fatal worsening. Journal of the College of Physicians and Surgeons Pakistan, 21(3), 182-183.
9. Kumar R, Garg P, Maurya V, Sahu RN, Mahapatra AK. Spontaneous bullet migration-uncommon sequelae of firearm injury to the brain. The Indian Journal of Neurotrauma. 2008 Dec 1;5(2):119-21.
10. AbdulAzeez MM, Dolachee AA, Huber PZ, Romario MF, Obaid HA, Arakwazi AH, Altaweel MM, Hoz SS. Intracranial ricocheted-bullet injuries: An overview and illustrative case. Journal of Acute Disease. 2018 Sep 1;7(5):186.
11. Zafonte RD, Watanabe T, Mann NR. Moving bullet syndrome: a complication of penetrating head injury. Archives of physical medicine and rehabilitation. 1998 Nov 1;79(11):1469-72.
12. Abdulrahman AL, Shudifat, Kefah Taha, et al. Migrated Intracranial Falling Bullet, Crossed Midline Twice in a Conscious Pa ent. Med Case Rep. 2016, 2:2.
13. Rapp LG, Arce CA, McKenzie R, Darmody WR, Guyot DR, Michael DB. Incidence of intracranial bullet fragment migration. Neurological research. 1999 Jul 1;21(5):475-80.
14. Abdali HA, Hoz SS, Moscote-Salazar LR. Cranial gravitational (falling) bullet injuries: Point of view. Journal of neurosciences in rural practice. 2018 Apr;9(2):278.
15. Organisation mondiale de la santé, World Health Organization, WHO Staff, WHO. The ICD-10 classification of mental and behavioural disorders: clinical descriptions and diagnostic guidelines. World Health Organization; 1992.
16. Rengachary SS, Carey M, Templer J. The sinking bullet. Neurosurgery. 1992;30:291–295.
17. Milhorat TH, Elowitz EH, Johnson RW,etal. Spontaneous movement of bullets in the brain. Neurosurgery. 1993;32:140–143.
18. Wasserman SM, Cohen JA. Spontaneous migration of an intracranial bullet fragment. Mt Sinai J Med. 1979;46:512–515.
19. Leibeskind AL, Anderson RD, Schechter MM. Spontaneous movement of an intracranial missile. Neuroradiology. 1973;5:129 –132.
20. Grant GA. Migrating intracranial bullets: a rare occurrence. World neurosurgery. 2012 Mar 1;77(3):479-80.
21. Moores LE. Practical Considerations for the Management of Migrating Intracranial Bullets. World neurosurgery. 2012 Mar 1;77(3):481-3.
22. Campbell E, Howard WP, Weary WB. Gunshot wound of the brain: report of two unusual complications: bifrontal pneumocephalus and loose bullet in the lateral ventricle. Arch Surg. 1942;44:789–798.
23. Furlow LT, Bender MB, Teuber HL. Moveable foreign body within the cerebral ventricle: a case report. J Neurosurg. 1947;4:380–386.
24. Lang EK. Acute hydrocephalus secondary to occlusion of the aqueduct by a bullet. J LA Med Soc. 1969;121:167–168.
25. Masoudi MS, Ghafarpasand F, Yousefi S. A Migrating Retained Intracranial Bullet Presenting with Hydrocephalus and Alterations of Consciousness: A Case Report and Literature Review. Journal of Archives in Military Medicine. 2017;5(3).
26. Raimondi AJ, Samuelson GH. Craniocerebral gunshot wounds in civilian practice. Journal of neurosurgery. 1970 Jun;32(6):647-53.
27. Taarnhoj P. Gravity Instrumental in Removing an Intracerebral Missile: Case Report. Journal of neurosurgery. 1962 Apr 1;19(4):351-2.

Romanian Neurosurgery (2020) XXXIV (1): pp. 158-163
DOI: 10.33962/roneuro-2020-025
www.journals.lapub.co.uk/index.php/roneurosurgery

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

Krishna Govind Lodha, Tarun Kumar Gupta, Gaurav Jaiswal, Yogendra Singh

RNT Medical College Udaipur, Rajasthan, INDIA

ABSTRACT

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

Aim & 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.

