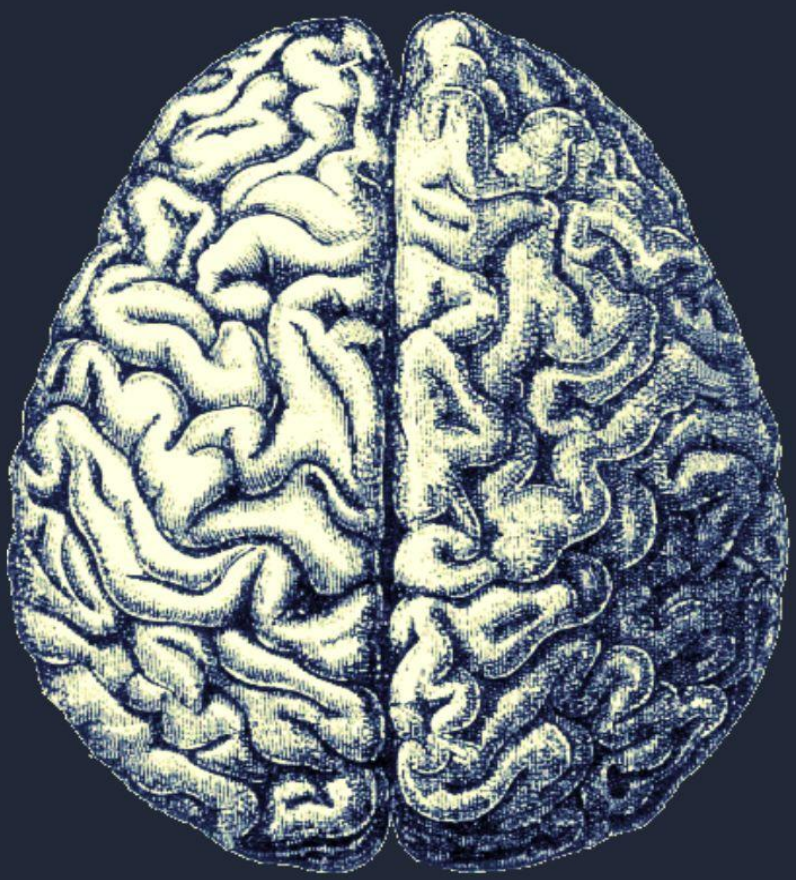
ISSN 1220-8841 (Print)
ISSN 2344-4959 (Online)

ROMANIAN NEUROSURGERY

Vol. XXXIX | No. 1 March 2025



The official journal of
"Romanian Society of Neurosurgery"
- Est. 1982 -

LONDON ACADEMIC PUBLISHING

Editorial and Advisory Board

ROMANIAN

NEUROSURGERY

Vol. XXXIX | No. 1 March 2025

London
Academic Publishing

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

First Printing: March 2025
London Academic Publishing Ltd.
27 Old Gloucester Street
WC1N 3AX
London, United Kingdom
Email: contact@lapub.co.uk

london-ap.uk
lapub.co.uk
journals.lapub.co.uk
journals.lapub.co.uk/index.php/roneurosurgery

Company Reg. No. 10941794
Registered in England and Wales

Contents

Romanian Neurosurgery (2025) XXXIX (1): pp. 9-13
DOI: 10.33962/roneuro-2025-001
www.journals.lapub.co.uk/index.php/roneurosurgery

Syringomyelia resulting from aneurysmal subarachnoid haemorrhage. One case report and literature review

Alexandru Ioan Bira[1], Horia Ples[1,2]

[1] Emergency County Hospital, Timisoara, ROMANIA
[2] "Victor Babes" University of Medicine and Pharmacy, Timisoara, ROMANIA

ABSTRACT

Syringomyelia, characterized by cystic cavitation in the spinal cord, can be primary or secondary to various conditions.[1] This article focuses on syringomyelia resulting from subarachnoid haemorrhage (SAH). A case study of a 45-year-old female with SAH is presented. Despite successful aneurysm clipping, the patient developed post-SAH syringomyelia, a rare complication occurring in less than 1% of SAH cases. [2] The pathophysiology involves arachnoiditis disrupting cerebrospinal fluid dynamics, leading to spinal cord tethering. Multiple hypotheses, including inflammatory responses and disruptions in glymphatic flow, contribute to syrinx formation.[4] Surgical options, from arachnoid lysis to various shunting procedures, aim to address progressive symptoms. The choice remains case-specific, with debates on long-term shunt efficacy. Overall, syringomyelia post-SAH poses diagnostic and therapeutic challenges, emphasizing the need for further research in understanding and managing this rare complication.

Keywords
syringomyelia,
cerebral aneurysm,
subarachnoid haemorrhage

Corresponding author:
Horia Ples

"Victor Babes" University of Medicine and Pharmacy, Timisoara, Romania

horia.ples@gmail.com

Scan to access the online version

INTRODUCTION

Syringomyelia or syrinx represent the cystic cavitation of the spinal cord.

Syrinx can be either primary, idiopathic, without an identifiable cause, or secondary to a myriad of conditions, including Chiari type I malformation, spinal cord trauma, inflammatory/ postinfectious conditions such as arachnoiditis, and both spinal and posterior fossa neoplasms, among others. [1]

Syringomyelia largely affects children and young adults. The prevalence has been estimated at 9 per 100,000 people, and an incidence of 0.44 cases per year has been cited in the literature, but epidemiological data on syringomyelia is limited. Chiari malformations are responsible for nearly half of the affected population, while spinal cord trauma and arachnoiditis account for another quarter of adult patients with syringes. [1]

Milhorat developed a classification of syringomyelia based on pathophysiology by comparing information obtained from 175

autopsies with clinical data on 927 patients with syringomyelia. Syringes are divided into four categories on the basis of pathologic findings: (1) dilations of the central canal that are anatomically continuous with the fourth ventricle (communicating syringomyelia); (2) noncommunicating syringomyelia, including dilations of the central canal that do not communicate with the fourth ventricle and extracanalicular syringes that originate in the spinal cord parenchyma and do not communicate with the central canal or fourth ventricle (primary parenchymal cavitations); (3) atrophic syringes occurring with myelomalacia (syringomyelia ex vacuo); and (4) neoplastic cyst (Table 1). [6]

In this article, we will focus our attention on syringomyelia resulting from subarachnoid hemorrhage.

Table 1.

Class Type of Syringomyelia
I. Communicating syringomyelia • Central canal dilations 1. Communicating hydrocephalus (posthemorrhagic, postmeningitic) 2. Complex hindbrain malformations (Chiari type II, encephalocele) 3. Dandy-Walker malformation
II. Noncommunicating syringomyelia • Central canal/paracentral syringes 1. Chiari malformation 2. Basilar invagination 3. Spinal arachnoiditis (posttraumatic, postmeningitic) 4. Extramedullary compression (spondylosis, tumors, cysts) 5. Tethered cord 6. Acquired tonsillar herniation (hydrocephalus, intracranial mass lesions) • Primary parenchymal cavitations 1. Spinal cord trauma 2. Ischemia/infarction 3. Intramedullary hemorrhage
III. Atrophic cavitations (syringomyelia ex vacuo)
IV. Neoplastic cavitations

Modified from Milhorat TH, Johnson RW, Milhorat RH, et al. Clinicopathological correlations in syringomyelia using axial magnetic resonance imaging. *Neurosurgery.* 1995;37:206-213.

CASE PRESENTATION

We present the case of a 45-year-old female patient, first admitted to our clinic in 2018 with subarachnoid hemorrhage Fischer Grade II, Hunt and Hess Grade II, and WFNS Grade I. Cerebral CT angiography revealed cerebral aneurysms of the anterior communicating artery and the left posterior inferior cerebellar artery.

Analyzing the subarachnoid hemorrhage pattern, the rupture of the anterior communicating artery aneurysm was identified, leading to surgical intervention for aneurysm clipping. The neurological outcome was favorable.

However, 7 days after the surgical procedure, the patient's neurological status deteriorated abruptly. A cerebral CT scan revealed infratentorial subarachnoid hemorrhage with intraventricular extension (Fischer Grade IV) and secondary hydrocephalus. Emergency neurosurgical intervention included the placement of an external ventricular drain and clipping of the left posterior inferior cerebellar artery aneurysm. Subsequent progress was slow, but favorable.

In the course of recovery, the patient required the placement of a permanent ventriculo-peritoneal shunt. She was discharged conscious, Glasgow Coma Scale (GCS): 15p, ambulating with assistance, and a modified Rankin Scale (mRS) score of 2, one month after admission.

In 2022, the patient presented to our clinic complaining of neck pain, paresthesia in the upper and lower limbs, progressive spastic tetraparesis and loss of thermal and pain sensitivity especially in upper limbs. The symptoms had started six months prior to the presentation. The MRI revealed obstruction of the outflow pathways of the fourth ventricle and holocord syringomyelia. (Fig. 1)

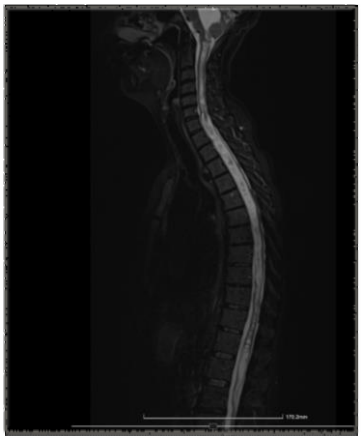


Figure 1.

Considering the imaging findings and the symptoms, a decision was made to perform posterior fossa decompression through adhesiolysis and

the placement of a cervical syringo-subarachnoid shunt via midline myelotomy and duraplasty.

Immediately postoperatively, there was an worsening of motor deficits (from muscular force 4/5 to 3/5), and she develop neurogenic bladder. After a few days, the symptoms subsided. The patient is discharged 10 days postoperative. At discharge, she is conscious, Glasgow Coma Scale (GCS) score of 15, mobilizing with assistance, with pre-operative spastic tetraparesis with overall muscle strength of 4/5, preservation of sensory disturbances, and improvement in pain symptoms.

At the 6-month follow-up, a reduction in the size of the syringomyelic cavity at the cervical level was observed, but the syringomyelia in the thoracic-lumbar region persisted. Currently, the patient is not inclined towards undergoing another intervention.

Discussion

Post-subarachnoid hemorrhage syringomyelia occurs in less than 1% of subarachnoid hemorrhage cases.[2] Given the rarity of this complication and the absence of specific guidelines and recommendations, we sought information in the literature. Searching on PubMed with the keywords "syringomyelia" and "subarachnoid hemorrhage," we identified 10 articles addressing this subject. Searching in the literature, we identified several risk factors in the development of this late complication, including ruptured aneurysms in the posterior circulation, intraventricular extension of hemorrhage, and the need for cerebrospinal fluid drainage [2]. These risk factors align with the case of our patient.

Given the physiopathology of this entity, it can be stated that the involved mechanism is not fully understood. Several hypotheses have been proposed.

It is considered that the presence of blood in the subarachnoid space acts as a trigger. Hemoglobin degradation products generate activation and recruitment of inflammatory cells. Ultimately, this inflammatory mechanism leads to fibroblast activation with collagen synthesis and the development of chronic arachnoiditis. The formation of adhesions in the subarachnoid space leads to tethering of the spinal cord and disturbances in cerebrospinal fluid dynamics. Adhesions in the cerebellomedullary cistern obstruct the outlet of the fourth ventricle thereby inhibiting the cerebrospinal fluid flowing out of the fourth ventricle and leading to hydrocephalus, which directly impacts and dilates the spinal cord central canal, thus ultimately leading to syringomyelia. In the case of our patient, this mechanism seems to be involved, considering the obstruction of the obex by adhesions.[2,6] (Fig. 2)

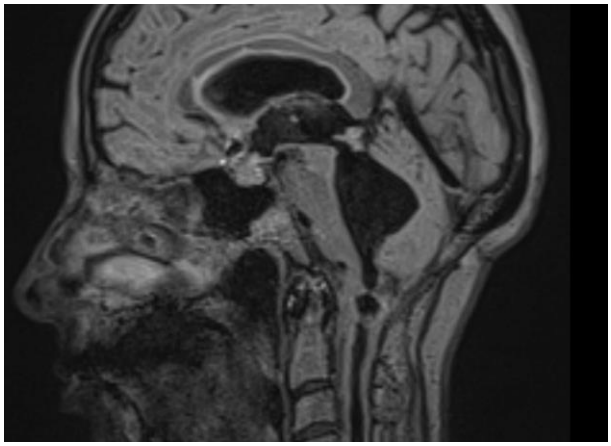


Figure 2.

This mechanism bears resemblance to the hydrodynamic theory proposed in the 1960s by Gardner. The theory suggested that cerebrospinal fluid (CSF) pulsations originating from the choroid plexus typically contribute to the expansion of the neural tube during development. According to the theory, imbalanced CSF pulsations between the supratentorial and infratentorial spaces during development could lead to the formation of a small posterior fossa, tonsillar ectopia, and the redirection of CSF from the fourth ventricle into the central canal due to the obstruction of fourth ventricular outflow tracts at the foramen magnum.[1]

Another mechanism that could be involved is represented by disruptions in the glymphatic flow. The glymphatic system is the internal mechanism through which the central nervous system accomplishes metabolic clearance. It has been demonstrated that within the spinal cord, there exists the perivascular network necessary for the functioning of this system. [7,8]

We know from studies conducted on the brain that the presence of blood in the subarachnoid space leads to a blockage of the glymphatic system pathways [8]. We have no reason to believe that, in the context of presence of subarachnoid hemorhage in spinal subarachnoid space, blood cannot disrupt the functioning of the spinal glymphatic flow.

In a 2017 study examining glymphatic system in normal pressure hydrocephalus, the intrathecal administration of an MRI contrast agent (gadobutrol), serving as a cerebrospinal fluid tracer, revealed indications of delayed glymphatic clearance in iNPH patients compared to a reference group[9]. Consequently, it is possible that the glymphatic flow may contribute to hydrocephalus in patients with subarachnoid hemorrhage, both in the acute phase and later through its permanent impairment. Although we are aware of the speculation we are raising, we consider that the disruption of glymphatic flow is involved in the development of syringomyelia following subarachnoid hemorrhage. Of course, the mechanism is not well-defined, and further studies are absolutely necessary.

Regarding the surgical treatment, the recomandation it is to treat the patient with progressive symptoms.

Surgical options for treatment of symptomatic syringomyelia include direct lysis of arachnoid adhesions with or without duraplasty and a variety of shunting procedures, including syringo-subarachnoid shunt, syringo-pleural, and syringo-peritoneal. [1,2,3]

Certain authors argue that the primary treatment objective should be the release of adhesions through duraplasty. Advocates of this approach assert that arachnoid lysis addresses the fundamental disruption in cerebrospinal fluid (CSF) flow and enlarges the subarachnoid space, as opposed to merely redirecting CSF. Klekamp and colleagues observed that among 67 patients treated for progressive symptoms, 97% of those who underwent syrinx shunting experienced recurrence during follow-up, whereas 78% of those who underwent arachnoid release and duraplasty remained stable.[10]

Shunting of the syrinx to the subarachnoid, pleural, or peritoneal space is still widely accepted as the treatment of choice for patients with syringomyelia caused by arachnoiditis. Shunting of the syrinx to the subarachnoid or peritoneal cavity was associated with a recurrence rate of 60%, whereas microsurgical dissection of the arachnoid scar and decompression of the subarachnoid space had a recurrence rate of 33%, with a mean follow-up period of 28 months. Successful long-term management of the syrinx was associated with microsurgical dissection of the arachnoid scar and decompression of the subarachnoid space. [2,3]

These data appear to be supported by the conclusions of other articles. In a study published in 2011 the conclusion is that shunting proves effective in alleviating the pressure of the syrinx on the spinal cord tissue, but its long-term impact is limited, with recurrence rates varying from virtually all patients experiencing a recurrence to 60% . This outcome is not unexpected, considering that shunting does not address the root cause of syrinx formation; rather, it merely alleviates the symptoms.[4]

Regarding the effectiveness of shunts no statistically significant difference in efficacy between these methods was highlighted.

In a meta-analysis published in 2021, assessing the complication rates of syringo-subarachnoid, syringo-peritoneal, and syringo-pleural shunts, regardless of the cause that led to the development of syringomyelia, syringopleural shunts may offer the lowest rate of reoperation.[6]

In conclusion, our opinion is that in the case of syringomyelia resulting from post-subarachnoid hemorrhage arachnoiditis, the primary goal of the intervention is the lysis of adhesions and the opening of the subarachnoid space. If distal to the site of adhesiolysis there are no other areas of chronic arachnoiditis, we recommend the placement of a syringo-subarachnoid shunt. When distant adhesions are evident on preoperating MR imaging, local lysis of adhesions is unlikely to open the subarachnoid space well enough for effective syrinx to subarachnoid drainage, so we recommand syringo-pleural or syringo-peritoneal shunt. [2]

Conclusions

Syringomyelia as a complication following subarachnoid hemorrhage is a rare occurrence, with limited reported cases in the literature, accounting for less than 1% of SAH patients. The exact pathophysiology of syringomyelia post-SAH is not fully understood but may involve arachnoiditis, which disrupts cerebrospinal fluid dynamics and leads to spinal cord tethering. Surgical management is a viable option for patients with progressive symptoms, and until this moment we have more questions than answers about this entity.

References

1. Youmans and Winn Neurological Surgery, 4-Volume Set, 7th Edition.
2. Syringomyelia and spinal arachnoiditis resulting from

aneurysmal subarachnoid hemorrhage: Report of two cases and review of the literature Taylor J. Abel, Matthew A. Howard, III, Arnold Menezes.
3. Spinal syringomyelia following subarachnoid hemorrhage Kinya Nakanishi ⇑ , Takuya Uchiyama, Naoki Nakano, Norihito Fukawa, Kimito Yamada, Tomonari Yabuuchi, Amami Kato.
4. Subarachnoid Hemorrhage-associated Arachnoiditis and Syringomyelia J. Eneling · S. Boström · S. Rossitti.
5. Syrinx shunts for syringomyelia: a systematic review and meta-analysis of syrin-gosubarachnoid, syringoperi-toneal, and syringopleural shunting. Robert J Rothrock, Victor M Lu, Allan D Levi.
6. Classification of syringomyelia , T H Milhorat.
7. The ultrastructure of spinal cord perivascular spaces: Implications for the circula-tion of cerebrospinal fluid Magdalena A Lam 1, Sarah J Hemley 2, Elmira Najafi 2, Nicole G F Vella 3, Lynne E Bilston 4, Marcus A Stoodley.
8. The Glymphatic System: A Beginner's Guide. Nadia Aalling Jessen 1, Anne Sofie Finmann Munk 2, Iben Lundgaard 2, Maiken Nedergaard.
9. Glymphatic MRI in idiopathic normal pressure hydrocephalus. Geir Ringstad,1,2 Svein Are Sirirud Vatnehol,3 and Per Kristian Eide.
10. Treatment of syringomyelia associated with arachnoid scarring caused by arach-noiditis or trauma, J Klekamp, U Batzdorf, M Samii, H W Bothe.

Romanian Neurosurgery (2025) XXXIX (1): pp. 14-21
DOI: 10.33962/roneuro-2025-002
www.journals.lapub.co.uk/index.php/roneurosurgery

Remember: 30 years since Constantin Arseni passed away. The founder of Romanian neurosurgery

Alexandru Vlad Ciurea[1,2,3], Horia Ples[4,5], Aurel Mohan[6,7], Milena-Monica Ilie[1], Matei Serban[1]

[1] Department of Neurosurgery, University of Medicine and Pharmacy "Carol Davila", Bucharest, ROMANIA
[2] Honorary Member of the Romanian Academy
[3] Neurosurgery Department, Sanador Clinical Hospital, Bucharest, ROMANIA
[4] Centre for Cognitive Research in Neuropsychiatric Pathology (NeuroPsy-Cog), "Victor Babes" University of Medicine and Pharmacy, Timisoara, ROMANIA
[5] Department of Neurosurgery, "Victor Babes" University of Medicine and Pharmacy, Timisoara, ROMANIA
[6] Faculty of Medicine and Pharmacy, University of Oradea, ROMANIA
[7] Department of Neurosurgery, Clinical Emergency Hospital Oradea, ROMANIA

Keywords
neurosurgery, Romanian Society of Neurosurgery, Constantin Arseni

Corresponding author:
Milena-Monica Ilie

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

ilie.milena@yahoo.com

Scan to access the online version

ABSTRACT

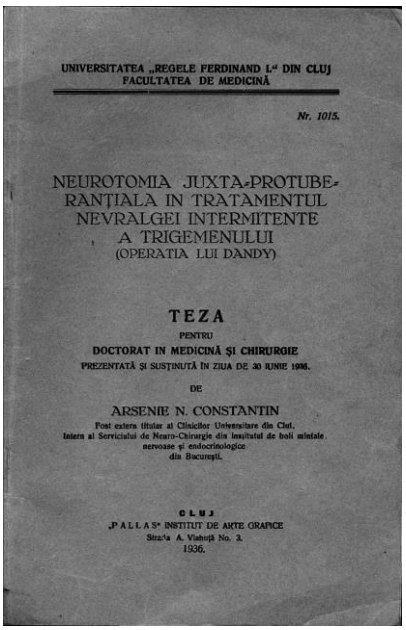
Professor Constantin Arseni (1912–1994) was a Romanian neurosurgeon whose pioneering contributions established the foundation for modern neurosurgery in Romania. Widely acknowledged as the founder of Romanian neurosurgery, his legacy reflects not only exceptional clinical expertise but also visionary leadership in the development of neurosurgical training programs, the establishment of scientific societies, and substantial contributions to international neurosurgical literature. Notably, in 1982, he founded the Romanian Society of Neurosurgery (RSN), a milestone that further cemented his influence in the field. His work and enduring legacy have profoundly shaped generations of neurosurgeons, both within Romania and internationally. Over the course of his illustrious career, Arseni authored an extensive body of scientific literature, spearheaded the establishment of one of Europe's most advanced neurosurgical departments, and played a pivotal role in advancing neurosurgical techniques and technologies.

EARLY LIFE AND EDUCATION

Constantin Arseni was born on February 3, 1912, in the small town of Dolhasca, situated in Suceava County, Romania [1]. Coming from a modest family background, Arseni exhibited an early passion for science and medicine, which guided his decision to pursue a medical education. He gained admission to the Faculty of Medicine in Cluj, one

of Romania's most esteemed medical schools, graduating in 1935 with high honors. During his academic journey, Arseni's interest in neurology and surgery began to crystallize, shaped by the remarkable medical advancements of the early 20th century. Influenced by groundbreaking developments in neuroscience and surgery both domestically and internationally, Arseni's academic curiosity and achievements propelled him toward specialized training in neurosurgery, a field still in its nascent stages at the time.

Upon earning his medical degree, Arseni concentrated his efforts on research in the nervous system and neurosurgery. His doctoral thesis, titled "Juxta-Protuberance Neurotomy in the Treatment of Intermittent Neuralgia of the Trigeminus," defended on June 30, 1936, represented one of the earliest significant Romanian studies on trigeminal neuralgia, a neurological disorder characterized by chronic facial pain (Photo 1) [2]. Captivated by the complexity of this condition, Arseni's research focused on novel surgical methods to interrupt nerve pathways associated with the disorder, laying the groundwork for advanced neurosurgical techniques that would later become standard practice. Recognized within the Romanian medical community, his thesis marked the inception of a distinguished and impactful career in neurosurgery.


UNIVERSITATEA „REGELE FERDINAND I" DIN CLUJ
FACULTATEA DE MEDICINĂ

Nr. 1015.

NEUROTOMIA JUXTA-PROTUBE-
RANȚIALA IN TRATAMENTUL
NEVRALGEI INTERMITENTE
A TRIGEMENULUI
(OPERAȚIA LUI DANDY)

TEZA
PENTRU
DOCTORAT IN MEDICINĂ ȘI CHIRURGIE
PREZENTATĂ ȘI SUSȚINUTĂ ÎN ZIUA DE 30 IUNIE 1936.
DE
ARSENIE N. CONSTANTIN
Fost extern titular al Clinicilor Universitare din Clul.
Intern al Serviciului de Neuro-Chirurgie din Institutul de boli mintale,
nervoase și endocrinologice
din București.

CLUJ
„PALLAS" INSTITUT DE ARTE GRAFICE
Strada A. Vlahuță No. 3.
1936.

Photo 1. "Juxta-Protuberance Neurotomy in the Treatment of Intermittent Neuralgia of the Trigeminus", on June 30, 1936.

Collaboration with Professor Dumitru Bagdasar

In the late 1930s, Constantin Arseni began working under the mentorship of Professor Dumitru Bagdasar, a distinguished Romanian neurosurgeon and one of the pioneers of modern neurosurgery in the country. Dumitru Bagdasar, a highly influential figure, had trained under Harvey Cushing, the American neurosurgeon universally recognized as the father of neurosurgery [1,3]. Cushing's profound influence on Dumitru Bagdasar, and subsequently on Arseni, was instrumental in shaping their approaches to the field. Recognizing Arseni's potential, Bagdasar invited him to join his team at the Central Hospital for Mental, Nervous, and Endocrinological Diseases in Bucharest. This collaboration led to the establishment of the first neurosurgical center in Bucharest under Bagdasar's leadership—a pivotal moment in Arseni's career (Photo 2).


Fațada clădirii în care a luat ființă în 1935 serviciul de neurochirurgie din București.

Photo 2. First Independent Neurosurgical Department in Bucharest – Central Hospital of Nervous Diseases, in 1935

During their partnership, Constantin Arseni and Dumitru Bagdasar collaborated on numerous groundbreaking projects addressing complex neurological disorders, including brain tumors, spinal injuries, and nerve lesions [1,3]. Together, they refined surgical techniques and pioneered new methods for managing these challenging conditions. Among their most significant contributions was the development of a novel surgical technique for treating craniostenosis, a congenital defect in which the bones of a baby's skull fuse prematurely. Alongside their colleague, Dr. State Drăgănescu, they introduced this groundbreaking approach, significantly improving patient outcomes and advancing the management of this condition [4].

One of their landmark achievements was the co-authorship of the first neurosurgical treatise in Romania and Southeastern Europe, published in French in 1948. This work represented a major step forward in the development of neurosurgery in Romania, solidifying their reputations as leaders in the field and advancing the discipline both nationally and regionally (Photo 3).

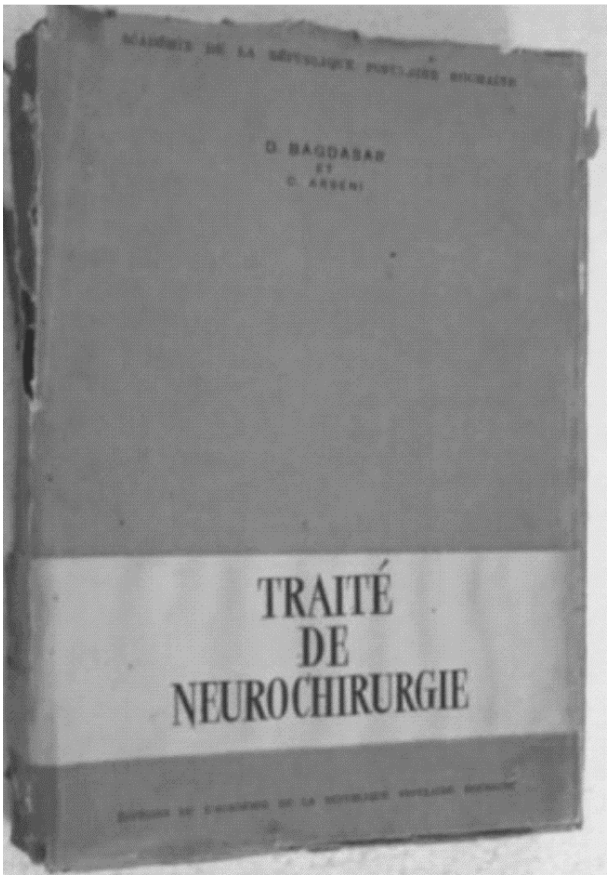


Photo 3. First "Traite De Neurochirurgie" (1948)

The Creation of the Neurosurgical Clinic in Bucharest

In 1946, following the death of Professor Bagdasar, Constantin Arseni took over the leadership of the Neurosurgical Service at the Central Hospital. At just 34 years old, he became the Head of Neurosurgery, a position that would allow him to shape the future of the field in Romania. Under his leadership, the neurosurgical service grew significantly. By 1964, the clinic had expanded to 220 beds, with specialized sections for brain tumors, spinal surgery, pediatric neurosurgery, craniocerebral and vertebral trauma, and vascular neurosurgery. Arseni's vision for the clinic was to create a comprehensive center for neurosurgery that could handle all types of neurological and neurosurgical cases, from the most common to the most complex (Photo 4).

One of Constantin Arseni's most significant contributions to Romanian neurosurgery was the introduction of advanced medical technologies. In 1982, he was instrumental in acquiring Romania's first CT scan device, which revolutionized diagnostic capabilities in the country. The CT scanner allowed for more accurate imaging of the brain and spinal cord, making it easier to diagnose and plan surgical interventions for conditions like tumors, trauma, and vascular malformations [7]. This technological leap significantly improved patient outcomes and placed Romania at the forefront of neurosurgical technology in Eastern Europe (Photo 5) [5].

Photo 4. Neurosurgical Clinic-1964

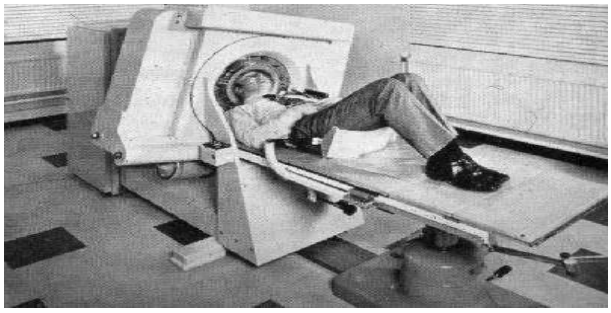


Photo 5. The first CT scan device in Romania-1982

Expansion of the Neurosurgical Clinic

Throughout the 1960s and 1970s, Constantin Arseni continued to expand the clinic, both in terms of physical capacity and the range of services offered. By 1975, the clinic had grown to 550 beds, making it one of the largest neurosurgical centers in Europe (Photo 6) [1].

Photo 6. The Neurosurgical Clinic-1975

The clinic was divided into seven departments, each specializing in a different aspect of neurosurgical care. This included a pediatric neurosurgery department, which was one of Arseni's personal passions. He believed that children with neurological conditions required specialized care, and he worked to develop new surgical techniques for treating congenital malformations, hydrocephalus, and brain tumors in pediatric patients.

Pediatric neurosurgery was one of the areas where Arseni made his most significant contributions. Together with Dr. Lenke Horvarth and his long-time collaborator Professor Alexandru Vlad Ciurea, Consantin Arseni helped to establish pediatric neurosurgery as a distinct subspecialty within Romania (Photo 7). His work in this field included the development of new surgical techniques for treating cranial deformities, congenital brain malformations, and tumors in children. Arseni was particularly interested in improving the outcomes for children with hydrocephalus, a condition in which excess fluid builds up in the brain [6,7,8,9].

Photo 7. The golden team of pediatric neurosurgery (Professor Consantin Arseni, Dr. Lenke Horvarth and Professor Alexandru Vlad Ciurea)

This prodigious activity of Prof. Constantin Arseni elevated the neurosurgery clinic, especially in its new building, to European and international recognition. Prof. Constantin Arseni developed all branches of neurosurgery, focusing particularly on intracranial tumors, for which he had a clinical, operative, and research inclination. Moreover, all cases of brain tumors operated on in the clinic were evaluated both clinically and anatomopathologically by Constantin Arseni and the head of the anatomopathology laboratory, Dr. Carp [10]. During this period of neurosurgical development, Prof. Constantin Arseni sent his collaborators abroad for experience exchanges (to France, Germany, Italy, and the USA). Thus, the future Prof. Dr. Leon Dănăilă was granted a nine-month assignment in New York at Bienville Hospital, which significantly contributed to his preparation in tumor and vascular microsurgery.

Unfortunately, the events of 1989 were entirely unfavorable for Prof. Constantin Arseni, as he was officially reproached for his collaboration with the state leadership. It should be noted that he was not a party member, and this collaboration was solely scientific in nature. Considering the slightly dark period following the regime change in 1989–1990, the personality of Prof. Constantin Arseni fell into obscurity, especially as his health gradually deteriorated, with several minor strokes and diabetic retinopathy, which halted his participation in neurosurgical interventions.

He passed away in 1994 due to a stroke, leaving behind an unforgettable legacy [1].

Scientific Contributions

Professor Constantin Arseni's scientific contributions were vast and influential. Over the course of his career, he authored or co-authored 54 monographs, treatises, and books, as well as numerous scientific papers published in Romanian and international medical journals [1]. His writings covered a wide range of topics within neurosurgery, from brain tumors to spinal cord injuries, and from vascular diseases of the brain to neuroendocrinology.

Among his most important publications are the following works:

1. Vertebral Sciatica (1948) – This was one of the first comprehensive studies on the treatment of sciatic pain caused by vertebral issues [11].
2. Vascular Diseases of the Brain and Spinal Cord (1965) – A major contribution to the understanding of vascular pathology in the central nervous system [12].
3. Pain (1967) – A groundbreaking study on the mechanisms and treatment of chronic pain in neurosurgical patients [13].
4. Neuro-Ophthalmologic Diagnosis (1967) – This book explored the neurological causes of vision problems and their treatment [14].
5. Spinal Neurological Pathology (1968) – A comprehensive study of spinal cord diseases and injuries [15].
6. Raised Intracranial Pressure (1972) – This work focused on the diagnosis and management of elevated intracranial pressure, a common problem in neurosurgery [16].

7. Craniocerebral Traumatology (1972) – A major work on the treatment of head injuries [17].
8. Intracranial Space-Occupying Processes (1973) – This two-volume work was a detailed study of brain tumors and other mass lesions in the brain [18,19].
9. Spinal Cord and Peripheral Nerve Injuries (1974) – A study of the treatment of traumatic injuries to the spinal cord and peripheral nerves [20].
10. Neurosurgical Semiology (1977) – This book focused on the clinical examination and diagnosis of neurosurgical conditions [21].
11. Pathologic Anatomy of Tumors of the Nervous System (1978) – A major work on the pathological aspects of brain and spinal tumors [10].
12. Cervical Vertebromedullary Pathology (1982) – This book explored the diagnosis and treatment of conditions affecting the cervical spine and spinal cord [22].
13. Neuroendocrinology (1988) – A comprehensive study of the relationship between the nervous system and the endocrine system, particularly in the context of tumors affecting the hypothalamus and pituitary gland [23].
14. Neurologic Semiology (1988) – This work focused on the clinical signs and symptoms of neurological diseases [24].

Notably, Professor Constantin Arseni's contribution to the development of the "Treatise of Neurology" between 1979 and 1982 stands out as a cornerstone of his academic achievements (Photo 8).

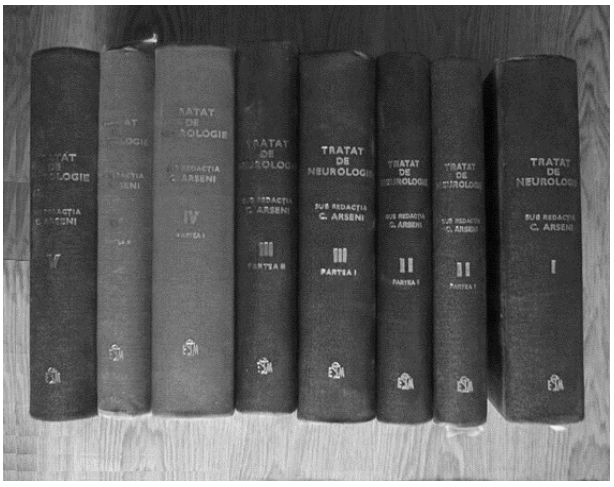


Photo 8. The "Treatise of Neurology" in 5 volumes, during the 1979-1982 period.

Through his exceptionally prolific scientific activity and membership on the editorial boards of numerous international journals, including ACTA NeuroChirurgica, Romanian neurosurgery gained significant international recognition. Prof. Arseni's relentless dedication as a scientific researcher has left an enduring impact on the field [25].

The scientific library curated by Professor Constantin Arseni exemplifies his devotion and multidisciplinary approach across all domains of the neurosciences (Photo 9).

Photo 9. Library created by Professor Constantin Arseni

Of particular importance was his focus on the pathology of brain tumors, evident in his extensive publications and the brain tumor classification he developed in collaboration with Dr. Carp in 1978 [10]. This classification, translated and internationally recognized, remains a benchmark in the field (Photo 10).

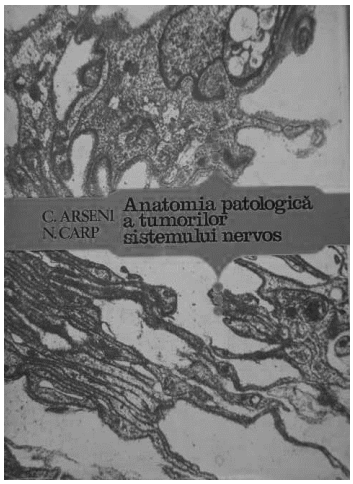


Photo 10. Arseni, C., & Carp, N. "Pathologic Anatomy of Tumors of the Nervous System". Romanian Academy Publishing House, 1978, Bucharest.

Furthermore, the "Museum of Encephalons", showcasing extremely complex pathological cases, stands as a testament to his scientific dedication to addressing intricate lesions of the nervous system (Photo 11).

Photo 11. Anatomo-pathological Museum created by Professor Constantin Arseni.

Establishment of the Romanian Society of Neurosurgery

In 1982, Arseni founded the Romanian Society of Neurosurgery, an organization that aimed to promote neurosurgical education, research, and clinical practice in Romania. The society was instrumental in connecting Romanian neurosurgeons with their international counterparts, fostering collaboration and knowledge exchange [26]. Arseni's leadership in the society helped to elevate the status of Romanian neurosurgery on the global stage. The first congress of the Romanian Society of Neurosurgery was held in Oradea Felix, and it attracted neurosurgeons from across Romania and Europe. Under Arseni's guidance, the society became a key platform for sharing new research, discussing advances in surgical techniques, and training the next generation of neurosurgeons [27].

Teaching and Training Future Generations

One of Constantin Arseni's greatest legacies was his role in training future generations of neurosurgeons. He believed strongly in the importance of education and mentorship, and he dedicated much of his career to teaching. As a professor of neurosurgery at "Carol Davila" University of Medicine and Pharmacy in Bucharest, Constantin Arseni trained hundreds of neurosurgeons who would go on to practice throughout Romania and beyond. His teaching was characterized by a rigorous approach to both theory and practice, and he emphasized the need for neurosurgeons to be well-versed in the latest research as well as highly skilled in the operating room.

Arseni played a key role in developing the neurosurgical training curriculum in Romania. His approach to training was systematic and thorough, and he established a curriculum that included multiple years of training in different subspecialties within neurosurgery. This curriculum included one year of training in neuro-traumatology, one year in brain tumors, one year in vascular neurosurgery, and six months in pediatric and functional neurosurgery. By creating a structured and comprehensive training program, Arseni ensured that Romanian neurosurgeons were among the best-trained in the world [1,27].

The entire neurosurgical community in Romania pays tribute to Professor Constantin Arseni, who established numerous departments and neurosurgical centers across the country, including in Ploiești, Brașov, Craiova, Sibiu, Galați, and Constanța.

