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

Romanian
NEUROSURGERY

Vol. XXXVII | No. 3 September 2023

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

LONDON ACADEMIC PUBLISHING

EDITORIAL AND ADVISORY BOARD

EDITOR-IN-CHIEF

Assoc. Prof. Vicentiu Saceleanu, MD, PhD
vicentiu.saceleanu@ulbsibiu.ro

ASSISTANT EDITORS

Andrei Marinescu
marinescu.andrei.alex@gmail.com

Cosmin Cindea
cindea.cos@gmail.com

Titus Fagarasi
fagarasi.titus@gmail.com

ADVISORY BOARD – ROMANIA

Prof. Horia Ples
Prof. Stefan I. Florian
Prof. Mircea Gorgan
Assoc. Prof. Lucian Eva
Prof. G.B.I. Iacob
Prof. I. Poeata
Prof. Aurel Mohan
Assoc. Prof. Adrian Balasa
MD Marius Dabija

ADVISORY BOARD - INTERNATIONAL

Prof. M.A. Arraez, Spain
Prof. H. Bertalanffy, Germany
Prof. J. Brotchi, Belgium
Prof. Y. Kato, Japan
Prof. U. Kehler, Germany
Prof. Christopher M. Loftus, USA
Dr M.R. Mahmud, Nigeria
Prof. P. Mertens, France
Prof. B.K. Misra, India
Prof. D.F. Muresanu, Romania
Prof. S.C. Robertson, USA
Prof. M. Samii, Germany
Prof. J. Schramm, Germany
Prof. M. Sindou, France
Prof. M. Tatagiba, Germany
Prof. M. Buchfelder, Germany
Prof. I. Solaroglu, Turkey
Prof. T.T. Wong, Taiwan

EMERITUS EDITORIAL BOARD

Prof. A.V. Ciurea,
Honorary Member of the Romanian Academy

Assoc. Prof. Habil. H. Ples, Romania,
Former *Editor-in-Chief*

Assoc. Prof. St. M. Iencean, Romania,
Former *Editor-in-Chief*

Prof. Al. Constantinovici_
Former *Editor-in-Chief*

FOUNDING EDITOR

Prof. Constantin Arseni

EXECUTIVE EDITOR

Madalin Onu, PhD

ROMANIAN

NEUROSURGERY

Vol. XXXVII | No. 3 September 2023

London
Academic Publishing

Copyright © 2023 Romanian Society of Neurosurgery &
London Academic Publishing

All rights reserved. This book or any portion thereof may not be reproduced or used in any manner whatsoever without the express written permission of the Romanian Society of Neurosurgery or the publisher except for the use of brief quotations in a book review or scholarly journal.

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

First Printing: September 2023
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

The opinions expressed in the published articles are the sole responsibility of the authors and do not reflect the opinion of the editors or members of the editorial board.

Contents

Romanian Neurosurgery (2023) XXXVII (3): pp. 249-251
DOI: 10.33962/roneuro-2023-045
www.journals.lapub.co.uk/index.php/roneurosurgery

A rare case of Yhwag gene mutation causing developmental and epileptic encephalopathy

Karina Lidia Gheorghita[1], A.V. Ciurea[2,4], R.E. Rizea[3,4]

[1] Private Practice, Bucharest, ROMANIA
[2] Sanador Clinical Hospital, Bucharest, ROMANIA
[3] Bagdasar-Arseni Hospital, Bucharest, ROMANIA
[4] "Carol Davila" University of Medicine and Pharmacy Bucharest, ROMANIA

ABSTRACT

Background: epileptic encephalopathy 56 or DEE is a rare disease characterized by early-onset treatment-refractory epilepsy accompanied by global developmental regression that has been shown to be caused by various mutations of the YWHAG gene.
Case presentation: We report a novel of a heterozygous mutation of YHWAG c.170G>A, p.(Arg57His), in a Caucasian male.
Conclusions: Our report further confirms that mutation of YWHAG results in developmental and epileptic encephalopathy.

Keywords
YWHAG,
developmental and epileptic encephalopathy,
neurocognitive disorder,
autistic spectrum disorder,
polymorphic seizures,
Arnold-Chiari malformation type I,
parietal dystrophy

Corresponding author:
R.E. Rizea

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

rizea.radu.eugen@gmail.com

Scan to access the online version

BACKGROUND

Developmental and epileptic encephalopathy 56 (DEE 56)is a severe genetic disorder with early-onset, refractory, polymorphic seizures and developmental delay. YWHAG, known as the Tyrosine 3-Monooxygenase/ Tryptophan 5 Monooxygenase Activation Protein Gamma, was recently introduced in the international databases as responsible for the symptoms of epileptic encephalopathy type 56. [1-4]

It belongs to the 14-3-3 family of proteins and it is highly expressed in brain, skeletal muscle, and heart. The 7q11.23 deletion variant was found to cause Williams Beuren syndrome, which associates infantile spasms and cardiomegaly, this being a YWHAG related gene.

Multiple variants of the YWHAG gene have been reported to cause autistic spectrum disorder accompanied by epilepsy refractory to treatment, the causal relationship between these two being still inconclusive.

CASE PRESENTATION

We report a rare case of a Caucasian male with early onset epilepsy, autistic spectrum disorder, several brain malformations and scoliosis with the heterozygous mutation of YHWAG gene c.170G>A, p.(Arg57His). [1]

We present a 21-year-old male, born naturally after extended labor, having at birth a weight of 2950 grams, a length of 52 cm and normal head circumference. A double circular cord is mentioned, but it did not require resuscitation. He had a normal development until six months old. After that he presented infantile spasms and developmental delay.

At the age of 11 months in a febrile context, the child presented the first generalized tonic-clonic convulsion, and the diagnosis of epilepsy was established, recommending treatment with Valproic Acid, but the child continued to present tonic- clonic generalized seizures.

Topiramate was initially added, without significant improvement. After that Levetiracetam was added to valproic acid, but the seizures continued with a frequency of 1 to 3-4 episode per month. Clobazam was added, with a slight improvement in the frequency of epileptic seizures.

After that, the child was recommended ketogenic diet.

In that time the child's seizures stopped for 8 months and 3 weeks, being the longest period of absence of crises since the debut.

The crises returned, most of the time in the form of absences, sometimes accompanied by pallor and gestural automatisms, lasting 1-2 min, with constant progression towards generalized tonic-clonic seizure, these having a frequency of 1 episode every 3 weeks to a month.

Four years after, Clobazam was replaced by lamotrigine and carbamazepine, with a slight improvement in the frequency of epileptic seizures.

A combination of valproic acid, carbamazepine, lamotrigine and pregabalin managed to keep the seizures under control for about 1 year.

During this time, the child underwent permanent language development therapy and physical therapy.

At the age of 15, the child presented frequent, daily, polymorphic, generalized and focal seizures, which is why it was recommended to perform a brain MRI which revealed Arnold Chiari I malformation and focal cortical dysplasia, indicating neurosurgical intervention.

In 2019, the patient underwent a neurosurgical intervention for cortical dysplasia, but the epileptic seizures reappeared 5 days later.

Later it was decided to carry out a genetic test of total exon sequencing, supplemented by total genomic sequencing which revealed the YWHAG mutation. Patient's parents and sister did not present the mutation.

Discussion

Considering the negative family history for genetic diseases, the patient's genetic samples were tested for de novo variants of recessive inheritance

Sequence analysis using Whole Exome Plus identified a heterozygous missense variant YWHAG c.170G>A, p.(Arg57His).

This variant is absent in the international genetic databases. YWHAG c.170G>A, p.(Arg57His) affects an amino acid. The YHWAG mutation was previously described as an epileptic and neurodevelopmental encephalopathy presenting with generalized tonic-clonic seizures, global developmental delay and aspects of autistic spectrum disorder.

There have been reported different missense variants affecting the same codon as de novo in patients with epilepsy and developmental issues. [1,4,7-9]

YWHAG, Tyrosine 3- Monooxygenase/ Tryptophan 5 Monooxygenase Activation Protein Gamma, belonging 14-3-3 protein family affects several cellular processes implicated in neurological and cancer diseases. This protein is expressed in brain and skeletal muscle. The p value of YWHAG gene in the gnomAD variant database is 0.96. This value shows intolerance for loss-of-function variation. The observed reference population in gnomAD is 54 and the expected number is 159, showing a Z score of 2.95, which indicates the intolerance of missense variation. [1,5-7]

Until now, only de novo heterozygous missense YWHAG variants have been reported to be associated with developmental and epileptic encephalopathy 56, characterized by polymorphic early-onset seizures such as focal seizures with motor debut, myoclonic seizures, limb jerks, absences, generalized tonic-clonic seizures, febrile seizures, developmental delay, associated with behavioral abnormalities such as autism, deficit of attention with or without hyperactivity and anxiety. Other described features include scoliosis, hyperlaxity of the joints, receptive and expressive language delay, hypotonic syndrome and different motor difficulties. [1,2,3,5-9]

Treatment and prognosis

In the present case, the patient required a complex therapeutic regimen of antiepileptics. Usually, epilepsy caused by YWHAG gene mutation requires complex combinations of antiepileptics including: valproic acid, levetiracetam, lamotrigine, carbamazepine, topiramate and lately stiripentol. Some of patients develop a phenomenon of tolerance to antiepileptic drugs, seizure control being obtained only for short periods of time. It is very important for patient to have multidisciplinary approach due to the particularity of each other's case. We believe that the main concern of the therapeutic team should be the early genetic diagnosis and the rigorous medicinal control of the seizures because it could allow the neuropsychomotor development of the patient and avoid the formation of a severe clinical picture. [1,2,3]

Conclusion

YWHAG c.170G>A, p.(Arg57His) is a highly pathogenic gene, that produces a genetic neurodevelopmental encephalopathy which requires a correct multidisciplinary approach where the main role is played by the specific neurological approach to epileptic manifestations. The risk of inheriting the variant and of being affected is 50%. We recommend genetic counseling for the patient and his family.

Abbreviations:
AD= autosomal dominant;
AR = autosomal recessive;
DEE= developmental and epileptic encephalopathy;
gnomAD = genome Aggregation Database;
gnomAD AC/AN = allele count/allele number in the genome Aggregation Database.

References

1. Zhi Yi, Zhenfeng Song, Jiao Xue et al. A Novel Heterozygous Missense Variant in YWHAG Gene Cause Early-Onset Epilepsy in a Chinese Family, 31 December 2020, PREPRINT (Version 1) available at Research Square [https://doi.org/10.21203/rs.3.rs-136482/v1]
2. Guella I, McKenzie MB, Evans DM, Buerki SE, Toyota EB, Van Allen MI et al: De Novo Mutations in YWHAG Cause Early-Onset Epilepsy. American journal of human genetics 2017; 101: 300-310.
3. Scheffer IE, Berkovic S, Capovilla G, Connolly MB, French J, Guilhoto L et al: ILAE classification of the epilepsies: Position paper of the ILAE Commission for Classification and Terminology. Epilepsia 2017; 58: 512-521.
4. Helbig I, Tayoun AA: Understanding Genotypes and Phenotypes in Epileptic Encephalopathies. Molecular syndromology 2016; 7: 172-181.
5. Cornell B, Wachi T, Zhukarev V, Toyo Oka K: Overexpression of the 14-3 3gamma protein in embryonic mice results in neuronal migration delay in the developing cerebral cortex. Neuroscience letters 2016; 628: 40-46.
6. Wachi T, Cornell B, Marshall C, Zhukarev V, Baas PW, Toyo-oka K: Ablation of the 14-3-3gamma Protein Results in Neuronal Migration Delay and Morphological Defects in the Developing Cerebral Cortex. Developmental neurobiology 2016; 76: 600-614.
7. Kanani F, Titheradge H, Cooper N, Elmslie F, Lees MM, Juusola J et al: Expanding the genotypephenotype correlation of de novo heterozygous missense variants in YWHAG as a cause of developmental and epileptic encephalopathy. American journal of medical genetics Part A 2020; 182:713-720.
8. Horie M, Suzuki M, Takahashi E, Tanigami A: Cloning, expression, and chromosomal mapping of the human 14-3-3gamma gene (YWHAG) to 7q11.23. Genomics 1999; 60: 241-243.
9. Komoike Y, Fujii K, Nishimura A, Hiraki Y, Hayashidani M, Shimojima K et al: Zebras gene knockdowns imply roles for human YWHAG in infantile spasms and cardiomegaly. Genesis (New York, NY : 2000) 2010; 48: 233-243.

Acknowledgements
We thank to the Blueprintgenetics testing team and the patient and his family for cooperation.

Romanian Neurosurgery (2023) XXXVII (3): pp. 252-255
DOI: 10.33962/roneuro-2023-046
www.journals.lapub.co.uk/index.php/roneurosurgery

Delayed diagnosis and management of a congenital intranasal meningoencephalocele in a 24-year-old male. A case report revisiting the 'bath-plug' technique

Ahtesham Khizar[1], Adeel Niaz[1,2], Syed Muhammad Azeem[1,3], Imran Ali[1], Khalid Mahmood[1]

[1] Punjab Institute of Neurosciences, Lahore, PAKISTAN
[2] Lahore General Hospital, Lahore, PAKISTAN
[3] Sir Ganga Ram Hospital/FJMU, Lahore, PAKISTAN

Keywords
congenital, intranasal, meningoencephalocele, CSF rhinorrhoea, endoscopy

Corresponding author:
Ahtesham Khizar

Punjab Institute of Neurosciences, Lahore, Pakistan

arwain.6n2@gmail.com

ABSTRACT

Background: Intranasal meningoencephalocele is a rare congenital malformation. It is defined by protrusion of cerebral tissue and meninges through a defect in the skull base. Serious complications usually occur in early childhood, however, symptoms occasionally can wait until adulthood.

Case Presentation: We report a case of a 24-year-old male who had a congenital intranasal meningoencephalocele with recurrent episodes of bacterial meningitis. After a delayed diagnosis, he eventually underwent Endoscopic Endonasal Repair of Meningoencephalocele via 'Bath-plug' technique.

Conclusions: To prevent improper management, it is crucial that ENT colleagues, to whom these cases are primarily present, are well informed about this unusual condition. For the purpose of preventing further neurological problems, thorough neuroimaging evaluations and successful surgical repair are important. The 'Bath-plug' technique for anterior skull base cerebrospinal fluid leak repair is effective for a wide range of situations.

Scan to access the online version

BACKGROUND

Intranasal meningoencephalocele is a rare condition and it is characterized by protrusion of the meninges into the nasal cavity through a defect in the cribriform plate of the ethmoid bone. Although it is typically a congenital defect, it can also occur as a result of trauma or persistently elevated intracranial pressure. Meningitis and rhinorrhea can be symptoms; however, the condition can also remain asymptomatic until adulthood. There have been reports of nasal obstruction or unilateral nasal congestion.[1]

Case presentation

A 24-year-old male presented to us through outpatient with complaints of right sided nasal discharge since childhood. This was a clear watery discharge and had a salty taste. There was also a complaint of right nasal obstruction and decreased sense of smell due to swelling inside the nose. There were some episodes of epistaxis as well. He had been hospitalized twice due to meningitis in the last 8 years and on the last occasion he remained on ventilatory support for 7 days. On his last visit to an ENT surgeon, he was diagnosed with an intranasal meningoencephalocele and referred to us for further management. His Magnetic Resonance Imaging (MRI) brain and paranasal sinuses revealed an intranasal meningoencephalocele (Figure.1).

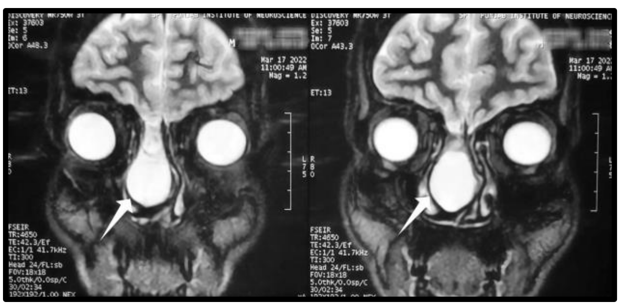


Figure 1. MRI Brain T2 weighted coronal image showing intranasal meningoencephalocele (pointed by white arrows).

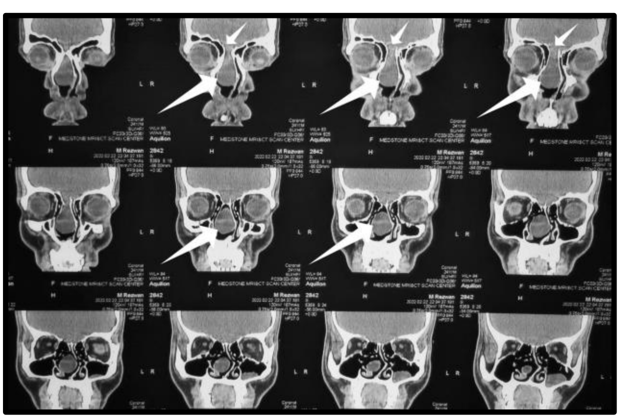


Figure 2. CT paranasal sinuses with FESS protocol coronal image showing defect in the anterior skull base (pointed by small white arrows) and intranasal meningoencephalocele (pointed by large white arrows)/

We performed a Computed Tomography (CT) paranasal sinuses with FESS (Functional Endoscopic Sinus Surgery) protocol for detailed study of meningoencephalocele (Figure. 2). After baseline investigations and anaesthetic fitness, he underwent Endoscopic Endonasal Repair of Meningoencephalocele via 'Bath-plug' technique. The defect was approximately 13 mm in size. Per-operative endoscopic views of intranasal meningoencephalocele can be seen in Figure. 3 A&B. The 'Bath-plug' technique consists of introducing a fat plug with a specially secured vicryl suture into the intradural space and then placing some traction on the suture to seal the defect similarly as a bath plug seals a bath. This repair was superimposed by dural sealant. Lumbar drain placed preoperatively was removed on the second postoperative day. Postoperative period was uneventful.

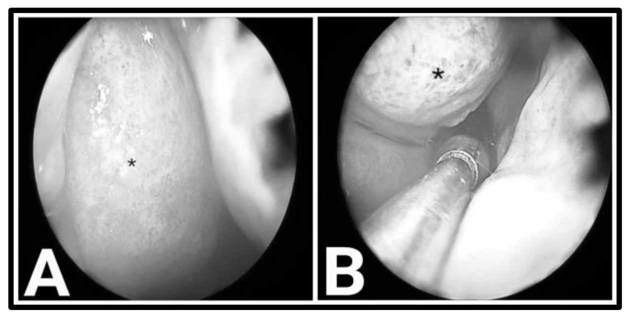


Figure 3. A & B: showing per-operative endoscopic views of intranasal meningoencephalocele (marked by black asterisks).

Discussion

Congenital malformations like intranasal meningoencephalocele are extremely uncommon. The signs and symptoms of an intranasal meningoencephalocele can be mistaken for nasal polyps. To prepare for the right surgical approach, it is crucial to identify the type of basal meningoencephalocele that is now present. CT is helpful to delineate the encephalocele sac. It also helps for the precise position and extent of the cranial bone defect. 3D CT is particularly useful in planning an operative technique.[1] To avoid fatal neurological complications from intranasal meningoencephalocele, a thorough neuroradiological assessment and appropriate surgical technique are crucial.[2]

Recurrent meningitis is a rare occurrence. Giunta et al[3] reported a young woman with an intranasal meningoencephalocele who experienced 18 episodes of meningitis. Recurrent meningitis following a number of nasal polypectomy procedures has also been reported due to erroneous diagnosis.[4] The significance of MRI in diagnosis and treatment is emphasised by these authors as there have been cases of intranasal meningoencephalocele that were initially diagnosed as nasal polyps and resulted in cerebrospinal fluid (CSF) rhinorrhea.[4] Meningoencephaloceles confined to the nose are uncommon in adults. Khan and Salahuddin[5] described the treatment of an adult with a large intranasal meningoencephalocele, and how removal of this lesion caused a gap in the cribriform plate, which was afterwards repaired with tissue and fibrin glue. Bykova et al[6] also reported an intranasal meningoencephalocele in a 64 year old female, which clinically resembled a nasal polyp but histological analysis revealed an aberrant glial tissue.

Meningoencephaloceles are thought to be caused by a developmental defect in the closure of the neural tube.[7] These may manifest as a single deformity, but complex malformations of the face, skull, and brain are also possible. Meningoencephaloceles are classified based on the location of the defect in the cranial bones.[7] Suwanwela et al[8] classified anterior basal meningoencephaloceles into four subgroups: 1) transethmoidal, 2) spheno-ethmoidal, 3) transsphenoidal and 4) frontosphenoidal or spheno-orbital. The fronto-ethmoidal region of the anterior skull has been identified in prior studies as being the area most commonly impacted. According to a theory, the rostral neuropore, which is the last portion of the neural tube to seal, is prone to aberrations throughout cranial development.[7]

Intranasal meningoencephaloceles can only be treated surgically. Transmaxillary route was used to execute the first successful intranasal meningoencephalocele surgery. An endoscopic transnasal technique can be considered to reduce surgical risks if the defect in the skull base is minimal.[2] For Endoscopic Endonasal Repair of Meningoencephalocele, a 'Bath-plug' technique has been reported in literature[9] and it entails inserting a fat plug with a specially secured vicryl suture into the intradural space and applying traction on the suture to seal the defect similarly to how a bath plug seals a bath. The 'Bath-plug' technique for closure of anterior skull base CSF leaks is a reliable technique for a large variety of cases.[10]

Conclusion

A congenital defect of the skull base, including meningoencephalocele, should be taken into consideration when a patient has recurrent intracranial infection but no history of immunodeficiency, cranial trauma, or neurosurgical operation. To prevent improper management, it is crucial that ENT colleagues, to whom these cases primarily present, are well informed about this unusual condition. For the purpose of preventing further neurological problems, thorough neuroimaging evaluations and successful surgical repair are important. The "Bath-plug" technique for anterior skull base CSF leak repair is effective for a wide range of situations.

Abbreviations:
CT: Computed Tomography;
MRI: Magnetic Resonance Imaging;
FESS: Functional Endoscopic Sinus Surgery;
CSF: Cerebrospinal fluid.

References

1. Kumar KK, Ganapathy K, Sumathi V, Rangachari V, Sundararajan I, Govindaraj R. Adult intranasal meningoencephalocele presenting as a nasal polyp. Journal of clinical neuroscience. 2005 Jun 1;12(5):594-6.
2. Hasegawa T, Sugeno N, Shiga Y, Takeda A, Karibe H, Tominaga T, Itoyama Y. Transethmoidal intranasal meningoencephalocele in an adult with recurrent meningitis. Journal of clinical neuroscience. 2005 Aug 1;12(6):702-4.
3. Giunta G, Pappagallo G, Piazza I, Girardi A. A rare case of recurrent meningitis: intranasal encephalocele. Minerva Medica. 1990 Jul 1;81(7-8):577-81.
4. López Aguado D, López Campos D, Pérez Piñero B, Campos Bañales ME. Meningoencefalocele intranasal. A propósito de un caso [Intranasal meningoencephalocele. A case report]. Acta Otorrinolaringol Esp. 2000;51(5):441-444.
5. Khan MA, Salahuddin I. Intranasal meningoencephalocele and the use of fibrin glue. Ear, nose & throat journal. 1997 Jul;76(7):464-7.
6. Bykova VP, Grobov AO, Kuznetsova IL, Serbina VI. An intranasal meningoencephalocele in a 64-year-old woman. Arkhiv Patologii. 1993 May 1;55(3):75-7.
7. Pollock JA, Newton TH, Hoyt WF. Transsphenoidal and transethmoidal encephaloceles: a review of clinical and roentgen features in 8 cases. Radiology. 1968 Mar;90(3):442-53.
8. Suwanwela C, Suwanwela N. A morphological classification of sincipital encephalomeningocele. J Neurosurg 1972; 36: 201–211.
9. Wormald PJ, McDonogh M. 'Bath-plug'technique for the endoscopic management of cerebrospinal fluid leaks. The Journal of Laryngology & Otology. 1997

Nov;111(11):1042-6.
10. Wormald PJ, McDonogh M. The bath-plug closure of anterior skull base cerebrospinal fluid leaks. Am J Rhinol. 2003;17(5):299-305.

Acknowledgements
We would like to acknowledge the efforts of Professor Dr. Khalid Mahmood for Endoscopic Endonasal Skull Base and Minimally Invasive Brain Surgery in Pakistan.

Romanian Neurosurgery (2023) XXXVII (3): pp. 256-262
DOI: 10.33962/roneuro-2023-047
www.journals.lapub.co.uk/index.php/roneurosurgery

A prospective observational study on evaluating the efficacy of bedside optic nerve sheath diameter in assessing clinical progression of patients admitted in neurosurgical ICU with comparisons to CT scans and GCS score

Sunil Munakomi[1], Dipak Chaulagain[2], Binod Bhattarai[1], Shashi Bhushan Shah[1]

[1] College of Medical Sciences, Bharatpur, NEPAL
[2] Uzhhorod National University, Uzhhorod, UKRAINE

ABSTRACT

Background: Ultrasound of the optic nerve sheath diameter (ONSD) is a non-invasive, repeatable tool that can be used to measure intracranial pressure in a dynamic way with high diagnostic accuracy. The goal of this study was to find out if a bedside ultrasonographic measurement of optic nerve sheath diameter (ONSD) can accurately predict the computed tomography (CT) findings of high intracranial pressure (ICP) and changes to the Glasgow Coma Scale (GCS) in adult head injury patients in the Neurosurgery ICU.

Methods: For 54 patients in the neurosurgical intensive care unit, we conducted a retrospective analysis of the results of cranial ultrasounds. Those under the age of 18 and those with apparent visual injuries were ineligible. Both horizontal and vertical optic nerve sheath diameters were measured 3 mm beneath the globe in each eye using a 7.5-10MHz ultrasonographic probe. A binocular change in optic nerve sheath diameter of more than 2.00 mm was deemed abnormal in two consecutive readings in the same patient. Patients in the neurosurgical ICU were given a GCS score, which was used to classify their level of brain damage as mild, moderate, or severe. The accuracy of the optic nerve sheath diameter was evaluated using cranial CT findings of shift, oedema, or effacement that suggested an increased intracranial pressure.

Results: The research has 54 participants. According to the results, 68.5% of those who took the study were men, while 31.5% of those who did so were women. Nearly 16.7 per cent of respondents were between the ages of 18 and 40, while 40.7% of respondents were between the ages of 40 and 60, and 42.6% of respondents were above 60. The significant change in ONSD- fall in GCS and CT-progression scan-findings correlation was very strong. When compared to CT scan progression, the ONSD bedside sonographic test had an 86.7% sensitivity and an 89.7% specificity for detecting elevated ICP. The Positive Predictive Value of the reduction in GCS with advancement in CT scan was 80% and the Negative Predictive Value was 89.7%, respectively.

Conclusions: The sensitivity, specificity, and positive predictive value of bedside ONSD ultrasonography in predicting high intracranial pressure are significant to those of progression in CT scan and drop in GCS. An ONSD bedside measurement may be used to determine elevated ICP since it is non-invasive and repeatable.

Keywords
intracranial pressure,
optic nerve,
ultrasonography

Corresponding author:
Sunil Munakomi

College of Medical Sciences,
Bharatpur, Nepal

sunilmunakomi@gmail.com

Scan to access the online version

Introduction

Detection of increasing intracranial pressure (ICP) has been a common non-invasive approach for evaluating optic nerve sheath diameter (ONSD). A frequent emergency is increased intracranial pressure. Poor clinical outcomes, such as a high mortality rate from various neurological illnesses, are associated with an elevated ICP [1].

This portion of the optic nerve is contained by cerebrospinal fluid as well as the optic nerve sheath (ONS), which is a membrane that is attached to the brain's dura mater (the outer membrane). To put it another way, the optic nerve sheath diameter varies within minutes when cerebrospinal fluid pressure changes in the perioptic subarachnoid space, which is an extension of the intracranial subarachnoid space [2-5].

Several factors, including age, gender, and severity of the injury, co-morbidities and concurrent anticoagulation, secondary insults and the initial Glasgow Coma Scale (GCS) score, the motor score and pupil reactivity, the type of lesion visualized on brain computed tomography (CT) scan, changes in intracranial pressure (ICP), and blood levels of specific proteins, influence the prognosis of a brain injury. According to research conducted using ultrasonography, there is an excellent link between intracranial pressure and the diameter of the optic nerve sheath [6,7].

The ICU routinely performs at least one first brain CT scan on patients with serious head trauma. The first ONSD measurement has not been studied in conjunction with the brain CT scan and GCS to our knowledge. Current research examined the relationship between first brain CT scan ONSD and GCS and the outcome of brain injury patients admitted to the ICU. When it comes to identifying high ICP and comparing it to CT findings of higher ICP and GCS alterations, this research was developed. If bedside ultrasonography guided measurement of ONSD could predict higher ICP in patients with any form of brain illness, then this research was successful.

Materials and methods:

In the Neurosurgery ICU of a tertiary care teaching hospital, this retrospective study was carried out between January 2019 and October 2020 after it was approved by the hospital's ethical committee. Those who had provided written informed consent and were suspected of having an elevated ICP were allowed to participate in this research. Patients came in all shapes and sizes, with a wide range of medical conditions. GCS and early CT scan data were used to estimate the extent of brain injury. Both eyes were examined using ONSD before a CT scan was performed on the head of all participants. All patients over the age of 18 who were admitted to a neurosurgical ICU with a suspected increase in ICP were included in the study. There was a strict exclusion policy for patients with serious head traumas, substantial eye injury, or a history of glaucoma or optic nerve illness. A linear ultrasonic probe with a 7.5-10 MHz bandwidth was used to assess ONSD. While laying down, all patients were screened. Over an upper eyelid that was completely closed, an ultrasound gel was placed without exerting any pressure. There was an anomalous mean binocular ONSD more than 5mm and a transverse ONSD of 3mm behind the retina. As long as the patient's midline shift was more than 3 millimeters and the CT scan result indicated an elevated ICP, the CT scan was considered to be positive. (GCS 13-15), (GCS 9-12), or (GCS severe) GCS scores were classed as mild, moderate, or severe (GCS 8 or less).

Age, gender, clinical diagnosis, GCS, and death were all recorded. In order to determine if there was any correlation between various parameters and ONSD levels, the collected data was analyzed. A binocular change in optic nerve sheath diameter of more than 2.00 mm was deemed abnormal in two consecutive readings in the same patient. On the basis of sensitivity and specificity, the ROC curve was utilized to determine the optimal ONSD cutoff point. Shorthand for data analysis is "bringing facts and figures together to address the research question." Analyzes a study on "optic nerve sheath diameter measures in neurosurgical intensive care unit (ICU) for ICP with connection to GCS and CT scan progression" Based on demographic and statement questions, a well-known instrument was created. This survey had a total of 54 answers. SPSS version 25 was used to record, tabulate, and analyze the data statistically. The Chi-square test was done, and a P value of less than 0.05 was thought to be significant. A ROC curve was made to find the ONSD cutoff point with respect to CT progression relation with drop in GCS that gives the best balance of sensitivity and specificity for this modality.

Results

Descriptive analysis is used to characterize the fundamental properties of the data in the research. The quantitative method is a field in which the descriptive data analysis takes its significance. It aims at summarizing a sample accessible to the researcher. It gives concise summaries of the Sample and also about the observation conducted on them. The descriptive analysis analyses the data to generate descriptions of the population, either via numerical computations or tables. They constitute the fundamental basis of any quantitative study of data. The summary data of the researcher is presented in the following tables that are exhibited and detailed below Table 1.

Table 1. Descriptive analysis of gender and age

Variables	*N (sample)*	Frequency	Percentage %
Gender	54		
Male		37	68.5%
Female		17	31.5%
AGE	54		
18-40		9	16.7%
40-60		22	40.7%
>60		23	42.6%

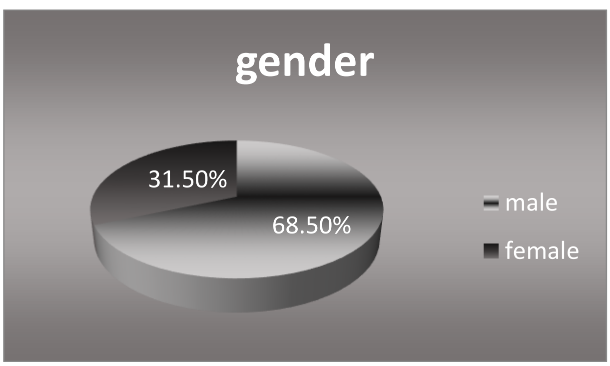


Figure 1. Pie chart showing distribution of participants according to gender.

From the Figure 1, we conclude the gender that about 68.5% of the respondents were males and 31.5% of the respondents were females participate in the survey.

From the Figure 2, we conclude the age of respondents almost 16.7% of the respondents were of age group between 18 and 40 years, 40.7% of the respondents were of age group between 40 and 60 years and 42.6% of the respondents age was more than 60 years.

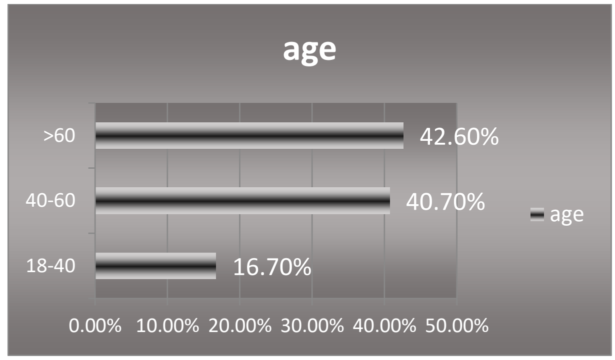


Figure 2. Bar Graph showing distribution of participants according to age groups

Inferential analysis:

Inferential statistics uses a sample of data from a population to draw inferences and predictions about that group. The estimate of hypotheses, parameters, or the testing of hypotheses are included in this section. Assessing the strength of a link between variables is made easier with its assistance the researcher conducts a series of tests to determine the importance of their findings.

Hypothesis:

H1: there's relation between change in ONSD with fall of GCS and CT scan progression

Table 2. Relationship between CT progression and drop in GCS

Crosstab relation between CT progression with Drop in GCS

			CT progression		Total
			No	Yes	
Drop GCS	No	Count	35	3	38
		% within CT progression	89.7%	20.0%	70.4%
	Yes	Count	4	12	16
		% within CT progression	10.3%	80.0%	29.6%
Total		Count	39	15	54
		% within CT progression	100.0%	100.0%	100.0%

From Table 2 we conclude that the sensitivity of drop in GCS to detect raised ICP was found to be approximately 80% and specificity was 89.7% when compared with progression in CT scan. The chi-square tests from table 3 shows highly significant (P-value <0.05) which indicates drop in GCS has significant relationship with the progression in CT scan.

Table 3. Chi-square tests with the relation of drop in GCS and progression in CT scan

Chi-Square Tests

	Value	df	Asymptotic Significance (2-sided)	Exact Sig. (2-sided)	Exact Sig. (1-sided)
Pearson Chi-Square	25.273[a]	1	.000		

Table 4. Relationship between change in ONSD and progression in CT scan

Crosstab relation between CT progression with change in ONSD

			CT progression		
			No	Yes	Total
Change in ONSD	No	Count	35	2	37
		% within CT progression	89.7%	13.3%	68.5%
	Yes	Count	4	13	17
		% within CT progression	10.3%	86.7%	31.5%
Total		Count	39	15	54
		% within CT progression	100.0%	100.0%	100.0%

Table 5. Chi-square tests with the relation of change in ONSD and progression in CT scan

Chi-Square Tests

	Value	df	Asymptotic Significance (2-sided)	Exact Sig. (2-sided)	Exact Sig. (1-sided)
Pearson Chi-Square	29.323[a]	1	.000		

From Table 4 we conclude that the sensitivity of significant change in ONSD to detect raised ICP was found to be approximately 86.7% and specificity was 89.7% when compared with progression in CT scan. The chi-square tests from table 5 shows highly significant (P-value <0.05) which indicates change in ONSD has significant relationship with the progression in CT scan.

Table 6. Area under the curve for change in ONSD with relation of drop in GCS and progression in CT scan.

Area Under the Curve

Test Result Variable(s)	Area
Drop in GCS	.849
Change in ONSD	.882

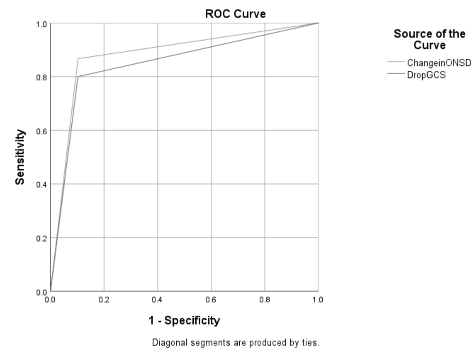


Figure 3. Receiver Operating Characteristic curve (ROC) analysis of change in ONSD with relation of drop in GCS and progression in CT scan.

The ROC for the change in ONSD had a high ability to discriminate between normal and high ICP, where the area under curve (AUC) value was 0.882 [Table 6] whereas drop in GCS while increasing ICP with respect to CT progression has AUC value was 0.849 [Table 6} and shows a good separation between true positives and false positives. Change in ONSD more than 2mm in consecutive times and drop in GCS in a same patient relation with CT progression has a good negative as well as positive predictive value [Figure 3].

Discussion

Even though there are other reliable ways to measure ICP, none of them are as good as ultrasonographic ONSD in a number of ways. For example, places that focus on neurocritical care are the only ones that offer invasive ICP monitoring [8].

It's possible that many institutions lack the specialized, high-priced equipment needed for this procedure, and the If a patient has coagulopathy, for example, invasive ICP monitoring may be impossible. It is possible to identify elevated ICP with CT and MRI scans without the need for intrusive procedures. The procedure may be challenging and time-consuming for patients in the critical care unit who need mechanical breathing or monitoring devices. There is a need to investigate noninvasive bedside techniques to ICP evaluation in clinical settings. An increase in intracranial pressure (ICP) may be accurately measured using noninvasive imaging techniques [9-11].This means that ultrasonographic ONSD evaluation is in high demand and may become the primary method of determining increased ICP [12].

An increase in intraocular pressure has been proven to prevent optic disc enlargement even in the presence of a high intraocular pressure (ICP), according to Brodsky et al. [13]. An A scan has been used by many groups to investigate the connection between ONSD and ICP, and different echography techniques have been used to do so. There is a positive linear association between these two variables in neurosurgical patients, as shown by Cennamo et al.[14], Gangemi et al.[15], and Tamburelli [16].

In our study, we took the reference of 2mm change in ONSD in consecutive measurement of the same patients. To verify this, Helmke and Hansen[17] used cadaver testing and found that the ONSD climbed by up to 60 percent when the globe was three millimeters away from the body, whereas it only rose by 35 percent when the globe was 10 millimeters away according to Liu and Kahn [18].

More than 2mm of ONSD change was detected in the control group over the age of 18 in our research (n=54). Neuropathology patients in the Neurosurgery ICU may be consistently predicted to have an elevated ICP using ultrasound-based ONSD estimate (about 86.7% sensitivity, and 89.7% specificity). Study results by Kimberly et al. found a strong agreement between the values of the ICP catheter and the results of the ONSD ultrasonographic examination, with a sensitivity of 88% and a specificity of 93% [19]. In their investigation, Tayal et al. found that ONSD had 100% sensitivity and 63% specificity for identifying increased ICP [20].

Using adult emergency room patients who had suffered head injuries, Tayal et al. performed a prospective, blinded observational research. They compared the ONSD results from USG with the elevated ICP seen on CT. In order to identify elevated ICP, the sensitivity was 100%, while the specificity was 63% [21]. An ultrasonographic assessment of the ONS diameter of detection for elevated ICP showed that ONSD>5.00mm had a higher ICP >20 mm, with pooled sensitivity of 90% (95 percent confidence interval: 80-95 percent) and specificity of 85%, as shown by Dubourg et al (95 percent CI: 73-93 percent). A positive ONSD is 51 times more common in individuals with elevated ICP [22].

We examined 54 individuals and found no correlation between ONSD results with the age or gender of the group. We also found a correlation between ONSD results and hemodynamic parameters, CT progression, and the patient's GCS at the time of evaluation, which may be attributed to the pathophysiological impact of these factors on intracranial pressure and ONSD. We utilized CT progression as a reference standard for comparing ONSD outcomes since CT scans of the head are used on a regular basis in neurosurgical ICUs to identify elevated ICP. CT scan has also been used as a reference diagnostic test in other research [23,24]. Neurosurgical competence, time for insertion, and lack of experience in all institutions make invasive monitoring of ICP the gold standard [25].

In our study, we also used a reference standard of change in ONSD 2mm while measuring consecutive time in the same patients was considered to be abnormal. The ROC for the change in ONSD readings had an AUC value of 0.882 with respect to CT progression and The ROC for drop in GCS relation with CT progression shows 0.849 and both test showed a good separation between true positives and false positives

These findings show that ultrasound measurements of ONSD may be an effective substitute for invasive ICP measurements or other imaging modalities. It is possible to estimate the ICP value of patients whose ICP measurement through lumbar puncture is problematic (in high risk, such as a hemodynamically unstable patient using this noninvasive technique [26]. A research found a substantial correlation between invasive ICP and both ONSD ultrasound and straight sinus systolic flow velocity. According to Robba et al. [27], ultrasonographic assessment of ONSD is noninvasive, safe, and rapid due to the easy visibility of the orbital window and the absence of problems. CT scans of the brain need time and money, and in certain emergency cases, there is a shortage of time for this procedure. An ultrasound-guided ONSD assessment may help identify increased ICP in the early stages of traumatic brain injury (TBI) and avoid further brain damage. There was no added advantage to therapy based on intracranial catheter measurement of ICP compared to clinical and imaging results alone, according to a major trial [28].

We do not use intrusive monitoring at our institution, and if we suspect that a patient's ICP is elevated, we do repeated CT scans. ONSD may be quite useful in detecting elevated ICP in these types of health care institutions so that efforts to lower ICP can be taken as soon as possible. There are a number of ways in which ONSD may be used to evaluate ICP, determine the next course of therapy, and send patients to higher facilities, and in situations when a CT scan is not accessible or if a tertiary care facility is a considerable distance away.

Conclusion

For identifying elevated ICP, ONSD measurement using ultrasound has a high degree of sensitivity, specificity, and connection with CT brain advancement and a decrease in GCS. Bedside measurement of ONSD is a good way to find out if ICP is too high because it is non-invasive, can be done more than once, doesn't use ionizing radiation, and can be used in many different situations. More research is needed to prove that it helps people with raised ICP. It also helps start treatment for a high ICP as soon as possible. It is quick, can be done at the bedside, doesn't hurt the patient, doesn't cost much, and doesn't use any ionizing radiation.

References

1. Juul N, Morris GF, Marshall SB, et al. Intracranial hypertension and cerebral perfusion pressure: influence on neurological deterioration and outcome in severe head injury. The Executive Committee of the International Selfotel Trial. J Neurosurg 2000;92:1–6.
2. Liu, D.; Kahn, M. Measurement and Relationship of Subarachnoid Pressure of the Optic Nerve to Intracranial Pressures in Fresh Cadavers. Am. J. Ophthalmol. 1993, 116, 548–556.
3. Helmke, K.; Hansen, H.C. Fundamentals of transorbital sonographic evaluation of optic nerve sheath expansion under intracranial hypertension. Pediatr. Radiol. 1996, 26, 701–705.
4. Maude, R.R.; Hossain, M.A.; Hassan, M.U.; Osbourne, S.; Langan Abu Sayeed, K.; Rezaul Karim, M.; Samad, R.; Borooah, S.; Dhillon, B.; Day, N.P.J.; et al. Transorbital sonographic evaluation of normal optic nerve sheath diameter in healthy volunteers in Bangladesh. PLoS ONE 2013, 8, e81013.
5. Hansen, H.-C.; Helmke, K. Validation of the optic nerve sheath response to changing cerebrospinal fluid pressure: Ultrasound findings during intrathecal infusion tests. J. Neurosurg. 1997, 87, 34–40.
6. Geeraerts T, Duranteau J, Benhamou D: Ocular sonography in patients with raised intracranial pressure: the papilloedema revisited. Crit Care 2008, 12:150.
7. Geeraerts T, Launey Y, Martin L, Pottecher J, Vigué B, Duranteau J, Benhamou D: Ultrasonography of the optic nerve sheath may be useful for detecting raised intracranial pressure after severe brain injury. Intensive Care Med 2007, 33:1704-17118. Citerio G, Bakker J, Bassetti M, et al. Year in review in Intensive Care Medicine 2014: I. Cardiac dysfunction and cardiac arrest, ultrasound, neurocritical care, ICU-acquired weakness, nutrition, acute kidney injury, and miscellaneous. Intensive Care Med 2015;41:179–191.
8. Geeraerts T, Launey Y, Martin L, et al. Ultrasonography of the optic nerve sheath may be useful for detecting raised intracranial pressure after severe brain injury. Intensive Care Med 2007;33:1704–1711.
9. Kimberly HH, Noble VE. Using MRI of the optic nerve sheath to detect elevated intracranial pressure. Crit Care 2008;12:181.
10. Das SK, Shetty SP, Sen KK. A novel triage tool: optic nerve sheath diameter in traumatic brain injury and its correlation to rotterdam Computed Tomography (CT) scoring. Pol J Radiol 2017;82:240–243.
11. Kristiansson H, Nissborg E, Bartek J Jr, et al. Measuring elevated intracranial pressure through noninvasive methods: a review of the literature. J Neurosurg Anesthesiol 2013;25:372–385.
12. Brodsky MC, Chen JJ, Wetjen NM. Optical coherence tomography for the noninvasive detection of elevated intracranial pressure a new role for the ophthalmologist? JAMA Ophthalmol 2017;135(4):329–330.
13. Cennamo G, Gangemi M, Stella L. The correlation between endocranial pressure and optic nerve diameter: An ultrasonographic study. Ophthal Echography 1987;7:603 6.
14. Gangemi M, Cennamo G, Maiuri F, D'Andrea F. Echographic measurement of the optic nerve in patients with intracranial hypertension. Neurochirurgia (Stuttg) 1987;30:53 5.
15. Tamburelli C, Aricle C, Mangiola A. CSF dynamic parameters and changes of optic nerve diameters measured by standardised echography. Ophthal Echography 1993;13:101 9.
16. Hansen HC, Helmke K. The subarachnoid space surrounding the optic nerves. An ultrasound study of the optic nerve sheath. Surg Radiol Anat 1996;18:323 8.
17. Liu D, Kahn M. Measurement and relationship of subarachnoid pressure of the optic nerve to intracranial pressures in fresh cadavers. Am J Ophthalmol 1993;116:548-56.
18. Kimberly HH, Shah S, Marill K, Noble V. Correlation of optic nerve sheath diameter with direct measurement of intracranial pressure. Acad Emerg Med 2008;15:201 4.
19. Tayal VS, Neulander M, Norton HJ, Foster T, Saunders T, Blaivas M. Emergency department sonographic measurement of optic nerve sheath diameter to detect findings of increased intracranial pressure in adult head injury patients. Ann Emerg Med 2007;49:508 14.
20. Tayal VS, Neulander M, Norton HJ, Foster T, Saunders T, Blaivas M. Emergency department sonographic measurement of optic nerve sheath diameter to detect findings of increased intracranial pressure in adult head injury patients. Ann Emerg Med 2007;49:508 14.