Keywords
tribal region, paediatric traumatic brain injury

Corresponding author:
Krishna Govind Lodha

RNT Medical College Udaipur, Rajasthan, India

kg_lodha@yahoo.co.in

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. Pediatric TBI has different Pathophysiology due to higher vascularity, plasticity and less rigidity of scalp .pediatric 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]

Scan to access the online version

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 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 month to 18 years with a mean age of 9.25 years. There was 245 males (71.01%) and 100 females (28.98%) with male to female ratio of 2.45:1.

Table 1. Outcome according to age

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

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.

Table 2. Mode of injury

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%)

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.

Table 3. Severity and outcome

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%)

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)

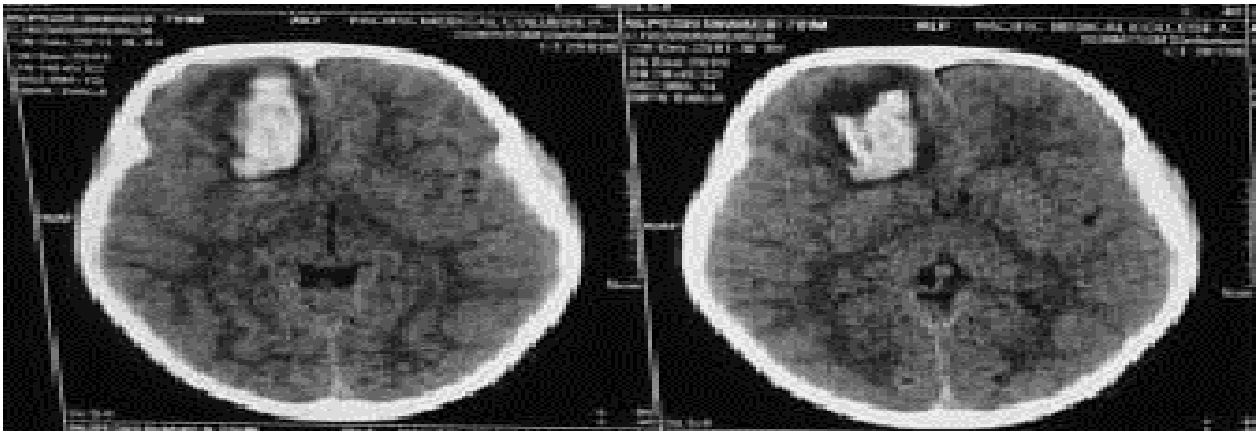


Figure 1. NCCT Head of 8 yrs old child admitted with h/o of fall from roof with GCS E2V3M5 Pupils b/l 3mm reacting to light.

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.

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

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

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).

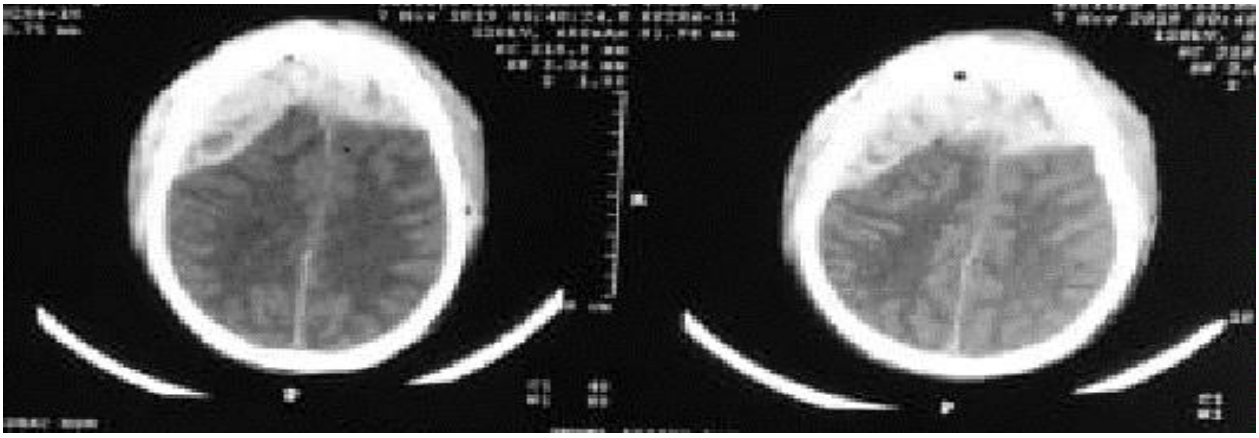


Figure 3 (a). NCCT Head of 15yrs male admitted with h/o RTA with GCS E2V1M4 Pupils b/l 3mm reacting to light undergone bifrontal craniectomy with evacuation of EHD. He discharged with GCS E4V5M6 and under gone 3D Mold customized cranioplasty after 4 months.