The leaders of these departments, all senior physicians, received their training under Professor Constantin Arseni's mentorship, honing their expertise in his clinic before advancing to leading roles in various academic and medical centers throughout Romania.

International Recognition and Legacy

Professor Arseni's contributions to neurosurgery were recognized not only in Romania but also internationally. His work earned him numerous awards and honors, including the Gheorghe Marinescu Award from the Romanian Academy for his contributions to neurosurgical research. In 1991, he was inducted as *A Full Member of The Romanian Academy***,** a testament to his lifelong dedication to advancing medical science. Arseni's work also opened the door for Romanian neurosurgeons to participate in international collaborations, and his influence extended well beyond the borders of Romania.

Arseni's influence on neurosurgery extended beyond his home country, and he became an internationally respected figure in the medical community. His work in introducing new surgical techniques and technologies, such as the CT scan in Romania, helped to bring Romanian neurosurgery in line with international standards and earned him a reputation as a forward-thinking and innovative surgeon.

Leadership and International Influence

Professor Constantin Arseni's influence extended far

beyond the walls of the neurosurgical clinic in Bucharest. Throughout his career, Arseni played an active role in the broader neurosurgical community, both in Romania and internationally. As a prominent figure in Romanian medicine, he helped to shape the direction of the country's medical education system, ensuring that future generations of neurosurgeons were well-trained and equipped with the latest knowledge and skills [28]

In addition to his contributions to medical education, Arseni was instrumental in establishing connections between Romanian neurosurgeons and their counterparts in other countries. His efforts to bridge the gap between Romanian neurosurgery and international practices helped to raise the standard of care in Romania and to integrate the country into the global neurosurgical community [29].

Conclusion

A Visionary Who Shaped the Future of Romanian Neurosurgery

Professor Constantin Arseni was not only a skilled neurosurgeon but also a visionary who helped to shape the future of Romanian medicine. His contributions to clinical practice, medical education, and scientific research have left an indelible mark on the field of neurosurgery, both in Romania and globally. As the founder of modern Romanian neurosurgery, Arseni played a central role in bringing advanced neurosurgical care to the country and ensuring that Romania was on the cutting edge of medical technology and research. His leadership, vision, and dedication to his patients and students have made him a lasting figure in the history of neurosurgery.

Constantin Arseni's work has not only improved the lives of countless patients, but it has also inspired generations of neurosurgeons to strive for excellence in their own practice. His legacy lives on in the institutions he helped build, the research he conducted, and the many neurosurgeons he trained. As we look back on his remarkable career, it is clear that Constantin Arseni's contributions to neurosurgery will continue to shape the field for years to come.

Due to his exceptional contributions across scientific, organizational, and administrative domains, Romanian neurosurgery honors his legacy appropriately by naming the current neurosurgical hospital after its founder: Emergency Hospital Bagdasar-Arseni.

References

1. Mohan, D., A. G. Mohan, M. V. Saceleanu, D. Bentia, A. V. Ciurea, 110 Years Since the Birth of Constantin Arseni – The Founder of Romanian Neurosurgery. The Medical-Surgical Journal, 2023, 127(1), 149–156. https://doi.org/10.22551/msj.2023.01.15
2. Arseni, C. Juxta-Protuberance Neurotomy in the Treatment of Intermittent Neuralgia of the Trigeminus. Romanian Journal of Medical Sciences, 1970, 12(4), 299–315.
3. Arseni, C., Ciurea, A. V. The History of Neurosurgery. Medical Publishing House, 1981, Bucharest.
4. Arseni, C., Drăgănescu, S. Craniostenosis: A New Surgical Method of Treatment. Romanian Academy Publishing House, 1969, Bucharest.
5. Ciurea, A. V., Arseni, C. The Role of CT Scanning in the Diagnosis of Intracranial Pathology in Romania. Neurosurgical Innovations Journal, 1989, 15(4), 185–205.
6. Arseni, C., Horvath, L. Cranial Deformities and Congenital Brain Malformations in Children. Romanian Academy Publishing House, 1975, Bucharest.
7. Ciurea, A. V., Arseni, C. Hydrocephalus in Children: Surgical Solutions and Long-Term Outcomes. Pediatric Neurosurgery Journal, 1985, 9(3), 100–120.
8. Arseni, C., Horvath, L., Ciurea, A. V. Neurosurgical Disorders in Infants and Young Children (0-3 years). Medical Publishing House, 1979, Bucharest.
9. Arseni, C., Horvath, L., & Ciurea, A. V. Pediatric Neurosurgical Pathology. Romanian Academy Publishing House, 1980, Bucharest.
10. Arseni, C., Carp, N. Pathologic Anatomy of Tumors of the Nervous System. Romanian Academy Publishing House, 1978, Bucharest.
11. Arseni, C., Iacob, T. Vertebral Sciatica. H. Welther Publishing House, 1948, Sibiu.
12. Arseni, C., Petrovici, I. N., Nass, F. Vascular Diseases of the Brain and Spinal Cord. Romanian Publishing House, 1965, Bucharest.
13. Arseni, C., Oprescu, I. Pain. Romanian Academy Publishing House, 1967, Bucharest.
14. Arseni, C. Neuro-Ophthalmologic Diagnosis. Romanian Academy Publishing House, 1967, Bucharest.
15. Arseni, C., Simionescu, M. Spinal Neurological Pathology. Romanian Academy Publishing House, 1968, Bucharest.
16. Arseni, C., Constantinescu, A. I. Raised Intracranial Pressure. Romanian Academy Publishing House, 1972, Bucharest.
17. Arseni, C., Oprescu, I. Craniocerebral Traumatology. Romanian Academy Publishing House, 1972, Bucharest.
18. Arseni, C., Constantinescu, A. I., Maretsis, M., Stanciu, M., & Voinescu, I. Intracranial Space-Occupying Processes, Volume 1. Romanian Academy Publishing House, 1973, Bucharest.
19. Arseni, C., Maretsis, M., Constantinescu, A. I.,

Constantinovici, A., & Ghitescu, M. Intracranial Space-Occupying Processes, Volume 2. Romanian Academy Publishing House, 1974, Bucharest.
20. Arseni, C., Constantinovici, A., Panoza, G. Spinal Cord and Peripheral Nerve Injuries. Romanian Academy Publishing House, 1974, Bucharest
21. Arseni, C., Constantinescu, A. I., Maretsis, M. Neurosurgical Semiology. Romanian Academy Publishing House, 1977, Bucharest.
22. Arseni, C., Panoza, G. Cervical Vertebromedullary Pathology. Romanian Academy Publishing House, 1981, Bucharest.
23. Arseni, C., Maretsis, M. Neuroendocrinology. Romanian Academy Publishing House, 1988, Bucharest.
24. Arseni, C., Popoviciu, L. Neurologic Semiology. Romanian Academy Publishing House, 1981, Bucharest.
25. Dinca, E. B., Banu, M., Ciurea, A. V. Constantin N. Arseni (1912-1994) Centenary: The Birth of Modern Neurosurgery in Romania. World Neurosurgery, 2014, 82(1-2), e371-373. https://doi.org/10.1016/j.wneu.2013.06.007
26. Mohan, D., Husti, I. L., Moisa, H., Ciurea, A., & Mult, H. C. The Legacy of Prof. Constantin Arseni: The Medical Architect Behind the Romanian School of Neurosurgery. Chirurgia (Bucur), 2014, 109(5), 705-708.
27. Mohan, A. G., Săceleanu, M. V., Marinescu, A. A., Popescu, M., & Ciurea, A. V. Neurosurgery in Romania in the Centenary of the Great Union (1918-2018). Romanian Journal of Morphology and Embryology, 2018, 59(4), 1299-1303.
28. Umansky, F., Constantin N. Arseni: A Leader of Romanian Neurosurgery. World Neurosurgery, 2014, 82(1-2), e153-154. https://doi.org/10.1016/j.wneu.2013.07.125.
29. Tomasello, F., Angileri, F. F. A Lesson from a Visionary Pioneer of Romanian Neurosurgery: Prof. Constantin Arseni. World Neurosurgery, 2014, 82(1-2), e151-152. https://doi.org/10.1016/j.wneu.2013.08.050

Romanian Neurosurgery (2025) XXXIX (1): pp. 22-27
DOI: 10.33962/roneuro-2025-003
www.journals.lapub.co.uk/index.php/roneurosurgery

Gemistocytic astrocytoma mimicking hypertensive haemorrhage. A rare case of tumour disguised as intracerebral haemorrhage

Alexandru Breazu[1], Cosmin Cindea[2], Sonia Lucaciu[1], Antonia Iliescu[2], Sabina Sasu[2], Vicentiu Saceleanu[2]

[1] County Clinical Emergency Hospital of Sibiu, ROMANIA
[2] "Lucian Blaga" University of Sibiu, ROMANIA

Keywords
craniotomy, gemistocytic astrocytoma, intracerebral haemorrhage, tumour mimicking stroke, brain gliomas, ICH

ABSTRACT

Gemistocytic astrocytoma is a rare variant of diffuse astrocytoma, characterized by a high proportion of gemistocytic cells, which exhibit aggressive behaviour and an increased risk of malignant transformation. Its clinical and radiological presentation can be misleading, especially when it mimics other intracerebral pathologies, such as hypertensive intracerebral haemorrhage (ICH). Differentiating between a primary haemorrhagic event and a haemorrhagic tumour remains a significant diagnostic challenge.

We report the case of a 56-year-old male with no prior medical history who presented with sudden-onset right-sided hemiparesis and severe speech disturbances. Initial neuroimaging revealed a deep intraparenchymal hematoma in the left internal capsule, and lenticular nucleus, strongly suggestive of a hypertensive haemorrhagic stroke. Despite intensive medical management, the patient's condition deteriorated, prompting further imaging studies, which raised suspicion of an underlying neoplastic process. Subsequent MRI findings indicated features atypical for a purely haemorrhagic lesion, necessitating neurosurgical intervention for definitive diagnosis.

The patient underwent a left fronto-temporo-parietal craniotomy, during which a tumour-like mass was encountered and completely resected. Histopathological analysis confirmed the diagnosis of gemistocytic astrocytoma. Postoperatively, the patient showed gradual neurological improvement, though residual deficits persisted.

This case highlights the complexity of differentiating a gemistocytic astrocytoma from a spontaneous hypertensive haemorrhage, particularly in patients without a prior oncological history. While intracerebral haemorrhage is commonly associated with chronic hypertension, intratumoural haemorrhage remains an important differential diagnosis, especially when imaging findings suggest a mass effect, perilesional oedema, or progressive neurological deterioration despite optimal medical therapy. MRI characteristics, such as hyperintense T2-weighted and FLAIR signals, can provide critical clues, but histopathological confirmation remains the gold standard.

Gemistocytic astrocytomas, though rare, should be considered in cases of unexplained intracerebral haemorrhage, particularly when imaging findings or clinical progression are atypical. This case underscores the importance of a

Corresponding author:
Cosmin-Nicodim Cindea

"Lucian Blaga" University of Sibiu, Romania

cindea.cos@gmail.com

Scan to access the online version

multidisciplinary approach involving neurology, neuroradiology, and neurosurgery to ensure timely diagnosis and appropriate management. A high index of suspicion is crucial to prevent delays in the recognition and treatment of haemorrhagic brain tumours, which can significantly impact patient outcomes.

Introduction

Astrocytomas are common primary brain tumors originating from astrocytes, glial cells that maintain neuronal function and brain homeostasis. Gemistocytic astrocytoma, a rare subtype of diffuse astrocytoma (WHO grade II), is distinguished by a high proportion of gemistocytic cells-large, eosinophilic astrocytes with eccentric nuclei [1]. These tumors typically arise in adults in their fourth and fifth decades, predominantly in the cerebral hemispheres, and exhibit a higher tendency for malignant transformation into glioblastoma, necessitating early detection and intervention [2].

While gemistocytic astrocytomas usually present with progressive neurological symptoms such as headaches, seizures, or focal deficits, they can occasionally mimic hypertensive intracerebral hemorrhage (ICH), creating diagnostic challenges. Although hypertensive ICH is the leading cause of spontaneous brain hemorrhage, affecting deep structures like the thalamus and basal ganglia, hemorrhagic tumors account for only 2-5% of spontaneous ICH cases. Tumor-related hemorrhage may result from fragile neovascularization, necrosis, or vascular invasion, leading to an acute presentation [3].

Imaging plays a crucial role in differentiating these conditions. While CT scans may reveal hyperdense lesions, MRI-particularly contrast-enhanced sequences-can indicate irregular enhancement, perilesional edema, and mass effect, raising suspicion of an underlying tumor. However, initial imaging may fail to detect tumors; in a study of 193 patients with primary malignant brain tumors, 9 had normal initial MRI scans, while 8 had abnormalities misinterpreted [4]. In gemistocytic astrocytomas, hyperintense signals on T1-weighted MRI, especially in cases with necrosis or infiltration, further complicate differentiation from ICH [5]. Follow-up imaging or histopathological confirmation remains the gold standard for diagnosis.

At the molecular level, gemistocytic astrocytomas frequently harbor TP53 mutations and IDH1/IDH2 pathway alterations, contributing to their aggressive behavior and poor prognosis. Despite their WHO grade II classification, they carry a significant risk of progression to glioblastoma, underscoring the importance of early diagnosis [6].

Delays in identifying tumor-related hemorrhages can impact treatment strategies and outcomes. While hypertensive hemorrhages are managed conservatively, hemorrhagic tumors often require surgical intervention, biopsy, or oncological treatment. Misdiagnosis may lead to delayed tumor detection, allowing disease progression. Even when imaging suggests hypertensive ICH, a neoplastic etiology should be considered, particularly if the clinical course is atypical.

We present the case of a 56-year-old male initially diagnosed with hypertensive ICH, whose progressive neurological decline and atypical imaging findings ultimately led to the diagnosis of gemistocytic astrocytoma. This case highlights the need to consider tumor-related hemorrhage in select spontaneous ICH cases and emphasizes the role of advanced neuroimaging and a multidisciplinary approach in distinguishing hemorrhagic strokes from brain tumors to ensure timely and accurate management.

Case Presentation

A 56-year-old male with no prior medical history experienced a sudden onset of right-sided hemiparesis and severe speech disturbances. His spouse reported a rapid neurological decline, prompting immediate transportation to the Emergency Department (ED). Upon arrival, the patient was somnolent and minimally cooperative, with a Glasgow Coma Scale (GCS) score of 7 and a National Institutes of Health Stroke Scale (NIHSS) score of 15, indicating a major neurological deficit.

The neurological examination revealed complete flaccid hemiplegia on the right, upper motor neuron (central) facial paresis on the right, right homonymous hemianopia, and profound mixed aphasia affecting both receptive and expressive language functions. Given the acute presentation and severe deficits, an intracerebral hemorrhage was highly suspected.

Consequently, an emergent non-contrast cranial CT scan was performed immediately upon admission. The imaging revealed a large deep intraparenchymal hematoma in the left internal

capsule and lenticular nucleus, measuring approximately 30-40 mL. The hematoma was accompanied by mild perilesional edema and moderate mass effect, although no signs of herniation were observed at that time. These findings strongly suggested a hypertensive intracerebral hemorrhage (ICH), particularly in the context of the patient's elevated blood pressure at presentation (Figure *1*).

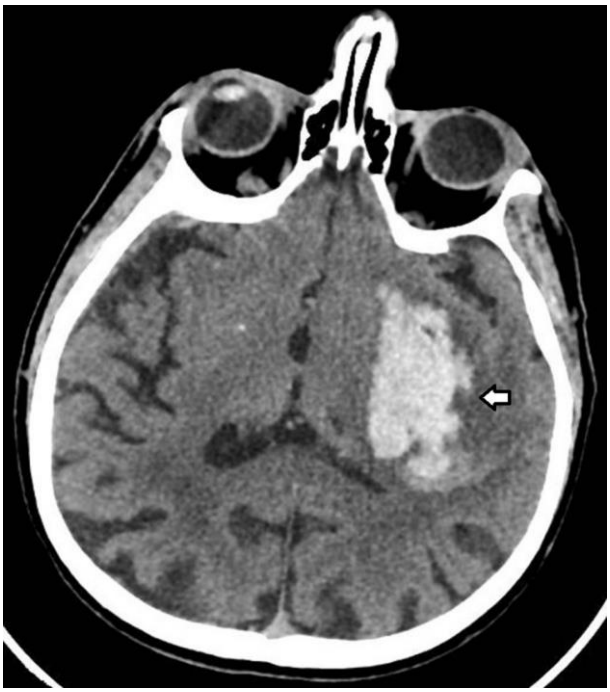


Figure 1. Initial cranial CT scan showing a deep intraparenchymal hematoma in the left internal capsule and lenticular nucleus

During the hospital course, while under conservative neurological management, the patient exhibited a slight deterioration in his neurological status. A repeat cranial CT scan was performed (Figure 2), which revealed an expansion of the intracerebral hemorrhage, accompanied by perilesional edema and a slightly mass effect on the midline structures.

Conservative management was continued over the following week as the patient's clinical status remained stable. A third cranial CT scan showed a cerebral hematoma in the process of resolving, without evidence of recent bleeding, but with significant cerebral edema and a midline shift (Figure 3).

This unfavorable imagistic evolution raised the possibility of surgical intervention. Consequently, an intravenous contrast-enhanced cranial MRI was performed for further evaluation (Figure 4).

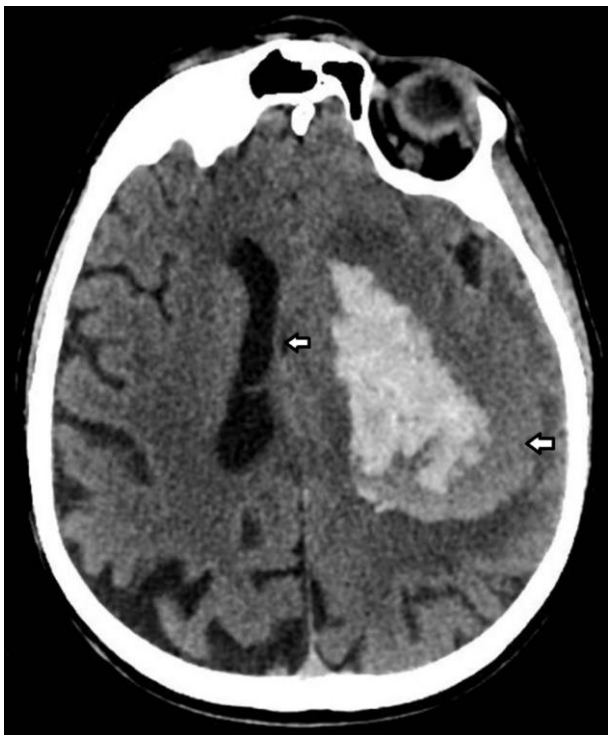


Figure 2. Follow-up CT scan showing an increase in hematoma size, extension into the left frontal and temporal lobes, and mass effect on the left lateral ventricle. Arrows indicating the expanding hematoma and accompanying midline shift.

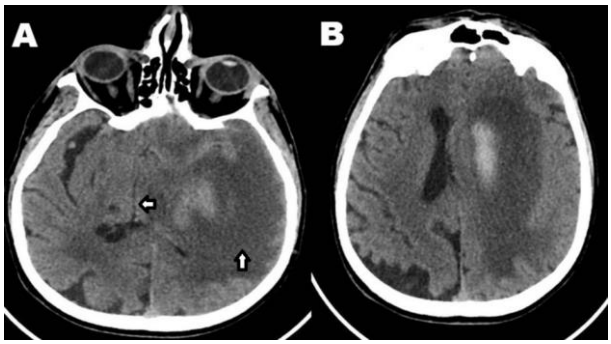


Figure 3. Follow-up CT scan showing a decrease in hemorrhagic density with extended perilesional edema, presenting a peritumoral-like appearance. (A) - large mass effect, (B) - compression of the left lateral ventricle.

Due to the worsening mass effect and persistent neurological deficits, the patient was transferred to the Neurosurgery Department for surgical evaluation. A left fronto-temporo-parietal craniotomy was performed under general anesthesia. Intraoperatively, a tumor-like mass with

hemorrhagic components was identified and completely resected along with hematoma evacuation. The tumor appeared well-vascularized and infiltrative, which raised concerns for a primary brain neoplasm rather than a simple hematoma.

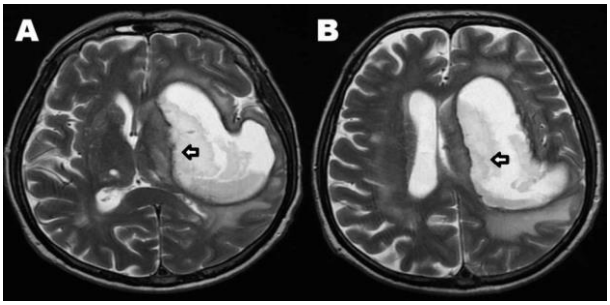


Figure 4. MRI scan showing a large hematoma with hyperintense signals on T2-weighted and FLAIR sequences, raising suspicion of a tumor. (A) sylvian valley - insular level; (B) left lateral ventricle compression

Following surgery, the patient was transferred to the Neurosurgery ICU for close monitoring. A postoperative CT scan showed a porencephalic cavity in the left temporo-insular region, with minimal adjacent hematoma and moderate perilesional edema, but no signs of progressive bleeding or infection (Figure 5). The resolution of mass effect was evident, confirming the successful removal of the underlying pathology.

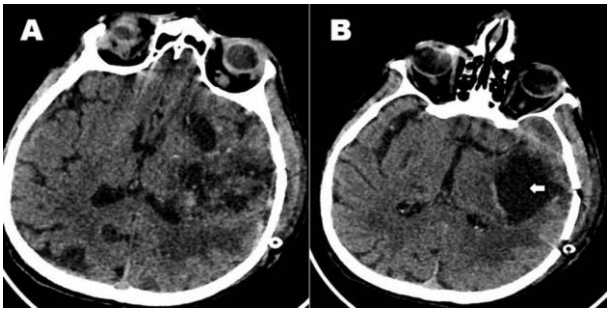


Figure 5. Postoperative CT scan showing porencephalic cavity and resolution of mass effect. (A) lateral ventricle level; (B) 3rd ventricle level

Over the next few days, the patient's condition gradually improved. He regained partial consciousness and became more cooperative, though severe mixed aphasia and right-sided hemiplegia persisted. Rehabilitation, including speech therapy and physical therapy, was initiated to aid neurological recovery.

At discharge, the patient was conscious and cooperative, with persistent right-sided hemiplegia and motor aphasia, but early signs of motor recovery were observed. He was referred for intensive neurological rehabilitation, with close outpatient follow-up scheduled to monitor his progress and assess for potential tumor recurrence.

Histopathological examination (Figure 6) of the excised tissue confirmed gemistocytic astrocytoma (WHO Grade II), characterized by abundant gemistocytic cells with eosinophilic cytoplasm and eccentric nuclei. The presence of tumor-associated hemorrhage explained the misleading presentation and radiological findings.

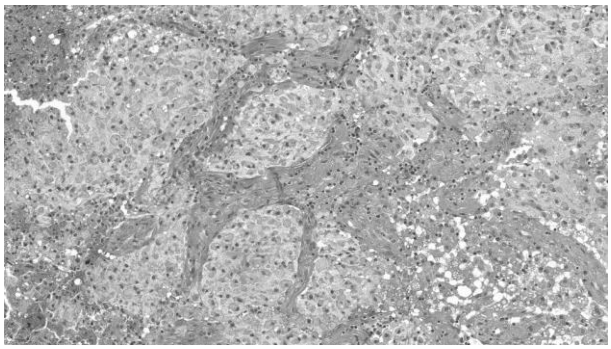


Figure 6. Histopathology 35.1x - Gemistocytic astrocytoma

This case highlights the diagnostic challenge posed by gemistocytic astrocytomas when they present as intracerebral hemorrhage, leading to an initial misdiagnosis of hypertensive stroke. The patient's progressive neurological deterioration and MRI findings raised suspicion of a neoplasm, ultimately confirmed through histopathological analysis after neurosurgical intervention. Recognizing such cases is crucial to avoid delays in diagnosis and ensure timely, appropriate management.

Discussion

The case of a gemistocytic astrocytoma presenting as a spontaneous hypertensive intracerebral hemorrhage (ICH) highlights the complexity of diagnosing brain tumors with atypical hemorrhagic presentations. While hypertensive hemorrhages typically occur in deep brain structures such as the thalamus, basal ganglia, and brainstem, hemorrhagic brain tumors are rare and can be misinterpreted as primary vascular events. Distinguishing between a hypertensive ICH and a

hemorrhagic tumor is challenging due to overlapping clinical and radiological features [7].

Tumor-associated hemorrhages account for 2-5% of spontaneous ICH cases, with gliomas, metastases, and vascular malformations being the most common causes. High-grade gliomas, particularly glioblastomas, have a higher propensity for bleeding due to fragile neovascularization and tumor necrosis. Low-grade gliomas, such as gemistocytic astrocytomas, rarely present with hemorrhage, making this case unusual. This suggests that hemorrhagic risk is influenced not only by tumor histology but also by factors such as location, vascular infiltration, and tumor biology [8].

The initial diagnosis of hypertensive hemorrhage was plausible given the capsulo-thalamic localization and history of hypertension. However, the patient's unfavorable clinical progression under conservative treatment and MRI findings raised suspicion of a tumor. Intracerebral hematomas and brain tumors share common imaging characteristics, including perilesional edema, mass effect, and secondary structural compression. Initial imaging can be inconclusive, as studies have shown that in 193 patients with primary malignant brain tumors, 9 had normal initial MRIs, and 8 had abnormalities that were not initially diagnosed [9]. This suggests that some tumors may grow rapidly and escape early detection. In this case, it cannot be ruled out that the hemorrhage was initially hypertensive and that the tumor developed later in the same location.

Neuroimaging is crucial for differentiation, but CT scans, while effective for detecting hematomas, may not reveal underlying neoplasms. MRI, particularly T2-weighted and FLAIR sequences, can provide critical clues such as heterogeneous signal intensities, irregular borders, and residual enhancement, suggestive of an underlying tumor. Perfusion-weighted imaging (PWI) and MR spectroscopy can aid differentiation, though they are not always available in emergency settings. Intraoperative findings confirmed the presence of a tumor, and histopathological analysis established the diagnosis of gemistocytic astrocytoma (WHO Grade II). Given the high risk of malignant progression, reported in 60-80% of cases within 2-5 years, early diagnosis and intervention are essential [9].

The role of surgery in hemorrhagic gemistocytic astrocytomas is debated. While gross total resection (GTR) is associated with better survival outcomes, complete excision is often challenging due to tumor infiltration. In cases where total resection is not feasible, adjuvant radiotherapy and temozolomide-based chemotherapy are standard treatment options. In this case, craniotomy and total tumor resection were justified due to mass effect, worsening neurological symptoms, and imaging findings suggestive of neoplasm [10].

Misdiagnosing a tumor-related hemorrhage as a primary stroke can lead to delays in appropriate treatment. Patients with suspected hypertensive ICH typically receive blood pressure management and supportive care, with hematoma evacuation in select cases. However, in cases of underlying tumors, delayed recognition may allow disease progression and worsen prognosis. A retrospective study by Nozaki et al. demonstrated that misdiagnosis of tumor-associated hemorrhages as strokes correlated with poorer survival outcomes [11]. Given that glioma-related hemorrhages occur in 3.7% to 12% of cases, even when imaging suggests a hypertensive ICH, an underlying tumor should remain a differential diagnosis. In cases of atypical evolution or poor response to conservative therapy, histological sampling during hematoma evacuation and post-operative MRI are critical for detecting hidden neoplasms.

Anticoagulation can exacerbate tumor-associated hemorrhages, as fragile tumor vasculature increases bleeding risk. The role of anticoagulation reversal in ICH raises concerns about undiagnosed gliomas presenting with early hemorrhage, potentially delaying tumor detection [12]. Further research should explore whether coagulation status influences glioma-associated hemorrhage and clinical progression.

Gliomas, including gemistocytic astrocytomas, have high malignant potential, requiring long-term imaging follow-up. Even low-grade tumors can progress to glioblastoma, emphasizing the need for MRI surveillance every 3-6 months [13, 15]. Recognizing hemorrhage as an early sign of tumor instability could refine treatment and monitoring strategies.

Long-term management includes close surveillance with MRI every 3-6 months due to the high risk of recurrence and malignant transformation. Neurorehabilitation, including speech and physical therapy, is essential for

functional recovery. Despite persistent aphasia and hemiplegia, early signs of motor improvement were observed, reinforcing the benefits of intensive rehabilitation [11, 14].

This case underscores the importance of a multidisciplinary approach involving neurologists, neurosurgeons, neuroradiologists, and pathologists to ensure accurate diagnosis and optimal treatment. While spontaneous ICH is often attributed to vascular causes, an underlying tumor should be considered, particularly when neurological deterioration is progressive, mass effect persists, or imaging features are atypical. Vigilance in cases of unexplained hemorrhage can facilitate timely intervention and improve patient outcomes.

Conclusions

This case underscores the diagnostic challenges of differentiating hemorrhagic tumors from primary intracerebral hemorrhage (ICH), particularly when initial imaging suggests a vascular origin. While spontaneous ICH is frequently attributed to hypertensive causes, this case highlights the need for a high index of suspicion in atypical presentations. The stepwise diagnostic approach, including MRI evaluation, ultimately led to the identification of an underlying neoplasm, which was confirmed intraoperatively and histopathologically.

Our findings reinforce the importance of multimodal imaging and clinical vigilance in cases where neurological deterioration persists despite conservative management. Early differentiation between tumor-associated hemorrhage and primary ICH is crucial, as treatment strategies differ significantly. A multidisciplinary approach integrating neurology, neurosurgery, and neuroradiology is essential to ensuring timely intervention and optimizing patient outcomes.

References

1. Avninder S, Sharma MC, Deb P: Gemistocytic astrocytomas: histomorphology, proliferative potential and genetic alterations--a study of 32 cases. J Neurooncol. 2006, 78:123-7. 10.1007/s11060-005-9077-x
2. Choi G, Park DH, Kang SH, Chung YG: Glioma mimicking a hypertensive intracerebral hemorrhage. J Korean Neurosurg Soc. 2013;54(2): 125-127. 10.3340/jkns.2013.54.2.125. Address for reprints: Dong-Hyuk Park, M.D., Ph.D., Department of Neurosurgery, Korea University Anam Hospital, Korea University College of Medicine. 73:136-705. 10.3340/jkns.2013.54.2.125
3. Thaler PB, Li JY, Isakov Y, Black KS, Schulder M, Demopoulos A: Normal or non-diagnostic neuroimaging studies prior to the detection of malignant primary brain tumors. J Clin Neurosci. 2012, 19:411-414. 10.1016/j.jocn.2011.09.002
4. Miller KD, Ostrom QT, Kruchko C: Brain and other. 20212021, 71:381-406. 10.3322/caac.21693
5. Krouwer HG, Davis RL, Silver P, Prados M: Gemistocytic astrocytomas: a reappraisal. J Neurosurg. 1991, 74:399-406. 10.3171/jns.1991.74.3.0399
6. Abe, K., Hasegawa, H., Kobayashi: A gemistocytic astrocytoma demonstrated high intensity on MR images. Neuroradiology. 32:166-167.
7. Babu R, Bagley JH, Park JG, Friedman AH, Adamson C: Low-grade astrocytomas: the prognostic value of fibrillary, gemistocytic, and protoplasmic tumor histology. J Neurosurg. 2013, 119:1251-7. 10.3171/2013.4.JNS122329
8. Hauser A, Dutta SW, Showalter TN, Sheehan JP, Grover S, Trifiletti DM: Impact of academic facility type and volume on post-surgical outcomes following diagnosis of glioblastoma. J Clin Neurosci. 2018, 47:103-10. 10.1016/j.jocn.2017.09.035
9. Bitoh S, Hasegawa H, Ohtsuki H, Obashi J, Fujiwara M, Sakurai M: Cerebral neoplasms initially presenting with massive intracerebral hemorrhage. Surg Neurol. 1984, 22:57-62. 10.1016/0090-3019(84)90230-1
10. Liang M, Gao C, Wang Y: Enhanced blood-brain barrier penetration and glioma therapy mediated by T7 peptide-modified low-density lipoprotein particles. Drug Deliv. 2018, 25:1652-63. 10.1080/10717544.2018.1494223
11. Cîndea, I. E., & Cîndea, C. (2024). Exploring language acquisition in infants: A review from neurolinguistic and cognitive perspectives. Analele Universității din Craiova. Seria Științe Filologice. Lingvistică, 46(1-2), 245–262. https://doi.org/10.52846/aucssflingv.v46i1-2.143
12. Nozaki T, Sato H, Yamazoe T, Namba H: Diffuse astrocytoma initially presenting as a massive intracerebral hemorrhage: case report. Neurol Med Chir (Tokyo). 2014 Jan. 10:86-88. 10.2176/nmc.cr.2013-0177
13. Roman, C., Mihaila, R., Cindea C: Anticoagulation Reversal in Intracerebral Hemorrhage: A Case Report on the Efficacy of Andexanet Alfa in an Apixaban-Treated Patient. Cureus. 16:74750-10. 10.7759/cureus.74750
14. CN Cîndea, A Săceleanu: Year Journey Through Brain Gliomas: Case Report; . Acta Medica Transilvanica. 2020, 10.2478/amtsb-2020-0044
15. Willman M, Willman J, Figg J: Update for astrocytomas: medical and surgical management considerations. Explor Neurosci. 2023;2: 1-26. 10.37349/en.2023.00009. Received: September 06. 2022, 10:2022. 10.37349/en.2023.00009

Romanian Neurosurgery (2025) XXXIX (1): pp. 28-34
DOI: 10.33962/roneuro-2025-004
www.journals.lapub.co.uk/index.php/roneurosurgery

Three decades of colloid cyst resection. A single-centre retrospective analysis of 59 cases (1990-2020)

William Mirfin-Boukouris[1], Mustafa Dashti[1], Ahmed Aly[2], Viswapathi Kandula[2]

[1] Keele University, School of Medicine, Staffordshire, UK
[2] Neurosurgery Department, University Hospital North Midlands, Stoke-on-Trent, UK

Abstract

Purpose: To analyse 59 colloid cyst resections over 30 years at a single institution.
Methods: Retrospective review of electronic health records, including surgical approaches: transcortical (18), transcallosal (36), and endoscopic (5).
Results: Mean age at resection: 45.8 years. Cyst diameter: 4-27mm. Headache was the primary symptom (57.6%). Complications included memory deficits, infection, and neurological deficits; no mortality. Most cases were high-risk per Colloid Cyst Risk Score. Histology revealed pseudostratified epithelium (35%) and unique eosinophils. Craniotomy rate: 93%. Endoscopy had the highest reoperation rate; the transcallosal approach had more seizures and infections. Post-operative short-term memory issues: 40% (craniotomy), 50% (endoscopy).
Conclusions: Findings largely align with literature, with notable differences in headache prevalence, gender ratio, histology, and endoscopy outcomes.
This version reduces the word count by about 40% while retaining the essential information from each section. It maintains the structure and key points of the original abstract, allowing readers to quickly grasp the study's scope, methods, main findings, and conclusions.

Keywords
colloid cyst, resection, transcallosal, endoscopic

Corresponding author:
William Mirfin-Boukouris

Neurosurgery Department, University Hospital North Midlands, Newcastle Road, Stoke-on-Trent, UK

willwgmb@hotmail.com

Scan to access the online version

Introduction

Benign and rare, the colloid cyst or neuroepithelial cyst is an intracranial tumour, representing 0.5%-2% of all brain tumors[1–3]. The aetiology behind colloid cysts is not well understood. They have been postulated to be congenital in origin, deriving from the embryological remnants of the third ventricle, from the tela choroidea or endoderm[4,5]. Cases have been reported of 1st degree family relatives having colloid cysts, including identical twins. However, as of August 2022, only 23 familial cases were reported in the literature, suggesting that this is unlikely to be merely a genetic phenomenon[6].

Colloid cysts typically occur in the anterior third ventricle, frequently causing obstructive hydrocephalus. The presence of ventriculomegaly can cause a wide range of symptoms due to increased intracranial

pressure[5]. The most common of these symptoms is that of a persistent headache[5,7], and these symptoms can aid the calculation of the Colloid Cyst Risk Score (CCRS)[7]. The Colloid cyst risk score is not an official Score recommended by NICE guidelines; there are multiple versions of it available and determine which patients are at most immediate risk of complications as a result of their colloid cyst, guiding treatment. CCRS gives one point for each of the following:

- Age <65 years – because of fear the cyst will get bigger over time as a younger person on average will live longer.
- Headache - a sign of hydrocephalus causing brain damage.
- Axial diameter 7mm or above - greater size can cause more complete obstruction of the ventricular system
- Location of the cyst – the anterior third of the third ventricle increasing blockage risk
- FLAIR hyperintensity suggesting calcification of brain matter

This is the version used by our trust; however, modified versions exist, such as mCCRS - score out of 7 instead of 5, where an extra point is given for being female as this is believed to increase risk and for axial diameter one point is given for over 10mm instead of 7mm and a second point is given if over 15mm [5]. In this retrospective review, we will calculate the CCRS for resection cases to see what proportion of the risk score encouraged resection and what proportion was due to other factors such as patient desire for tumour resection for psychological reasons. However, this paper will primarily communicate the experience of our neurosurgical unit in resecting these colloid cysts.

One of the most severe consequences of concern in patients presenting with an untreated, predominantly large colloid cyst over 10mm is acute hydrocephalus and brain herniation. 86% of patients who die directly from their colloid cyst report having headaches as a symptom of brain damage before death, as shown in a systematic review of fatal colloid cysts in 2017 of literature over 15 decades [8]. These cysts are often found to be over 10mm in diameter [9]. The mechanism of sudden death is uncertain. One theory suggests that ventriculomegaly causes the mass effect of the cyst against the hypothalamus, triggering catecholamine release. This is linked to increased intracellular calcium and reactive nitrogen species, causing lipid peroxidation in cardiomyocytes and potentially resulting in cardiac arrest [8]. A less commonly argued hypothesis is that lumbar puncture prior to Computer tomography is a precipitant for sudden death linked to colloid cysts [8].

However, despite the risk of mortality, the risk of surgery must also be counterbalanced. Historical management favoured resection, but recent data supports initial conservative monitoring as many cysts remain asymptomatic.

A recent retrospective study with 82 patients found that only three required surgery[9]. The Colloid Cyst risk score is used to help gauge whether surgery is preferable. Alternative options include open craniotomy (interhemispheric transcallosal and transcortical) or endoscopic resection, which have a lower recurrence but higher complication rate like Transcranial resection or with lower complication but a greater chance of future surgery in the case of endoscopic resection; however, the expertise of the surgeon is the most important factor according to a recent meta-analysis [10]. Aspiration can be used to drain the cyst; however, it is known to have a higher recurrence, limiting its usefulness as a long-term solution; hence, this latter option is less likely to be used [4]. This is also reflected in our unit's choice of intervention.

Methods and Materials

In this study, we present a retrospective review of all patients at our hospital who underwent colloid cyst resection of the third ventricle. Our data covers 59 cases over 30 years, from January 1991 to November 2021. Sex was defined as biological unless otherwise specified in the iPortal records (no such specifications occurred in this study).

Due to the extended retrospective period, not all scans were readily available, as medical records were only accessible back to 2004. Consequently, the Colloid Cyst Risk Score (CCRS) could not be retrospectively calculated for all cases. Post-surgical follow-up, examining clinical features and CT radiological scans, was conducted for an average of 21.5 months for cases without recurrence. Seven cases are still ongoing and were not included in this calculation.