21. Dubourg J, Javouhey E, Geeraerts T, Messerer M, Kassai B. Ultrasonography of optic nerve sheath diameter for detection of raised intracranial pressure: A systematic review and meta analysis. Intensive Care Med 2011;37:1059 68.
22. Ohle R, McIsaac SM, Woo MY, Perry JJ. Sonography of the optic nerve sheath diameter for detection of raised intracranial pressure compared to computed tomography: A systematic review and meta analysis. J Ultrasound Med 2015;34:1285 94.
23. Tayal VS, Neulander M, Norton HJ, Foster T, Saunders T, Blaivas M. Emergency department sonographic measurement of optic nerve sheath diameter to detect findings of increased intracranial pressure in adult head injury patients. Ann Emerg Med 2007;49:508 14.
24. Liu D, Li Z, Zhang X, Zhao L, Jia J, Sun F, et al. Assessment of intracranial pressure with ultrasonographic retrobulbar optic nerve sheath diameter measurement. BMC Neurol 2017;17:188.
25. Liu D, Li Z, Zhang X, Zhao L, Jia J, Sun F, et al. Assessment of intracranial pressure with ultrasonographic retrobulbar optic nerve sheath diameter measurement. BMC Neurol 2017;17:188.
26. Robba C, Cardim D, Tajsic T, Pietersen J, Bulman M, Donnelly J, et al. Ultrasound non invasive measurement of intracranial pressure in neurointensive care: A prospective observational study. PLoS Med 2017;14:e1002356.
27. Soliman I, Johnson GG, Gillman LM, Zeiler FA, Faqihi F, Aletreby WT, et al. New optic nerve sonography quality criteria in the diagnostic evaluation of traumatic brain injury. Crit Care Res Pract 2018;2018:3589762. doi: 10.1155/2018/3589762

Romanian Neurosurgery (2023) XXXVII (3): pp. 263-271
DOI: 10.33962/roneuro-2023-048
www.journals.lapub.co.uk/index.php/roneurosurgery

An anencephalic heterophagus or parasitic twin. A case report with literature review

Ahtesham Khizar[1], Soha Zahid[2], Filza Shahid[2], Madeha Amjad[1], Pradhumna Kumar Yadav[3]

[1] Institute of Medical Sciences, Islamabad, PAKISTAN
[2] Jinnah Medical and Dental College, Karachi, PAKISTAN
[3] Janaki Health Care and Research Centre, Janakpur, NEPAL

ABSTRACT
A case of 6-week-old female twins is presented. Twin 1 was fully formed, whereas twin 2 was parasitic, adhered to the left flank, and had anencephaly and complete upper limbs. Depending on the twins' overall condition, the recommendation is to separate conjoined twins as soon as possible for a better outcome.

Keywords
anencephaly,
heteropagus,
parasitic twins

Corresponding author:
Ahtesham Khizar

Punjab Institute of Neurosciences,
Lahore, Pakistan

arwain.6n2@gmail.com

Scan to access the online version

INTRODUCTION

With an estimated prevalence of fewer than 0.1 per 100,000 births, heteropagus or parasitic twinning is a very unusual disorder. Ambroise Pare, a 16th-century French surgeon, described an acephalus body linked to the abdomen of one of his patients, which is perhaps the earliest plausible mention of the ailment.[1] The symmetry, site of fusion, and grade of duplication of conjoined twins are characterised, with thoracopagus being the most common kind (40%), followed by thoraco-omphalopagus (28%). Asymmetrically conjoined twins also known as heteropagus or parasitic twins are difficult to categorise.[2]

According to Spencer[3], parasitic twins are conjoined twins that are linked in one of the same anatomical sites as "complete" conjoined twins. One of the twins has a significant disability and is referred to as a "parasite." While certain congenital anomalies, such as heart malformations, may be present, its corresponding "autosite" must be largely intact. To describe parasitic twins, the terms "heteropagus," "asymmetric conjoined," "partial or incomplete conjoined," "parasitic," and "exo-parasitic" are all used interchangeably.[1]

The largest documented series on the incidence of heteropagus twinning comes from a database of 7.9 million births collected over an eight-year period in the 1970s in the United States. The true incidence of heteropagus twins was estimated to be 0.05 to 0.1 per 100,000 in this US study.[5] An incidence of 0.02 per 100,000 was discovered in a more recent European research of approximately 5 million newborns.[4] The frequency of heteropagus pairs as a fraction of all conjoined twins has been estimated to be between 4.5 and 15%.[5] In symmetric conjoined twins (72%), there is a significant female preponderance; however, in heteropagus twins, the gender difference is less prominent. In a 2001 study of 157 heteropagus twins, 51% were female.[3]

CASE PRESENTATION

Female twins, 6 weeks old, were presented to us at the emergency department. The twins were conjoined and parasitic. There was no significant history of any antenatal checkups during the pregnancy. No record of any antenatal ultrasound was available. Twins were born at full term and without any medical assistance at home. According to the family, the twin 1 (complete twin) cried after a few minutes of birth. The birth weight of the twins was unknown. On presentation to the emergency, the twins were ordered to have baseline investigations, X-rays and then an Magnetic Resonance Imaging (MRI) lumbar spine without contrast. On examination, the complete twin had a well-formed head, torso, upper limbs, and lower limbs. A parasitic twin was attached to the left flank and presented with an anencephalic head and normal upper limbs as shown in Figure.1 (A,B&C). On MRI, a scoliotic deformity of the thoracolumbar spine was noted in twin 1. There was a convexity towards the left side of the spinal cord terminating at L3/L4 level.

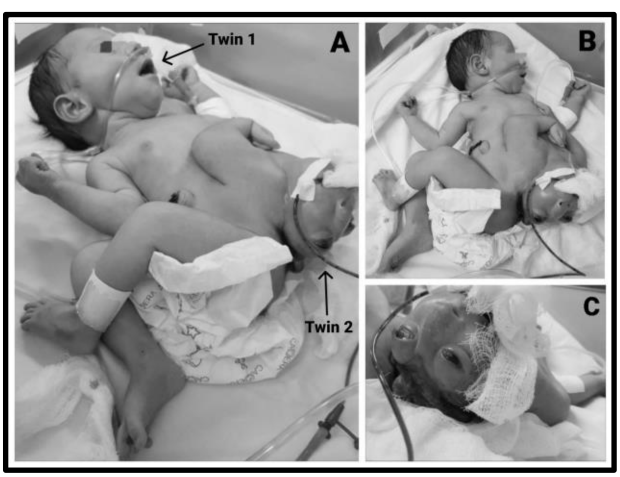


Figure 3. Twin 1: Complete twin (Autosite), Twin 2: Partial twin (Parasite).

There was a partial widening of the spinal canal along with partial defective posterior elements. Twin 2, the partial twin, had non visualisation of brain matter as well as the spinal canal. After sufficient radiological reports and clinical evaluation, elective surgery was scheduled to separate the conjoined twins. Before the surgery could take place, there were episodes of repeated apnea attacks. Even though sufficient resuscitative measures were taken, unfortunately, the twins died before the planned surgery.

DISCUSSION

The spontaneous occurrence of twinning, both monozygotic and dizygotic, is being highlighted. Conjoined twinning, on the other hand, happens only when the twinning event occurs during the primitive streak stage of development, which occurs around 13-14 days after conception in humans, and is only associated with the monoamniotic monochorionic form of placentation. Humans are said to be the species with the highest rate of conjoined twinning. While monozygotic twinning can be created in animals in the lab using a number of chemicals, the mechanism of spontaneous twinning in humans is unknown. All agents that can cause twinning are teratogenic, and they most likely do so by interfering with the spindle apparatus.[2]

According to the findings of the greatest study to date, the spontaneous birth rate is around 10.25 per million. Thoraco-omphalopagus (28%), thoracopagus (18.5%), omphalopagus (10%), parasitic twins (10%), and craniopagus (6%) were the most common types seen. Approximately 40% of these babies were stillborn, while 60% were born alive, albeit only about 25% of those who made it to birth lived long enough to be surgical candidates. Conjoined twinning is caused by the incomplete division of the embryonic axis, and all conjoined twins are symmetrical, with the exception of parasitic conjoined twins, and "the same parts are always attached to the same parts."[1] Table.1 shows a comprehensive evaluation of the literature on heteropagus twins from 2001 to 2022.

Conjoined twins are characterised based on their symmetry, fusion site, and duplication grade. In contrast to conjoined twins, who have the same locations fused, parasitic twins are a subset of conjoined twins who join "asymmetrically," with their own varieties and classification.[1] The anencephalic parasitic twin was connected to the left flank of the autosite in our reported case, an unusual presentation that has never been defined or classified before.

Table 1. Literature review on heteropagus twins from 2001 to 2022

Authors	Year	Type of heteropagus	Gender	Autosite abnormalities	Parasite features
EK Cury[13]	2001	1.Epigastric heteropagus 2.Epigastric heteropagus 3.Epigastric heteropagus	1.M 2.M 3.F	1.ASD 2.Omphalocele 3.Absent 1/3rd sternum	BUE, BLE, bowel, bladder, kidney, ureter, pelvis, external genitalia & patent urethra
DK Gupta[14]	2001	1.Epigastric heteropagus 2.Epigastric heteropagus	1.M 2.M	1.Omphalocele 2.VSD, PDA	UUE
P Raynal[15]	2001	Epigastric heteropagus	M	Omphalocele	Size: 12×10 cm, BLE, bowel, pelvis & imperforate anus
JK Mahajan[16]	2002	Ischiopagus	F	Omphalocele, single ovary, hemiuterus	BLE, bowel, bladder, ovary×1, hemiuterus, pelvis, external genitalia & patent urethra
DA De Ugarte[17]	2002	1.Epigastric heteropagus 2.Epigastric heteropagus	M	Inferior sternal cleft, VSD, PFO, TGA, PDA, COA, omphalocele, meckel diverticulum	Rudimentary BUE, complete BLE, palpable spine, liver, gallbladder, kidney, ureter, bladder, pelvis, testes×2 (undescended) & external genitalia
Martinez[1]	2003	1.Epigastric heteropagus 2.Acardiac acephalus	-	-	-
A Tongsin[18]	2003	Epigastric heteropagus (4)	-	Congenital heart defect, omphalocele	-
JR Corona-Rivera[19]	2003	Ischiopagus	M	Small left diaphragmatic defect, omphalocele, exstrophy of the cloaca, and lumbar meningomyelocele	Complete left LE, hemipelvis, lumbosacral vertebral column, spinal cord, and one kidney with ureter & adrenal gland
AP MacKenzie[20]	2004	Epigastric heteropagus	F	Bilateral choroid plexus cysts, dextrocardia, omphalocele	BLE & pelvis
AT George[21]	2004	Epigastric heteropagus	M	Talipes equinovarus	BLE, accessory abdominal compartment, bowel, liver, gallbladder, cystic duct, pelvis, testes×2, phallus & empty scrotum
SK Ratan[22]	2004	Rachipagus	F	Spinal dysraphism, tethered cord	Size: 8×8 cm, ULE (fused feet), vertebrae×2, bowel, pelvis, bladder & fallopian tissue
AK Bangroo[23]	2004	Epigastric heteropagus	M	-	UUE, BLE & phallus

K Fujimori[24]	2004	Omphalopagus	F	Single ventricle, hypoplastic aorta	BUE (limb buds), alobar holoprosencephaly, cleft palate/lip & hypoplastic lung
RC Ribeiro[25]	2005	Xipho Omphalopagus	M	Unilateral renal agenesis	Hydrocephalus, hypertelorism, choanal atresia, micrognathia, trachea, rudimentary heart, bowel & bladder
M Bhansali[26]	2005	1.Epigastric heteropagus 2.Epigastric heteropagus 3.Epigastric heteropagus	1.F 2.M 3.F	1.Omphalocele 2.Omphalocele 3.Unknown	1.BUE, BLE, atretic rectum, absent anus, kidney×2, pelvis 2.BLE (limb buds) 3.BLE, pelvis, external genitalia
H Rode[27]	2006	Ischiopagus (4) Not explained (5)	F	-	-
R Chadha[28]	2006	Rachipagus	F	Spinal dysraphism	Size: 11×8 cm, anal dimple, external genitalia, empty scrotum & digits×3
Shibata[1]	2006	Craniopagus	F	None	Multiple cranial malformation, hair, scalp, rudimentary trachea, lungs, esophagus, stomach & bowel
Y Kanamori[29]	2006	Epigastric heteropagus	F	Abducted/flexed LE	BUE, BLE (fused), lung, stomach, bowel, liver, pancreas, kidney, ureter, bladder, pelvis, external genitalia & ovaries×2
J Hager[30]	2007	Epigastric heteropagus	F	VSD	BUE, BLE, bowel, imperforate anus, multicystic kidney, pelvis, external genitalia & blind urethra
CM Snelling[31]	2008	Rachipagus	M	High arched palate, retrognathia, posteriorly set ears, simian crease, spinal dysraphism, meningocele, hydromyelia, split, tethered cord	Size: 4×4×2 cm, amorphous with skin, bone, digit, clavicle & scapula
G Albert[32]	2008	Rachipagus	F	Spinal dysraphism, myelocystocele, teratoma	Size: 8×9.5× 2.5 cm, amorphous
E Satter[33]	2008	Omphalopagus	F	-	Size: 15×9 cm, amorphous with skin, hair, bone, rudimentary URT, bowel & liver

XL Hu[34]	2009	Epigastric heteropagus	M	-	Limbs, abdomen, buttocks & male external genitalia
S Sanoussi[35]	2010	1.Rachipagus 2.Rachipagus	1.M 2.M	1.Meningocele, spinal lipoma 2. Spina bifida	1.Complete LE & rudimentary genitalia 2.Dysmorphic UE fused at the midline
AM Abubakar[36]	2011	Epigastric heteropagus	M	Ventral hernia of a healed exomphalos	BLE, rudimentary UUE, pelvis, external genitalia & imperforate anus
S Das[37]	2011	Epigastric heteropagus	M	Omphalocele	Rudimentary limbs, auricles, and hairs on the scalp
J Zhang[38]	2011	Rachipagus	F	Spina bifida, diplomyelia, scoliosis, tethered cord, & cardiac VSD	Soft-tissue mass including fully developed breast glandular tissue with an external nipple-areola complex, uterine tube & duct, bone & cartilage
M Qasim[39]	2011	Epigastric heteropagus	M	-	Size: 6x4 cm, rudimentary UE, LE, head & external genitalia
H Ozkan-Ulu[40]	2011	Epigastric heteropagus	M	Cardiac malformation	UUE & rudimentary external genitalia
RV Reddy[41]	2011	Pyopagus	-	-	Rudimentary bony hemipelvis, BLE with gross deformity of right UE, intergluteal fold without anal orifice, partial gastrointestinal system & rudimentary bladder
A Solak[42]	2012	Rachipagus	F	Spina bifida	Dysmorphic & immobile UE with 4 small buds at the base
JT Xie[43]	2012	1.Epigastric heteropagus 2.Epigastric heteropagus	1.M 2.M	1.None 2.Omphalocele	1.BUE, BLE, pelvis, scrotum, and penis 2.Immobile LE, buttocks, perineum & male genitalia
K Kesan[44]	2013	Epigastric heteropagus	M	Omphalocele	-
G Komla[45]	2013	1.Epigastric heteropagus 2.Epigastric heteropagus 3.Omphalopagus	1.F 2.M 3. -	1.Omphalocele 2.Omphalocele and fibrous mitral valves 3.Systolic murmur	1.BUE & BLE 2.BUE & ULE 3.BUE & ULE
DR Calderoni[46]	2014	Thoraco-omphalopagus	M	Omphalocele	BUE, BLE, external genitalia, scrotum & anus
Y Bayri[47]	2014	Rachipagus	M	Lumbosacral meningocele, unfused	Accessory LE & rudimentary hemipelvis

				vertebral arches between L1 & S1	
EC Gokcen[48]	2015	Ischiopagus	F	-	BLE, rudimentary LE
D Baskaran[49]	2015	Epigastric heteropagus	M	-	Gastroschisis, BUE, BLE, pelvis
AA Navaei[50]	2015	1.Rachipagus 2.Rachipagus	1.F 2.M	1.None 2.Neurogenic bladder	1.Fully formed LE & hemipelvis 2.Soft-tissue mass with rudimentary scrotum & penis as well as bony structures resembling a scapula & rib
A Sahlu[51]	2016	Rachipagus		Lipomyelomeningocele and tethered cord	Foot, ankle, lower leg & a small sinus that resembled an anus
K Ahmed[52]	2016	Thoracic heteropagus	M	Giant omphalocele	Hypoplastic BUE, thorax, misshapen trunk, small abdomen, pelvis, BLE, male genitalia, empty scrotum & imperforate anus
KN Rattan[53]	2016	Rachipagus	F	-	Ulcerated, multicystic mass with rudimentary limb-like structures including bone & cartilage
P Raj[54]	2017	Epigastric heteropagus	M	-	BLE, rudimentary UE, pelvis, scrotum, penis & imperforate anus
A Raheja[55]	2017	Rachipagus	-	-	-
A Khushdil[56]	2018	Epigastric heteropagus	M	Omphalocele	LE, UE, pelvis & male genitalia
M Malik[57]	2018	Epigastric heteropagus	M	Omphalocele	BLE, rudimentary UE, external genitalia, scrotum & absent testes
N Khavanin[58]	2018	Rachipagus	F	-	BLE, cervicothoracic diplomyelia, pelvis & rudimentary scapula
AR Muhelo[59]	2018	Ischiopagus	F	-	BLE
MS Falyoun[60]	2020	Epigastric heteropagus	M	Omphalocele	BLE, external genitalia, scrotum, absent testes & anus
AB Pati[61]	2020	Rachipagus (2)	-	-	-
RCS Alves[62]	2020	Asymmetrical	F	Anencephaly	BUE, BLE, rudimentary external genitalia & pelvis

MH Takrouney[12]	2020	1.Asymmetrical 2.Epigastric heteropagus 3.Epigastric heteropagus	1.F 2.M 3.M	1.Unexplained 2.Omphalocele 3.Omphalocele	1.Incomplete head & soft tissue swelling 2.BLE, rudimentary UE & external genitalia 3.BLE, rudimentary gut

"Fission" and "Fusion" are the two basic ideas for the embryologic origin of conjoined twins. Incomplete fission of the blastocyst inner cell mass at the primitive streak stage, 13 to 15 days postfertilization, results in two foci of axial growth that preserve a connection at some point, according to proponents of the former theory.[6] "Fusion," on the other hand, refers to the coalescence of two initially separate inner cell masses at a later stage.[3] Donitz et al.[7] proposed the most widely accepted theory for the formation of asymmetry between autosite and parasite after these occurrences. They hypothesised that vascular impairment causes the parasitic twin's tissue to become reliant on autosite collaterals. The starved section of the parasite's body experiences selective ischemic atrophy.[1]

Asymmetric conjoined twins, like their symmetric counterparts, were traditionally thought to be universally monozygotic. Recent studies that used deoxyribonuleic acid (DNA) polymerase chain reaction to analyse renal tissue and blood cells from the autosite and parasite support this conclusion.[8,9,10] One group, on the other hand, has discovered a parasite with scrotal skin that is related with a female autosite.[11]

Depending on the location of the attachment and the shared inner parts, surgery to separate conjoined twins might be simple or complex. Rectal manometry can only be employed if the anal canal is impacted or implicated during separation, and the surgery often leads in the death of one or both of the conjoined twins, especially if they are attached at the head or share a vital organ.[12] For a better outcome, we urge that conjoined twins be separated as soon as feasible, depending on their overall condition. Recently, endoscopic minimally invasive techniques like laparoscopy and robotic surgery have been used in the preoperative and operational management to examine internal organs and determine whether or not connections exist, as well as to treat connections. The mainstay of the separation procedure for external connections is open surgery.[12]

Conclusion

An anencephalic parasitic twin attached to the autosite's left flank has never been classified or reported before. Separating conjoined twins as soon as possible, depending on their general condition, is recommended for a better outcome. Depending on the location of the attachment and the shared inner parts, surgery to separate conjoined twins might be simple or complex. Endoscopic minimally invasive procedures can now be employed for internal organ assessment and surgery planning, thanks to recent advancements.

References

1. Sharma G, Mobin SS, Lypka M, Urata M. Heteropagus (parasitic) twins: a review. Journal of pediatric surgery. 2010 Dec 1;45(12):2454-63.
2. Kaufman MH. The embryology of conjoined twins. Child's Nervous System. 2004 Aug;20(8):508-25.
3. Spencer R. Parasitic conjoined twins: external, internal (fetuses in fetu and teratomas), and detached (acardiacs). Clin Anat. 2001;14(6):428-44.
4. Martínez-Frías ML, Bermejo E, Mendioroz J, Rodríguez-Pinilla E, Blanco M, Egüés J, et al. Epidemiological and clinical analysis of a consecutive series of conjoined twins in Spain. J Pediatr Surg. 2009;44(4):811-20.
5. Edmonds LD, Layde PM. Conjoined twins in the United States, 1970–1977. Teratology. 1982 Jun;25(3):301-8.
6. Potter EL. Multiple pregnancies and conjoined twins. Pathology of the Fetus and Infant. 1975:207-37.
7. Donitz W. Beschreibung und Erlauterung von Doppelmissgeburten, Dritte Abhandlung, Fünfter Fall. Archiv für Anatomie. Physiologie und Wissenschaftlige Medicin. 1866:534-44.
8. Gupta DK, Lall A, Bajpai M. Epigastric heteropagus twins– a report of four cases. Pediatric surgery international. 2001 Jul;17(5):481-2.
9. Hwang EH, Han SJ, Lee JS, Lee MK. An unusual case of monozygotic epigastric heteropagus twinning. Journal of pediatric surgery. 1996 Oct 1;31(10):1457-60.
10. Fujimori K, Shiroto T, Kuretake S, Gunji H, Sato A. An omphalopagus parasitic twin after intracytoplasmic sperm injection. Fertility and sterility. 2004 Nov 1;82(5):1430-2.
11. Chadha R, Lal P, Singh D, Sharma A, Choudhury SR. Lumbosacral parasitic rachipagus twin. Journal of Pediatric Surgery. 2006 Jan 1;41(1):e45-8.

12. Takrouney MH, Ibrahim IA, Abdel-Ghaffar HS, et al. Conjoined twins: A report of four cases. Int J Surg Case Rep. 2020;73:289-293.
13. Cury EK, Schraibman V. Epigastric heteropagus twinning. Journal of pediatric surgery. 2001 Jul 1;36(7):1-3.
14. Gupta DK, Lall A, Bajpai M. Epigastric heteropagus twins–a report of four cases. Pediatric surgery international. 2001 Jul;17(5):481-2.
15. Raynal P, Petit T, Ravasse P, Herlicoviez M. A rare case of epigastric heteropagus twinning. Journal de Gynecologie, Obstetrique et Biologie de la Reproduction. 2001 Feb 1;30(1):65-9.
16. Mahajan JK, Devendra K, Mainak D, Rao KL. Asymmetric conjoined twins: atypical ischiopagus parasite. Journal of Pediatric Surgery. 2002 Oct 1;37(10):1-3.
17. De Ugarte DA, Boechat MI, Shaw WW, Laks H, Williams H, Atkinson JB. Parasitic omphalopagus complicated by omphalocele and congenital heart disease. Journal of pediatric surgery. 2002 Sep 1;37(9):1357-8.
18. Tongsin A, Niramis R, Rattanasuwan T. Epigastric heteropagus twins: a report of four cases. J Med Assoc Thai. 2003;86 Suppl 3:S605-S609.
19. Corona-Rivera JR, Corona-Rivera E, Franco-Topete R, Acosta-León J, Aguila-Dueñas V, Corona-Rivera A. Atypical parasitic ischiopagus conjoined twins. J Pediatr Surg. 2003;38(2):e3.
20. MacKenzie AP, Stephenson CD, Funai EF, Lee MJ, Timor-Tritsch I. Three-dimensional ultrasound to differentiate epigastric heteropagus conjoined twins from a TRAP sequence. American journal of obstetrics and gynecology. 2004 Nov 1;191(5):1736-9.
21. George AT, Varkey S, Kalam A, Surendran N. Left over bowels: an unique complication in a surviving twin. Postgraduate medical journal. 2004 Dec 1;80(950):736-7.
22. Ratan SK, Rattan KN, Magu S, Rohilla S, Purwar P, Mathur SK. Thoracolumbar rachipagus parasite. Pediatric Surgery International. 2004 Apr;20(4):298-300.
23. Bangroo AK, Khetri R, Tiwari S. Epigastric heteropagus: a parasitic twin. Journal of Indian Association of Pediatric Surgeons. 2004;9(3):148-50.
24. Fujimori K, Shiroto T, Kuretake S, Gunji H, Sato A. An omphalopagus parasitic twin after intracytoplasmic sperm injection. Fertility and sterility. 2004 Nov 1;82(5):1430-2.
25. Ribeiro RC, Maranhão RF, Moron AF, Leite MT, Cordioli E, Hisaba W, Martins JL. Unusual case of epigastric heteropagus twinning. Journal of Pediatric Surgery. 2005 Mar 1;40(3):E39-41.
26. Bhansali M, Sharma DB, Raina VK. Epigastric heteropagus twins: 3 case reports with review of literature. Journal of pediatric surgery. 2005 Jul 1;40(7):1204-8.
27. Rode H, Fieggen AG, Brown RA, Cywes S, Davies MR, Hewitson JP, Hoffman EB, Jee LD, Lawrenson J, Mann MD, Matthews LS. Four decades of conjoined twins at Red Cross Children's Hospital-lessons learned. South African Medical Journal. 2006 Oct 18;96(9):931-40.
28. Chadha R, Lal P, Singh D, Sharma A, Choudhury SR. Lumbosacral parasitic rachipagus twin. Journal of Pediatric Surgery. 2006 Jan 1;41(1):e45-8.
29. Kanamori Y, Tomonaga T, Sugiyama M, Hashizume K, Goishi K, Haga N. Bizarre presentation of epigastric heteropagus: report of a case. Surgery today. 2006 Oct;36(10):914-8.
30. Hager J, Sanal M, Trawöger R, Gassner I, Oswald E, Rudisch A, Schaefer G, Mikuz G, Sergi C. Conjoined epigastric heteropagus twins: excision of a parasitic twin from the anterior abdominal wall of her sibling. European journal of pediatric surgery. 2007 Feb;17(01):66-71.
31. Snelling CM, Ellis PM, Smith RM, Rossiter JP. Lipomatous lumbar mass with an attached digit and associated split cord malformation. Canadian journal of neurological sciences. 2008 May;35(2):250-4.
32. Albert G, Campos M, Menezes A, Vogel T, Weinstein S: Rachipagus parasite associated with myelocystocele and diastematomyelia. Pediatr Neurosurg 44:418–421, 2008
33. Satter E, Tomita S. A case report of an omphalopagus heteropagus (parasitic) twin. Journal of Pediatric Surgery. 2008 Jun 1;43(6):e37-9.
34. Hu XL, Song LY, Zhao LS, Ning PR. Zhonghua Bing Li Xue Za Zhi. 2009;38(7):491-492.
35. Sanoussi S, Rachid S, Sani CM, Mahamane B, Addo G: Rachipagus: a report of two cases - thoracic and lumbar. J Surg Tech Case Rep 2:27–29, 2010
36. Abubakar AM, Ahidjo A, Chinda JY, et al. The epigastric heteropagus conjoined twins. J Pediatr Surg. 2011;46(2):417-420.
37. Das S, Ghosh D, Biswas S, et al. Heteropagus twins-a tale of two cases. Indian J Surg. 2011;73(3):217-220.
38. Zhang J, Duan H, Zhang Y, Yi Z, Bao S: Parasitic rachipagus conjoined twins with spina bifida, diplomyelia, scoliosis, tethered cord syndrome, and ventricular septal defect—case report. Neurol Med Chir (Tokyo) 51:736–739, 2011
39. Qasim M, Shaukat M. Epigastric heteropagus twin. APSP J Case Rep. 2011;2(3):24.
40. Ozkan-Ulu H, Yilmaz Y, Sari FN, Altug N, Uras N, Dilmen U. An unusual case of heteropagus: autosite with a complex cardiac malformation. Pediatr Neonatol. 2011;52(6):358-360.
41. Reddy VR, Chakrabarti SK, Singh MCh, Devi PP, Singh CK, Sharma NG. Asymmetric conjoined twins: pyopagus parasite. J Indian Med Assoc. 2011;109(5):351-352.
42. Solak A, Ergün S, Polat I, Sahin N, Genç B: A rare form of heteropagus twinning: three-armed infant with spinal dysraphism. Case Rep Pediatr 2012:831649, 2012
43. Xie JT, Zhou L, Yang ZL, Sun HY. Epigastric heteropagus conjoined twins: two case studies and associated DNA analysis. Clinics (Sao Paulo). 2012;67(5):527-529.
44. Kesan K, Gupta A, Gupta RK, et al. Gluteal flap for omphalocele repair in a case of epigastric heteropagus: A novel approach for surgical management. Indian J Plast Surg. 2013;46(1):127-129.
45. Komla G, Komla A, Kpatekana S, et al. Experience of managing conjoined and parasitic twins from a developing country. APSP J Case Rep. 2013;4(3):52.

46. Calderoni DR, Mizukami A, Nunes PH, Kharmandayan P. Thoraco-omphalopagus asymmetric conjoined twins: report of a case and complete review of the literature. J Plast Reconstr Aesthet Surg. 2014;67(1):e18-21.
47. Bayri Y, Tanrıkulu B, Ekşi MS, Dağçınar A: Accessory lower limb associated with spina bifida: case report. Childs Nerv Syst 30:2123–2126, 2014
48. Gokcen EC, Wamisho BL. Delayed presentation of a heteropagus (parasitic) twin: a case report of a 17-year-old patient. J Pediatr Orthop B. 2015;24(6):567-572.
49. Baskaran D, Aleem MA, Ravi R. Parasitic twin with gastroschisis is one of the rarest variant of conjoined twins: a case report. Indian J Surg. 2015;77(Suppl 1):90-91.
50. Navaei AA, Habibi Z, Moradi E, Nejat F: Parasitic rachipagus twins; report of two cases. Childs Nerv Syst 31:1001–1003, 2015
51. Sahlu A, Mesfin B, Tirsit A, Debebe T, Wester K. Parasitic twin—a supernumerary limb associated with spinal malformations. A case report. Acta neurochirurgica. 2016 Mar;158(3):611-4.
52. Ahmed K, Mahdi BD, Hayet Z, Hamdi L, Mohamed J, Riadh M. Thoracic heteropagus conjoined twins associated to an omphalocele: Report of a case and complete review of the literature. Afr J Paediatr Surg. 2016;13(4):209-212.
53. Rattan KN, Singh J, Dalal P, Sonika P, Rattan A: Sacral rachipagus parasite: a case report. J Neonatal Surg 5:16, 2016
54. Raj P, Birua H. Epigastric Heteropagus Twin. J Neonatal Surg. 2017;6(2):47. Published 2017 Apr 15.
55. Raheja A, Mahapatra AK. Rachipagus parasitic twin. Neurol India. 2017;65(6):1443-1444.
56. Khushdil A, Niaz H, Ahmed Z. Epigastric Heteropagus Conjoined Twins. J Coll Physicians Surg Pak. 2018;28(3):S42-S43.
57. Malik M, Bahadur Singh U, Hedge S, Mahajan JK, Samujh R. Omphalocele and epigastric heteropagus: implications and treatment. Oxford Medical Case Reports. 2018 Oct;2018(10):omy074.
58. Khavanin N, Ruge JR, Vicari FA, Belin EJ, Kellogg RG, Steinberg JP. Parasitic rachipagus conjoined twin: case report. J Neurosurg Pediatr. 2018;22(3):313-316.
59. Muhelo AR, Montemezzo G, Da Dalt L, et al. Successful management of a parasitic ischiopagus conjoined twins in a low-income setting. Clin Case Rep. 2018;6(2):385-390. Published 2018 Jan 10.
60. Falyoun MS, Mashlah Q, Aldiri Q, Mohamad AR, Daowd LK, Ayash M. Epigastric heteropagus and omphalocele. Journal of Surgical Case Reports. 2020 Oct;2020(10):rjaa437.
61. Pati AB, Mahalik SK, Naik S, Das K. Heteropagus Twins: A Tale of Two. Neurol India. 2020;68(6):1453-1455.
62. Alves RCS, Petry AUS, Marçal JMB, Hochhegger B, Damin AS, Roehe AV. Asymmetric parasitic twins - Heteropagus. Rev Assoc Med Bras (1992). 2020;66(11):1526-1529

Romanian Neurosurgery (2023) XXXVII (3): pp. 272-277
DOI: 10.33962/roneuro-2023-049
www.journals.lapub.co.uk/index.php/roneurosurgery

The central sulcus. Perioperative identification and surgical implication

Teeba A. Al-Ageely[1], Abdullah A. Abdulhaleem[1], Sara A. Mohammed[1], Mustafa Ismail[1], Hagar A. Algburi[1], Hayder R. Salih[2], Samer S. Hoz[3]

[1] College of Medicine, University of Baghdad, IRAQ
[2] Department of Neurosurgery. Neurosurgery Teaching Hospital, Baghdad, IRAQ
[3] Department of Neurosurgery, University of Cincinnati College of Medicine, Cincinnati, OH, USA

Keywords
cerebral cortex,
central sulcus,
craniometry,
neuroradiology,
neurosurgical approaches

Corresponding author:
Mustafa Ismail

College of Medicine, University of Baghdad, Iraq

mostafa.ismail1700d@
comed.uobaghdad.edu.iq

Scan to access the online version

ABSTRACT
The central sulcus is an important anatomical landmark the location of most of the anatomical structures and cortical lesions are described by their relation to the central sulcus [9,19]. During direct observation of the cerebral cortex, it is not always easy to understand the cortical anatomy of the sulci and gyri due to the presence of arachnoid matter. Furthermore, there often is anatomical variation in this region [13]. Therefore, this paper presents the crucial methods for identifying the central sulcus's exact anatomical location as it is critical for the neurosurgical team and to discuss its surgical implications.

PERTINENT ANATOMY

The central sulcus is located between the frontal and the parietal lobes of the brain, separating the primary motor cortex in the precentral gyrus anteriorly from the primary sensory cortex in the postcentral gyrus posteriorly [9,19]. Classically, the central sulcus is divided into three parts: an upper curvature with an anteriorly directed convexity, a lower curvature that is - although less well defined - also anteriorly directed, and in between, there's a middle curvature with a posteriorly directed convexity. The central sulcus is generally continuous, although an interruption is seen in approximately 1% of cases [10,19]. The approximate percentage of the depth of the superior peak of the central sulcus is 36% of the sulcal length, the inferior peak at 62% of the sulcal length, and the 'Pli de Passage Fronto-Pariétal Moyen' (PPFM), which is a surface landmark of the primary motor area of the hand was at 47% of sulcal length. The mean sulcal depth varies slightly between the two hemispheres being 16.6 ± 1.3 mm for the left central sulcus and 16.4 ± 1.2 mm for the right sulcus [3].

The inferior end of the central sulcus often doesn't reach the sylvian fissure, although there's usually a small bridge connecting the inferior ends of the precentral and post-central gyri; however, in 19% of cases, there was an anastomosis with the sylvian fissure, in 17% with the anterior subcentral sulcus, and in 7% with the posterior subcentral sulcus. The superior end of the central sulcus, however, extends into the medial surface in the majority of cases, and in the few in which it doesn't reach the superior margin, it can end in a bifurcation. An anastomosis with the cingulate sulcus was only described by Benedikt. The inferior and superior ends have numerous variations in shape (straight shaped, T shaped, Y shaped) and direction, whether anteriorly or posteriorly. The central sulcus can connect with the precentral gyrus, postcentral gyrus, and small free sulcus over the precentral gyrus. The side branches of the central sulcus vary greatly in their location and number. But due to the variability and inconsistency, their clinical and anatomical significance is scarce [10].

Similar variations of the normal sulcal anatomy in the central lobe of the brain were reported by observing the sulcal patterns while studying the in vivo magnetic resonance imaging (MRI) data. The different criteria between the two studies included the straight-shaped inferior end and T-shaped inferior end of the central sulcus, along with the number of sulcal connections with small free sulcus in the left precentral gyrus. Yet, the most observed difference was related to the number of side branches and connections, with more depressions seen in the post-mortem description than in the MRI analysis, which can be attributed to the lower spatial resolution in MRI data [7,10].

Preoperative identification

Perioperative identification of the central sulcus can be done using craniometry which utilizes certain craniometric points as the landmarks from which measurements of the skull and facial structures to analyze specific osseous features in different populations. The application of the craniometric points established the footing of modern neurosurgery to tailor craniotomies in specific areas of attention [11,16].

The Tylor-Haughton method utilized different lines to localize the central sulcus using the Frankfurt plane (which is a line extending from the inferior margin of the orbit to the superior margin of the external auditory meatus), the distance between the two points, the nasion and the inion (Na-In) along the calvarium is divided in quarters (25%-50%-75%), the line from the middle of the orbit to the 75% mark of the (Na-In), which corresponds to the sylvian fissure from the orbit to posterior ear line (a perpendicular line from the mastoid directed upward). The central sulcus is situated 2cm posterior to the 50% mark between (Na-In) corresponding to the superior Rolandic point (SRP), to the connection of the sylvian fissure and condyle line (which is a perpendicular line extending from the mandibular condyle directing upward), corresponding to the inferior Rolandic point (IRP) [16].

According to Broca, the central sulcus is identified by an oblique line between two main points: the superior Rolandic point (SRP) and the inferior Rolandic point (IRP). The SRP can be located from the uppermost point of the second vertical line at its connection with the horizontal line intersecting the Bregma. The IRP is situated at the intersection of the horizontal line from the external angular process of the frontal bone to the lambda suture and the auriculo-bregmatic line [16].

Rothon's method of detecting the central sulcus includes three lines. The first one is the (Na-In) line. The second line represents the sylvian fissure which is an oblique line extending from the outer angular process of the frontal bone to the three-fourths point of the (Na-In) line. The third line is a tilted line extending between the midpoint of the zygomatic arch to the halfway point of the (Na-In) line. The intersection of this last line with the 50% mark on the Na-In line corresponds to the SRP. The IRP is defined at the meeting of the midpoint of the third line with the sylvian fissure [16].

Several methods based on imaging modalities such as computerized tomography (CT) or MRI have been suggested to help neurosurgeons and neuroradiologists in accurately localizing the central sulcus. On conventional CT and MRI, the central sulcus is defined indirectly in relation to cortical or commissural landmarks, which are often easily identified in the normal brain [19].

Radiological efforts to identify the central sulcus are imminent, including the localization of the central sulcus by detecting specific radiological signs. One of the most commonly suggested methods consists of localizing the precentral knob, which can be recognized on the axial plane by the form of the Greek letter inverted 'Omega', which is named the 'Omega sign' [13]. The contralateral Omega sign can be applied to clarify the topographic location of a lesion, giving a quick idea of the relationship between the pathology and the precentral and postcentral gyri preoperatively [2] (Figure 1A).

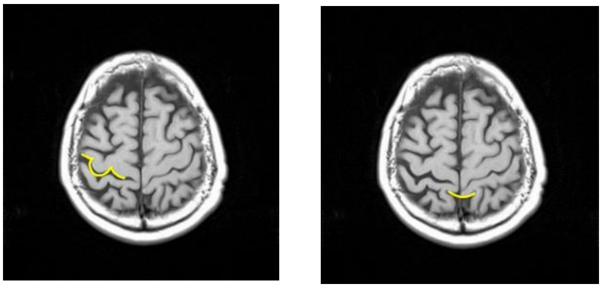


Figure 1. 1A: Brain MRI (Axial section) showing: Omega sign. **1B**: Brain MRI (Axial section) showing: Bracket sign.

While the Omega sign may be a useful anatomic landmark for the preoperative localization of the hand motor area, precise localization of the sign depends on the experience, anatomical distortion, and the need for multiple scanning planes [20].

Other less reliable signs from the axial aspect include the Bracket sign, the upper and the lower T signs, the lower L sign, the M sign, the bifid postcentral gyrus sign, the thin postcentral gyrus sign, the midline sulcus sign, and the 'white grey sign' [5,8,13,15,18]. The 'U sign' characterizes the subcentral gyrus, which is a U-shaped gyrus surrounding the most inferolateral extent of the central sulcus [8,18] (Figure 1B-E).

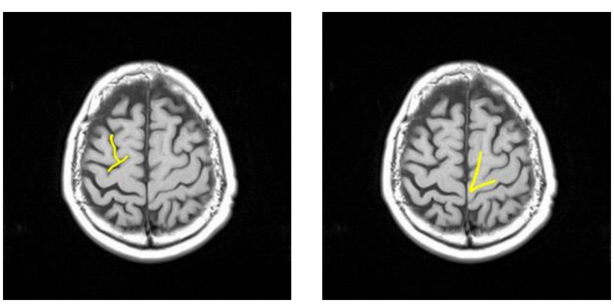


Figure 1. 1C: Brain MRI (Axial section) showing: Upper 'T' sign. **1D**: Brain MRI (Axial section) showing: 'L' sign.

These radiological signs are inconsistently helpful in identifying the central sulcus, and their reported presence ranged between 54.5% and 98.9% [18]. Notably, most of these signs are based on the superior aspect of the central sulcus. The 'invisible cortex sign', on the other hand, is seen as iso-intensity in the peri-rolandic cortices relative to the adjacent white matter on diffused weighted imaging (DWI) MRI. It can be of value in detecting the inferolateral part of the central sulcus on a single axial image without needing to rely on the more superior aspects of the sulcus [15].

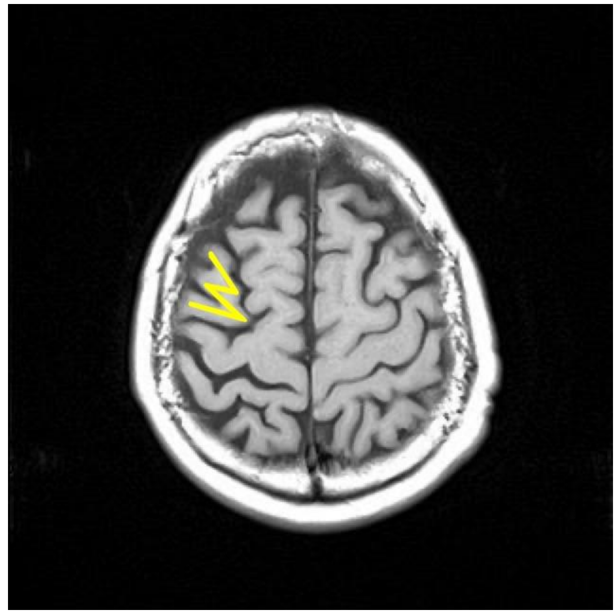


Figure 1. 1E: Brain MRI (Axial section) showing: 'M' sign.

The central sulcus can also be identified by localizing the central sulcal vein using functional MRI (fMRI) [19]. Complex finger movements can be studied to detect the function of the primary motor hand area also by using fMRI and positron emission tomography (PET) scan [20].

INTEROPERATIVE IDENTIFICATION

Intraoperative identification of deep-seated or very small cortical pathologies may be difficult because of the absence of visible anatomical landmarks. Technical development in computer-assisted surgery (CAS) has led to the introduction of frameless image-guided navigation systems for identifying small or deep-seated lesions and intraoperative anatomical guidance [6].

The brain is considered a geometric bulk that can be divided by three imaginary interconnecting spatial planes (horizontal, frontal, and sagittal) based on the Cartesian coordinate system. Any point within the brain can be localized by measuring its distance along these three intersecting planes. Neuro-navigation provides accurate surgical orientation by referencing this coordinate system of the brain with a parallel coordinate system of the three-dimensional image data of the patient; therefore, these images become precise point-to-point maps of the corresponding site within the brain [4,14].

Certain intraoperative electrophysiological mapping techniques are combined with neuro-navigation to relate the functional regions to the site of the lesion and to the patient's distinct anatomy [6]. Neuro-navigation reduces the length of surgery, lowers the risk of wound infections, and shortens the time of hospital stay. In addition, it reduces the risk of neurological morbidity by allowing the neurosurgeon to assess the relationship between the lesion and the surgical approach to protect the neighboring critical brain structures [1].

Surgical implications

The operative neurosurgical approaches that expose the central sulcus are classified according to the specific segment of the central sulcus exposed.

Figure 2. 2A: The surgical approaches that are exposing the central sulcus: Typical pterional approach showing the inferior end of the central sulcus (red arrow). **2B**: The surgical approaches that are exposing the central sulcus: The para sagittal approach shows the superior end of the central sulcus (red arrow). **2C**: The surgical approaches that are exposing the central sulcus: The paramedian approach shows the middle and the medial segment of the central sulcus (red arrow).