Figure 3 (b). Intraoperative photograph of 3D customised cranioplasty flap of above mentioned patient.

Table 5. Intervention and outcome

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%)

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.

Table 6. Intervention and outcome

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

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 Comman 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 Comman cause of pediatric head injury [7]. This peculiarly occure 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 out-come as described by Beca et [9].

The patient who had a GCS of 13-15 (218) had a poor out come in 8 (3.67%), followed by GCS of 9-12 (92) who had a poor outcome in 22(23.91%) followed be GCS of 8 or less then 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 Comman 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 Childers in our study irrespective to severity of the injury while author include only sever cases of head injury in peadtric 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].

Table 7. Glasgow outcome scale

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

GOS: Glasgow Outcome Scale

Conclusion

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.

Financial support and sponsorship: Nil
Conflicts of interest: There are no conflicts of interest

References

1. 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]
2. 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
3. Jennet B. Epidemiology of head injury. Arch Dis Child 1998; 78:403-6
4. 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]
5. International Institute for Population Studies. Mumbai, India: NF, India; 1998-9.
6. 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.
7. Sambasivan M. Epidemeology-Pediatric head injuries. Neurol India 1995; 43:57-8.
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.
9. Somatosensory evoked potentials for Prediction of outcome in acute severe brain injury. J Pediatr. 1995; 126:44–9. [Pub Med: 7815222]
10. 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]
11. 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]
12. Tomber g T, Rink U, Tikk A. Computerized tomography and prognosis in pediatric head injury. Acta Neurochir (Wien) 1996; 138:543-48.
13. 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]
14. 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.

Romanian Neurosurgery (2020) XXXIV (1): pp. 164-168
DOI: 10.33962/roneuro-2020-026
www.journals.lapub.co.uk/index.php/roneurosurgery

Resection of giant invasive thoracic schwannoma. Case report

A. Khelifa[1], F. Aichaoui[1], I. Assoumane[2], M. Al-Zekri[1], T. Bennafaa[1], A. Morsli[1]

[1] Neurosurgical Department of BEO University Hospital, Algiers, Algeria
[2] Department of Surgery, National Hospital of Niamey, Niger

Abstract
Even though spinal nerve sheath tumours, presented especially by schwannomas, are considered to be mostly benign; they can gain a huge size and have an invasive behaviour, causing spinal cord compression, bone destruction, and make the total removal of the tumour a real challenge for the surgeon. This type of tumours is recently described as giant invasive spinal schwannoma (GISS), this type rarely reported in the thoracic region; deserve a special studying vis-a-vis the diagnosis and the management of both the tumour and the bone destruction.

Keywords
schwannoma,
nerve sheath tumours,
spinal cord compression

Corresponding author:
Khelifa Adel

Neurosurgical Department of BEO University Hospital, Algiers, Algeria

drkhelifaadel@gmail.com

Introduction

Even though spinal nerve sheath tumors, presented especially by schwannomas, are considered to be mostly benign; they can gain a huge size and have an invasive behavior, causing spinal cord compression, bone destruction, and make the total removal of the tumor a real challenge for the surgeon. This type of tumors is recently described as giant invasive spinal schwannoma (GISS), this type rarely reported in the thoracic region (2); deserve a special studying vis-a-vis the diagnosis and the management of both the tumor, and the bone destruction.