Resections were performed either via craniotomy or endoscopically. The craniotomy approaches included:

Transcortical: Involves creating a bone flap over the frontal area and navigating through the brain parenchyma to access the ventricle.

Transcallosal: Accesses the ventricle through the corpus callosum.

Endoscopic resection, a minimally invasive technique, utilizes endoscopes and specialized instruments inserted through a burr hole to access the ventricle and resect the cyst. Neuronavigation was employed for both open and endoscopic procedures.

Results

The mean age at resection was 45.8 years (range 17-76, SD 15.86). The age distribution of patients is shown in (Figure 1) including one patient under 18 years old. There was no significant difference between males (31 cases) and females (28 cases) in our study. The cyst sizes ranged between 4-27mm. A recurring theme in our analysis is that our sample size is too small for any statistically significant interpretation.

Figure 1. Shows the distribution of patients by age.

Histological findings (Table 1) included pseudostratified cuboidal epithelium in 35% of cases and columnar epithelium in 29%. Other findings were eosinophils, goblet cells, cilia, blood, and psammoma bodies.

Common presenting clinical features (Table 2) included headaches in most cases. Other frequent symptoms were memory problems, balance issues, collapse, visual disturbance, nausea, and vomiting. Interestingly, in six cases, the cyst was found incidentally, with two of these patients later found to have headaches. There were also singular symptoms like numbness. Common post-resection complications (Table 3) included memory problems, infection, neurological deficits, and seizures. Six patients passed away during follow-up; however, none were found to have died from causes related to the colloid cyst or its resection.

The Colloid Cyst Risk Score (CCRS) was calculable in 32 cases (not all radiology scans were available over the 30 years). The majority scored three or above, with a mode of 4 (Figure 2). Only 11 resections had a CCRS of 1 or 2. We investigated potential correlations between CCRS and post-surgical complications, as well as between cyst diameter and complication likelihood or symptoms, but found no significant correlations.

Table 1. Shows the percentages of histological findings of resected cysts.

Histology	%
Pseudostratified cuboidal	35%
Columnar	29%
Uncertain	12%
Cuboidal	9%
Other	6%
Cuboidal and columnar	6%
Pseudostratified columnar	3%

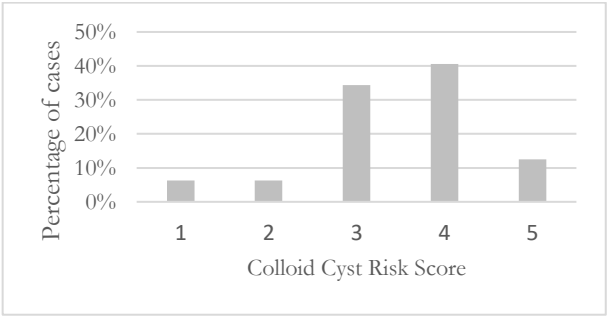


Figure 2. Show the percentages of colloid cyst risk scores in operated patients.

Table 2. Shows the frequency of patients' presenting symptoms.

Symptoms	
Headache	34
Visual Disturbances	10
Nausea & Vomiting	10
Hearing deficits	3
Seizures	2
Sensory Disturbances	4
Incidental Findings	6
Gait Disturbance	7
Balance Disturbance	12
Incontinence	6
Cognition	1
Confusion	4

Memory Issues	14
Collapse	12
Other	22

Table 3. Shows the frequency of complications postoperatively.

Complications	
Infection	7
Transient Neurological Deficits	7
Long-Term Neurological Deficits	4
Seizure	2
Short-term Memory Deficits	22
Long-Term Memory Deficits	7

Craniotomy was performed in 93% of cases (18 transcortical, 36 transcallosal), while only 7% were endoscopic (5 cases). Most patients were white (85%). Common radiological findings included ventriculomegaly, mass effect, ischemia, and haemorrhage.

We analysed complications by surgical approach. Memory issues occurred in 50% of endoscopic, 37.5% of transcortical, and 40% of transcallosal cases. Seizures were noted twice as often after the transcallosal approach. Postoperative infection rates were: endoscopic 0%, transcortical 11.11%, and transcallosal 11.11%. Return to theatre rates were: endoscopic 20%, transcortical 0%, and transcallosal 13.89%. Most resections were routine (72.22%), followed by urgent (18.52%) and semi-urgent (9.26%).

The complication rate was highest for the transcallosal approach, followed by endoscopic and transcortical. The most frequent approach was transcallosal, followed by transcortical and endoscopic.

Discussion

Our results represent the unique experience of our unit and, while not universally applicable, provide valuable insights and draw important parallels to existing literature.

At our centre, the mean age at resection was 45.8 years (range: 17-76, SD: 15.86); (see Fig. 1). This is in line with the statistics reported within other case reviews. Colloid cysts are reported to account for around 0.5%-2% of all brain tumours [1–3]. These findings reported by other papers are comparable to our centre's experience.

The resected cases had a CCRS score mode of 4, and very few scored 1 or 2, (see Figure 2). This makes logical sense as symptomatic patients would be expected to be more eager to have a resection than asymptomatic patients and symptomatic patients mostly scored 4 or 5 in research on risk analysis for colloid cysts[11]. We investigated whether higher CCRS and cyst diameter correlated with an increased risk of post-surgical complications, which might influence surgical decisions. However, no significant correlations were found. Colloid cysts are mostly found incidentally while investigating other conditions. There is a 5-15% chance within five years of diagnosis that progression will have taken place requiring management; conservative management is therefore often opted for, including neuroimaging [5]. Another paper found that 3/82 of conservatively managed patients required surgical management over ten years [9]. This shows how a large quantity of patients are best managed conservatively, which validates the management of our unit.

The histological findings were coherent with the rest of the literature, pseudostratified cuboidal or columnar with occasional evidence of calcification, cilia and goblet cells[12]. We found no evidence in the literature for the presence of eosinophils in histological samples of colloid cysts, which might suggest inflammation due to cellular damage. Whether this was due to hydrocephalus or surgery is uncertain, and this matter would benefit from further research. The features found frequently included cilia and goblet cells, supporting claims that colloid cysts share developmental origins with sinus epithelium[13].

Hyperintense was the most common FLAIR sign, followed by hypointense, Mirroring other retrospective research of resections[3]. This makes logical sense as a higher CCRS encourages resection, and Hyperintense FLAIR scores an extra point.

Of the 59 cases in our study, there did not seem to be a significant difference in prevalence between sexes, with 31 cases in males and 28 in females. The literature has found no definitive answer to the query as to whether one sex is more likely to have colloid cysts. A 105-patients study from Mumbai, India, found there was a 3:2 ratio for male: female[3]. Contrary to this, an 84-patients retrospective study found no link [14]. Another paper found that in 82 patients, conservatively managed men were more likely than women to be represented[9].

There was little diversity data available on the ethnic background of patients, preventing us from

making comparisons based on ethnicity as the vast majority were white due to the local population being white. Of the patients within our centre, no patients reported having any family members who had also previously suffered from a colloid cyst. Despite evidence of this being available in the literature[6]. Overall research on genetic aetiology has rarely yielded fruit.

The symptoms found within our retrospective review show that the symptoms were similar to those of other comparable research [1–3]; for instance, headache has been the most common finding, [15]. The same conclusion can be drawn from the post-surgical complications.

Patients presented with various signs suggestive of increased intracranial pressure. This included headache, hydrocephalus, vertigo, diplopia, mass effect, behaviour change, memory deficit, oedema, a decrease in Glasgow Coma Scale (GCS) score, sensory or motor loss, tinnitus, incontinence, syncope, and confusion. The most common symptom was headache appearing in 57.63% of our recorded cases, which was also found in a smaller retrospective study 90% of the time [2], as well as the more extensive Mumbai study of 105 cases [3] where it appeared in 92% of cases. This suggests documenting headaches at our centre is done less commonly, perhaps with a threshold of higher intensity of pain, or that patients in the other studies were effectively managed more conservatively for longer. The Mumbai paper had several patients come in comatose and several died, unlike 0% in our study where resection were rarely urgent and hence symptoms less pronounced. Patients at our Hospital are operated on if they are determined to be at moderate or high risk. Except for headache, the symptoms closely correlated with fatal outcomes were not common at our centre: change in gait, decreased GCS, seizure, emesis and nausea to a degree [8]. In the Mumbai paper, 4.7% of patients out of 105 cases died from surgery [8], this is higher than in our data sample, where 0% died because of surgery; it should be acknowledged the medical demands are likely much higher in Mumbai.

The radiological findings are in keeping with the rest of the literature: ventriculomegaly was often found logically following the tumour's obstruction of the foramen of Monroe.

Larger tumours found in older patients might be expected to be due to a greater length of time for the tumour to grow, It may seem intuitive to conclude that the risk of death would therefore be higher at greater age, but the literature suggests that it is more likely to happen earlier in life [9]. This is coherent with the congenital hypothesis about their origins as if the colloid cyst is congenital in origin then the harm caused would be more likely to transpire in the younger years of life. Our data shows no significant correlation of age at resection against diameter. This does not concord with the hypothesis that they enlarge to become a problem across a lifespan. According to the literature, people under 18 are said to be less likely to have hydrocephalus [16]. Our data only had one patient 18 or under, so we cannot comment on this reliably. According to a recent meta-analysis, transcallosal, endoscopic and transcortical approaches did not show any difference in the chance of seizure [10]. Other studies found that compared to other approaches, transcallosal increased the risk of seizures [3,9]. Our data only showed two seizures in resected patients, both transcallosal. We cannot weigh in on this controversy decisively with this sample size. Research has shown an increased risk of short-term memory loss with the transcallosal approach [9,10,17,18]. Our data showed short-term memory issues: endoscopic 50%, transcortical 37.5%, and transcallosal 40%; our data does not support this proposed hypothesis; one possibility for the difference is surgical techniques and ability have changed over the decades confounding results. Our Data shows that transcallosal had the highest mean complication rate, followed by endoscopic and finally transcortical. Other research found that endoscopic should have a lower but statistically insignificant complication rate than transcranial[19], which does not contradict our results.

According to several studies, endoscopic resection is commonly linked to a lower complication rate, yet surprisingly, some research has been linked to an increased risk of infection like ventriculitis and meningitis [18–20]. Our data disagrees; Infection as a complication happened the least endoscopically, 0% and 11.11% transcallosal and 11.11% transcortical, supporting the general notion that endoscopic is less likely to cause infection due to smaller incision. Endoscopic was more likely to require a return to theatre at 20% compared to 0% transcortical and 13.8% transcallosal. The surgical methodology has been 93.22% craniotomy and only

6.68% endoscopic because the former has a lower risk of return to theatre at any point after surgery than the latter, which is why it is opted for.

Overall, our data shows transcortical had the lowest risk of return to theatre whilst still having a lower risk of infection than transcallosal yet is only used half as much as transcallosal; furthermore, transcallosal appears to have the highest complication rate, whereas transcortical had the lowest suggesting our unit might possibility benefit from opting for a transcortical approach more often, likely due to higher surgical skill level despite been use less often than transcallosal. Endoscopic is rarely used, so whilst endoscopic could be suggested to have a lower chance of complications due to smaller incisions, this is less important than the surgeon's skill with a given technique.

Conclusion

Our unit's 30-year experience with 59 colloid cyst resections largely affirms findings reported in the literature while providing valuable insights into our specific management practices. We found no notable sex difference in colloid cyst prevalence or resection rates, with 31 male and 27 female patients. The mean age at resection was 45.8 years, and most patients had a high Colloid Cyst Risk Score (CCRS) of 3 or 4, indicating higher-risk cases.

Histological findings were consistent with the literature, except for the presence of eosinophils, which was not reported in other studies and may warrant further investigation. Cyst diameters ranged from 4 to 27 mm, aligning with ranges reported in other studies. Presenting symptoms were generally as expected, but headaches were reported in only 57.63% of our cases, significantly lower than the 90-92% reported in other studies. Radiological findings were typical, with hyperintense Fluid-Attenuated Inversion Recovery (FLAIR) signals being most common, and ventriculomegaly frequently observed.

Of the 59 cases, 36 (61%) were resected using the transcallosal approach, 18 (31%) transcortical, and only 5 (8%) endoscopically. Post-operative seizures were rare, with only two cases observed, both following the transcallosal approach. Post-operative infections were most common in transcallosal and transcortical approaches (both 11.11%), while no infections were observed in endoscopic cases. Short-term memory loss was observed in 50% of endoscopic cases and approximately 40% of craniotomy cases (37.5% transcortical, 40% transcallosal). The rate of return to the theater was highest for the endoscopic approach (20%), compared to 13.89% for transcallosal and 0% for transcortical approaches.

References

1. Kondziolka D, Lunsford LD. Microsurgical resection of colloid cysts using a stereotactic transventricular approach. Surg Neurol. 1996;46(5):485-492. doi:10.1016/S0090-3019(96)00201-7
2. Jeffree RL, Besser M. Colloid cyst of the third ventricle: a clinical review of 39 cases. Journal of Clinical Neuroscience. 2001;8(4):328-331. doi:10.1054/jocn.2000.0800
3. Desai KI, Nadkarni TD, Muzumdar DP, Goel AH. Surgical management of colloid cyst of the third ventricle—a study of 105 cases. Surg Neurol. 2002;57(5):295-302. doi:10.1016/S0090-3019(02)00701-2
4. Yadav YR, Yadav N, Parihar V, Kher Y, Ratre S. Management of colloid cyst of third ventricle. Turk Neurosurg. Published online 2014. doi:10.5137/1019-5149.JTN.11086-14.1
5. Burhan Janjua M, Reddy S, El Ahmadieh TY, et al. Inchoate guidelines of endoscopic resection of colloid cysts. Journal of Clinical Neuroscience. 2020;71:1-8. doi:10.1016/j.jocn.2019.12.017
6. Calderon C, Fernandez-de Thomas R, De Jesus O. Familial colloid cysts of the third ventricle: Case report and literature review. Asian J Neurosurg. 2020;15(02):414-417. doi:10.4103/ajns.AJNS_332_19
7. Alford EN, Rotman LE, Lepard JR, Agee BS, Markert JM. Interrater and Intrarater Reliability of the Colloid Cyst Risk Score. Neurosurgery. 2020;86(1):E47-E53. doi:10.1093/neuros/nyz399
8. Lagman C, Rai K, Chung LK, et al. Fatal Colloid Cysts: A Systematic Review. World Neurosurg. 2017;107:409-415. doi:10.1016/j.wneu.2017.07.183
9. Velicu MA, Rossmann K, Vahedi A, et al. On Natural History and Management of Colloid Cysts: Time to Rethink? World Neurosurg. 2023;170:e188-e199. doi:10.1016/j.wneu.2022.10.094
10. Elshamy W, Burkard J, Gerges M, et al. Surgical approaches for resection of third ventricle colloid cysts: meta-analysis. Neurosurg Rev. 2021;44(6):3029-3038. doi:10.1007/s10143-021-01486-5
11. Zeineddine HA, Westmark K, Khanpara S, et al. Risk Analysis and Management of Third Ventricular Colloid Cysts. World Neurosurg. 2021;146:e1071-e1078. doi:10.1016/j.wneu.2020.11.090
12. Khanpara SD, Day AL, Bhattacharjee MB, Riascos RF, Fernelius JP, Westmark KD. The Variable Appearance of Third Ventricular Colloid Cysts: Correlation with Histopathology and the Risk of Obstructive

Ventriculomegaly. American Journal of Neuroradiology. 2020;41(10):1833-1840. doi:10.3174/ajnr.A6722

13. Lach B, Scheithauer BW, Gregor A, Wick MR. Colloid cyst of the third ventricle. J Neurosurg. 1993;78(1):101-111. doi:10.3171/jns.1993.78.1.0101
14. Camacho A, Abernathey C, Kelly P, Laws E. Colloid Cysts: Experience with the Management of 84 Cases Since the Introduction of Computed Tomography. Neurosurgery. 1989;24:693-700.
15. Desai KI, Ch M, Nadkarni TD, Muzumdar DP, Goel AH. Surgical Management of Colloid Cyst of the Third Ventricle-A Study of 105 Cases. Acta Neurol Scand. 2016;135(4):484-487.
16. McCrea HJ, Lara-Reyna J, Perera I, et al. Colloid cysts of the third ventricle in children. J Neurosurg Pediatr. 2021;27(6):700-706. doi:10.3171/2020.10.PEDS18458
17. Sampath R, Vannemreddy P, Nanda A. Microsurgical Excision of Colloid Cyst With Favorable Cognitive Outcomes and Short Operative Time and Hospital Stay. Neurosurgery. 2010;66(2):368-375. doi:10.1227/01.NEU.0000363858.17782.82
18. Milligan BD, Meyer FB. Morbidity of Transcallosal and Transcortical Approaches to Lesions in and Around the Lateral and Third Ventricles: A Single-Institution Experience. Neurosurgery. 2010;67(6):1483-1496. doi:10.1227/NEU.0b013e3181f7eb68
19. Connolly ID, Johnson E, Lamsam L, Veeravagu A, Ratliff J, Li G. Microsurgical vs. Endoscopic Excision of Colloid Cysts: An Analysis of Complications and Costs Using a Longitudinal Administrative Database. Front Neurol. 2017;8. doi:10.3389/fneur.2017.00259

Romanian Neurosurgery (2025) XXXIX (1): pp. 35-41
DOI: 10.33962/roneuro-2025-005
www.journals.lapub.co.uk/index.php/roneurosurgery

Carpal Tunnel syndrome surgery. A 10-year retrospective analysis of 442 cases

Sonia Lucaciu[1], Antonia Iliescu[2], Cosmin-Nicodim Cindea[2], Diana Elena Teacoe[1], Alexandru Breazu[1], Claudia Elena Popa[2], Titus Fagarasi[1], Vicentiu Saceleanu[2]

[1] County Clinical Emergency Hospital of Sibiu, ROMANIA
[2] "Lucian Blaga" University of Sibiu, UK

ABSTRACT

Carpal Tunnel Syndrome (CTS) is the most common peripheral nerve entrapment neuropathy, caused by compression of the median nerve at the wrist. It leads to hand pain, numbness, tingling, and weakness, often affecting the thumb, index, middle, and ring fingers. CTS frequently requires surgical treatment (carpal tunnel release) when symptoms are moderate to severe or unresponsive to conservative measures.

We analysed a cohort of 442 CTS patients treated surgically over the last 10 years at the County Clinical Emergency Hospital of Sibiu (312 women, 130 men; 308 urban residents, 134 rural; average age 59 years) and compared the outcomes and characteristics with findings from the literature. This report examines risk factors and comorbidities associated with CTS, surgical outcome metrics (recurrence, failure, complications), bilateral involvement patterns, laterality, urban-rural differences, and occupational contributions, supported by recent studies.

Keywords
carpal tunnel,
electromyography,
surgical complications,
peripheral nerve surgery,
decompression,
risk factors

Corresponding author:
Cosmin-Nicodim Cindea

"Lucian Blaga" University of Sibiu, Romania

cindea.cos@gmail.com

Scan to access the online version

INTRODUCTION

Carpal Tunnel Syndrome (CTS) is the most common peripheral nerve entrapment neuropathy, caused by compression of the median nerve at the wrist. It leads to hand pain, numbness, tingling, and weakness, often affecting the thumb, index, middle, and ring fingers. CTS frequently requires surgical treatment (carpal tunnel release) when symptoms are moderate to severe or unresponsive to conservative measures.

We analyzed a cohort of 442 CTS patients treated surgically over the last 10 years at the *County Clinical Emergency Hospital of Sibiu* (312 women, 130 men; 308 urban residents, 134 rural; average age 59 years) and compared the outcomes and characteristics with findings from the literature. This report examines risk factors and comorbidities associated with CTS, surgical outcome metrics (recurrence, failure, complications), bilateral involvement patterns, laterality, urban–rural differences, and occupational contributions, supported by recent studies.

Patient Demographics and Risk Factors for CTS

Gender and Age

In our series, about 70% of patients were female (312 women vs. 130 men), reflecting the well-known female predominance in CTS. Population studies show women have significantly higher incidence rates of CTS than men (approximately 2–3 times higher) [1]. Hormonal and anatomical differences are thought to contribute, and CTS most often manifests in middle age (commonly in the 40–60 year range) [2,3]. Our patients' average age was 59, consistent with the typical age of CTS sufferers (CTS is uncommon in youth and increases in prevalence until late midlife) [4].

Common Risk Factors and Comorbidities

Multiple medical and lifestyle factors contribute to CTS risk, with most patients in our cohort having at least one known risk factor.

- **Diabetes Mellitus:** Diabetics have a 90% higher risk of developing CTS due to chronic hyperglycemia-induced neuropathy. In our series, 126 patients (28.5%) had diabetes, aligning with its known high co-incidence with CTS [5].
- **Obesity:** Excess weight increases intracarpal pressure, raising CTS risk. Studies confirm a strong correlation between high BMI and CTS, which was evident in our patient group, where 172 patients (39%) were obese [6].
- **Thyroid Disorders:** Hypothyroidism can cause median nerve compression through myxedematous changes, a pattern observed in 41 patients (9.3%) in our cohort [7].
- **Rheumatoid and Inflammatory Arthritis:** Chronic synovitis from RA compresses the median nerve, commonly leading to CTS. Other inflammatory conditions like gout and psoriatic arthritis may have similar effects. In our cohort, 38 patients (8.6%) had RA or another inflammatory arthritis.
- **Pregnancy and Hormonal Factors:** Hormonal fluctuations, fluid retention, and anatomical changes in pregnancy and menopause increase CTS risk, as noted in 67 female patients (15.2%), who reported onset or worsening of symptoms during pregnancy or menopause.
- **Repetitive Hand Use and Occupational Factors:** Jobs involving repetitive wrist motions (e.g., factory work, typing, vibrating tools) are significant CTS risk factors. Many patients had prolonged exposure to such tasks. In our study, 185 patients (41.9%) had occupations associated with CTS risk. Prior wrist injuries (fractures, sprains) were also observed as contributors in 34 cases (7.7%) [8].

Other comorbidities in our series included hypertension (198 patients, 44.8%) and osteoarthritis (89 patients, 20.1%), both common in older populations. Chronic kidney disease, hormonal contraceptive use, and connective tissue disorders are additional potential risk factors reported in literature (1). Our patient cohort—older, predominantly female, and with high diabetes and obesity prevalence—mirrors established epidemiological data on CTS risk factors.

Bilateral Involvement and Hand Laterality

Prevalence of Bilateral CTS

CTS frequently affects both hands, either simultaneously or sequentially. Studies report that 50–80% of CTS patients develop bilateral involvement. In our cohort, 225 patients (54.7%) eventually had symptoms in both hands, and 94 patients (22%) required surgery on both hands. Many underwent surgery on one hand and later developed significant symptoms in the opposite hand, aligning with clinical observations that CTS often progresses over time [9].

Impact on Treatment

The high bilateral prevalence influenced surgical strategies. Staged surgery was the preferred approach, addressing the more symptomatic or non-dominant hand first, followed by the second hand after sufficient recovery. This method optimized postoperative function and minimized disability during healing. Simultaneous bilateral CTR was rare due to potential recovery challenges. Staged surgery outcomes were generally successful, but careful monitoring for contralateral symptom progression was essential [10].

Dominant vs. Non-dominant Hand (Right vs. Left)

The right hand was affected in 266 cases (60.2%) compared to 176 cases (39.8%) on the left, reflecting the predominance of right-hand dominance and greater cumulative hand use. This distribution aligns with literature reporting a slightly higher right-hand involvement in CTS [11]. However, bilateral CTS and variable presentation highlight the importance of

evaluating both hands in all patients, even if initial symptoms are unilateral.

Surgical Outcomes: Recurrence and Failure Rates

Carpal tunnel release (CTR) is highly effective, with most patients achieving significant symptom relief. However, a small subset experiences recurrence or requires additional interventions.

- **Success Rate:** The vast majority of our 442 patients had significant improvement. Only 32 patients (7.2%) reported persistent symptoms, and 9 patients (2%) required revision surgery. These results align with literature indicating that 69% of patients become symptom-free long-term, with nearly all showing partial improvement [12].
- **Recurrence Rates:** True recurrence (symptom return after a pain-free interval) is uncommon when the ligament is fully released. In our cohort, 9 patients (2%) required reoperation, matching reported revision rates of 1–5% . Including minor symptom persistence, recurrence rates can reach 10–25% in broader studies [13].
- **Risk Factors for Recurrence/Revision:**
  - **Incomplete Decompression:** The leading technical cause of recurrence, found in all revision cases in our study.
  - **Male Sex:** Males had a slightly higher revision rate, reflecting trends seen in larger studies [14].
  - **Rheumatoid Arthritis:** Present in 38 patients (8.6%), RA was linked to a higher likelihood of persistent CTS and revision surgery [15].
  - **Smoking:** Identified in 14 revision cases, smoking is associated with poorer surgical outcomes.
  - **Bilateral Surgery:** Simultaneous bilateral CTR is linked to higher revision risk; our staged approach mitigated this.
  - **Endoscopic Technique:** Though not used in our cohort, endoscopic CTR has a higher reported recurrence rate than open CTR [16].

These findings emphasize that proper surgical technique and patient risk management are crucial to minimizing recurrence and optimizing long-term outcomes.

Surgical 'Failures' or Persistent Symptoms

A small subset of patients did not experience full symptom relief post-surgery. In our cohort, 14 patients (3.2%) had persistent significant symptoms in the early postoperative period. Contributing factors included:

- **Misdiagnosis or Concurrent Neurologic Conditions:** Conditions such as cervical radiculopathy, diabetic peripheral neuropathy, or ulnar nerve compression can mimic CTS. Proper preoperative diagnosis using clinical exams and EMG helped prevent unnecessary surgeries in many cases [17].
- **Anatomical Variants:** Rare nerve anomalies like bifid median nerves or median-ulnar anastomoses can complicate diagnosis and surgery, leading to persistent symptoms. These variants must be considered in refractory cases [18].
- **Scar Tissue and Fibrosis:** Postoperative fibrosis can re-compress the nerve, occasionally requiring revision surgery. One patient (0.2%) in our study needed a secondary procedure to release excessive scar tissue.
- **Intraoperative Nerve Injury:** Nerve damage is exceedingly rare (<1%) in CTR. We recorded no cases of iatrogenic nerve laceration, likely due to careful intraoperative visualization and the open surgical technique used in all cases.

Postoperative Complications

Carpal tunnel release is a low-risk surgery with minimal complications. Among 442 patients, we observed:

- **Infection Rates:** Only 2 patients (0.5%) developed superficial wound infections, both resolving with antibiotics. No deep infections or abscesses were recorded. This aligns with literature indicating infection rates of <1% in CTR cases. A multicenter study of 3,003 CTR surgeries found infection rates of 0.37%, with deep infections occurring in <0.5% of cases [19].
- **Risk Factors for Infection:** Infection was rare regardless of comorbidities. One infected patient had diabetes, but studies suggest no significant difference in infection rates between diabetic and non-diabetic CTS patients. Routine prophylactic antibiotics were not used, consistent with best practices given the low infection risk. Some literature suggests younger males have a slightly higher risk of complications post-CTR, but severe

cases (e.g., requiring reoperation) remain under 0.1% [20].

- **Other Complications:** No cases of permanent nerve or tendon injury were recorded. Minor issues, such as transient pillar pain or scar tenderness, occurred in 38 cases (8.6%), resolving within weeks to months. Our complication rates were at or below published benchmarks, which estimate major complications (nerve, vessel, or tendon injury) in 0.9% of CTR cases and minor complications (e.g., wound issues, transient pain) in ~10% [21]. The use of open mini-incision techniques in all patients likely contributed to the low complication rate.

Urban vs. Rural Disparities in Surgery Rates

Our dataset showed a notable urban-rural disparity: 308 patients (69.7%) were from urban areas, while 134 (30.3%) were from rural regions. This suggests higher surgery rates or better healthcare access in urban populations. Several factors likely explain this trend:

- **Healthcare Access:** Urban residents have closer access to hospitals, hand surgeons, and nerve conduction studies, facilitating earlier diagnosis and treatment. In contrast, rural patients may delay care due to long travel distances or limited specialist availability. A U.S. study found only ~5% of CTR surgeries were performed on rural patients at tertiary centers, highlighting access disparities [22].
- **Awareness and Referrals:** Urban patients may be more aware of CTS and its treatment options, with higher referral rates from primary care providers. Rural patients may attribute symptoms to physical labor and seek care later, often with more severe symptoms.
- Occupational Factors: Urban jobs (e.g., assembly line, office work) involve repetitive hand movements, increasing CTS risk. Rural occupations, while physically demanding, may not always involve repetitive hand motions to the same extent. However, some rural industrial areas report high CTS incidence [23].
- **Healthcare Infrastructure and Socioeconomics:** Urban areas generally have more surgeons and shorter wait times for elective procedures. Rural patients may face delays due to fewer specialists. Socioeconomic factors also play a role—urban patients may have better insurance coverage and financial means for surgery [24].

Interestingly, rural patients in our cohort were generally older and had lower BMIs than urban patients. This mirrors studies indicating rural CTS patients often present later and may have atypical symptoms [25]. To improve equity in CTS care, initiatives such as telemedicine consultations and surgical outreach programs could help bridge this gap.

These findings highlight the need for improved CTS awareness, early diagnosis, and accessible treatment options in rural populations to prevent long-term disability.

Occupational and Professional Factors

Repetitive strain and occupational hand use are well-known contributors to CTS. In our study, 185 patients (41.9%) had jobs involving repetitive wrist motions, such as factory work, office typing, or manual labor. Based on literature, nearly 47% of CTS cases are considered work-related [26], suggesting that ~200 patients in our series may have developed CTS due to occupational strain.

- **High-Risk Professions**: Common CTS-related jobs include assembly line workers, cashiers, office workers, and manufacturing laborers. Certain industries report 13–15 CTS cases per 1,000 workers annually.
- **Physical Demands:** Jobs involving vibrating tools, repetitive grasping, and fine motor tasks (e.g., sewing, cleaning) also contribute to CTS risk. Our urban hospital likely treated many such workers.
- **Interaction with Medical Risk Factors:** Work-related CTS was often seen in patients with additional risk factors like diabetes, obesity, or rheumatoid arthritis. A diabetic factory worker or an obese office clerk may experience more severe CTS due to the combination of systemic and occupational strain.

To reduce work-related CTS, ergonomic interventions, wrist splints, and employer-supported modifications should be promoted. Patients returning to high-risk jobs post-surgery benefited from workplace adjustments to prevent recurrence. 40–50% of our CTS cases were likely occupational in origin, underscoring CTS as both a medical and workplace health issue. Improved workplace

ergonomics and early intervention can help reduce the burden of CTS and the need for surgery.

Cross-Reference with Broader Studies and Our Dataset Comparison

Our 442-patient dataset over 10 years aligns with broader CTS studies in several key aspects:

- Gender Ratio: Our female-to-male ratio was 2.4:1 (312 women, 130 men), consistent with epidemiologic reports showing 2:1 to 4:1 ratios. Studies indicate 60–75% of CTS patients are female, supporting our findings [27].
- Age Profile: The average age was 59 years (range 20–87), comparable to large studies where most CTS surgeries occur between 50–70 years [28]. Elderly patients (>75 years) had longer hospital stays (2.6 vs. 1.7 days), likely due to monitoring for comorbidities.
- Urban vs. Rural Distribution: Our 69.7% urban, 30.3% rural patient split highlights an access gap. While less extreme than the 95:5 ratio seen in some U.S. tertiary centers, it suggests rural patients may delay surgery or face referral barriers [29].
- Bilateral Cases: 225 patients (50.9%) had bilateral CTS, with 94 (22%) undergoing staged bilateral surgeries. This matches studies showing 50–80% bilateral involvement on nerve studies, though not all require immediate surgery [30].
- Recurrence and Revision Rates: Our 1–2% revision rate aligns with published 1.5% rates for reoperation. Most cases of recurrence in our series were due to incomplete decompression or underlying conditions, similar to reported findings.
- Comorbidities: Our high prevalence of diabetes (28.5%), obesity (39%), and RA (8.6%) mirrors literature indicating metabolic and inflammatory disorders increase CTS risk.

CASE REPORTS

To illustrate real-world clinical scenarios, we present two representative cases highlighting key aspects of bilateral CTS presentation, surgical management, and postoperative outcomes.

Case 1

Bilateral CTS in a 69-Year-Old Male with Diabetes A 69-year-old right-handed male with type 2 diabetes and hypertension presented with progressive numbness, tingling, and grip weakness in both hands, more severe on the left. Symptoms interfered with daily activities, including buttoning his shirt and gripping objects. Nerve conduction studies confirmed severe CTS bilaterally, with prolonged median nerve latencies. Conservative treatments, including wrist splints and NSAIDs, provided little relief.

He initially underwent open carpal tunnel release (CTR) on the left hand, experiencing significant improvement in sensation and function. Postoperatively, he had delayed wound healing, likely due to his diabetic status, but no infection. He engaged in hand therapy to restore full dexterity. One year later, he developed worsening symptoms in his dominant right hand, necessitating a second CTR. The staged approach allowed uninterrupted hand function during recovery, and the second surgery also resulted in symptom relief. Follow-up at six months postoperatively showed full functional recovery, though mild residual numbness persisted in the fingertips. This case highlights diabetes as a risk factor for delayed recovery and reinforces the common bilateral nature of CTS, necessitating careful monitoring for progression in the contralateral hand.

Case 2

Bilateral CTS in a 72-Year-Old Female with Obesity A 72-year-old woman with obesity and hypertension developed progressive left-hand CTS symptoms, characterized by nocturnal pain and clumsiness while performing fine motor tasks. She had a gradual decline in hand strength and occasional hand swelling. Electrophysiologic studies confirmed moderate-to-severe median nerve compression bilaterally, though symptoms were more pronounced on the left.

She underwent staged CTR, starting with the more affected left hand. Postoperative recovery was uneventful, with full symptom resolution within six weeks. Physical therapy was initiated to improve grip strength. Within a year, she developed similar symptoms in the right hand, which was subsequently treated with CTR. Healing was smooth, and she regained full function without complications. Despite obesity being a recognized risk factor for CTS, it did not delay her postoperative recovery. At one-year follow-up, she reported complete resolution of symptoms in both hands, with no recurrence. This

case further demonstrates the high bilateral occurrence of CTS and the effectiveness of staged surgery in elderly patients, allowing gradual functional restoration without significant disability during recovery.

Both cases emphasize the importance of staged bilateral surgical planning, risk factor consideration (diabetes, obesity), and tailored patient management to optimize recovery and function. These cases also illustrate that while comorbidities may prolong healing, staged CTR remains a highly effective approach for managing bilateral CTS in medically complex patients.

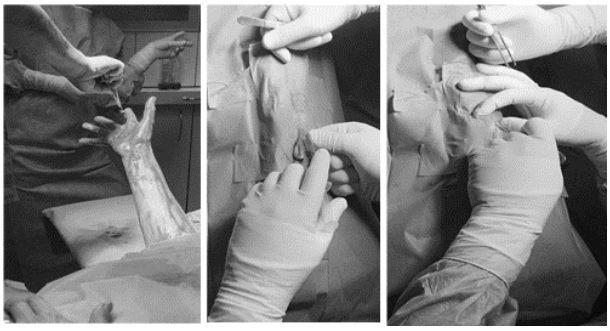


Figure 1. Surgical skin antisepsis, local lidocaine anesthesia, and skin incision performed.

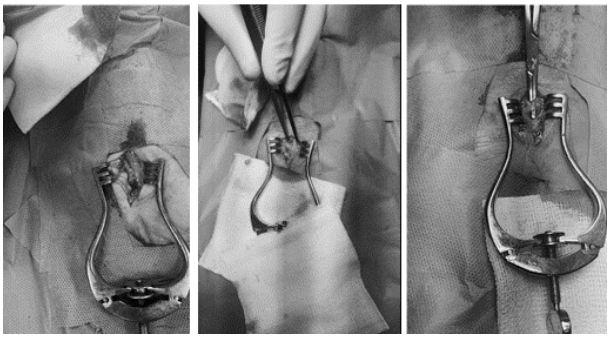


Figure 2. Surgical retractor, layer dissection, and exposure of the flexor retinaculum.

CONCLUSION

Key Findings

Our 442-patient analysis confirms that carpal tunnel release is highly effective, with outcomes aligning with international literature. The patient profile was predominantly female (70.6%), older adults (mean age 59 years), with common comorbidities like diabetes (28.5%), obesity (39%), and RA (8.6%). Bilateral CTS was frequent (50.9%), with 22% requiring staged surgery. The right hand was affected in 60.2% of cases. Surgery led to symptom relief in nearly all patients, with low recurrence (1–2%) linked to incomplete decompression, RA, or smoking. Complications were rare (infection 0.5%, no major nerve injuries). Urban patients comprised 69.7%, reflecting better access to care, while rural patients were underrepresented. Occupational factors played a role in 41.9% of cases.

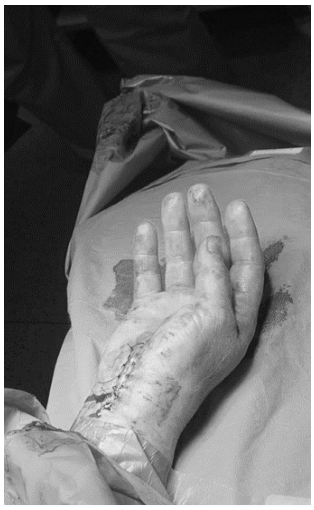


Figure 3. Skin closure with sutures.

Clinical Implications

Managing modifiable risk factors such as diabetes, obesity, and RA may improve outcomes. Open CTR remains the gold standard, offering low complication rates and high success. Patients should be aware of recurrence risks in RA and smokers. The urban-rural gap underscores the need for better rural access to care, and workplace interventions could prevent work-related CTS.

Final Thoughts

CTS is influenced by medical, occupational, and lifestyle factors. Our findings confirm high surgical success, the importance of early diagnosis, and the need for equitable access. CTR provides lasting relief with minimal risk, emphasizing the value of prevention, conservative management, and improved rural healthcare access.

REFERENCES

1. Mitake T, Iwatsuki K, Hirata H. Differences in characteristics of carpal tunnel syndrome between male and female patients. J Orthop Sci. 2020;25(5):843-846.
2. Genova A, Dix O, Saefan A, Thakur M, Hassan A. Carpal Tunnel Syndrome: A Review of Literature. Cureus. 2020;12(3):e7333.
3. Wipperman J, Goerl K. Carpal tunnel syndrome: Diagnosis and management. Am Fam Physician. 2016;94(12):993-999.
4. Sevy JO, Sina RE, Varacallo MA. Carpal Tunnel Syndrome. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2023 Aug.