The approaches that expose the inferior end of the central sulcus include the typical Pterional approach, which can also reveal the subcentral gyrus and the cortical area surrounding the sylvian fissure [12]. The approaches that expose the superior end of the central sulcus include the parasagittal approach (with or without transcallosal approach) around the midline of the cranial vault for lesions along the midline and the interhemispheric fissure, therefore, it can reveal the upper part of the central sulcus [1]. The approaches that expose the middle segment of the central sulcus include the paramedian retro-coronal approach for eloquent and peri-eloquent cortical lesions (Table 1) (Figure 2).

Table 1. Neurosurgical approaches revealing the central sulcus according to the segment exposed.

Exposed segment of the central sulcus	**Surgical approach**	**Description of the approach**
Inferior end of the central sulcus	Typical Pterional approach	Pterional craniotomies expose the Sylvian fissure with the inferior part of the middle frontal gyrus, inferior frontal gyrus, superior temporal gyrus, and the middle temporal gyrus. This approach is indicated for vascular pathologies arising from the circle of Willis, lesions located in or around the cavernous sinus, the sella turcia, and the parasellar and subfrontal regions (12).
The superior end of the central sulcus	Parasagittal approach with or without transcallosal approach	The parasagittal approach is indicated in craniotomies to reach the midline of the cranial vault, which require safe exposure of the superior sagittal sinus or its boundaries. Used mainly in common lesions of the midline of the cranial vault, including parasagittal and Falcine meningiomas, along with lateral and third ventricle lesions, which often directly involve the superior sagittal sinus (1).
The middle segment of the central sulcus	Paramedian retro-coronal approach	It is the parasagittal approach when the targeted lesions are cortical or subcortical. Particularly when the lesions are around the central sulcus, i.e., lesions deep or within the precentral and postcentral gyrus. These pathologies can

		be approached by parasagittal or paramedian craniotomy located behind the coronal suture. This approach will expose the middle segment of the central sulcus and does not include the lateral end or the medial end segments of the sulcus.

Limitations

There are multiple shortcomings in almost all the methods for central sulcus identification, including the numerous anatomical variations and inaccuracy of the imaging modalities, the invasiveness, time consuming, as well as unavailability of fMRI or neuro-navigation [10,18,20].

Considering the risk and the eloquence of the area and the lack of the exact operative knowledge for most neurosurgeons in the pericentral area. Recognition of all the variations and revision of all the identification methods aid the neurosurgeons in identifying the central sulcus will be of critical value for the success of the surgery and improving the patients' outcome.

Based on the above, although the advancement of imaging techniques to has a huge value in localizing the central sulcus, however, central sulcus identification still forms a surgical challenge for most neurosurgeons. Utilizing preoperative craniometry, preoperative imaging identification methods, intraoperative neuro-navigation, intraoperative neurophysiological electrophysiological monitoring, and cortical stimulation as a collaborative effort will definitely increase the safety of the surgery in any approach targeting the pericentral areas.

Conclusion

Although the central sulcus is a very important anatomical landmark, variability in its parts and anatomy can make it difficult to readily identify it. Integrating the preoperative and intraoperative techniques described can minimize the complications of false identification in surgery, and further understanding of the variability in the anatomy and its surgical implication helps increase the safety of surgery.

Abbreviations

CAS: Computed assisted surgery,
CT: Computed tomography,
DWI: Diffuse weighted imaging,
fMRI: Functional magnetic resonance imaging,
IN: Inion,
IRP: Inferior Rolandic point,
MRI: Magnetic resonance imaging,
Na: Nasion,
PET: Positron emission tomography,
PPFM: Pli de Passage Fronto-Pariétal Moyen,
SRP: Superior Rolandic point.

References

1. Burke JF, Han SJ, Han JH, McDermott MW. Two-part parasagittal craniotomy. Cureus. 2014 Aug 18;6(8).
2. Campero A, Ajler P, Martins C, Emmerich J, de Alencastro LF, Rhoton Jr A. Usefulness of the contralateral Omega sign for the topographic location of lesions in and around the central sulcus. Surgical Neurology International. 2011;2.
3. Cykowski MD, Coulon O, Kochunov PV, Amunts K, Lancaster JL, Laird AR, Glahn DC, Fox PT. The central sulcus: an observer-independent characterization of sulcal landmarks and depth asymmetry. Cerebral cortex. 2008 Sep 1;18(9):1999-2009.
4. Golfinos JG, Fitzpatrick BC, Smith LR, Spetzler RF. Clinical use of a frameless stereotactic arm: results of 325 cases. Journal of Neurosurgery. 1995 Aug 1;83(2):197-205.
5. Kaneko OF, Fischbein NJ, Rosenberg J, Wintermark M, Zeineh MM. The "white gray sign" identifies the central sulcus on 3T high-resolution T1-weighted images. American Journal of Neuroradiology. 2017 Feb 1;38(2):276-80.
6. Krombach GA, Spetzger U, Rohde V, Gilsbach JM. Intraoperative localization of functional regions in the sensorimotor cortex by neuronavigation and cortical mapping. Computer Aided Surgery: Official Journal of the International Society for Computer Aided Surgery (ISCAS). 1998;3(2):64-73.
7. Mellerio C, Lapointe MN, Roca P, Charron S, Legrand L, Meder JF, Oppenheim C, Cachia A. Identification of reliable sulcal patterns of the human Rolandic region. Frontiers in Human Neuroscience. 2016 Aug 17;10:410.
8. Naidich TP, Blum JT, Firestone MI. The parasagittal line: an anatomic landmark for axial imaging. American journal of neuroradiology. 2001 May 1;22(5):885-95.
9. Oberman DZ, Rasmussen J, Toscano M, Goldschmidt E, Ajler P. Computed tomographic localization of the central sulcus: a morphometric study in adult patients. Turk Neurosurg. 2018 Jan 1;28(6):877-81.
10. Ono M, Kubik S, Abernathey CD. Atlas of the cerebral sulci. Thieme; 1990.
11. Ribas GC. Applied cranial-cerebral anatomy: brain architecture and anatomically oriented microneurosurgery. Cambridge University Press; 2018 Mar 1.
12. Roberto RR, Ricky C, Ioannis K, Adib A, McDermott M. Immersive Surgical Anatomy of the Frontotemporal-

Orbitozygomatic Approach. Cureus. 2019;11(11).

13. Rodrigues T, Rodrigues M, Paz D, Costa MD, Santos B, Braga V, Paiva Neto MD, Centeno R, Cavalheiro S, Chaddad-Neto F. Is the omega sign a reliable landmark for the neurosurgical team? An anatomical study about the central sulcus region. Arquivos de Neuro-Psiquiatria. 2015;73:934-8.
14. Selesnick SH, Kackar A : Image guided surgical navigation in otology and neurootology. American Journal of Otology 1999; 20 : 688-697.
15. Su S, Yang N, Gaillard F. Invisible cortex sign: a highly accurate feature to localize the inferolateral central sulcus. Journal of medical imaging and radiation oncology. 2019 Aug;63(4):439-45.
16. Vigo V, Cornejo K, Nunez L, Abla A, Rubio RR. Immersive Surgical Anatomy of the Craniometric Points. Cureus. 2020 Jun 15;12(6).
17. Wadley J, Dorward N, Kitchen N, Thomas D. Pre-operative planning and intra-operative guidance in modern neurosurgery: a review of 300 cases. Annals of the Royal College of Surgeons of England. 1999 Jul;81(4):217.
18. Wagner M, Jurcoane A, Hattingen E. The U sign: tenth landmark to the central region on brain surface reformatted MR imaging. American Journal of Neuroradiology. 2013 Feb 1;34(2):323-6.
19. Yousry TA, Schmid UD, Schmidt D, Hagen T, Jassoy A, Reiser MF. The central sulcal vein: a landmark for identification of the central sulcus using functional magnetic resonance imaging. Journal of neurosurgery. 1996 Oct 1;85(4):608-17.
20. Zakaria HM, Massa PJ, Smith RL, Moharram TH, Corrigan J, Lee I, Schultz L, Hu J, Patel S, Griffith B. The reliability of identifying the Omega sign using axial T2-weighted magnetic resonance imaging. The Neuroradiology Journal. 2018 Aug;31(4):345-9.

Romanian Neurosurgery (2023) XXXVII (3): pp. 278-282
DOI: 10.33962/roneuro-2023-050
www.journals.lapub.co.uk/index.php/roneurosurgery

Non-missile penetrating spinal injury with an impaled knife. An uncommon injury with an unlikely outcome

Bruno Arekhandia[1], Williams Achibiri[1,2], Muyiwa Kayode Rotimi[3], Edward Jolayemi[1], Bamidele Adebayo[2], Omotayo Ojo[1,4], Okezie Obasi Kanu[1,4]

[1] Neurosurgery Unit, Department of Surgery, Lagos University Teaching Hospital, Idi-Araba, Lagos State, NIGERIA
[2] Neurosurgery Unit, Department of Surgery, Federal Medical Centre, Abeokuta, NIGERIA
[3] Department of Anaesthesia, Lagos University Teaching Hospital, Idi-Araba, Lagos State, NIGERIA
[4] Neurosurgery Division, Department of Surgery, College of Medicine, University of Lagos, Idi-Araba, Lagos State, NIGERIA

Keywords
non-missile penetrating spinal injury (NMPSI), impaled knife, Brown-Sequard-plus syndrome, complete spinal cord injury

Corresponding author:
Okezie Obasi Kanu

College of Medicine University of Lagos, Nigeria

drkanu@gmail.com

Scan to access the online version

ABSTRACT

Retention of all or part of the weapon following a non-missile penetrating spinal injury is a rare occurrence. The authors report a case of thoracic spinal cord stab injury in a young man. The patient presented with a knife blade lodged in his back and with Brown-Sequard-plus syndrome. At surgery, the wound with the knife in situ was explored and the knife was removed. Although the wound healed without evidence of cerebrospinal fluid leakage or infection, he developed a complete neurologic deficit post-operatively. This was an unlikely outcome for incomplete spinal cord injury resulting from non-missile penetrating spinal injury, historically known to have a favourable outcome. Pre-operatively, patients with incomplete neurological injury following penetrating spine injury with the retained foreign body should be specifically counselled on the possibility of a worsened neurological outcome after surgical intervention.

INTRODUCTION

Spinal cord injury is associated with long-term disability and substantial economic burden; disproportionately affecting younger populations mainly in their third decade. (1,2) While blunt trauma accounts for most traumatic spinal injuries, penetrating injuries represent higher rates of morbidity, disability, and financial burden.(1) Furthermore, non-missile-penetrating spinal cord injuries are rare and account for less than 1.5% of the total penetrating injuries.(2) Most of the weapons are already withdrawn by the attacker in spinal stab injuries, but rarely may get impacted into the bone and is retained either as a whole or as fragments requiring surgery.(3,4) Spinal cord injury resulting from stab wounds are usually incomplete with most cases appearing typically as the Brown-Sequard-plus syndrome – a term reserved for the less pure forms of the original description of cord hemisection.(5) Neurologic recovery overall fares well generally for these injuries – with up to 97% significant recovery in a series.(6) On the other hand, Lipschitz opined that if spinal injury due to stab wound is neurologically complete over 24 hours after injury neurological recovery was rare, and if present, negligible.(7) Literature on outcome specifically for penetrating spinal cord injury with retained weapon is unsurprisingly rare, due to the uncommon nature of these injuries.

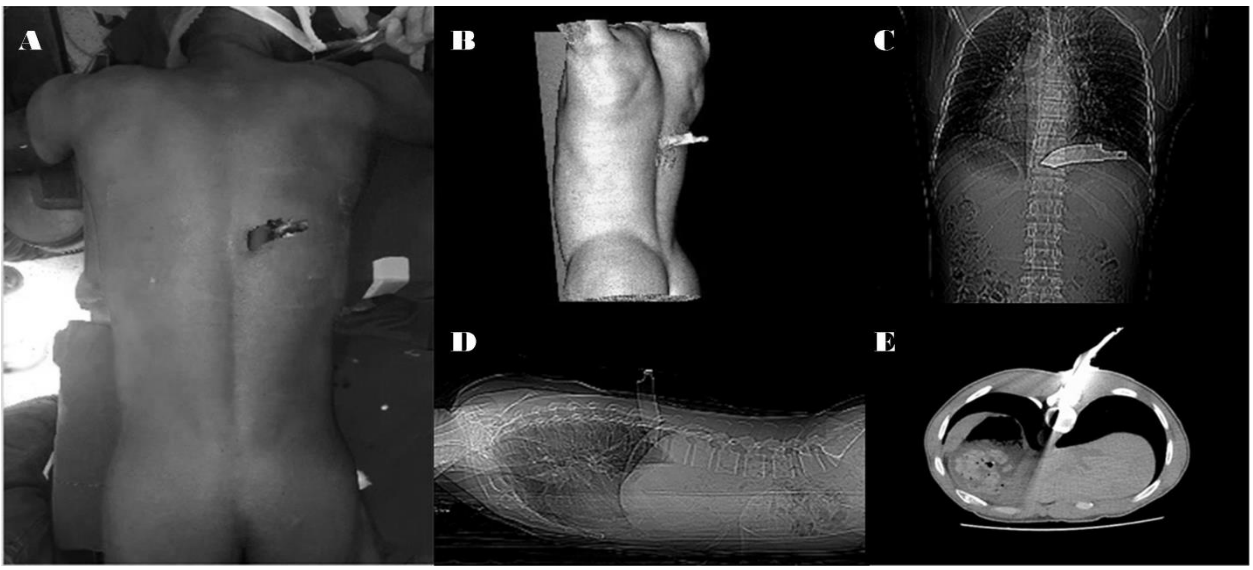


Figure 1. Non-missile Penetrating Spinal Injury. **A**: Patient in prone position showing the point of entry; **B-E**: Reconstructed images showing direction and depth of entry.

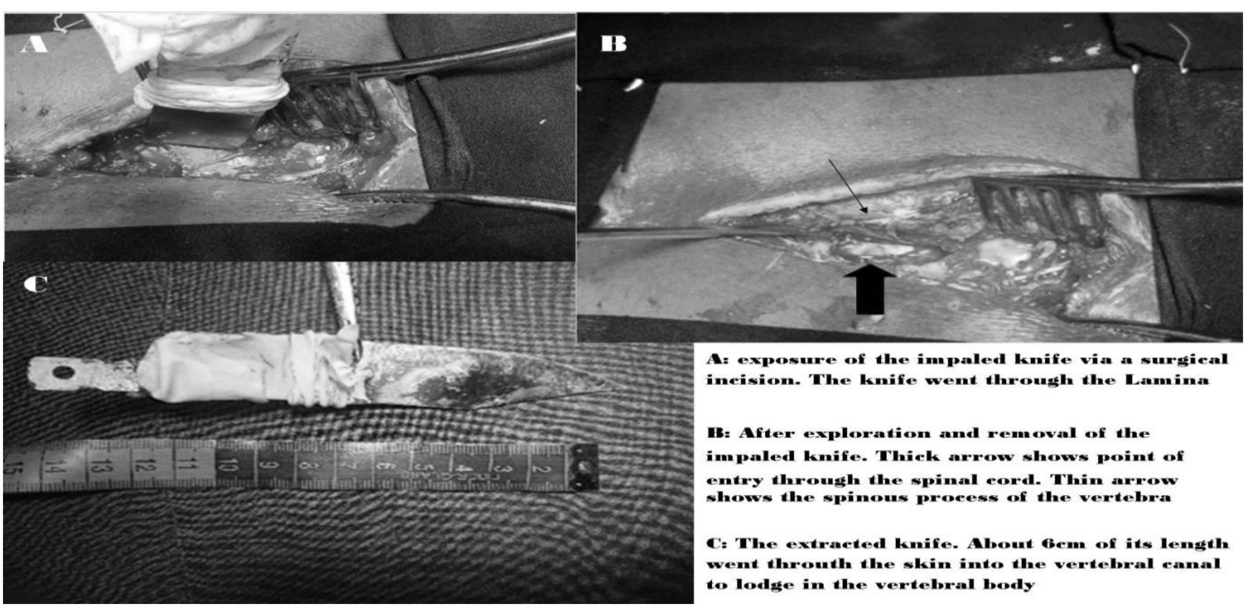


Figure 2. Intraoperative images of non-missile penetrating spinal injury. **A-B**: Surgical exposure and extraction; **C**: Impaled knife after extraction.

CASE REPORT

A 40-year-old man was stabbed in the back with a knife by unknown assailants who also hit his waist with a block, four hours prior to presentation. He sustained a single stab wound which bled minimally. He perceived immediate paraesthesia in the left lower limb but no weakness or sphincteric compromise. There were no significant injuries involving other regions of the body.

On examination he was a young man, fully conscious and lying prone. His heart rate was 90 beats per minute, respiratory rate of 23 cycles per minute and arterial blood pressure was 100/65mmHg. His Glasgow coma score was 15 and he had intact higher mental function. There was normal sensation to light touch and pin prick on the right side of the body and sensation up to L2 on the left, as well as hypoesthesia down to L5. (Motor examination was made difficult by the prone positioning) but power in the lower limb muscle groups were at least 3 in the left lower limb and at most 2 in the right lower limb. There was no evidence of acute urinary retention prior to urethral catheterization and rectal tone was normal.

A knife blade was seen stuck in the T10/T11 interspace to the right of the midline, with clotted blood surrounding it. (Figure 1A) A diagnosis of penetrating thoracic spinal injury with Brown-Sequard-plus syndrome – grade C on the ASIA scale – was made.

A thoracic spine CT was performed considering the obvious risk of an MRI in the presence of a ferromagnetic object; this showed a 6 cm hyperdense foreign body, consistent with a knife, entering the right T11 lamina. A radiograph improved on the poor visualization of the path of the foreign body, occasioned by ferromagnetic streak artefact on the CT, and clearly showed the downward and medial trajectory of the knife. (Fig. 1B-E)

He was worked up for planned extraction of the retained knife.

In the operating room, pre-oxygenation before anaesthetic induction, and tracheal intubation with the aid of a laryngeal mask airway, were done in lateral position as patient could not lie supine. At surgery, under intraoperative magnification, the stab wound was extended and the underlying paravertebral muscle was dissected along the tract of the knife down to the vertebral lamina. The knife's entry point was found entirely in to the lamina lateral to the spinous of T11 on the right. Although rigidly held in position, attention was paid to avoid moving with knife while dissecting, to avoid spinal cord damage. Right-sided T10 hemi-laminectomy was done using Kerrison punches. Following adequate exposure, the stuck tip of the knife piece was easily extracted with minimal additional bleeding that was readily controlled. The estimated entire retained length deep to the skin was 6cm (Figure.2 A-C). Inspection of thecal sac showed a small longitudinal split with minimal cerebrospinal fluid leak, which was sealed with Surgicel. Copious irrigation, and then meticulous soft tissue closure was done in layers after a closed passive drain was left in situ.

Immediate and subsequent postoperative neurological examination revealed a sensory level of T10 and power in both lower limbs was 0 in all muscle groups respectively. Wound drain was minimal, and no evidence of post-operative cerebrospinal fluid leak or wound infection was present.

A post-operative magnetic resonance scan could not be done due to financial constraint. He has been followed up for two months post-injury during which the neurological injury was yet to evolve from the E rating on the ASIA scale, while his bladder remains neurogenic.

Discussion

Apart from the longest case series on penetrating spinal cord injury which was published by Peacock et al from South Africa, the global literature contains only rare case reports and small case series on NMPSI.(8)

Spinal cord injury resulting from stab wounds are usually incomplete.(9) In the South African study earlier referred to, Peacock and colleagues found out that half of the 450 patients from a total of 1,600 patients had Brown-Sequard syndrome. Most of the injuries occurred in the thoracic region, and knives were found to be the mostly used object, both in this series and in the next most impactful series on this subject matter. (10, 11)

Usually, the weapon enters the spinal canal via the interlaminar region between the spinous process and collide with the bony elements of the spine, and is compelled through a trajectory that guides the knife to only one side of the spinal cord, thereby resulting in Brown-Sequard syndrome.(9,12) Our patient's injury was most likely through this mechanism.

The proposed mechanisms of injury to the cord include direct damage by the weapon or in-driven bone fragments, damage of the vascular supply of the cord or coup-countercoup spinal contusions.(3) The first is an immediate mechanism and the second is a delayed mechanism.(12) The occurrence of neurologic deficit immediately after the assault suggested a direct cord injury in our patient, and the subsequent deterioration was likely of a vascular cause or secondarily from cord oedema.

While there is significant variability in penetrating spinal trauma management practices due to paucity of data on penetrating spine trauma generally, existing literature provides some guidance on surgical indication, reserving surgical intervention for progressive neurological deficit, spinal instability, or infection control.(1) Additionally, for foreign bodies lodged in the spinal canal the need for surgery is obvious to prevent infection, myelopathy and delayed neurological deficit.(3) The objectives of the surgical treatment are therefore to decompress the spinal cord, remove the foreign body, control infection and prevent cerebrospinal fluid leakage.(13)

Generally, in the management of retained foreign bodies, the manipulation or blind removal of a retained weapon before a careful evaluation can cause a significant bleeding, because of the theoretical plugging effect of the weapon over adjacent vessels.(14) Therefore, the standard of care in retained foreign body injuries to the spine remains extraction performed on the operating table.(3) The options for removal of the foreign body were summarised by Sobnach to include simple extraction, wound exploration and extraction, or open operation and extraction.

Arguments in favour of simple extraction hold the position that open surgical explorations will add little benefits if any, and have surgical risks in neurologically intact spinal stab wound patients, but added that during simple extraction the operation team must be ready for urgent exploration of the wound in the event of brisk bleeding or CSF leakage after withdrawal.(3)

One of the challenges with patients with the perioperative management of a penetrating spinal injury with an impaled knife is that of positioning for endotracheal intubation. Tracheal intubation in the lateral position is difficult because the laryngeal view is compromised during direct laryngoscopy.(15) There are several reports of successful ventilation with the laryngeal mask airway (LMA).(16) The left lateral position is favoured because of the relative difficulty in the right lateral position was attributed to the positioning of the tongue, which has a tendency to slip off the laryngoscope blade while the blade is inserted from the right side of the tongue.(17)

At surgery, identification and repair of dural tears is done to prevent the formation of CSF fistulas, pseudomeningoceles, and intradural infections.(18) In the case we presented oxidized regenerated cellulose and meticulous soft tissue closure was sufficient to prevent CSF leakage post-operatively.

The neurological outcomes from penetrating spine injury other than those presenting with Brown Sequard syndrome, are worse than those of blunt injuries.(9) In Peacock's series, about half of the patients had Brown-Sequard syndrome, and two-thirds of them had a positive functional recovery.(10) This promising neurological recovery has been attributed to the neuronal plasticity of the spinal cord.(9)

In illustrating both the favourable outcome in incomplete penetrating spinal cord injuries as well as beneficial role of surgery in their management, the report by Manzone and colleagues comes to mind. They reported a 22-year old man who sustained a thoracic spinal cord stab injury with a concealed broken knife tip which was retained for several months before surgery; neurologic improvement was noted to be rapid after operative removal of the retained object. (19)

Nasser et al. reported a unique case in a 34-year old male, whose neurological status essentially improved post-operatively to ASIA E, from a pre-operative status of C, in spite of injuries caused by the ice pick fragment penetrating posteriorly through the spinal canal into the aorta.(20)

Rabiu and colleagues reported a case of C4 Brown-Sequard syndrome with the impaling object – a screw driver – in situ, who had some neurologic improvement post-surgery, but later died from unrelated causes.(8)

The absence of a similar favourable outcome in our patient was envisaged during the process of acquiring informed consent for surgery, and he was so counselled.

An understanding of the pathophysiological mechanism underlying this uncommon outcome was partly made difficult by the patient's failure to carry out post-operative spine MRI. Possibilities may include firstly, the mass effect from an intraspinal haematoma following release of the theoretical plugging effect by the weapon over vessels within the cord. Another possible mechanism probably involved the artery of Adamkiewicz, considering the proximity of the injury to artery.(20) Indeed the artery of Adamkiewicz and the aorta are the most commonly injured vascular structures from NMPSI.(12) It must be stated however that the loss of sacral root functions in the patient post-operatively did not support the second consideration because injury to the artery of Adamkiewicz usually presents with signs of thoracic watershed ischemia — paraplegia with relative sparing of the sacral roots.(21) .

Conclusion

This case reflects some of the difficulties encountered in an attempt to describe an unlikely outcome in a rare entity in a resource-poor setting. The reality of a worsened outcome following operative retrieval of a retained foreign body in the spine should be specifically brought to the fore in counselling patients needing intervention.

References

1. Morrow KD, Podet AG, Spinelli CP, Lasseigne LM, Crutcher CL, Wilson JD, et al. A case series of penetrating spinal trauma: comparisons to blunt trauma, surgical indications, and outcomes. Neurosurgical Focus. 2019 Mar;46(3):E4.
2. Kramer MM, Acker A, Ohana N. Penetrating Spinal Cord Injury. In: Dionyssiotis Y, editor. Essentials of Spinal Cord Injury Medicine [Internet]. InTech; 2018 [cited 2021 Dec 28]. Available from:
3. http://www.intechopen.com/books/essentials-of-spinal-cord-injury-medicine/penetrating-spinal-cord-injury
4. Sakar M, Dogrul R, Niftaliyev S, Bayri Y, Dagcınar A. Direct withdrawal of a knife lodged in the thoracic spinal canal in a patient with normal neurologic examination: is it safe? Spinal Cord Ser Cases. 2016 Dec;2(1):16009.
5. Prasad BC, Vemula RC, Varaprasad G. Non-missile Penetrating Spinal Injury with an Impaled Knife: Case Report. Indian J Surg. 2013 Jun;75(3):237–8.
6. Gulamhuseinwala N, Terris J. Evolving presentation of spinal canal penetrating injury. Injury. 2004 Sep;35(9):948–9.
7. Wallace DJ, Sy C, Peitz G, Grandhi R. Management of non-missile penetrating spinal injury. Neurosurg Rev. 2019 Dec;42(4):791–8.
8. Waters RL, Sie I, Adkins RH, Yakura JS. Motor recovery following spinal cord injury caused by stab wounds: A multicenter study. Spinal Cord. 1995 Feb;33(2):98–101.
9. Rabiu TB, Aremu AA, Amao OA, Awoleke JO. Screw driver: an unusual cause of cervical spinal cord injury. BMJ Case Rep. 2011 Sep 5;2011:bcr0620114309.
10. Pascual MG, Alcácer VS, Pomares MV, Alberola MA. Brown-Séquard-plus syndrome after a stab injury. NeuroRehabilitation. 2011;29(4):353-7.
11. Peacock WJ, Shrosbree RD, Key AG. A review of 450 stab wounds of the spinal cord. S Afr Med J. 1977 Jun 25;51(26):961-4.
12. Lipschitz R, Block J. Stab wounds of the spinal cord. Lancet. 1962 Jul 28;2(7248):169-72.
13. Fiani B, Figueras RA, Stefano F, Gautam N, Khan A, Soula M. Nonmissile penetrating spinal injuries: Mechanisms, expectations, and management. Surg Neurol Int. 2020 Nov 25;11:406.
14. Gul S, Dusak A, Songur M, Kalayci M, Acikgoz B. Penetrating Spinal Injury With a Wooden Fragment: A Case Report and Review of the Literature. Spine. 2010 Dec;35(25):E1534–6.
15. Obrador LZ, Dolores Pe´rez Dı´az M, Ayuso MC. Retained Weapon Injuries: An Unusual Presentation with a Good Prognosis. Clin Surg. 2016; 1: 1224.
16. Komatsu R, Kamata K, You J, Sessler DI, Kasuya Y. Airway Scope for Tracheal Intubation in the Lateral Position. Anesthesia & Analgesia. 2011 Apr;112(4):868–74.
17. McCaul CL, Harney D, Ryan M, Moran C, Kavanagh BP, Boylan JF. Airway Management in the Lateral Position: A Randomized Controlled Trial: Anesthesia & Analgesia. 2005 Oct;101(4):1221–5.
18. Bhat R, Sanickop CS, Dhorigoll MG, Umrani V, Suresh SN. Comparison of ease of intubation in right and left lateral position using C-MAC videolaryngoscope. Anaesthesia, Pain and Intensive Care. 2013;17:162-165.
19. Sarica C. Steel Rod Impalement Injuries Involving the Spine: Case Reports and Literature Review. Ulus Travma Acil Cerrahi Derg [Internet]. 2018 [cited 2021 Dec 28]; Available from:
20. https://www.journalagent.com/travma/pdfs/UTD-83727-CASE_REPORTS-SARICA.pdf
21. Manzone P, Domenech V, Forlino D. Stab Injury of the Spinal Cord Surgically Treated: Journal of Spinal Disorders. 2001 Jun;14(3):264–7.
22. Nasser R, Nakhla J, Sharif S, Kinon M, Yassari R. Penetrating thoracic spinal cord injury with ice pick extending into the aorta. A technical note and review of the literature. Surg Neurol Int. 2016;7(29):763.
23. Barbano RL. Mechanical and Other Lesions of the Spine, Nerve Roots, and Spinal Cord. In: Goldman's Cecil Medicine [Internet]. Elsevier; 2012 [cited 2022 Jan 21]. p. 2258–69. Available from:
24. https://linkinghub.elsevier.com/retrieve/pii/B9781437716047004073.

Romanian Neurosurgery (2023) XXXVII (3): pp. 283-287
DOI: 10.33962/roneuro-2023-051
www.journals.lapub.co.uk/index.php/roneurosurgery

Diastasis coronal suture fracture with frontoparietal (vertex) epidural hematoma in adult. A rare case

M. Aga Firza Diandra, T. Akmal Kausar, Reyhan Aristo, Luthfy Farhan, Mahyudanil

Department of Neurosurgery, Universitas Sumatera Utara, Medan, Indonesia, INDONESIA

ABSTRACT

Introduction: Vertex epidural hematoma (VEDH) is an uncommon presentation of extra-axial hematomas and comprises 1-8% of all extradural hematomas. It can represent a surgical dilemma regarding when and how to operate, particularly considering the potential implication of the superior sagittal sinus (SSS). It is estimated that up to 85%-95% of EDHs are associated with skull fractures. Nonetheless, diastasis of cranial sutures in adults has scarce scientific reports, let alone with associated epidural hematoma.

Cases: We reported a 42-year-old male patient who came with a main complaint of a lacerated wound on his head due to an accident when he repaired the truck tire. His GCS score was 15 without any neurological deficit. A Head CT scan revealed a hyperdense biconvex lesion on the frontoparietal (vertex), frontal sinus depressed fracture and diastasis fracture of the coronal suture. Craniotomy evacuated EDH and craniectomy elevated depressed fracture with cranialization of frontal sinus were performed. Postoperative there is no surgery complication.

Discussion: Vertex epidural hematoma (VEDH) frequently causes a diagnostic dilemma, both clinically and radiographically. Clinically, VEDH are often indicated by elevated intracranial pressure (a headache which is usually severe and unrelenting, nausea, visual impairment, and vomiting). Those resulted from compression of the venous outflow at the SSS and subsequently decreased absorption and outflow of cerebrospinal fluid (CSF), which may not contribute to the establishment of a specific diagnosis. Vertex fractures are present in most cases with a linear fracture line crosses (usually horizontally) the sagittal suture overlying the hematoma or diastasis of the coronal and/or sagittal suture, with or without significant external sub-galeal hematomas, indicating a vertex impact.

Conclusion: Vertex epidural hematoma is a rare case. In this case, the patient had GCS score 15 without any neurological deficit. The prompt decision to surgery is vital for good outcomes for the patient with VEDH.

Keywords
craniotomy,
vertex epidural hematoma,
diastasis fracture,
good outcome

Corresponding author:
Reyhan Aristo

Department of Neurosurgery,
Universitas Sumatera Utara, Medan,
Indonesia, Indonesia

aristoreyhan14@gmail.com

Scan to access the online version

INTRODUCTION

Vertex epidural hematoma (VEDH) is a relatively uncommon type of posttraumatic intracranial hematoma; constituting roughly 2.5% of all epidural hematomas, and it has certain features which are distinct from EDH at other sites.[1, 2] It is recognized as a separate entity, which has both diagnostic and therapeutic challenges due to its unique pathogenesis and location.

VEDH may be missed in the routine axial computerized tomography (CT) scan because of its location, and bleeding usually derives from the superior sagittal sinus, and its presentation is more often acute than chronic.[1, 2] Fractures are often found, but diastasis of the sagittal suture in adults is quite unique.

Case report

We report a 42 year-old male patient that came to the ER with the chief complaint of lacerated wound on his head due to an accident that happened when he was repairing his truck tire. On admission, his GCS score was 15, and no neurological deficit was found. CT scan of the head revealed a hyperdense biconvex lesion on the fronto-parietal (vertex), frontal sinus depressed fracture and diastasis with depressed fracture of coronal suture.

Figure 1. Clinical Presentation in the ER.

Figure 2. Schedel X-ray.

We performed craniotomy, EDH evacuationis done and craniectomy elevated depressed fracture with cranialization of frontal sinus. EDH with approximately 50 mL in volume evacuated using controlled suction and irrigation; fracture fragments were removed by bone rongeur and venous bleeding from the fracture was controlled with bone wax.

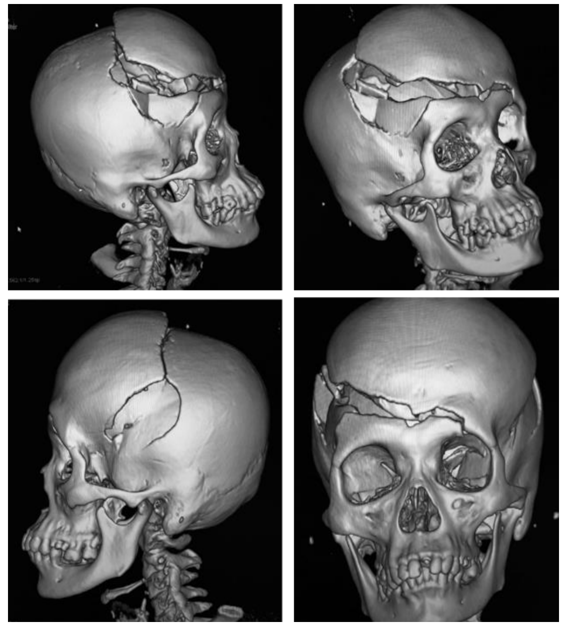


Figure 3. 3D reconstruction.

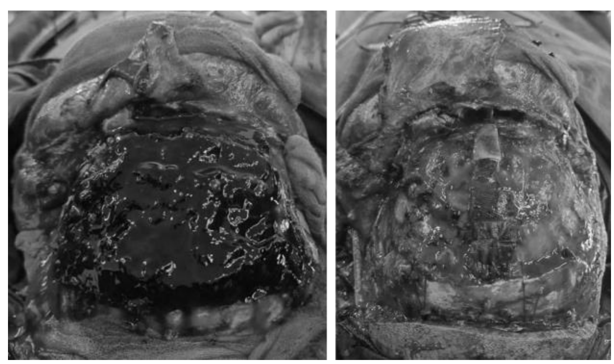


Figure 4. Intraoperative picture of the VEDH,

Following the surgery, the patient returned to the intensive care unit (ICU). He was extubated approximately 48 hours postoperatively; and by day 5 he has GCS of 15 and no neurological deficit was found. He was discharged on the 7th day.

At the time of 1 month follow-up, we performed non-contrast head CT and described cranial defect of right and left frontoparietal bone.

Discussion

Epidural hematoma (EDH) is a common traumatic sequel, and it is estimated that up to 85-95% of EDHs are associated with skull fracture.[2] On the other hand, vertex epidural hematoma (VEDH) is a relatively uncommon type of posttraumatic intracranial hematoma, and it has several unique features that render it a separate entity.[1, 2] It occurs in the quadrangular area of the skull, bounded anteriorly by bregma and coronal sutures; posteriorly by lambda and lambdoid suture; laterally by the pariteal eminence.[1] The source of epidural hematoma are any of the following: 1) a tear in the sagittal sinus, which is the most common source, 2) bleeding from the skull fracture line itself, 3) dural stripping from the innter table of the skull, 4) bleeding from the diseased vascular skull bone, 5) arteriovenous fistula of meningeal artery created by a laceration of dura underlying a linear skull fracture.[1, 3, 4]

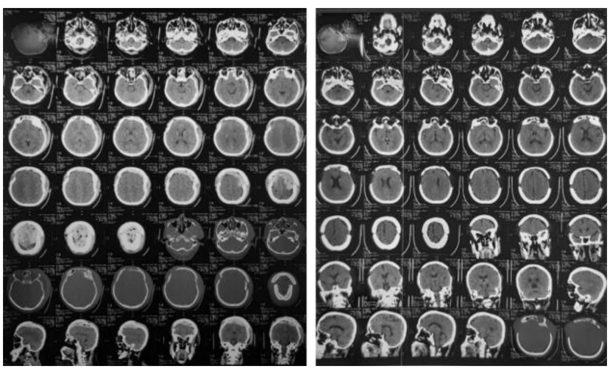


Figure 5. Pre (left) and postoperative (right) CT scans; note the hyperdense biconvex lesion on the vertex seen on preoperative CT.

Figure 6. Clinical presentation at 1 month follow-up.

VEDH is a rare type of posttraumatic intracranial hematoma; in a report by Wylen and Nanda, they reported that VEDH account for 1.3 to 8.2% of all traumatic intracranial hematomas, while vertex intracranial epidural hematoma (VEH) account 8% of all epidural hematomas.[5, 6]

The clinical manifestations of VEDH is highly variable; they may present either in the immediate posttraumatic period, or later. Borzone et al reported that 9 out of 14 patients presented in the acute phase, while Ramesh et al presented that 22 out of 29 patients presented within the first 24 hours with severe headache.[1, 7] Severe headache is considered to be major symptom of VEDH, and there seems to be a pattern where this headache gradually increases in its intensity, from mild to severe.[2, 3, 8] Headache was the presenting symptom in all cases; papilledema was present in five patients and lower limb weakness in five patients. None of them had any cranial nerve involvement.[1]

The pathogenesis of headache in these cases could be either due to dural irritation around the SSS, which is rich in pain sensitive fibers; this meningeal layer has sensory innervationfrom trigeminal nerve; or raised intracranial pressure (ICP) due to obstruction of SSS; while the progressive intensity of this headache is thought to be due to the gradual detachment of the dura mater covering the SSS.[2]

Some other patients may present with other features of increased ICP; a headache which is usually severe and unrelenting, nausea, visual impairment, and vomiting.[3, 8] Those were results from compression of the venous outflow at the SSS and subsequently decreased absorption and outflow of cerebrospinal fluid (CSF), which may not contribute to the establishment of a specific diagnosis.

Other presenting feature of VEDH may include lower limb weakness, either unilateral or bilateral; while some other may also have upper-limb weakness or hemiparesis.[5, 9, 10] This is because of the pressure given directly to the motor cortex representing the leg area. The presence of pure motor weakness without sensory involvement should alert the attending physician to the possibility of the intracranial cause of the weakness.[1] Another possibility of VEDH feature is cranial nerve involvement, which is quite unusual in VEDH, but there has been a report of VEDH presenting with unilateral third nerve palsy.[9]

Acute VEDH might go overlooked on classic CT scans, due to the blind spot near the top of the calvaria, because fresh blood of acute VEDH is isodense with the nearby bone of the upwardly coning skull and might somehow overlap with it, unless coronal and sagittal reconstructions are made from the axial scans.[2] This, unfortunately, requires studies with thin slices; otherwise the reconstruction will have somewhat poor quality. Even if noticed, the fresh blood might be mistaken for hyperostosis, meningioma, dural lymphoma, or plasmocytoma.[2] In routine axial CT scan, VEDH may be seen as 1) vague hyperdense lesion in the highest slices, which often be dismissed as an artifact; 2) fracture line is running across the vault of the skull on either side; 3) diastasis of coronal or sagittal suture; this findings can be confirmed with directional coronal CT.[1]

MRI scans are very useful in diagnosing VEDH, due to its multi-planar capabilities and lack of bone artifacts; however it is not used routinely because of the longer time taken and it costs a lot, even though it has its own advantages; for example in the bone window view it may unveil a fracture line and contrast-enhanced MRI may help the preoperative planning with detailed assessment of the relations with adjacent structures; yet, directional coronal CT is still the preferred radiological investigation in suspected cases of VEDH.[1] Recognizing the cranial sutural diastasis is also not easy. To date, there is no study providing normal limits of width of adult cranial sutures.[2]

The management of VEDH is better considered on a case to case basis.[1] Factors which determine the management of VEDH are 1) the size of the VEDH, 2) the rapidity of evolution of the VEDH, 3) the location of the hematoma, and 4) the clinical presentation of the patient.[1] Smaller hematomas are likely to resolve spontaneously, while rapidly evolving hematomas often than not are fatal; hence the need for immediate evacuation.

VEDH in the posterior is often more severe in symptoms and usually it require surgical evacuation. Common indications for immediate VEDH evacuation surgery are 1) deteriorating consciousness, 2) features of severly increased ICP, 3) features of focal neurological deficit, and 4) hematoma measuring more than 30 mL in volume.[1,11] This surgical management include wide craniotomy, extending across the midline to include the margins of the hematoma, evacuation of the hematoma, and controlling the source of the bleeding.

High suspicion, right diagnosis, close monitoring, and surgical intervention when required lead to a good outcome in VEDH.[1] The following should make one suspect the presence of VEDH: 1) direct impact on the vertex, 2) fracture line running across the vertex, 3) coronal or sagittal suture diastasis, 4) the patient presenting with features of increased ICP and or limb weakness.[1]

Conclusion

VEDH is a relatively uncommon type of posttraumatic intracranial hematoma, accounted for 1.3 to 8.2% of all traumatic intracranial hematomas.[1, 2, 5, 6] The clinical manifestations of VEDH is highly variable; they may present either in the immediate posttraumatic period, or later, with severe headache is considered to be major symptom of VEDH, and there seems to be a pattern where this headache gradually increases in its intensity, from mild to severe, caused by elevated ICP, therefore, clinical presentation of VEDH can be signs of elevated intracranial pressure.[2, 3, 8]

Acute VEDH might go overlooked on classic axial CT scan due to the blind spot, hence directional coronal CT and MRI are the preferred radiological investigation in suspected cases of VEDH.[1]

The management of VEDH is better considered on a case to case basis.[1] High suspicion, right diagnosis, close monitoring, and surgical intervention when required lead to a good outcome in VEDH.[1] The following should make one suspect the presence of VEDH: 1) direct impact on the vertex, 2) fracture line running across the vertex, 3) coronal or sagittal suture diastasis, 4) the patient presenting with features of increased ICP and or limb weakness.[1].

References

1. Ramesh V, Kodeeswaran M, Deiveegan K, Sundar V, Sriram K. Vertex epidural hematoma: An analysis of a large series. Asian Journal of Neurosurgery. 2017;12(02):167-171.
2. Klepinowski T, Kawalec P, Larysz M, Sagan L. Acute-on-Chronic Vertex Epidural Hematoma with Diastasis of the Sagittal Suture in an Adult. World Neurosurgery. 2020;139:245-249.
3. COLUMELLA F, DELZANNO GB, NICOLA GC. [Rare form of epidural hematoma: epidural hematoma at the vertex]. Sist Nerv. 1959 Mar-Apr;11(2):104-18. Italian. PubMed PMID: 13668671.
4. Matsumoto K, Akagi K, Abekura M, Tasaki O. Vertex epidural hematoma associated with traumatic arteriovenous fistula of the middle meningeal artery: a case report. Surg Neurol. 2001 May;55(5):302-4. PubMed PMID: 11516475.
5. Wylen EL, Nanda A. Vertex epidural hematoma with coronal suture diastasis presenting with paraplegia. J Trauma. 1998 Aug;45(2):413-5. PubMed PMID: 9715209.
6. Navarro JN, Alves RV. Vertex epidural hematoma: A rare cause of post-traumatic headache and a diagnostic challenge. Surg Neurol Int. 2016 May 6;7(Suppl 10):S276-

8. doi: 10.4103/2152-7806.181982. eCollection 2016. PubMed PMID: 27213114.

7. Borzone M, Gentile S, Perria C, Rivano C, Rosa M. Vertex epidural hematomas. Surg Neurol. 1979 Apr;11(4):277-84. PubMed PMID: 441913.
8. Miller DJ, Steinmetz M, McCutcheon IE. Vertex epidural hematoma: surgical versus conservative management: two case reports and review of the literature. Neurosurgery. 1999 Sep;45(3):621-4; discussion 624-5. PubMed PMID: 10493382.
9. Alexander GL. EXTRADURAL HAEMATOMA AT THE VERTEX. J Neurol Neurosurg Psychiatry. 1961 Nov;24(4):381-4. PubMed PMID: 21610904; PubMed Central PMCID: PMC495403.
10. Messori A, Pauri F, Rychlicki F, Veronesi V, Salvolini U. Acute posttraumatic paraplegia caused by epidural hematoma at the vertex. AJNR Am J Neuroradiol. 2001 Oct;22(9):1748-9. PubMed PMID: 11673172.
11. Bullock MR, Chesnut R, Ghajar J, Gordon D, Hartl R, Newell DW, et al. Surgical management of acute epidural hematomas. *Neurosurgery.* 2006;58:S7–15.