Case presentation

The patient is a girl of 30 years old without past medical history, who consulted for a weakness of the lower limbs appeared 5 months before she consults and with a recent worsening; the clinical exam at the admission found a patient who present paraplegia with urinary urgency. Spinal CT then MRI was performed, objectified a spinal cord

Scan to access the online version

compression by an intra and extra spinal giant dumped shape process extending to the left thoracic cavity at the level of Th 8 and Th 9, measuring: 73x47x45 mm; hypointense on T1 weighted images, with heterogeneous signal on T2 weighted images and intense enhancement after gadolinium injection; the process was responsible of a destructive scalloping on the left pedicle and partial destruction of the posterior wall of Th 8, the tumor extends posteriorly to the left paraspinal muscles, and on the thoracic cavity in contact with the aorta (figures 1,2). There were no signs of neurofibromatosis. We operated the patient with the help of the thoracic surgery colleagues, under general anesthesia, and selective intubation of the right lung. Together we performed a total removal of the tumor through a combined approach. First a posterior midline approach was performed through which a spinal cord decompression was obtained, and then through a left posterolateral thoracotomy passing through the sixth interrib space a total removal of the tumor was achieved (figure 3). The thoracic part of the tumor was well encapsulated and easily dissected, but the intra spinal part lacks a capsule and was totally excised with piecemeal removal. A thoracic drain was left. The histological exam found a WHO grade I schwannoma. On post operative the patient was diagnosed with an atelectasis of the left lung which was managed with fibroscopic aspiration and steroids. The patient was oriented to physical medicine where she progressively improved, gaining control on her urinary behavior, then 5 weeks later she was able to walk. Post operative imaging performed 5 months later objectified a total removal of the tumor without residual or recurrence (Figure 4).