5. Sanjari E, Raeisi Shahraki H, Khachatryan LG, Mohammadian-Hafshejani A. Investigating the association between diabetes and carpal tunnel syndrome: A systematic review and meta-analysis. PLoS One. 2024;19(4):e0299442.
6. Shiri R, Pourmemari MH, Falah-Hassani K, Viikari-Juntura E. The effect of excess body mass on the risk of carpal tunnel syndrome: a meta-analysis of 58 studies. Obes Rev. 2015;16(12):1094-1104.
7. Shiri R. Hypothyroidism and carpal tunnel syndrome: a meta-analysis. Muscle Nerve. 2014;50(6):879-883.
8. Guan W, Lao J, Gu Y, Zhao X, Rui J, Gao K. Case-control study on individual risk factors of carpal tunnel syndrome. Exp Ther Med. 2018;15(3):2761-2766.
9. Bhanja D, Freedman Z, Daggubati L, Moeckel C, Rizk E. Simultaneous versus staged bilateral carpal tunnel release via open and endoscopic surgeries: a retrospective propensity score-matched analysis. J Neurosurg. 2024;140(5):1414-1422.
10. Osei DA, Calfee RP, Stepan JG, Boyer MI, Goldfarb CA, Gelberman RH. Simultaneous bilateral or unilateral carpal tunnel release? A prospective cohort study of early outcomes and limitations. J Bone Joint Surg Am. 2014;96(11):889-896.
11. Cîndea, I. E., & Cîndea, C. (2024). Exploring language acquisition in infants: A review from neurolinguistic and cognitive perspectives. Annals of the University of Craiova. Series Philology. Linguistics, 46(1-2)
12. Carrillo-Ruiz JD, Cedeño DL, Martínez-Navarro MA, et al. Positive sensory symptoms in surgically managed patients with carpal tunnel syndrome: a long-term follow-up. Exp Ther Med. 2024;(in press).
13. Krieger Y, Ofri M, Sagi G, et al. Long-Term Functional Outcomes and Quality of Life Following Carpal Tunnel Release Surgery. Int J Environ Res Public Health. 2022;21(9):1203.
14. Neuhaus V, Christoforou D, Cheriyan T, Mudgal CS. Evaluation and treatment of failed carpal tunnel release. Orthop Clin North Am. 2012;43(4):439-447.
15. Saceleanu, V., Paz, R., García, J., Rivero, Y., Cîndea, C.-N., Cacciotti, I., & Monzón, M. (2021). Production of Synthetic Models for Neuro-Oncology Training by Additive Manufacturing. Applied Sciences, 11(24), 11823. https://doi.org/10.3390/app112411823
16. Furniss D, Taylor C, Giuffrida V, et al. Serious postoperative complications and reoperation after carpal tunnel decompression in England: a nationwide cohort analysis. Lancet Rheumatol. 2020;3(1):e49-e57.
17. Westenberg RF, Oflazoglu K, de Planque CA, Jupiter JB, Eberlin KR, Chen NC. Revision carpal tunnel release: risk factors and rate of secondary surgery. Plast Reconstr Surg. 2020;145(5):1204-1214.
18. Santosa KB, Chung KC, Waljee JF. Complications of compressive neuropathy: prevention and management strategies. Hand Clin. 2015;31(2):139-149.
19. Saceleanu, C., Cindea, C., and Saceleanu, A. (2018). Pediatric traumatic brain injury: pathophysiology, treatment and case reports. Acta Medica Transilvanica 23(3): 48–51. amtsibiu.ro
20. Keith MW, Masear VR, Chung KC, et al. AAOS Clinical Practice Guideline on diagnosis of carpal tunnel syndrome. J Am Acad Orthop Surg. 2009;17(6):389-396.
21. Kim YS, Ko HC, Son BC. Carpal Tunnel Syndrome Caused by High Division of the Median Nerve Proximal to the Carpal Tunnel: A Case Report. The Nerve. 2018;4(1):1-4.
22. Harness NG, Inacio MC, Pfeil FF, Paxton LW. Rate of infection after carpal tunnel release surgery and effect of antibiotic prophylaxis. J Hand Surg Am. 2010;35(2):189-196.
23. Furniss D, et al. Serious postoperative complications and reoperation after carpal tunnel decompression... Lancet Rheumatol. 2020;3(1):e49-e57.
24. Santosa KB, et al. Complications of compressive neuropathy... Hand Clin. 2015;31(2):139-149.
25. Earp BE, Blazar PE, Zhang D. Rural-Urban Disparities in the Surgical Treatment of Carpal Tunnel Syndrome in the United States. Cureus. 2024;16(7):e65687.
26. Mondelli M, Giannini F, Giacchi M. Carpal tunnel syndrome incidence in a general population. Neurology. 2002;58(2):289-294.
27. Kim CY, Wiznia DH, Wang Y, et al. The Effect of Insurance Type on Patient Access to Carpal Tunnel Release Under the Affordable Care Act. J Hand Surg Am. 2016;41(4):503-509.e1.
28. Earp BE, et al. Rural-Urban Disparities... Cureus. 2024;16(7):e65687.
29. Luckhaupt SE, Dahlhamer JM, Ward BW, et al. Prevalence and work-relatedness of carpal tunnel syndrome in the working population, United States, 2010 National Health Interview Survey. Am J Ind Med. 2013;56(6):615-624.
30. Bagatur AE, Zorer G. The carpal tunnel syndrome is a bilateral disorder. J Bone Joint Surg Br. 2001;83(5):655-658.

Romanian Neurosurgery (2025) XXXIX (1): pp. 42-45
DOI: 10.33962/roneuro-2025-006
www.journals.lapub.co.uk/index.php/roneurosurgery

A case report of a large gluteal schwannoma with pelvic extension

Nenad Lalović[1,2], Ognjen Čančar[1,2], Nikolina Dukić[2,3], Dragana Sokolović[2,4], Jelena Vladičić[2,3], Vuk Aleksić[1,2,5]

[1] Department of Surgery, University Hospital Foča, Bosnia and Herzegovina
[2] Faculty of Medicine; University of East Sarajevo, Foča, Bosnia and Herzegovina
[3] Department of Oncology, University Hospital Foča, Bosnia and Herzegovina
[4] Department of Pharmacology, University Hospital Foča, Bosnia and Herzegovina
[5] Department of Neurosurgery, Clinical Hospital Centre Zemun, Belgrade, Serbia

Keywords
schwannomas, peripheral nerves, sciatica

Corresponding author:
Vuk Aleksić

Department of Neurosurgery, Clinical Hospital Centre Zemun, Belgrade, Serbia

aleksicvuk@hotmail.com

Abstract

Benign schwannomas are slow-growing, painless tumours originating from Schwann cells, which form the sheaths of peripheral nerves. These tumours are relatively rare, with an incidence of 1-3 cases per 100,000 individuals annually. While they are most commonly found in the head, neck, and spine, schwannomas can also occur in the extremities, particularly in the upper limbs. Tumours in the pelvis and gluteal region are less frequent but are clinically significant due to their potential to compress adjacent structures.

We present a case of a 74-year-old male patient who underwent surgery for a large schwannoma in the gluteal region, extending into the pelvis at the level of the piriformis muscle and sciatic foramen. Initially misdiagnosed and treated as sciatica, this case highlights the importance of considering schwannomas in the differential diagnosis of patients presenting with neurological symptoms in unusual locations. Sciatic symptoms that do not respond to conservative treatment should be further investigated, and a thorough palpation of tender and painful points should always be performed to aid in the potential diagnosis of a local soft tissue tumour.

Scan to access the online version

Introduction

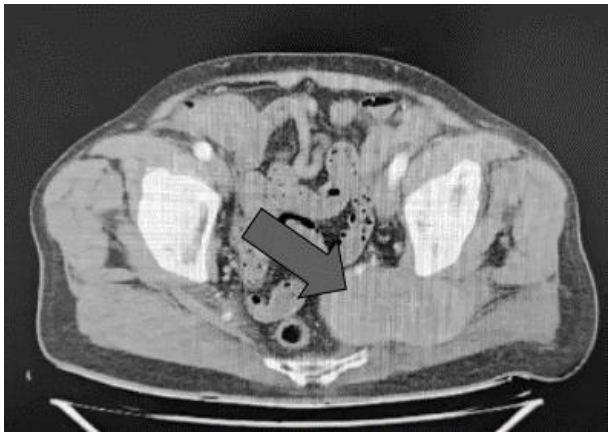
Benign schwannomas are slow-growing, painless tumors that originate from Schwann cells, which form the sheaths of peripheral nerves. These tumors are most commonly observed in females between their 20s and 40s. While the typical size does not exceed 6 cm in diameter, cases with larger tumors, reaching up to allmost 30 cm, have also been documented (1). Schwannomas frequently occur in the region of the head, neck and spine, while the localization of these tumors in the extremities is somewhat less common (2). Schwannomas are more

frequently found in the superficial layers of the body along peripheral nerve locations. However, they can also develop in deeper, less visible regions like the retroperitoneum or mediastinum. In such hidden locations, diagnosing schwannomas is often postponed due to the lack of noticeable clinical symptoms until the tumor grows significantly larger, leading to compression of intra-abdominal and intra-pelvic structures (1, 3, 4). The majority of schwannomas are asymptomatic; however, they may lead to functional impairments and pain, depending on the specific nerve involved (5). In locations where schwannomas can grow asymptomatically for years, clinical symptoms typically appear when exceptionally large tumors begin to compress intra-abdominal or pelvic organs (1).

We present a 74-year-old male patient with a massive gluteal schwannoma extending into the pelvis in the projection of the piriformis muscle.

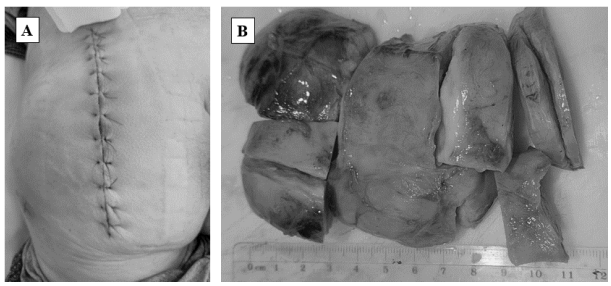
CASE REPORT

The case presents a 74-year-old patient who sought medical attention due to pain in the gluteal region and difficulty sitting, accompanied by numbness in the left leg. The patient had previously undergone several months of treatment for lumbosciatic symptoms, which did not respond well to the prescribed therapy. Due to the emergence of symptoms, including difficulty and pain during sitting and since deep palpation of the gluteal region revealed a painful mass, while deep abdominal palpation was painless and unremarkable, a computed tomography (CT) scan of the abdomen and pelvis was performed. The scan revealed an extensive well defined tumor measuring approximately 15 x 8 x 5 cm in the right gluteal region, with extension into the pelvis, predominantly in the region of the left piriformis muscle and sciatic foramen (picture 1). The tumour appeared hyper-vascular. The radiological diagnosis was a mass of neurogenic origin, most likely the sciatic nerve and suggested neurofibroma or schwannoma, while our initial clinical diagnosis of a soft tissue sarcoma involving the left gluteal region and pelvis was considered. Due to the clear visualization on the CT scan and the patient's pronounced claustrophobia, it was decided not to proceed with further radiological evaluation, specifically not to perform an magnetic resonance imaging (MRI).



Picture 1. A CT scan of the pelvis, with an arrow marking a large, well-defined tumor, which is partly located in the pelvis and partly in the gluteal region, in the projection of the piriformis muscle and the sciatic foramen.

A two-stage surgery was planned, with the first stage involving the resection of the tumor from the gluteal region. The patient was operated on by a team consisting of a general surgeon, orthopedic surgeon, and neurosurgeon. The first stage of the surgery involved a linear incision in the gluteal region (Picture 2A), in the projection of the sciatic nerve. Through careful dissection, which included both blunt and sharp techniques, the tumor was removed in one large piece (Picure 2B), and there was no need for a second surgical stage. It was impossible to trace the originating nerve. The patient recovered well, without neurological deficits, and three months post-surgery, there were no signs of tumor recurrence on the follow-up CT scan. The patient remained symptom-free, with a normal neurological examination. Histopathology confirmed the diagnosis of a schwannoma, and further clinical follow-up was recommended.



Picture 2. (A) The image shows a linear incision in the left gluteal region in the projection of the sciatic nerve, one day after surgery. **(B)** The image shows the tumor removed in its entirety, and then cut by the pathologist for further histopathological examination.

Discussion

Schwannomas are relatively uncommon benign tumors, with an estimated incidence of 1-3 cases per 100,000 individuals annually. These tumors typically arise from Schwann cells in peripheral nerves and are most commonly found in the head, neck, and spine. In terms of extremity schwannomas, these tumors are more commonly observed in the upper limbs, though they can also be found in the lower extremities. These schwannomas generally present as slow-growing, well-circumscribed, and mobile masses, often palpable under the skin. Schwannomas located in the pelvis and gluteal region are much less frequent but are clinically significant due to their potential to cause compression of adjacent structures (6, 7). Although these tumors are more commonly observed in female patients at a younger age, typically between the 2nd and 4th decades of life, we present a rare case of schwannoma in a 74-year-old male patient, with a specific localization in the gluteal region, pelvis, and in the projection of the piriformis muscle and sciatic foramen. The tumor is of considerable size, indicating that it has been asymptomatic for a relatively long period.

While the exact pathogenesis of schwannomas remains unclear, their development may be associated with specific gene mutations. Schwannomas of peripheral nerves are typically well-defined, encapsulated masses that are usually round and connected to the nerve. These tumors originate from Schwann cells and are often located eccentrically, affecting one or two fascicles while leaving the other neural fascicles of the nerve intact and displaced. Schwannomas form within the endoneurium and are encased by the perineurium and fibrous epineurium, which collectively surround and encapsulate the tumor (8). In the case of retroperitoneal, pelvic, sacral, and gluteal schwannomas, the average growth rate of the tumor is about 2 mm per year, e.g. schwannomas have a slow growth rate. Additionally, these schwannomas display strong concealment abilities due to the large pelvic space, and thus are associated with a range of clinical symptoms, such as compression of pelvic organs or nerve tissue, bone destruction, sciatica, lower back pain, difficulty with urination and defecation due to bladder and rectal compression, and weakness in the lower limbs (8, 9). In our patient, the size of the tumor suggests that it was likely asymptomatic for several years. Symptoms then developed, which were initially interpreted as sciatica. However, as the patient did not respond to standard therapy after several months, a follow-up examination was performed, during which a painful palpable mass was found in the gluteal region. This led to further radiological evaluation.

Computed tomography (CT) and magnetic resonance imaging (MRI) are commonly employed imaging modalities for assessing soft tissue tumors. However, CT's diagnostic utility is limited by its relatively low resolution and suboptimal soft tissue contrast, often failing to clearly depict the stromal heterogeneity characteristic of schwannomas. Enhanced resolution can often be achieved with the use of intravenous contrast agents, such as iopamino. MRI, when available, is considered the preferred imaging technique (10). In our case, CT imaging clearly revealed a large, well-defined, and demarcated tumor in the left gluteal region, extending into the pelvis at the level of the sciatic foramen. The tumor's clear delineation, without surrounding soft tissue reaction, raised suspicion of a schwannoma, although our initial diagnostic assumption was a sarcoma. Due to the clear visualization on CT and the patient's claustrophobia, we opted not to perform an MRI. The subsequent course of action was surgical treatment. Initially misdiagnosed and treated as sciatica, this case highlights the importance of considering schwannomas in the differential diagnosis of patients presenting with neurological symptoms in unusual locations. Sciatic symptoms that do not respond to conservative treatment should be further investigated, and a thorough palpation of tender and painful points should always be performed to aid in the potential diagnosis of a local soft tissue tumor, subsequently followed up with the necessary radiological diagnostics.

Complite surgical removal is the treatment of choice for of large schwannomas. Some authors recommend wide local excision for retroperitoneal and pelvic schwannomas, believing that malignancy can never be entirely ruled out. However, tumor recurrence or malignant transformation is extremely rare in benign schwannomas (1, 11). Successful surgical resection of retroperitoneal or pelvic schwannomas requires thorough and meticulous preoperative planning. The patient should be well-informed about the potential risks of residual

functional impairment, which may arise from nerve damage or muscle fibrosis. Challenges in tumor excision often stem from the tumor's proximity to surrounding neurovascular structures and its blood supply. Ideally, the surgical team should include different specialists such as neurosurgeon, general surgeon, vascular surgeon, and urologyst. The anesthesiologist should also have experience managing procedures with a high risk of significant blood loss. Surgery in multiple stages is often used for similar tumors (1). In our case, the clinical diagnosis initially suggested a sarcoma, but radiological evaluation, specifically the CT scan, led to the conclusion that the tumor was most likely a schwannoma. As a result, a team consisting of a neurosurgeon, general surgeon, and orthopedic surgeon was assembled, along with an experienced anesthesiology team, and a two-stage surgical approach was planned. However, through a combination of sharp and blunt dissection, the tumor was completely removed via a posterior approach in a single stage. The nerve origin was not identified, even though preoperative suspicion pointed to the sciatic nerve. The patient's preoperative symptoms completely resolved, and the neurological signs, primarily the tingling in the left leg, were most likely a result of compression on the sciatic nerve.

Although large schwannomas of the same location have been described in the literature, in our case, the tumor was one of the largest schwannomas of this location reported so far.

References

1. Ohene-Yeboah M, Adjei E, Stalsberg H. A giant guteal schwannoma with extension into the pelvis: a case report. West Afr J Med. 2009;28(3):185-188.
2. Prelić J, Radojević M, Mihajlović M, Ćulafić R, Aleksić V. Ulnar nerve schwanomma: Case report. Materia medica. 2021;37(3):2081-2085.
3. Maleux G, Brys P. Sampson I, Sciot R, Baert AL. Giant schwannoma of the lower leg. Eur. Radio. 1997;7:1031-1034.
4. Herrington JL, Edwards LW. Massive retroperitoneal neurilemoma, with emphasis on technical problems encountered during surgical removal. Surgery. 1965;57:366-369.
5. Aleksić V, Prelić J, Radojević M, Ćulafić R. Schwannoma of the digital nerve. A rare case report. Romanian neurosurgery. 2021;35(4):490-492.
6. Majumder A, Ahuja A, Chauhan DS, Paliwal P, Bhardwaj M. A clinicopathological study of peripheral schwannomas. Med Pharm Rep. 2021;94(2):191-196.
7. Kumar R, Ranjan R, Jeyaraman M, Kumar S. Tibial Nerve Schwannoma: An Unexplained Cause of Lateral Foot Pain - A Rare Case Report and Review. J Orthop Case Rep. 2020;10(9):1-6
8. Zhou S, Wan S, Li L, Dong W, Ma X, Chu H, Zhong Y. Rare retroperitoneal giant sacral schwannoma: A case report. Oncol Lett. 2024;27(6):261.
9. Tahta A, Dinc C, Ozdenkaya Y, Cakir A. Giant sacral schwannoma causing bilateral hydronephrosis: Case report and review of the literature. World Neurosurg. 2020;142:184–187.
10. Hide IG, Baudouin CI, Murray SA, Malcolm AJ. Giant ancient schwannoma of the pelvis. Skeletal Radio. 2000;29: 538–542.
11. Pinson W, Remine G , Fletcher W,Braasch J. Long-term results withprimary retroperitoneal tumors. ArchSurg. 1989;124: 1168–1173.

Romanian Neurosurgery (2025) XXXIX (1): pp. 46-48
DOI: 10.33962/roneuro-2025-007
www.journals.lapub.co.uk/index.php/roneurosurgery

Brown spinal tumour secondary to primary hyperparathyroidism, a primary entity that we should not forget. Case report

Luis Avellaneda[1], Sofia Roberto Vargas[2], Nicolas Melo[3], Santiago V. Puerta[4], Jorge Humberto Aristizabal[5]

[1] Neurosurgery Resident, Department of Neurosurgery, Universidad del Bosque, Bogotá, COLOMBIA.
[2] Primary Care Physician, Department of Neurosurgery, Universidad del Bosque, Bogotá, COLOMBIA
[3] Primary Care Physician, School of Medicine, Universidad de La Sabana, Chía, COLOMBIA
[4] Neurosurgeon, Department of Neurosurgery, Fundación Cardio Infantil, Bogotá, COLOMBIA
[5] Neurosurgeon, Department of Neurosurgery, Fundación Cardio Infantil, Bogotá, COLOMBIA

Keywords
hyperparathyroidism, osteitis fibrosa cystica, parathyroid hormone, parathyroid neoplasms, sacrum

Corresponding author:
Nicolas Melo Sierra

School of Medicine,
Universidad de La Sabana,
Chía, Colombia

drnicolasmelo@gmail.com

ABSTRACT

Introduction: A thorough study of lytic lesions can guide us toward a more accurate aetiology in spinal column lesions. The use of imaging techniques and blood chemistry studies is useful when considering brown tumours as part of the differential diagnosis.

Clinical Case: A 43-year-old woman with multiple comorbidities presented with lumbar pain. A lytic lesion in the body of S2 was documented, and thoracoabdominal lesions were initially ruled out. However, during her stay, a malignant thyroid lesion was documented.

Discussion: Hyperparathyroidism, whether primary (adenomas, hyperplasia, or carcinoma) or secondary (vitamin D deficiency or chronic kidney disease), causes skeletal alterations in approximately 16% of cases. Among its manifestations are parathyroid tumours, which primarily affect the pelvis, rib arches, facial bones, and long bones, with less frequency in the spine. Clinically, these present progressive pain and neurological deficits. Diagnosis requires paraclinical tests and imaging studies. Treatment should address the underlying cause of hyperparathyroidism, with options ranging from medical therapies to surgical interventions.

Conclusion: Thyroid carcinoma as a manifestation of a brown tumour at the sacral level is a rare presentation. We consider it pertinent to perform extension studies when evaluating patients with multiple comorbidities and a single lytic lesion.

Scan to access the online version

INTRODUCTION

Brown tumor (BT) refers to the presence of one or multiple bone lesions, usually with a lytic appearance. This is generally due to metabolic conditions caused by alterations in parathyroid hormone

(PTH) levels, with primary hyperparathyroidism (PHPT) and secondary hyperparathyroidism (SHPT) being the only two causes described so far (1). It is considered that most cases of BT are primarily caused by PHPT with an incidence of up to 13%, compared to SHPT cases which account for less than 5% of the incidence (2,3).

Elevated serum PTH levels are considered to increase osteoclast activity, causing increased bone resorption and decreased mineralization, resulting in changes in trabecular bone, fibrous cystic osteitis, and hemorrhages with hemosiderin deposits, which gives it a brownish appearance; hence its name brown tumor (3, 4).

The following presents the case of a woman with a lytic bone lesion in the sacrum (S2), which was initially considered malignant in etiology. However, given the paraclinical and imaging findings, the possibility of a BT secondary to renal failure in hemodialysis was considered. Nevertheless, upon re-evaluating the case and performing extension studies, it was evident that the patient had a parathyroid lesion, later identified as a thyroid carcinoma. Therefore, the final diagnosis was BT due to PHPT.

Clinical case

A 43-year-old female with a history of tertiary hyperparathyroidism, stage V chronic kidney disease on hemodialysis, hypertension, and chronic heart failure presented to the emergency department with six days of severe lumbosacral axial pain, without signs of radicular compression. Physical examination revealed normal strength (5/5) in all four extremities, preserved sensation, muscular atrophy, hypotonia, and an antalgic gait.

A simple magnetic resonance imaging of the sacrococcygeal spine revealed a multilocular cystic lesion on the left lateral aspect at the level of S2. Additionally, PTH levels were found to be 2062.9 pg/ml, serum calcium was 9.5 mg/dl, and serum phosphorus was 7.2 mg/dl. Due to these findings, and considering the patient's history and symptoms, the initial possibility was a tertiary hyperparathyroidism due to mediastinal parathyroid adenomas and chronic kidney disease. Therefore, treatment was initiated with Sevelamer 800 g orally every 8 hours and vitamin D 5600 IU orally daily; parathyroidectomy was not possible due to an ongoing infectious process. Furthermore, due to the absence of spinal cord compression and pathological fractures, surgical management of the spine was not required, and multimodal analgesic management was the alternative implemented.

Discussion

Hyperparathyroidism, whether of primary origin related to single or multiple adenomas, hyperplasia or carcinoma; or of secondary origin due to vitamin D deficiency or chronic kidney disease, causes skeletal system alterations in approximately 16% of cases (5). Among its manifestations, BT stands out, with an incidence of less than 5% in PHPT and approximately 15% in SHPT (5, 6), mainly affecting the pelvis, costal arches, facial bones and long bones; while a lower prevalence is observed in the spine, mainly affecting the thoracic and lumbar regions, and only four cases have been reported at the sacral level (5, 7, 8, 9).

From a clinical standpoint, vertebral BT commonly presents with progressive pain and neurological deficits, which can manifest as alterations in strength, sensation, and even impaired sphincter control; depending on the location of the lesion and the presence of spinal cord compression and pathological fractures. Likewise, it is necessary to perform paraclinical tests, such as measuring serum hormone levels, calcium and phosphorus, in addition to imaging, with the aim of providing an accurate diagnosis and distinguishing vertebral BT from its main mimics, which include giant cell tumor, giant cell reparative granuloma, multiple myeloma, and metastatic lesions (6). In our case, it correlates with the clinical presentation described in the literature when there is involvement of the S2 nerve roots.

Regarding the paraclinical results, it is common to observe elevated calcium and PTH levels, associated with low phosphorus levels (5). On the other hand, among the useful diagnostic images, there is the X-ray, where one or more well-defined and expansive lesions are usually evident; while in the computed tomography, bone erosion can be evidenced. For its part, magnetic resonance imaging allows visualizing hypointense lesions in the T1 sequence and hypo or hyperintense in the T2 sequence, with or without signs of spinal cord compression (6). Finally, bone scintigraphy shows diffuse systemic hypermetabolism. Although these examinations have advantages, their reliability is not absolute, so it is recommended to take a biopsy of the lesion, where multinucleated osteoclasts, spindle-shaped

stromal cells and a fibrous matrix can be observed (10). Correlating it with the presented case, changes in biochemical parameters were evidenced associated with extremely high serum hormonal levels, which does not agree with the expected results, but, when analyzing the case, it is determined that these findings are given by the history of chronic renal failure with hemodialysis requirement. On the other hand, the diagnostic images coincide with the descriptions found in the literature (Figure 1).

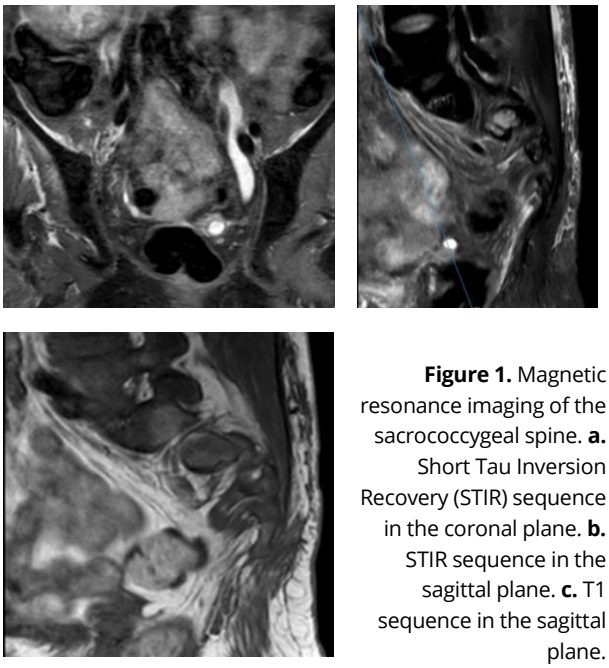


Figure 1. Magnetic resonance imaging of the sacrococcygeal spine. **a.** Short Tau Inversion Recovery (STIR) sequence in the coronal plane. **b.** STIR sequence in the sagittal plane. **c.** T1 sequence in the sagittal plane.

Regarding BT treatment, management should be etiological, addressing the underlying cause of hyperparathyroidism. In primary cases, phosphate binders, vitamin D, and calcimimetics are used, while in secondary cases, parathyroid surgery is employed (9, 11). This approach demonstrates positive outcomes, with normalization of calcium and PTH levels, as well as improvement in bone density and healing of pathological fractures (10). Specifically in spinal lesions, the surgical option may include tumor resection or decompression, depending on the size, location, and degree of associated neurological deficit (5).

Conclusion

In conclusion, primary TP should be considered in pathologies related to PTH homeostasis, such as HPTP and HPTS. The importance of distinguishing its origin lies in the fact that treatment varies, ranging from pharmacological management to surgical intervention of the primary tumor and TP. It is essential to conduct further studies on this entity to expand the bibliographic base and, ultimately, standardize its management.

Abbreviations

BT Brown tumor
PHPT Primary hyperparathyroidism
PTH Parathyroid hormone
SHPT Secondary hyperparathyroidism
STIR Short Tau Inversion Recovery

References

1. Yokota N, Kuribayashi T, Nagamine M, Tanaka M, Matsukura S, Wakisaka S. Paraplegia caused by brown tumor in primary hyperparathyroidism: Case report. Journal of Neurosurgery. 1989;71(3):446–8.
2. Tayfun H, Metin O, Hakan S, Zafer B, Vardar A. Brown tumor as an unusual but preventable cause of spinal cord compression: Case report and review of the literature. Asian J Neurosurg. 2014;9(01):40–4.
3. Alfawareh MD, Halawani MM, Attia WI, Almusrea KN. Brown Tumor of the Cervical Spines: A Case Report with Literature Review. Asian Spine J. 2015;9(1):110.
4. Khalatbari MR, Moharamzad Y. Brown Tumor of the Spine in Patients With Primary Hyperparathyroidism: Spine. 2014;39(18):E1073–9.
5. Altan L, Kurtoğlu Z, Yalçınkaya U, Aydınlı U, Ertürk E. Brown tumor of the sacral spine in a patient with low-back pain. Rheumatol Int. 2007;28(1):77–81.
6. Hu J, He S, Yang J, Ye C, Yang X, Xiao J. Management of brown tumor of spine with primary hyperparathyroidism: A case report and literature review. Medicine. 2019;98(14):e15007.
7. Araújo SMHA, Bruin VMS, Nunes AS, Pereira ENS, Mota ACM, Ribeiro MZD, et al. Multiple brown tumors causing spinal cord compression in association with secondary hyperparathyroidism. Int Urol Nephrol. 2013;45(3):913–6.
8. Tarrass F, Ayad A, Benjelloun M, Anabi A, Ramdani B, Benghanem MG, et al. Cauda equina compression revealing brown tumor of the spine in a long-term hemodialysis patient. Joint Bone Spine. 2006;73(6):748–50.
9. Muppidi V, Meegada SR, Rehman A. Secondary Hyperparathyroidism. In: StatPearls. Treasure Island (FL): StatPearls Publishing; 2024. Available from: http://www.ncbi.nlm.nih.gov/books/NBK557822/
10. Hoshi M, Takami M, Kajikawa M, Teramura K, Okamoto T, Yanagida I, et al. A case of multiple skeletal lesions of brown tumors, mimicking carcinoma metastases. Arch Orthop Trauma Surg. 2008;128(2):149–54.
11. Pokhrel B, Leslie SW, Levine SN. Primary Hyperparathyroidism. In: StatPearls. Treasure Island (FL): StatPearls Publishing; 2024. Available from: http://www.ncbi.nlm.nih.gov/books/NBK441895/.

Romanian Neurosurgery (2025) XXXIX (1): pp. 49-55
DOI: 10.33962/roneuro-2025-008
www.journals.lapub.co.uk/index.php/roneurosurgery

Symptomatic relief of pain following percutaneous vertebroplasty compared to conservative management in patients with osteoporotic vertebral compression fractures. A prospective cohort study from a low-middle-income country

Pradhumna Kumar Yadav[1,2,5], Ahtesham Khizar[1,3], Bushra Tariq Ali[1,2], Hammad Mehtab[1,4]

[1] Pakistan Institute of Medical Sciences (PIMS), Islamabad, PAKISTAN
[2] Shaheed Zulfiqar Ali Bhutto Medical University (SZABMU), Islamabad, PAKISTAN
[3] Punjab Institute of Neurosciences, Lahore, PAKISTAN
[4] Fauji Foundation Hospital, Rawalpindi, PAKISTAN
[5] National Trauma Centre, Kathmandu, NEPAL

Keywords
osteoporosis, osteoporotic fractures, vertebroplasty

Corresponding author:
Ahtesham Khizar

Department of Neurosurgery
Punjab Institute of Neurosciences, Lahore, Pakistan

arwain.6n2@gmail.com

ABSTRACT

Objectives: To determine the efficacy of percutaneous vertebroplasty in pain management over conservative management in patients with osteoporotic vertebral compression fractures.

Materials and Methods: A prospective cohort study was conducted from 13th December 2018 to 12th December 2019 at PIMS/SZABMU, Islamabad, Pakistan. A total of 76 patients (Aged: 35-75 years) of both genders having osteoporotic vertebral compression fractures involving a maximum of two vertebrae were enrolled. Patients were divided equally into two groups. One group was managed surgically through vertebroplasty (Group A) and the other group was managed conservatively (Group B). All the patients were asked about the intensity of pain, assessed by Visual Analogue Scale (VAS) score at the presentation and after 24 hours, 3rd and 6th week of given treatment and compared using independent sample t-test in both groups. Complications were also assessed and compared in both groups.

Results: In group A, dorsal vertebrae were involved in 23.7%, lumbar vertebrae in 68.4% and dorsal/lumbar vertebrae in 7.9% of cases. In group B, dorsal vertebrae were involved in 21.1%, lumbar vertebrae in 60.5% and dorsal/lumbar vertebrae in 18.4% of cases. At baseline, mean VAS was 8.01 ± 0.99 in group A and it was 8.35 ± 0.75 in group B. At 24 hours after the intervention, mean VAS was 4.37 ± 0.79 in group A and it was 7.29 ± 1.21 in group B. At 3 weeks after the intervention, mean VAS was 4.03 ± 0.85 in group A and it was 6.37 ± 0.91 in group B and at 6 weeks after the intervention, mean VAS was 3.87 ± 1.09 in group A and it was 5.37 ± 0.97 in group B. The overall complication rate was 10.5% at 24 hours in group A and it was 5.3% in group B. At 3 weeks, the complication rate was 5.3% in group A and it was 28.9% in

Scan to access the online version

group B. At 6 weeks, the complication rate was 21.1% in group A and it was 55.3% in group B patients.

Conclusion: Mean VAS score was found to be significantly lower in patients who underwent vertebroplasty as compared to those managed conservatively. Overall complication rate was similar in both groups at 24 hours; however, it was significantly lower at 3rd and 6th weeks in patients who underwent vertebroplasty as compared to those managed conservatively.

INTRODUCTION

Osteoporosis is one of the most common diseases of elderly and post-menopausal women characterised by fragile bone with increased susceptibility to fracture. The fracture may be at multiple skeletal sites, most commonly involving the spine, hip and wrist.[1] Osteoporotic vertebral compression fractures (OVCF) are the most common type of osteoporotic fracture accounting about 1.4 million worldwide every year.[2,3] Studies have shown that among individuals above 50 years of age about one fourth will sustain at least one vertebral fracture over lifetime.[4] They are considered to be low energy fractures which are more frequently seen in post-menopausal women and its prevalence increases with age in both genders.[5]

Osteoporotic vertebral compression fractures result in serious health concerns such as pain, disability and often mortality[6] and so the quality of life is markedly decreased in patients with OVCF. Conservative management such as rest, immobilisation, analgesics, bisphosphonates have been widely used but remain ineffective.[7] Furthermore, if a patient is of old age, bed rest and decreased activity leads to associated pneumonia, decubitus ulcer, venous thromboembolism and even death.[8] So, considering all these disadvantages, Percutaneous Vertebroplasty (PVP) is considered to be another alternative. It is minimally invasive procedure, which involves injection of cement (polymethylmethacrylate, PMMA) into the OVCF resulting in immediate pain relief.[3] Vertebroplasty was initially used in the treatment of hemangioma about 25 years ago and with time it is being used in osteoporotic fractures starting from late 1990's.

According to published data, there is considerable relief of pain following PVP compared to conservative treatment for OVCF.[4] Given the scenario of high rate of morbidity associated with non-operative management, PVP seems to be the best alternative for not only the pain management but also for return of functional independence, decreased rate of readmission, and fewer admission to skilled nursing or long term care facilities.

MATERIALS AND METHODS

Study design: Prospective cohort study.

Setting: Department of Neurosurgery, Pakistan Institute of Medical Sciences (PIMS)/Shaheed Zulfiqar Ali Bhutto Medical University (SZABMU), Islamabad**,** Pakistan.

Duration of Study: One year (13th December, 2018 to 12th December, 2019).

Sample Size: Calculated by WHO sample size calculator with the following parameters:

- Sample size (n)=76 i.e, 38 in each group
- Level of significance = 5%
- Power of test=80%
- Test value of population (Mean value of VAS score after 24hrs in PVP group) p1: 4.7
- Anticipated population (Mean value of VAS score after 24hrs in conservative treatment group) p2: 7.1

Sampling Technique: Non-probability based consecutive sampling.

Sample Selection

Inclusion Criteria

1. Patients of both genders (Age: 35-75 years) having osteoporotic vertebral compression fracture.
2. Osteoporotic vertebral compression fracture involving maximum of two vertebrae.
3. VAS score at presentation ≥4.

Exclusion Criteria

1. Patients who did not give consent.
2. Patients having osteoporotic vertebral fractures involving more than two vertebrae.
3. Patients who had untreatable coagulopathy.
4. Patients who had spinal cord compression syndrome.
5. Patients having any pre-existing infection at the surgical site.

Data Collection Procedure

This study was conducted after taking approval from the hospital's ethical review committee. Patients admitted to Neurosurgery ward who had osteoporotic vertebral compression fracture as

evidenced by thorough clinical examination, X-rays and Computed Tomography (CT), Magnetic Resonance Imaging (MRI), dual-energy x-ray absorptiometry (DEXA) scan, serum calcium and Vitamin D3 levels were included in the study. Informed written consent was taken from each patient. Initial data about age, sex, contact number and date of admission were recorded on predesigned proforma. Patients were divided into two groups. One group was managed conservatively and the other was operated via vertebroplasty. Vertebroplasty involves the percutaneous injection of bone cement under image guidance into a fractured vertebra whereas conservative management includes pain control and activity modification. Oral analgesics are first-line therapy for the relief of acute pain. Options include acetaminophen, ibuprofen, naproxen, mild opioids combined with acetaminophen, or mixed mechanism drugs (e.g, tramadol, tapentadol), centrally acting analgesics whose mode of action is based both on mu-opioid receptor binding and monoamine (serotonin and norepinephrine) reuptake blockade. Both the groups were asked about the severity of pain which was assessed by VAS score at the presentation and after 24 hours, 3rd week and 6th week of given treatment. Any complication associated with the drugs such as dizziness, headache, gastric upset was noted. Also the complications associated with vertebroplasty such as cement leakage, adjacent fracture, infection were noted in the subsequent follow up.

Data Analysis Procedure

Collected data was analysed with SPSS version 26. Mean and standard deviation were calculated for numerical variables like, age and pain score. Frequency and percentages were presented for categorical variables like, gender and complications in both groups. Independent sample t-test was used to compare pain between both groups. Chi-square test was applied to compare complication rate between both groups. P-value <0.05 was considered significant.

Results

Demography of the selected population

A total of seventy six (n=76) patients (Age: 35-75 years) of both genders having osteoporotic vertebral compression fracture involving a maximum of two vertebrae were enrolled into this study. All the enrolled patients had moderate degree of pain (VAS≥4). Patients were divided equally (n=38 in each group) into two groups. One group was managed surgically through vertebroplasty procedure (Group A) and the other group was managed conservatively (Group B). Gender distribution was similar in both groups. There were 39.5% (n=15/38) males and 60.5% (n=23/38) females in group A and 36.8% (n=14/38) males and 63.2% (n=24/38) females in group B (Table. 1). Age distribution was also similar in both groups. Mean age of Group A patients was 62.3 years ± 13.4 SD while it was 65.8 years ± 15.1 SD in group B patients (Table. 2).