Romanian Neurosurgery (2023) XXXVII (3): pp. 288-291
DOI: 10.33962/roneuro-2023-052
www.journals.lapub.co.uk/index.php/roneurosurgery

Feasibility and safety of using the radial artery approach for the Wada test. A case report

Krunal Moradiya[1], Shahram Hadidchi[2], Parthasarathi Chamiraju[3]

[1] Michigan State University College of Osteopathic Medicine, East Lansing, MI, USA
[2] Department of Radiology, Detroit Medical Center, Detroit, MI, USA
[3] Department of Neurosurgery, Detroit Medical Center, Detroit,MI, USA

Keywords
Endovascular,
neurointervention,
transradial catheterization,
transradial neurointervention,
transradial Wada test

Corresponding author:
Krunal Moradiya

Michigan State University College of Osteopathic Medicine, East Lansing, MI, USA

moradiya@msu.edu

Scan to access the online version

Abstract

The Wada test is used to lateralize the dominant hemisphere in epilepsy patients prior to surgery. Either amobarbital, methohexital, or propofol is injected via a transfemoral catheter into a unilateral carotid artery, briefly suppressing the ipsilateral cerebral hemisphere to allow functional evaluation of the opposite side. Here, we highlight and propose a radial artery approach for the Wada test, allowing easier arterial access, better patient comfort, and ample procedural efficacy.

Introduction

The Wada test is conducted by a neurologist, neurointerventionalist, and neuropsychologist. Electroencephalogram (EEG) leads are placed on the patient for monitoring during the test. Preliminary cerebral angiogram is performed through transfemoral catheterization of the internal carotid artery (ICA), confirming adequate perfusion of a unilateral hemisphere. Afterwards, either amobarbital, methohexital or propofol is dispensed to suppress activity of the ipsilateral hemisphere [5]. Contralateral hemiparesis will then be observed, indicating that the medication has taken effect. Next, the neuropsychologist administers a series of speech, memory, and motor assessments to evaluate functionality of the contralateral hemisphere, which remains “awake”. After allowing the patient to recover, the entire procedure is repeated to evaluate the hemisphere not yet anesthetized. As language and memory are controlled by the dominant hemisphere, relative aphasia experienced during the test allows lateralization. Typically, the Wada test is conducted using a transfemoral artery approach (TFA). Here, we describe a case of the Wada test performed using a radial artery approach. The value of this approach in neurointervention will also be discussed.

CASE REPORT

A 63-year-old woman with intractable temporal lobe seizures was evaluated by the epileptologist and worked up for surgical treatment. The patient was admitted to the epilepsy monitoring unit to lateralize seizure location. Clinical semiology and electrographic findings suggested temporal lobe epilepsy.

Wada testing was requested as part of the pre-operative evaluation by the epileptologist. The patient was kept NPO for 12 hours. With the patient supine on the angiography table, the right wrist and hand were prepped and draped in the usual sterile fashion. 10 ml of 2% lidocaine was administered subcutaneously at the puncture site. 100 μg of nitroglycerin and 5 mg of verapamil were also provided. Under sonographic guidance, vascular access was obtained via a single-wall puncture of the right radial artery and a 5 F Pinnacle sheath (Terumo, Tokyo, JA) was introduced. 3,000 units of heparin were administered intravenously following sheath placement. A right radial artery angiogram was performed. The 0.038" guidewire and Simmons II diagnostic catheter were then advanced into the right ICA (RICA) using a Simmons curve in the aorta, and digital angiograms were obtained. Initially, 3 mg of methohexital were injected intra-arterially, followed by another 2 mg after a few minutes. Neuropsychological testing was performed before and after the second 2 mg injection.

After 10 minutes, the catheter was placed into the left ICA (LICA) via the Simmons curve and additional cerebral angiograms were taken (Figure 1). Neuropsychological testing was performed. 3 mg methohexital were then injected intra-arterially and more neuropsychological testing was performed. The patient remained under EEG surveillance throughout the entire procedure.

Each instance of neuropsychological testing involved assessment of the upper extremity motor function to confirm laterality. For the left side, laterality was evaluated by asking the patient to squeeze the epileptologist's hand and raise their own arm. For the right side, the neurointerventionalist checked the patient's force by asking her to squeeze his hand, which maintained hemostasis and kept the catheter insertion site safe. On the right side, the patient was not asked to raise her arm given the risk of catheter displacement.

Following completion of the entire procedure, heparin was not reversed with protamine. The sheath was removed, and hemostasis was obtained by application of a TR band (Terumo, Tokyo, JA), followed by manual compression. Total fluoroscopy time was 7.5 minutes, and total procedure time was around 90 minutes. The patient was found to have a right dominant hemisphere, determined to be the side of the epileptogenic zone.

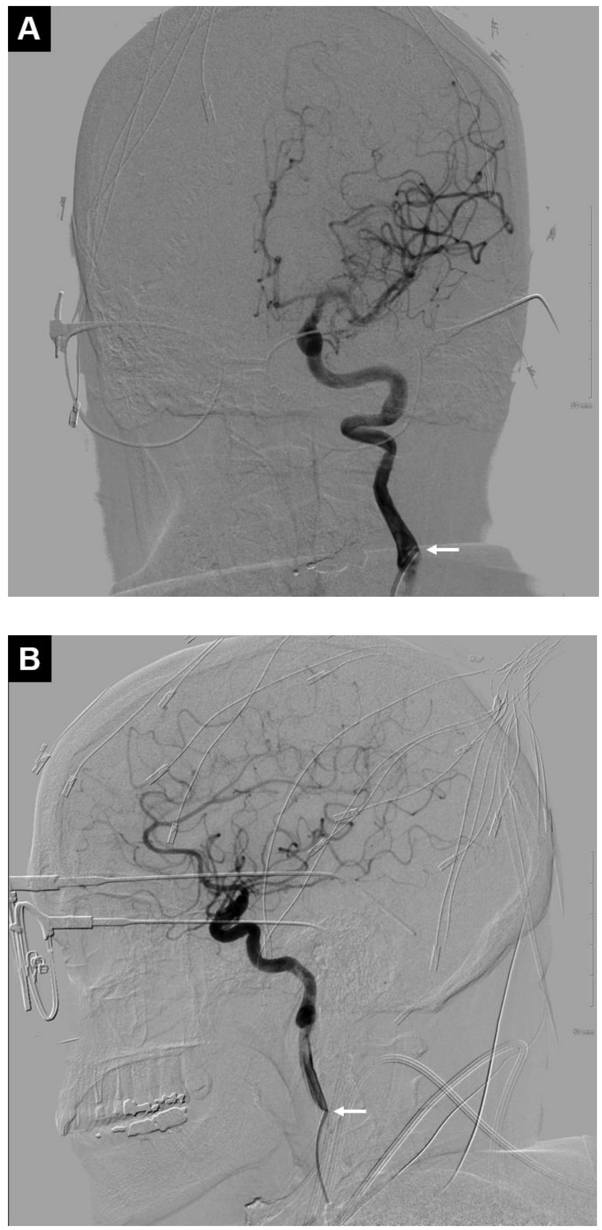


Figure 1. Frontal (A) and lateral (B) angiograms taken prior to methohexital administration, demonstrating patency of cranial vessels. The catheter tip (white arrows) is seen in the left internal carotid artery. Cranial EEG leads are best seen on lateral view. During the angiogram, the patient was wearing

glasses because she was asked to recognize objects held in front of her, in order to examine her speech and memory.

Discussion

In current practice, transfemoral catheterization of the ICA is the mainstay for most neurointerventional procedures, including the Wada test. TFA is preferred due to familiarity of the anatomy among neurointerventionalists and the large vessel diameter, allowing deployment of wider catheters [9]. However, TFA has several limitations, and a new approach has been described.

As an alternative to TFA, radial artery catheterization is viable for the Wada test. Among the cardiology literature, the transradial artery approach (TRA) has been used in coronary angiography and intervention for many years. Multicenter studies led by interventional cardiologists have shown that TRA can reduce vascular complications by at least 60% compared to TFA [1]. There are several factors that make TRA feasible. Firstly, the radial artery is situated away from crucial nerves, veins, and organs, which decreases the risk of access-site complications. Also, the superficial location of the radial artery permits better compression of bleeding for establishment of hemostasis. With TFA, patients may need to tolerate bed rest with uncomfortable groin compression for several hours if percutaneous closure devices are not used [8,9]. Hemostasis in TRA is more easily attainable, using manual compression or basic compression devices [4,6,8]. In our case, combined use of a TR Band and manual compression was sufficient for establishing hemostasis. By reaching rapid hemostasis, patients experience greater comfort and satisfaction due to the ability to ambulate more quickly [6]. Moreover, collateral circulation between the palmar branches of the radial and ulnar arteries lessens the risk of ischemic complications. A pre-procedural Allen's test can confirm the presence of collateral circulation [9]. By utilizing an end artery, TFA may increase the risk of ischemia.

Access-site hematomas are the most common minor complications of TRA, but they are typically inconsequential and easily treated [9]. Rather, the most salient complications of TRA are radial artery spasm (RAS) and radial artery occlusion (RAO). RAS occurs at rates anywhere between 14-30% [1,9] and is attributed to the high density of adrenoreceptors in the tunica media. Lidocaine and prophylactic antispasmolytics (i.e., verapamil and/or nitroglycerin) can mitigate RAS upon catheter introduction [1,9]. If RAS occurs, antispasmolytics can be administered until resolution is seen clinically and angiographically [7]. RAO is a post-procedural complication of TRA occurring at rates up to 33% [1,4,7,9]. RAO involves excess hemostatic compression and/or blood flow stagnation promoting transmural thrombus formation [4]. Although RAO is often asymptomatic due to the collateral blood supply, the radial artery could become precluded from future interventional use [1,4,9]. Radial patency can be improved periprocedurally through best practices like ulnar artery compression and prophylactic heparin [4,9]. For both RAS and RAO, ultrasound guidance can decrease the number of TRA puncture attempts, preventing arterial spasm and stenosis [1,6,9]. The wide variation in the incidence of RAS and RAO may be due to differences in study methodology, use of prophylaxis, diagnostic definition, and access-site techniques. With proper technique and prophylaxis, the risk of RAS and RAO can be tempered. With RAO specifically, the prevalence may be reduced to less than 1% of patients by using patent hemostatic techniques [7].

Apart from our case, TRA for the Wada test has not been described in the literature. With the use of appropriate medical prophylaxis and sonographic guidance, right radial access was achieved in our experience without complication of RAS or RAO. Additionally, our fluoroscopy time of 7.5 minutes is comparable to the range of 5.46+/-4.21 minutes found in other studies deploying TRA in cerebral angiography [8]. Fluoroscopy times can differ depending on the number and type of vessels catheterized using a right TRA, especially when targeting left-sided supra-aortic vessels. In using a right radial approach, the formation of a Simmons curve in the aorta is crucial for selective catheterization of the left subclavian artery, left vertebral artery, and LICA [7,9]. In our case, a Simmons curve was employed to catheterize the LICA during the Wada test. We acknowledge the inherent difficulty of selective LICA catheterization. In a series of 1,240 patients, one center reported a failure rate of 17.6% in attempted LICA catheterization using a right TRA. Tortuous angulature encountered during the catheterization attempt and the lack of dedicated catheter designs were cited as reasons for failure [3]. Tortuous angulature is seen in some patients with aberrant aortic arch anatomy and exacerbates the learning curve of TRA [7]. The formation of a Simmons curve in the aorta is often necessary for selective vessel catheterization and further increases the learning curve for inexperienced operators [7,9]. Indeed, the steep learning curve of TRA and lack of specialized catheter equipment are primary reasons for non-adoption of TRA among many neurointerventionalists [2]. However, the learning curve can be overcome as expertise is developed through procedural volume [2,7,9], resulting in decreased crossover rates to TFA and fluoroscopy times during angiography [7]. Currently, the success rate of TRA for cerebral angiography is around 92.7-99% [7,8].

Conclusion

We propose increased adoption of TRA for the Wada test. In recent times, TRA has been successfully used for cerebral angiography. Angiography is a primary segment of the Wada test, along with amobarbital/methohexital administration. At present, a 5 F Simmons II is a satisfactory choice for both segments of the test. As more specialized transradial kits and catheter designs are developed, TRA will only become more practical to perform. Given the straightforward nature of the Wada test, TRA is a feasible approach as it provides greater patient comfort and reduces access-site complications. Both novel and seasoned neurointerventionalists should make efforts to adopt TRA, especially for procedures like the Wada test.

Abbreviations

Electroencephalogram = EEG
Internal carotid artery = ICA
Left internal carotid artery = LICA
Nothing per os = NPO
Transfemoral artery approach = TFA
Transradial artery approach = TRA
Radial artery spasm = RAS
Radial artery occlusion = RAO

References

1. Brunet MC, Chen SH, Peterson EC. Transradial access for neurointerventions: management of access challenges and complications. J Neurointerv Surg. 2020;12(1):82-86.
2. Das S, Ramesh S, Velagapudi L, et al. Adoption of the Transradial Approach for Neurointerventions: A National Survey of Current Practitioners. J Stroke Cerebrovasc Dis. 2021;30(3):105589.
3. Jo KW, Park SM, Kim SD, Kim SR, Baik MW, Kim YW. Is Transradial Cerebral Angiography Feasible and Safe? A Single Center's Experience. J Korean Neurosurg Soc. 2010;47(5):332-337.
4. Pancholy SB, Shah S, Patel TM. Radial Artery Access, Hemostasis, and Radial Artery Occlusion. Interv Cardiol Clin. 2015;4(2):121-125.
5. Patel A, Wordell C, Szarlej D. Alternatives to sodium amobarbital in the Wada test. Ann Pharmacother. 2011;45(3):395-401.
6. Satti SR, Vance AZ. Radial Access for Neurovascular Procedures. Semin Intervent Radiol. 2020;37(2):182-191.
7. Snelling BM, Sur S, Shah SS, et al. Transradial cerebral angiography: techniques and outcomes. J Neurointerv Surg. 2018;10(9):874-881.
8. Wang Z, Xia J, Wang W, et al. Transradial versus transfemoral approach for cerebral angiography: A prospective comparison. J Interv Med. 2019;2(1):31-34.
9. Zalocar LAD, Doroszuk G, Goland J. Transradial approach and its variations for neurointerventional procedures: Literature review. Surg Neurol Int. 2020; 11:248..

Romanian Neurosurgery (2023) XXXVII (3): pp. 292-297
DOI: 10.33962/roneuro-2023-053
www.journals.lapub.co.uk/index.php/roneurosurgery

Key considerations for nutritional management in traumatic brain injury. A narrative review

A.C.C. Martínez[1], L.I.A. Ortiz Sierra[2], A.F.R. Galeano[3], D.A.B. Cundar[4], M.G.O. Sierra

[1] School of Medicine, Universidad de Cartagena, Cartagena, COLOMBIA
[2] School of Medicine, Fundación Universitaria San Martin, Bogotá, COLOMBIA
[3] School of Medicine, Universidad el Bosque, Bogotá, COLOMBIA
[4] School of Medicine, Universidad de Antioquia, Medellin, COLOMBIA
[5] Department of Neurosurgery, Universidad Centroccidental Lisandro Alvarado - Hospital Central Antonio María Pineda, Barquisimeto, VENEZUELA

Keywords
traumatic brain injury,
diet,
food,
nutrition,
neurosurgery,
nervous system diseases

Corresponding author:
Michael Gregorio Ortega Sierra

Universidad Centroccidental Lisandro Alvarado - Hospital Central Antonio María Pineda, Barquisimeto, Venezuela

mortegas2021@gmail.com

Scan to access the online version

ABSTRACT

Patients with traumatic brain injury experience complications and sequelae that lead to dysfunction and an uncertain prognosis. Brain injury induces a state of hypermetabolism and hypercatabolism, increasing the energy requirements of patients and predisposing them to malnutrition if appropriate nutritional support is not initiated early. To investigate this issue, we conducted a narrative review through December 2022. Poor nutrition in neurosurgical patients elevates mortality rates and significantly amplifies the risk of post-surgical complications. Recent studies have revealed that patients with traumatic brain injury experience an increase in metabolism of up to 250%, resulting in significant nitrogen loss. These patients should consume no less than 15% of total calories in the form of protein, with amino acid intake approaching 2 g/kg based on the patient's ideal weight being beneficial. Studies have compared the effectiveness of parenteral and enteral nutrition in these patients, with enteral nutrition providing superior benefits. Enteral nutrition is consistent with human physiology and supports a healthy gastrointestinal tract, modulates the immune system, and reduces the risk of liver cholestasis, which is higher in parenteral nutrition.

INTRODUCTION

Traumatic brain injury (TBI) creates specific nutritional requirements in patients that demand continuous medical monitoring [1]. Approximately 66% of neurosurgical patients experience weight loss during their hospitalization due to iatrogenic malnutrition caused by inadequate nutritional evaluation. Neurosurgical patients have unique metabolic needs because neurological injury triggers a state of hypercatabolism and hypermetabolism. Hypermetabolism accelerates the depletion of lipid and protein reserves, hindering the healing process and increasing the risk of infection [2]. Additionally, this state increases hormone levels, such as anti-diuretic hormone (ADH) and aldosterone [3], which can lead to malnutrition if not accompanied by an adequate nutritional supply [2]. The extent of these alterations varies depending on the injury's severity. Approximately 50% of post-surgical neurosurgery patients experience vomiting, nausea, bloating, and increased residual gastric volume, indicating that they do not tolerate enteral nutrition [3,4]. However, a combination of enteral and parental nutrition has been linked to several benefits, including attenuation of the hypercatabolic response, prevention of intestinal atrophy and muscle loss, and improved prognosis [2]. This study aims to describe the nutritional support utilized in neurosurgical patients.

THE METABOLIC PATTERN OF BRAIN INJURY

TBI can trigger an inflammatory response that leads to various physiological changes at the cardiovascular and neuroendocrine levels, including vasodilation, increased microvascular permeability, cellular activation, and coagulation activation [5]. These changes result in the release of hormones and neurotransmitters, increasing the need for fluids and oxygen consumption to cope with the induced stress. The stressor can be an injury, mechanical disorder, or chemical changes. The metabolic alteration varies depending on the severity and duration of the event, the type of damage, and the nutritional status of the patient before the injury [6].

In humans, the metabolic response following major injury or trauma is characterized by three phases: The Ebb phase, the Flow or catabolic phase, and the anabolic phase [7-9]. During the Ebb phase (within 24–48 hours), there is a decrease in metabolic rate, and the patient presents a clinical picture characterized by hypovolemia, hypotension, and tissue hypoxemia. In this phase, the patient is in a state of shock and requires resuscitation and life support measures [7-9]. The Flow phase is divided into an acute and an adaptive response. In the acute response, the body seeks hemodynamic stabilization, and in the adaptive response, there is an increase in energy expenditure, oxygen consumption, cardiac output, and a decrease in systemic vascular resistance. At this stage, it is essential to start nutritional support to take advantage of the increased metabolic rate [7-9].

Patients undergoing neurosurgery commonly exhibit a state of hypermetabolism and hypercatabolism, which does not necessarily correlate with the severity of the injury as measured by the Glasgow Coma Scale (GCS). Surprisingly, patients with lower degrees of coma (GCS, 4-5) often exhibit higher energy expenditure than those with higher levels of coma (GCS, 8-11), while patients with intermediate levels of coma (GCS, 6-7) demonstrate energy expenditure levels that fall between these extremes [10].

HYPERMETABOLISM

Patients with this type of pathology have an elevated resting energy expenditure and increased oxygen consumption. To prevent adverse events such as pain and seizures, sedation should be used to achieve a decrease in energy expenditure, which can increase by up to 140%. In sedated patients, energy consumption is typically around 25 Kcal/kg weight/day, while patients who are hypothermic and in a barbiturate-induced coma can maintain energy consumption in the range of 80% to 100%, requiring only an intake of 20 to 22 Kcal/kg weight/day [11].

HYPERCATABOLISM

During the acute phase response to injury, there is a significant release of amino acids from skeletal muscles and lipids, which leads to increased catabolism. Nitrogen loss in urine output is a manifestation of this hypercatabolism and becomes significant when it exceeds 0.2 grams per kg of body weight per day between the second and third week after injury. This increase in nitrogen loss is associated with a direct increase in morbidity and mortality in patients with inadequate nutrition. The increase in blood glucose levels is mediated by catecholamines and cortisol, as well as alterations in insulin utilization in the periphery. This leads to a shift from aerobic to anaerobic metabolism, in which the active metabolite lactate is created. Lactate is capable of inducing tissue acidosis, leading to cell destruction. However, there is no clear relationship between glycemic control and improvement in the clinical course of the injury [11].

MALNUTRITION

In neurosurgical patients, poor nutrition increases mortality and significantly raises the risk of post-surgical complications. In 1993, it was proposed that around 50% of mechanically ventilated patients have malnutrition problems. Therefore, it is essential to determine the specific energy requirement for each patient according to their metabolic condition. To calculate the energy intake, it is necessary to consider the body mass index. Patients exceeding 25 kg/m2 require 10% less energy intake than their actual weight, whereas those below 20 kg/m2 need an intake 10% higher than their real weight. Additionally, measuring body temperature is crucial since its increase is associated with higher energy requirements [1,4].

Recent studies have reported increased metabolism and nitrogen loss in neurosurgical patients, mainly in those who have suffered a TBI. In these patients, metabolism could increase up to 250%. For such patients, the protein intake should not be below 15% of the total calories consumed, and amino intake approaching 2 g/kg based on the ideal weight of the patient is beneficial [2,4].

GLYCEMIC CONTROL

Hyperglycemia is linked to a poor prognosis in patients with TBI. While strict glycemic control may result in severe reduction of glucose at the cerebral level, the aim is to manage hyperglycemia by monitoring brain glucose levels through microdialysis [12]. In neurosurgery, intraoperative glucose levels should be maintained below 180 mg/dl and above 140 mg/dl, and monitoring is essential during the procedure to prevent hyper- or hypoglycemic states that are associated with unfavorable outcomes [13]. There are two approaches for controlling intraoperative hyperglycemia: long-acting insulin with an insulin correction scheme or continuous infusion of intravenous insulin. Additionally, to achieve good glycemic control, factors such as the use of corticosteroids, steroids, and anesthesia should be considered since they can affect serum glucose levels [13].

ESTIMATED ENERGY NEED AND EVALUATION OF THE NUTRITIONAL STATE

Severe metabolic stress often affects individuals with severe acute illness or trauma. While such patients are typically admitted without a prior history of malnutrition, the massive inflammatory response observed under such conditions often limits the effectiveness of nutritional interventions and contributes to the rapid development of malnutrition. Additionally, periods of interrupted feeding, which are necessary to accommodate the various medical and surgical interventions required to stabilize these patients, also contribute to malnutrition, despite physicians' best efforts to provide adequate calories and other nutrients [14]. Thus, frequent and intensive monitoring of critically ill patients is required to determine the actual level of nutrients provided and ensure that the patient's needs are adequately addressed [15].

The brain primarily relies on glucose for energy, requiring approximately 25% of all available glucose in the body. Despite this high demand, glycogen stores in the central nervous system are limited, making neurons entirely dependent on constant, adequate cerebral blood flow. In a patient with brain trauma, determining the amount of energy and protein required is essential because it can reduce morbidity and mortality by nearly 50% and reduce the length of stay in the intensive care unit [3]. Calculating the ideal requirements for these patients is challenging due to variables such as weight, medication, and body temperature. Thus, formulas such as the Harris-Benedict and Penn State equations are used to estimate the energy requirement or an estimate of 20-30 Kcal/kg/day. However, it is believed that the most suitable method to determine energy needs is the indirect calorimeter, although there is insufficient evidence to support its use due to variation according to the patient's condition, such as the use of a mechanical ventilator, renal therapy, or movements during physical therapy [16].

ENTERAL AND PARENTERAL NUTRITION

Traumatic brain injury (TBI) induces a hypermetabolic and hypercatabolic state that increases energy needs and causes severe loss of protein mass, leading to acute malnutrition. This condition increases morbidity and mortality, worsens prognosis, and elevates the risk of complications such as infections, delayed healing, weak respiratory muscles, and prolonged ICU stay [11, 17].

Several studies have compared parenteral and enteral nutrition and established that enteral nutrition is more beneficial for TBI patients. Enteral nutrition is consistent with human physiology, promotes gastrointestinal tract health, modulates the immune system, and reduces the risk of liver cholestasis and epithelial damage compared to parenteral nutrition. The Society for Medicine and Critical Care (SCCM) and the American Society for Enteral and Parenteral Nutrition (ASPEN) recommend starting enteral nutrition within 24-48 hours of hemodynamic stabilization to preserve the intestinal epithelium and prevent enterobacteria from entering the bloodstream [18].

Härtl et al [17] studied the effects of early nutrition in the first two weeks after trauma and found that patients who did not receive nutrition during the first week had significantly higher mortality rates. This correlation was directly related to the daily caloric loss experienced by patients. Even when controlling for parameters such as hypotension or increasing the Glasgow Coma Scale (GCS), the probability of death remained high if early nutrition was not provided.

Enteral nutrition provides several benefits, including improving gastrointestinal function, which can still be compromised in patients receiving parenteral nutrition [19,20]. In a study conducted on rats with brain trauma, the timing and composition of enteral nutrition were found to be important factors in preserving the intestinal mucosa, with early nutrition and the addition of immune nutrients being particularly beneficial [11].

The administration route for enteral nutrition is chosen based on criteria such as timing, delayed gastric emptying, and the risk of aspiration. The preferred routes are gastric and jejunal, with gastric administration being more physiological but also more prone to aspiration and nosocomial pneumonia, while jejunal administration allows for earlier initiation and greater caloric intake but may inhibit intestinal motility [21-26]. Additionally, the type of enteral formula and specific nutrients such as glutamine must be considered when selecting the appropriate supplementation [11].

SPECIALIZED NUTRITIONAL SUPPORT

Individualized metabolic management is crucial in compensating for the hypermetabolic state of patients through specialized nutritional support. Carbohydrates should contribute most of the energy intake, with a maximum dose of 4-6 g/kg weight/day. Glycemic monitoring and insulin therapy are essential to maintain levels lower than 110 mg/dl. The lipid intake should represent only 20-30% of the total caloric intake at 1-1.5 g/kg/day, while high protein intake is necessary, with a minimum of 1.5g protein/kg body weight/day. However, establishing the optimal amount of protein required to establish nitrogen balance in these patients is challenging [5]. The problem of iatrogenic malnutrition in neurosurgical patients is underestimated, and the relationship between nutritional status and outcomes has gained more attention recently. Adequate nutrition and appropriate metabolic care are essential for critically injured patients, including neurosurgical patients. It is crucial to recognize nutritional deficiency in a timely manner by assessing nutritional status through various means [27-30].

OTHER CONSIDERATIONS

Neurosurgical patients often require non-oral nutritional support due to altered mental status, intubation, or dysphagia. Nasogastric feeding is often preferred over total parenteral nutrition due to its lower cost and complication rate, as well as its demonstrated ability to deliver adequate calories and amino acids via the enteral route [27,28,30]. Malnutrition has been found to have a significant impact on neurological outcomes, with studies suggesting that undernourishment is associated with longer hospital stays and higher rates and severity of infections [31-34]. Early feeding has been linked to lower infection rates and improved outcomes in terms of survival and disability, highlighting the importance of early nutritional intervention for malnourished patients [5]. Critically ill neurosurgical patients with malnutrition are also at a higher risk for postoperative complications, making timely assessment and intervention crucial for reducing morbidity and mortality [27,28,35,36]. In a retrospective study of patients with intracranial hemorrhage (ICH), an increase in enteral nutrition caloric intake was associated with a favorable Glasgow Coma Scale (GCS) score at discharge (≤ 25 vs 25 Kcal/kg/48 hrs), underscoring the importance of appropriate caloric intake in improving outcomes [12,37].

SPECIAL CONSIDERATIONS IN TRAUMATIC BRAIN INJURY

Nutritional therapy is critical for traumatic brain injury patients due to widespread metabolic, immune, gastrointestinal, and neurological dysfunction following injury [27,28,30]. Studies show that energy requirements increase substantially following head injury [5]. The acute metabolic response in TBI involves the release of inflammatory cytokines, stress hormones, and other mediators, leading to a serious catabolic state that can deplete energy stores and body mass [1,2,3]. The energy expenditure in TBI is influenced by various factors, such as neurologic status, intracranial pressure, medical or surgical therapy, and the presence of infection or renal failure [28,30]. Without nutritional intervention, hypercatabolism can lead to a loss of 10-15% in one week and over 30% in 2-3 weeks, with villous atrophy, epithelial cell reduction, and edema in villous interstitial tissue [2,3,4].

However, there are several challenges to advancing our understanding of traumatic brain injury, designing public policy, and producing high-quality evidence [38,39]. Efforts must continue to address this disease's burden in low- and middle-income countries, where there are no technological tools to control morbidity, mortality, and disability [40,41].

Conclusions

Establishing an early and adequate nutritional therapy is crucial for improving the general condition of patients with traumatic brain injury who require continuous monitoring. Their prognosis is directly dependent on the hypermetabolic state that results in malnutrition and leads to various complications, prolonged stay in the intensive care unit, and increased risk of mortality. Providing the patients with the necessary caloric and protein requirements through proper nutritional support can significantly improve their outcomes.

References

1. Annette H, Wenström Y. Implementing clinical guidelines for nutrition in a neurosurgical intensive care unit. Nursing & health sciences. 2005; 7(4):266-272.
2. Riboldi BP, Contini B, dos Santos FT, et al. Nutrição e neurocirurgia: uma revisão. Arquivos Brasileiros de Neurocirurgia: Brazilian Neurosurgery. 2011;30(02): 55-59.
3. Abdelmalik PA, Dempsey S, Ziai W. Nutritional and Bioenergetic Considerations in Critically Ill Patients with Acute Neurological Injury. Neurocritical care. 2017;27(2):276-286.
4. Oertel MF, Hauenschild A, Gruenschlaeger J, Mueller B, Scharbrodt W, Boeker DK. Parenteral and enteral nutrition in the management of neurosurgical patients in the intensive care unit. J Clin Neurosci. 2009; 16(9):1161-7
5. García De Lorenzo y Mateos A, Rodríguez Montes JA. Head trauma and nutritional management of the neurological patient in a critical status. Nutrición Hospitalaria. 2009; 2(2):106-113.
6. McClave SA, Taylor BE, Martindale RG, et al. Guidelines for the Provision and Assessment of Nutrition Support Therapy in the Adult Critically Ill Patient: Society of Critical Care Medicine (SCCM) and American Society for Parenteral and Enteral Nutrition (A.S.P.E.N.). JPEN J Parenter Enteral Nutr. 2016; 40(2):159-211.
7. Walrand S, Gaulmin R, Aubin R, Sapin V, Coste A, Abbot M. Nutritional factors in sport-related concussion. Neurochirurgie. 2021; 67(3):255-258.
8. Brunicardi F, Andersen D, Billiar T, et al. Schwartz's principles of surgery, 10e: McGraw-hill; 2014.
9. McHoney M, Eaton S, Pierro A. Metabolic response to surgery in infants and children. Eur J Pediatr Surg. 2009; 19(5):275-85.
10. Foley N, Marshall S, Pikul J, Salter K, Teasell R. Hypermetabolism following moderate to severe traumatic acute brain injury: a systematic review. J Neurotrauma. 2008; 25(12):1415-31.
11. Ponce y Ponce de León G, Mayagoitia Witrón JJ, Cornejo Bravo JM, Pérez Morales ME. Enteral nutrition in patients with traumatic head injury: systematic review of clinical trials. Revista Iberoamericana de las Ciencias de la Salud. 2016; 4(7):1-4.
12. Menon DK, Ercole A. Critical care management of traumatic brain injury. Handbook of clinical neurology. 2017; 140:239-274.
13. Daniel R, Villuri S, Furlong K. Management of hyperglycemia in the neurosurgery patient. Hospital practice. 2017; 45(4):150-157.
14. White JV, Guenter P, Jensen G, Malone A, Schofield M. Consensus statement: Academy of Nutrition and Dietetics and American Society for Parenteral and Enteral Nutrition: characteristics recommended for the identification and documentation of adult malnutrition (undernutrition). JPEN Journal of parenteral and enteral nutrition. 2012; 36(3):275-283.
15. Jensen GL, Wheeler D. A new approach to defining and diagnosing malnutrition in adult critical illness. Current opinion in critical care. 2012; 18(2):206-211.
16. Chapple LS, Deane AM, Heyland DK, et al. Energy and protein deficits throughout hospitalization in patients admitted with a traumatic brain injury. Clinical nutrition (Edinburgh, Scotland). 2016; 35(6):1315-1322.
17. Härtl R, Gerber LM, Ni Q, Ghajar J. Effect of early nutrition on deaths due to severe traumatic brain injury. Journal of neurosurgery. 2008; 109(1):50-56.
18. Reintam Blaser A, Starkopf J, Alhazzani W, et al. Early enteral nutrition in critically ill patients: ESICM clinical

practice guidelines. Intensive care medicine. 2017; 43(3):380-398.

19. Rodriguez-Zapata MF, Valdes-Moreno PL, Baldosea-Mena Y, Lozada-Martinez ID. Regarding: Nutritional factors in sport-related concussion. Neurochirurgie. 2021: S0028-3770(21)00164-8.
20. Ortega-Sierra MG, Durán-Daza RM, Carrera-Patiño SA, Rojas-Nuñez AX, Charry-Caicedo JI, Lozada-Martínez ID. Neuroeducation and neurorehabilitation in the neurosurgical patient: programs to be developed in Latin America and the Caribbean. J Neurosurg Sci. 2021. Online ahead of print
21. Black EK, Phillips JK, Seminetta J, Bailes J, Lee JM, Finan JD. The effect of dietary supplementation with high- or low-dose omega-3 fatty acid on inflammatory pathology after traumatic brain injury in rats. Transl Neurosci. 2021; 12(1):76-82.
22. Yang J, Wang K, Hu T, Wang G, Wang W, Zhang J. Vitamin D3 Supplement Attenuates Blood-Brain Barrier Disruption and Cognitive Impairments in a Rat Model of Traumatic Brain Injury. Neuromolecular Med. 2021. Online ahead of print
23. Xiong W, Qian K. Low-Protein, Hypocaloric Nutrition with Glutamine versus Full-Feeding in the Acute Phase in ICU Patients with Severe Traumatic Brain Injury. Neuropsychiatr Dis Treat. 2021; 17:703-710.
24. Chen X, Wu S, Chen C, Xie B, Fang Z, Hu W, et al. Omega-3 polyunsaturated fatty acid supplementation attenuates microglial-induced inflammation by inhibiting the HMGB1/TLR4/NF-κB pathway following experimental traumatic brain injury. J Neuroinflammation. 2017; 14(1):143.
25. Lewis SR, Schofield-Robinson OJ, Alderson P, Smith AF. Enteral versus parenteral nutrition and enteral versus a combination of enteral and parenteral nutrition for adults in the intensive care unit. Cochrane Database Syst Rev. 2018; 6(6):CD012276.
26. Marik PE, Hooper MH. Normocaloric versus hypocaloric feeding on the outcomes of ICU patients: a systematic review and meta-analysis. Intensive Care Med. 2016; 42(3):316-323.
27. Institute of Medicine (US) Committee on Nutrition, Trauma, and the Brain. Nutrition and Traumatic Brain Injury: Improving Acute and Subacute Health Outcomes in Military Personnel. Erdman J, Oria M, Pillsbury L, editors. Washington (DC): National Academies Press (US); 2011.
28. DePalma RG. Combat TBI: History, Epidemiology, and Injury Modes. In: Kobeissy FH, editor. Brain Neurotrauma: Molecular, Neuropsychological, and Rehabilitation Aspects. Boca Raton (FL): CRC Press/Taylor & Francis; 2015. Chapter 2.
29. Kurtz P, Rocha E. Nutrition Therapy, Glucose Control, and Brain Metabolism in Traumatic Brain Injury: A Multimodal Monitoring Approach. Front Neurosci. 2020; 14:190.
30. Adesanya A, Sullivan R, Stawicki S, Evans D. Nutrition in Traumatic Brain Injury: Focus on the Immune Modulating Supplements. 2014.
31. Chacón-Aponte AA, Durán-Vargas ÉA, Arévalo-Carrillo JA, Lozada-Martínez ID, Bolaño-Romero MP, Moscote-Salazar LR, et al. Brain-lung interaction: a vicious cycle in traumatic brain injury. Acute Crit Care. 2022; 37(1):35-44.
32. Acevedo-Aguilar L, Gaitán-Herrera G, Reina-Rivero R, Lozada-Martínez ID, Bohorquez-Caballero A, Paéz-Escallón N, et al. Pulmonary injury as a predictor of cerebral hypoxia in traumatic brain injury: from physiology to physiopathology. J Neurosurg Sci. 2021; online ahead of print.
33. Lozada-Martinez ID, Ortega-Sierra MG, Daza-Vega MM, Cárdenas-Hormiga LN, Contreras-Saldarriaga JE, Garcia-Moron CE, et al. Basic considerations on growth hormone deficiency in traumatic brain injury. Romanian Neurosurgery. 2021; 35(4):483-9.
34. Reina-Rivero R, Rodriguez-Gutierrez MM, Ortega Sierra MG, Jamioy-Cabrera DF, Villegas-Lora JF, Caycedo-Montiel DM, et al. Multimodal neurological monitoring in neurotrauma: Theoretical considerations for a practical approach. Romanian Neurosurgery. 2021; 35(4):531-7.
35. Lozada-Martínez I, Galeano-Buelvas A, Pearson-Arrieta A, Diaz-Castillo O, Ortega-Sierra M, Robledo-Arias J, et al. Talk and Die Syndrome: A comprehensive review. Romanian Neurosurgery. 2021; 35(3):262-9.
36. Lozada Marintez ID, Ortega-Sierra MG, Zambrano-Arenas MDDP, Páez-Escallón N, Perea-Martinez DG, Caldera-Arrautt YS, et al. Essentials of cerebral fat embolism syndrome: A hidden enemy in trauma. Romanian Neurosurgery. 2021; 35(3):316-20.
37. Mass-Ramirez SM, Lozada Marintez ID, Bohorquez-Rivero J, Garcia-Ballestas E, Bustos D, Poveda-Castillo IT, et al. Gastroparesis as a complication in the patient with traumatic brain injury. Romanian Neurosurgery. 2021; 35(3):345-51.
38. Ortega-Sierra MG, Durán-Daza RM, Carrera-Patiño SA, Rojas-Nuñez AX, Charry-Caicedo JI, Lozada-Martínez ID. Neuroeducation and neurorehabilitation in the neurosurgical patient: programs to be developed in Latin America and the Caribbean. J Neurosurg Sci. 2021. Online ahead of print
39. Silva-Rued ML, Ramírez-Romero A, Guerra-Maestre LR, Forero-Hollmann ÁM, Lozada-Martínez ID. The need to develop specialized surgical centers: the evidence that surgical diseases cannot wait. Int J Surg. 2021; 92:106036.
40. Pérez-Fontalvo NM, De Arco-Aragón MA, Jimenez-García JDC, Lozada-Martinez ID. Molecular and computational research in low- and middle-income countries: Development is close at hand. J Taibah Univ Med Sci. 2021; 16(6):948-949.
41. ozada-Martinez ID, Ortega-Sierra MG, Hernández-Morales EY, Gonzales-Méndez R, Mejias-Riverón K, Argüello-Manrique M, et al. Letter to the Editor. Latin American collaboration in international neurotrauma registries: a strategy from the academy. J Neurosurg. 2022; online ahead of print.

Romanian Neurosurgery (2023) XXXVII (3): pp. 298-300
DOI: 10.33962/roneuro-2023-054
www.journals.lapub.co.uk/index.php/roneurosurgery

Orbital lipoma. A rare case report from India with review of literature

Nabajyoti Borah, Inamul Haque, Navanil Barua, Ananya Barman, Sneha Gang, Shabnam Akhtar Ahmed

GNRC Hospitals, Dispur-781006, Assam, INDIA

ABSTRACT

Orbital lipomas are uncommon benign tumors with only a few reported cases in literature. They are usually asymptomatic and exhibit gradual growth. We rendered such a rare case of primary lipoma in the orbital region with interpreted symptoms of ocular pain, reduced vision, and mild proptosis. Right fronto-orbital craniotomy with gross total excision of the tumor was done. Histological examination suggested a final diagnosis of lipoma. Post-operatively, the patient developed mild ptosis. During follow-up, ptosis completely subsided and significant improvement in vision was noted.

Keywords
craniotomy,
orbital lipoma,
surgical excision

Corresponding author:
Nabajyoti Borah

Department of Neurosurgery, GNRC Hospitals, Dispur, Assam, India

nabajyoti.borah@gnrchospitals.com

Scan to access the online version

INTRODUCTION

Lipomas are typical mesenchymal neoplasms which mostly occur in the subcutaneous tissues in the neck, shoulder, and back and are usually seen in persons of 40 years and above (1). Even though there is presence of abundant fat in the periorbital and the retrobulbar space, however, lipomas in the orbital region are still extremely rare (2). The incidence of primary orbital lipomas was reported to be ~ 0.6 % in a pooled orbital tumor series (2). These rare benign neoplasms typically appear as well-circumscribed masses on computed tomography (CT) and magnetic resonance imaging (MRI) (3). On conventional histology, the orbital lipomas closely resemble normal orbital fat thereby making their pathological entity doubtful (4). Rather than infiltration, the lipomas in the orbit, however, displaces the surrounding tissue and often leads to exopthalmos (5). Here in, we present a rare case of orbital lipoma diagnosed and treated in our hospital.

CASE REPORT

A 51 year old female patient presented in our Neurosurgery OPD with a history of right-sided ocular pain and reduced vision for six months. On eye examination, mild proptosis was present with visual acuity of R 6/24 and L 6/6. The extraocular movements were normal and full in every direction. She was investigated with MRI Brain and both orbits (P + C) with MR-angiography which showed right sided retrobulbar homogenously enhancing ovoid mass lesion in the intraconal compartment extending into orbital apex region and causing mass effect in optic nerve and adjacent extra ocular muscles (Fig. 1). Surgical intervention was planned after all necessary routine investigation and pre-anesthetic check-up. Right fronto-orbital craniotomy was done in two pieces. Extended removal of the roof and the lateral wall of the orbit were further done with the help of rongeur. Periorbita opened in a cruciat manner and extended with the help of a rongeur carefully without exerting much pressure on the intraorbital contents.

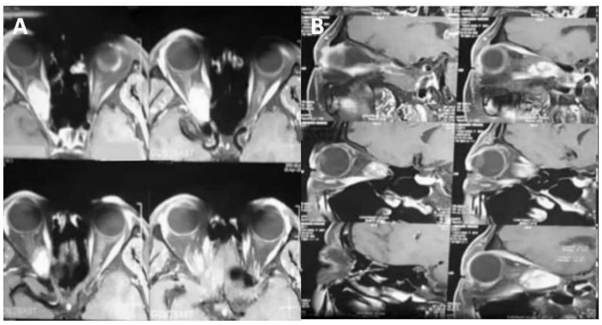


Figure 1. MRI images showed right sided orbital tumour.

Figure 2. Intra-operative image following right fronto-orbital craniectomy.

The dissection started medial to levator palpebrae superioris muscle and in between the superior rectus and medial rectus muscle. Tumor was localized super medial to the optic nerve. Internal decompressor followed by extra capsular dissection done. Tumor was found to be soft, avascular, and capsulated (Fig. 2). Total excision of the tumor was performed and homeostasis achieved (Fig. 3). Periorbital repaired with 4-0 vinyl sutures. Orbital root was reconstructed and frontal bone flap replaced. Wound was closed in layers with a subgaleal drain in situ. Post-operative recovery was uneventful. The patient developed a mild ptosis in the right eye. She was discharged in a stable condition on the 6th post operative day. Histopathological examination of the tumor tissue was suggestive of lipoma (Fig. 4). On follow up visit after two months, the patient recovered from the ptosis and showed significant improvement in vision.

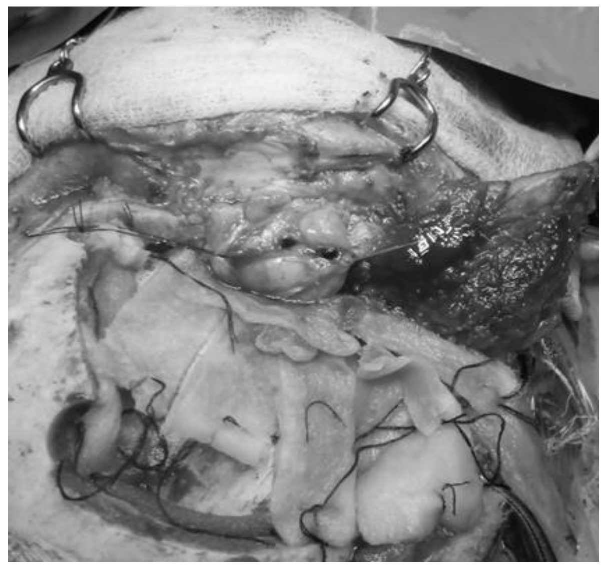


Figure 3. Intra-operative image showed tumor cavity following total excision.

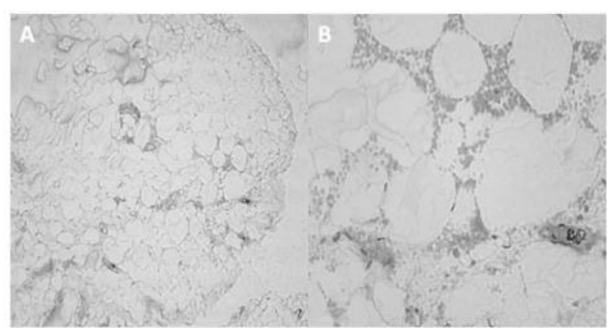


Figure 4. A) Histopathological examination of the lesion (H&E; 100X) that showed mature fat cells (adipocytes) surrounded by connective tissue. **B)** Histopathological examination of the lesion (H&E; 400X) that showed blood vessels and inflammatory cells.

Discussion

Lipomas are encapsulated benign tumors of fat cells (adipocytes) separated by fibrous septa6. They most commonly occur in the subcutaneous tissue while lipomas in the orbit are rarely found in spite of presence of abundant lipomatous tissue (2, 7). A pooled incidence of 0.6 % has been reported in literature in a large orbital lipoma tumor series (2). However, as lipomas are very much similar to normal orbital fat, therefore, many such cases may have been misrepresented in the pre-imaging era (2). So far orbital lipoma incidence has not been reported from India. As such this is the first reported case of true orbital lipoma from this country.