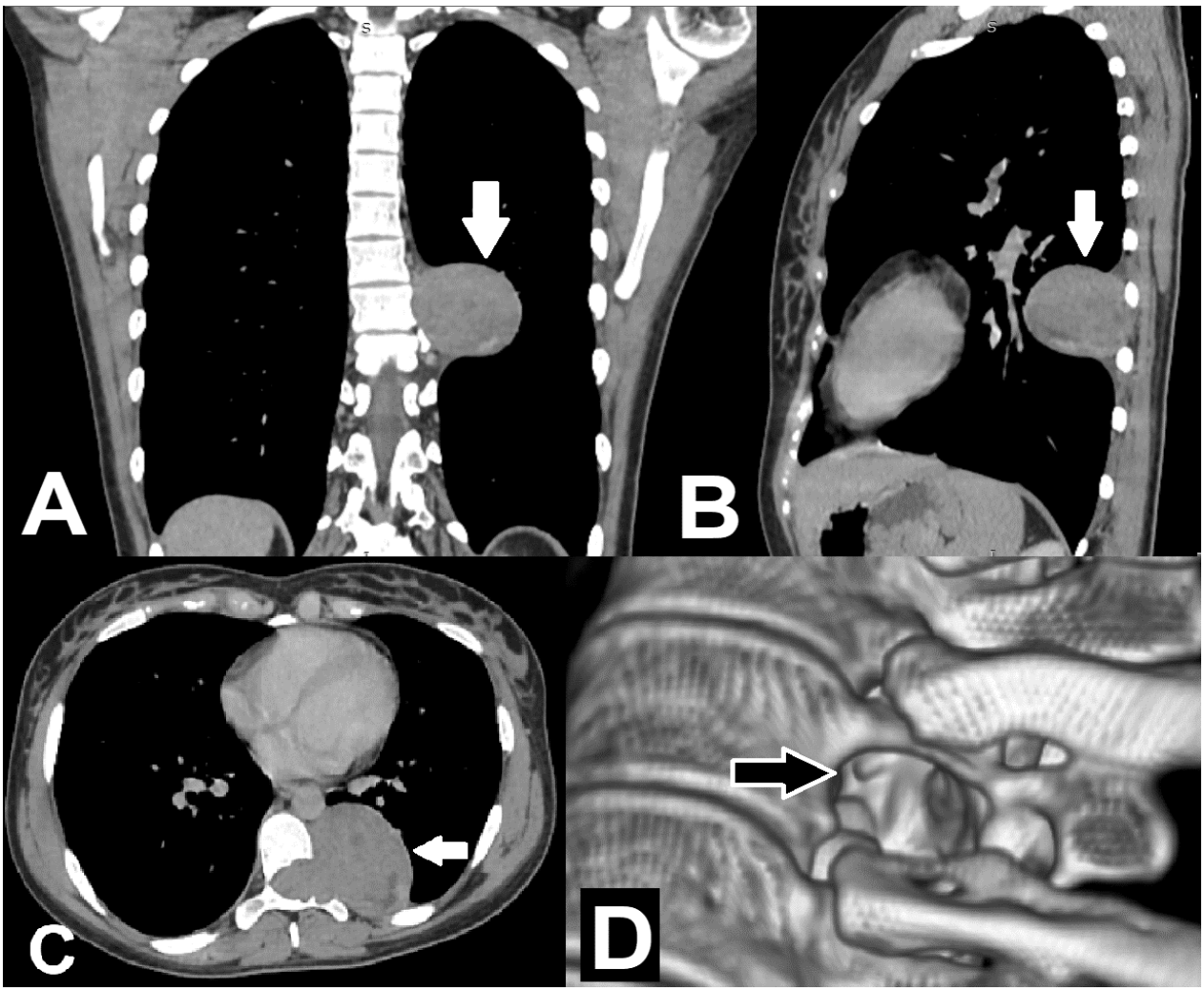


Figure 1. Preoperative thoracic CT; A: coronal reconstruction, B: sagittal reconstruction, C: axial slide; and D: 3D reconstruction. Note the extension of the tumor (white arrow) and the amount of bone destruction on the axial and 3D views.

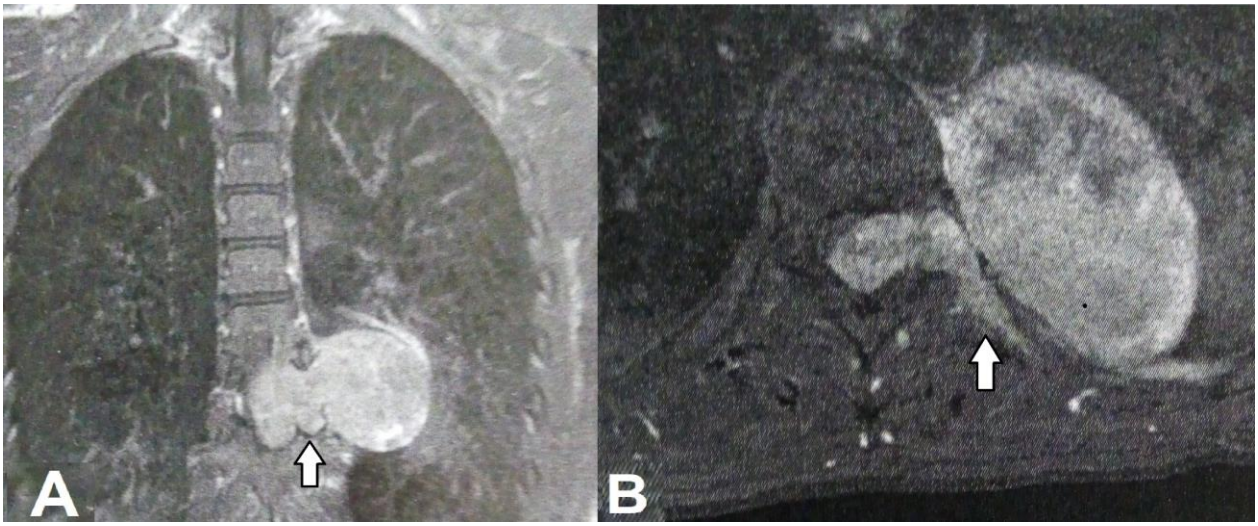
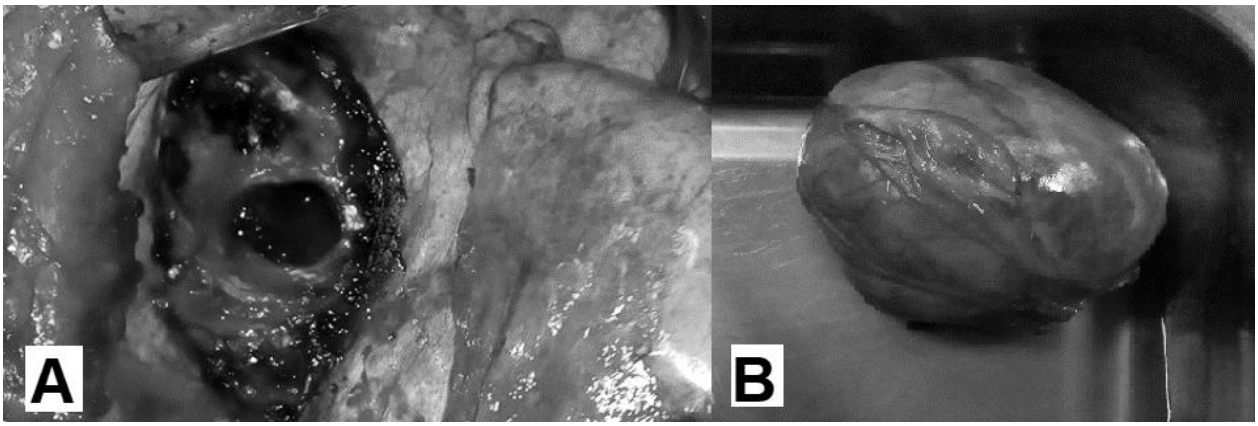