Baseline patient characteristics in both groups

In group A patients, dorsal vertebrae were involved in 23.7% (n=9/38), lumbar vertebrae in 68.4% (n=26/38) and both dorsal/lumbar vertebrae in 7.9% (n=3/38) of cases. In group B patients, dorsal vertebrae were involved in 21.1% (n=8/38), lumbar vertebrae in 60.5% (n=23/68) and both dorsal/lumbar vertebrae in 18.4% (n=7/38) of cases (Table. 3). Analysis of comorbid conditions at baseline revealed that comorbids were present in 63.2% (n=24/48) of cases in group A and 63.2% (n=24/48) of cases in group B patients (Table. 4). At baseline, mean VAS was 8.01 ± 0.99 in group A and it was 8.35 ± 0.75 in group B patients (Table. 5).

Mean VAS Score at different time intervals:

Mean VAS score was estimated at different time intervals. At 24 hours after the intervention, mean VAS was 4.37 ± 0.79 in group A and it was 7.29 ± 1.21 in group B (p=0.001, Table. 5), at 3 weeks after the intervention, mean VAS was 4.03 ± 0.85 in group A and it was 6.37 ± 0.91 in group B (P=0. 001, Table. 4) and at 6 weeks after the intervention, mean VAS was 3.87 ± 1.09 in group A and it was 5.37 ± 0.97 in group B (P=0. 001, Table. 5). Mean VAS was found to be significantly lower at all time intervals in patients who underwent vertebroplasty procedure (group A) as compared to those managed conservatively (group B).

Complication rate in both groups:

Overall complication rate was 10.5% (n=4/38) (cement leakage) at 24 hours in group A patients and it was 5.3% (n=2/38) (2 DVT) in group B patients (P=0.395, Table. 6). At 3 weeks, complication rate was 5.3% (n=2/38) in group A patients (DVT, sleep disorder [mainly insomnia]) and it was 28.9%

(n=11/38) in group B patients (5 pressure sores, 3 pneumonia, 2 depression, 1 DVT) (P=0.006, Table. 6). At 6 weeks, complication rate was 21.1% (n=8/38) in group A patients (3 UTI, 2 pneumonia, 2 sleep disorder [mainly insomnia], 1 DVT) and it was 55.3% (n=17/38) in group B (6 pneumonia, 5 pressure sores, 4 constipation, 2 depression) patients (P=0.002, Table. 6). Overall complication rate was similar (P>0.05) in both groups at 24 hours, however, it was significantly lower (P<0.05) at 3 and 6 weeks in patients who underwent vertebroplasty procedure (group A) as compared to those managed conservatively (group B).

Table 1. Gender distribution in both the study groups.

Gender	Groups		Total	P-value
	Vertebro-plasty	Conser-vative		
Male	15 (39.5%)	14 (36.8%)	29 (38.2%)	0.813329
Female	23 (60.5%)	24 (63.2%)	47 (63.2%)	
Total	38 (100%)	38 (100%)	76 (100%)	

Table 2. Age distribution in both the study groups

Groups	n	Mean Age ± SD (years)
Vertebroplasty	38	62.3 ± 13.4
Conservative	38	65.8 ± 15.1

Table 3. Vertebrae involved in both the study groups

Diagnosis	Groups		Total	P-value
	Vertebro-plasty	Conser-vative		
Dorsal	9 (23.7%)	8 (21.1%)	17 (22.4%)	0.398022
Lumbar	26 (68.4%)	23 (60.5%)	49 (64.5%)	
Dorsal + Lumbar	3 (7.9%)	7 (18.4%)	10 (13.2%)	
Total	38 (100%)	38 (100%)	76 (100%)	

Table 4. Baseline Comorbidities in both the study groups

Comorbidities	Groups		Total
	Vertebro-plasty	Conser-vative	
None	14 (36.8%)	14 (36.8%)	28 (36.8%)
Hypertension	12 (31.6%)	16 (42.1%)	28 (36.8%)
Diabetes	4 (10.5%)	0 (0%)	4 (5.3%)
*HTN + DM	8 (21.1%)	5 (13.2%)	13 (17.9%)
*HTN + DM + IHD	0 (0%)	3 (7.9%)	3 (7.9%)
Total	38 (100%)	38 (100%)	76 (100%)

*DM: Diabetes Mellitus, HTN: Hypertension, IHD: Ischemic Heart Disease

Table 5. Mean VAS at different time intervals in both groups

VAS	Groups	Mean VAS ± SD	P-value t-test
Baseline	Vertebroplasty	8.01 ± 0.99	0.101
	Conservative	8.35 ± 0.75	
24 Hours	Vertebroplasty	4.37 ± 0.79	0.001
	Conservative	7.29 ± 1.21	
3 Weeks	Vertebroplasty	4.03 ± 0.85	0.001
	Conservative	6.37 ± 0.91	
6 Weeks	Vertebroplasty	3.87 ± 1.09	0.001
	Conservative	5.37 ± 0.97	

Table 6. Complications at 24 hours in both the study groups

Compli-cations	Groups		Total	P-Value Chi-square
	Vertebro-plasty	Conser-vative		

24 Hours	Present	4 (10.5%)	2 (5.3%)	6 (7.9%)	0.395
	Absent	34 (89.5%)	36 (94.7%)	70 (92.1%)	
3 Weeks	Present	2 (5.3)	11 (28.9%)	13 (17.1%)	0.006
	Absent	36 (94.7%)	27 (71.1%)	63 (82.9%)	
6 Weeks	Present	8 (21.1%)	21 (55.3%)	29 (38.2%)	0.002
	Absent	30 (78.9%)	17 (44.7%)	47 (61.8%)	

DISCUSSION:

Given the scenario of high rate of morbidity associated with non-operative management, PVP has been reported as the best alternative to conservative management for the management of OVCF for not only pain management but also for return of functional independence, decreased rate of readmission and fewer admission to skilled nursing or long term care facilities. In the current study, we determined the efficacy of PVP in pain management over conservative management in patients with OVCF. The baseline characteristics of patients in both the groups were comparable. Our study showed that patients who underwent PVP had greater pain relief in terms of VAS score at 24 hours, 3 and 6 weeks compared to conservative management. Per-operative fluoroscopic imaging of Vertebroplasty of one of our cases is shown in Fig. 1 A&B.

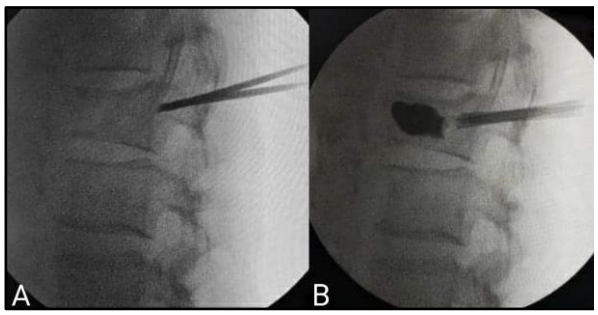


Figure 1. A&B: Per-operative fluoroscopic imaging of Vertebroplasty at L1 vertebral level.

Our study results are similar with other reports in the literature. Mattie R et al.[9] in their meta-analysis compared PVP with conservative treatment for the management of osteoporotic compression fractures in terms of pain relief. They had included eleven (n=11) trials comprising 1048 subjects. Patients treated with PVP (n=531) showed significantly lower intensity of pain when compared with patients treated conservatively at 1 to 2 weeks, 2 to 3 months, and 12 months. They concluded that the pain relieving effect of PVP exceeded the effect of conservative management in patients with osteoporotic compression fractures up to one year after the treatment. Our study showed similar results upto 6 weeks of follow up. We did not follow our patients up to one year. In fact several other reports showed similar results in studies by Liu et al.[10], Xie et al.[11], Luo W et al.[12], Lou S et al.[13], Zuo et al.[14], Zuo RS[15] and Chen LX[16]. In the pooled analysis by Zuo XH et al.[14], they evaluated the efficacy and safety in percutaneous vertebroplasty for osteoporotic vertebral compression fractures in comparison with conservative treatment, kyphoplasty and nerve block. They included eighteen (n=18) trials comprising 1994 patients. They concluded that both vertebroplasty and kyphoplasty had better performance than conservative management in terms of short and long-term pain relief.

However, randomised controlled trials by Kallmes et al.[17] and Buchbinder et al.[18] showed that vertebroplasty offered no benefit over sham procedure/simulated procedure over varying time intervals in terms of pain relief, physical function and quality of life. However, the sham/simulated procedure offered in these studies are difficult to achieve in surgical practice and ethically unacceptable. Furthermore, Samuel Butler[19] stated that these two studies had samples of fewer than 300 patients in both the vertebroplasty and control groups, and that many patients were unwilling to accept randomisation, particularly those in severe pain. As a result of the small sample size and selection bias, these studies were deemed untrustworthy. Further, if we observe the statistical trend in the above studies, PVP shows higher rate of clinical improvement of pain compared to conservative or sham treatment.

In Cochrane review of twenty-one trials, Buchbinder R et al.[20,21] compared benefits and side effects of vertebroplasty for treatment of osteoporotic vertebral fractures with placebo (sham), conservative care or some other

intervention. There were five trials that compared vertebroplasty with placebo (n=541 subjects), eight trials comparing vertebroplasty with conservative care (n=1136 subjects), seven trials comparing it with kyphoplasty (n=968 patients) and one trial was included that compared vertebroplasty with facet joint glucocorticoid injection (n=217 patients). Authors found no remarkable benefit in terms of pain relief when compared with a sham procedure and conservative care. The heterogeneity of included data could not be ignored and warrants a large-scale single randomised controlled trial.

Zhang L et al.[22] in another meta-analysis summarised current best evidence on the efficacy of PVP and conservative treatment (CT) for pain management and functional results among OVCFs patients. Their analysis revealed that PVP had benefits on pain relief at 1 week and 1 month, but not at 3 months along with improved quality of life, without increasing the incidence of vertebral fracture compared with the CT group. Wang D et al.[23] recently designed a clinical trial comparing the efficiency and safety of vertebroplasty versus conservative treatment for acute OVCFs. The primary outcome was pain relief at 1 month and 1 year, measured with the VAS score. The preliminary results showed vertebroplasty provides a rapid decrease of pain and an early return to daily life activities compared with the control group. However, detailed results are yet awaited. The mechanism of pain relief that occurs within minutes to hours after vertebroplasty remains unknown. Following the injection of the cement, pain pathways in the surrounding tissue appear to be altered in response to mechanical, chemical, vascular, and thermal stimuli.[24]

In summary, results of the present study and bulk of evidence cited in the literature suggest that percutaneous vertebroplasty has been consistently showing better results in terms of pain relief associated with osteoporotic vertebral compression fractures when compared with conservative treatment. Present study has some limitations. Firstly, the sampling technique can result in some bias and the sample size was relatively smaller, yet sufficient enough for interpretation. Nonetheless, it is not wise to extend our results to the general population; a larger sample size is needed for that purpose. Secondly, we did not use a placebo (sham) procedure. Thirdly, we did not follow our patients beyond 6 weeks and hence, were not able to evaluate long-term efficacy and risk of subsequent fractures associated with the procedure. We suggest future studies addressing these limitations taking larger sample size and taking into account the placebo effect, longer durations of follow up and comparison with other treatment modalities for pain relief like, nerve block and kyphoplasty.

Conclusion

Mean VAS score was found to be significantly lower ($P<0.05$) in patients who underwent vertebroplasty procedure as compared to those managed conservatively. Overall complication rate was similar ($P>0.05$) in both groups at 24 hours, however, it was significantly lower ($P<0.05$) at 3rd and 6th weeks in patients who underwent vertebroplasty procedure as compared to those managed conservatively.

List of abbreviations

VAS: Visual Analogue Scale
OVCF: Osteoporotic vertebral compression fractures
PVP: Percutaneous Vertebroplasty
PMMA: Polymethylmethacrylate
CT: Computed Tomography
MRI: Magnetic Resonance Imaging
DEXA: Dual-energy x-ray absorptiometry
DVT: Deep Vein Thrombosis

References

1. Savage JW, Schroeder GD, Anderson PA. Vertebroplasty and kyphoplasty for the treatment of osteoporotic vertebral compression fractures. J Am Acad Orthop Surg. 2014;22(10):653-664. doi:10.5435/JAAOS-22-10-653
2. Goldstein CL, Chutkan NB, Choma TJ, Orr RD. Management of the Elderly With Vertebral Compression Fractures. Neurosurgery. 2015;77 Suppl 4:S33-S45. doi:10.1227/NEU.0000000000000947
3. Guo JB, Zhu Y, Chen BL, et al. Surgical versus non-surgical treatment for vertebral compression fracture with osteopenia: a systematic review and meta-analysis. PLoS One. 2015;10(5):e0127145. doi:10.1371/journal.pone.0127145
4. Mattie R, Laimi K, Yu S, Saltychev M. Comparing Percutaneous Vertebroplasty and Conservative Therapy for Treating Osteoporotic Compression Fractures in the Thoracic and Lumbar Spine: A Systematic Review and Meta-Analysis. J Bone Joint Surg Am. 2016;98(12):1041-1051. doi:10.2106/JBJS.15.00425
5. Ruiz Santiago F, Santiago Chinchilla A, Guzmán Álvarez L, Pérez Abela AL, Castellano García Mdel M, Pajares López M. Comparative review of vertebroplasty and kyphoplasty. World J Radiol. 2014;6(6):329-343. doi:10.4329/wjr.v6.i6.329
6. Zhao S, Xu CY, Zhu AR, et al. Comparison of the efficacy

and safety of 3 treatments for patients with osteoporotic vertebral compression fractures: A network meta-analysis. Medicine (Baltimore). 2017;96(26):e7328. doi:10.1097/MD.0000000000007328

7. Yuan WH, Hsu HC, Lai KL. Vertebroplasty and balloon kyphoplasty versus conservative treatment for osteoporotic vertebral compression fractures: A meta-analysis. Medicine (Baltimore). 2016;95(31):e4491. doi:10.1097/MD.0000000000004491
8. Yang EZ, Xu JG, Huang GZ, et al. Percutaneous Vertebroplasty Versus Conservative Treatment in Aged Patients With Acute Osteoporotic Vertebral Compression Fractures: A Prospective Randomized Controlled Clinical Study. Spine (Phila Pa 1976). 2016;41(8):653-660. doi:10.1097/BRS.0000000000001298
9. Mattie R, Laimi K, Yu S, Saltychev M. Comparing Percutaneous Vertebroplasty and Conservative Therapy for Treating Osteoporotic Compression Fractures in the Thoracic and Lumbar Spine: A Systematic Review and Meta-Analysis. J Bone Joint Surg Am. 2016;98(12):1041-1051. doi:10.2106/JBJS.15.00425
10. Liu J, Li X, Tang D, et al. Comparing pain reduction following vertebroplasty and conservative treatment for osteoporotic vertebral compression fractures: a meta-analysis of randomized controlled trials. Pain Physician. 2013;16(5):455-464.
11. Xie L, Zhao ZG, Zhang SJ, Hu YB. Percutaneous vertebroplasty versus conservative treatment for osteoporotic vertebral compression fractures: An updated meta-analysis of prospective randomized controlled trials. Int J Surg. 2017;47:25-32. doi:10.1016/j.ijsu.2017.09.021
12. Luo W, Cui C, Pourtaheri S, Garfin S. Efficacy of Vertebral Augmentation for Vertebral Compression Fractures: A Review of Meta-Analyses. Spine Surg Relat Res. 2018;2(3):163-168. doi:10.22603/ssrr.2017-0089
13. Lou S, Shi X, Zhang X, Lyu H, Li Z, Wang Y. Percutaneous vertebroplasty versus non-operative treatment for osteoporotic vertebral compression fractures: a meta-analysis of randomized controlled trials. Osteoporos Int. 2019;30(12):2369-2380. doi:10.1007/s00198-019-05101-8
14. Zuo XH, Zhu XP, Bao HG, et al. Network meta-analysis of percutaneous vertebroplasty, percutaneous kyphoplasty, nerve block, and conservative treatment for nonsurgery options of acute/subacute and chronic osteoporotic vertebral compression fractures (OVCFs) in short-term and long-term effects. Medicine (Baltimore). 2018;97(29):e11544. doi:10.1097/MD.0000000000011544
15. Zhu RS, Kan SL, Ning GZ, et al. Which is the best treatment of osteoporotic vertebral compression fractures: balloon kyphoplasty, percutaneous vertebroplasty, or non-surgical treatment? A Bayesian network meta-analysis. Osteoporos Int. 2019;30(2):287-298. doi:10.1007/s00198-018-4804-2
16. Chen LX, Li YL, Ning GZ, et al. Comparative efficacy and tolerability of three treatments in old people with osteoporotic vertebral compression fracture: a network meta-analysis and systematic review. PLoS One. 2015;10(4):e0123153. doi:10.1371/journal.pone.0123153
17. Kallmes DF, Comstock BA, Heagerty PJ, et al. A randomized trial of vertebroplasty for osteoporotic spinal fractures. N Engl J Med. 2009;361(6):569-579. doi:10.1056/NEJMoa0900563
18. Kallmes D, Buchbinder R, Jarvik J, et al. Response to "randomized vertebroplasty trials: bad news or sham news?". AJNR Am J Neuroradiol. 2009;30(10):1809-1810. doi:10.3174/ajnr.A1887
19. Munk PL, Liu DM, Murphy KP, Baerlocher MO. Effectiveness of vertebroplasty: a recent controversy. Can Assoc Radiol J. 2009;60(4):170-171. doi:10.1016/j.carj.2009.08.001
20. Buchbinder R, Johnston RV, Rischin KJ, et al. Percutaneous vertebroplasty for osteoporotic vertebral compression fracture. Cochrane Database Syst Rev. 2018;11(11):CD006349. doi:10.1002/14651858.CD006349.pub4
21. Buchbinder R, Osborne RH, Ebeling PR, et al. A randomized trial of vertebroplasty for painful osteoporotic vertebral fractures. N Engl J Med. 2009;361(6):557-568. doi:10.1056/NEJMoa0900429
22. Zhang L, Zhai P. A Comparison of Percutaneous Vertebroplasty Versus Conservative Treatment in Terms of Treatment Effect for Osteoporotic Vertebral Compression Fractures: A Meta-Analysis. Surg Innov. 2020;27(1):19-25. doi:10.1177/1553350619869535
23. Wang D, Cang D, Wu Y, Wang S. Therapeutic effect of percutaneous vertebroplasty and nonoperative treatment on osteoporotic vertebral compression fracture: A randomized controlled trial protocol. Medicine (Baltimore). 2020;99(27):e20770. doi:10.1097/MD.0000000000020770
24. Medical Advisory Secretariat. Percutaneous vertebroplasty for treatment of painful osteoporotic vertebral compression fractures: an evidence-based analysis. Ont Health Technol Assess Ser. 2010;10(19):1-45. PMID: 23074396

Romanian Neurosurgery (2025) XXXIX (1): pp. 56-60
DOI: 10.33962/roneuro-2025-009
www.journals.lapub.co.uk/index.php/roneurosurgery

Outcome of endovascular treatment of giant aneurysm. A retrospective study

Rahul Singh, Surendra Jain, Ashok Gandhi, Suresh Kumar Choudhary, Hariom Meena

Department of Neurosurgery, SMS Medical College, Adarsh Nagar, Jaipur, Rajasthan, INDIA

ABSTRACT

Introduction: Very large and giant aneurysms are among the most challenging cerebrovascular pathologies in neurosurgery. The risk of aneurysmal rupture compounds with an increase in the size of the aneurysm thus warranting appropriate intervention.

Objective: To analyze the outcome of endovascular treatment of giant aneurysm.

Materials and Methods: A retrospective study was conducted at the Department of Neurosurgery. 35 cases were selected from the database with radiological diagnoses of giant aneurysm referred to our departments from 2016 to 2023.

Results: The patients mainly belonged to >60 years age group (37.14%) and had a mean age of 49 years, with a slight female preponderance (57.14%). Aneurysms were mainly located in patients who had left internal carotid artery (ICA) supraclinoid aneurysms (14.29%) and right middle carotid artery (MCA) bifurcation aneurysms (14.29%). The patients predominantly underwent simple coiling (45.7%) and stent-assisted coiling (42.8%). Of 35 patients, 2 (5.7%) died. The recurrence was higher in posterior circulation aneurysms with, 75% in basilar top aneurysms (3 out of 4 cases), 50% in posterior communicating artery aneurysms (one out of 2 cases), 37.9% in remaining cases (11 out of 29 patients).

Conclusion: Giant aneurysm is associated with reasonably high morbidity and mortality. The aneurysms are found most often in the anterior circulation, while the recurrence is mainly observed in the posterior circulation have more chances of recurrence. However, favourable outcome was frequently observed (94.3%).

Keywords
intracranial giant aneurysm,
clipping,
coiling,
bypass,
flow diverter

Corresponding author:
Rahul Singh

SMS Medical College,
Adarsh Nagar, Jaipur, Rajasthan,
India

rahuladvance89@gmail.com

INTRODUCTION

Giant intracranial aneurysms (GIAs), defined as greater than 25 mm, are rare intracranial lesions. Giant cerebral aneurysms account for ~5% of all intracranial aneurysms. [1-3] They occur more commonly in the 5th-7th decades and are more common in females. [2] Patients can present with symptoms and signs of mass effect or subarachnoid hemorrhage. [1,2] GIAs are typically saccular in shape, though they can also be fusiform or serpentine in morphology. [1] They are thought to develop via two pathways: internal elastic lamina *de novo* defect and enlargement from a smaller aneurysm. [2]

Compared to non-giant cerebral aneurysms, GIAs are more commonly located in the posterior circulation, with an incidence of around 35%. [3]

The appearance can vary depending on whether the aneurysm is non-thrombosed, partially thrombosed, or completely thrombosed. Radiographically, non-thrombosed GIAs appear as well-defined, slightly hyperdense, round extra-axial masses, and may demonstrate a peripheral calcified rim. [2]

The treatment of GIAs includes both endovascular and open surgical techniques, with endovascular options generally associated with lower morbidity. [3] Available therapeutic approaches include reconstructive techniques (such as clipping, coiling, stent-assisted coiling, and flow diversion) and deconstructive techniques (including parent artery occlusion, sometimes combined with bypass surgery, and flow modification strategies). [4] Given the poor natural course of GIAs, aggressive treatment is often recommended to achieve both aneurysm occlusion and relief of mass effect. [5,6] However, the risks of treatment must be carefully weighed against the potential benefits.

Reconstructive techniques are usually the preferred treatment strategy for intracranial aneurysms, as these procedures preserve the patency of the parent vasculature and maintain cerebral blood flow distal to the aneurysm. [7-9] Direct surgical clip ligation of the neck with preservation of the parent vasculature remains the ideal reconstructive treatment strategy in the majority of very large and giant vascular aneurysms. On the other hand, coil embolization has proven ineffective in the treatment of very large and giant aneurysms. Intravascular coil embolization has significant limitations in wide-necked aneurysms due to comparably lower packing densities and subsequently higher rates of recanalization. [10]

Deconstructive treatments are considered only when reconstructive methods are not feasible or would result in unacceptable morbidity. The present study aims to describe the characteristics of patients with giant intracranial aneurysms and to assess treatment outcomes.

Material and methods

This retrospective study reviewed electronic medical records of patients diagnosed with GIAs (≥25 mm) from 2016 to 2023 at the Trauma Centre, Department of Neurosurgery, in a tertiary care hospital. The study included patients with radiological diagnoses confirmed by digital subtraction angiography (DSA) or computed tomography angiography (CTA) and underwent endovascular treatment. Only patients with available pre- and post-operative DSA and computed tomography (CT) scan (from either our institution or external sources) and those undergoing their first surgical intervention were included. Exclusion criteria included a history of trauma or iatrogenic injury, poor prognosis conditions (e.g., severe heart disease, cancer), connective tissue disorders, and patients lost to follow-up. Patients with incomplete or erroneous clinical, radiological, or surgical data were also excluded. Necessary permission was obtained from the Institutional Ethics Committee and patient consent was waived due to the retrospective nature of the study.

Participants and assessment

This study involved the review of 35 cases of GIA with data collected on patient demographics, clinical presentation, aneurysm rupture, and aneurysm configuration, as well as treatment modalities and outcomes. Diagnostic imaging, including DSA, magnetic resonance imaging (MRI), CT scan, and CTA was independently analyzed by a neuroradiologist and a neurosurgeon. Treatment decisions were based on the location and morphology of the aneurysms, and procedures such as DSA, coiling, stent-assisted coiling, balloon-assisted coiling, and flow diversion were performed as appropriate. All patients completed regular follow-up assessments. Outcomes were evaluated using the Glasgow Outcome Scale (GOS), with a good outcome defined as a final GOS of 4 or 5, and a poor outcome as a final GOS of less than 4.

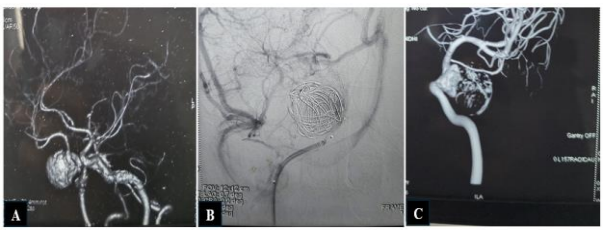


Figure 1. Digital subtraction angiography of giant basilar top aneurysm. Pre-operative (A), post-operative stent assisted coiling of a patient showing occlusion of aneurysm (B), and at 1 year illustrating residual filling of the aneurysm.

Statistical analyses

Descriptive statistics were used.

Results

The mean age of the study population was 49 years (range, 22–66 years). The patients were mainly in the >60 years age group (37.14%), with slight female preponderance (57.14%).

The majority of patients had left internal carotid artery (ICA) supraclinoid aneurysms (14.29%) and right middle carotid artery (MCA) bifurcation aneurysms (14.29%). While the least common locations were right ICA cavernous giant segment aneurysms (5.71%), and others (2.86%) (Table 2).

The patients predominantly underwent simple coiling (45.7%) and stent-assisted coiling (42.8%) (Table 3).

Of 35 patients, 2 (5.7%) died. One had a right middle cerebral artery bifurcation giant aneurysm and underwent simple coiling. The other had a ruptured left supraclinoid giant aneurysm and underwent balloon-assisted coiling. Additionally, two patients (5.7%) experienced weakness in the left upper limb, and one patient (2.8%) had weakness in both the left upper and left lower limbs. Additionally, one patient (2.8%) developed left eye ptosis, while another patient (2.8%) experienced right eye enophthalmos. The recurrence rate was higher in posterior circulation aneurysm with, 75% in basilar top aneurysm (3 out of 4 cases), 50% in posterior communicating artery aneurysm (one out of 2 cases), 37.9% in remaining cases (11 out of 29 patients) (Table 4).

Table 1. Demographic characteristics

Characteristics	Number
Sex, n (%)	
Male	15 (42.85%)
Female	20 (57.14%)
Mean age, years	49
Age, n (%)	
<30 years	7 (20.00%)
31- 50 years	8 (22.86%)
51-60 years	7 (20.00%)
>60 years	13 (37.14%)

Table 2. Diagnosis of patients with aneurysm

Diagnosis, n (%)	n (=35)
Left ICA aneurysm	4 (11.43%)
Right ICA aneurysm	3 (8.57%)
RICA Cavernous Giant Segment Aneurysm	2 (5.71%)
Right ICA Supraclenoid giant aneurysm	4 (11.43%)
Right MCA Bifurcation aneurysm	5 (14.29%)
Left ICA Supraclenoid giant aneurysm	5 (14.29%)
Left ICA Cavernous Giant Aneurysm	1 (2.86%)
Right ophthalmic Artery Giant aneurysm	1 (2.86%)
Right PCOM artery aneurysm	1 (2.86%)
Left ophthalmic Artery giant aneurysm	1 (2.86%)
Left PCOM artery aneurysm	1 (2.86%)
Giant ACOM aneurysm	4 (11.43%)
Baislar Top Giant Aneurysm	3 (8.57%)

ACOM: Anterior communicating artery, ICA: Internal carotid artery, PCOM: Posterior communicating artery

Table 3. Treatment of patients with aneurysm

Treatment, n (%)	n (=35)
Simple coiling	16 (45.7%)
Stent assisted Coiling	15 (42.8%)
Flow diverter	3 (8.5%)
Balloon Assisted Coiling	1 (2.8%)

Table 4. Outcome of patients with aneurysm

Outcome	Number
Dead	2 (5.7%)
Left Upper limb weakness	2 (5.7%)
Left Upper limb + Left lower limb weakness	1 (2.8%)
Left eye ptosis	1 (2.8%)
Right eye enophthalmos	1 (2.8%)

Discussion

Giant cerebral aneurysms have a poor natural history, with high risk of subarachnoid hemorrhage or progressive symptoms of mass effect. Several endovascular techniques may be applied for treatment, depending on location, size, anatomy and presence of collateral circulation. The authors reviewed clinical experience of 35 very large and giant aneurysms and presented their perspective on the present state of the outcome in endovascular therapy for these aneurysms.

In our study, patients who were treated for Giant aneurysm had a mean age of 49 years (range, 22–66 years). Majority of patients belonged to 51- 60 year age group with female preponderance (57.14%). This data was supported by study of Chalouhi et al., [11] and Dutta et al. [12] where 64.3% were female with a mean age of 47.8 years. A meta-analysis of nearly 4000 patients found that aneurysm growth was associated with both increasing age and female sex. [13] The risk of developing a brain aneurysm increases with age, particularly after 40 years, likely due to the progressive weakening of blood vessel walls over time as they endure the constant pressure

of blood flow. Although aneurysms generally affect males more than females, with a male-to-female ratio of approximately 4:1, females tend to experience worse outcomes. Available literature suggests that ruptured aneurysms occur in females more frequently and also at smaller diameters compared to males. [1]

In our study, the majority of patients had left ICA supraclinoid aneurysms and right MCA bifurcation aneurysms. Similarly, Serbinenko et al. found giant aneurysm in internal carotid artery. [14] Similar study states that these lesions are found most often in the anterior circulation, affecting the ICA, MCA, and ACA, [1,2] while in the posterior circulation, they most commonly occur at the basilar artery, vertebrobasilar junction, PCA, and PICA. [2]

Large and giant aneurysms can be managed with various treatment options like open surgery, endovascular therapy, or a combined approach. Although endovascular modalities like stenting, coiling, and flow diverters have favorable clinical and angiographic outcomes, surgical clipping continues to be the first management option in experienced hands. In our study, maximum patients i.e. 45.7% underwent simple coiling, followed by 42.8% cases who underwent stent assisted coiling. Others had balloon assisted occlusion and FD.

Treatment strategy is planned by assessing factors such as age, comorbidities, size, location, morphology, projection of the aneurysm, neck-to-dome ratio, aneurysmal characteristics such as thrombosis and calcification, collateral circulation, and the presence of critical perforating vessels arising from the aneurysm wall. [1,12]

Among all patients, two died, in which 1 had right MCA bifurcation giant aneurysm who underwent simple coiling. Other one had ruptured left supraclenoid giant aneurysm who underwent Balloon assisted coiling. Sughrue et al., in their study of 140 patients with 141 giant aneurysms treated surgically, reported a mortality rate of 13%, morbidity of 9%, and a favorable outcome in 81% of cases. [15] Similarly, Sharma et al. reported a mortality rate of 9%, and a morbidity rate of 12% for giant aneurysms treated surgically, with favorable outcomes in 86% of patients. [16] Our study also had comparable results with a favorable outcome in 91% with 9% mortality.

Recurrence rate was higher in posterior circulation aneurysm with, 75% in basilar top aneurysm (3 out of 4 cases), 50% in posterior communicating artery aneurysm (one out of 2 cases), 37.9% in remaining cases (11 out of 29 patients)

Conclusion

Giant aneurysms are associated with reasonably low morbidity and mortality. Cases reported in our institution occurred in the 5^{th}-7^{th} decades and are more common in females. These lesions are found most often in the anterior circulation, affecting the ICA and MCA. Recurrence rate is higher in posterior circulating aneurysm, maximum with basilar top aneurysm (75%), DSA remains a "gold standard" in the diagnosis and treatment planning of giant aneurysms. Endovascular coiling was frequently used as treatment technique for large and giant aneurysms, especially in the setting of subarachnoid hemorrhage. Our study had a favorable outcome in 94.3%. Moving forward, there is a critical need for further research into newer treatment modalities, such as flow diverters, which may offer improved outcomes for managing giant aneurysms.

Abbreviations

DSA	Digital subtraction angiography
ICA	Internal Carotid Artery
MCA	Middle Carotid Artery
GIA	Giant intracranial aneurysm

Acknowledgements

Authors would like to thank Dr. Vikas S. Sharma (MD), CEO, Maverick Medicorum® (India), for medical writing services.

References

1. UCAS Japan Investigators; Morita A, Kirino T, Hashi K, Aoki N, Fukuhara S, Hashimoto N, et al. The natural course of unruptured cerebral aneurysms in a Japanese cohort. N Engl J Med. 2012;366(26):2474-82. https://doi.org/10.1056/NEJMoa1113260
2. Sundt TM Jr, Piepgras DG. Surgical approach to giant intracranial aneurysms. Operative experience with 80 cases. J Neurosurg. 1979;51(6):731-42. https://doi.org/10.3171/jns.1979.51.6.0731
3. Kalani MY, Zabramski JM, Hu YC, Spetzler RF. Extracranial-intracranial bypass and vessel occlusion for the treatment of unclippable giant middle cerebral artery aneurysms. Neurosurgery 2013;72:428–35, discussion 435–36. https://doi.org/10.1227/NEU.0b013e3182804381

4. Van Rooij WJ, Sluzewski M. Endovascular treatment of large and giant aneurysms. AJNR Am J Neuroradiol 2009;30:12–18. https://doi.org/10.3174/ajnr.A1267
5. Roy D, Milot G, Raymond J. Endovascular treatment of unruptured aneurysms. Stroke 2001;32:1998–2004. https://doi.org/10.1161/hs0901.095600
6. Chalouhi N, Jabbour P, Singhal S, Drueding R, Starke RM, Dalyai RT, et al. Stent-assisted coiling of intracranial aneurysms: predictors of complications, recanalization, and outcome in 508 cases. Stroke 2013;44:1348–53. https://doi.org/10.1161/STROKEAHA.111.000641
7. Altman DG. Practical Statistics for Medical Research. Boca Raton, Florida: Chapman & Hall/CRC; 1999. https://doi.org/10.1201/9780429258589
8. Parkinson RJ, Eddleman CS, Batjer HH, Bendok BR. Giant intracranial aneurysms: endovascular challenges. Neurosurgery. 2008;62(6 Suppl 3):1336-45. https://doi.org/10.1227/01.neu.0000333798.67209.1f
9. Gruber A, Killer M, Bavinzski G, Richling B. Clinical and angiographic results of endosaccular coiling treatment of giant and very large intracranial aneurysms: a 7-year, single-center experience. Neurosurgery. 1999;45(4):793-803. https://doi.org/10.1097/00006123-199910000-00013
10. Sluzewski M, Menovsky T, van Rooij WJ, Wijnalda D. Coiling of very large or giant cerebral aneurysms: long-term clinical and serial angiographic results. AJNR Am J Neuroradiol 2003;24:257–62. PMID: 12591644; PMCID: PMC7974128.
11. Chalouhi N, Tjoumakaris S, Gonzalez LF, Dumont AS, Starke RM, Hasan D, et al. Coiling of large and giant aneurysms: complications and long-term results of 334 cases. AJNR Am J Neuroradiol. 2014;35(3):546-52. https://doi.org/10.3174/ajnr.A3696
12. Dutta G, Singh D, Jagetia A, Srivastava AK, Singh H, Kumar A. Endovascular management of large and giant intracranial aneurysms: Experience from a tertiary care neurosurgery institute in India. J Cerebrovasc Endovasc Neurosurg. 2021;23(2):99-107. https://doi.org/10.7461/jcen.2021.E2020.08.008
13. Brinjikji W, Zhu YQ, Lanzino G, Cloft HJ, Murad MH, Wang Z, Kallmes DF. Risk Factors for Growth of Intracranial Aneurysms: A Systematic Review and Meta-Analysis. AJNR Am J Neuroradiol. 2016;37(4):615-20. https://doi.org/10.3174/ajnr.A4575
14. Serbinenko FA, Filatov JM, Spallone A, Tchurilov MV, Lazarev VA. Management of giant intracranial ICA aneurysms with combined extracranial-intracranial anastomosis and endovascular occlusion. J Neurosurg 1990;73:57–63. https://doi.org/10.3171/jns.1990.73.1.0057
15. Sughrue ME, Saloner D, Rayz VL, Lawton MT. Giant intracranial aneurysms: evolution of management in a contemporary surgical series. Neurosurgery. 2011;69(6):1261-70; discussion 1270-1. https://doi.org/10.1227/NEU.0b013e31822bb8a6
16. Sharma BS, Gupta A, Ahmad FU, Suri A, Mehta VS. Surgical management of giant intracranial aneurysms. Clin Neurol Neurosurg. 2008;110(7):674-81. https://doi.org/10.1016/j.clineuro.2008.04.001

Romanian Neurosurgery (2025) XXXIX (1): pp. 61-65
DOI: 10.33962/roneuro-2025-010
www.journals.lapub.co.uk/index.php/roneurosurgery

Spontaneous extradural hematoma in sickle cell anaemia-a stroke mimic. A report of two cases

Adegboye Olakunle Michael, Ahmed Mahmud, Adeleke Nurudeen Abiola, Enaworu Oghenevwoke, Nnara Stanley Onyeka, Wahab Kolawole Wasiu, Odebode Timothy Olugbenga

Division of Neurosurgery, Department of Surgery, University of Ilorin Teaching Hospital, Ilorin, Kwara State, NIGERIA

ABSTRACT

In the absence of trauma, extradural hematoma is rarely considered a cause of hemiplegia in a sickle cell disease patient, rather a cerebrovascular accident, as this occurs in about a quarter of sickle cell disease patients. We report two sickle cell anaemia patients who were initially diagnosed as cases of stroke having presented with hemiplegia/hemiparesis without a prior history of trauma to the head. Cranial computed tomographic scans however revealed extradural hematomas and both of them underwent surgical evacuation of the hematomas with subsequent neurologic recovery.

Keywords
CT scan, hemiplegia, sickle cell anaemia, spontaneous extradural hematoma, stroke

✉

Corresponding author:
Adegboye Olakunle Michael

University of Ilorin Teaching Hospital, Ilorin, Kwara State, Nigeria

omadegboye@gmail.com

INTRODUCTION

Sickle cell disease (SCD) is a hematological condition with multi-systemic manifestations, including central nervous system. Neurological complications are among the most devastating manifestations of the disease and these include stroke, vascular malformations and cranial neuropathies.[1,2] Jeffrey reported that as many as 11% of patients with SCD will develop stroke by age 20 years while the incidence increases to 24% by age 45 years.[3]

Spontaneous extradural hematoma (EDH) is one of the underrecognized complications of sickle cell disease and rarely considered a cause of limb weakness.[4] Delay in making diagnosis of extradural hematoma could be dangerous as there could be expansion of the hematoma with subsequent coning and death. While ischemic and most times, haemorhagic stroke may not be amenable to surgical procedures but rather a long time of physical therapy which may leave the patients with significant residual deficit, surgical evacuation of extradural hematoma may have dramatic post-operative recovery once the offending compressive lesion is removed.

CASE PRESENTATION

Case 1

An 18-year-old right-handed known sickle cell anemia (SCA) male patient diagnosed in childhood who presented to medical emergency room with a 3-day history of generalized headache, weakness of left upper and lower limbs and altered sensorium of two days. He had no history of seizure, vomiting and no preceding history of trauma to the head. He was being managed by the medical team as a case of right hemispheric stroke.