Orbital lipomas are well-circumscribed masses that originate from the anterior orbit (2, 5). They may be clinically perceptible before the onset of proptosis (2, 5). In general, they show no symptoms until the tumor becomes enlarged (2, 5). In certain rare cases, orbital lipomas outgrows and compresses the optic nerve which in turn causes disturbances in ocular functions such as decreased vision, visual field defects, and relative afferent pupillary defect (8). Prolonged pressure can also result in displacement of orbital structures. Previous studies have reported about four well-documented cases of conventional orbital lipoma within the age range of 11 to 72 years and comprising of two males and two females (4, 9, 10, 11). All four patients exhibited gradual but persistent swelling of the eyes or proptosis. In our case, presented symptoms were ocular pain, decreased visual acuity, and mild proptosis.

On CT scans, conventional orbital lipomas are seen as distinct low attenuation lesions of varying densities with the occasional presence of a finely defined border (11, 12) They do not enhance in imaging upon administration of contrast agents. On MRI, typically lipomas are hyperintense on T1 weighted images and hypointense after fat suppression (4). They are normally indistinct on T2 weighted images (4). In our case, T1 and T2 weighted MRI images showed mixed signal intensities (hypo and hyper intense) lesion and post contrast T1 with fat suppression. MRI images showed enhancing lesion which was suggestive of possible atypical lipoma. Histologically, true lipomas consist of a fine capsule encompassing lobulated fatty tissue (13). Our histological examination of the tumor gave a final diagnosis of lipoma.

Gross total surgical excision is the preferred treatment of choice for conventional orbital lipoma (2). Surgical intervention gives very good long term outcome (2). It also rules out the possibility of malignancy, establishes histological diagnosis, and provides relief from any persistent symptoms (5, 14). In our case too, the lipoma was surgically excised not only for histological diagnosis but also to obtain better outcome. The result was obvious during the follow up visit where the patient showed significant improvement in vision in the right eye.

Conclusions

Orbital lipomas are extremely rare benign neoplasms. There are very few reported cases in literature. They appear as distinctive well-circumscribed mass lesions. In general, they are asymptomatic and demonstrate chronic and gradual onset of growth with dislocation of orbital structures. Surgical excision is the preferred approach for the management of these lesions for better prognosis.

References

1. Stock N. Adipocytic tumors. Ann Pathol 2015;35(1):41-53.
2. Shah NB, Chang WY, White VA, et al. Orbital lipoma: 2 cases and review of literature. Ophthal Plast Reconstr Surg 2007;23(3):202-205.
3. Chorost MS, Yakopson V, Chou E. Posttraumatic encapsulated orbital lipoma. Mil Med 2020;185(9-10):e1876-e1879.
4. Behrendt S, Werner JA, Jänig U. Lipoma of the orbit. Orbit 1996;15(2):101-104.
5. Shrestha P, Shrestha GB. Orbital lipoma as an uncommon cause of unilateral proptosis: a case report. Int Med Case Rep J 2020;415-418.
6. Jones IS, Reese A, Kraut J. Orbital Rhabdomyosarcoma: an Analysis of 62 Cases. Am J Ophthalmol 1966;61(4):721-736.
7. Ali SF, Farber M, Meyer DR et al. Fibrolipoma of the orbit. Ophthal Plast Reconstr Surg 2013; 29(3):e79-e81.
8. Chi MJ, Roh JH, Lee JH, et al. A case of orbital lipoma with exophthalmos and visual disturbance. Jpn J Ophthalmol 2009; 53(4):442-444.
9. Koganei Y, Ishikawa S, Abe K, et al. Orbital lipoma. Ann Plast Surg 1988; 20(2):173-182.
10. Poweleit AC. Congenital lipoma of the orbit. J Ky Med Assoc 1978;76(1):20.
11. Brown HH, Kersten RC, Kulwin DR. Lipomatous hamartoma of the orbit. Arch Ophthalmol 1991;240-243.
12. Mawn LA, Jordan DR, Olberg B. Spindle-cell lipoma of the preseptal eyelid. Ophthal Plast Reconstr Surg 1998;14(3):174-177.
13. Weiss SW, Goldblum JR. Benign lipomatous tumors. In: Weiss SW, Goldblum JR, eds. Enzinger and Weiss's Soft Tissue Tumors. 4th ed. St. Louis: Mosby 2001;574-581.
14. Reibel JF, Greene WM. Liposarcoma arising in the pharynx nine years after fibrolipoma excision. Otolaryngol Head Neck Surg 1995;112(4):599-602.

Romanian Neurosurgery (2023) XXXVII (3): pp. 301-304
DOI: 10.33962/roneuro-2023-055
www.journals.lapub.co.uk/index.php/roneurosurgery

A rare case of primary orbital Ewings sarcoma with intracranial extension

Tamajyoti Ghosh[1], Binoy Kumar Singh[2], Aishik Mukherjee[1], Biswajit Dey[3]

[1] Department of Neurosurgery, NEIGRIHMS, INDIA
[2] Associate Professor, Department of Neurosurgery, AIIMS Raipur, INDIA
[3] Department of Pathology, NEIGRIHMS, INDIA

Keywords
non-axial proptosis,
small round blue cell tumours,
skeletal metastases,
extraosseous Ewing's sarcoma,
orbital decompression surgery

Corresponding author:
Tamajyoti Ghosh

Department of Neurosurgery,
NEIGRIHMS, India

tamajyoti@gmail.com

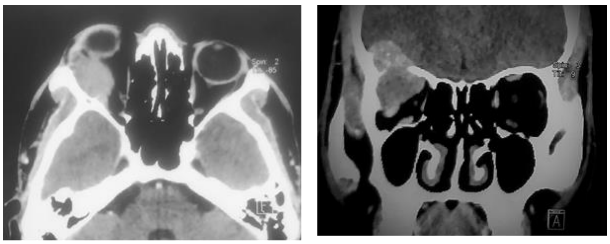
Scan to access the online version

ABSTRACT
Primary Ewings sarcomas of orbit with intracranial extension are extremely rare with few cases reported to date. They form an important differential diagnosis in aggressive orbital tumours presenting in young males. We presented the case report of a young male presenting with right eye proptosis. The patient underwent complete tumour excision. Biopsy was suggestive of Ewings sarcoma. Metastatic workup showed extensive lesions in lungs and bones. Six months post-operative follow-up with radiation and chemotherapy showed disease remission. CD99 is a key IHC marker in differentiating from other similar tumours. Total tumour excision with adjuvant radiation and chemotherapy may be helpful in survivability in such patients but overall prognosis still remains poor.

INTRODUCTION
Ewings sarcoma is a malignant tumour of bone with mean age of presentation of 10-14 years in male and 5- 9 years in female [1]. Ewings sarcoma is commonly seen in lower extremities in 35% , chest (21%), pelvis (17%), spine (10%), upper extremity (9%), head and neck (6%) and abdomen in 2% cases [2]. Rare case reports of Primary Ewings sarcoma of orbit are present [3]. Genetically Ewings sarcoma is due to cytogenetic translocation , t(11;22) (q24;q12) [4].

CASE PRESENTATION
Here we present the case report of 29 year old male who presented with history of proptosis of right eye for last 1 year. There is also associated history of double vision of lateral and medial gaze. On examination his right eye was non axially proptosed medially and downward. There was restriction of movement in eye in lateral, medial and upward gaze. His visual acquity of Right eye was 6/12 and left eye was 6/6. However there was no cranial nerve palsy on examination. His vitals were stable and no abnormality detected on rest of his systemic examination. Opthalmology consultation was taken and fundus examination was normal.

Figure 1. Severe downward proptosis right eye.



Caption Caption

Figure 2. Contrast MRI showing enhancing Right orbital lesion with intracranial extension.

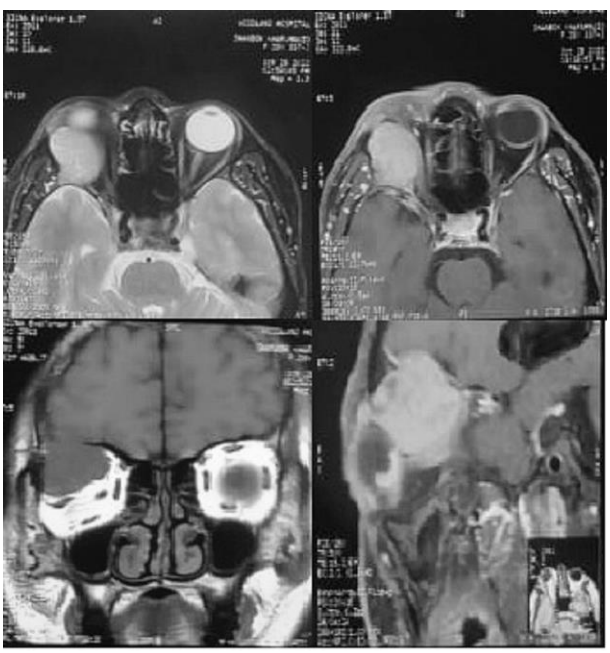


Figure 3. CT scan brain showing the Right frontal bone erosion and intracranial extension.

We requested urgent magnetic resonance imaging of his brain and orbit which was suggestive of T1 hyperintesnse and T2 isointense homogenous lesion in the superolateral extraconal compartment of right orbit measuring 3.4X 2.6X 3.47 in dimensions. There was extension of the lesion through roof of orbit to Anterior cranial fossa. Based on MR findings a provisional diagnosis of high grade lacrimal tumour was made.

Figure 4. Showing complete resolution of proptosis and opthalmoparesis postoperatively.

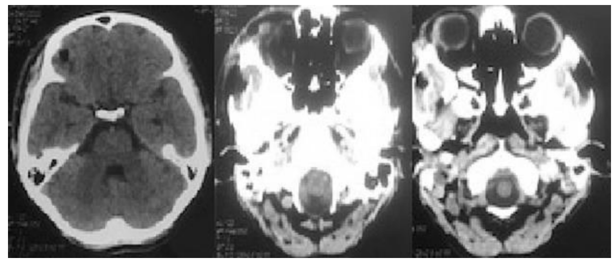


Figure 5. Post operative CT scan showing complete excision of tumour along with intracranial extension.

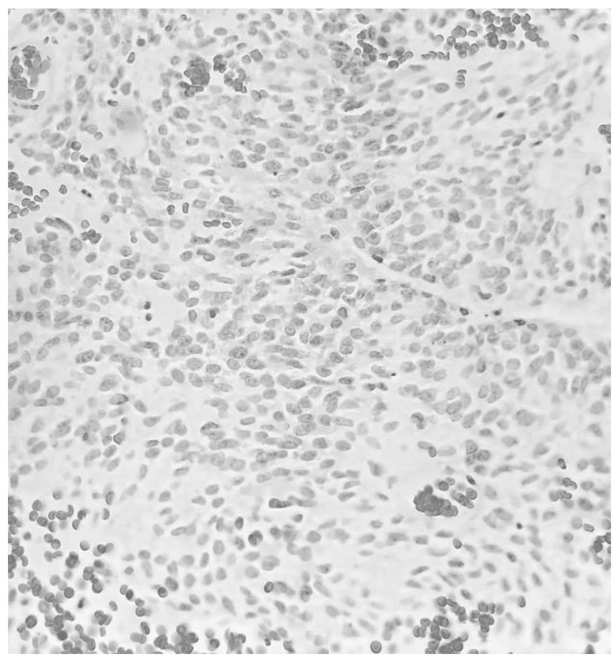


Figure 6. Histiopathology showing small round cells arranged in sheets (H&E stain 400X).

Rest of his blood investigations were unremarkable. He underwent Right lateral orbitotomy and tumour excision. Intraoperatively tumour was soft, vascular, pinkish in colour and suckable. Erosion and extracranial extension was seen in right orbital roof and gross total excision of tumour was done. Post operatively patient recovered well. His vision in right eye improved to 6/6 and opthalmoparesis resolved completely.

Tumour histiopathology report was suggestive poorly differentiated small round cell tumour. IHC showed strong positivity towards CD 99 and a final diagnosis of Ewing sarcoma was made.

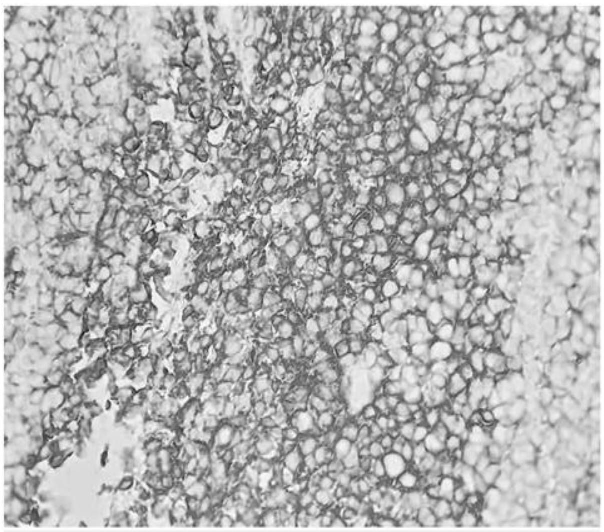


Figure 7. Tumour cells showing strong membranous positivity for CD99 (IHC 400X).

Patient underwent complete metatstatic workup and multiple lesions were detected in bilateral lung and lytic lesions in multiple ribs and pelvis. Patient underwent radiotherapy followed by chemotherapy with vincristine , adriamycin and cyclophosphamide alternating with etoposide and ifosamide . Patient has been on followup for last 6 months and is doing well.

Discussion

Primary orbital Ewing with intracranial extension is extremely rare and only few case reports are present [5, 6]. Orbital Ewings sarcoma most commonly occurs in young male similar to our case [7]. In our case tumour was located in superolateral quadrant of orbit which confers with the most common location in other reported cases [8, 9]. Patient usually presents with proptosis and the median duration of presentation is 9 weeks [7], however our patient presented to us at an advanced stage of disease with distant metastasis. Metastasis from primary orbital Ewings is seen in 20-25% cases [1]. Our patient underwent surgical excision followed by radiotherapy and chemotherapy according to current treatment strategy for Ewings Sarcoma [1]. CD99 is the key IHC marker which is invariably positive in Ewings sarcoma and in our case helped to differentiate it from Neuroblastoma [10]. Orbital Ewings is considered to be less aggressive than Ewings at other location [10] and patient usually have survival more than 6 months in most of the cases. Similar findings were seen in our case where our patient is doing well on his 6 month follow up.

Conclusions

Primary Ewings Sarcoma with intracranial extension is extremely rare condition with only few case reports available in literature. One should consider Primary Ewings sarcoma of orbit as differential diagnosis for orbital tumours in young male patients especially with intracranial extension and distant metastasis. Despite multimodality treatment involving surgical excision and adjuvant chemo-radiotherapy these tumours are rapidly progressive and invariably fatal.

References

1. Carvajal R, Meyers P: Ewing's sarcoma and primitive neuroectodermal family of tumors . Hematol Oncol Clin North Am. 2005, 19(3):501-525. 10.1016/j.hoc.2005.03.004.
2. Le Deley MC, Paulussen M, Lewis I, Brennan B, Ranft A, Whelan J .: Cyclophosphamide compared with ifosfamide in consolidation treatment of standard-risk Ewing sarcoma: results of the randomized noninferiority Euro-EWING99-R1 trial.. J Clin Oncol . 2014, 32(23):2440-2448. 10.1200/JCO.2013.54.4833. Epub 2014 Jun 30
3. Woodruff G, Thorner P, Skarf B: Primary Ewing's sarcoma of the orbit presenting with visual loss . Br J Ophthalmol. 1988, 72:786-792. 10.1136/bjo.72.10.786.
4. Ewing J : Classics in oncology. Diffuse endothelioma of bone. James Ewing. Proceedings of the New York PathologicalSociety,1921.CACancerJClin.1972,Mar-Apr;22(2):95-98. 10.3322/canjclin.22.2.95.
5. Abbasi S, Cheema A: Ewing sarcoma of orbit with intracranial extension . Pak J Ophthalmol. 2015, 31:111-14.
6. Naqvi SH, Hameed S, Naqvi SH, Musani MA: Primary Ewing's sarcoma of the orbit with intracranial
7. extension abutting the temporal lobe: a rare case report. Int Ophthalmol. 2014, 34:1137-1140. 10.1007/s10792-014-9937-5
8. Kaliki S, Rathi SG , Palkonda VAR: Primary orbital Ewing sarcoma family of tumors: a study of 12 cases . Eye. 2018, 32:615-621. 10.1038/eye.2017.278 .
9. Harbert F, Tabor GL Jr : Ewing's tumor of the orbit, report of two cases . Am J Ophthalmol. 1950, Aug 33(8):1219-1225. 10.1016/0002-9394(50)90991-3.

10. Yamada S, Takahashi H: A dissected case of Ewing's sarcoma (reticulum cell sarcoma) of the orbit . Hirosaki Med J. 1957, 8:202-207.
11. Alfaar AS, Zamzam M, Abdalla B, Magdi R, El-Kinaai N: Childhood Ewing sarcoma of the orbit . J Pediatr Hematol Oncol. 2015, 37(6):433-437. 10.1097/MPH.0000000000000346.

Romanian Neurosurgery (2023) XXXVII (3): pp. 305-311
DOI: 10.33962/roneuro-2023-056
www.journals.lapub.co.uk/index.php/roneurosurgery

Stepwise approach and management of traumatic brain injury during pregnancy. A tertiary apex trauma centre experience

Sarita Kumari, Manish Jaiswal, Bal Krishna Ojha, Chhitij Srivastava

King George's Medical University, U.P., Lucknow, Uttar Pradesh, INDIA

ABSTRACT
Introduction. Trauma is the leading cause of non-obstetric maternal mortality and affects up to 8% of all pregnancies. Pregnant patients with traumatic brain injury (TBI) are a vulnerable population and management is complex with multiple special considerations. Recommendations proposed for the management of TBI patients are non-applicable to pregnant women as often have been excluded from these major trials. Evidence on TBI management in pregnant women is limited and mostly based on clinical experience.
Aim and objective. This study is to share our experiences in the management of TBI during pregnancy at a tertiary trauma centre in north India to formulate a stepwise approach consisting of different tiers of treatment.
Methods. Case records of thirty pregnant patients with traumatic brain injury admitted at the tertiary apex trauma centre of northern India during the period of January 2015 to June 2022 were retrospectively analyzed.
Results. Road traffic accidents (specifically two-wheelers) were the most common cause of TBI in pregnancy (80%). 60% suffered from moderate to severe TBI. Operative neurosurgical intervention was required in 30% of cases. During hospital stay, 27 patients (90%) had continuation of pregnancy. Nine patients (30%) who expired, belonged to moderate to severe TBI.
Conclusion. Moderate or severe TBI in pregnancy is associated with unfavourable maternal and fetal outcomes. The complex physiological modifications occurring in pregnancy have a crucial role in the management and require a stepwise approach. Fetal concerns and the paucity of high-quality evidence further complicate the issue and more studies including this group specifically are required.

Keywords
pregnancy,
traumatic brain injury,
Glasgow coma scale

Corresponding author:
Manish Jaiswal

King George's Medical University, U.P., India

manishjaiswal@kgmcindia.edu

Scan to access the online version

INTRODUCTION

Traumatic brain injury (TBI) resulting from Road traffic accidents (RTA) is a major cause of morbidity and mortality in the world especially in developing countries [1]. When coupled with pregnancy it imposes a significant physiological alteration that may confuse and complicate the clinical evaluation, resuscitation and definitive management of traumatic brain injury. According to the 2019 statistics provided by the Ministry of Road transport and Highways transport of India, there were 21794 RTA related female deaths in India, with 7632 deaths attributed to two wheelers related accidents [1]. Accurate prediction of pregnancy and its outcome after traumatic brain injury is largely unknown [2]. In the treatment of TBI with pregnancy, it should always be remembered that there are two patients being treated, the mother and the foetus. Resuscitation of the mother is the main action which has profound secondary implications on the foetus, and is followed by intrauterine resuscitation. Decision-making regarding such patients is a multidisciplinary effort requiring cooperation between neurosurgeons, gynaecologists, neonatologists, and anaesthetists [3]. Recommendations proposed for the management of TBI patients are not applicable to pregnant women as often have been excluded from these major trials. Evidence on TBI management in pregnant women is limited and mostly based on clinical experience.

Aim and objective

This retrospective study is to share our experiences in the management of TBI during pregnancy at tertiary trauma centre in north India so that we can formulate a stepwise approach consisting of different tiers of treatment based on pathophysiological knowledge, common sense in the selection of the best combination of monitoring and medical-surgical intervention, according to the different status of pregnancy, risks and benefits for both mother and foetus.

Materials and methods

All the patients of pregnancy with TBI admitted and treated at tertiary apex trauma centre of northern India during the period of January 2015 to June 2022 were included in this retrospective study. This trauma centre serves the northern part of the country as a tertiary institute and referral centre.

The data was collected from the case records available in the record section of the Department of Neurosurgery. Epidemiological and clinic-radiological parameters were tabulated and the outcome was analysed on the basis of morbidity & mortality. Follow up records were updated till December, 2022.

As with non-pregnant patients, initial evaluation and management was done in the emergency room. Airway, breathing and circulation were secured. Patients with Glasgow Coma Scale (GCS) score of less than 8 or with aspiration were intubated and those with a GCS score of more than 8 kept on oxygen mask at 5 litre/min to keep the target Oxygen Saturation (SPO2) >95% [4]. Intravenous access was secured with 2 large bore (18 Gauze) lines. Once the patient was stabilized a secondary survey of history, clinical examination and relevant investigations was done.

The initial targets for resuscitation were (1) systolic blood pressure of 80–100 mmHg, (2) SpO2>95%, (3) haematocrit 25–36%, (4) platelet count >50,000/ cell mm3, (5) normal serum calcium, (6) temperature >35°C, (7) Prevention of metabolic acidosis and elevated serum lactate and (8) adequate analgesia [5].

Emergency Computerised Tomogram (CT) scan of head with protective abdominal lead shield and ultrasound of the abdomen were acquired in each case. Indications of CT scan were a GCS score of less than 15, a history of loss of consciousness for more than 30 minutes, a history of seizure, bradycardia, anisocoria and high velocity injuries. CT scan was obtained with shielding. Ultrasound abdomen with Doppler study was repeated in patients with a pregnancy of gestational age less than 28 weeks, patients who underwent a procedure or had deterioration in GCS score and in hemodynamically unstable patients. Magnetic Resonance Imaging (MRI) Brain was not done routinely due to technical feasibility. It was conducted in patients who were stable, conscious and oriented without neurological deficit but with persistent symptoms. Apart from radiological investigations blood investigations included Complete Blood Count, Coagulation profile, serum creatinine and urea, serum electrolytes, viral markers and Blood Grouping. Cross match was also sought in relevant patients.

Secondary survey physical examination of pregnant head injury patients was similar to non-pregnant ones and followed the same principles of full exposure with inspection and palpation followed by evaluation of patient by the neurosurgical team. A full obstetrician's examination was sought. Antiepileptic drug and osmotic cerebral dehydrants were used judiciously. The preferred antiepileptic drug was Levetiracetam. Indications of surgical intervention were similar to non-pregnant patients. Indications of emergency lower segment caesarean section were low GCS of mother having a pregnancy of Gestational Age (GA) >28wks).

Foetal wellbeing was ensured by evaluation and resuscitation of foetus performed simultaneously with maternal evaluation and resuscitation. We used external foetal assessment and ultrasound to assess foetal wellbeing. If the foetus was pre-viable, interventions were limited and focused mainly on maternal resuscitation.

RESULTS

30 patients of TBI in pregnancy were analysed during the study period, with ages ranging from 18 to 41 years with a median age of 25 years. Patients were admitted for 4 days on an average (with a range of 1-24 days) with different mode of injuries & 80% of injuries were a result of RTAs (specifically two-wheeler related) [Figure 1].

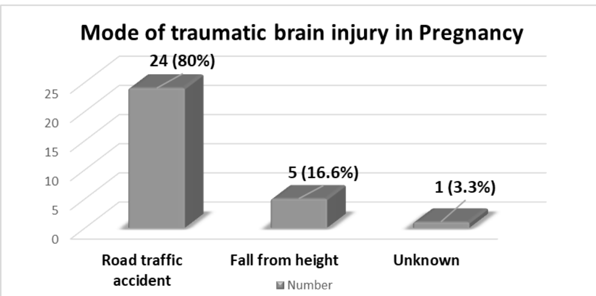


Figure 1. Mode of traumatic brain injury in pregnancy.

The time lag between injuries to admission for 29 of these patients ranged from 6 hours to 12 days, the data of time lag for 1 patient was not available as she was found in a field with no attendant to give history.

Six of our patients (28.6%) were primigravida, in 8 patients (38.1%) it was their second pregnancy, six patients were in their third pregnancy (28.6%) and one patient was pregnant for the 4th time. The month of gestation ranged from 2-9 with a median of 6 months. A majority of patients (36.67%) presented in the second trimester of pregnancy [Table 1].

Table 1. Incidence of traumatic brain injury in relation to trimester of pregnancy

Trimester of Presentation	Number of pregnant women	Percentage
First Trimester of pregnancy	10	33.33%
Second Trimester of pregnancy	11	36.67%
Third Trimester of pregnancy	9	30%

The GCS score at admission ranged from 4-15 with a median of 12 [Figure 2]. One patient had a mandibular fracture in addition to head injury. All 30 patients showed foetal cardiac activity at admission.

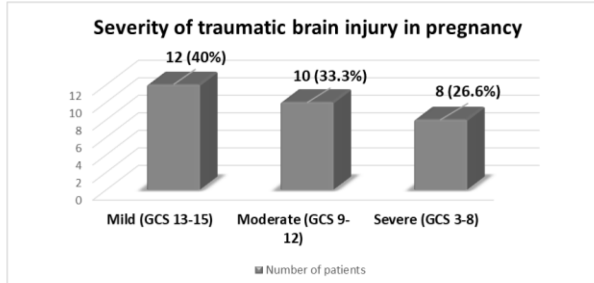


Figure 2. Severity of traumatic brain injury in pregnancy.

Ten of our patients had bilateral lesions (33.3%), and twenty (66.6%) had unilateral injuries (left sided in 12 and right sided in 8). The lesions were supratentorial in all 30 patients. Nine (30%) patients expired in hospital, out of which 4 patients were those who had undergone neurosurgical intervention, while 5 were those who had been managed conservatively. Out of the 9 (30%) patients who expired, 6 (20%) were those with a severe head injury, while 3 (10%) were those with moderate head injury. All patients with mild head injuries survived [Table 2].

Table 2. Radiological findings & mortality in traumatic brain injury during pregnancy.

CT head radiological findings	Left	Right	Bilateral	Total =30	Surgery done in	Expired
Acute SDH	3	2	0	5	4	2 (surgical)
EDH	1	2	0	3	2	1 (surgical)
Intra Cerebral Hematoma	6	4	6	16	1	3 (conservative)
Depressed fracture	1	0	0	1	1	-
Pneumocephalus	-	-	1	1	-	-
IVH	-	-	1	1	-	1(Conservative)
DAI			2	2	-	1 (conservative)

Hemispheric Infarct	1	0	0	1	1	1 (surgical)
	12	8	10 (33%)	30	9	9(30%)

Among the survivors, 19 out of 21 patients had a continuation of pregnancy, while 2 patients required medical termination of pregnancy. After discharge, all 21 patients were given antiepileptic drug in the form of Levetiracetam.

Out of 21 patients discharged, 17 were discharged in full GCS with a live foetus in utero, while the remaining 4 were discharged with a GCS of 14, 13, 10 and 9 respectively. All the 21 patients were alive at follow up, and their foetuses survived to term and were alive and well at follow up except for one who expired on day 10 after birth due to pneumonia.

Discussion

Consideration of physiological changes caused by pregnancy

Physiological changes during pregnancy must be in our minds while managing the TBI as there are some specific risks related to these changes. We have tried to elaborate these risks based on our experience in [Table 3].

Table 3. Consideration of Physiological Changes Caused by Pregnancy

Physiological change	Cause	Effect	Risk
Decreased systemic vascular resistance	To optimize uteroplacental blood flow	Capillary engorgement & tissue edema (Upper airway) Low blood pressure in first trimester	Risk of failed intubation Hemodynamic instability
Increased circulatory blood volume	Due sodium retention via R-A-A system activation	Relative anemia state	Delay in hemorrhagic shock recognition
Concentration of clotting factor raised	To prevent acute blood loss after delivery	Prothrombotic state	Risk of DVT & DIC
Diaphragm upward displacement	Due to uterine enlargement	Increased metabolic rate and oxygen consumption	Rapid episode of desaturation during intubation

Priorities in initial resuscitation

As per ATLS protocol initial resuscitation is done by ABCDE approach in trauma patients, but there is specific need of this population (TBI in Pregnancy) during trauma which must be tackled simultaneously for successful initial resuscitation [Table 4].

Table 4. Priorities in Initial Resuscitation during trauma with pregnancy

ABCDE approach	Specific need of this population	Management
A	augmented risk of failed intubation and acidic gastric contents aspiration compared to the non -pregnant population	Early placement of an endotracheal and a nasogastric tube is recommended to secure the airway
B	Risk of maternal and fetal hypoxia	Oxygen supplementation is mandatory to maintain a saturation > 95%
C	Risk of supine hypotension & decreased venous return Avoid high dose norepinephrine (risk of decrease placental perfusion)	Left tilt or manual uterine displacement Use of Phenylephrine is safe Use of O negative blood
D	Quick but accurate neurological assessment (GCS limitation) Distress of fetus evaluation	Obstetric evaluation of the fetus and USG
E	Entire exposure and examination	special regard to thorax, abdomen, pelvic and perineal regions to rule out fetal injuries. Avoiding extreme hypothermia

The main outcome of neurosurgical management in pregnant patients with traumatic brain injury is to maintain maternal and foetal survival [6,7]. The timing of surgery is a big challenge for neurosurgeons. In the first trimester, there is highest risk of spontaneous abortion caused by general anaesthesia (risk ratio = 1.58) [8]. In addition, a study has reported the incidence of spontaneous abortion of 15%–20% and a risk of congenital abnormalities of 3%–5% when the surgery is performed in the first trimester before 13 weeks of gestation [5]. Gestational age ranging from 13 to 23 weeks is usually a safe period for surgery for trauma cases in pregnancy [9,10]. Once the foetus becomes viable (>24 weeks), three risks of complications have to be faced, namely (1) supine hypotension, (2) neurodevelopmental delay in offspring and (3) premature delivery [5]. If trauma occurs with a viable foetus (>32 weeks of gestation), the usual clinical decision is to terminate the pregnancy by delivering the foetus especially in cases of acute neurological worsening. The preferred method is a caesarean section under general anaesthesia followed by neurosurgery [11,12,13]. Categorisation of mother & foetus risk: benefit, based on trimester of pregnancy and clinical status is formulated by us for management strategy [Table 5].

Table 5. Categorizing the mother and the Fetus Risks and Benefits during traumatic brain injury

Trimester & Clinical status	Monitoring & assessment	Protocol and purpose	Treatment option
During first & second trimester: Fetus is considered non-viable (<23 week)	Maternal assessment & stabilization In case of neurosurgical intervention	ATLS protocol Brain trauma foundation guideline	Optimizes fetal status Postoperative & ICU care
Third trimester: fetus is considered viable (>23 week)			
if mother clinical status is stable	Monitor the viability of fetus and documentation	To establish the appropriate time and method delivery	(Caesarean section alone or with neurosurgery)
if maternal condition is critical or cerebral damage leads to maternal brain death			Urgent caesarean delivery

Computed tomography scan is the neuroimaging of choice to evaluate TBI

CT scan head is frequently delayed due to fear of expose the developing fetus to harmful ionizing radiations. Ionizing radiations have a dose-dependent teratogenic and carcinogenic potential. NCCT Head, expose the fetus to a radiation amount below the threshold of 5,000 m-rad, considered safe for fetal damage [Table 6].

Table 6. Estimated fetal radiation adsorbed doses during some common radio-diagnostic procedures

Radiological procedure	m-rad
CT Head	0
CT Chest	16
CT Abdomen	3000

Determine fetal viability though fetal monitoring is fundamental

Serial echo-graphic approaches, electronic fetal heart rate monitoring (EFM), obstetric anamnesis and physical examination are various tools to determine the foetal viability. Abnormal heart rate pattern is early warning signal of maternal hemodynamic compromise and uteroplacental hypoperfusion, it allows to rapidly assess and prevent reversible causes such as hypoxia and acidosis

Avoid secondary insults of systemic origin

An injury whose effects do not occur at the time of the trauma but becomes evident in the following hours or days leads to secondary insults. GHOST-CAP mnemonic (Glycemia, Hb, Oxygen, Sodium, Temperature, Comfort, Arterial pressure & PaCo2) highlights eight pivotal elements which should be regularly assessed during patient's ICU stay to prevent secondary insults of systemic origin.

When to induce labor? Natural or caesarean section?

Preterm delivery is considered best, when delivery improve mother's prognosis. If definitive treatment can be safely delayed, and the gestational age is appropriate, to guarantee in-utero fetal development, additional stay in the ICU is recommended. No further prolongation of pregnancy is necessary after 32 weeks of gestation. Cesarean section should be reserved to cases when maternal injuries result in severe complications. In case of maternal stability, induced vaginal delivery should be considered in view of a non-viable fetus. Previous case reports and their management approach of TBI in pregnancy has been summarized in Table 7.

Table 7. Previous case reports of traumatic brain injury in pregnancy [7,9,14,15,16,17]

Reference	GCS	Gestational Age (in weeks)	Timing of Surgery	Outcome
Cirak et al.[7]	NA NA	38 39	CS Per vagina	Both Alive Both Alive
Satapathy et al.[9]	11 14 7	24 16 28	Craniotomy Both Conservative	Both Alive Both Alive
Dawar et al.[14]	11	36	CS->Craniotomy	Both Alive
Whitney et al.[15]	3	20	ICP monitor	Foetal Complication (Germinal Matrix haemorrhage)
Goldschlager et al[16]	9	34	CS-> craniotomy	Both Alive
Darlan et al.[17]	6 6	18 20	Craniotomy ICP monitor	NA NA

In our study, the main corroborative factor that appeared to affect the outcome was the admitting GCS score of the patient, as maximum morbidity as well as mortality was in those patients who had suffered a severe head injury, followed by patients with moderate head injury.

CONCLUSIONS

Moderate or severe TBI in pregnancy is associated with unfavorable maternal and fetal outcomes. The complex physiological modifications occurring in pregnancy have a crucial role in the management. Fetal concerns, and paucity of high-quality evidence further complicate the issue and more studies including this group specifically are required.

Abbreviations

RTA-Road Traffic accidents;
MTP- Medical Termination of Pregnancy;
LSCS- Lower segment caesarean section;
GCS- Glasgow Coma Scale;
TBI- Traumatic Brain Injury;
SPO2- Oxygen Saturation;
CT- Computerised Tomogram;
MRI-Magnetic Resonance Imaging;
GA- Gestational Age;
CS-Caesarean section;
ICP-Intracranial pressure;
NA-Not available.

REFERENCES

1. https://morth.nic.in/sites/default/files/RA_Uploading.pdf
2. El-Kady D, Gilbert WM, Anderson J, Danielsen B, Towner D, Smith LH. Trauma during pregnancy: an analysis of maternal and fetal outcomes in a large population. Am J Obstet Gynecol. 2004 Jun;190(6):1661-8. doi: 10.1016/j.ajog.2004.02.051. PMID: 15284764.
3. Macarthur A. Craniotomy for suprasellar meningioma during pregnancy: role of fetal monitoring. Can J Anaesth. 2004 Jun-Jul;51(6):535-8. English, French. doi: 10.1007/BF03018393. PMID: 15197113.
4. Jain V, Chari R, Maslovitz S, Farine D; Maternal Fetal Medicine Committee, Bujold E, Gagnon R, Basso M, Bos H, Brown R, Cooper S, Gouin K, McLeod NL, Menticoglou S, Mundle W, Pylypjuk C, Roggensack A, Sanderson F. Guidelines for the Management of a Pregnant Trauma Patient. J Obstet Gynaecol Can. 2015 Jun;37(6):553-74. English, French. doi: 10.1016/s1701-2163(15)30232-2. PMID: 26334607.
5. Reddy SV, Shaik NA, Gunakala K. Trauma during pregnancy. J Obstet Anaesth Crit Care 2012;2:3-9
6. ACOG Committee on Obstetric Practice. ACOG committee opinion. Nonobstetric surgery in pregnancy. Number 284, August 2003. Int J Gynaecol Obstet. 2003

Oct;83(1):135. doi: 10.1016/s0020-7292(03)00391-6. PMID: 14635614.
7. Cirak, Bayram et al. Neurosurgical procedures in pregnancy. Acta Cirúrgica Brasileira [online]. 2003, v. 18, n. 1. Epub 19 Feb 2003. ISSN 1678-2674. https://doi.org/10.1590/S0102-86502003000100002.
8. Duncan PG, Pope WD, Cohen MM, Greer N. Fetal risk of anesthesia and surgery during pregnancy. Anesthesiology. 1986 Jun;64(6):790-4. doi: 10.1097/00000542-198606000-00019. PMID: 3717642.
9. Satapathy MC, Mishra SS, Das S, Dhir MK. Emergency management strategy for pregnant head trauma victims–Case reports and review of literatures. Indian Journal of Neurotrauma. 2014 Jun;11(01):45-8.
10. Liu PL, Warren TM, Ostheimer GW, Weiss JB, Liu LM. Foetal monitoring in parturients undergoing surgery unrelated to pregnancy. Can Anaesth Soc J. 1985 Sep;32(5):525-32. doi: 10.1007/BF03010803. PMID: 4041953.
11. Qaiser R, Black P. Neurosurgery in pregnancy. Semin Neurol. 2007 Nov;27(5):476-81. doi: 10.1055/s-2007-991129. PMID: 17940927.
12. Ng J, Kitchen N. Neurosurgery and pregnancy. J Neurol Neurosurg Psychiatry. 2008 Jul;79(7):745-52. doi: 10.1136/jnnp.2007.117002. PMID: 18559459.
13. Wang LP, Paech MJ. Neuroanesthesia for the pregnant woman. Anesth Analg. 2008 Jul;107(1):193-200. doi: 10.1213/ane.0b013e31816c8888. PMID: 18635488.
14. Dawar P, Kalra A, Agrawal D, Sharma BS. Decompressive craniectomy in term pregnancy with combined cesarean section for traumatic brain injury. Neurol India. 2013 Jul-Aug;61(4):423-5. doi: 10.4103/0028-3886.117588. PMID: 24005739.
15. Whitney N, Raslan AM, Ragel BT. Decompressive craniectomy in a neurologically devastated pregnant woman to maintain fetal viability. J Neurosurg. 2012 Mar;116(3):487-90. doi: 10.3171/2011.11.JNS11707. Epub 2011 Dec 23. PMID: 22196099.
16. Goldschlager T, Steyn M, Loh V, Selvanathan S, Vonau M, Campbell S. Simultaneous craniotomy and caesarean section for trauma. J Trauma. 2009 Apr;66(4):E50-1. doi: 10.1097/TA.0b013e318031cc98. PMID: 19088560.
17. Darlan D, Prasetya GB, Ismail A, Pradana A, Fauza J, Dariansyah AD, Wardana GA, Apriawan T, Bajamal AH. Algorithm of Traumatic Brain Injury in Pregnancy (Perspective on Neurosurgery). Asian J Neurosurg. 2021 May 28;16(2):249-257. doi: 10.4103/ajns.AJNS_243_20. PMID: 34268147; PMCID: PMC8244712.
18. Sharma R, Grover VL, Chaturvedi S. Health-risk behaviors related to road safety among adolescent students. Indian J Med Sci. 2007 Dec;61(12):656-62. PMID: 18174635.

Romanian Neurosurgery (2023) XXXVII (3): pp. 312-316
DOI: 10.33962/roneuro-2023-057
www.journals.lapub.co.uk/index.php/roneurosurgery

Epidemiology of head injury in a tertiary care hospital in south India

Anand Shanmugaiah, R. Rajkumar

Institute of Neurosurgery, Madras Medical College, INDIA

ABSTRACT

Introduction: The annual incidence of traumatic brain injury is around 28-70 million in the world which causes morbidity and mortality. In India, around 1 million suffer from severe head injury and the mortality rate is around 1 lakh per year. As time progresses there is an observable increase in rates of Head injuries. This study focuses on the epidemiology of traumatic head injury in a tertiary care centre (Madras Medical College, Chennai- India)

Materials and methods: This study is a prospective study covering all head injury cases admitted from January 2022 – December 2022 in our tertiary care centre. These patients are treated according to the protocol of the Institute of Neurosurgery. This study covers the management, discharge and follow-up of these patients.

Results: The total number of patients admitted with head injuries from January 2022 to December 2022 was 2061. Among them, RTA tops head injury mortality and morbidity. Among RTA, Two-wheeler-associated injuries were most common. Drunken driving, and not wearing helmets and protective gear played a vital role in head injuries.

Conclusion: Primary prevention can be the key to reducing the national burden of head injuries.

Keywords
head injury,
injuries,
prevention

Corresponding author:
R. Rajkumar

Institute of Neurosurgery, Madras Medical College, India

ra.kumar4410@gmail.com

Scan to access the online version

INTRODUCTION

Head injuries are defined as: 'An alteration in brain function, or other evidence of brain pathology, caused by an external force[1] . Of all the injuries, head injury accounts for most cases of death and disability. 28 to 70 million individuals worldwide sufferred from head injury in 2018[2]. Every year October 17 is celebrated as world trauma day. It highlights the increasing rate of accidents and the need to prevent it. The major cause of these deaths were traffic accident (43.9%) followed by accidental fall (5.1%). The majority of deaths were in the age of 30-45[4]. A total of 10,50,945 assault cases were registered during 2019 which accounted for 32.6% of total IPC crimes, out of which hurt (5,45,061 cases)accounted for maximum number of cases i.e. 51.9%, followed by cases of causing death by negligence and others[5]. It is estimated that the total costs of road traffic injuries alone is about 3% of GDP in India[6].Road travel seem to be the preferred choice with over 60% using personal vehicles to commute. The Industrial movements of goods through Roadways are also on the rise with over 2 billion metric tons of freight being transported every year.The total number of vehicles in

FISCAL year 2019 is around 295.8 million. The Indian automobile manufacturers produced a whooping 26.36 million motor vehicles in 2019-2020[7].

AIM

To study the Epidemiology of Head Injury in our tertiary care centre, Madras Medical College and hospital, Chennai.

MATERIALS AND METHODS

This is a prospective study covering all head injury cases admitted from january 2022 - December 2022 in a tertiary care centre, Madras Medical College and hospital, Chennai. IEC clearance was taken, as also consent from study subjects. The treatment is initiated upon arrival in Zero delay ward and they are triaged by emergency team. These patients are treated according to the protocol of Institute of Neurosurgery. This study covers the management, discharge and follow up of these patients at 1 month, 3 months and 6 months. During the study period, 2061 cases of head injury were admitted in our tertiary care center. Those patients who were brought dead following injury and Head injury cases wherein the patients/attenders didn't give consent for the study were excluded. Socio economic status, mechanism of injury,treatment and follow up information were recorded using a structured questionnaire. Data was entered in Excel sheets and appropriate analysis done using SPSS software.

RESULTS

RTA cases were the most common cause of Head Injury with 1426 cases, Assault 334 cases, fall 257 cases and other causes were 44.

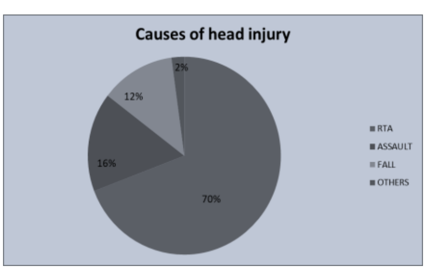


Figure 1, Table 1. Causes of head injury.

Head injury cause	Total	Percentage
RTA	1426	70%
Assault	334	16%
Fall	257	12%
Others	44	2%

Table 2. Mode of RTA

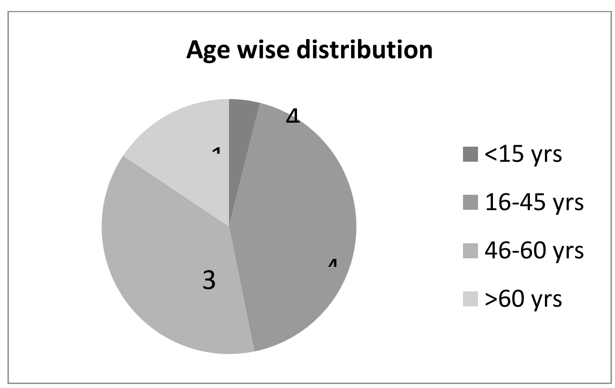
Mode of travel	Number	Percentage
Two wheeler	1155	81%
3 and 4 wheeler , heavy vehicle	185	13%
Pedestrian	86	6%

80% of victims of head injury due to assault under the influence of alcohol. Further in 95% of cases, injuries were caused by known persons to the victims.

Fall from height was the cause of injury in 213 (83%) followed by self fall at ground level in 44 (17%). Of the 2061 cases admitted 1786 cases are male and 275 were female. 81 cases were less than 15 yrs of age, 884 cases was between 16-45 years of age, 774 cases was between 46-60 years of age and 322 cases was more than 60 years of age.

Table 3, Figure 2. Age wise distribution

Age	Number	Percentage
< 15 years	81	4%
16 – 45 years	884	43%
45 – 60 years	774	38%
> 60 years	322	15%



There are various methods of classifying the severity of head injury, to highlight the amount of disruption of brain parenchyma. These are the Glasgow Coma Scale (GCS), the Abbreviated Injury Severity Score (AIS), and so on.The aim of categorisation is to predict the outcome[13]. In our study, GCS scale is followed to classify the head injury.