Figure 2. Preoperative T1 injected MRI; A: coronal slide, B: axial slide. Note the soft tissues infiltration (the arrow).



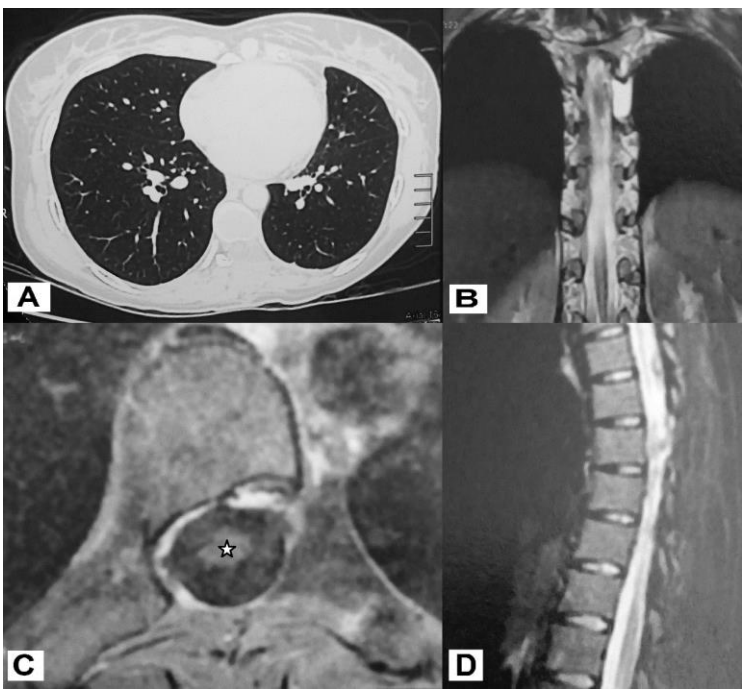


Figure 3. Peroperative views, A: a view through the interrib approach after removing the tumor; B: the intrathoracic part of the tumor. Note on the left image, the widened opening through which the tumor got through to invade the thoracic cavity; the black halo presents the parietal insert of the tumor.

Figure 4. Post-operative images, A: axial slide of a thoracic CT, B: T2 WI MRI coronal slide, C: T1 injected image axial slide, and D: T2 WI sagittal slide. Note the total removal, no residual or recurrence tumor, and the spinal cord decompression (the star).

Discussion

Nerve sheath tumor could be either schwannoma or neurofibroma. Neurofibromas produce a fusiform enlargement of the nerve where schwannomas are more smooth globoid tumors that develop eccentrically (1). Histological exams of schwannomas find usually elongated bipolar cells with fusiform darkly staining nuclei arranged in compact interlacing fascicles that tend to palisade (1). Nerve sheath tumors are mostly intradural but in 10% to 15% they are intra and extra dural in dumbbell shape (1). Nerve sheath tumors are malignant in 2.5% of cases and half of these cases are Neurofibromas (1). In 2001 Sridhar et al proposed a classification for benign nerve sheath tumors with a precise definition of the giant and the invasive spinal schwannomas (table 1); in the same paper the authors reported for the first time, cases of giant thoracic invasive spinal schwannomas (2). Giant invasive spinal schwannoma (GISS) although a benign lesion put the surgeon face to many difficulties related to the size, the infiltration, and the invasive nature. Big lesions need more exposure, though the selection of an adequate approach is mandatory. Midline approach might be sufficient (2) but for more exposure a lateral extracavitary or extracavitary costotrans-versectomies approaches provide access to the extraforaminal extension of the tumor (3); for tumor extending beyond the vertebral body a combined approach is recommended (4). Total removal is not possible in all cases (2,4), and that is mainly because the tumor might lack a capsule (2). The complications related to infiltration are represented especially by bone diffusion, in fact, some amount of bone destruction might jeopardize spinal stability and though the protection of neural structures, motion, and might cause some deformities. The evaluation of the consequences of bone infiltration classes these lesions into stable and unstable. Classically spine surgeons use Denis classification based on the three columns; Kostuik divided those columns on tow, right and left zones and considered a destruction of three or more of these six zones as an unstable lesion (5). Only instable lesions need spinal instrumentation. The lesion of our patient is considered stable, moreover some amount of infiltration is tolerable in the semi rigid spine (from Th3 to Th10) more than in the junctional or on the mobile spine, and that is the first criteria of stability evaluation in "The spinal instability neoplastic score (SINS)" adopted by some spine surgeons (3,6). Total removal of the tumor is the only factor related to long term outcome (2,4), in case where it is not possible, a decompression of neural structures is the priority, but a repeat surgery might be necessary (2).