A cranial computerized tomography (CT) scan done however showed biconvex hyperdense right parietal mass lesion (measuring 78 mls) with a significant midline shift (12mm) and effacement of ipsilateral lateral ventricle, suggestive of an extradural hematoma with mass effect and overlying subgaleal hematoma (figure 1A-B). There was no overlying skull fracture. These radiological findings necessitated a consult to our unit (Neurosurgical team).

Upon our review, his Glasgow coma score was 9 and he had left spastic hemiplegia and a right sided parietal minimal diffuse scalp swelling. He was afebrile, anicteric but pale. His packed cell volume (PCV) was 24% (his stable PCV was 23-25%), platelet count and clotting profile were within normal limit.

He underwent emergency right sided parietal craniotomy and evacuation of the hematoma under general anaesthesia. Intraoperative findings were minimal subgaleal clotted blood and massive parietal extradural hematoma, compressing the underlying dura and brain (figure 2A-C). The bone flap appeared grossly normal with punctuate bleeding from the craniotomy edges.

Postoperatively, he made significant improvement, with full regain of consciousness within 2 days and gradual improvement of power in the left upper and lower limbs. On 12th day post - operative day, power had improved to 4 (from pre-operative power of 0) in the left upper and lower limbs. He was then discharged home for outpatient physiotherapy and follow up at the neurosurgery clinic. A clinic follow-up at three weeks post-operative revealed full power in all the limbs and he had no complaints. A repeat cranial CT scan at five weeks post-operative revealed no residual or recurrent extradural hematoma. (Figure 1C)

Case 2

A 10-year-old right-handed known SCA male patient (diagnosed at the age of three years), who presented to the emergency paediatric unit with history of abdominal pain, passage of dark coloured urine and weakness of right upper and lower limbs. He was pale with PCV of 18% (his stable PCV was not known). There was no antecedent history of trauma to the head or abdomen. He was diagnosed with hyperhaemolytic and vaso-occlusive crises and left hemispheric stroke. He could not do cranial CT scan due to financial constraints. He had exchange blood transfusion by the paediatric team and was discharged home after nine days of admission

He however represented after two weeks with history of headache and persistence of the weakness of the right upper and lower limbs. At this time, the mother was persuaded and he was able to do a cranial CT scan which showed a left parietal extradural hematoma and an overlying subgaleal hematoma without overlying skull fracture (figure 3A-C). Full blood count revealed adequate platelet count (206,000 cells/uL) and the clotting profile was within normal limit. A review by our team on being consulted revealed a fully conscious boy with normal pupils and right hemiparesis (power was grossly 3 in the right upper and lower limbs). He also had minimal left parietal scalp swelling.

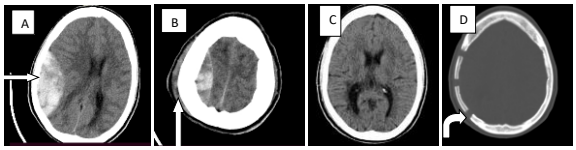


Figure 1. Pre-operative non-contrast cranial axial CT scan showing right parietal extradural hematoma with mass effect (A) and overlying subgaleal hematoma (B), fifth week post-operative cranial axial CT scan showing no residual or recurrent hematoma (C), bone window showing craniotomy burr hole sites (D).

He had surgical intervention under general anaesthesia. At surgery, we found minimal subgaleal hematoma, brittle dusky left parietal bone **(**figure 4A**)** and extradural clotted blood with lysed liquid portion (figure 4B). Due to the unhealthy nature of the bone, the parietal skull bone was excised until healthy bleeding edges (figure 4C) were reached and the hematomas were evacuated.

He made good recovery post-operatively with complete resolution of headache and gradual

resolution of the right hemiparesis. He was discharged home 10 days post-operatively with full power in all muscle groups of the limbs and residual left parietal skull defect of 8cm by 6cm size and counselled for later cranioplasty.

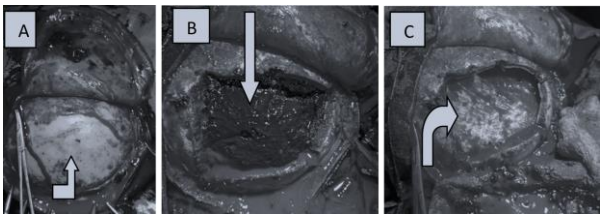


Figure 2. Intra-operative images showing healthy looking skull bone (A), massive extradural hematoma(B) and compressed dura/brain post evacuation of the hematoma (C).

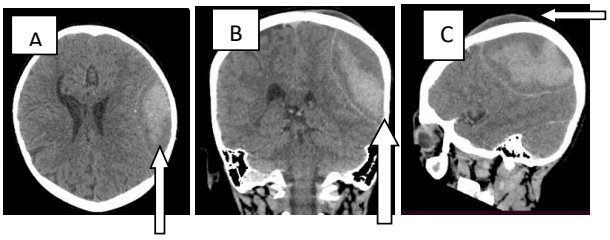


Figure 3. Non-contrast cranial axial (A) and coronal (B) CT scan showing right parietal extradural hematoma, the sagittal view (C) showed overlying subgaleal hematoma)**.**

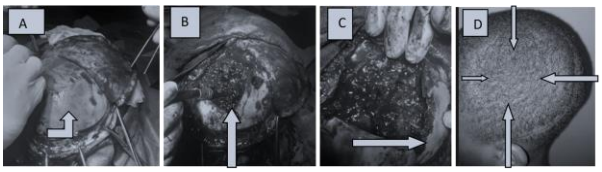


Figure 4. Intra-operative images showing dusky left parietal skull bone, suggestive of skull infarction (A), extradural hematoma(B) and bleeding bone edges, post craniectomy (C), three months post-operative clinical photograph showing left parietal skull defect with overlying scalp depression (D).

Discussion

Stroke, a significant cause of morbidity and mortality, is relatively common in sickle cell disease patients, affecting up to a quarter of these patients.[3,5] This could be in form of silent infarct, overt ischemic or hemorrhagic stroke[5–7] Overt stroke could manifest clinically with limb weakness, focal seizure, cranial nerve deficit among others.[3]

Intracranial EDH usually occurs following trauma and cases of spontaneous EDH are rare. The causes of spontaneous EDH could include coagulopathies, vascular malformations of the dura, dural/skull metastasis and infectious processes of the skull.[8] In the absence of the above aetiological factors and without history of trauma diagnosis of EDH becomes rarer. Spontaneous EDH is a rare complication in SCD patients, suspicion of its diagnosis may become submerged if the patients present with history suggestive of stroke.

Though the pathogenesis of spontaneous EDH in SCD patients has not been clearly defined, certain mechanisms have been proposed. These mechanisms include infarction of the skull bone, disruption of the diploic vein, periosteal elevation and cortical margin disruption and subsequent bleeding into either the extradural or subgaleal space or both.[9] Other suggested mechanism is that the skull could act as extramedullary hematopoietic site in SCD patients and during the period of hemolytic crisis, hyperproliferation and sudden expansion of the bone marrow in response to anemia could lead to expansion of the diploe, disruption of the diploic veins, disruption of inner and outer cortical skull margins and subsequent extravasation of blood.[10] Both of these mechanisms could explain co-existence of subgaleal and extradural hematomas in the same patient as found in the two patients presented. Although the pathogenesis of the spontaneous EDH could not be explained by any of these two mechanisms in the first patient presented, it could be argued that the finding of overlying necrosed bone flap at surgery could be explained by the infarction theory.

As a rare complication of SCD, the possibility of spontaneous EDH may be overlooked if the patients present with history suggestive of stroke. The clinical manifestation of spontaneous EDH varies. In a recent literature review in 2023 of 25 cases of spontaneous EDH by Lintz and Blum, the commonest presenting complaint was headache which occurred in 10 (40%), followed by VOC (32%), 12% of the patients presented with coma.[11] None of the 25 patients in the review presented with hemiparesis/hemiplegia, although Iversen *et al* reported a case of massive spontaneous EDH in a SCD patient who presented with headache, coma, dilated pupils and hemipareisis.[5] The index report is one of the few reports in which patients presented with limb weakness mimicking stroke.

Some pathomechanisms of spontaneous EDH in SCD patients suggest that there could be concomitant subgaleal hematoma in the same

patient. Page *et al* reported that concomitant subgaleal hematoma is found in 50% of SCD patients with spontaneous EDH.[12] The two patients in this case report had scalp hematomas. The presence of more sinister physical signs such as limb weakness may divert the attention of the attending physician from noticing a scalp swelling especially when it is subtle. We suggest that presence of scalp swelling should be specifically sought for in a SCD patient presenting with limb weakness.

The management of extradural hematoma depends on several factors, such as volume of the hematoma, presence or otherwise of mass effect, level of consciousness, presence or absence of neurologic deficit.[11] While some surgeons surgically manage these patients, some toe the pathway of conservative care. Chaurasiya *et al* opined that patients with large extradural hematoma with mass effect and significant neurologic deficit warrant surgical evacuation.[13] Both patients we presented in this report underwent surgical evacuation due to the large volume of hematoma with mass effect, presence of neurologic deficit and altered consciousness in the first patient and neurologic deficit in the second patient. Lint and Blum reported that 13 of 25 patients they reviewed were managed surgically while the remaining 12 patients had conservative care.[11]

The outcomes of patients with extradural hematoma varies, mortality ranges between 20% and 33% in the literature.[11,12,14] Prognosis largely depends on early diagnosis and promptness of management institution. Therefore, it is important to have a high index of suspicion and low threshold for requesting cranial CT scan in SCD patients who present with features suggestive of spontaneous EDH in order to avoid preventable mortality and/morbidity. Page *et al* reported a large EDH in a SCD which was only diagnosed at autopsy with features of brain herniation and infarction of the skull overlying the hematoma.[12]

Conclusion

The presence of limb weakness in sickle cell disease patient may not always be due to stroke. Unfortunately, little attention may be given to the possibility of surgically treatable extradural hematoma, especially in the absence of history of head trauma. Failure of early diagnosis of extradural hematoma and prompt surgical evacuation may lead to mortality. A search for a possible spontaneous extradural hematoma through cranial computed tomography scan should be commenced in earnest as surgical evacuation of such could be life-saving and produce good neurologic recovery.

References

1. Ahmed KA, Bello-Manga H, Jordan LC. Sickle cell anemia and early stroke detection and prevention in Nigeria. Front Stroke. 2024;(3):1368576
2. Alrayyes S, Baghdan D, Haddad RY, Compton AA, Mohama S, Goreishi R, et al. Sickle cell disease; An overview of the disease and its systemic effects. Disease-a-Month. 2018;64(6):283–89.
3. Switzer JA, Hess DC, Nichols FT, Adams RJ. Pathophysiology and treatment of stroke in sickle-cell disease : present and future. Lancet Neurol. 2006;5(6):501–12.
4. Debaun MR, Kirkham FJ. Central nervous system complications and management in sickle cell disease. Blood, J Am Soc Hematol. 2016;127(7):829–38.
5. Iversen PO, Jacob M, Makame J, Abisay M, Yonazi M, Schuh A, et al. A massive extradural hematoma in sickle cell disease and the importance of rapid neuroimaging. Case Rep Hematol. 2019;2019(1):1742472
6. Runge A, Brazel D, Pakbaz Z. Stroke in sickle cell disease and the promise of recent disease modifying agents. J Neurol Sci. 2022;442(2022):120412.
7. Light J, Boucher M, Baskin-Miller J, Winstead M. Managing the cerebrovascular complications of sickle cell disease: Current Perspectives. J Blood Med. 2023;14:279–93.
8. Babatola BO, Salman YA, Abiola AM, Okezie KO, Oladele AS. Spontaneous epidural haematoma in sickle cell anaemia: Case report and literature review. J Surg Tech Case Rep. 2012;4(2):135–37.
9. Resar LM, Oliva MM, Casella JF. Skull infarction and epidural hematomas in a patient with sickle cell anemia. Journal of pediatric hematology/oncology. 1996; 18(4):413–15.
10. Dahdaleh NS, Lindley TE, Kirby PA, Oya H, Howard MA. A "neurosurgical crisis" of sickle cell disease: Case report. J Neurosurg Pediatr. 2009;4(6):532–35.
11. Lintz VC, Blum PB. Spontaneous epidural hematoma in a patient with sickle cell anemia - Case report. Hematol Transfus Cell Ther. 2024;46:S274-77.
12. Page C, Gardner K, Height S, Rees DC, Hampton T, Thein SL. Nontraumatic extradural hematoma in sickle cell anemia: a rare neurological complication not to be missed. Am. J. Hematol. 2014;89(2):225–27.
13. Chaurasiya M, Bhardwaj S, Naik D. Can spontaneous bifrontal extradural haematoma be a sole presentation in previously undiagnosed sickle cell disease ? A rare case report and literature review. Interdiscip Neurosurg Adv Tech Case Manag. 2021;25:101230.

14. Hamm J, Rathore N, Lee P, Leblanc Z, Lebensburger J, Meier ER, et al. Cranial epidural hematomas : A case series and literature review of this rare complication associated with sickle cell disease. Pediatr Blood Cancer. 2017;64(3):1–8.

15. Shah D, Reddy H, Kumar S, Acharya S. Sickle cell disease presenting as extradural hematoma : An extremely rare fatal crisis. cureus. 2022;14(7):1–5.

Romanian Neurosurgery (2025) XXXIX (1): pp. 66-69
DOI: 10.33962/roneuro-2025-011
www.journals.lapub.co.uk/index.php/roneurosurgery

Sagittal alignment in lumbar spinal canal stenosis

Hermann Adonis N'da, Meleine Landry Konan, Kporou Landry Drogba, Joel N'guessan Brou, Dominique N'dri Oka

Service de neurochirurgie, CHU de Yopougon, Abidjan, Côte d'Ivoire

Abstract

Objective: To evaluate the impact of lumbar canal stenosis on the sagittal balance of patients by studying the parameters of sagittal alignment.

Methods: This single-centre prospective study, conducted from January 2020 to January 2022, included 47 patients presenting with lumbar canal stenosis confirmed by MRI and documented spinal balance assessed by full spine X-ray in standing profile. Patients with lumbo-radicular pain post-lumbar arthrodesis surgery were also included. The sagittal alignment parameters assessed were pelvic incidence (PI), pelvic tilt (PT), sacral slope (SS), lumbar lordosis (LL), L4-S1 lordosis (LL4-S1), PI-LL mismatch, thoracic kyphosis (TK), sagittal vertical axis (SVA), and TPA.

Results: The mean pelvic incidence (PI) was 60.97° (SD 13.93°), with Class IV incidence in 36.2% of cases. Pelvic tilt (PT) averaged 18.65° (SD 10.91°), with values <10° in 19%, 10-25° in 53%, and >25° in 28% of patients. Sacral slope (SS) was 42.87° (SD 8.49°), with 53.2% in the 35-45° range. The measured lumbar lordosis (LL) was 46.10° (SD 17.84°), significantly lower than the theoretical LL (p=0.001). L4-S1 lordosis averaged 38.85° (SD 15.34°), also less than the theoretical value (p=0.017). PI-LL mismatch >10° was present in 60% of cases. Thoracic kyphosis (TK) averaged 14.57° (SD 11.29°), and sagittal vertical axis (SVA) was >5 cm in 81% of patients. T1 pelvic angle (TPA) was <20° in 57.4%.

Conclusion: The analysis revealed that lumbar canal stenosis significantly affected spinal architecture, primarily through the loss of lumbar lordosis, and had a notable impact on thoracic kyphosis. Therapeutic approaches should not only focus on root decompression but also on restoring lumbar lordosis, with careful consideration of the arthrodesis level.

Keywords
lumbar canal stenosis,
lumbar lordosis,
pelvic incidence,
pelvic tilt,
sagittal balance

Corresponding author:
Hermann Adonis N'da

Service de neurochirurgie, CHU de Yopougon, Abidjan, Côte d'Ivoire

omadegboye@gmail.com

Scan to access the online version

Introduction

Lumbar spinal canal stenosis is one of the most degenerative spine diseases. It's the most common cause of spine surgery for people over 65 years old (4,14). Lumbar spine canal stenosis is characterized by morphological changes of the intervertebral disc, the ligamentum flavum and the facet joints. The result of these changes is the reduction of the useful diameter of the vertebral canal thus causing spine and neurogenic pain (12). Apart from neurological damage, the modifications caused by stenosis have an impact on spinal architecture.

The biomechanical organization of the spine includes the sagittal spinal alignment defined as harmonious succession sagittal spinal curvatures. (5)

There is a strong statistical link between sagittal alignment and pre or post operative quality of life (15). Spinal imbalance induces spinal pain, a source of functional disability well evaluated by the ODI (6). Maintaining or restoring optimal spinal alignment is one of the major therapeutic goals in spinal stenosis.

Lumbar canal stenosis poses the problem of the evaluation of its impact on the lumbar spinal architecture but also far from the initial lumbar site.

The aim of this study was to evaluate the impact of lumbar canal stenosis on the sagittal balance of patients by studying the parameters of sagittal alignment.

Material and method

This was a single-center prospective study carried out from January 2020 to January 2022. The inclusion criteria grouped together patients seen in neurosurgery consultation for stenosis of the lumbar canal documented by an MRI, patients whose spinal balance was documented by a full spine X-ray in standing profile as well as patients operated for lumbar canal stenosis with an arthrodesis and presenting with lumboradicular pain. According to these inclusion criteria, the sample for this study was 47 patients, M= 21, F= 26. The data was collected on a survey form guaranteeing the anonymity of the patients. It included patients sociodemographic characteristics (age, gender, occupation). The sagittal and global alignment parameters studied were pelvic incidence (PI), pelvic tilt (PT), sacral slope (SS), L1-S1 lumbar lordosis (LL), L4-S1 lordosis (LL4-S1), PI minus LL (PI-LL), thoracic kyphosis (TK), sagittal vertical axis (SVA) and TPA.

The data was analyzed using IBM SPSS 26 software. For quantitative variables, the numbers and percentages were calculated. For the qualitative variables we calculated the followed by the standard deviations and the extremes. We used Khi^2 test or the exact test of fisher to cross two qualitative variables. The difference between the measured values and the theoretical values were analyzed by the T-student test. The significance threshold was set at 5% ($p \leq 0.05$).

Results

The mean PI was of 60.97° (minus=35°, maximum=90.6°, SD=13,93°). Class IV incidence was mostly seen (36,2%). The PT was in value 18.65°+/-10.91°. Its distribution showed extreme between 1° to 45.10°. PT was less than 10 in 19%, between 10° to 25° in 53% and over than 25° in 28%. The SS was of 42,87°+/- 8.49. SS was small (less than 35°) in 17%, middle (between 35° to 45°) in 53.2% and over 45° in 29.8%. the measured LL was 46.10°+/-17.84° (13° to 94.3°). The measured LL was 21.4°±15.41° in women and 18.30°±16.70° in men (P=0.662). The theoretical LL was 67.15°±7.46° (55° to 85.6°). This LL was 68.01°±7.03° in women and 65.98°±8.02° in men (p=0.362). The theoretical LL was greater than measured LL (p=0.001). The L4-S1 lordosis was measured to 38.85°±15.34° (2.30° to 77.40°) and this lordosis was less than the theoretical one, 44.76±4.97° (p=0.017). The loss of lumbar lordosis is summarized in the table 1. PI-LL was 20.12°±16.07° (0.6° to 59.6°). PI-LL up to 10° (PI-LL>10°) was seen in 60 % of cases. The thoracic kyphosis (TK) was 14.57°±11.29° (0.8° to 50.4°). TK was less than 45° in 93.6% of cases, up to 45° in one case and equal to 45° in two cases (4.3%). The SVA was 16.01±14.12 cm (0.1 to 69.4°). It was greater than 5 cm in 81% and lower than 5 cm in 19%.

The Pelvic angle of T1 was 17.97°±11.59° (0.3 to 46.4°). TPA was less than 20° in 57.4% of cases. The Table 2 summarizes the characteristics of all patients in this cohort.

Table 1. Assessment of loss of lordosis according to theoretical L1-S1 lordosis.

	Number	%
Lost of lordosis>30°	16	34.04
Lost of lordosis beetwen 20° and 30°	11	23.41
Lost of lordosis between 10° to 20°	05	10.63
No lost of lordosis	15	31.92

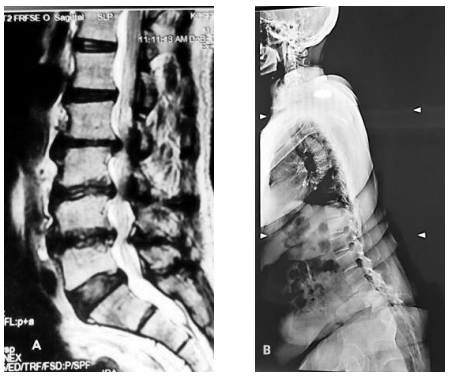


Figure 1. Sagittal MRI with L1-S1 spinal canal stenosis (A**)** with important degenerative changes in spine architecture on full spine radiography in standing profile (**B**).

Discussion

Lumbar canal stenosis is a common degenerative disease which causes significant pain and functional disability (2,3). This condition is responsible for spinal structural and architectural modifications. Evaluating its impact on spinal alignment is needed to establish spinal and pelvic parameters of the target population. Savadogo et al defined LL of subsaharians Africans according to pelvic parameters (12).

Table 2. Summary of the study patient characteristics.

N°	Age/sex	PI	PT	SS	LL1-S1	TLL1-S1	LL4-S1	TK	SVA	TPA	PI-LL	LL1-S1-TL1-S1
1	64/F	78°	36°	42	25°	83°	11°	3°	61	46.4°	53°	-58°
2	39/F	35°	1°	36	59°	55°	42°	22.7°	9.4	7.4°	-24°	4°
3	17/F	60°	3°	57	85°	65°	40°	12.2°	21	9.1°	-25°	20°
4	58/F	52°	26°	26°	13°	62°	49°	6.7°	26	32.9°	39°	-49°
5	34/F	58°	8°	50°	73°	63°	60°	28,9°	25	3.3°	-15°	10°
6	23/F	61.8°	11.1°	50.7°	57.2°	66.8°	51°	18.1°	17	1.5°	4.6°	-9.6°
7	63/F	82°	45.1°	36.9°	49.1°	77°	12.1°	8.1°	24	44.6°	32.9°	-27.9°
8	55/M	42.6°	12.4°	30.2°	55.1°	57.6°	22.4°	6.9°	18	12.9°	-12.5°	-2.5°
9	57/F	47.9°	11.5°	36.4°	49.5°	62.9°	48.7°	8.2°	17	0.3°	-4.2°	-13.4°
10	48/M	37.1°	7.4°	29.7°	48.5°	57.1°	37.5°	4.5°	6.8	0.9°	-11.4°	-8.6°
11	53/F	85.9°	33.8°	52°	55.7°	80.9°	51.9°	11.8°	11	24.9°	30.2°	-25.2°
12	22/F	63.4°	18.4°	45°	94.3°	68.4°	55.1°	9.3°	14	9.7°	-30.9°	25.9°
13	74/M	48°	7°	41°	34°	58°	29°	2.9°	15	13.3°	14°	-24°
14	40/F	72°	15°	57°	79°	72°	65°	23.3°	12	7.6°	-7°	7°
15	48/F	44.2°	4.4°	48.6°	54.2°	59.2°	31.5°	9.9°	16	7.8°	-10°	-5°
16	40/F	53.7°	10.9°	42.8°	21.5°	63.7°	2.3°	23°	28	20.6°	32.2°	-42.2°
17	34/F	58.7°	17.7°	41.2°	57.6°	63	27.6°	13.8°	0.2	12.6°	1.1°	-5.4°
18	42/M	43.1°	12.6°	30.06°	44.4°	58.1°	36.6°	18.1°	0.4	9.3°	-1.3°	-13.7°
19	39/M	53.6°	8.6°	45°	66.1°	63.6°	43.5°	18.7°	15	1°	-12.5°	2.5°
20	57/F	54.1°	12.1°	42°	24.1°	64.1°	42.9°	18°	11	15.3°	30°	-40°
21	55/M	77.4°	22°	55.4°	68°	77.4°	65°	30.4°	8.2	14.2	9.4°	-9.4°
22	63/F	58.4°	46.1°	46.2°	40.4°	63.4°	35.4°	9.4°	20	16.9°	18.4°	-23°
23	50/M	90.6°	26.2°	64.4°	65.2°	85.6°	61.5°	0.8°	4.3	21.2°	25.4°	-20.4°
24	60/F	73°	34°	39°	57.2°	73°	20.3°	41.7°	43	43.1°	15.8°	-15.8°
25	48/M	44.2°	4.4°	39.8°	54.2°	59.2°	31.5°	10.1°	16	7.8°	-10°	-5°
26	38/M	58.4°	13.4°	45°	56.5°	63.4°	36°	19.8°	1.1	9.2°	1.9°	-7.1°
27	52/M	64.8°	21.6°	43.2°	53.8°	69.8°	28.4°	16.9°	37	29.4°	9°	-16°
28	44/F	70.4°	28.7°	41.7°	50.4°	70.4°	33°	21°	0.7	20.6°	20°	-20°
29	63/F	46.9°	11.6°	35.3°	55.7°	61.9°	42.7°	2.8°	20	13.4°	-8.8°	-6.2°
30	58/M	60.3°	16.3°	44°	40.3°	65.3°	34.3°	6.3°	28	21.9°	20°	-25°
31	73/F	66°	23.1°	42.9°	19.5°	71°	28.3°	19.6°	10	23°	46.5°	-51.5°

The PI distribution showed large PI in 15.90%, a middle PI in 65.90% and a low grade PI in 18.20%. They concluded that the expected LL of Subsaharian Africans was in fact mostly middle (12). PI distribution was identical in our sample with middle grade PI in 49%. Lim et al (10) evaluated spinopelvic alignment between degenerative spondylolisthesis (SPDL) and lumbar canal stenosis (SCL) on a sample of 142 patients. PI was 56.10 +/-10 in SPDL and 49.60 +/-12 in SCL. According to them SCL spinopelvic alignment was relatively well compensated by a pelvic retroversion. We can concluded that the possibility of pelvic retroversion is more important in SCL even when the PI was lower than in SPDL. In fact we observed that, the sagittal misalignment was compensated by a pelvic retroversion in 28% of patients of our cohort. We had also 19% of pelvic anteversion. They was no difference in pelvic adaptation between men and women (p=0.522).

The global L1-S1 lordosis stayed low according to PI. The theoretical L1-S1 lordosis was significantly superior to the measured one (p<0.005) in 87.2%. Thus the stenosis had a strong impact on the lumbar lordosis leading to a loss of it in the majority of patients (8,9).

Frequently it existed a transition from lordosis to kyphosis (2,3,11). Apart from absolute values of lumbar lordosis the measured LL was statistically

inadapted to PI in 60% thus testifying to this loss of lumbar lordosis. We noted also a statistically significant loss of adequacy between the segmental L4-S1 lordosis and the global lordosis L1-S1 (p=0.017). Distal lordosis was no longer synchronous with overall lumbar lordosis and therefore with other pelvic parameters. This state led to a forward shift of the head far from the axis of pelvis thus attested in our series by the lengthening of the SVA indicating severe anterior imbalance. We thus found 81% of patients having an imbalance according to SVA. This percentage was reduced to 42.6 when we use the T1 slope (TPA) ignoring the bias induced by pelvic retroversion. The TPA was 17.9°+/- 11.6° in lumbar canal stenosis. Hasegawa et al found 17.6° in degenerative spondylolisthesis and 35.6° lumbar canal stenosis with degenerative scoliosis (7). Despite the average value of the TPA, we noticed a global imbalance in 42.6% (TPA>20°).

One of the affected alignment parameters was thoracic kyphosis (1). In our series there was a loss of thoracic kyphosis in 93.6%. Overall, 95.7% were unbalanced at the thoracic spinal level, reflecting an adaptation phenomenon.

Conclusion

The analysis of the different parameters revealed that stenosis of the lumbar canal had a strong impact on spinal architecture. The key point of this impact seemed to be the loss of lumbar lordosis and this stenosis had a significant impact on the thoracic kyphosis.

The pain observed in this pathology was not only linked to the radicular compressive phenomenon but to the cascade of events aimed at rebalancing the spine. The spinal imbalance was global in the majority of cases. Therapeutic treatment in addition to root release should aim to restore lumbar lordosis with the problem of choosing the level of arthrodesis.

References

1. Barrey C, Jund J, Noseda O, Roussouly P. Sagittal balance of the pelvis-spine complex and lumbar degenerative diseases. A comparative study about 85 cases. Eur Spine J. 2007;16 (9):1459-67.
2. Conway J, Tomkins CC, Haig AJ. Walking assessment in people with lumbar spinal stenosis: Capacity, performance, and self-report measures. Spine J. 2011;11 (9):816-23.
3. Deyo RA. Treatment of lumbar spinal stenosis: a balancing act. Spine J. 2010;10 (7):625-7.
4. Deyo RA, Gray DT, Kreuter W, Mirza S, Martin BI. United states trends in lumbar fusion. Surgery for degenerative conditions. Spine (Phila 1976). 2005;30 (12):1441-5.
5. Duval-Beaupère G, Schmidt C, Cosson P. A barycentremetric study of the sagittal shape of Spine and pelvis: the conditions required for an economic standing position. Ann Biomed. Eng. 1992;20 (4):451-62.
6. Fairbank JC, Pynsent PB. The Owestry disability index. Spine (Phila Pa 1976). 2000;25 (22): 2940-52.
7. Hasegawa K, Okamoto M, Hatsushikano S, Watanabe K, Ohashi M, Vital J-M, Dubousset. Jean. Compensation for standing posture by whole-body sagittal alignment in relation to health-related quality of life. Bone Joint J. 2020;102-B(10):1359-67.
8. Jackson RP, Mcmanus AC. Radiographic analysis of sagittal plane alignment and balance in standing volunteers and patients with low back pain matched for age, sex, and size. A prospective controlled clinical study. Spine (Phila Pa 1976). 1994;19 (14):1611-8.
9. Korovessis P, Dilmas A, Iliopoulos P, Lambiris E. Correlative Analysis of lateral of lateral vertebral radiographic variables and medical outcomes study short form health survey: a comparative study in asymptomatic volunteers versus with low back pain. J Spinal disord. Tech. 2002;15 (5):384-90.
10. Li JK, Kim SM. Comparaison of sagittal spinopelvic alignment between lumbar degenerative spondylolisthesis and degenerative spinal stenosis. J Korean Neurosurg Soc. 2014;55 (6):331-6.
11. Oniankitan O, Magnan A, Fianyo E, Mijiyawa M. Lumbar spine stenosis in an outpatient clinic in lome. Med Trop. 2007;67 (3):263-6.
12. Otani K, Kikuchi S, Yabuki S, Igarashi T, Nikaido T, Watanabe K, konno S. Lumbar spinal stenosis has a negative impact on quality of life compared with other comorbidities: an epidemiological cross-sectional study of 1862 community-dwelling individuals. Sci World J. 2013:1-9. DOI: 10.1155/2013/590652
13 Savadogo B. Evaluation de la lordose lombaire du sujet Africain subsaharien à partir des paramètres pelviens. Thèse de doctorat d'état en médecine, Université Felix Houphouet Boigny, Côte d'ivoire; 2021.
14. Steurer J, Nydegger A, Held U, Brunner F, Hodler J, Porchet F, Min K, Mannion AF, Michel B and the Lumbsten research collaboration. Lumbsten: the lumbar stenosis outcome study. BMC Musculoskelet Disord.2010;11:1-6.
15. Ulrich CT, Schr RT, Jesse CM, Fichtner J, Raabe A, Payer M,Tessitore E, Hausmann ON. Equilibre sagittal et instabilité posturale comme critères de la chirurgie du rachis. Swiss Med Forum. 2021;21 (31–32):536–40.

Romanian Neurosurgery (2025) XXXIX (1): pp. 70-74
DOI: 10.33962/roneuro-2025-012
www.journals.lapub.co.uk/index.php/roneurosurgery

Carcinoma metastasis mimicking meningioma. Challenges and doubts

Marcel Sincari[1], Margarida Conceição[1], Mark-Daniel Sincari[2]

[1] Neurosurgery Department, Unidade Local de Saúde Dão Lafões, Viseu, PORTUGAL
[2] Faculdade de Medicina da Universidade de Coimbra, PORTUGAL

ABSTRACT

The scientific approach to dural metastases mimicking meningiomas holds significant relevance, as these cases pose considerable challenges to clinicians in routine practice. Differentiating between these two distinct pathologies is critical, particularly when conservative management is considered for patients diagnosed with non-surgical meningiomas. Misdiagnosis in such scenarios can lead to detrimental outcomes for the patient, underscoring the need for vigilant follow-up in cases exhibiting suspicious imaging patterns.

While biopsy could provide definitive diagnosis in uncertain cases, it is often avoided due to its inherent risks, especially in elderly patients and in cases where the meningiomas are located in surgically challenging regions. Consequently, most conservatively managed cases are presumed to be typical meningiomas, and invasive diagnostic measures are typically not pursued unless absolutely necessary [1].

However, carcinomas from various primary sites including the breast, prostate, gallbladder, larynx, and less commonly, Ewing's sarcoma or melanoma can rarely present as dural metastases, especially in the parasagittal convexity [2,3,4]. These metastases can closely mimic meningiomas both clinically and radiologically. A study of 1,000 meningioma cases diagnosed between 2004 and 2010 revealed that 20 (2%) were ultimately found to mimic, with histological diagnoses including gliosarcoma, Rosai-Dorfman disease, hemangiopericytoma, osteosarcoma, medulloblastoma, adenocarcinoma, and nonseminomatous germ cell tumours [5].

Among these, adenocarcinomas are the most common metastatic tumours mimicking meningiomas. These lesions, like meningiomas, exhibit attachment to the dura, a dural tail, and contrast enhancement [6]. Such imaging characteristics can make distinguishing metastatic tumors from meningiomas exceedingly difficult using standard neuroimaging techniques [7,8]. Even intraoperatively, dural metastases can appear identical to meningiomas, complicating diagnosis further [4]. Both conditions may share features such as a solid structure, limited diffusion of water molecules, extensive peritumoral edema, and similar contrast enhancement patterns [9].

The pathways for metastatic spread include arterial and venous routes, particularly via Batson's venous plexus [10]. Cases of cerebrospinal fluid (CSF) dissemination have not been described.

This report describes three cases of dural metastases mimicking meningiomas, with locations including the temporal region, the cavernous sinus, and the cervicothoracic dura.

Keywords
dura,
meningioma,
metastasis,
carcinoma

Corresponding author:
Marcel Sincari

Neurosurgery Department, Unidade Local de Saúde Dão Lafões, Viseu, Portugal

sincari1973@gmail.com

Scan to access the online version

Case 1.

41 years old lady with known breast cancer, mastectomy 5 years ago came for headache and MRI revealed a meningioma mimicking temporal lesion.

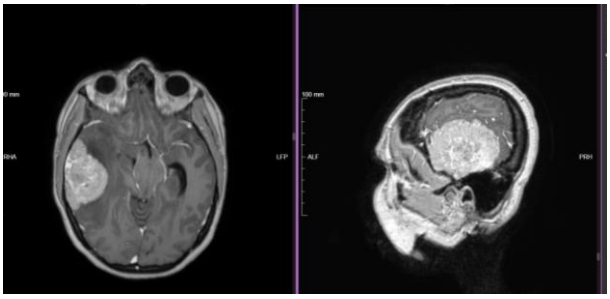


Figure 1. MRI of carcinoma metastasis mimicking meningioma.

She was selected for surgery, it was done complete removal, histological analysis was carcinoma HER2 negative with focal neuro-endocrine differentiations.

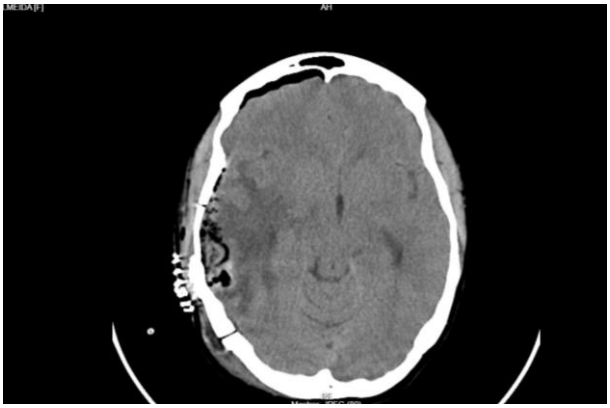


Figure 2. Postoperative CT scan showing total tumor removal.

Case 2

A 65-year-old male presented with sudden-onset, two-month progression of progressive vision loss in the right eye. Upon arrival at our service, he could only distinguish fingers at 10 cm. The ophthalmological findings were as follows: visual acuity deficit in the finger-counting maneuver (right eye), direct pupillary afferent defect (DPAR) in the right eye, and limitation of extraocular movements consistent with right third cranial nerve palsy. Fundoscopy was normal. Optical coherence tomography (OCT) showed bilateral increased retinal nerve fiber layer thickness with early ganglion cell layer loss temporally in the right macula.

He was referred for neurosurgical evaluation, which led to neuroimaging studies (head CT scan and MRI), demonstrating a right cavernous sinus lesion centered on the anterior clinoid process, extending to the sphenoid ridge and orbital apex, encasing the right internal carotid artery (ICA), and indenting the prechiasmatic segment of the right optic nerve and ipsilateral chiasm. While preparing for surgery, a mass was found in the left lung on a chest X-ray. Further investigation with a TAP CT scan revealed a solid spiculated lesion in the left upper lung lobe, with necrotic supraclavicular lymphadenopathy, bilateral adrenal metastasis (4.5 cm left, 5.0 cm right), and a lytic lesion in the right iliac bone.

The cranial lesion was removed through a right pterional craniotomy, classified as "Simpson Grade II," which revealed significant invasion of the right cavernous sinus. The conclusion of the anatomopathological report was mucinous adenocarcinoma. There was no immunophenotype that allowed for the safe identification of the primary origin. Immunohistochemistry showed immunoreaction for CAM 5.2 and K7, with only faint staining for GATA3 in the supporting cells. The study was negative for CK20, TTF1, CDX2, and SATB2, suggesting a pulmonary primary origin.

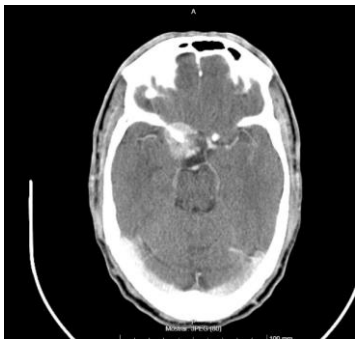


Figure 3. The initial head CT scan demonstrated a right cavernous sinus lesion mimicking meningioma.

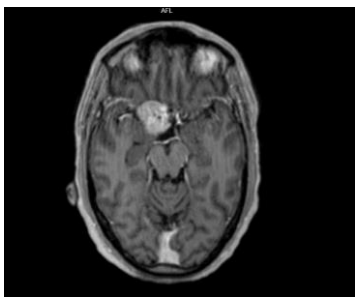


Figure 4. MRI Brain

MRI Brain confirmed a 2.9 × 2.6 × 1.6 cm right cavernous sinus lesion centered on the anterior clinoid process, extending to sphenoid ridge and orbital apex. Encasing the right internal carotid artery (ICA). Indenting the prechiasmatic segment of the right optic nerve and ipsilateral chiasm. Angiographic study: 70% stenosis of the clinoid segment of the right ICA without occlusion.