Table 4, Figure 3. Severity of head injury

GCS at admission	Severity of head injury	Number of cases	Percentage
13-15	Mild	1071	52%
9-12	Moderate	639	31%

8 and <8	Severe	351	17%

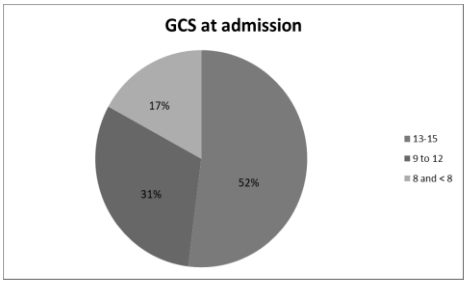


Table 5. CT findings

CT Finding	Number of cases	Percentage
Extradural hematoma	91	4%
Subdural hematoma	542	26%
Contusions and intracerebral hemorrhage	569	27%
Subarachnoid hemorrhage	268	14%
Diffuse axonal injury (CT showing IVH, brainstem contusions)	116	6%
Cranial bone fractures	435	21%
Diffuse cerebral edema	40	2%

Among the head injuries- acute subdural hemorrhage, subarachnoid hemorrhage, cerebral contusions and calvarial fractures were predominantly encountered.

Out of 2061 cases admitted, 1648 cases were managed conservatively and 413 cases were operated. Of the 413 cases, decompressive craniectomy was done in 194 cases of acute SDH and in 60 cases of basifrontal contusions and in 52 cases of ICH, craniotomy and evacuation of hematoma done in 44 cases of EDH, wound debridement and excision of fracture fragment done in 63 cases of calvarial depressed fractures.

Table 6: Management of head injury

Management	Number of cases
Conservative	1648
Surgery	413

Table 7: Surgical management

Injury	Surgical management	Number of cases
SDH	Decompressive craniectomy	194
Basifrontal contusion	Decompressive craniectomy	60
Intracerebral hemorrhage	Decompressive craniectomy	52
Extra dural hemorrhage	Craniotomy and evacuation of EDH	44
Calvarial depressed fractures	Wound debridement and excision of fracture fragment	63

Mortality statisctics: There was 352 (17%) in hospital deaths in patients admitted with head injury.

Table 8: Causes of mortality

Cause of mortality	Number of cases	Percentage
RTA	211	60%
Fall	88	25%
Assault	4	1%
Others	49	14%

Table 9: GOS - Discharge & Follow-up

Time	GOS-1	GOS-2	GOS-3	GOS-4	GOS-5
During hospital stay	352(17%)	19 (1%)	247 (12%)	309 (15%)	1134 (55%)
1 month later	14 (0.6%)	-	142 (8.4%)	208 (12%)	1345 (79%)
3 months later	-	-	101 (6%)	154 (9%)	1440 (85%)
6 months later	-	-	67 (4%)	118 (7%)	1510 (89%)

GOS 1 - Death

GOS 2 - Persistent vegetative state- unresponsive, speechless, may open eyes

GOS 3 - Severe disability (conscious but disabled)-dependent for daily support.

GOS 4 - Moderate disability (disabled but independent)

GOS 5 - Good recovery-resumption of normal life despite minor deficits.

Discussion

Traumatic brain injury is defined as "An occurrence of injury to the head (arising from blunt or penetrating trauma or from acceleration-deceleration forces) by an external agent with at least one of the following: Observed or self-reported alteration of consciousness or amnesia due to head trauma, and/or,

• Neurological or neuropsychological changes (determined from neurologic and neuropsychological examinations) or

• diagnosis of skull fracture or intracranial lesions (determined from radiological examination or other neuro- diagnostic procedures) that

could be attributed to head trauma and/or

• Occurrence of death resulting from trauma with head injury or traumatic brain injury listed on the Death Certiicate, Autopsy Report, or Medical Examiner's Report in the sequence of conditions that resulted in death." [14]

The causes of head injury were RTA, Assault, Fall, Train accidents, animal attack, sporting activity, fall of objects. The major cause of head injury causing Mortality and morbidity is RTA. The external causes of injury, pattern and circumstances was made as per ICD- 10 classification methods[15].

In our study, we admitted a total of 2061 patients with head injury. RTA was the major cause of head injury accounting for 1426 cases and others were 635. We excluded brought dead, patients whom didn't give consents and unknown patient. In the study, males were affected 6.5 times higher than females. This is more compared to previous studies. In that study conducted in a level 1 trauma centre in India, the results showed that the male:female ratio was 5:1[16] .The age group between 15-60 yrs we had 1658 patients followed by 322 patients in >60 age group and 81 cases in below <15 age group. The leading cause was observed to be RTA with 1426 cases (70%), 334 cases (16%) of assault and 257 cases (12%) of fall. The leading cause in RTA was motorcycle accidents 1340 cases (94%). Alcohol consumption and not wearing protective gears were found to be significant contributing factor to Head injury morbidity and mortality.

ENT bleed and scalp injuries were noted in majority of cases. Swelling and abrasions of face was found in 95% of patients in the present study. History of seizures was present in 86 patients. Scalp injuries were found to be common in previous studies which is in accordance to the present study[17,18,19].

On admission, 351 cases (17%) were of severe head injury, 639 cases (31%) were of moderate head injury and 1071 cases (52%) were of mild head injury. On average, patients were admitted for 10 days. 20% (413 patients) of the head injury patients underwent surgery and rest 80% (1648 patients) were treated conservatively. In the present study, 17% died (352 patients). The mortality was seen in patients with severe head injuries and low GCS score. Mortality in the present study was common in the age group of 41-60. It is in contrast to the study [17,20] where common age group being 31-40.

In CT findings- SDH, contusion and SAH dominated the cause for mortality. 352 patients died between 3-7 day of admission. In follow-up, 22 patients died. At the end of 6 months 89% of cases (1510 patients) showed good outcome. This is similar to the previous study[21].

Conclusion

Head injury is one of the leading cause of major public health problem in India. Road accidents are the major cause of morbidity and mortality. Alcohol and not using protective gears contribute to Head injury. It requires researchers, policy makers and surveillance programs to implement effective evidence-based interventions. Prevention and care, follow up of head injury patients are a multidisciplinary area and requires inter-sectoral co-ordination for planning. By improving our system with better reporting and documentation of head injury cases, we will be able to make decisions in planning and make appropriate multimodality approaches to reduce the morbidity and mortality of head injury cases with available resources.

References

1. Menon DK, Schwab K, Wright DW, Maas AI, Demographics and Clinical Assessment Working Group of the International and Interagency Initiative toward Common Data Elements for Research on Traumatic Brain Injury and Psychological Health Position statement: definition of traumatic brain injury. Arch Phys Med Rehabil. 2010;91:1637-40.
2. Estimating the global incidence of traumatic brain injury. Dewan MC, Rattani A, Gupta S, et al. https://pubmed.ncbi.nlm.nih.gov/29701556/ J Neurosurg. 2018:1–18. – PubMed.
3. Report of technical group on population projection november 2019,by National commission on population, Ministry of health & family welfare,india., based on 2011 census.
4. Accidental deaths and suicides in india - 2019,by National crime records bureau,india.(Empowering Indian Police with information technology).
5. Crime in India-2019. Volume 1 https://ncrb.gov.in/sites/default/files/CII%202019% 20Volume%201.pdf VI.Planning Commission Report of the working group on prevention of road accidents and injuries, Government of India, 2002.
6. https://morth.nic.in/road-accident-in-india

7. Roadways- The world Factbook.,www.cia.gov. Archived from the original on 2021-07-12.
8. Report of technical group on population projection november 2019,by National commission
9. population, Ministry of health & family welfare,india., based on 2011 census https://en.wikipedia.org/wiki/Coimbatore.
10. The Hindhu, newspaper dated,june 29,2018.
11. GO (Ms) No 214, dated 05.06.2018, health and family welfare department,tamilnadu, India. XIII.Teasdale G, Jennett B. Assessment of coma and impaired consciousness a practical scale. Lancet1974; 2:81-4. The Hindhu, newspaper dated,june 29,2018.
12. Centre for Disease Control and World Health Organization. Standards for Surveillance in Neurotrauma,2000 (unpublished document). Epidemiology of head injuries, National institute of mental health and Neurosciences, Bangalore 1994.
13. World Health Organization. International classification of diseases 10th revision, 1996.
14. Rahman MA, Ramazan. Patterns of head injuries (cranio-cerebral) in road traffic accidents in Riyadh region of KSA. Dinajpur Med Coll J 2015;8:195-9.
15. Tandle RM, Keoliya AN. Patterns of head injuries in fatal road traffic accidents in a rural district of Maharashtra autopsy based study. J Indian Acad Forensic Med 2011;33:228-31.
16. Munakomi S. A comparative study between Marshall and Rotterdam CT scores in predicting early deaths in patients with traumatic brain injury in a major tertiary care hospital in Nepal. Chin J Traumatol 2016;19:25-7.
17. Epidemiological study of Trauma brain injury in Trauma centre, New Delhi, in Journal of emergency Trauma & shock 2015 Jul-Sep.

Romanian Neurosurgery (2023) XXXVII (3): pp. 317-327
DOI: 10.33962/roneuro-2023-058
www.journals.lapub.co.uk/index.php/roneurosurgery

Percutaneous pedicle screw fixation. A comprehensive review of techniques, outcomes, and advancements

Muhammad Jehanzeb, Ahtesham Khizar, Naeem Ul Hassan, Khawar Anwar, Moin Islam Arain, Asif Bashir

Punjab Institute of Neurosciences, Lahore, PAKISTAN

ABSTRACT

Percutaneous pedicle screw fixation (PSF) has emerged as a promising alternative to traditional open surgical approaches for spinal stabilization. Pedicle screws are inserted through percutaneous access sites without substantial soft tissue dissection. To ensure proper screw insertion, the procedure employs fluoroscopic or image-guided navigation devices. The popularity of percutaneous PSF has accelerated because of its prospective advantages and the mounting body of research demonstrating its effectiveness and safety. However, there are a few drawbacks to the procedure, such as a longer learning curve for surgeons, poor visibility when inserting screws, and a requirement for specialized tools and imaging guiding systems. This article provides a comprehensive review of the technique of percutaneous PSF, its clinical outcomes, and recent advancements in the field. It also aims to analyze the efficacy, safety, and limitations of percutaneous PSF, as well as explore the evolving technologies and techniques that have contributed to its improved application.

Keywords
spine,
lumbar vertebrae,
thoracic vertebrae,
pedicle screws,
fracture fixation

Corresponding author:
Muhammad Jehanzeb

Department of Neurosurgery.
Punjab Institute of Neurosciences,
Lahore, Pakistan

drjehanzeb28@gmail.com

Scan to access the online version

INTRODUCTION

Spinal instability and deformities, such as degenerative disc disease, spondylolisthesis, and spinal fractures, often require surgical intervention for stabilization and restoration of spinal alignment. Traditional open surgical approaches for pedicle screw fixation (PSF) have been widely used and proven effective. However, these approaches are associated with significant tissue disruption, prolonged hospital stays, higher complication rates, and prolonged recovery periods.[1,2] Percutaneous PSF, also known as minimally invasive PSF, has emerged as an alternative technique to address these drawbacks. It involves the insertion of pedicle screws through percutaneous access points without extensive soft tissue dissection. The technique utilizes fluoroscopic or image-guided navigation systems to facilitate accurate screw placement.[3,4]

The significance of percutaneous PSF lies in its potential to provide several advantages over open surgical techniques. Firstly, the percutaneous approach minimizes tissue damage, leading to reduced blood loss, postoperative pain, and soft tissue complications. This approach allows for preservation of the paraspinal muscles, which can help maintain spinal stability and enhance postoperative functional outcomes. Secondly, the reduced tissue disruption may result in shorter hospital stays, faster recovery, and improved patient satisfaction. Furthermore, percutaneous PSF offers the potential for reduced intraoperative radiation exposure for both patients and surgical staff.[3]

Understanding the background and significance of percutaneous PSF is crucial for healthcare professionals and researchers involved in spinal surgery. It enables them to make informed decisions regarding surgical approaches, consider alternative techniques, and contribute to the ongoing advancements and developments in spinal stabilization procedures.[5,6] The adoption of percutaneous PSF has gained momentum due to its potential benefits and growing evidence supporting its safety and efficacy. However, the technique also presents challenges, including a steeper learning curve for surgeons, limited visualization during screw placement, and the need for specialized instruments and imaging guidance systems. Therefore, further research and exploration are needed to optimize the technique, refine patient selection criteria, and evaluate long-term outcomes. By discussing the current challenges and limitations associated with percutaneous PSF, this article aims to identify potential areas for future research and development. Exploring emerging technologies, refining techniques, and addressing limitations can contribute to further improving patient outcomes and advancing the field.

Discussion

Biomechanical principles underlying percutaneous pedicle screw fixation

Percutaneous PSF is a technique used to achieve spinal stability and fusion by placing screws through percutaneous access points into the pedicles of vertebral bones. The biomechanical principles underlying this technique are essential to understand its efficacy and to optimize surgical outcomes. The key biomechanical principles associated with percutaneous PSF are given below:

a. **Screw purchase and stability:** The primary biomechanical objective of PSF is to achieve secure screw purchase within the pedicle and maximize stability. Screw purchase refers to the engagement of the screw threads within the pedicle bone, allowing for effective load transfer and resistance to pull-out forces.[7]

b. **Load sharing and stability:** Percutaneous PSF provides load sharing and stability to the spinal column. The screws are inserted into the pedicles of adjacent vertebrae, allowing for the transfer of axial and torsional loads. By engaging the pedicles, the screws enhance the stability of the construct, reducing motion at the treated segment and promoting fusion.[7,8]

c. **Correction of spinal deformities:** PSF is frequently employed to correct spinal deformities such as scoliosis, kyphosis, and spondylolisthesis. The technique allows for three-dimensional deformity correction by applying corrective forces to the vertebrae through the screws and connecting rods.[9]

d. **Three-column concept:** Percutaneous PSF follows the three-column concept, which aims to restore and maintain the stability of the anterior, middle, and posterior columns of the spine. The screws inserted into the pedicles provide posterior column stability, complementing the anterior column support provided by interbody fusion or other anterior procedures.[10,11]

e. **Screw-bone interface:** The success of percutaneous PSF relies on the biomechanical stability of the screw-bone interface. Achieving adequate purchase within the pedicle is essential to prevent screw loosening, pull-out, or breakage. Factors influencing the screw-bone interface include screw size, design, thread profile, cortical bone integrity, and bone quality.[12]

f. **Stress distribution and preservation of spinal motion segments:** PSF should distribute stresses evenly across the instrumented spinal segments, minimizing stress concentrations and avoiding adjacent segment degeneration. The technique aims to preserve the range of motion of the instrumented spinal segments.[7]

g. **Biomechanical considerations for osteoporotic spines:** Osteoporosis is characterized by a decrease in bone density and quality, which can compromise the screw-bone interface and stability of PSF. Osteoporotic vertebrae have lower bone mineral density, diminished trabecular bone structure, and increased susceptibility to screw loosening, pullout, or vertebral body fracture. Therefore, careful screw trajectory planning and selection of appropriate screw size and design are crucial in osteoporotic spines.[7]

Percutaneous Pedicle Screw Fixation Techniques

Percutaneous PSF techniques have evolved to improve surgical outcomes and minimize tissue trauma compared to traditional open approaches. The use of image guidance and navigation systems further enhances the accuracy and safety of percutaneous pedicle screw placement. A description of percutaneous techniques and the role of image guidance and navigation systems is given below:

A. **Percutaneous techniques:** Percutaneous PSF involves the insertion of screws through small incisions, typically aided by fluoroscopy or navigation systems, without extensive soft tissue dissection. Key steps include:

   a. **Patient positioning:** The patient is positioned in a prone position on the operating table, allowing access to the targeted vertebrae.
   b. **Percutaneous access points:** Small incisions are made near the targeted vertebrae to access the pedicles. Dilators or cannulas are then used to create a pathway to the pedicle entry point.
   c. **Pedicle screw insertion:** Pedicle screws of appropriate length and diameter are inserted into the pedicles under fluoroscopic guidance or navigation assistance. The screws are carefully advanced, ensuring proper engagement within the pedicle.
   d. **Rod placement and fixation:** Once the screws are in place, connecting rods or plates are attached to achieve spinal alignment and stability. The rods are contoured to match the natural curvature of the spine.
   e. **Closure:** The incisions are closed, and appropriate postoperative care is provided to the patient.

B. **Image guidance and navigation systems:** Image guidance and navigation systems play a crucial role in percutaneous PSF, providing real-time visualization and assisting in accurate screw placement. These systems utilize preoperative imaging such as Computed Tomography (CT) or Magnetic Resonance Imaging (MRI) scans and intraoperative imaging such as fluoroscopy or 3D navigation to guide the surgical procedure. Key components include:

   a. **Fluoroscopy:** Fluoroscopy provides live X-ray imaging during the surgery, enabling the surgeon to visualize the bony landmarks, pedicles, and screw trajectories. It helps ensure accurate screw placement and assess the quality of screw purchase.
   b. **3D Navigation systems:** 3D navigation systems use preoperative imaging data to create a virtual 3D representation of the patient's spine. Real-time tracking of instruments and screws is facilitated using optical or electromagnetic sensors. The system provides guidance and feedback to assist the surgeon in precise screw placement.
   c. **Robotic-assisted navigation:** Robotic-assisted navigation systems utilize robotic arms and intraoperative imaging to guide the surgeon's movements. The robotic system assists in preoperative planning, intraoperative screw trajectory guidance, and screw insertion.
   d. **Optical navigation:** Optical navigation systems use infrared cameras and specialized instruments with reflective markers to track their positions relative to the patient's anatomy. The system provides real-time feedback on the instrument's location and trajectory.[13,14]

Explanation of procedural steps and instrumentation

Percutaneous PSF is a minimally invasive technique used for spinal stabilization and fusion. The procedure involves specific steps and utilizes specialized instrumentation. Here is an explanation of the procedural steps and instrumentation commonly employed in percutaneous PSF:

a. **Preoperative planning:** CT or MRI scans are obtained to evaluate the spinal anatomy, assess the pedicle morphology, and determine the appropriate screw size and trajectory.
b. **Patient positioning:** The patient is positioned prone on the operating table, with padding and

supports to maintain proper alignment and accessibility to the targeted vertebrae.

c. **Percutaneous access/skin incisions:** Small incisions, typically ranging from 1 to 2 cm in length, are made near the targeted vertebrae to access the pedicles.
d. **Pedicle preparation and screw placement:** 1) Pedicle identification: Fluoroscopic guidance or navigation systems are used to identify the entry points of the pedicles. 2) Pedicle preparation: Sequential dilators or cannulas are inserted through the incisions to create a pathway to the pedicles while minimizing soft tissue disruption. 3) Screw insertion: Pedicle screws, with self-drilling and self-tapping features, are carefully inserted into the pedicles. Fluoroscopic guidance or navigation assistance is used to ensure accurate screw placement and trajectory.
e. **Rod placement and fixation:** 1) Rod bending: Connecting rods, typically made of titanium or stainless steel, are contoured to match the natural curvature of the spine and correct any deformities. 2) Rod insertion: The rods are inserted into the previously placed rod connectors or connectors on the pedicle screws. 3) Rod fixation: Rods are secured in place using set screws or locking mechanisms on the rod connectors, providing stability and maintaining proper alignment.
f. **Closure and postoperative care:** The incisions are closed using sutures or adhesive strips, and sterile dressings are applied. Postoperative care includes monitoring for complications, pain management, and rehabilitation protocols tailored to the patient's specific condition.[15,16]

Comparison of percutaneous techniques with open surgical approaches

Percutaneous PSF techniques have gained popularity as an alternative to open surgical approaches for spinal stabilization. Several studies have compared the outcomes and advantages of percutaneous techniques with traditional open surgical approaches. Here, we provide a comparison between percutaneous techniques and open surgical approaches, supported by relevant references.

a. **Surgical trauma and complications:** Percutaneous techniques involve smaller incisions, minimal soft tissue dissection, and reduced muscle damage compared to open approaches. This results in decreased blood loss, lower infection rates, and shorter hospital stays.As compared to Open Surgical Approaches. Open surgical approaches require larger incisions, extensive muscle dissection, and more tissue trauma. These factors can contribute to increased blood loss, higher infection rates, and longer hospital stays.[17]
b. **Surgical time and operative efficiency:** Percutaneous techniques generally require a shorter surgical time compared to open approaches. The reduced soft tissue dissection and simpler instrumentation can contribute to faster procedure as compared to Open Surgical Approaches. Open surgical approaches typically involve more extensive exposure, requiring longer surgical times. The complexity of soft tissue handling and the need for additional steps may contribute to increased operative time.[18]
c. **Postoperative pain and recovery:** Percutaneous techniques are associated with less postoperative pain due to reduced muscle damage and tissue trauma. Patients may experience a quicker recovery, earlier ambulation, and a faster return to daily activities as compared to Open Surgical Approaches. Open surgical approaches can result in more postoperative pain due to extensive soft tissue dissection. The recovery period may be longer, with delayed mobilization and slower return to daily activities.[19]
d. **Correction of spinal deformities:** Percutaneous techniques can achieve comparable deformity correction to open surgical approaches in select cases. However, complex deformities may require open surgery for better visualization and three-dimensional correction. Open surgical approaches allow direct visualization of the spinal deformity, facilitating extensive correction, osteotomies, and fusion. These approaches are often preferred for complex deformities requiring substantial correction.[20,21]

Advantages and limitations of percutaneous pedicle screw fixation

Percutaneous PSF has gained popularity as a minimally invasive technique for spinal stabilization. Here is a discussion of the advantages and limitations of percutaneous pedicle screw fixation:

A. Advantages:

a. **Minimally invasive approach:** Percutaneous PSF involves smaller incisions and reduced soft tissue disruption compared to open surgical approaches. This results in less postoperative pain, reduced blood loss, shorter hospital stays, and faster recovery for patients.[3,23]
b. **Preservation of muscle and soft tissue:** The percutaneous technique minimizes disruption to the surrounding muscles and soft tissues, leading to decreased muscle trauma and potentially faster rehabilitation. Preservation of the muscle and soft tissue structures can help maintain spinal stability and reduce the risk of muscle atrophy.[22]
c. **Reduced infection rates:** The smaller incisions and limited soft tissue dissection associated with percutaneous pedicle screw fixation have been linked to lower rates of surgical site infections compared to open techniques.[22]
d. **Improved cosmesis:** The smaller incisions in percutaneous PSF result in less visible scarring and improved cosmetic outcomes compared to open surgery.[3]
e. **Reduced surgical morbidity:** Percutaneous PSF has been associated with lower rates of complications such as blood loss, wound complications, and postoperative infections compared to open techniques, resulting in reduced surgical morbidity.[22,23]

B. Limitations:

a. **Technical challenges:** Percutaneous PSF requires advanced surgical skills and familiarity with fluoroscopic guidance or navigation systems. The procedure can be technically demanding and may have a learning curve for surgeons.[3]
b. **Limited visualization:** The limited visualization provided by percutaneous techniques may make it challenging to address complex spinal pathologies or ensure precise screw placement. In some cases, conversion to an open technique may be necessary for optimal visualization and screw placement.[22]
c. **Radiation exposure:** The use of fluoroscopic guidance or navigation systems in percutaneous PSF exposes the patient and surgical team to ionizing radiation. Proper radiation safety measures should be taken to minimize radiation exposure.[3]
d. **Fusion rates:** Although percutaneous PSF has shown comparable fusion rates to open techniques in several studies, the long-term durability and stability of fusion achieved with percutaneous methods are still subjects of ongoing research and debate.[6,22]
e. **Limited indications:** Percutaneous PSF may not be suitable for all spinal pathologies or complex deformities. Cases involving severe instability, significant vertebral destruction, or extensive multi-level involvement may require open surgical approaches for optimal outcomes.[3]

It is important to consider the advantages and limitations of percutaneous PSF on a case-by-case basis, taking into account the patient's specific condition and individual risk factors. Collaboration between the surgeon and the patient is essential in making informed decisions about the most appropriate surgical approach.

Advancements in Percutaneous Pedicle Screw Fixation

Percutaneous PSF has undergone advancements and refinements over time, leading to improved techniques and outcomes. Here are some notable advancements in percutaneous PSF:

a. **Navigation and image guidance:** The integration of navigation systems and image guidance technologies, such as fluoroscopy, CT scans, or intraoperative 3D imaging, has enhanced the accuracy and precision of percutaneous pedicle screw placement. These technologies allow real-time visualization of the surgical field, aiding in optimal screw trajectory and reducing the risk of misplacement.[3,22]
b. **Robotic assistance:** Robotic-assisted percutaneous PSF is an emerging advancement that offers increased precision and control during screw placement. Robotic systems provide intraoperative navigation, image guidance, and robotic arm assistance, enhancing the surgeon's ability to achieve accurate screw trajectories.

Robotic assistance can potentially improve the safety and efficacy of percutaneous PSF.[23]

c. **Cement augmentation:** Cement augmentation techniques, such as cement-augmented screws or vertebroplasty/kyphoplasty, have been applied in percutaneous PSF to enhance screw stability and fixation, especially in osteoporotic spines. The use of bone cement can improve screw purchase within the pedicle and increase construct rigidity. This advancement has shown promising results in improving screw pullout strength and reducing the risk of screw loosening or failure.[24,25]

d. **Expandable pedicle screws:** Expandable pedicle screws are designed to be inserted in a compact form and then expanded within the pedicle, providing improved anchorage and fixation. These screws allow for greater bone-screw interface contact, potentially enhancing biomechanical stability. Expandable pedicle screws have shown promise in improving screw pullout strength and reducing the risk of screw loosening or migration.[24,25]

e. **Biomechanical studies and finite element analysis:** Advancements in biomechanical studies and finite element analysis have contributed to a better understanding of the biomechanical properties and load-sharing characteristics of percutaneous pedicle screw fixation. These studies help optimize screw design, implant selection, and surgical techniques, leading to improved construct stability and patient outcomes.[26] Biomechanical studies involving in vitro testing, cadaveric studies, and animal models provide valuable insights into the load-sharing characteristics, stability, and strength of percutaneous PSF. These studies help assess the effects of various factors, such as screw design, screw length and diameter, bone quality, and loading conditions, on the biomechanical performance of the construct. They aid in optimizing screw placement techniques, evaluating the risk of screw pullout or failure, and determining the appropriate surgical parameters for achieving desired outcomes.[26,27] Finite element analysis (FEA) is a computational technique widely used in biomechanical studies of percutaneous pedicle screw fixation. FEA allows for the creation of a virtual model of the spinal segment, incorporating anatomical details, material properties, and boundary conditions. Through FEA, researchers can simulate and analyze the distribution of stresses and strains within the spinal construct, predict the response to different loading scenarios, and evaluate the impact of various variables on the biomechanical performance. This analysis provides insights into the stress distribution along the screws, vertebral bodies, and intervertebral discs, helping optimize the design, placement, and fixation techniques.[28,29] Overall, biomechanical studies and finite element analysis play a critical role in improving our understanding of percutaneous PSF, guiding surgical decision-making, and enhancing patient outcomes.

These advancements in percutaneous PSF aim to enhance surgical precision, optimize screw placement, improve construct stability, and expand the indications for minimally invasive spinal surgeries. However, further research and long-term clinical studies are needed to validate these advancements and evaluate their impact on patient outcomes.

Overview of emerging technologies, such as Robotic Assistance and Augmented Reality

Emerging technologies are transforming the field of spinal surgery, offering new tools and techniques that improve surgical precision, patient outcomes, and surgical workflow. Two notable emerging technologies in spinal surgery are robotic assistance and augmented reality. Here is an overview of these technologies:

A. **Robotic Assistance:** Robotic-assisted spinal surgery involves the use of robotic systems to enhance surgical precision, accuracy, and safety. These systems provide surgeons with real-time navigation, image guidance, and robotic arm assistance, allowing for improved screw placement, optimal trajectory, and increased procedural efficiency. Advantages of robotic assistance are:

   a. **Improved accuracy:** Robotic systems enable highly accurate preoperative planning and intraoperative execution, resulting in precise screw placement and reduced risk of complications.

b. **Enhanced safety:** Real-time intraoperative navigation and feedback help prevent potential errors, such as screw misplacement or neural damage.
c. **Workflow efficiency:** Robotic assistance can streamline surgical workflow, decrease surgical time, and potentially reduce radiation exposure for both the patient and surgical team.[30,31]

B. **Augmented Reality:** Augmented reality (AR) is a technology that overlays virtual information onto the real-world surgical view, providing surgeons with real-time guidance and anatomical visualization during surgery. AR systems utilize advanced imaging techniques, such as preoperative CT/MRI scans, to project 3D virtual models onto the surgical field, assisting in accurate anatomical localization and surgical navigation. Advantages of augmented reality are:

a. **Enhanced visualization:** AR technology allows surgeons to visualize and interact with anatomical structures, implants, and surgical plans in real-time, improving accuracy and decision-making.
b. **Precise instrument guidance:** AR systems provide real-time visual guidance for instrument navigation, aiding in precise screw placement, tumor resection, or spinal deformity correction.
c. **Surgical education and training:** AR can facilitate surgical education and training by providing a virtual environment for surgical simulation and rehearsal.[32,33]

Both robotic assistance and augmented reality hold great potential in advancing spinal surgery by improving surgical accuracy, safety, and outcomes. However, it is important to note that these technologies are still evolving, and further research, clinical validation, and widespread adoption are needed to fully assess their impact on patient outcomes and to determine their optimal applications in the field of spinal surgery.

Clinical outcomes and complications

Literature review evaluating the efficacy and safety of percutaneous PSF demonstrates that percutaneous PSF has gained popularity as a minimally invasive technique for spinal stabilization. Numerous clinical studies have been conducted to evaluate the efficacy and safety of this procedure. Here is a review of selected clinical studies that have examined the outcomes of percutaneous PSF:

Tian NF et al.[5] in their meta-analysis compared the accuracy of pedicle screw insertion using different assisted methods, including percutaneous techniques. The study found that percutaneous pedicle screw insertion had comparable accuracy to open techniques and other assisted methods, with lower radiation exposure and reduced blood loss.

Titan F et al.[34] in their meta-analysis compared percutaneous PSF with open techniques for thoracolumbar fractures. The study found that percutaneous fixation resulted in similar clinical outcomes, including pain relief, functional improvement, and complication rates, with shorter surgical time, reduced blood loss, and shorter hospital stays compared to open techniques.

Phan K et al.[15] in their systematic review and meta-analysis compared percutaneous PSF with open techniques for thoracolumbar fractures. The study found that percutaneous fixation had similar clinical outcomes, including pain relief, functional improvement, and complication rates, with reduced blood loss, shorter hospital stays, and lower infection rates compared to open techniques.

These studies suggest that percutaneous PSF is associated with favorable patient outcomes, including comparable fusion rates and functional improvements compared to open techniques, along with benefits such as reduced complications, less blood loss, less radiation exposure, shorter surgical time and shorter hospital stays.

Evaluation of the impact of advancements on surgical outcomes

The advancements in percutaneous PSF techniques and technologies have the potential to significantly impact surgical outcomes in terms of accuracy, safety, patient recovery, and long-term success. Several studies have evaluated the impact of these advancements on surgical outcomes, and here we present a summary of their findings:

a. **Accuracy and screw placement:** The integration of navigation systems and robotic assistance has been shown to improve the accuracy of screw placement in percutaneous PSF. Tian NF et al.[14] conducted a meta-analysis comparing minimally invasive techniques with open surgery and found that navigation-guided percutaneous PSF had a higher accuracy rate and lower screw misplacement rate compared to conventional techniques.

b. **Surgical efficiency and time:** Robotic-assisted percutaneous pedicle screw fixation has shown potential in improving surgical efficiency and reducing operative time. Huang J et al.[23] reported in their review that robotic-assisted procedures were associated with shorter surgical times compared to conventional techniques. The advanced imaging and robotic arm assistance provided by robotic systems contribute to a more streamlined workflow, leading to potential time savings.
c. **Patient-specific planning and outcomes:** The use of patient-specific planning and customized screw trajectories in percutaneous pedicle screw fixation has shown promising outcomes. Preoperative planning of pedicle screw placement done using a 3D printed spine model has been shown to increase the accuracy of pedicle screw placement, with a 91% acceptance rate, according to a study by Xu W et al.[35] on patients with middle-upper thoracic spine trauma.
d. **Cement augmentation and stability:** The use of cement augmentation techniques, such as cement-augmented screws or vertebroplasty/kyphoplasty, has been shown to enhance screw stability and reduce complications, particularly in osteoporotic patients. Liu D et al.[28] conducted a systematic review and meta-analysis and reported that cement augmentation significantly improved screw pullout strength and reduced the risk of screw loosening or failure.

These studies highlight the positive impact of advancements in percutaneous pedicle screw fixation techniques on surgical outcomes, including improved accuracy, surgical efficiency, patient-specific planning, and enhanced screw stability.

Future directions and challenges

Percutaneous PSF has shown promising results in various studies and has gained popularity in clinical practice. However, ongoing research aims to further enhance the technique and explore potential areas for improvement. Here, we discuss some of the current research trends and areas for advancement:

a. **Navigation and Robotics:** Ongoing research focuses on the integration of navigation systems and robotic assistance to improve the accuracy and safety of percutaneous pedicle screw placement. These technologies offer real-time intraoperative guidance, improved screw trajectory planning, and reduced radiation exposure. Studies are investigating the effectiveness and cost-effectiveness of these advanced techniques in achieving optimal outcomes.[23]
b. **Augmented Reality (AR) and Virtual Reality (VR) :** The integration of AR and VR technologies holds great promise for enhancing surgical planning, visualization, and intraoperative guidance in percutaneous pedicle screw fixation. AR/VR can provide surgeons with real-time feedback, improved depth perception, and enhanced anatomical visualization. These technologies have the potential to improve accuracy, reduce surgical time, and facilitate the adoption of percutaneous techniques in more complex cases.[32,33]
c. **Biomechanical studies and Material innovation:** Further biomechanical studies and material innovation can contribute to the refinement of percutaneous PSF. Evaluating the biomechanical stability of percutaneous constructs under different loading conditions and studying the long-term effects on fusion rates and implant failure can provide valuable insights. Additionally, advancements in implant materials and coatings can enhance the osseointegration of percutaneous pedicle screws and improve their overall biomechanical performance.[26,28,29]
d. **Patient selection and Outcome measures:** Ongoing research emphasizes the importance of appropriate patient selection criteria and standardized outcome measures to evaluate the effectiveness of percutaneous PSF. Studies are investigating patient-specific factors, such as age, bone quality, and pathology, to determine the ideal candidates for percutaneous techniques. Additionally, research focuses on developing standardized outcome measures to assess clinical outcomes, fusion rates, functional outcomes, and complications consistently.[36]

e. **Cost-effectiveness analysis:** The economic impact of percutaneous PSF compared to open surgery is an important area of investigation. Studies are assessing the cost-effectiveness and cost-benefit of percutaneous techniques by considering factors such as operative time, length of hospital stay, reoperation rates, and overall healthcare costs. These analyses contribute to healthcare decision-making and resource allocation.[36]
f. **Learning curve and Technical expertise:** The percutaneous approach requires specialized training and technical expertise. Surgeons must become proficient in percutaneous techniques, including image guidance and navigation systems. The learning curve for percutaneous PSF can be steep, and initial cases may have longer operative times and higher complication rates. However, with experience, surgeons can overcome these challenges and achieve satisfactory outcomes.[37]
g. **Limited visualization and exposure:** Compared to open surgery, percutaneous techniques provide limited visualization and exposure of the surgical site. This can make it challenging to address complex anatomical variations, severe deformities, or multi-level pathologies. In such cases, open surgery may be more appropriate to achieve optimal correction and decompression.[28]
h. **Risk of screw misplacement:** The percutaneous technique relies heavily on fluoroscopic or intraoperative navigation guidance for screw placement. However, despite these aids, there is a risk of screw misplacement, especially in cases with complex anatomy or poor visualization. Screw malposition can lead to complications such as neurovascular injury, implant failure, or inadequate biomechanical stability.[5]
i. **Limited accessibility and versatility:** Percutaneous techniques may not be suitable for all patients and pathologies. Factors such as severe obesity, extensive scarring, or anatomical variations can limit the accessibility and feasibility of percutaneous pedicle screw fixation. Additionally, percutaneous techniques may have limitations in addressing complex spinal deformities or extensive multi-level pathologies, where open surgery may be more appropriate.
j. **Limited ability for direct decompression:** Percutaneous PSF focuses primarily on achieving spinal stability and fusion but may not directly address neural decompression. In cases where neural compression is a significant concern, additional minimally invasive or open decompression techniques may be necessary.
k. **Patient-specific planning and 3D printing:** Advancements in patient-specific planning and 3D printing technology can facilitate precise preoperative planning and improve screw trajectory accuracy in percutaneous PSF. The use of patient-specific anatomical models and guides, created through 3D printing, allows for personalized surgical approaches. This technology enables surgeons to anticipate anatomical variations, optimize screw placement, and improve overall surgical outcomes.[35]
l. **Advanced Navigation and Robotics:** Continued advancements in navigation systems and robotic assistance are expected to refine percutaneous pedicle screw fixation. Real-time feedback, improved accuracy, and enhanced precision offered by these technologies can further optimize screw placement and reduce the risk of complications. Additionally, the integration of artificial intelligence and machine learning algorithms may enable automated screw trajectory planning, improving surgical efficiency.[23]

Exploring these future directions in technique refinement and patient selection has the potential to enhance the efficacy, safety, and applicability of percutaneous PSF in clinical practice. These advancements aim to improve surgical outcomes, expand the patient population eligible for percutaneous techniques, and enhance the overall efficacy of the procedure.

Conclusions

Percutaneous PSF is shown to be an effective and safe technique for achieving spinal stability and fusion. Clinical studies have reported high fusion rates and satisfactory functional outcomes in various spinal pathologies, including degenerative conditions, trauma, and deformities. Comparative studies and meta-analyses have consistently shown that percutaneous techniques offer comparable or even superior outcomes compared to open surgical approaches. These studies have demonstrated shorter operative times, reduced blood loss, decreased postoperative pain, and faster recovery with percutaneous pedicle screws fixation. Patient outcomes following percutaneous PSF are generally positive. The majority of patients experience improved pain relief, restoration of spinal alignment, and functional recovery. However, careful patient selection is crucial to ensure optimal outcomes, and certain patient factors, such as obesity or extensive scarring, may limit the suitability of percutaneous techniques. While percutaneous PSF has demonstrated favorable outcomes, there are potential complications and limitations associated with the technique. These include screw misplacement, neurovascular injury, inadequate decompression, and limited versatility in addressing complex spinal deformities or multi-level pathologies. Advancements in image guidance, navigation systems, augmented reality, and robotic assistance are enhancing the accuracy and precision of percutaneous PSF. Patient-specific planning and 3D printing technologies are also facilitating personalized surgical approaches. Furthermore, ongoing research is exploring the impact of these advancements on surgical outcomes and expanding the eligibility of percutaneous techniques to more challenging cases. In short, percutaneous PSF is a valuable surgical technique for achieving spinal stabilization and fusion. It offers several advantages over open surgery, including reduced tissue trauma, faster recovery, and comparable outcomes. However, careful patient selection and surgeon expertise are crucial for optimizing outcomes and minimizing complications. Continued research and technological advancements hold promise for further refining the technique and expanding its applications in the future.

List of Abbreviations
PSF: Pedicle screw fixation;
CT: Computed Tomography;
MRI: Magnetic Resonance Imaging;
FEA: Finite Element Analysis;
AR: Augmented Reality;
VR: Virtual Reality.

References

1. Anand N, Baron EM, Khandehroo B, Kahwaty S. Long-term 2- to 5-year clinical and functional outcomes of minimally invasive surgery for adult scoliosis. Spine (Phila Pa 1976). 2013;38(18):1566-1575.
2. doi:10.1097/BRS.0b013e31829cb67a
3. Hu X, Ohnmeiss DD, Lieberman IH. Robotic-assisted pedicle screw placement: lessons learned from the first 102 patients. Eur Spine J. 2013;22(3):661-666. doi:10.1007/s00586-012-2499-1
4. Phan K, Rao PJ, Kam AC, Mobbs RJ. Minimally invasive versus open transforaminal lumbar interbody fusion for treatment of degenerative lumbar disease: systematic review and meta-analysis. Eur Spine J. 2015;24(5):1017-1030. doi:10.1007/s00586-015-3903-4
5. Kehr, P. Daniel H. Kim, Alexander R. Vaccaro, Richard G. Fessler, Kris E. Radcliff (Eds): Spinal instrumentation: challenges and solutions. Eur J Orthop Surg Traumatol 29, 237 (2019). doi:10.1007/s00590-018-2256-1
6. Tian NF, Huang QS, Zhou P, et al. Pedicle screw insertion accuracy with different assisted methods: a systematic review and meta-analysis of comparative studies. Eur Spine J. 2011;20(6):846-859. doi:10.1007/s00586-010-1577-5
7. Youssef JA, Orndorff DO, Patty CA, et al. Current status of adult spinal deformity. Global Spine J. 2013;3(1):51-62. doi:10.1055/s-0032-1326950
8. Cho W, Cho SK, Wu C. The biomechanics of pedicle screw-based instrumentation. J Bone Joint Surg Br. 2010;92(8):1061-1065.
9. doi:10.1302/0301-620X.92B8.24237
10. Christodoulou E, Chinthakunta S, Reddy D, et al. Axial pullout strength comparison of different screw designs: fenestrated screw, dual outer diameter screw and standard pedicle screw. Scoliosis. 2015;10:15. doi:10.1186/s13013-015-0039-6
11. Liljenqvist UR, Halm HF, Link TM. Pedicle screw instrumentation of the thoracic spine in idiopathic scoliosis. Spine (Phila Pa 1976). 1997;22(19):2239-2245. doi:10.1097/00007632-199710010-00008
12. Allen BL Jr, Ferguson RL. The Galveston technique for L rod instrumentation of the scoliotic spine. Spine (Phila Pa 1976). 1982;7(3):276-284. doi:10.1097/00007632-198205000-00014
13. Perna F, Borghi R, Pilla F, Stefanini N, Mazzotti A, Chehrassan M. Pedicle screw insertion techniques: an update and review of the literature. Musculoskelet Surg. 2016;100(3):165-169. doi:10.1007/s12306-016-0438-8
14. Liebschner MA, Rosenberg WS, Keaveny TM. Effects of bone cement volume and distribution on vertebral stiffness after vertebroplasty. Spine (Phila Pa 1976). 2001;26(14):1547-1554.
15. doi:10.1097/00007632-200107150-00009
16. Rajasekaran S, Vidyadhara S, Ramesh P, Shetty AP. Randomized clinical study to compare the accuracy of navigated and non-navigated thoracic pedicle screws in deformity correction surgeries. Spine (Phila Pa 1976). 2007;32(2):E56-E64. doi:10.1097/01.brs.0000252094.64857.ab
17. Tian NF, Xu HZ. Image-guided pedicle screw insertion accuracy: a meta-analysis. Int Orthop. 2009;33(4):895-903. doi:10.1007/s00264-009-0792-3

18. Phan K, Rao PJ, Mobbs RJ. Percutaneous versus open pedicle screw fixation for treatment of thoracolumbar fractures: Systematic review and meta-analysis of comparative studies. Clin Neurol Neurosurg. 2015;135:85-92. doi:10.1016/j.clineuro.2015.05.016
19. Wang J, Zhou Y, Zhang ZF, Li CQ, Zheng WJ, Liu J. Comparison of one-level minimally invasive and open transforaminal lumbar interbody fusion in degenerative and isthmic spondylolisthesis grades 1 and 2. Eur Spine J. 2010;19(10):1780-1784. doi:10.1007/s00586-010-1404-z
20. Santoni BG, Hynes RA, McGilvray KC, et al. Cortical bone trajectory for lumbar pedicle screws. Spine J. 2009;9(5):366-373. doi:10.1016/j.spinee.2008.07.008
21. Ahn Y, Lee SH, Park WM, Lee HY, Shin SW, Kang HY. Percutaneous endoscopic lumbar discectomy for recurrent disc herniation: surgical technique, outcome, and prognostic factors of 43 consecutive cases. Spine (Phila Pa 1976). 2004;29(16):E326-E332. doi:10.1097/01.brs.0000134591.32462.98
22. Kim YJ, Lenke LG, Cheh G, Riew KD. Evaluation of pedicle screw placement in the deformed spine using intraoperative plain radiographs: a comparison with computerized tomography. Spine (Phila Pa 1976). 2005;30(18):2084-2088. doi:10.1097/01.brs.0000178818.92105.ec
23. Wang MY, Mummaneni PV. Minimally invasive surgery for thoracolumbar spinal deformity: initial clinical experience with clinical and radiographic outcomes. Neurosurg Focus. 2010;28(3):E9. doi:10.3171/2010.1.FOCUS09286
24. Meyer B, Thomé C, Vajkoczy P, et al. Lumbar dynamic pedicle-based stabilization versus fusion in degenerative disease: a multicenter, double-blind, prospective, randomized controlled trial. J Neurosurg Spine. 2022;1-10. doi:10.3171/2022.2.SPINE21525
25. Xie Q, Zhang J, Lu F, Wu H, Chen Z, Jian F. Minimally invasive versus open Transforaminal lumbar Interbody fusion in obese patients: a meta-analysis. BMC Musculoskelet Disord. 2018;19(1):15. Published 2018 Jan 17. doi:10.1186/s12891-018-1937-6
26. Huang J, Li Y, Huang L. Spine surgical robotics: review of the current application and disadvantages for future perspectives. J Robot Surg. 2020;14(1):11-16. doi:10.1007/s11701-019-00983-6
27. Singh V, Mahajan R, Das K, Chhabra HS, Rustagi T. Surgical Trend Analysis for Use of Cement Augmented Pedicle Screws in Osteoporosis of Spine: A Systematic Review (2000-2017). Global Spine J. 2019;9(7):783-795. doi:10.1177/2192568218801570
28. Feng Q, Zhang Z, Wang D, Feng J. Comparing the efficacy and safety of cement-augmented fenestrated pedicle screws and conventional pedicle screw in surgery for spinal metastases: a retrospective comparative cohort study. Transl Cancer Res. 2022;11(12):4397-4408. doi:10.21037/tcr-22-2631
29. Kahaer A, Zhou Z, Maitirouzi J, et al. Biomechanical Investigation of the Posterior Pedicle Screw Fixation System at Level L4-L5 Lumbar Segment with Traditional and Cortical Trajectories: A Finite Element Study. J Healthc Eng. 2022;2022:4826507.
30. doi:10.1155/2022/4826507
31. Matsukawa K, Yato Y, Imabayashi H, Hosogane N, Asazuma T, Nemoto K. Biomechanical evaluation of the fixation strength of lumbar pedicle screws using cortical bone trajectory: a finite element study. J Neurosurg Spine. 2015;23(4):471-478. doi:10.3171/2015.1.SPINE141103
32. Liu D, Zhang XJ, Liao DF, et al. Biomechanical Comparison of Pedicle Screw Augmented with Different Volumes of Polymethylmethacrylate in Osteoporotic and Severely Osteoporotic Synthetic Bone Blocks in Primary Implantation: An Experimental Study. Biomed Res Int. 2016;2016:9627504. doi:10.1155/2016/9627504
33. Chen HC, Wu JL, Huang SC, et al. Biomechanical evaluation of a novel pedicle screw-based interspinous spacer: A finite element analysis. Med Eng Phys. 2017;46:27-32. doi:10.1016/j.medengphy.2017.05.004
34. Lonjon N, Chan-Seng E, Costalat V, Bonnafoux B, Vassal M, Boetto J. Robot-assisted spine surgery: feasibility study through a prospective case-matched analysis. Eur Spine J. 2016;25(3):947-955. doi:10.1007/s00586-015-3758-8
35. Lee NJ, Leung E, Buchanan IA, et al. A multicenter study of the 5-year trends in robot-assisted spine surgery outcomes and complications. J Spine Surg. 2022;8(1):9-20. doi:10.21037/jss-21-102
36. McCloskey K, Turlip R, Ahmad HS, Ghenbot YG, Chauhan D, Yoon JW. Virtual and Augmented Reality in Spine Surgery: A Systematic Review. World Neurosurg. 2023;173:96-107. doi:10.1016/j.wneu.2023.02.068
37. Burström G, Persson O, Edström E, Elmi-Terander A. Augmented reality navigation in spine surgery: a systematic review. Acta Neurochir (Wien). 2021;163(3):843-852. doi:10.1007/s00701-021-04708-3
38. Tian F, Tu LY, Gu WF, et al. Percutaneous versus open pedicle screw instrumentation in treatment of thoracic and lumbar spine fractures: A systematic review and meta-analysis. Medicine (Baltimore). 2018;97(41): e12535. doi:10.1097/MD.0000000000012535
39. Xu W, Zhang X, Ke T, Cai H, Gao X. 3D printing-assisted preoperative plan of pedicle screw placement for middle-upper thoracic trauma: a cohort study. BMC Musculoskelet Disord. 2017;18(1):348. doi:10.1186/s12891-017-1703-1
40. Lee TJ, Thomas AA, Grandhi NR, et al. Cost-effectiveness Applications of Patient-reported Outcome Measures (PROMs) in Spine Surgery. Clin Spine Surg. 2020;33(4):140-145. doi:10.1097/BSD.0000000000000982
41. Yang P, Chen K, Zhang K, Sun J, Yang H, Mao H. Percutaneous short-segment pedicle instrumentation assisted with O-arm navigation in the treatment of thoracolumbar burst fractures. J Orthop Translat. 2019;21:1-7. doi:10.1016/j.jot.2019.11.002.