	Classification
Type I	intraspinal tumor, < 2 vertebral segments in length; a: intradural; b: extradural.
Type II	intraspinal tumor > 2 vertebral segments in length (**giant tumor**)
Type III	intraspinal tumor with extension into nerve root foramen
Type IV	intraspinal tumor with extraspinal extension (dumbbell tumors); a: extraspinal component < 2.5 cm; b: extraspinal component > 2.5 cm (giant tumor)
Type V	tumor with erosion into the vertebral body (**giant invasive tumor**), lateral and posterior extensions into myofascial planes

Table 1. Sridhar et al classification for benign nerve sheath tumors (2).

Conclusion

Giant invasive spinal schwannoma is a benign lesion with high bone destruction potential; its infiltration nature to the surrounding soft tissues could make the total resection a real challenge. Knowing the real instable spinal lesions will orient the indication to spinal instrumentation.

References

1. Basheal M. Agrawal, Barry D. Birch, Paul C. McCormick, Daniel K. Resnick, E.C. Benzel. Intradural Extramedullary Spinal Lesions. Spine Surgery. Techniques, Complication Avoidance and Management. 3d edition. CHAPTER 102. P992.
2. Sridhar K, Ramamurthi R, Vasudevan MC, Ramamurthi B: Giant invasive spinal schwannomas: definition and surgical management. J.Neurosurg. 2001, 94:210–215.
3. Edison P. Valle-Giler, Juanita Garces, Roger D. Smith, Wale A. R. Sulaiman. One-Stage Resection of Giant Invasive Thoracic Schwannoma: Case Report and Review of Literature. The Ochsner Journal 14:135–140, 2014.
4. Nam Hun Yu, Soo Eon Lee, Tae-Ahn Jahng, Chun Kee Chung. Giant Invasive Spinal Schwannoma: Its Clinical Features and Surgical Management. Neurosurgery, Volume 71, Issue 1, July 2012, Pages 58–67, https://doi.org/10.1227/NEU.0b013e31824f4f96.
5. William F. Lavelle; E.C. Benzel. Thoracic and

Thoracolumbar Spinal Tumors: Regional Challenges. Spine Surgery. Techniques, Complication Avoidance and Management. 3d edition. CHAPTER 113. P1117.

6. Shandy Fox, Michael Spiess, Luke Hnenny, and Daryl. R. Fourney. Spinal Instability Neoplastic Score (SINS): Reliability Among Spine Fellows and Resident Physicians in Orthopedic Surgery and Neurosurgery. Global Spine Journal. 2017, Vol. 7(8) 744-748. DOI: 10.1177/2192568217697691.

Guidelines for authors

1. Ethics

The publication of an article in Romanian Neurosurgery is a direct reflection of the quality of the work of the authors. The prevention of publication malpractice is first the responsibility of every author and also of our editorial board. Authors must submit accurate information and sufficient details, presenting its objective significance; unethical behaviour is unacceptable.

Plagiarism in all its forms constitutes unethical publishing behaviour and is unacceptable.

For Romanian Neurosurgery the publication ethics and publication malpractice statement are consistent with the recommendations and guidelines of the Committee on Publication Ethics, the World Association of Medical Editors, the International Committee of Medical Journal Editors and Consolidated Standards of Reporting Trials.