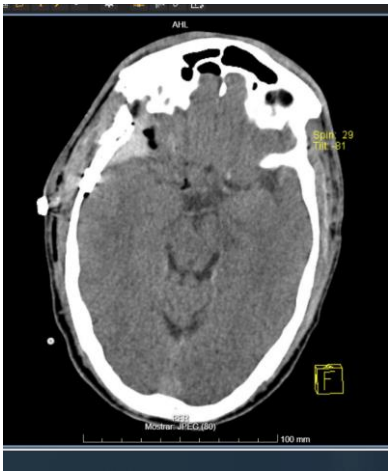


Figure 5. Postoperative CT scan, revealing removal of the mass.

Case 3

37-year-old lady with known systemic cancer, breast invasive carcinoma HER2 positive (score 3+) with negative progesterone and estrogenic receptors, treated surgically two years before diagnosing the pathological fracture Th12, associated with Th11 vertebral body infiltration.

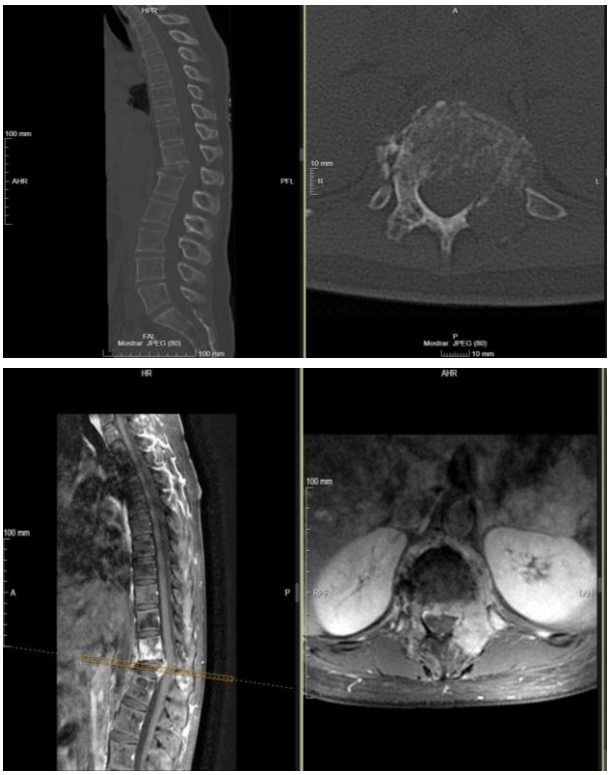


Figure 6. Th12 pathological fracture with posterior structures infiltration and Th11 vertebral body metastasis

The main complaint was uncontrolled pain, Asia E. She was selected for surgical treatment: posterior approach, Th12 vertebrectomy, posterior fixation, and Th10-L2, arthrodesis with expandable cylinder, Th11-L1, and cement augmented screws in Th11, L1 after radio ablation of metastasis of Th11 vertebral body.

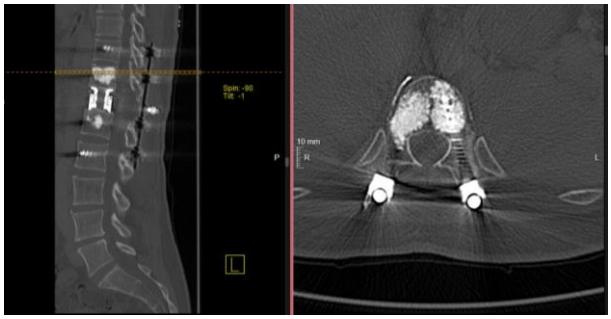


Figure 7. Postoperative CT scan: vertebrectomy Th12, good Th11 vertebral body cement filling after radio ablation.

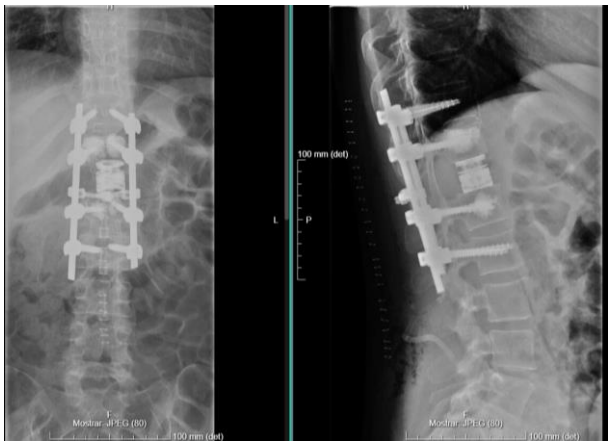


Figure 8. Postoperative X-Ray

She did well during 2 years, reduced painkillers, using only in SOS. She came back with progressive weakness of inferior limbs and was diagnosed with intramedullary metastasis (conus)

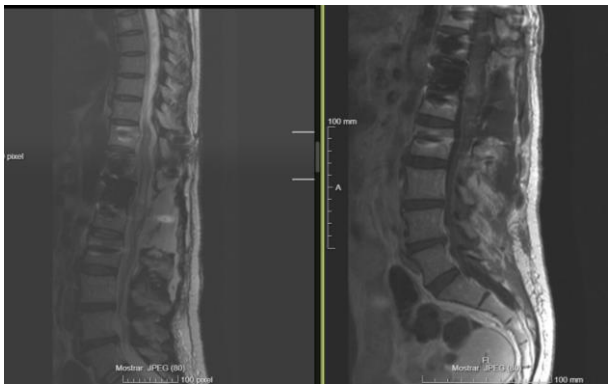


Figure 9. Conus medullary metastasis

She was submitted to another surgery-microscopic removal of intramedullary metastasis with no neurological improve. Nine months after, a patient is almost paraplegic with recurrence of the metastasis.

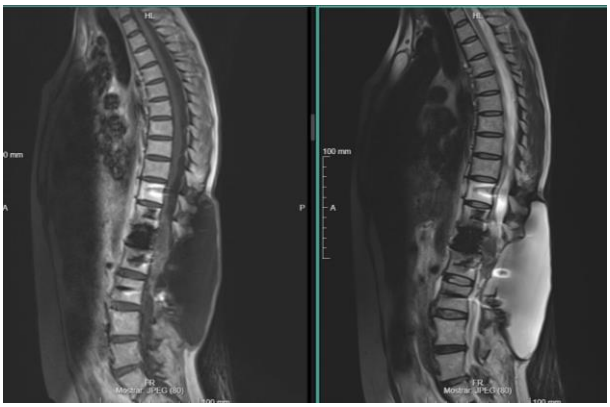


Figure 10. Medullary metastasis recurrence.

We did another attempt to remove the metastasis with good imageology result as seen in the figure.

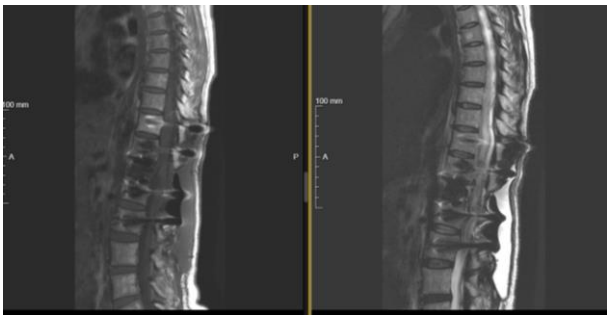


Figure 11. Postoperative MRI after second metastasis removal

But the patient was gradually worsening and another MRI revealed multiple cardiothoracic intradural and cranial metastasis with dural implantation, mimicking meningiomas. In this case we support the theory of CRF dissemination.

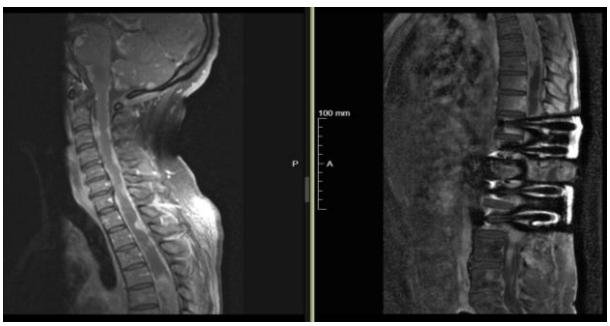


Figure 12. Cervicodorsal and endocranial dural metastasis mimicking meningiomas.

Conclusions

In cases of patients with diagnosed systemic cancer, the radiological finding of an intracranial mass with contrast enhancement must be considered as a metastasis until histological proof, even when a meningioma is suspected. And in contrast, in usual meningioma cases we should always pay attention to histological results, especially in younger patients and if metastasis is found we should check them for primary source. Metastasis misinterpreted as meningioma can delay surgery and consequently have a deleterious impact on patient care, being imperative to distinguish them from a meningioma. There is no standard algorithm treatment dural metastasis, surgical resection and radiation therapy are used in cases of dural metastasis, surgical treatment being the most efficacious, especially when the lesion is removable and systemic disease can be suppressed.

References

1. Natasha M. Savage1, Cargill H. Alleyne2, John R. Vender2, Ramon Figueroa3, Hui Zhang4, Thomas A. Samuel4, Suash Sharma1. Dural-based metastatic carcinomas mimicking primary CNS neoplasia: report of 7 cases emphasizing the role of timely surgery and accurate pathologic evaluation. Int. J Clin Exp Pathol 2011;4(5):530-540
2. Kleinschmidt-DeMasters BK. Dural metastases. A retrospective surgical and autopsy series. Arch Pathol Lab Med 2001; 125: 880-887.oma.23;31;40
3. Lippman SM, Buzaid AC, Iacono RP, Steinbronn DV, Stanisic TH, Rennels MA, Yang PJ, Garewal HS, Ahmann FR. Cranial metastases from prostate cancer simulating meningioma: report of two cases and review of the literature. Neurosurgery 1986; 19: 820-823
4. Tagle P, Villanueva P, Torrealba G, Huete I. Intracranial metastasis or meningioma? An uncommon clinical dilemma. Surg Neurol 2002; 58: 241-245
5. Ghosal N, Dadlani R, Gupta K, Furtado SV, Hegde AS (2012) A clinicopathological study of diagnostically challenging meningioma mimics. J Neurooncol 106(2):339–352
6. Quint DJ, McGillicuddy JE. Meningeal metastasis of the cerebellopontine angle demonstrating „dural tail" sign. Can Assoc Radiol J 1994;45:40–3
7. Bademci G, Bozdogan O, Berdan F, Evliyaoglu C. Extremely delayed renal cell carcinoma metastasis mimicking convexity meningioma. Neurocirugia 19: 562-564, 2008
8. Schmidt GW, Miller NR. Metastatic breast carcinoma mimicking basal skull meningioma. Clin Ophthalmol 1: 343-346, 2007

9. Wu H, Beylerli O, Gareev I, Beilerli A, Ilyasova T, Talybov R, Sufianov A, Guo X. Are there reliable multiparametric MRI criteria for differential diagnosis between intracranial meningiomas and solitary intracranial dural metastases? Oncol Lett. 2023 Jun 28;26(2):350. doi: 10.3892/ol.2023.13936. PMID: 37427340; PMCID: PMC10326821

10. Mitchell RA, Dimou J, Tsui A, Kavar B. Metastatic prostate adenocarcinoma invading an atypical meningioma. J Clin Neurosci 2011;18:1723-5

Romanian Neurosurgery (2025) XXXIX (1): pp. 75-78
DOI: 10.33962/roneuro-2025-013
www.journals.lapub.co.uk/index.php/roneurosurgery

Sellar space-occupying lesion: not always a pituitary tumour!

Sharad Pandey[1], N. Monica[1], Nirupma P. Khan[2], Anurag Anand[1]

[1] Department of Neurosurgery, A.B.V.I.M.S. and Dr Ram Manohar Lohia Hospital, New Delhi, INDIA
[2] Department of Pathology, A.B.V.I.M.S. and Dr Ram Manohar Lohia Hospital, New Delhi, INDIA

ABSTRACT

Background and objective: Hypophysitis is an inflammatory disease of the pituitary gland that is clinically and radiologically similar to pituitary tumours. We are reporting a case of xanthogranulomatous hypophysitis which was confused as a pituitary neoplasm preoperatively.

Materials and methods: A 56-year-old woman presented with extreme tiredness and visual disturbances for 4 months and had visited multiple doctors for the same. NCCT head and CEMRI done preoperatively were suggestive of sellar SOL. The patient was optimised preoperatively and underwent trans-nasal transsphenoidal surgery was performed. Histologic examination of the tissue was s/o xanthogranulomatous hypophisitis

Conclusion: We have described an unusual inflammatory lesion of the pituitary in the sellar region that was mimicking neoplasm. A high level of clinical suspicion and knowledge regarding the differential diagnosis of the sellar region is necessary for correct diagnosis and management.

Key message: All the sellar space occupying lesions are not always pituitary adenomas. Rare entities like xa thogranulomatous hypophisitis should be thought of when we encounter patients with hypocortisolism.

Keywords
sellar SOL, pituitary, xanthogranulomatous hypophisitis

Corresponding author:
Sharad Pandey

A.B.V.I.M.S. and Dr Ram Manohar Lohia Hospital,
New Delhi, India

drsharad23@yahoo.com

INTRODUCTION

Pituitary hypophysitis is one of the rare, inflammatory condition that may present both clinically and radiologically similar to a pituitary tumor (1). Clinically, it might present with f/o headaches and/ or visual disturbance due to the local mass effect and compression of the optic chiasm. Inflammatory infiltration can lead to pituitary dysfunction in these patients.

Xanthogranulomatous hypophysitis, is an inflammatory disease showing granulomas with foamy macrophages, multinucleated giantcells, epithelioidcells, and infiltration of lymphocytes (1). Hypophysitis can be histologically divided into the five distinct categories: lymphocytic, granulomatous, xanthomatous, necrotising and xanthogranulomatous hypophysitis (XGH). Xanthogranulomatous

type is a mixture of the xanthomatous and granulomatous subtypes (1,2). Based on their anatomical location and areas of infiltration, they can also be classified as either adenohypophysitis or infundibuloneurohypophysitis (1,2). Most common type of hypophisitis is lymphocytic hypophysitis (1) and XGH is rare type. On histopathological evaluation of Xanthogranulomatous hypophysitis, cholesterol clefts, haemosiderin deposits, multinucleated giant cells, macrophage accumulation and fibrous proliferation can be seen (1,3). Currently the pathogenesis of this condition is not very well described and aetiologically it can either be a primary (most common) or a secondary hypophysitis. Primary hypophysitis is autoimmune in origin. Primary hypophisitis can occur in isolation or as a part of systemic disease such as polyglandular autoimmune syndromes or IgG4 systemic disease (1)(4). Secondary hypophysitis might occur as a consequence of local inflammation caused by lesions including craniopharyngioma (CP)/Rathke's cleft cyst or as a part of systemic diseases such as tuberculosis, sarcoidosis, Wegener's granulomatosis or syphilis (1) (4) (5).

We report here a case of xanthogranulomatous hypophysitis in a 56-yr-old woman, who presented with symptoms mimicking pituitary neoplasms.

CASE REPORT

A 56-yr-old woman came with c/o extreme tiredness and visual disturbances in the form of reduced peripheral vision since 4 months associated with headache and on and off vomiting. There was h/o significant weight loss. Patient had visited a physician with these complaints and underwent a thorough examination by which she was found to have hypothyroidism was started on replacement Thyroxine 25mcg OD and . Patient visual distubances continued to worsen for which she consulted an ophthalmologist who evaluated and advised a MRI orbit which showed an incidental pituitary lesion. On further hormonal work up by an endocrinologist she was found to have hypocortisolism with baseline hormone profile values being as follows: Replacement hydrocortisone of 10mg in the morning and 5 mg in the evening was started with which patient improved significantly with reduced apathy and regained appetite with weight gain.

On further revaluation with CEMRI brain, there was a heterogeneously enancingT2/T1 isointense lesion seen in the sella and suprasellar region, measuring 2.3x1.5x2.0cm. It was causing widening of the sella(Fig 1a) . Anterior pituitary gland couldnot be identified separately from the lesion and pituitary stalk was involved. Superiorly, lesion was extending upto the floor of 3rd ventricle causing displacement and compression of optic chiasm. On right lateral side, lesion extends beyond the lateral tangent line into the superior cavernous sinus compartment, partially encasing the right cavernous ICA(KNOSP grade 3A). On left side, it is extending between intercarotid line and lateral tangent (KNOSP grade 2) (Fig 1b). The minimum diameter between the cavernous segment of both ICA was 14mm

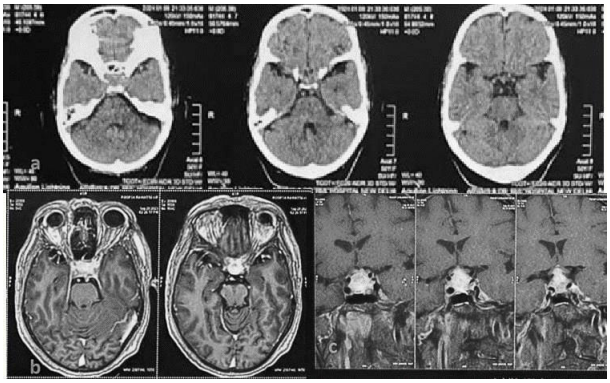


Figure 1. (**a**) NCCT head showing isodense sellar space occupying lesion. (**b**) MRI axial image of Isointensense sellar lesion. (**c**) MRI coronal image of Knosp grade 3A on the right and grade 2 on the left.

Hormone profile was done and showed results as mentioned in the Table 1.

Table 1. Pre operative hormone profile reports

Hormone	IGF-1(ng/ml)	Prolactin (ng/ml)	fT3 (pg/ml)	fT4 (ng/ml)	TSH (mcUI/ml)	ACTH (pg/ml)	Cortisol (mcg/dl)
Levels (N)	44-210	1.8-20.30	2.30-4.20	0.89-1.76	0.55-4.76	<46	2.5-25
Patient Report	35.30	3.40	1.52	0.33	2.15	27.20	0.75

Preoperatively patient was started on Tab. Hisone 10mg in the morning and 5mg in the night and Tab. Thyroxinbe 25mcg OD daily and was optimized and was taken up for surgery

Patient underwent Transsphenoidal tumor decompression and intraoperatively there was a fibrotic firm tumor which was moderately vascular . Near total tumor decompression was done and

tissue was fixed immediately in formalin and sent for histopathological examination. And on Histopathological examination , there was Rathke's cleft in the pituitary tissue (Fig. 2b) highlighted by synaptophysin on IHC surrounded and infiltrated by foamy histiocytic collection (xanthoma cells) and along with both Langerhans type 2 foreign body giant cells surrounded by lymphocytes with no caseous necrosis seen (Fig. 2a).

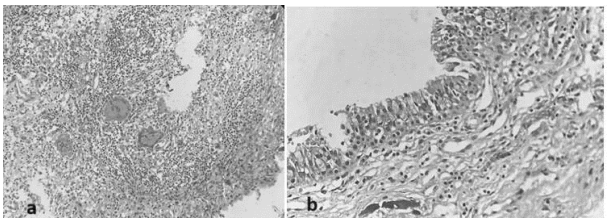


Figure 2. (**a**) Histological features showing xanthogranulomatous lesion (10x). (**b**) Simple cyst lined by single layer of columnar cell supported by fibrocollagenous stroma (40x).

Postoperatively patient developed diabetes insipidus which was managed with the help of endocrinologist. Rest of the postoperative period was uneventful. Replacement steroid and thyroxine is being continued in the postoperative period and the post op hormone profile was as mentioned below (Table 2).

Table 2. Postoperative hormone profile

Hormone	IGF - 1 (ng/ml)	Prolactin (ng/ml)	fT3 (pg/ml)	fT4 (ng/ml)	TSH (mcUI/ml)	ACTH (pg/ ml)	Cortisol (mcg/dl)
Levels (N)	44-210	1.8-20.30	2.30-4.20	0.89-1.76	0.55-4.76	<46	2.5=25
Patient Report	75.4	2.14	1.92	0.49	0.41	3.2	18.5

Discussion

Hypophysitis can be due to number of inflammatory processes that are different in etiology, pathogenesis, and morphology [8]. Idiopathic or primary inflammatory lesions of the hypophysis include lymphocytic hypophysitis, granulomatous hypophysitis, xanthomatous hypophysitis, xanthogranulomatous hypophysitis, and necrotizing hypophysitis [2,4,7].

Granulomatous hypophysitis is histologically characterized by aggregates of multinucleated giant cells, macrophages, and extensive plasma cell infiltration [3,6]. The histologic findings consisted of foamy histiocytic (xanthomatous) infiltration without evidence of associated adenomas, cyst, hemorrhagic infarct, granulomas, Langerhans cells, neutrophilic exudates, or Michaelis– Gutmann bodies.

In our case, the MRI findings showed homogenously isointense lesion in the sellar and suprasellar region causing widening of sella from which anterior pituitary couldn't be visualised separately.

And the histopathological specimen in our case included Rathke's tissue highlighted by synaptophysin on IHC and infiltrated by foamy histiocyte collection (xanthoma cells) and long with both Langhan's giant cells and foreign body type giant cells surrounded by lymphocytes.

The pathological mechanisms underpinning autoimmune pituitary disorders merit some discussion. The present patient did not have a history of autoimmune disorders, but her sex and age may suggest the progression of LH to XGH by the time of presentation. However, infection or systemic disease may also have been involved in the etiology. Systemic conditions such as tuberculosis, sarcoidosis, and other granulomatous diseases were excluded as etiologic factors in the present patients.

Surgical resection is typically the only treatment that is required. However, anti-inflammatory and immunosuppressive drugs may be indicated in patients with residual or recurrent hypophysitis

The overall prognosis for XH is good, but improvement of pituitary function after transsphenoidal surgery has been reported in less than 50% of the cases in the literature. Chronic inflammation may result in destruction and fibrosis of the pituitary gland (6)

In summary, we described an unusual inflammatory lesion of the pituitary mimicking a neoplasm in the sellar region. A high level of clinical suspicion of the inflammatory disorder is necessary to provide the correct diagnosis and choose optimal management. (8)

Conclusion

In the present report, we have described a patient with a rare pituitary pathology who presented with hypopituitarism and visual impairment and was misdiagnosed as having a macroadenoma. Postoperative histological evaluation revealed an XGH of a remodeled RPC. Following surgical treatment, the patient had persistent pituitary

dysfunction. Surgical intervention early in the development of such lesions may have beneficial effects on pituitary function, because chronic inflammation leads to the destruction of the pituitary gland and permanent pituitary dysfunction

REFERENCES:

1. Yokoyama S, Sano T, Tajitsu K, Kusumoto K. Xanthogranulomatous hypophysitis mimicking a pituitary neoplasm. Endocrine Pathology. 2004 Dec;15:351-7.
2. Tashiro T, Sano T, Xu B, Wakatsuki S, Kagawa N, Nishioka H, Yamada S, Kovacs K. Spectrum of different types of hypophysitis: a clinicopathologic study of hypophysitis in 31 cases. Endocrine pathology. 2002 Sep;13:183-95.
3. Sugata S, Hirano H, Yatsushiro K, Yunoue S, Nakamura K, Arita K. Xanthogranuloma in the Suprasellar Region—Casae Report—. Neurologia medico-chirurgica. 2009;49(3):124-7.
4. Leporati P, Landek-Salgado MA, Lupi I, Chiovato L, Caturegli P. IgG4-related hypophysitis: a new addition to the hypophysitis spectrum. The Journal of Clinical Endocrinology & Metabolism. 2011 Jul 1;96(7):1971-80.
5. Mittal R, Kalra P, Dharmalingam M, Verma RG, Kulkarni S, Shetty P. Lymphocytic hypophysitis masquerading as pituitary adenoma. Indian Journal of Endocrinology and Metabolism. 2012 Dec 1;16(Suppl 2):S304-6.
6. Gutenberg A, Hans V, Puchner MJ, Kreutzer J, Brück W, Caturegli P, Buchfelder M. Primary hypophysitis: clinical-pathological correlations. European Journal of Endocrinology. 2006 Jul;155(1):101-7.
7. Tashiro T, Sano T, Xu B, Wakatsuki S, Kagawa N, Nishioka H, Yamada S, Kovacs K. Spectrum of different types of hypophysitis: a clinicopathologic study of hypophysitis in 31 cases. Endocrine pathology. 2002 Sep;13:183-95.
8. Burt MG, Morey AL, Turner JJ, Pell M, Sheehy JP, Ho KK. Xanthomatous pituitary lesions: a report of two cases and review of the literature. Pituitary. 2003 Sep;6:161-8.

Romanian Neurosurgery (2025) XXXIX (1): pp. 79-81
DOI: 10.33962/roneuro-2025-014
www.journals.lapub.co.uk/index.php/roneurosurgery

A rare case of cerebral astroblastoma

Binoy Singh, Tamajyoti Ghosh, Biswajit Dey

AIIMS Raipur, INDIA

ABSTRACT

Cerebral Astroblastomas are rare central nervous system tumours constituting 0.45-2.8% of neuroglial tumours Recent 2021 WHO classification of Brain tumours has described Astroblastoma as "other neuroepithelial tumours" with MN1 rearrangement. Because of its rare entity and indistinctive radiological and pathological features diagnosis and further management continue to remain a challenge. Ependymoma and angiocentric glioma are the most important differential diagnoses of Astroblastoma and at times pre-operative differentiation is challenging. Here we discuss the case of a 22-year-old female with features of headache and vomiting. Histopathological examination revealed Cerebral Astroblastoma MN1 altered. Post-operative imaging suggested gross total resection of the tumour and the patient was subjected to radiotherapy of 54Gy in the tumour bed. 1-year follow-up of the patient showed no recurrence.

Keywords
brain tumour, immunohistochemistry, ependymoma

Corresponding author:
Tamajyoti Ghosh

AIIMS Raipur, India

tamajyoti@gmail.com

INTRODUCTION

Cerebral Astroblastoma are rare Central nervous system tumour constituting 0.45-2.8% of neuroglial tumours (1) . Recent 2021 WHO classification of Brain tumour has described Astroblastoma as "other neuroepithelial tumours" with MN1 rearrangement (2). Because of its rare entity and indistinctive radiological and pathological features diagnosis and further management continues to remain a challenge (3). Here we discuss the case of a 22 year old female with features of headache and vomitting. Histopathological examination revealed Cerebral Astroblastoma MN1 altered.

CASE REPORT

We report the case of a 22 year old female who presented with features of raised intracranial features. On clinical examination she was conscious, oriented with no focal neurological deficit. Her fundoscopy was normal. Contrast Magnetic resonace study of her brain revealed T1 hypointense , T2 hyperintense lesion in left pearietal lobe.

The lesion was heterogenously contrast enhancing with bubbly appearance within the lesion. GRE images was suggestive of punctate calcification.

A differential diagnosis of Supratentorial Ependymoma was made. patient underwent Left parietal craniotomy and tumour excisiom.

Scan to access the online version

Tumour was vascular, soft to firm with well defined brain tumour interface. On histopathological examination perivascular rossettes with broad, thick process were seen.

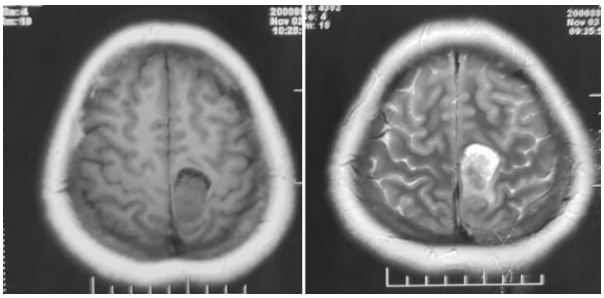


Figure 1a and 1b: T1 hypointense and T2 hyperintense lesion in left parietal lobe

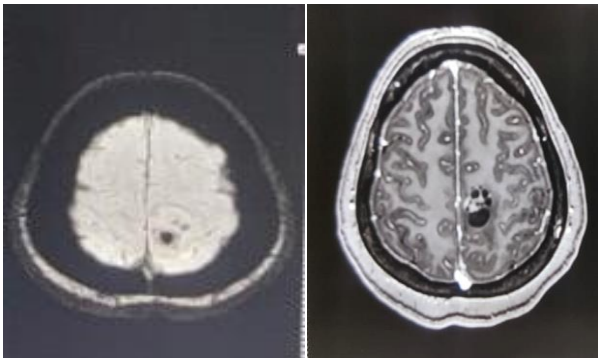


Figure 2a and 2b: Left parietal lesion showing punctate calcification and heterogenously enhancement with characteristic bubbly appearance

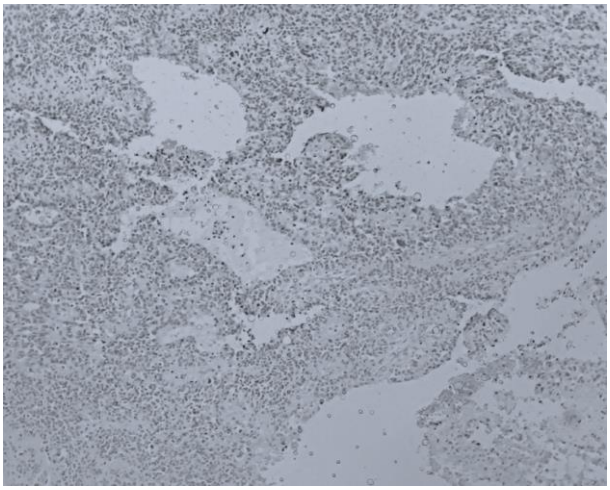


Figure 3: Histopathology showing Perivascular pseudorosettes with thick processes (H & E, 100x)

Immunohistochemistry study of the specimen showed GFAF, OLIG2, EMA and Vimentin positive.

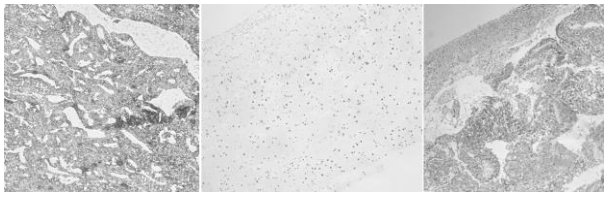


Figure 4: IHC panel showing GFAF, OLIG2 and EMA positive

ATRX retained and absence of IDH1 and P53 mutation differntiated it from ependymoma. Fluorescence in situ hybridization (FISH) showed MN1 break apart in more than 30% of tumour cells. A final diagnosis of astroblastoma, MN1 altered was made. Post operative imaging suggested gross total resection of tumour and the patient was subjected to radiotherapy of 54Gy in the tumour bed. 1 year followup of the patient showed no recurrence.

Discussion

Astroblastoma are extremely rare CNS tumours having bimodal age ditribution incidence peak in between 5-10 years and then in young adults between 21-30 years (4) . They are more frequently seen in females (1:11)(5) . They are generally supratentorial in location more frequently found in frontal lobe (6). Headache, seizure , vomiting and focal neurological deficit are the most frequent presenting symptoms (7). Ependymoma and angiocentric glioma are the most important differential diagnosis of Astroblastoma and at times pre operative differtiation is challenging (8). Astroblastoma are exclusively supratentorial and peripheral in location (7). They are characteristically large , well demarcated, lobulated mass with areas of calcificationmhaving solid component with little vasogenic edema unlike ependymoma (9), (10). On Histological examination both astroblastoma and ependymoma have pseudorosettes but absence of fibrillary background and presence of short and broad cytoplasmic processes helps in differntiation of astroblastoma (9), (10).

They show IHC positivity towards GFAF, EMA, Pancytokeratin, Oligodendrocyte transcription factor 2, vimentin and negative for IDH1/2 and TP53 mutation (11). They are classified into low garde and high grade based upon presence of mitosis, Ki67 index , degree of cellular pleomorphism and areas of necrosis. High grade Astroblastoma have anaplastic nuclear features, increased mitosis >5 per 10 HPF, microvascular proliferation and necrosis with

palisading with Ki67 index >10% (12). Our case had areas of focal necrosis and frequent mitosis >5 per HPF with Ki67 index of >10% classifying it as high grade. 2021 WHO CNS classifcation has classifed astroblastoma as seperate entity of tumours under "other neuroepeithelial tumours" (2). MN1 rearrarangement has defined Astroblastoma as either MN1 altered where rearrangement is seen or Astroblastoma NOS where MN1 alteration cannot be tested (2). As these are rare tumour optimal management strategies still remains unclear. However total surgical resection is preferred followed by adjuvant radiotherapy in high grade lesions. Role of chemotherapy remains unclear (13).

Conclusion

Cerebral Astroblastomas are extremely rare CNS neoplasms with non-distinct Radiological and pathological features thus making their diagnosis and further management quite challanging. Astroblastoma should be one of the differentials in young females with supratentorial neoplasms having MRI characteristics of Ependymoma. Histopathological examination and IHC and cytogenetics study plays a crucial role in diagnosis and differntiation of Astroblastoma.

Refrences

1. Eom KS, Kim JM, Kim TY. A cerebral astroblastoma mimicking an extra-axial neoplasm.J Korean Neurosurg Soc. 2008;43:205–208. doi: 10.3340/jkns.2008.43.4.205.
2. Louis DN, Wesseling P, Aldape K, et al. cIMPACT-NOW update 6: new entity and diagnostic principle recommendations of the cIMPACT-Utrecht meeting on future CNS tumor classification and grading. Brain Pathol. 2020;30:844–856. doi: 10.1111/bpa.12832.
3. Agarwal V, Mally R, Palande DA, Velho V. Cerebral astroblastoma: A case report and review of literature. Asian J Neurosurg. 2012;7:98–100.
4. Shen F, Chen LC, Yao Y, Zhou LF. Astroblastoma: rare incidence and challenges in the pattern of care.World Neurosurg. 2014;82:0–7. doi: 10.1016/j.wneu.2014.03.008.
5. Port JD, Brat DJ, Burger PC, Pomper MG. Astroblastoma: Radiologic-Pathologic Correlation and Distinction from Ependymoma. Am J Neuroradiol. 2002;23:243–7.
6. Singh DK, Singh N, Singh R, Husain N. Cerebral astroblastoma: A radiopathological diagnosis. J Pediatr Neurosci. 2014;9:45–7.
7. Ahmed KA, Allen PK, Mahajan A, Brown PD, Ghia AJ. Astroblastomas: a surveillance, epidemiology, and end results (SEER)-based patterns of care analysis. World Neurosurg. 2014;82:e291–7.
8. Shen F, Chen L-c, Yu Y, Zhou L-F. Astroblastoma: Rare Incidence and Challenges in the Pattern of Care. World Neurosurg. 2014;82(1/2):e125–7.
9. Alaraj A, Chan M, Oh S, Michals E, Valyi-Nagy T, Hersonsky T. Astroblastoma presenting with intracerebral hemorrhage misdiagnosed as dural arteriovenous fistula: review of a rare entity. Surg Neurol. 2007;67:308–13.
10. Notarianni C, Akin M, Fowler M, Nanda A. Brainstem astroblastoma: a case report and review of the literature. Surg Neurol. 2008;69:201–5.
11. Dey B, Dutta S, Saurabh A, Raphael V, Khonglah Y. Cerebral Astroblastoma: A Rare Tumor. Cureus. 2021 Jul 12;13(7):e16323. doi: 10.7759/cureus.16323. PMID: 34395111; PMCID: PMC8355865.
12. Hammas N, Senhaji N, Alaoui Lamrani MY, Bennis S, Chaoui EM, El Fatemi H, Chbani L. Astroblastoma - a rare and challenging tumor: a case report and review of the literature. J Med Case Rep. 2018;12:102. doi: 10.1186/s13256-018-1623-1.
13. Salvati M, D'Elia A, Brogna C, et al. Cerebral astroblastoma: analysis of six cases and critical review of treatment options. J Neurooncol 2009; 93:369-78.

Romanian Neurosurgery (2025) XXXIX (1): pp. 82-85
DOI: 10.33962/roneuro-2025-015
www.journals.lapub.co.uk/index.php/roneurosurgery

Cranioplasty: autologous bone graft vs. titanium mesh. Comparative cross-sectional study done in single centre

Binit Kedia, Kritesh Goel, Krishna Govind Lodha, Gaurav Jaiswal

RNT Medical College, INDIA

ABSTRACT

Background: Cranioplasty (CP) is a neurosurgical procedure performed after decompressive craniectomy using autologous bone graft or various artificial materials.

Objective: To determine differences in complications between patients who underwent CP using an autologous bone flap versus titanium mesh and to identify significant risk factors for post-CP complications.

Study design: Comparative cross-sectional study.

Methods: A total of 38 patients, 29 males (76.4%) and 9 females (23.6%), were included in this study. All patients underwent cranioplasty with titanium mesh or autologous bone graft.

Results: The results were compared between autografts and titanium implants. Autologous bone graft was used in 76.3% of the patients and titanium implant was used in 23.7%. Different complications occurred in 5.26% of the patients in both groups, 2.63% in the autologous group and 2.63% in the titanium mesh group respectively. Infection occurred in the surgical site in 5.26% of the patients in both groups (similar). Cranioplasty infection occurred in 2.63% of the patients who underwent autologous transplantation. One patient developed a hematoma in both groups. One patient underwent autologous bone graft removal, and one patient underwent mesh removal.

Practical implication: Titanium mesh cranioplasty is an essential procedure for junior neurosurgeons to learn and achieve good results, shorten hospital stays, and save hospital resources.

Conclusion: Titanium mesh cranioplasty has similar complications to autologous bone cranioplasty.

Keywords
autograft,
bone resorption,
cranioplasty,
haematoma,
infection,
titanium mesh

Corresponding author:
Binit Kedia

RNT Medical College, India

bkedia1989@gmail.com

Scan to access the online version

INTRODUCTION

Neurotrauma is a leading cause of death and disability in India[1]. Cranioplasty is the treatment of the skull defect after a previous surgery due to trauma. This surgery is performed after treatment and stabilization of the original pathology such as cerebral edema, brain tumor, traumatic brain injury, or infarction leading to craniectomy. This is followed by cranioplasty to protect brain parenchyma, provide cosmesis, and to avoid the effect of atmospheric pressure on cranial

fluid dynamics such as trephine syndrome. The materials used for cranioplasty are divided into two categories: autologous bone or synthetic replacement. Autologous bone grafting uses the patient's own resected skull; it is both cost-effective and a physiological alternative to synthetic products[2].

Cranioplasty is usually performed 2–3 months after craniectomy, so bone preservation should be considered in advance. Autologous bone flaps can be preserved by cryopreservation (placing the bone flap in a special refrigerator according to a pre-planned protocol) or by placing the bone flap in a subcutaneous compartment in the abdominal wall. Many synthetic materials are available for cranioplasty, such as titanium mesh, polymethyl-methacrylate, hydroxyapatite cement, and polyetheretherketone (PEEK). There is currently no single device that provides all the required features for calvarial substitute. Therefore, neurosurgeons choose materials that are effective and have reduced complications[3]. Titanium mesh is easily available, strong with malleable properties, has a low infection rate, and facilitates cosmetic restoration.

The purpose of this study is to compare the complications of cranioplasty using autologous bone graft with titanium mesh implants. It also aims to show which cranioplasty method is effective, has the least complications, and provides the best results.

Methods

This is a comparative cross-sectional study conducted between February 2022 and January 2024 at the Department of Neurosurgery, RNT Medical College, Udaipur. Population Group: Patients over 10 years of age who underwent autologous bone/titanium mesh cranioplasty in our hospital were included. A total of 38 patients were selected, comprising 29 males and 9 females.

Data collection process

After approval by the Research Committee, patients' previous clinical data were collected along with related demographic information and relevant operative characteristics such as cranial defect size and Glasgow Coma Scale score (preoperative and postoperative). Acute surgical complications such as hospital-acquired infection or surgical site infection, hematoma, deep vein thrombosis, cerebrospinal fluid leak, bone resorption, readmissions, reoperations, cranioplasty flap removal, cranioplasty flap infection, duration of stay in the neurosurgical ward, and discharge were noted.