Romanian Neurosurgery (2023) XXXVII (3): pp. 328-332
DOI: 10.33962/roneuro-2023-059
www.journals.lapub.co.uk/index.php/roneurosurgery

From low back pain to ochronosis. A case of late diagnosed alkaptonuria

Turkan Turgay, Erdal Yayla, Murat Baloglu

[1] SANKO University, Faculty of Medicine, Department of Physical Medicine and Rehabilitation, Gaziantep, TURKEY
[2] SANKO University, Faculty of Medicine, Department of Neurosurgery, Gaziantep, TURKEY
[3] Eskisehir City Hospital, Department of Neurosurgery, Eskisehir, TURKEY

Keywords
Alkaptonuria, homogentisate 1,2, dioxygenase, ochronosis, low back pain

Corresponding author:
Murat Baloglu

Eskişehir City Hospital, Department of Neurosurgery, Turkey

apopitozis@gmail.com

ABSTRACT

Background: Alkaptonuria is a rare metabolic autosomal recessive disorder. Its aetiology involves homogentisate 1,2 dioxygenase (HGD) deficiency resulting in homogentisic acid accumulation in the connective tissues (ochronosis). The classic triad of the disease is: i) homogentisic aciduria, ii) bluish-black pigmentation in tissues such as the sclera, cornea, cartilage and skin, and iii) degenerative arthropathy usually in the fourth decade of life.

Case: In this case report, we present a 41-year-old man with diffuse musculoskeletal pain and additional clinical features in tissues such as the ear and urinary tract who was diagnosed late (>30 years). He was diagnosed with alkaptonuria based on clinical findings and elevated urinary homogentisic acid levels.

Conclusion: This case report underscores the need for early diagnosis of alkaptonuria, which can help in managing symptoms and improving the quality of life of patients. Further research is needed to develop more targeted therapies for alkaptonuria, which can help slow down the progression of joint degeneration and improve overall patient.

INTRODUCTION

Alkaptonuria, also known as ochronosis, is a rare metabolic autosomal recessive disorder. The disease is characterized by accumulation of homogentisic acid in collagenous tissues such as cartilage. This is due to a deficiency in the HGD 1,2 enzyme involved in phenylalanine and tyrosine metabolism. In addition to bluish-black cartilage discoloration, progressive ochronotic arthropathy is observed. This may lead to calcification of tendons, ligaments and intervertebral discs, degeneration of large joints, inflammation and bone resorption. The diagnosis is based on clinical history, urinary homogentisic acid level, histopathologic and genetic examination. Although treatment options are limited, early diagnosis can prevent joint and spinal deformities and organ dysfunction. We aimed to present a patient with a delayed

Scan to access the online version

diagnosis of alkaptonuria with musculoskeletal, ear and urinary findings.

41-year-old male, university graduate, suffering from severe low back pain for the past three months was admitted to our clinic. It was learned that his complaints had been intermittent for 8 years. He described morning stiffness for 15-20 minutes and pain that subsided with rest. He had a 5-year history of a lumbar discectomy and right renal calculi. Family history revealed persistent dark urine and suspicion of rheumatologic disease in all siblings.

Clinical evaluation revealed a bluish coloration of the ear cartilages (Figure 1). Physical examination revealed decreased lumbar lordosis, thoracic kyphoscoliosis, limited and painful lumbar spine movements in all directions. The patient's lumbar Schober measurement was 2.0 cm.

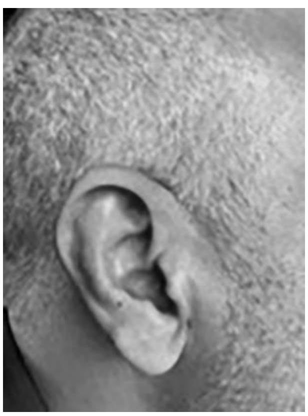


Figure 1. Bluish discoloration of the cartilage of the external ear.

Lumbar vertebral radiography showed intervertebral disc calcification, vacuum phenomenon, degenerative changes and a stone in the lower pole of the right kidney. Pelvic radiography showed bilateral sacroiliac joint narrowing, increased sclerosis and similar degenerative changes (Figure 2).

Figure 2. Narrowing of intervertebral disc spaces, disc calcifications, vacuum phenomenon, osteophytic degenerative changes on lumbar vertebral radiography, narrowing of sacroiliac and hip joint space, increased sclerosis on pelvic radiography.

Lumbar magnetic resonance imaging (MRI) showed mild distortion of the lumbar axis, kyphotic appearance at the thoracolumbar level, degenerative changes in the lumbar vertebrae with marginal osteophytes, Schmorl's nodules, narrowing of the disc spaces, disc protrusions compressing the talar sac, and degenerative changes in the facet joints (Figure 3). At the L4-L5 level, there was a left laminectomy operation defect and a paracentral protrusion at this level. This compressed the tibial sac and left L5 nerve root. When the patient's past imaging studies were examined, lower abdominal computed tomography showed prostatic calcification. The patient's urine turned dark brown when left to stand (Figure 4).

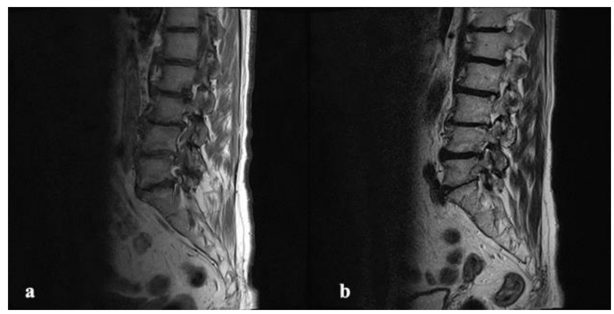


Figure 3. T1 and T2 weighted sagittal lumbar MRI image

Figure 4. Patient's dark brown urine at 0 and 18 hours.

A homozygous c.175delA/p.Ser59AlafsTer52 mutation was detected in the exons of the HGD gene of the patient in genetic analysis performed at an external center. Family screening was performed and it was found that his brother had a homozygous mutation and his sister had a heterozygous mutation. The patient was informed about alkaptonuria. Protein-free diets were started. He was referred to ophthalmology, cardiology, nephrology and urology for control and follow-up.The patient reapplied to our clinic 6 months later with right knee pain. Physical examination revealed prepatellar swelling and patellofemoral crepitation. Right knee MRI revealed thinning cartilage and osteonecrosis (grade 4 chondromalacia patella), the largest being 5 mm in size in the subchondral bone of the patella joint (Figure 5).

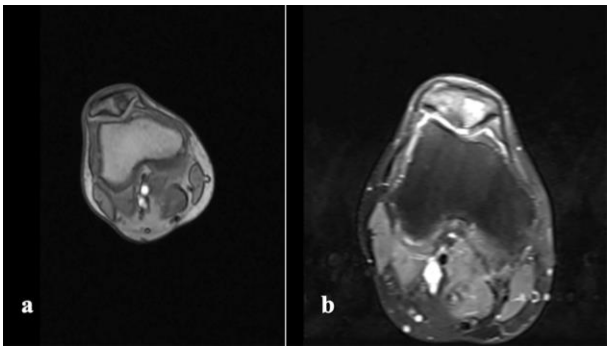


Figure 5. T1 and T2 weighted axial patellofemoral MRI image of the patient.

Characteristic clinical and radiologic findings suggested a possible diagnosis of alkaptonuria. The patient was started on 1 g/day vitamin C, analgesics, and nonsteroidal anti-inflammatory drugs. The patient was enrolled in a physical therapy program.

Discussion

The strength of this case is that it reflects the ability to classify a rare disease with a global prevalence of 1:100,000-250,000 (1) with possible suspicious and seemingly independent symptoms and signs. In addition, the patient underwent family screening with genetic examination after clinical diagnosis. In this way, the diagnosis of alkaptonuria was confirmed with a differential diagnosis of rheumatologic disease. Early treatment options were offered to his siblings.

The pathophysiology of alkaptonuria involves a deficiency in the enzyme homogentisate 1,2-dioxygenase. This enzyme deficiency leads to the accumulation of homogentisic acid in the body, which accumulates and deposits in bones and cartilage. This accumulation can lead to arthritis in affected joints and also cause a condition known as ochronosis, where the skin and other tissues darken 1.

As seen in our case, alkaptonuria is mostly asymptomatic in childhood and diagnosis is often delayed. Early diagnosis is key to preventing joint and spinal deformities and organ dysfunction. Many patients are also misdiagnosed with osteoarthritis. Differential diagnosis of ochronosis from diseases that cause disc calcification in the spine such as spondylosis, ankylosing spondylitis, pseudogout, hemochromatosis, amyloidosis and hypervitaminosis D should be made. Other features of alkaptonuria include aortic stenosis, vascular calcifications and a dark urine appearance due to HGA in the urine. Molecular genetic testing is not the gold standard for alkaptonuria diagnosis, as some polymorphisms may not cause the disease. Therefore, measurement of urinary HGA remains the classic diagnostic approach.

Patients are often not aware of the disease because of delayed referrals, misdiagnoses, and lack of awareness. However, when signs and symptoms reappeared later in life, the disease cannot be ignored. Genetic testing can provide the patient and their family members with disease information and help them make more informed medical and personal decisions.

There is currently no gold standard treatment for alkaptonuria. The development of disease sequelae can limit the available treatment options for alkaptonuria, including physical therapy, surgery, and analgesics. Nevertheless, dietary interventions may be effective in reducing phenylalanine and tyrosine intake that leads to homogentisic acid buildup within the body. By doing so, adverse effects associated with alkaptonuria could potentially be minimized through implementation of such measures. Nitisinone inhibits 4-hydroxyphenylpyruvate, the enzyme responsible for HGA production, and is currently approved for the treatment of hereditary tyrosinemia type 1. Studies have suggested that nitisinone may also be effective in reducing HGA levels in alkaptonuria patients. In several studies, nitisinone reduced urine and blood HGA levels by 95 percent 2. On the other hand, some argue that nitisinone may have potential benefits in the treatment of arthritis associated with alkaptonuria.

Although nitisinone did not show a significant improvement in a randomized clinical trial by Barconi et al (hip range of motion and other measures of musculoskeletal function), it is possible that different dosages or treatment regimens may yield better results 3. Treatment includes a low protein diet, ascorbic acid and lifestyle changes.

Dietary restriction of tyrosine and phenylalanine cannot reverse arthropathy, but may prevent clinical and radiologic progression. Ascorbic acid, which inhibits the enzyme catalyzing the oxidation of HGA to the polymer with collagen affinity, is given. However, its efficacy for ochronosis has not been proven 4. Therefore, although some treatments exist for alkaptonuria, more research is needed to develop effective therapies to manage the condition's joint-related complications.

Laura Groseanu et al. 5 initially diagnosed spondyloarthropathy; ankylosing spondylitis in a 55-year-old male patient with diffuse calcifications, who had onset of joint problems at an early age, morning stiffness, back and lower back pain, gradually increasing limitation of motion in the hip. However, intervertebral disc calcifications, history of kidney stones, aortic stenosis, pigmentation of the sclera, ear cartilage and hands suggested a late diagnosis of alkaptonuria.

Taşkıran et al., on the other hand, ordered urinalysis with a prediagnosis of HGA deficiency in a 71-year-old male patient who was incidentally observed to have blue pigments in the nose, cheeks and ears on routine physical examination, and who was diagnosed with hand arthrosis, dorsal kyphosis and bilateral hip prosthesis surgery due to early osteoarthritis 15 years ago on detailed physical examination and history, and made a diagnosis of delayed diagnosed alkaptonuria 6.

In a study including 58 patients with alkaptonuria, it was shown that only 12 (21%) of the patients were diagnosed before the age of 1 year. The remaining 32 patients (55%) were diagnosed because of dark urine, 26 patients (45%) were diagnosed because of chronic joint pain, and it was found that low back pain started before the age of 40 in 33 of 35 patients (94%) over the age of 40 7. As in our case, narrowing of the disc space, disc calcifications and disc fusion were observed at an early age. Although alkaptonuria shows spinal involvement similar to the clinical features of ankylosing spondylitis, kyphosis, loss of height and limitation in lumbar flexion, peripheral joint effusions, damage to the vertebrae and large joints differ 8. In the disease, joint symptoms typically begin in the third or fourth decade of life and progress until chronic pain leads to knee, hip or shoulder replacement; on average, this occurs at age 55 7.

Other manifestations include aortic or mitral valve calcification or insufficiency and sometimes aortic dilatation; kidney stones, prostate stones and hypothyroidism. Osteoarthritis, ankylosing spondylitis, Paget's disease, acute porphyria, valvular heart disease, rheumatoid arthritis, and melanosarcoma should be considered in the differential diagnosis. Although alkaptonuria does not seem to affect life expectancy, it has serious negative effects on quality of life. Complications include kidney, salivary gland, gallbladder and prostate stones, tendon and ligament tears, osteopenia and fractures, aortic valve calcification and stenosis, and amyloidosis 9.

Conclusions

As a result, the diagnosis and treatment of patients with alkaptonuric ochronosis, a rare inherited disorder, is highly complex. Early diagnosis is the key to the successful treatment of alkaptonuria. There is currently no effective treatment for the disease. In order to detect the disease as early as possible in the community, screening and genetic counseling should be carried out. A multidisciplinary approach aimed at improving quality of life and reducing morbidity should be the priority in the treatment of alkaptonuria patients.

References

1. Mistry JB, Bukhari M, Taylor AM. Alkaptonuria. Rare Dis. 2013;1:e27475.
2. Teke Kisa P, Eroglu Erkmen S, Bahceci H, et al. Efficacy of Phenylalanine- and Tyrosine-Restricted Diet in Alkaptonuria Patients on Nitisinone Treatment: Case Series and Review of Literature. Ann Nutr Metab. 2022;78(1):48-60.
3. Braconi D, Laschi M, Taylor AM, et al. Proteomic and redox-proteomic evaluation of homogentisic acid and ascorbic acid effects on human articular chondrocytes. J Cell Biochem 2010;111:922–32.
4. Rudebeck M, Scott C, Sireau N, Ranganath L. A patient survey on the impact of alkaptonuria symptoms as perceived by the patients and their experiences of receiving diagnosis and care. JIMD Rep. 2020;53(1):71-79.
5. Groseanu, Laura et al. A late and difficult diagnosis of ochronosis. J Med Life. 2010;3:(4):437-43.
6. Taşkıran E, Taş MY, Hışmi B, İnce FD, Ekmekçi S, Solakoğlu et al. A late diagnosis of alkaptonuria in an elderly patient. Eur Geriatr Med. 2017;8 (2):178-180.
7. Phornphutkul, Chanika et al. Natural history of alkaptonuria. N Engl J Med. 2002;347(26):2111-21.

8. Khan AH, Afroze B, Majid H, Zaidi Y, Jamil A, Jafri L. Musculoskeletal manifestations in Alkaptonuria: A cross-sectional study. Medicine. 2021;100(51):e28241.

9. Keller JM, Macaulay W, Nercessian OA, Jaffe IA. New developments in ochronosis: review of the literature. Rheumatol Int. 2005;25(2):81-85.

Romanian Neurosurgery (2023) XXXVII (3): pp. 333-339
DOI: 10.33962/roneuro-2023-060
www.journals.lapub.co.uk/index.php/roneurosurgery

Impact of gross-total resection versus other extent of resections for the overall survival of anaplastic astrocytoma. A systematic literature review

Dipak Chaulagain[1], Volodymyr Smolanka[1], Andriy Smolanka[1], Sunil Munakomi[2]

[1] Neurosurgery Department, Uzhhorod Regional Clinical Centre of Neurosurgery and Neurology, Uzhhorod National University, Uzhhorod, UKRAINE
[2] Neurosurgery Department, College of Medical Science, Teaching Hospital, Bharatpur, NEPAL

Keywords
anaplastic astrocytoma,
gross total resection,
subtotal resection,
overall survival

Corresponding author:
Dipak Chaulagain

Uzhhorod Regional Clinical Centre of Neurosurgery and Neurology, Uzhhorod National University, Uzhhorod, Ukraine

neurodipak@gmail.com

Scan to access the online version

ABSTRACT

Aim: We aim to assess the overall survival (OS) in patients with Anaplastic Astrocytoma (AA) undergoing gross total resection (GTR) as compared to partial resection (PR) subtotal resection (STR), or biopsy.

Methods: This systematic review followed Preferred Reporting Items for Systematic Review and Meta-analyses (PRISMA) guidelines. An electronic search from PubMed/Medline was conducted from their inception to 26th April 2022. We included AA patients undergoing any surgical intervention resulting in GTR, PR, STR or biopsy. We did not include letters, case reports, abstracts, conference papers, reviews, and studies where full text was unavailable. We included only those articles which were published in English.

Results: Five cohorts were used in this study. Two studies assessed OS in GTR, PR/STR and biopsy, while one study compared GTR and STR/biopsy. Another study was used to compare OS between GTR and local excision/STR, and another was used to assess the complications/benefits of these surgeries. Three studies showed a significant increase in OS in patients who underwent GTR compared to the other interventions, while one study showed a non-significant effect on OS (p= 0.249).

Conclusion: Our study concluded a significant increase in OS when patients with AA had GTR instead of STR, PR or biopsy. Although these surgeries might carry some disadvantages, GTR allows a more positive effect on neurological status. Still, more studies need to be conducted to assess the efficiency of these surgeries.

INTRODUCTION

Anaplastic Astrocytoma (AA) is a malignant primary brain tumor originating from astrocytic cells, with an annual incidence of 14 per 100,000 person-years [1]. AA is classified as a rapidly infiltrating WHO grade III tumor with increased cellularity, nuclear atypia, marked mitotic activity, presence of GFAP markers, and no microvascular proliferation

[2]. AA is associated with mutations of TP53, ATRX, and isocitrate dehydrogenase enzymes (IDH1 and IDH 2) [3]. IDH is a key metabolic enzyme in Kreb's cycle that catalyzes the conversion of isocitrate to α-ketoglutarate. Parenteral astrocytoma, endometrial cancer, and melanoma are risk factors for AA [4]. AA presents at a mean of 41 years of age with clinical symptoms including focal seizures, generalized seizures, progressive cognitive deterioration, headache, and blurred vision [5]. The gold standard for diagnosis of AA is a brain MRI which shows a hypointense lesion on T1-weighted images and a hyperintense lesion on T2-weighted images [6].

AA has a better prognosis and overall mean survival than grade IV glioblastoma. Favorable factors for a better prognosis include younger age (<50 years), peripherally located tumor in an accessible area, and intact neurological function [7]. In a study comparing prognostic markers in grade III gliomas, the median survival for patients <40 years was about 65.5 months, and 4.4 months for patients >60 years of age [8]. Tortosa et al. assessed clinical, and radiological factors with prognostic value in anaplastic gliomas. Factors associated with increased survival were age <49 years (P < 0.03), postoperative KPS score >80 (P < 0.007), and no ring enhancement (p = 0.03) [9].

The extent of tumor resection has improved survival in glioma patients. Gross total rection (GTR) and Subtotal resection (STR) are the surgical options for AA patients. Tortosa et al. found no improvement in survival when comparing GTR, STR, and biopsy in anaplastic glioma patients [9]. Another study compared biopsy with resection of the tumor in improving survival in malignant glioma patients. Analysis showed a significantly higher survival (p = 0.0015) in patients treated with resection, even after excluding patients with age >65 years or KPS <70 to eliminate the possible risk of bias [10]. Vuorinen et al. found significantly longer survival (171 vs 85 days, p = 0.035) in malignant glioma while comparing resection of the tumor and radiotherapy with biopsy and radiotherapy [11].

No review has assessed the role of the extent of surgical resection in improving overall survival in AA patients. Therefore, this review aims to compare GTR with any other partial surgical resection in improving overall survival in grade III AA patients

Method

1. Data sources and search strategy

This systematic review followed Preferred Reporting Items for Systematic Review and Meta analyses (PRISMA) guidelines [12]. An electronic search from PubMed/Medline was conducted from their inception to 26th April 2022 using the search string: (Astrocytoma OR grade III astrocytoma OR grade 3 astrocytoma OR Anaplastic Astrocytoma OR grade III glioma OR high-grade glioma) AND (Gross total resection OR subtotal OR partial resection) AND (extent). Furthermore, we manually screened the cited articles from previous meta-analyses, systematic review, cohort studies including retrospective or prospective studies, and other review articles to identify any suitable studies.

2. Study selection

All studies were included if they met the following eligibility criteria: which can be given as PECOS: 1) P (Population): Patients with Anaplastic Astrocytoma (Grade III tumor); 2) I (Intervention): Gross total resection; 3) C (Control): partial surgical resection intervention; 4) O (Outcome): relative overall survival between the Gross total resection vs any other partial surgical resection intervention 5) S (Studies): Randomized Controlled Trials and Cohort studies published in English.

3. Data extraction and quality assessment of studies

Two reviewers independently searched electronic databases. Studies searched were exported to the EndNote Reference Library software version 20.0.1 **(**Clarivate Analytics**)**, and duplicates were screened and removed.

Three reviewers did data extraction and quality assessment of included studies simultaneously and independently. Newcastle-Ottawa Scale (NOS) was used to assess the quality of the cohort studies. NOS score <6 was considered high risk for bias, 6-7 was moderate, and a score >7 was considered a low risk of bias (Table 1).

4. Statistical analysis

The results were formed by using a qualitative analysis. We intended to summarize the findings of the articles to synthesise the results. It was selected as it incorporated the results from all articles and highlighted all the differences and similarities between the study findings. However, since the included studies showed heterogeneity among their evaluation criteria and results, with not all studies containing appropriate findings to carry out a meta-analysis, we decided to do a systematic review instead.

RESULTS

1. Literature search results

The initial search of the three electronic databases yielded 860 potential studies. After exclusions based on titles and abstracts, the full texts of 136 studies were read for possible inclusion. A total of 5 studies remained for qualitative analysis. Figure 1 summarizes the results of our literature search.

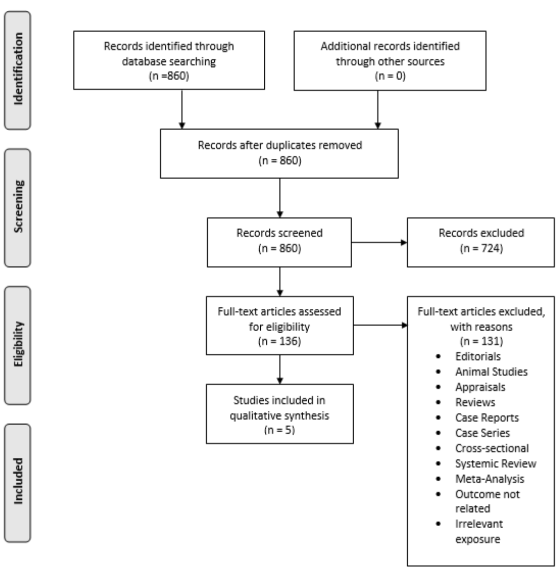


Figure 1. Prisma Flow Chart

2. Study characteristics

Table 2 provides the basic characteristics of included studies. All studies were cohort. We included 2,932 patients in our study. Mean percentage of females in our study was 43.5%. Three studies were from USA and one from Spain and another one from Germany. Mean age was 39.49 years. Further details of each study are provided in Table 1.

3. Quality assessment

All studies had a Low Risk of Bias (Table 1).

4. Result of qualitative analysis

We included five studies to assess the overall survival (OS) in patients, with Anaplastic Astrocytoma, when they underwent gross total resection (GTR) as compared to partial resection (PR) or subtotal resection (STR) or biopsy [13-17]. Nagy et al. and Capellades et al. assessed OS in GTR, PR/STR [13,16], and biopsy while McCrea et al. reported a comparison between GTR and STR/biopsy [14]. Padwal et al. compared OS between GTR and local excision/STR [15]. We included Groshev et al. to describe the complications/benefits of these surgeries [16].

Nagy et al. described OS in 2, 3 and 5 years with GTR giving 42% survival chance in 5 years compared to 29% and 9% 5 years survival in STR and biopsy, respectively [13]. Similarly, Padwal et al. reported a statistically significant increase in OS who had GTR instead of local excision/STR ($p < 0.0001$) [15]. McCrea et al also showed significant 2 years OS in Anaplastic Astrocytoma patients who underwent GTR rather than biopsy [14]. However, Capellades et al showed a statistically non-significant median OS chance between the three surgeries ($p = 0.249$) [16].

Groshev et al reported positive and negative outcomes of doing these surgeries. Neurological status was markedly improved in the patients, and seizures also ended after the surgery. However, few cases were seen where a transient motor weakness was resolved later. In addition, no cases of permanent speech impairment were recorded [17].

DISCUSSION

In this systematic review, we assess the effectiveness of GTR in comparison to partial resection in patients with grade III AA. The evidence from four studies suggested significantly better survival in the GTR group than the PR group. Groshev et al. highlighted the neurological outcomes in both groups. GTR group was associated with better neurological status; however, transient motor weakness was observed.

Neurological resection is the standard treatment approach for WHO grade III AA, followed by postoperative radiation therapy [18]. However, resection alone is considered the most effective treatment approach [19]. In 2016, Brown et al. evaluated the association of the extent of resection with survival in glioblastoma. The meta-analysis results showed a significant decrease in mortality at 1-year and 2-year in GTR compared to subtotal resection and biopsy [20]. Another systematic review and meta-analysis compared the GTR with supratotal resections in glioblastoma multiforme, suggested a median improvement of 10.5 months in supratotal resection, and showed a statistically significant 35% lower risk of mortality with supratotal resection than GTR. However, the study results were restricted by the clinical and methodological heterogeneity among the included articles [21]. Bond et al. found GTR as a superior technique to subtotal resection in adult pilocytic astrocytoma. In our literature search, no previously published meta-analysis or systematic review evaluated the OS and neurological outcome in patients with grade III AA who underwent GTR or STR [22].

Table 1. Quality assessment of cohorts

Study	Selection (Maximum 4)				Comparability (Maximum 2)	Outcome (Maximum 3)			Total score
	Representativeness of the Exposed Cohort	Selection of the Non-Exposed Cohort	Ascertainment of Exposure	Demonstration That Outcome of Interest Was Not Present at Start of Study	Comparability of Cohorts on the Basis of the Design or Analysis	Assessment of Outcome	Was Follow-Up Long Enough for Outcomes to Occur	Adequacy of Follow Up of Cohorts	
Nagy et al., 2009	1	1	1	1	1	1	1	1	8
McCrea et al., 2015	1	1	1	1	2	1	1	1	9
Padwal et al., 2016	1	1	1	1	2	1	1	1	9
Capellades et al., 2017	1	1	1	1	2	1	1	1	9
Groshev et al., 2017	1	1	1	1	1	1	1	1	8

Table 2. Baseline demographic characteristics

Study name	Year	Study type	Country	Duration	Number of patients (n)	Female (%)	Mean age (years)	Type of surgery	Net Risk of Bias	Other notes
Nagy et al.	2009	Cohort	Germany	Jan 1988-Jan 2007	104	45	42	Biopsy, Subtotal Resection, and Gross Total Resection	Low Risk	Median overall survival was 32 months after gross total resection, 36 months after subtotal resection, and 12 months after biopsy. Radiotherapy improved overall survival in grade III gliomas
McCrea et al.	2015	Cohort	USA	1988-2010	22	44	10.5	Biopsy, Subtotal Resection, and Gross	Low Risk	Patients treated with Gross total resection had a median overall survival of 3.4

								Total Resection		years, 1.6 years with subtotal resection, and 1.3 years with a biopsy. Females had a better overall survival
Padwal et al.	2016	Cohort	USA	1999-2010	2755	44.36	49.6	Biopsy, Subtotal Resection, Gross Total Resection, and no surgery	Low Risk	The median survival for patients who underwent gross total resection and subtotal resection was 64 and 24 months, respectively. Patients, less than 50 years of age had better survival
Capellades et al.	2017	Cohort	Spain	2005-2014	37	43.2	N/A*	Biopsy, Subtotal Resection, and Gross Total Resection	Low Risk	Among stage III patients, there was no significant difference in overall survival when resection was attempted (P=0.1). The statistical power may not have been sufficient to detect a difference with only 37 patients.
Groshev et al.	2017	Cohort	USA	Sep 2012-Feb 2015	14	41	55.86	Subtotal Resection, Near-total Resection, and Gross Total Resection	Low Risk	93% of patients had either gross total or near-gross total, which represents a favorable extent of resection. The extent of resection was better than the published outcomes of gross total near-total resection at 91%

N/A*= Not available

Groshev et al. included 18% WHO grade III astrocytoma patients, other resectable tumors; metastasis to the brain, gliomas, WHO grade I, grade II, and grade IV. They reported that GTR was associated with neurological status, including motor and sensory deficits improvement. Overall, 5 of 76 patients developed transient motor weakness, and one developed a transient speech deficit; however, deficits were resolved within two months postoperatively [17]. McCrea et al. found a median OS of 3.4 years in the GTR group compared to the median 1.6 years OS in STR and 1.3 years in the biopsy group. In addition, they found significantly longer OS in the female patient (8.1 years) in comparison to male patients (2.4 years). However, the OS was also significantly correlated with tumor location [14]. Similar results were reported by Padwal et al., they found a significant correlation between OS and extent of resection [15]. Nagy et al. found that median OS was 32 months after GTR, 36 months after STR, and 12 months after biopsy in grade III AA [13].

The results of the quantitative analysis suggested improvement in the OS in GTR; however, due to the heterogeneity among the results of included studies, strong results could not be predicted. In addition, only one study highlighted the postsurgical neurological outcomes. Therefore, more extensive trials and prolonged follow-ups are needed to evaluate OS associated with GTR and STR in grade III AA.

Limitations

Our study was limited by the following factors: (a) few articles were included in our manuscript; (b) only cohorts were included; (c) only one article discussed issues in the surgeries but was also not clear on which grade was more affected. However, these articles were pivotal in doing this review.

Conclusions

Our study concluded a significant increase in OS when patients with AA had GTR instead of STR, PR or biopsy. Although these surgeries might carry some disadvantages, GTR allows a more positive effect on neurological status. Still, more studies need to be conducted to assess the efficiency of these surgeries.

References

1. Ostrom QT, Patil N, Cioffi G, Waite K, Kruchko C, Barnholtz-Sloan JS. CBTRUS Statistical Report: Primary Brain and Other Central Nervous System Tumors Diagnosed in the United States in 2013-2017. Neuro Oncol. 2020 Oct 30;22(12 Suppl 2):iv1-iv96. doi: 10.1093/neuonc/noaa200. PMID: 33123732; PMCID: PMC7596247.
2. Louis DN, Perry A, Reifenberger G, von Deimling A, Figarella-Branger D, Cavenee WK, Ohgaki H, Wiestler OD, Kleihues P, Ellison DW. The 2016 World Health Organization Classification of Tumors of the Central Nervous System: a summary. Acta Neuropathol. 2016 Jun;131(6):803-20. doi: 10.1007/s00401-016-1545-1. Epub 2016 May 9. PMID: 27157931.
3. Watanabe K, Sato K, Biernat W, Tachibana O, von Ammon K, Ogata N, Yonekawa Y, Kleihues P, Ohgaki H. Incidence and timing of p53 mutations during astrocytoma progression in patients with multiple biopsies. Clin Cancer Res. 1997 Apr;3(4):523-30. PMID: 9815715.
4. Hemminki K, Li X, Collins VP. Parental cancer as a risk factor for brain tumors (Sweden). Cancer Causes Control. 2001 Apr;12(3):195-9. doi: 10.1023/a:1011275704954. PMID: 11405324.
5. Caccese M, Padovan M, D'Avella D, Chioffi F, Gardiman MP, Berti F, Busato F, Bellu L, Bergo E, Zoccarato M, Fassan M, Zagonel V, Lombardi G. Anaplastic Astrocytoma: State of the art and future directions. Crit Rev Oncol Hematol. 2020 Sep;153:103062. doi: 10.1016/j.critrevonc.2020.103062. Epub 2020 Jul 17. PMID: 32717623.
6. Cha S. Neuroimaging in neuro-oncology. Neurotherapeutics. 2009 Jul;6(3):465-77. doi: 10.1016/j.nurt.2009.05.002. PMID: 19560737; PMCID: PMC5084183.
7. Gorlia T, Delattre JY, Brandes AA, Kros JM, Taphoorn MJ, Kouwenhoven MC, Bernsen HJ, Frénay M, Tijssen CC, Lacombe D, van den Bent MJ. New clinical, pathological and molecular prognostic models and calculators in patients with locally diagnosed anaplastic oligodendroglioma or oligoastrocytoma. A prognostic factor analysis of European Organisation for Research and Treatment of Cancer Brain Tumour Group Study 26951. Eur J Cancer. 2013 Nov;49(16):3477-85. doi: 10.1016/j.ejca.2013.06.039. Epub 2013 Jul 26. PMID: 23896377.
8. Perry A, Jenkins RB, O'Fallon JR, Schaefer PL, Kimmel DW, Mahoney MR, Scheithauer BW, Smith SM, Hill EM, Sebo TJ, Levitt R, Krook J, Tschetter LK, Morton RF, Buckner JC. Clinicopathologic study of 85 similarly treated patients with anaplastic astrocytic tumors. An analysis of DNA content (ploidy), cellular proliferation, and p53 expression. Cancer. 1999 Aug 15;86(4):672-83. doi: 10.1002/(sici)1097-0142(19990815)86:4<672::aid-cncr17>3.0.co;2-g. PMID: 10440696.
9. Tortosa A, Viñolas N, Villà S, Verger E, Gil JM, Brell M, Caral L, Pujol T, Acebes JJ, Ribalta T, Ferrer I, Graus F. Prognostic implication of clinical, radiologic, and pathologic features in patients with anaplastic gliomas. Cancer. 2003 Feb 15;97(4):1063-71. doi: 10.1002/cncr.11120. PMID: 12569607.
10. Laws ER, Shaffrey ME, Morris A, Anderson FA Jr. Surgical management of intracranial gliomas--does radical resection improve outcome? Acta Neurochir Suppl. 2003;85:47-53. doi: 10.1007/978-3-7091-6043-5_7. PMID: 12570137.

11. Vuorinen V, Hinkka S, Färkkilä M, Jääskeläinen J. Debulking or biopsy of malignant glioma in elderly people - a randomised study. Acta Neurochir (Wien). 2003 Jan;145(1):5-10. doi: 10.1007/s00701-002-1030-6. PMID: 12545256.
12. Hutton B, Salanti G, Caldwell DM, et al. The PRISMA extension statement for reporting of systematic reviews incorporating network meta-analyses of health care interventions: checklist and explanations. Ann Intern Med. 2015;162(11):777-784. doi:10.7326/M14-238
13. Nagy M, Schulz-Ertner D, Bischof M, et al. Long-term outcome of postoperative irradiation in patients with newly diagnosed WHO grade III anaplastic gliomas. Tumori. 2009;95(3):317-324.
14. McCrea HJ, Bander ED, Venn RA, et al. Sex, Age, Anatomic Location, and Extent of Resection Influence Outcomes in Children With High-grade Glioma. Neurosurgery. 2015;77(3):443-453. doi:10.1227/NEU.0000000000000845
15. Padwal JA, Dong X, Hirshman BR, Hoi-Sang U, Carter BS, Chen CC. Superior Efficacy of Gross Total Resection in Anaplastic Astrocytoma Patients Relative to Glioblastoma Patients. World Neurosurg. 2016;90:186-193. doi:10.1016/j.wneu.2016.02.078
16. Capellades J, Puig J, Domenech S, et al. Is a pretreatment radiological staging system feasible for suggesting the optimal extent of resection and predicting prognosis in glioblastoma? An observational study. J Neurooncol. 2018;137(2):367-377. doi:10.1007/s11060-017-2726-z
17. Groshev A, Padalia D, Patel S, et al. Clinical outcomes from maximum-safe resection of primary and metastatic brain tumors using awake craniotomy. Clin Neurol Neurosurg. 2017;157:25-30. doi:10.1016/j.clineuro.2017.03.017
18. Walker, M. D., Strike, T. A., & Sheline, G. E. (1979). An analysis of dose-effect relationship in the radiotherapy of malignant gliomas. International journal of radiation oncology, biology, physics, 5(10), 1725–1731. https://doi.org/10.1016/0360-3016(79)90553-4
19. Bauman, G. S., Ino, Y., Ueki, K., Zlatescu, M. C., Fisher, B. J., Macdonald, D. R., Stitt, L., Louis, D. N., & Cairncross, J. G. (2000). Allelic loss of chromosome 1p and radiotherapy plus chemotherapy in patients with oligodendrogliomas. International journal of radiation oncology, biology, physics, 48(3), 825–830. https://doi.org/10.1016/s0360-3016(00)00703-3
20. Brown, T. J., Brennan, M. C., Li, M., Church, E. W., Brandmeir, N. J., Rakszawski, K. L., Patel, A. S., Rizk, E. B., Suki, D., Sawaya, R., & Glantz, M. (2016). Association of the Extent of Resection With Survival in Glioblastoma: A Systematic Review and Meta-analysis. JAMA oncology, 2(11), 1460–1469. https://doi.org/10.1001/jamaoncol.2016.1373
21. Jackson, C., Choi, J., Khalafallah, A. M., Price, C., Bettegowda, C., Lim, M., Gallia, G., Weingart, J., Brem, H., & Mukherjee, D. (2020). A systematic review and meta-analysis of supratotal versus gross total resection for glioblastoma. Journal of neuro-oncology, 148(3), 419–431. https://doi.org/10.1007/s11060-020-03556-y
22. Bond, K. M., Hughes, J. D., Porter, A. L., Orina, J., Fang, S., & Parney, I. F. (2018). Adult Pilocytic Astrocytoma: An Institutional Series and Systematic Literature Review for Extent of Resection and Recurrence. World neurosurgery, 110, 276–283. https://doi.org/10.1016/j.wneu.2017.11.102

Romanian Neurosurgery (2023) XXXVII (3): pp. 340-348
DOI: 10.33962/roneuro-2023-061
www.journals.lapub.co.uk/index.php/roneurosurgery

Symptomatic calcified chronic subdural hematoma treated surgically. A case report with review of the literature

Pradhumna Kumar Yadav[1], Ahtesham Khizar[2], Seeneen Bukhari[3], Somraj Lamichane[1]

[1] National Trauma Center, Kathmandu, NEPAL
[2] Punjab Institute of Neurosciences, Lahore, PAKISTAN
[3] Ameer-ud-din Medical College, Lahore, PAKISTAN

ABSTRACT

Background: Calcified chronic subdural hematoma (CSDH) is unusual. The majority of calcified CSDHs are found near the convexity, and the amount of calcification varies greatly. The progression of calcification in a CSDH is unknown. Seizures, mental and physical impairment, hemiparesis, and gait abnormalities are some of the symptoms; nevertheless, some people are asymptomatic. Surgical intervention is preferred for a calcified CSDH that is expanding. However, there is no agreement on surgical therapy for asymptomatic calcified CSDHs.

Case Presentation: A 69-year-old man was reported to have had behavioural abnormalities and seizures in the past couple of years. He had a three-year history of minor head trauma. On clinical examination, he appeared confused and had no hemiparesis. His Computed Tomography (CT) brain revealed a calcified frontoparietal CSDH on the left side. A calcified CSDH was discovered in his brain by Magnetic Resonance Imaging (MRI) as well. This patient underwent a left-sided frontoparietal craniotomy, and the calcified CSDH was removed to the greatest extent possible. It was firmly attached to the underlying surface of the brain parenchyma. The postoperative period was straightforward, and the patient's behavioural alterations improved noticeably.

Conclusions: We described a patient who had a rare, persistent, calcified CSDH that was successfully removed, leading to a noticeable recovery. Based on our assessment of the literature and our patient's experience, we believe surgical treatment for calcified CSDH is viable and typically results in neurological improvement.

Keywords
chronic subdural hematoma, calcification, ossification, traumatic brain injury, craniotomy

Corresponding author:
Ahtesham Khizar

Punjab Institute of Neurosciences, Lahore, Pakistan

arwain.6n2@gmail.com

Scan to access the online version

INTRODUCTION

Chronic subdural hematoma (CSDH) usually results from trivial head trauma or from use of anticoagulants and is more common in older age. Symptoms take weeks or months to appear. While CSDH is very common, calcified CSDH is a rare entity with some patients being asymptomatic and others having neurological symptoms such as balance problems, weakness or numbness, confusion, dizziness, behavioural abnormalities, and seizures. The mechanism of calcification in CSDH is likely to be multifactorial, involving both vascular and metabolic factors.[1,2]

CSDH is surgically treated to reduce pressure on the brain. Burr hole drainage is a popular surgical technique, and a drain is typically left in place to let the blood to drain out; a craniotomy is seldom ever necessary for a straightforward CSDH. The neurosurgical removal of the calcified hematoma frequently necessitates craniotomy, which can be difficult due to the calcified hematoma's adhesion to the meninges and underlying brain tissue. As calcified hematoma removal is difficult due to calcified hematoma adhesion to the underlying cortex, surgery is only suggested when acute or progressive neurological symptoms emerge.[3,4] According to reports, the incidence of calcified CSDH ranges from 0.3% to 2.7%.[1,4-11] Despite the fact that surgical treatment for the CSDH is largely acknowledged, there is still some debate over its application.[11]

Case presentation

A 69-year-old man went to the outpatient clinic with concerns of behavioural changes and seizures during the previous couple of years. He had a history of trivial head trauma three years prior. He had also been seeing a psychiatrist for his behaviour troubles. He was on Levetiracetam for seizures. He seemed disoriented and had no hemiparesis on clinical evaluation. His Computed Tomography (CT) brain plain revealed a left sided frontoparietal calcified CSDH, as indicated in Figures 1 A & B.

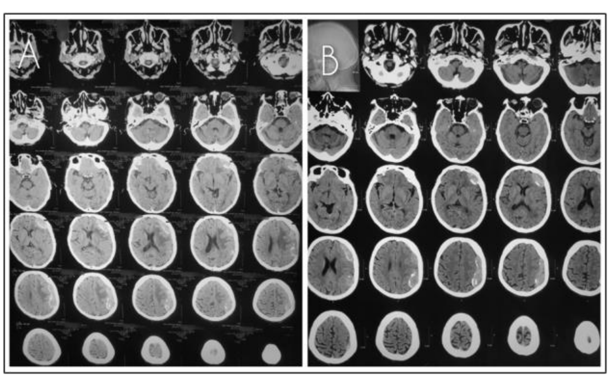


Figure 1. A & **B**: CT scan brain plain showing left sided frontoparietal calcified chronic subdural hematoma.