Links:
Committee on Publication Ethics
(COPE): http://www.publicationethics.org
World Association of Medical Editors
(WAME): http://www.wame.org
International Committee of Medical Journal Editors
(ICMJE): http://www.icmje.org

2. Enclosed Letter

In addition to the manuscript, the Editorial Board should receive an enclosed letter containing the exclusive reservation of copyright guaranteed by all authors whose manuscripts have already been accepted. If the paper was completely or partially published or exposed previously, a copy or a photocopy of it should be also sent. The technical reports should contain a declaration concerning the financial sources that cover the costs necessary for instruments and methodology acquisition.
In order to illustrate different cases, photos of identifiable patients will not be published without their legal consent or that of their legal representative. The letter containing this consent together with the manuscripts should be sent to the editorial office.
If the author wishes his unpublished manuscripts returned, please note this in the enclosed letter.

3. Sending of manuscripts

Authors shall ensure that the article has been "spell and grammar checked" prior to the submission.
The manuscript sent for publishing must be submitted in English. The manuscript will be typed without formatting, with a 1-line space. Please enclose 1 copy of the manuscript, tables, graphics and photos. After publishing, the paper and pictures become the property of Romanian Neurosurgery.

There are no article processing charges (NO APCs) and no article submission charges.

4. Manuscript elaboration

Paper sent for publishing should be in accordance with international standards of manuscript submittance. These standards are mentioned in "British Medical Journal" 1988; 296: 404-405 or in "Annals of Internal Medicine" 1988; 108:258-265. The authors are responsible for the accuracy of the information contained in the essay.

4.1. The title page should contain the whole title of the essay and complete names of authors with their academical degrees. If it is necessary, the department, the hospital or the institution where the search has been undertaken, should be also mentioned.

4.2. Please include an additional page containing the title of the essay and the author responsible for correcting of any and of all mistakes and for maintaining correspondence. The address, phone and fax number should be included (e-mail be available).

4.3. A summary, no longer than 300 words, should be written on a separate page. Key-words, no more than 7, should be listed in alphabetical order on the same page. The use of keywords should be approved by the "Index Medicus".

4.4. Text. The introduction should specify the purpose of the paper. The content and the method should give a minute account of the work methodology so that the experiment conclusion could be reproduced and checked up on in other centres. The experiments and medical studies performed on human beings should respect the principles specified in "The Declaration of Helsinki", whereas the experiments done on animals should be in accordance with "The Principles Charta of Animal's Care and Use". The results should not contain references to previous studies. The discussions should reflect the main features of the research.

5. References

- typed on 1 line;
- quoted in alphabetical order;
- explanatory footnotes are not accepted;
- unpublished data and personal papers should be quoted inside the text and not in the bibliography;
- entries to the bibliography should appear in the following order: the authors, the title of the essay, the title of the periodical (abbreviated according to the list of abbreviation specified in the "Index Medicus"), the volume number, the page number and the data publication.

6. Tables and pictures

There should be 1 copy sent. Each table with its own title should be submitted on separate pages. The photos, radiographies, CT scans should be labelled on the back with

the number of each picture, corresponding with the number included inside the text, and the author's name. The label will be placed on the top of the picture. The drawings may be sent on vellum paper, tracing paper or transparent paper. The use of pictures belonging to other publications is accepted, only if mentioning the original source. Further, the partial use of any previously published text is accepted with the approval of its author and editor. All pages should be consecutively numbered, starting with the title page.

7. Abbreviations

Abbreviations should be consistently used throughout the text and established in a fixed form from the beginning.

8. Submission

Manuscripts should be sent to the Editor:
Dr St. M. Iencean
Romanian Neurosurgery
"Gr.T. Popa" University of Medicine and Pharmacy
Universitatii Street, No. 16, 700115, Iasi, Romania
or by e-mail to ***mirceasteffan@yahoo.com***.

9. Disclaimer

The editors disclaim any responsibility for opinions expressed in the papers.

10. The review process

After the manuscript submission, the peer review process is broken down into the following steps:
1. The Editor assigns Reviewers to the manuscript.
2. The Reviewers review the manuscript.
3. The Editor drafts a decision to be sent to the author/authors.

The review process takes between three weeks and two months.