Instrument development

Decompressive craniectomy in patients with severe head trauma caused significant midline shift due to hematoma, contusion, tumor, or stroke causing intracranial pressure despite adjuvant medical treatment. In patients who underwent unilateral frontotemporoparietal craniectomy with durotomy or duroplasty, the bone flap edges were smoothed and placed in the subcutaneous abdominal compartment. The material used for CP depends on the surgeon's preference and the duration of DC and CP. In this study, we used titanium mesh plates in cases with a duration of 1.5 months to 22.5 months. We used a sterile clipper to remove the patient's hair before performing CP. The previous skin scar was reopened, and dissection was done in fibrous tissue between the dura and galea for the placement of the titanium mesh or autologous bone graft. In the case of using a bone flap, the flap was washed with normal saline solution. The flap was placed and fixed with multiple mini plates and screws in its original position using titanium plates and screws. The galea was then closed using vicryl, and skin closed with silk sutures.

Data Analysis

Frequencies or percentages of variables and means and standard deviations for continuous variables were used to present data.

Results

A total of 38 patients were selected to compare the results of autologous bone graft and titanium implants, comprising 76.4% male and 23.6% female. In 76.3% of patients, autologous bone graft was used, while titanium implant was used in 23.7% of patients. The overall complication rate was 5.26%, with 2.63% in the autologous group and 2.63% in the titanium mesh group. The incidence of surgical site infection was the same in both groups at 5.26%. Hematoma occurred in one patient of each group. Removal of the autologous bone graft was performed in one patient, and mesh removal in one patient in the other arm of the study. Bone resorption was observed in 5 patients, all of whom had autologous bone graft.

Table 1. Gender distribution

Gender	Frequency	Percentage
Male	29	76.4
Female	9	23.6
Total	38	100.0

Table 2. Frequency of autologous and titanium implant used

Type of Implant	Frequency	Percentage
Autologous Bone Graft	29	76.4
Titanium Implant	9	23.6
Total	38	100.0

Table 3. Indications

Indication	Frequency	Percentage
Traumatic Brain Injury	28	73.69
Stroke	3	7.89
Haemorrhage	5	13.15
Tumour	2	5.27
Total	38	100.0

Table 4. Postoperative complications

Complication	Frequency	Percentage
No Complication	36	94.72
Surgical Site Infection	1	2.64
Cranioplasty Infection	1	2.64
Total	38	100.0

Discussion

This comparative cross-sectional study compares autologous cranioplasty with titanium implants. Titanium mesh serves as an alternative to autologous bone flaps, offering advantages of bioinertness and less tissue reaction, especially in large bony defects.

In this study, one case of surgical site infection was observed in a patient with autologous cranioplasty and one case in a patient with titanium implants, resulting in rates of 2.63% for both. According to a study by Conen et al., the incidence of infection in implants ranges from 3-15%[4,5]. In our study, two patients (5.26%), one from each group, underwent cranioplasty removal due to infection.

Other studies suggest that debridement and bone preservation with intravenous and oral antibiotics are effective in 91% of patients[6]. These small studies confirm that implant retention or immediate exchange is an alternative to removal.

Overall, the results suggest no significant advantage of using autologous bone compared to titanium mesh after decompressive craniectomy. However, autologous cranioplasty often provides better cosmetic results and has evidence of bony fusion on radiographic examination of reconstructed anterior segment[7]. Some studies indicate problem is around 58.3% when autologous bone flap are used and 55% with titanium mesh[8]. In our study, equal complication rates were observed between the two materials.

Conclusion

In our study, cranioplasty using titanium mesh demonstrates complication rates similar to those associated with autologous bone grafting. Additionally, employing autologous bone, when feasible, can lead to reduced medical costs.

Study limitations

This study has several limitations. First, it includes data from a single tertiary care institution. Furthermore, obtaining patient consent may depend on the nature of the neurotrauma, making it difficult in some cases due to differences in GCS scores when caregiver consent was required.

References

1. Raja IA, Vohra AH, Ahmed M, Mb B. Neurotrauma in Pakistan. World Journal of Surgery. 2001 Sep 1;25(9):1230.
2. Piitulainen JM, Kauko T, Aitasalo KMJ, Vuorinen V, Vallittu PK, Posti JP. Outcomes of cranioplasty with synthetic materials and autologous bone grafts. World Neurosurg. 2015 May;83(5):708-14. doi: 10.1016/j.wneu.2015.01.014.
3. Meyer H, Khalid SI, Dorafshar AH, Byrne RW. The Materials Utilized in Cranial Reconstruction: Past, Current, and Future. Plast Surg (Oakv). 2021 Aug;29(3):184-196. doi: 10.1177/2292550320928560.
4. Conen A, Raabe A, Schaller K, Fux CA, Vajkoczy P, Trampuz A. Management of neurosurgical implant-associated infections. Swiss Medical Weekly. 2020 Apr 20;150(w20208):w20208.
5. Conen A, Fux CA, Vajkoczy P, Trampuz A. Management of infections associated with neurosurgical implanted devices. Expert Review of Anti-infective Therapy. 2017 Mar 4;15(3):241-55.
6. Yadla S, Campbell PG, Chitale R, Maltenfort MG, Jabbour P, Sharan AD. Effect of early surgery, material, and method of flap preservation on cranioplasty infections: a systematic review. Neurosurgery. 2011 Apr 1;68(4):1124-30.

7. Kim J, Kim JH, Kim JH, Kwon TH, Roh H. Outcomes of Cranioplasty Using Autologous Bone or 3D-Customized Titanium Mesh Following Decompressive Craniectomy for Traumatic Brain Injury: Differences in Complications. J Trauma Inj. 2019;32(4):202-209.

8. Kim J, Kim JH, Kim JH, Kwon TH, Roh H. Outcomes of Cranioplasty Using Autologous Bone or 3D-Customized Titanium Mesh Following Decompressive Craniectomy for Traumatic Brain Injury: Differences in Complications. J Trauma Inj. 2019;32(4):202-209.

Romanian Neurosurgery (2025) XXXIX (1): pp. 86-90
DOI: 10.33962/roneuro-2025-016
www.journals.lapub.co.uk/index.php/roneurosurgery

Management of intracranial lesions during pregnancy

Reeka Pradhan[1], Rajendra Shrestha[2]

[1] Paropkar Maternity and Women's Hospital, Thapathali, NEPAL
[2] Department of Neurosurgery. NAMS, Bir Hospital, NEPAL

ABSTRACT

Introduction. Intracranial lesions during pregnancy are very rare conditions in obstetrical and neurosurgical practice. There is no precise protocol for the management of intracranial lesions during pregnancy yet. The aim of this study was to elaborate on pregnant patients with intracranial pathology in order to achieve better outcomes in the future.

Methods. This is a descriptive cross-sectional study conducted among patients presented with intracranial lesions at the Department of Neurosurgery at the National Academy of Medical Science, Bir Hospital. The data was collected. retrospectively from hospital data thirty patients with intracranial lesions during pregnancy from 2015 to 2024. We analyzed the treatment decisions and obstetrical and neurosurgical outcomes. All patients were evaluated with either computed tomography, magnetic resonance imaging, or both.

Results. Among the admitted patients, the youngest patient was 20 years and the oldest was 42 years, of which 2, 13 and 15 patients were diagnosed in the first, second and third trimesters, respectively. The distribution of neurosurgical problems was as follows: subarachnoid haemorrhage (SAH) n=6, cerebral venous thrombosis (CVT) n=6, brain tumour n=5, trauma n=6, Intracranial haemorrhage (ICH) n=2, VP shunt n=2, tuberculoma n=1, neurocysticercosis n=1, and pituitary adenoma n=1. Fifteen patients (50%) underwent brain operation, eleven patients (36%) had conservative therapy, one patient died and four patients were terminated during pregnancy.

Conclusion. Multidisciplinary teams are not only required to successfully diagnose and treat intracranial lesions but also to safeguard both the mother and child.

Keywords
intracranial lesions, management, neurosurgeons, obstetricians, pregnancy

Corresponding author:
Reeka Pradhan

Paropkar Maternity and Women's Hospital, Thapathali, Nepal

reekapradhan@gmail.com

Scan to access the online version

INTRODUCTION

Intracranial lesions during pregnancy are very uncommon. Such kind of lesions can be life threatening for both mother and fetus. Anatomical and physiological changes lead to aggravate certain intracranial lesions. The biological changes including hormonal, metabolic, and immunological during pregnancy can stimulus women to a higher incidence of neurological problems like subarachnoid hemorrhage (SAH), stroke, pituitary apoplexy, venous sinus thrombosis, pseudotumor cerebri, neoplasms, pre-eclampsia and eclampsia etc. (4, 6, 13, 21) Such kind of complications in pregnancy bring special attention from super specialists due to the worsening of the clinical picture for both the mother and the fetus. There is not solid evidence

whether brain lesions should be addressed during pregnancy or after termination of pregnancy, so there is controversy to the mode of management. This is a challenge for both the obstetrician and neurosurgeon. It is one of the alarmed forms of neurosurgical and obstetrical problems that not only causes significant morbidity and mortality but also results in poor socioeconomic outcome.

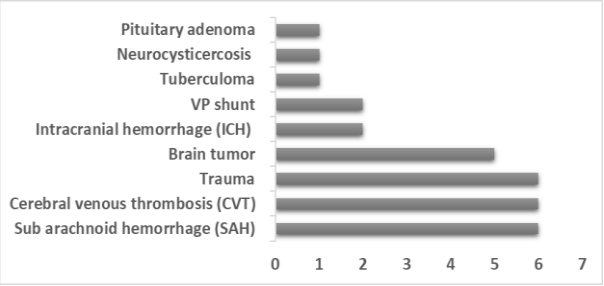
Materials and methods

A retrospective study was done on thirty patients with intracranial lesions during pregnancy who were admitted to the neurosurgical department from 2015 to 2024. All patients were evaluated with either computed tomography (CT), or magnetic resonance imaging (MRI), or both. CT scan brain (non-contrast) was done as a part of routine evaluation with precautions when presented in the emergency department (ER). MRI Brain is the ideal diagnosis tool of intracranial lesions during pregnancy. Patient characteristics like age, gestation age, history of hypertension, GCS on admission, pupil reactivity, laboratory tests, CT scan and MRI results were evaluated. The consciousness level was measured by the Glasgow Coma Scale (GCS). Outcome assessment was based on the data from the medical records of patients during their hospitalization, Glasgow Outcome Score (GOS) and 6-month period after discharge. All the pregnant women with brain lesions were admitted in the neurosurgical intensive care during the study period and were comprised in the study. Postpartum women with intracranial lesions were excluded from our study. Our results are statistically analyzed with SPSS Software.

Results

Two (7%) patients were identified in the first trimester, 13 (48%) and 15 (44%) patients presented in the second and the third trimester respectively. Their clinical features were focal neurologic symptoms (n = 10, 37%), epileptic seizures (n = 11, 41%) and others common symptoms like headache, nausea, vomiting (n = 8, 30). The distribution of neurosurgical patterns was as follows (Table 1): AVM / SAH aneurysm n=6 (figure 1), cerebral venous thrombosis (CVT) n=6, brain tumor n=5, trauma n=6, ICH n=2, VP shunt n=2, tuberculoma n=1, neurocysticercosis n=1, and pituitary adenoma n=1. We noted that three cases of aneurysm and 2 cases of AVM were managed surgically while one patient died due to posterior fossa massive intracranial hemorrhage but the baby survived. All cerebral venous thrombosis (CVT) cases were managed conservatively. There were five cases of brain tumor which were managed with antiepileptic medicines, among them, four were histologically meningioma and one was low grade glioma. There were six trauma cases during pregnancy, among them two cases underwent craniotomy with spontaneous abortion, two cases underwent craniotomy with simultaneous early cesarean section, and two cases were conservatively managed. There were total of two cases of intracranial hemorrhage among which one case underwent evacuation and the other was managed conservatively due to minimum volume. Two cases of congenital hydrocephalus with ventriculoperitoneal (VP) shunt malfunction were managed with emergency VP shunting. Both cases of tuberculoma, cases of neurocysticercosis and pituitary adenoma were managed conservatively. All of these three cases were kept in regular follow up with extra precautions.

Table 1. Distribution of neurosurgical lesions during pregnancy



Discussion

The first case of a pregnant woman with a brain tumor was well-defined by Bernard in 1898. Hagedoorn elaborated effects of pregnancy on intracranial meningioma in 1937 (19, 28, 30). The association between pregnancy and women with meningioma was first described by Cushing and Eisenhardt. Pregnancy may stimulate an underlying brain lesion by factors such as steroid mediated growth, hemodynamic changes and immunogenic changes. Fast growth tumors and its subsequent vasogenic edema can lead to raise intracranial pressure (ICP). However, the ideal time to decide operation during pregnancy is still controversial. It is highly praised to delay surgery if possible until after the first trimester to decrease the risk of miscarriage.

Surgery can be a life-saving or life-changing intervention during the second and third trimesters, (1, 5, 19).

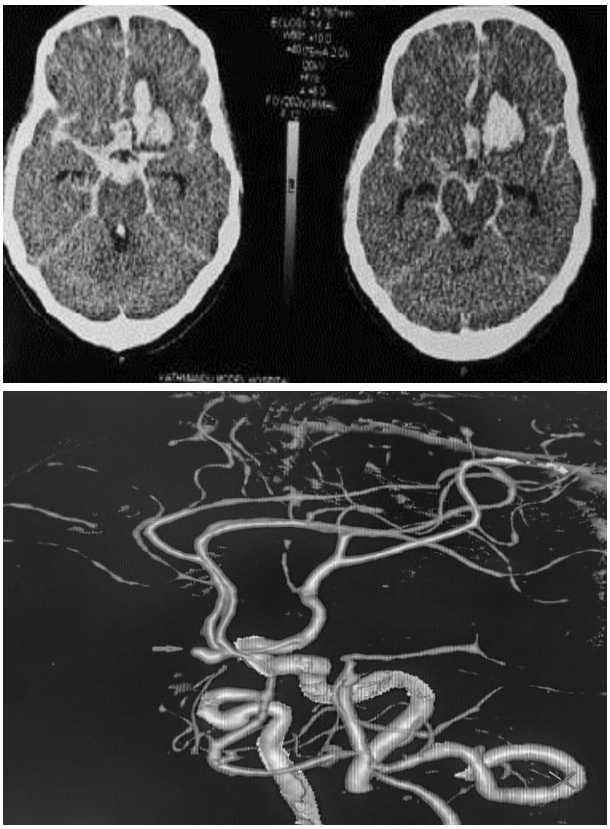


Figure 1. Preoperative axial computed tomography (CT) scans (A) showing diffuse subarachnoid hemorrhage with intra parenchymal hematoma and Computed tomography angiography (CTA) (B) demonstrating anterior communicating aneurysm

It is quite challenging to diagnose intracranial lesions during pregnancy. The clinical presenting of raise intracranial pressure like headache, nausea, altered sensorium, focal seizures, lateralizing neurological deficits, abnormal fundoscopic findings suggest an intracranial mass. Their management should not be misdiagnosed with chronic hypertension, gestational hypertension, hyperemesis gravidarum, pre-eclampsia or eclampsia or puerperal psychosis (10).

CT and MRI revealed the maximum number of diagnostic information. MRI is better over CT because of its greater sensitivity, higher image resolution and the absence of radiation. So, MRI brain is the ideal the diagnosis tool of intracranial lesions during pregnancy. The diagnosis of CVT should be assessed by MRI/magnetic resonance venography (MRV) or CT venography. Homocysteine levels should be calculated, as homocysteinemia may be linked with peripartum CVT. These days, CT scan is not chosen of investigation to avoid the risk of radiation damage (2, 3, 8, 14).

In this study, there were five brain lesions, among them four of common histological type, meningioma, and one was low grade glioma, all of which were managed with antiepileptic medicines. meningiomas are known to express progesterone receptors, which leads to increased severity during pregnancy (12, 25). When superimposed on the physiologic changes of pregnancy, symptoms can be triggered and quite severe in pregnant patients. The management protocol for intracranial tumors during pregnancy should be depends on patient's presentation, gestational age, localization of the tumor, and others medical related factors. Surgical intervention is the best approach for intracranial meningioma but surgical plan should be avoided during pregnancy, when possible, because of the increased risk to both mother and fetus (16, 18). Postpartum surgery of meningioma is recommended by authors in the English literatures. (23, 26).

Cerebral venous thrombosis (CVT) accounts for 6% to 64% of all pregnancy associated strokes. It is related with a hypercoagulable state in 64% of cases and in 73% happened in postpartum (9, 27). Its presentation is same as others presentation like with headache, seizures, focal neurologic signs or ICH. IV heparin, fluid administration, antibiotics for infection, and measures to reduce the increased intracranial pressure are all used in management of CVT. Oral anticoagulation is usually given for 6 months in patients with idiopathic venous thrombosis and indefinitely in those with a persistent or familial thrombophilic state. Mortality from pregnancy associated with CVT is 10% to 50%, with a case-fatality rate in the range of 4% to 36%. (9, 20) The risk for hemorrhagic conversion has been used as the main limitation and dispute against anticoagulation. In our study, all cerebral venous thrombosis (CVT) cases were managed conservatively.

In our study, the most common non traumatic causes of SAH in pregnancy were due to underlying AVMs and aneurysms. There were six cases of non-traumatic SAH among which three were of aneurysm and 2 were of AVM, all of which were managed

surgically while one patient died due to posterior fossa massive SAH. The chances of aneurysm rupture increase several folds during pregnancy and increasing with gestational age until it peaks at 30 to 34 weeks. Dias and Sekhar (7) described that the mortality of pregnancy related aneurysmal SAH to be 35%, with a fetal mortality to be 17%. In a retrospective study of 118 patients, 90% of aneurysmal bleeding occurred during pregnancy, 2% during labor, and 8% postpartum. Six percent of AVM associated ICH occurs during labor and 94% during pregnancy. The treatment of SAH or ICH is not others than surgery during pregnancy. Before and after securing the aneurysm, these patients must stay in the neurosurgical intensive care with monitoring. Clipping of the aneurysm can be achieved in any stage of pregnancy and is related with lower maternal and fetal morbidity and mortality (11, 31).

Trauma is accounted in 8% of all pregnancies. It can be life alarming for both the mother and fetus. It is not only associated with maternal but also fetal morbidity and mortality. It has been elaborated that trauma increases the incidence of spontaneous abortion (SAB), preterm birth, preterm premature rupture of membranes (PPROM), placental abruption, uterine rupture, stillbirth and cesarean delivery (24). The rate of fetal mortality after maternal blunt trauma is 3.4 to 38.0 %, generally from placental abruption, maternal shock, and maternal death. Early evaluation and treatment of the mother in trauma should be addressed as soon as possible, as maternal shock is connected with 80% fetal mortality (15, 17). In our study, there were six cases of trauma during pregnancy. Two trauma cases underwent craniotomy with spontaneous abortion, two underwent craniotomy with simultaneously early cesarean section, and two cases were conservatively managed.

In our study, there were two cases of congenital hydrocephalus (HCP) which were previously shunted and presented with shunt malfunction. They presented with features of raised ICP, which was reconfirmed with MRI. Both cases were managed with emergency VP shunt surgery. HCP is a neurosurgical disorder that is defined as the presence of an abnormal collection of cerebrospinal fluid (CSF) inside the brain ventricles (22, 29). We consider spontaneous vaginal delivery to be the best way to terminate the pregnancy in HCP but neurosurgical intervention is indicated in cases of acute neurological conditions. No standard procedure has been established yet for shunt malfunctions during pregnancy. However, low rates of shunt malfunctions and revisions were found in pregnant women.

A large retrospective population-based study of 644 patients diagnosed with intracranial lesions during pregnancy demonstrated no fundamental association between adverse outcomes and the neurosurgical procedures carried out during the immediate neonatal period. The management protocol for intracranial tumors during pregnancy should be adapted according to patient presentation and status. The general principle of surgery is cesarean section as first surgery and then the neurosurgical intervention when the patient's neurological status and the gestational age allow. (5,7,11). Our study recommendation is postpartum neurosurgical intervention when possible. Emergency neurosurgical interventions should be made in case of patients with malignant tumors, rupture aneurysm, active hydrocephalus or benign intracranial lesions with signs of impending herniation, and progressive neurological deficits. Finally, pregnant with brain lesions require an individualized approach of the institute for their care under the steering of multidisciplinary team. (12, 19)

Conclusion

Multidisciplinary teams including neurosurgeons, neurologists, obstetricians, perinatologists, anesthesiologists, genetics counselors and other health-care professionals' efforts are needed not only to successfully diagnose and treat the underlying pathology but also to ensure the safety of the mother and her unborn child.

References

1. Alluwimi I, Al-Anazi AR. Meningioma in pregnancy. Bahrain Madical Bulletin. 2004 June; 26 (2): 1-8.
2. American College of Radiologists. ARC practice guideline for imaging pregnant or potentially preg nant adolescents and women with ionizing radia tion. American College of Radiologists Council 2008;26:23–37
3. Chen MM, Coakley FV, Kaimal A, Laros RK Jr. Guidelines for computed tomography and mag netic resonance imaging use during pregnancy and lactation. Obstet Gynecol 2008;112:333–40

4. Contag SA, Mertz HL, Bushnell CD. Migraine during pregnancy: Is it more than a headache? Nat Rev Neurol. 2009;5:449–56.
5. Cushing H, Eisenhardt L. Meningiomas. Their classification, regional behaviour, life history, and surgical end result. Springfield, Illinois: 1938.
6. De Luca GC, Bartleson JD. When and how to investigate the patient with headache. Semin Neurol. 2010;30:131–44.
7. Dias MS, Sekhar LN. Intracranial hemorrhage from aneurysms and arteriovenous malformations during pregnancy and the puerperium. Neurosurgery. 1990;27: 855–65.
8. Ducray F, Colin P, Cartalat-Carel S, Pelissou-Guyotat I, Mahla K, Audra P, et al. Management of malignant gliomas diagnosed during pregnancy. Rev Neurol (Paris) 2006;162:322–9.
9. Ferro JM, Canhao P, Stam J, Bousser MG, Barinagarrementeria F ISCVT Investigators. Prognosis of Cerebral vein and dural sinus thrombosis: Results of the International Study on Cerebral Vein and Dural Sinus Thromobosis (ISCVT) Stroke. 2004;35:664–70.
10. Fox AW, Diamond ML, Spierings EL. Migraine during pregnancy: Options for therapy. CNS Drugs. 2005;19: 465–81
11. Giannotta SL, Daniels J, Golde SH, Zelman V, Bayat A. Ruptured intracranial aneurysms during pregnancy. A report of four cases. J Re prod Med. 1986;31:139–47.
12. Gurcay AG, Bozkurt I, Senturk S, et al. Diagnosis, Treatment, and Management Strategy of Meningioma during Pregnancy. Asian J Neurosurg. 2018;13(1):86–89. doi: 10.4103/1793-5482.181115.
13. Haas JF, Janisch W, Staneczek W. Newly diagnosed primary intracranial neoplasms in pregnant women: A population-based assessment. J Neurol Neurosurg Psychiatry. 1986;49:874–80.
14. Harandou M, Madani N, Labibe S, Messouak O, Boujraf S, Benkirane S, Neuroimaging findings in eclamptic patients still symptomatic after 24 hours: A descriptive study about 19 cases. Ann Fr Anesth Reanim 2006;25:577-83.
15. Huls CK, Detlefs C. Trauma in pregnancy. Semin Perinatol. 2018;42:13–20.
16. Christensen J, Hansen L, Garne E. Congenital hydrocephalus-prevalence and prognosis. Mortality and morbidity in a population-based study. Ugeskr Laeger. 2003;165(5):466–469.
17. Isla A, Alvarez F, Gonzalez A, Garcia-Grande A, Perez-Alvarez M, Garcia-Blazquez M. Brain tumor and pregnancy. Obstet Gynecol. 1997;89:19–23
18. Lucia A, Dantoni SE. Trauma management of the pregnant patient. Crit Care Clin. 2016;32:109–17.
19. Lynch JC, Gouvea F, Emmerich JC, Kokinovrachos G, Pereira C, Welling L, et al. Management strategy for brain tumour diagnosed during pregnancy. Br J Neurosurg. 2011;25:225–30.
20. Ng J, Kitchen N. Neurosurgery and pregnancy. J Neurol Neurosurg Psychiatry. 2008;79:745–52.
21. Pabinger I, Grafenhofer H, Kyrle PA, Quehenberger P, Mannhalter C, Lechner K, et al. Temporary increase in the risk for recurrence during pregnancy in women with history of venous thromboembolism. Blood. 2002;100: 1060–2.
22. Physiological changes in pregnancy and their influence on the endocrine investigation. Morton A, Teasdale S. Clin Endocrinol (Oxf) 2022;96:3–11.
23. Priddy BH, Otto BA, Carrau RL, Prevedello DM. Management of skull base tumors in the obstetric population: A case series. World Neurosurg. 2018;113:e 373–82.
24. Rodrigues, A.J.; Waldrop, A.R.; Suharwardy, S.; Druzin, M.L.; Iv, M.; Ansari, J.R.; Stone, S.A.; Jaffe, R.A.; Jin, M.C.; Li, G.; et al. Management of brain tumors presenting in pregnancy: A case series and systematic review. Am. J. Obstet. Gynecol. MFM 2021, 3, 100256.
25. Rosa ML, Loaiza S, Zambrano MA, Escobar MF. Trauma in pregnancy. Clin Obstet Gynecol. 2020;63:447–54.
26. Schrell UM, Adams EF, Fahlbusch R, Greb R, Jirikowski G, Prior R, et al. Hormonal dependency of cerebral meningiomas. Part 1: Female sex steroid receptors and their significance as specific markers for adjuvant medical therapy. J Neurosurg. 1990;73:743–9
27. Sharma JB, Roy KK, Mohanraj P, Kumar S, Karmakar D, Barua J. Pregnancy outcome in pituitary tumors. Arch Gynecol Obstet. 2009;280:401–4.
28. Shehata HA, Okosun H. Neurological disorders in pregnancy. Curr Opin Obstet Gynecol. 2004;16:117–22.
29. Silberstein SD. Headaches in pregnancy. Neurol Clin. 2004;22:727–56
30. Stagno V, Navarrete EA, Mirone G, Esposito F. Management of hydrocephalus around the world. World Neurosurg. 2013;79(2 Suppl):S23.e17–20
31. Stevenson CB, Thompson RC. The clinical management of intracranial neoplasms in pregnancy. Clin Obstet Gynecol. 2005;48:24–37
32. Trivedi RA, Kirkpatrick PJ. Arteriovenous malformations of the cerebral circulation that rupture in pregnancy. J Obstet Gynaecol. 2003;23:484–9.

Romanian Neurosurgery (2025) XXXIX (1): pp. 91-94
DOI: 10.33962/roneuro-2025-017
www.journals.lapub.co.uk/index.php/roneurosurgery

Giant sacral schwannoma mimicking teratoma. A case report

Vissa Shanthi, V. Siva Kumar, Madhu Kumar Vengala, Nandyala Rukmangadha, Bodagala Vijaya Lakshmi Devi, V.V. Ramesh Chandra

Sri Venkateswara Institute of Medical Sciences, Tirupathi, Andhra Pradesh, INDIA

ABSTRACT

Background: Sacral schwannomas are uncommon, constituting 1-5% of schwannomas. They present with vague symptoms. Schwannomas occurring in sacral and presacral regions present with enormous dimensions and are difficult to manage.

Case summary: We present a case of schwannoma occurring in the sacral region extending into the presacral region in a 45-year-old female which was diagnosed as teratoma on imageology. The lesion was excised and on histopathological examination, diagnosis of schwannoma was considered

Conclusion: Sacral schwannoma with cystic degeneration should be considered as a differential diagnosis for solid and cystic lesions of the sacral region. MRI and CT scanning will be helpful in diagnosis.

Keywords
schwannoma,
sacral,
spinal

Corresponding author:
Shanthi V.

Sri Venkateswara Institute of Medical Sciences, Tirupathi, Andhra Pradesh, India

santhijp@gmail.com

Scan to access the online version

INTRODUCTION

Schwannomas are benign tumours originating from schwann cells. These tumours are more common in head and neck, posterior mediastinum and extremities. sacral intraspinal schwannomas are relatively infrequent and accounts for 1-5% of spinal tumours. [1] Schwannomas in spinal cord often results in extensive bony destruction and are detected when they attain enormous dimensions and are termed as Giant schwannoma. 3 types of sacral schwannomas have been described based on anatomical findings i.e. retroperitoneal schwannoma, intra spinal schwannoma and spinal schwannoma. Clinical features vary depending upon type of sacral schwannoma. Retroperitoneal schwannoma has slow growth and presents with non-specific symptoms. [2] Intraspinal schwannoma is an intraosseous lesion presenting with mild low back pain. Presenting with neurological deficit is unusual. [3] spinal schwannoma rise from spinal nerve root and present as dumb-bell tumour. Dumb-bell schwannomas occurring in sacral region comprise 4% of all spinal schwannomas. [4] We are presenting a rare case of giant sacral schwannoma arising from spinal cord mimicking sacral teratoma on imageology.

Case report

A 45 years old female came with chief complaints of pain in the right parasacral region since one month. Pain is radiating to right leg, aggravated on prolonged standing. Patient has history of irregular menstrual cycles from 2 months and dyspareunia. There is no history of abdominal pain, vomiting, fever and melena. Bowel and bladder movements are normal. No history of loss of appetite. Her obstetric history was P4L4.

On local examination (per rectum) induration is felt at 10'0 clock position.

Systemic examination: Per abdomen examination revealed ill-defined palpable mass in the pelvic region with variable consistency. Mild tenderness is present. Bowel sounds are normal.

Tone, bulk and power of both lower limbs are normal. Haematological parameters are with in normal limits (Haemoglobin: 13.1g%, RBC count 4.48 million/microliter, Total count – 6500/ microliter, ESR-16mm/hour, PCV- 39%, MCV – 87fl, MCH 27pg, Platelet count – 3.15 lakhs/microliter). Serum Urea – 13mg/dl, serum creatinine – 0.5 micromoles/L, Serum electrolytes like serum sodium – 142 mmol/L, Serum potassium – 4.1mmol/L, Serum chloride – 106mmol/L, Serum chloride 106mmol/L. random plasma glucose is 101mg/dL.

Contrast enhancing computed tomography (CECT) revealed large lobular sacral lesion with cortical destruction and presacral soft tissue component measuring 8.2X8.5X7.2cms suggesting teratoma.

Magnetic resonance imaging of pelvis shows well defined altered signal intensity lesion of size 8.5X7.1X7ms which is heterogeneously hyper intense on T2, hypo and isointense on T1 showing areas of restricted diffusion on DW1 and heterogenous enhancement post contrast with few cystic areas with in are noted (Figure 1). Lesion is arising from spinal cord at S2-S3 vertebral body region causing expansion of spinal canal at that region, involving S2 vertebral body extending into right neural foramina at S2-S3 level with extension into right presacral region. Lesion is causing displacement of uterus and rectum anterolateral with maintained fat planes. Clinical diagnosis of sacral teratoma is considered.

Excision of retroperitoneal tumor was done which is 8x8cms well encapsulated tumor at the level of S2-S3 with pedicle extending into right sacral foramina.

Grossly we received encapsulated soft tissue mass measuring 7X6X4.5cms. Cut section of lesion is lobulated, grey white with focal cystic and calcified areas (Figure 2).

Microscopically, capsulated lesion was composed of hyper and hypocellular areas with spindle shaped cells. Few foci shows cells showing palisading with verocay bodies (Antoni A areas) (Figure 3). Many foci showed cystic spaces with adjacent hyalinised tissue (Figure 4). Foci of calcification are also noted (Figure 5). Congested blood vessels with hyalinised walls are noted. On immunohistochemistry, spindle shaped cells were positive with S-100. These histopathological features are consistent with schwannoma (Figure 6).

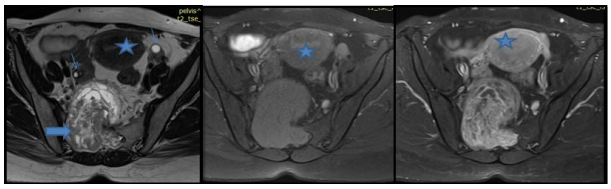


Figure 1. MRI pelvis - T2W, T1W and post contrast T1W axial images showing large heterogeneously enhancing lesion (thick arrow) in the spinal canal extending into presacral region through widened right neural foramina suggestive of neurogenic tumour. normal appearing uterus and ovaries (thin arrow) are seen separately

Discussion

Spinal schwannomas are more common in thoracic or lumbar region when compared to sacral region. [5] Retroperitoneal schwannomas are encapsulated tumors, often located in presacral and Para spinal areas. Pathogenesis of schwannoma is not clear but their occurrence may be associated with gene mutation. [6]

Peripheral nerve schwannoma arise from Schwann cells, involving one or two fascicles and displacing the other fascicles of involved nerve. They occur within endoneurium. Perineurium and fibrous epineurium surrounds the tumor and encapsulates it. [7]

Sacral schwannomas have low growth rate and have the ability to conceal due to the large pelvic space which leads to delayed diagnosis. Retroperitoneal schwannoma have mean growth rate of 1.9mm/year. Majority of these tumors are

solitary (96%) and only 4% are plexiform. [8] According to anatomic location, sacral schwannomas are divided into retroperitoneal, intraosseous, and dumbbell type. Retroperitoneal schwannomas are located in posterior peritoneum, outside the spinal cord. Intra osseous type are located in and invade spinal canal. Dumbbell type schwannoma extends outside the spinal cord through intervertebral foramen. [9]

Sacral schwannoma is asymptomatic when it is smaller in size but as the size increases patient presents with radicular pain to legs and reaches an enormous size at the time of presentation. Definite criteria for the term giant schwannoma is not defined but commonly accepted criteria is tumor extending into more than two vertebral bodies, extending into adjacent myofascial planes and extra spinal extension should be more than 2.5cms. [10]

Degenerative changes such as cyst formation can occur either because of degenerative changes in Antoni B areas or due to ischemic necrosis. Other degenerative changes are necrosis, fibrosis, haemorrhage, calcification and heterogenous cellularity which gives heterogenous signal intensities of contrast enhanced T 1 weighted images and T 2 weighted images. MRI features of typical schwannoma and giant sacral schwannoma are similar except when there is degenerative changes in giant sacral schwannoma. [11]

Patients with sacral schwannoma present with various clinical symptoms like nerve tissue or pelvic organ compression, sciatica, lower back pain, bone destruction, bladder and rectum compression and weakness in lower limbs. [12]

Common neoplastic lesions which have sacral origin are malignant tumors like chondrosarcoma, chordomas and metastatic lesion, while common benign tumors having sacral origin are giant cell tumor, osteoblastoma, aneurysmal bone cyst where as schwannoma are very rare. Schwannomas with degenerative changes can mimic other tumors in imageology as was in our case which was clinically diagnosed as teratoma. However histopathologically it is easy to differentiate the tumor from other entities

Complete resection of tumor is the treatment of choice to prevent the recurrence. However in giant invasive sacral schwannoma complete resection is difficult due to invasive growth pattern, hence piecemeal subtotal excision is choice and can achieve good outcome. Malignant transformation and local recurrence are extremely rare.

Conclusion

Sacral schwannoma are rare benign tumors and their clinical diagnosis is frequently delayed, as the symptoms are produced only when it reaches huge size. Sacral schwannoma with cystic degeneration should be considered as differential diagnosis for solid and cystic lesions of sacral region. MRI and CT scanning will be helpful in diagnosis. Specifically, CT scanning demonstrates bony destruction and helps for preoperative planning. Complete resection is treatment of choice but piecemeal subtotal excision in tumors with invasive growth pattern also achieves good outcome.

References

1. Cagatay Ozturk, Cuneyt Mirzanli, Omer Karatoprak, Mehmet Tezer, Mehmet Aydogan, Azmi Hamzaoglu. Giant sacral schwannoma: A case report and review of literature. Acta Orthop.Belg. 2009;75: 705-710
2. Theodosopoulos T, Stafyla V K, Tsiantoula P, Yiallourou A, Marinis A, Kondi Pafitis Aet al. Special problems encountering surgical management of large retroperitoneal schwannoma. World J Surg Oncol, 2008;6:107
3. Salvant JB, Young HF. Giant intrasacral schwannoma: an unusual cause of lumbosacral radiculopathy Surg Neurol. 1994;41:411-3
4. Ozawa H, Kokubun S, Aizawa T, Hoshikawa T, Kawahara C. Spinal dumb bell tumor: an analysis of a series of 118 cases. J Neurosurg spine 2007;7:587-93
5. Han-Alee, Se Jeong Jeon, See Sung Choi, Hye- Wonkim, HunSoo Kim. A purely cystic giant sacral schwannoma mimicking bone cyst. A case report. J Korean Soc Radiol.2014;71(1):1-5
6. Shunchang Zhou, Shenvi Wan, Liang Li, Wei Dong, Xuhui Ma, Haibu chu, Yuxu Zhong. Rare retroperitoneal giant sacral schwannoma: A case report. Oncol Lett. 2024;27(6):261. doi: 10.3892/ol.2024.14394. PMID: 38646494; PMCID: PMC11027106.
7. Suárez C, López F, Rodrigo JP, Mendenhall WM, de Bree R, Mäkitie AA, Vander Poorten V, Takes RP, Bondi S, Kowalski LP, Shaha AR, Fernández-Alvarez V, Gutiérrez JC, Zidar N, Chiesa-Estomba C, Strojan P, Sanabria A, Rinaldo A, Ferlito A. Benign Peripheral Non-cranial Nerve Sheath Tumors of the Neck. Adv Ther. 2022 Aug;39(8):3449-3471. doi: 10.1007/s12325-022-02191-5. Epub 2022 Jun 11. PMID: 35689724.
8. El Sayed L, Masmejean EH, Parfait B, Kalamarides M, Biau D, Peyre M. Natural history of peripheral nerve schwannomas. Acta Neurochir (Wien). 2020

Aug;162(8):1883-1889. doi: 10.1007/s00701-020-04430-6. Epub 2020 Jun 16. PMID: 32556523.

9. Handa K, Ozawa H, Aizawa T, Hashimoto K, Kanno H, Tateda S, Itoi E. Surgical Management of Giant Sacral Schwannoma: A Case Series and Literature Review. World Neurosurg. 2019 Sep;129:e216-e223. doi: 10.1016/j.wneu.2019.05.113. Epub 2019 May 21. PMID: 31125780
10. Sridhar K, Rammurthi R, Vasudevan M C, Ram murthy B, Giant invasive spinal schwannomas: definition and surgical management. J Neurosurg 2001;942Suppl:210-215.
11. Yu HN, Lee SE, Jahng TA, Chung CK. Giant invasive spinal schwannoma: its clinical features and surgical management. Neurosurgery 2012;71: 58-67
12. Khan U A, Ismayl G, Malik I. Giant sacral schwannoma treated with a 360 approach: A rare case and systematic review of the literature. World Neurosurg.2018;115:65-72

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.

Acceptable percentage of resemblance - 5%.
Duplicate content in research papers shall be considered only up to 5% of the total content.

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

The manuscript sent for publishing must be submitted in English.

Authors shall ensure that the article has been spell and grammar checked prior to the submission.

The manuscript will be typed without formatting, with 1-line space. Please enclose one 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 using the online submission platform or to the Editor:

Vicentiu Saceleanu, MD Dr.
(vicentiu.saceleanu@ulbsibiu.ro)

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.