His Magnetic Resonance Imaging (MRI) brain revealed a calcified CSDH, as seen in Figures. 2 A, B, C, and D. This patient had left sided frontoparietal craniotomy after counselling and signed informed permission, and the calcified CSDH was expunged as much as feasible. It was rigorously adhered to the underlying surface of the brain parenchyma and so that part did not dislodge. The postoperative phase was simplistic, and the patient's behavioural changes improved significantly.

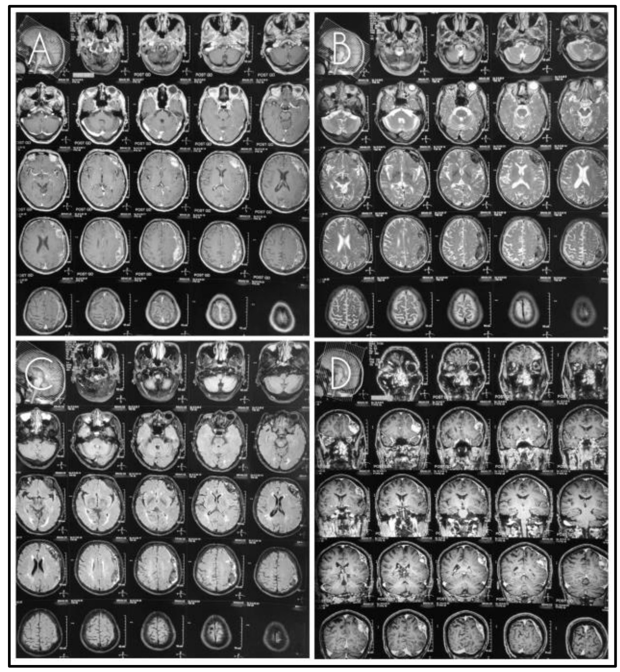


Figure 2. A: MRI Axial T1 weighted image with contrast, **B**: MRI Axial T2 weighted image, **C**: MRI FLAIR image, **D**: MRI Coronal T1 weighted image with contrast showing left sided frontoparietal calcified chronic subdural hematoma

Discussion

CSDH is one of the most common consequences of mild head trauma. Coagulation disorders, therapeutic anticoagulant use, intracranial hypotension (secondary to overdrainage in shunted patients), chronic alcoholism, vascular malformations, and primary and metastatic tumours may all play a role in the aetiology of CSDH.[12] An "Armoured brain" or "Matrioska head" is one in which the surface of the brain is covered by a calcified wall. Despite being thin, this calcified region is said to have strong adhesions to the dura mater and the surface of the brain. The risk of harming the underlying cortex prevents regular surgical removal of the calcification. It is advised that patients who are elderly, asymptomatic, and whose neurological state has not changed suddenly or significantly be monitored. It has been found that symptomatic and young patients require surgical treatment.[4,6,7,10,12,13,14]

The term "calcified CSDH" refers to the 0.5% to 2.0% of CSDH patients that experience calcification.[15] Von Rokitansky wrote about calcified CSDH as an autopsy finding in 1884.[1,10] The first instance of an armoured brain or calcified CSDH was described by Goldhan in 1930.[1,10,16,17] The mechanism of calcification development in CSDH is not established.[1,10,18] However, the hematoma may develop progressively from hyalinization to calcification and, eventually, ossification due to tissue irritation. Calcification after a hemorrhage often takes 6 months to several years to occur. Poor circulation and absorption into the subdural space, vascular thrombosis, an inherent metabolic tendency to calcification, the presence of a hematoma in the subdural space for an extended period of time, stagnant blood due to adequate arterial supply but insufficient venous return, a thick connective tissue membrane, and other local factors are thought to contribute to the development of CSDH calcification.[1,10]

Table 1: A comprehensive literature review through the PubMed and Google Scholar databases showing cases of calcified/ossified CSDH from 2000 to 2023.

S.no.	Authors' name	Year reported	No. of cases reported	Gender/Age (y=years m=months)	Cause	Treatment offered
1.	Imaizumi S et al.[1]	2001	5	i.M/3.8y ii.M/2.6y iii.M/78y iv.M/69y v.M/77y	i.Shunting ii.Trauma + Shunting iii.Unknown iv.Bleeding diathesis v.Trauma	i.Craniotomy ii.Craniotomy iii.Craniotomy iv.Craniotomy v.Craniotomy
2.	Gotoh M et al.[20]	2001	1	M/54y	Lymphoma	Craniotomy
3.	Narsinghani U et al.[21]	2002	1	M/5y	Unknown	Unknown
4.	Sgaramella E et al.[3]	2002	1	Unknown	Unknown	Conservative
5.	Wakamoto H et al.[22]	2003	1	M/46y	Trauma	Craniotomy
6.	Al Wohaibi M et al.[23]	2003	1	M/10m	Shunting	Conservative
7.	Jong-Soo P et al.[24]	2003	1	M/37y	Unknown	Craniotomy
8.	Moon HG et al.[25]	2003	1	M/67y	Unknown	Craniotomy
9.	Yang HZ et al.[11]	2004	1	M/59y	Unknown	Craniotomy
10.	Sato K et al.[26]	2005	1	M/50y	Unknown	Craniotomy
11.	He XS et al.[27]	2005	1	M/15y	Shunting	Craniotomy
12.	Per H et al.[6]	2006	1	M/3.5y	Trauma	Craniotomy
13.	Tatli M et al.[18]	2006	1	M/16y	Unknown	Craniotomy
14.	Dimogerontas G et al.[28]	2006	1	M/43y	Shunting	Conservative
15.	Moon KS et al.[13]	2007	1	M/47y	Unknown	Craniotomy

16.	Evans SJ[29]	2007	1	F/21y	Shunting	Conservative
17.	Dammers R et al.[4]	2007	1	M/67y	Trauma	Craniotomy
18.	Amr R et al.[30]	2008	1	F/30y	Shunting	Conservative
19.	Papanikolaou PG et al.[31]	2008	1	M/33y	Shunting + Trauma	Conservative
20.	Kaplan M et al.[12]	2008	1	M/22y	Trauma	Craniotomy
21.	Galldiks N et al.[7]	2010	1	M/86y	Trauma	Burr holes
22.	Rao Z et al.[9]	2010	1	M/75y	Unknown	Craniotomy
23.	Oda S et al.[19]	2010	1	F/32y	Unknown	Craniotomy
24.	Petraglia AL et al.[32]	2010	1	F/38y	Shunting	Conservative
25.	Turgut M et al.[33]	2010	1	F/30y	Trauma	Craniotomy
26.	Akhaddar A et al.[34]	2011	1	M/7y	Shunting	Conservative
27.	Rahman A et al.[35]	2012	1	M/65y	Unknown	Craniotomy
28.	Juan WS et al.[36]	2012	2	i.M/10y ii.F/35y	i.Shunting ii.Shunting + Aneurysmal bleed	i.Craniotomy ii.Craniotomy
29.	Sugita Y et al.[37]	2012	1	M/77y	Lymphoproliferative disorder + Trauma	Craniotomy
30.	Taha MM et al.[10]	2012	1	M/12y	Shunting	Conservative
31.	Pappamikail L et al.[38]	2013	1	M/73y	Unknown	Craniotomy
32.	Tandon V et al.[39]	2013	1	F/54y	Trauma	Conservative
33.	Garg K et al.[67]	2013	1	M/24y	Shunting	Conservative
34.	Chaudhry FS et al.[40]	2013	1	M/47y	Arrested hydrocephalus	Conservative
35.	Goyal PK et al.[41]	2013	1	F/15y	Unknown	Craniotomy
36.	Salunke P et al.[14]	2013	1	M/15y	Shunting	Burr holes
37.	Ito M et al.[42]	2014	1	M/72y	Unknown	Craniotomy
38.	Cai J et al.[43]	2014	1	M/77y	Glioblastoma	Craniotomy
39.	Arán-Echabe E et al.[44]	2014	1	Unknown	Unknown	Unknown
40.	Djoubairou BO et al.[8]	2015	1	M/22y	Shunting	Conservative
41.	Gupta SK et al.[45]	2015	1	M/30y	Shunting	Craniotomy

42.	Yang X et al.[2]	2015	1	F/54y	Infection	Craniotomy
43.	Siddiqui SA et al.[17]	2016	1	M/30y	Shunting	Craniotomy
44.	Li H et al.[46]	2017	1	M/61y	Trauma	Craniotomy
45.	Viozzi I et al.[47]	2017	1	F/15y	Shunting	Conservative
46.	Keser N et al.[48]	2017	1	F/35y	Trauma	Craniotomy
47.	Chan ZW et al.[66]	2018	1	Unknown	Shunting + Glioblastoma	Craniotomy
48.	Satyarthee GD et al.[49]	2018	1	M/8y	Shunting	Burr hole Craniostomy
49.	Qin G et al.[50]	2019	1	M/58y	Skull angiosarcoma	Craniotomy
50.	Fang J et al.[51]	2019	1	F/7y	Trauma	Craniotomy
51.	Ding H et al.[16]	2019	1	M/15y	Postoperative	Conservative
52.	Liu X et al.[52]	2019	1	M/46y	Lymphoproliferative disorder	Craniectomy
53.	Tian W et al.[53]	2019	1	M/49y	Unknown	Craniotomy
54.	Turgut M et al.[54]	2019	1	M/59y	Trauma	Craniotomy
55.	Rong J et al.[55]	2020	1	M/46y	Trauma	Craniotomy
56.	Snopko P et al.[5]	2020	1	F/81y	Unknown	Craniotomy
57.	Songnatsiri P et al.[56]	2020	1	M/83y	Trauma	Craniotomy
58.	Zhang S et al.[57]	2020	1	M/60y	Unknown	Craniotomy
59.	Marini A et al.[65]	2020	1	M/15y	Shunting	Craniotomy
60.	Bhardwaj S et al.[58]	2020	1	M/25y	Shunting	Craniotomy
61.	Pakrasi R et al.[59]	2021	1	M/75y	Unknown	Craniotomy
62.	Prasad PK et al.[60]	2021	1	M/50y	Unknown	Craniotomy
63.	Chaulagain D et al.[15]	2022	1	M/65y	Unknown	Craniotomy
64.	Bett D[61]	2022	1	M/96y	Unknown	Craniotomy
65.	Girotto AL[62]	2022	1	M/62	Unknown	Craniotomy
66.	Wang H et al.[63]	2023	1	M/69y	Unknown	Craniotomy
67.	Mansour M et al.[64]	2023		F/34y	Trauma	Craniotomy

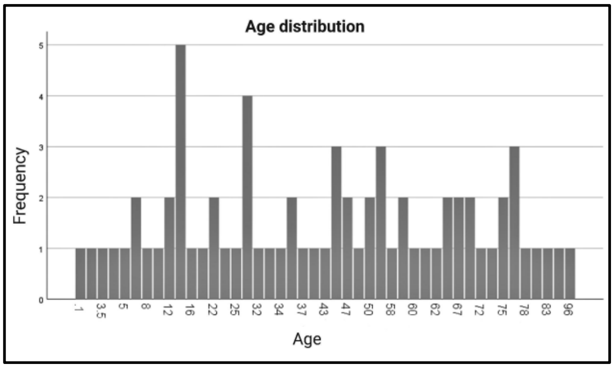


Figure 3: Age distribution among the reviewed cases.

Table 1: A comprehensive

Total no. of cases	72	
Mean age (years)	42.57 ± 25.62 SD (mini. 10m, max. 96y)	
Gender distribution	Male	55 (76.4%)
	Female	14 (19.4%)
	Unknown	3 (4.2%)
Cause distribution	Shunting	21 (29.2%)
	Trauma	16 (22.2%)
	Shunting + Trauma	2 (2.8%)
	Tumour related	3 (4.2%)
	Bleeding diathesis	1 (1.4%)
	Lymphoproliferative disorder	2 (2.8%)
	Arrested hydrocephalus	1 (1.4%)
	Infection	1 (1.4%)
	Postoperative	1 (1.4%)
	Unknown	24 (33.3%)
Treatment offered	Craniotomy	51 (70.8%)
	Craniectomy	1 (1.4%)
	Craniostomy	1 (1.4%)
	Burr holes	2 (2.8%)
	Conservative	15 (20.8%)
	Unknown	2 (2.8%)

Conclusions

We presented a patient who had an uncommon, calcified CSDH that was successfully removed, resulting in a satisfactory recovery. Based on the literature analysis and our patient's experience, we feel that surgical therapy for calcified CSDH is viable and commonly results in neurological improvement.

Abbreviations

CSDH: Chronic subdural hematoma;
CT: Computed Tomography;
MRI: Magnetic Resonance Imaging;
SPSS: Statistical Package for Social Sciences.

References

1. Imaizumi S, Onuma T, Kameyama M, Naganuma H. Organized chronic subdural hematoma requiring craniotomy--five case reports. Neurol Med Chir (Tokyo). 2001;41(1):19-24. doi:10.2176/nmc.41.19
2. Yang X, Qian Z, Qiu Y, Li X. Diagnosis and Management of Ossified Chronic Subdural Hematoma. J Craniofac Surg. 2015;26(6):e550-e551. doi:10.1097/SCS.0000000000002025
3. Sgaramella E, Sotgiu S, Miragliotta G, Fotios Kalfas Crotti FM. "Matrioska head". Case report of calcified chronic subdural hematoma. J Neurosurg Sci. 2002;46(1):28-31.
4. Dammers R, ter Laak-Poort MP, Maas AI. Neurological picture. Armoured brain: case report of a symptomatic calcified chronic subdural haematoma. J Neurol Neurosurg Psychiatry. 2007;78(5):542-543. doi:10.1136/jnnp.2006.108662
5. Snopko P, Kolarovszki B, Opsenak R, Hanko M, Benco M. Chronic calcified subdural hematoma - case report of a rare diagnosis. Biomed Pap Med Fac Univ Palacky Olomouc Czech Repub. 2020;164(2):209-212. doi:10.5507/bp.2019.041
6. Per H, Gümüş H, Tucer B, Akgün H, Kurtsoy A, Kumandaş S. Calcified chronic subdural hematoma mimicking calvarial mass: a case report. Brain Dev. 2006;28(9):607-609. doi:10.1016/j.braindev.2006.03.012
7. Galldiks N, Dohmen C, Neveling M, Fink GR, Haupt WF. A giant bilateral calcified chronic subdural hematoma. Neurocrit Care. 2010;12(2):272-273. doi:10.1007/s12028-009-9303-z

8. Djoubairou BO, Gazzaz M, Dao I, Mostarchid BE. Chronic calcified extradural and subdural hematoma following a ventriculoperitoneal shunt placement. Neurol India. 2015;63(2):282-283. doi:10.4103/0028-3886.156316

9. Rao Z, Li J, Yin H, You C. Huge calcified chronic subdural haematoma. Br J Neurosurg. 2010;24(6):722-723. doi:10.3109/02688697.2010.520763

10. Taha MM. Armored brain in patients with hydrocephalus after shunt surgery: review of the literatures. Turk Neurosurg. 2012;22(4):407-410. doi:10.5137/1019-5149.JTN.4782-11.1

11. Yang HZ, Tseng SH, Chen Y, Lin SM, Chen CJ. Calcified chronic subdural hematoma-Case report. Tzu Chi Med J. 2004 Aug 1;16(4):261-5. doi:10.6440/TZUCMJ.200408.0261

12. Kaplan M, Akgün B, Seçer HI. Ossified chronic subdural hematoma with armored brain. Turk Neurosurg. 2008;18(4):420-424.

13. Moon KS, Lee JK, Kim TS, et al. Contralateral acute subdural hematoma occurring after removal of calcified chronic subdural hematoma. J Clin Neurosci. 2007;14(3):283-286. doi:10.1016/j.jocn.2005.11.016

14. Salunke P, Aggarwal A, Madhivanan K, Futane S. Armoured brain due to chronic subdural collections masking underlying hydrocephalus. Br J Neurosurg. 2013;27(4):524-525. doi:10.3109/02688697.2013.769043

15. Chaulagain D, Munakomi S, Bhattarai B, Shah SB. Calcified Chronic Subdural Hematoma: A Case Report. Eastern Green Neurosurgery. 2022 Dec 31;4(1). doi:10.3126/egn.v4i1.51008

16. Ding H, Liao S, Liu L. A case with a postoperative rapidly calcified subdural hematoma. Br J Neurosurg. 2019;1-3. doi:10.1080/02688697.2019.1689230

17. Siddiqui SA, Singh PK, Sawarkar D, Singh M, Sharma BS. Bilateral Ossified Chronic Subdural Hematoma Presenting as Diabetes Insipidus-Case Report and Literature Review. World Neurosurg. 2017;98:520-524. doi:10.1016/j.wneu.2016.11.031

18. Tatli M, Guzel A, Altinors N. Spontaneous acute subdural hematoma following contralateral calcified chronic subdural hematoma surgery: an unusual case. Pediatr Neurosurg. 2006;42(2):122-124. doi:10.1159/000090468

19. Oda S, Shimoda M, Hoshikawa K, Shiramizu H, Matsumae M. Organized chronic subdural haematoma with a thick calcified inner membrane successfully treated by surgery: a case report. Tokai J Exp Clin Med. 2010;35(3):85-88. PMID: 21319032.

20. Gotoh M, Tsuno K, Handa A, Nishiura T, Ishimitsu H, Nishida A. No Shinkei Geka. 2001;29(3):259-264. PMID: 11321796

21. Narsinghani U, Burson G, Schmidt MB, Anand KS. Case of the month. Symptomatic chronic calcified subdural hematoma in a 5-year-old child. J Ark Med Soc. 2002;98(9):300-303. PMID: 11881264

22. Wakamoto H, Miwa T, Orii M, Miyazaki H, Ishiyama N. No Shinkei Geka. 2003;31(3):269-273. PMID: 12684980

23. Al Wohaibi M, Russell N, Al Ferayan A. A baby with an armoured brain. CMAJ. 2003;169(1):46-47. PMID: 12847041

24. Jong-Soo P, EUN-LK S, Dong-Won K, Sang-Pyo K. Calcified Chronic Subdural Haematoma associated with intra cerebral haematoma. J Korean Neurosurg Soc. 2003;34:177-8.

25. Moon HG, Shin HS, Kim TH, Hwang YS, Park SK. Ossified chronic subdural hematoma. Yonsei Med J. 2003;44(5):915-918. doi:10.3349/ymj.2003.44.5.915

26. Sato K, Yamada M, Shimzu S, et al. No Shinkei Geka. 2005;33(8):805-808. PMID: 16095211

27. He XS, Zhang X. Giant calcified chronic subdural haematoma: a long term complication of shunted hydrocephalus. J Neurol Neurosurg Psychiatry. 2005;76(3):367. doi:10.1136/jnnp.2004.043174

28. Dimogerontas G, Rovlias A. Bilateral huge calcified chronic subdural haematomas ['armoured brain'] in an adult patient with a coexistent VA shunt infection. Br J Neurosurg. 2006;20(6):435-436. doi:10.1080/02688690601101796

29. Evans SJ. Armored brain. Neurology. 2007;68(22):1954. doi:10.1212/01.wnl.0000265518.75600.e8

30. Amr R, Maraqa L, Choudry Q. 'Armoured brain'. A case report of a calcified chronic subdural haematoma. Pediatr Neurosurg. 2008;44(1):88-89. doi:10.1159/000110672

31. Papanikolaou PG, Paleologos TS, Triantafyllou TM, Chatzidakis EM. Shunt revision after 33 years in a patient with bilateral calcified chronic subdural hematomas. Case illustration. J Neurosurg. 2008;108(2):401. doi:10.3171/JNS/2008/108/2/0401

32. Petraglia AL, Moravan MJ, Jahromi BS. Armored brain: A case report and review of the literature. Surg Neurol Int. 2011;2:120. doi:10.4103/2152-7806.84391

33. Turgut M, Samancoğlu H, Ozsunar Y, Erkuş M. Ossified chronic subdural hematoma. Cent Eur Neurosurg. 2010;71(3):146-148. doi:10.1055/s-0030-1253346

34. Akhaddar A, Baallal H, Elasri A, Elmostarchid B, Boucetta M. Large bilateral calcified subdural hematomas. Headache. 2011;51(9):1440-1441. doi:10.1111/j.1526-4610.2011.01865.x

35. Rahman A, Haque M, Bhandari PB. Calcified chronic subdural haematoma. BMJ Case Rep. 2012;2012:bcr0120125499. doi:10.1136/bcr.01.2012.5499

36. Juan WS, Tai SH, Hung YC, Lee EJ. Multiple tenting techniques improve dead space obliteration in the surgical treatment for patients with giant calcified chronic subdural hematoma. Acta Neurochir (Wien). 2012;154(4):707-710. doi:10.1007/s00701-011-1234-8

37. Sugita Y, Ohta M, Ohshima K, et al. Epstein-Barr virus-positive lymphoproliferative disorder associated with old organized chronic subdural hematoma. Pathol Int. 2012;62(6):412-417. doi:10.1111/j.1440-1827.2012.02824.x

38. Pappamikail L, Rato R, Novais G, Bernardo E. Chronic calcified subdural hematoma: Case report and review of the literature. Surg Neurol Int. 2013;4:21. doi:10.4103/2152-7806.107548
39. Tandon V, Garg K, Mahapatra AK. 'Double skull' appearance due to calcifications of chronic subdural hematoma and cephalhematoma: a report of two cases. Turk Neurosurg. 2013;23(6):815-817. doi:10.5137/1019-5149.JTN.6189-12.1
40. Chaudhry FS, Biller J, Loftus CM, Braca JA 3rd. Armored brain. Arq Neuropsiquiatr. 2013;71(10):823. doi:10.1590/0004-282X20130131
41. Goyal PK, Singh D, Singh H, Dubey J, Tandon M. Armoured brain of unknown etiology. Asian J Neurosurg. 2013;8(3):165. doi:10.4103/1793-5482.121695
42. Ito M, Saito S, Kondo R, Nagahata M, Kayama T. No Shinkei Geka. 2014;42(8):717-722. PMID: 25087759
43. Cai J, Zhang Y, Bai X, et al. Postoperative hemorrhage in an elderly patient with a glioblastoma multiform and a calcified chronic subdural hematoma. World J Surg Oncol. 2014;12:110. doi:10.1186/1477-7819-12-110
44. Arán-Echabe E, Frieiro-Dantas C, Prieto-González Á. Hematoma subdural cronico calcificado: cerebro blindado [Chronic calcified subdural haematoma: armoured brain]. Rev Neurol. 2014;58(9):420-421.
45. Gupta SK, Pandia MP. Anesthetic management of a case of armored brain. Saudi J Anaesth. 2015;9(1):89-90. doi:10.4103/1658-354X.146323
46. Li H, Mao X, Tao XG, Li JS, Liu BY, Wu Z. A Tortuous Process of Surgical Treatment for a Large Calcified Chronic Subdural Hematoma. World Neurosurg. 2017;108:996.e1-996.e6. doi:10.1016/j.wneu.2017.09.023
47. Viozzi I, van Baarsen K, Grotenhuis A. Armored brain in a young girl with a syndromal hydrocephalus. Acta Neurochir (Wien). 2017;159(1):81-83. doi:10.1007/s00701-016-2991-1
48. Keser N, Celikoglu E, Aydın İH, Ozbay N. The outcome of a pregnant woman with a minor head injury: an ossified subdural hematoma (OSDH). J Surg Case Rep. 2017;2017(3):rjx048. doi:10.1093/jscr/rjx048
49. Satyarthee GD, Lalwani S. Armored Brain Associated with Secondary Craniostenosis Development at 7-year following Ventriculoperitoneal Shunt Surgery during Infancy: Extremely Unusual Association and Review. Asian J Neurosurg. 2018;13(4):1175-1178. doi:10.4103/ajns.AJNS_263_16
50. Qin G, Xu P, Lan S, et al. Skull angiosarcoma associated with a calcified chronic subdural hematoma. Br J Neurosurg. 2019;1-4. doi:10.1080/02688697.2019.1594693
51. Fang J, Liu Y, Jiang X. Ossified Chronic Subdural Hematoma in Children: Case Report and Review of Literature. World Neurosurg. 2019;126:613-615. doi:10.1016/j.wneu.2019.03.144
52. Liu X, Zhou J, Shen B, et al. Ossified Chronic Subdural Hematoma and Subsequent Epstein-Barr Virus-Positive Large B-Cell Lymphoma: Case Report and Literature Review. World Neurosurg. 2019;130:165-169. doi:10.1016/j.wneu.2019.07.011
53. Tian W, Meng X, Zou J. A Giant Ossified Chronic Subdural Hematoma. J Coll Physicians Surg Pak. 2019;29(9):905. doi:10.29271/jcpsp.2019.09.905
54. Turgut M, Yay MÖ. A Rare Case of Ossified Chronic Subdural Hematoma Complicated with Tension Pneumocephalus. J Neurol Surg Rep. 2019;80(4):e44-e45. doi:10.1055/s-0039-1694738
55. Rong J, Zhou W, Di G, Jiang X. A case of giant calcified chronic subdural hematoma. Asian J Surg. 2022;45(10):1870-1871. doi:10.1016/j.asjsur.2022.04.003
56. Songnatsiri P, Ounahachok T, Atikankul T, Thananon J, Sookpotarom P. An early presentation of a large calcified chronic subdural hematoma presenting as an inner eggshell in an elderly patient: A case report. Int J Surg Case Rep. 2020;73:150-153. doi:10.1016/j.ijscr.2020.07.005
57. Zhang S, Wang X, Liu Y, Mao Q. Resection of a huge calcified chronic subdural haematoma: case report. Br J Neurosurg. 2020;34(2):229-231. doi:10.1080/02688697.2018.1432749
58. Bhardwaj S, Sinha VD, Shekhawat JS, Gora NK. Large Chronic Calcified Subdural Hematoma with Empyema-A Long-Term Complication following Shunt Surgery. J Neurosci Rural Pract. 2020;11(4):673-674. doi:10.1055/s-0040-1713717
59. Pakrasi R, Pandey P, Das S, Datta S, Saha D. Calcified chronic subdural hematoma: illustrative case. J Neurosurg Case Lessons. 2021;2(15):CASE21468. doi:10.3171/CASE21468
60. Prasad PK, Tripathi M, Joshi A. Chronic Psychosis Unveiling the Diagnosis of the Armored Brain. Neurol India. 2021;69(5):1430-1431. doi:10.4103/0028-3886.329615
61. Bett D. Calcified Chronic Subdural Hematoma: Case Report And Review Of Literature . EAJNS. 2022 Dec. 6;1(Supp 2):45. Available from: https://theeajns.org/index.php/eajns/article/view/73
62. Girotto AL, Gago G. Surgical Management of the Armored Brain: Case Report of the Treatment of a Chronic Calcified Subdural Hematoma. Arquivos Brasileiros de Neurocirurgia: Brazilian Neurosurgery. 2022 Nov 29. doi:10.1055/s-0042-1758711
63. Wang H, Xu X, Yi Y, Liu B. An extensive calcified chronic subdural hematoma. Asian J Surg. 2023;S1015-9584(23)00760-1. doi:10.1016/j.asjsur.2023.05.089
64. Mansour M, Adi M, Mousa H, et al. A post-traumatic ossified subdural chronic hematoma successfully managed in a 34-year-old woman: a case report. Ann Med Surg (Lond). 2023;85(4):1026-1029. doi:10.1097/MS9.0000000000000304
65. Marini A, Spennato P, Aliberti F, et al. Brain Herniation into the Subdural Space: Rare Iatrogenic Complication of Treatment of a Giant Calcified Subdural Hematoma.

World Neurosurg. 2020;140:65-70. doi:10.1016/j.wneu.2020.05.057

66. Chan ZW, Gallo P. Chronic calcified subdural haematoma found after presentation of symptomatic glioma. BMJ Case Rep. 2018;2018:bcr2018224929. doi:10.1136/bcr-2018-224929

67. Garg K, Singh PK, Singla R, et al. Armored brain-massive bilateral calcified chronic subdural hematoma in a patient with ventriculoperotoneal shunt. Neurol India. 2013;61(5):548-550. doi:10.4103/0028-3886.121950

Romanian Neurosurgery (2023) XXXVII (3): pp. 349-353
DOI: 10.33962/roneuro-2023-062
www.journals.lapub.co.uk/index.php/roneurosurgery

Post-irradiation bilateral basal ganglia calcification in a patient with cerebral metastasis. A case report and review of literature

Akhlaque Hossain Khan[1], Urmeeta Dutta[1], Alok Jha[1], Tandin Wangyel Dorji[1], Farheen Khan[2], Bipin Chaurasia[3]

[1] Department of Neurosurgery, Bangabandhu Sheikh Mujib Medical University, BANGLADESH
[2] Popular Medical College and Hospital, Dhaka, BANGLADESH
[3] Department of Neurosurgery, Neurosurgery Clinic, Birgunj, NEPAL

Keywords
basal ganglia calcification,
post irradiation,
computed tomography

Corresponding author:
Bipin Chaurasia

Neurosurgery Clinic,
Birgunj, Nepal

trozexa@gmail.com

ABSTRACT
Background: Basal Ganglia calcification is now being diagnosed with increasing frequency with the widespread application of computed tomography (CT) scan in clinical practice. One of the rare causes of bilateral basal ganglia calcification is post-irradiation sequelae. So far, there are a total of 11 reported cases of post-radiotherapy basal ganglia calcification.
Case report: Here, we report another case of bilateral basal ganglia calcification following radiotherapy for cerebral metastasis from Ca- ovary along with a review of the other reported cases. The exact pathogenesis of this condition is not clear. It appears, however, to be related to radiation vasculitis of the small vessels of the brain with resultant hyalinization and calcification. Conclusions: A long-term follow-up study would be necessary to evaluate the significance and implication of post-irradiation calcification of the grey matter.

INTRODUCTION

Basal ganglia calcification is seen in approximately 1% of all CT scans of the brain (3). It is seen more frequently in older patients and is considered a normal incidental and idiopathic finding in an elderly patient but should be considered pathological in persons younger than 40 years unless proved otherwise (2). There are many pathological causes e.g. toxic, infectious, metabolic, hypoxic, inherited and idiopathic. The pathogenesis though exactly unknown, is assumed to be deposition of a colloid material in and around the finer cerebral blood vessels, with subsequent hyalinization and calcification of this colloid (1). Many patients are found to be asymptomatic; while others present with seizure, weakness, movement symptoms e.g. unsteadiness, walking difficulties, tremors, rigidity and others

Scan to access the online version

demonstrate psychiatric symptoms e.g. psychosis, mood disorders or dementia (6). CT scan is considered the gold standard for diagnosis which identifies calcifications as hyperdense lesions typically bilateral and symmetrical, most frequently involving basal ganglia, but also dentate nuclei, thalamus and subcortical white matter (4). In the basal ganglia region, the most common site of calcification is globus pallidus followed by putamen and caudate nucleus (21,5,17,19,20).

CASE REPORT

A 35 years old, non-diabetic, normotensive, non-asthmatic lady (para 2+0) presented to our department with the complaints of recurrent attacks of generalized tonic clonic seizure, occasional headache and vomiting for last 4 months. She had a history of total abdominal hysterectomy with bilateral salpingo-oophorectomy with omentectomy with appendicectomy due to papillary serous cystadenocarcinoma of ovary with multiple hepatic metastases 4 years back. Following surgery, she received 6 cycles of adjuvant chemotherapy (Carboplatin+Paclitaxel) within the next 6 months. After that, she didn't have regular follow up and again presented with the afore mentioned complaints. On examination, she was conscious (GCS 15) with intact higher psychic function and intact cranial nerves.

There was right sided hemiparesis with muscle power MRC grade 4 in all groups; with normal tone and jerks. She underwent an MRI(figure 1 and figure 2) of brain with contrast and it revealed ring enhancing irregular mass in the left frontal lobe with perilesional edema and was diagnosed as a case of cerebral metastasis from recurrence of the Ca-ovary. At that time, tumor marker CA 125 was markedly elevated: 127 U/mL (Normal upto 0-35U/Ml) (12). After initial resuscitation with injectable anticonvulsants and steroids, she was referred to the clinical oncology department and there she was treated with palliative radiotherapy to whole brain; 3DCRT (Photon, 6 MV LINAC), 300 cGy per fraction. She received ten fractions equating to 3000 cGy of total radiation exposure within 14 days.

She had an uneventful recovery (with persistence of hemiparesis) after completing scheduled doses of radiotherapy and was discharged to home with advice for follow up CT scans at every 6 months interval. During her first follow up (after 6 months), she complained of occasional headache. Muscle power of left upper and lower limb was as before (MRC grade 4) with no new neurological deficit and no constitutional symptom. CT scan of brain with contrast was done and it revealed hypodense area indicating radiation necrosis in the previous tumor bed. Moreover, there was calcification in the Globus pallidus of both basal ganglia region which was not evident in previous imaging studies(figure 3).

Biochemical parameters were evaluated and it revealed normal renal function and normal serum calcium, inorganic phosphate and parathyroid hormone. On the basis of above findings, post radiotherapy bilateral basal ganglia calcification was diagnosed.

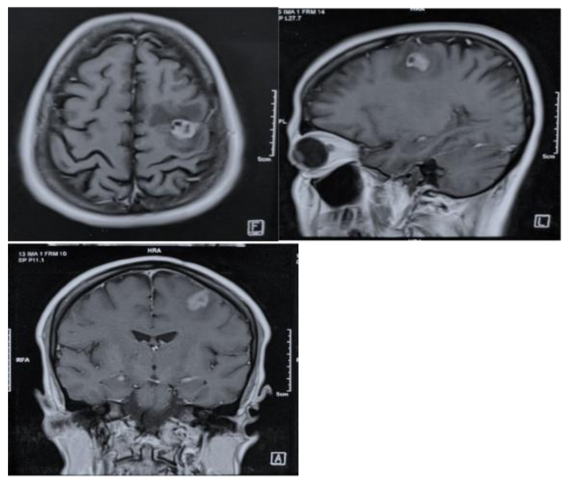


Figure 1. MRI of brain with contrast showing left frontal metastatic SOL with peri lesional edema with no other significant abnormality (before radiotherapy).

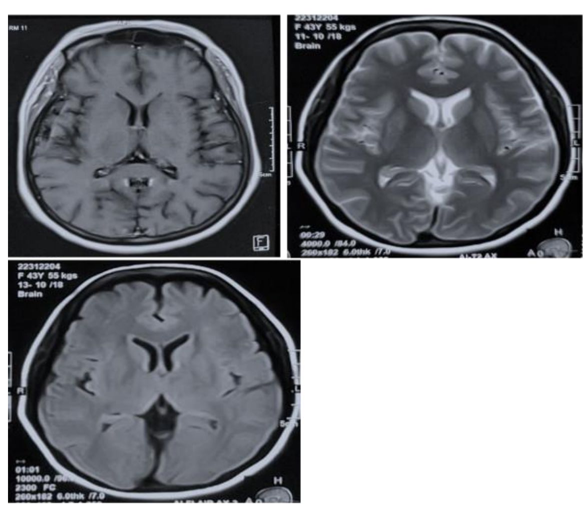


Figure 2. T1,T2, FLAIR sequences of MRI showing no significant abnormality of the basal ganglia region in the same patient (before Radiotherapy).

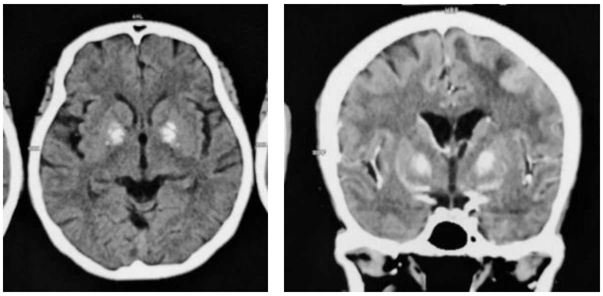


Figure 3. Post radiotherapy CT scan of brain showing symmetrical irregular hyperdensities in both Globus pallidus indicating bilateral basal ganglia calcification.

Discussion

Microscopic description of bilateral basal ganglia calcification was first made by Virchow (10) In 1935 Fritzsche (14) was the first to report roentgenologic appearance of basal ganglia calcification in 3 siblings. Its occurrence in idiopathic hypoparathyroidism was described by Eaton, Camp and Love in 1939 (13).

Basal ganglia calcification in pseudohypoparathyroidism was first reported by Sprague, Hains, and Power in 1945 (15). Its occurrence subsequent to radiation therapy was first described by Harwood-Nash and Reilly in 1970 (12). Intrathecal administration of Methotrexate may produce intracranial calcification (18). Kramer and Lee described a case with diffuse calcification in the basal ganglia and cerebral cortex subsequent to irradiation and methotrexate (intrathecal) treatment for acute lymphocytic leukemia (9). Nakagaki et al. (16) described monkey brain damage from radiation in the therapeutic range. At six months after 6,000 rad, widely scattered punctate necrotic lesions, 1 mm or less, were demonstrated, whereas, 1 year later almost complete mineralization of necrotic lesions with innumerable minute deposits of calcium and iron were observed. Vasculitis and telangiectasia were also present.

Table 1. Previously reported cases of post irradiation basal ganglia calcification (5, 17, 19,20).

Case No	Author	Age (yrs.)	Sex	Pathology & site	Radiotherapy: source, field, site & dose	Duration following radiotherapy	S calcium and PO4
1	Harwood-Nash, Reilly. 1970	9	F	Glioblastoma multiforme in suprasellar region	External cobalt 60. 5 X 5 cm^2 4500 rad/4wks	3 yrs	Normal
2	Harwood-Nash, Reilly. 1970	3	M	Histiocytosis involving multiple areas of the calvarium	200 kV X ray various ports 300 rad X 3 1250 next yr, 1750 following yr	9 yrs	Normal
3	Kramer and Lee 1974	10	M	CNS involvement of lymphocytic leukaemia	External cobalt 60 550 rad Methotrexate	10 months	Normal
4	Numaguchi et al. 1974	9	F	Astrocytoma in Thalamic region	Supervoltage 2 MeV resonant transformer 7 X 7 cm^2 6000 rad/6 wks	6 yrs	Normal
5	Lee and Suh 1976	12	F	Spongioblastoma polare, Right optic nerve	External cobalt 60. 5 X 5 cm^2 5500 rad/5wks	10 yr	Normal
6	Lee and Suh 1976	16	M	Medulloblastoma cerebellar vermis	Total brain and spine axis 4000 rad. 4700 rad to vermis/ 4 wks	14 yrs	Normal
7	A D J Pearson, Campbell et al. 1983	2	-	Medulloblastoma	8 MeV Lateral 18 X 18 cm; 36 doses	5 yrs	-
8	A D J Pearson, Campbell et al. 1983	3	-	Medulloblastoma	8 MeV Lateral 18 X 19 cm; 39 doses	5 yrs	-

9	A D J Pearson, Campbell et al. 1983	4	-	Medulloblastoma	4.2 MeV Lateral 19 X 15 cm; 30 doses	5 yrs	-
10	Terry Lichtor, Robert L. Wollmann et al. 1984	14	-	Medulloblastoma	Total brain 4000 rads and 4000 rads to spine; 27 doses	12 yrs	Normal
11*	P Sanchetee, S Venkataraman et al. 1999						

The pathogenesis of calcification in the grey matter is not clear. Microscopically, vasculitis, fibroblastic process, hyalinization, and calcification were observed in the brains of both humans and animals following irradiation to the heads (17). The possibility of an autoimmune reaction localized to the irradiated demyelinated tissue with subsequent accumulation of abnormal metabolites was suggested by Lampert et al. (7). Babbitt et al. (8) proposed that the ferrocalcinosis is the end result of a process of circulatory disturbance resulting in local anoxia and necrosis. Harwood-Nash and Reilly (12) postulate that hypersensitivity of small vessels of the basal ganglia to radiation produces vascular damage with hypoxia resulting in calcification. A long term follow-up study would be necessary in order to assess the significance and implication of post-irradiation calcification in the grey matter.

Our reported case of post radiotherapy bilateral basal ganglia calcification is about a middle aged female with secondary brain tumor while previously reported cases were children or adolescents with primary brain tumors. Among the previously reported cases (table 1), three of the patients had poor attention and hyperactivity disorders with endocrine abnormalities (5) while our patient did not have any specific sign or symptom.

Conclusion

Basal ganglia calcification may lead to various motor and psychiatric symptoms. While treating any patient with neuraxial radiotherapy, possibility of basal ganglia calcification might be kept in mind. Whether the incidence of basal ganglia calcification is radiation dose related or not, needs to be further studied. All of such patients should be kept under close follow up regimen to ensure quality of life.

References

1. John C. Bennett, Roy H, Maffly, Howard L, Steinbach: The significance of bilateral basal ganglia calcification, Radiology, Vol 72, No 3 (1959).
2. Haaga JR, Boll D: CT and MRI of the whole body, Mosby (2009).
3. Gaillard F, Saber M, Smith H: Basal ganglia calcification, Reference article, Radiopaedia.org (May, 2008).
4. Giulia Donzuso, Giovanni Mostile, Mario Zappia: Basal ganglia calcifications (Fahr's syndrome): related conditions and clinical features, Neurol Sci. August 2019: 40(11).
5. A D J Pearson, A N Campbell, V L McAllister, G L Pearson: Intracranial calcification in survivors of childhood medulloblastoma, Archives of Disease in Childhood, 58, 133-136 (1983).
6. WebMD editorial contributors: What is basal ganglia calcification? WebMD, Brain and Nervous system (November 2020).
7. Lampert, M. I. Tom and W. D. Rider, Disseminated demyelination of the brain following Co60(Gamma) radiation, AMA Arch. Pathol. 68, 322 (1959).
8. E. V. Medill, Bilateral symmetric calcification of basal ganglia associated with parathyroid insufficiency, Bit. J. Radial. 24, 685 (1951).
9. S. Kramer and K. F. Lee, Complications of radiation therapy central nervous system, Semin. Roentgenol 9. 75 (1974).
10. R Virchow, Kalk-metastasen. Virchows Arch Path. Anat. 8, 103 (1855).
11. https://emedicine.medscape.com/article/2087557-overview

C. C. F. Harwood-Nash and B. J. Reilly, Calcification of basal ganglia following radiation therapy, Am. J. Roentgenal. 108. 392 (1970).

12. L. M. Eaton, J. D. Camp and J. G. Love, Symmetrical cerebral calcification. particularly of basal ganglia, demonstrable roentgenographically, Arch. Neurol. Psychiat. 41. 921 (1939).
13. R. Fritzsche, Eine familiLr auftretende Form von Oligophrenie mit roentgenologisch nachweisbaren symmetrischen Kalkablagerungen im Gehirn, besonders in den Stammganglien. Schweiz. Arch. Neural. I/. Psychiat. 35, 1 (1935).
14. R. G. Sprague, S. F. Haines and M. H. Power, Metabolic effects of parathyroid hormone, dihydrotachys- terol and calciferol in a case of pseudo-hypoparathyroidism. J. Lab. C/in. Med. 30. 361 (1945).
15. H. Nakagaki, G. Brunhart, T. L. Kemper and W. F. Caveness. Monkey brain damage from radiation in the therapeutic range, J. Neurosurg. 44. 3 (1976).
16. K. Francis Lee, Jung Ho Suh: CT evidence of grey matter calcification secondary to radiation therapy, Pergamon Press, Vol. 1. pp. 103~ 110 (1976).
17. H. Norrell, C. B. Wilson, D. E. Slagel and D. B. Clark, Leukoencephalopathy following the administration of methotrexate into the cerebrospinal fluid in the treatment of primary brain tumors, Cancer 33. 923 (1974).
18. Terry Lichtor, Robert L. Wollmann, Frederick D. Brown: Calcified Basal Ganglionic Mass 12 Years After Radiation Therapy for Medulloblastoma, Surg Neurol 21:373-6 (1984).
19. P Sanchetee, S. Venkataraman, C mohan, D D Shetty, G Iyengar: Basal ganglia calcification, J Assoc Physicians India (1999).
20. H Forstl, B Krumm, S Eden, K Kohlmeyer: Neurological disorders in 166 patients with basal ganglia calcification: a statistical evaluation (1990).

Romanian Neurosurgery (2023) XXXVII (3): pp. 354-369
DOI: 10.33962/roneuro-2023-063
www.journals.lapub.co.uk/index.php/roneurosurgery

Radiation exposure in spine surgeries. A review of risks, consequences, and prevention strategies

Muhammad Jehanzeb, Ahtesham Khizar

Punjab Institute of Neurosciences, Lahore, PAKISTAN

ABSTRACT
Radiation exposure is a significant concern in spine surgeries due to the extensive use of fluoroscopic imaging. This review aims to evaluate the risks, consequences, and prevention strategies associated with radiation exposure in spine surgeries. The risks of radiation exposure include potential biological damage to patients and surgical staff, such as skin burns, cataracts, and an increased risk of developing cancer. The consequences of radiation exposure can have long-term health implications and may result in substantial healthcare costs. To mitigate the risks, various prevention strategies are recommended. These include optimizing the use of fluoroscopy by adopting low-dose imaging techniques, minimizing the duration of exposure, and ensuring proper shielding of personnel. Additionally, the use of alternative imaging modalities, such as intraoperative three-dimensional (3D) navigation systems, can reduce reliance on fluoroscopy and subsequently decrease radiation exposure. Furthermore, implementing a culture of radiation safety through education, training, and awareness programs is crucial. This involves educating surgeons, nurses, and other healthcare professionals about the potential risks, proper use of equipment, and radiation protection measures. Strict adherence to radiation safety guidelines and continuous monitoring of radiation doses are essential to ensure the well-being of both patients and healthcare providers. In short, radiation exposure in spine surgeries poses significant risks and potential consequences. However, with the adoption of appropriate prevention strategies, such as optimizing imaging techniques, implementing alternative modalities, and fostering a culture of radiation safety, the potential risks can be mitigated. By prioritizing radiation safety, healthcare facilities can provide better outcomes for patients and minimize the long-term health implications associated with radiation exposure in spine surgeries.

Keywords
radiation exposure,
radiation injuries,
prevention and control,
risk,
spine

Corresponding author:
Muhammad Jehanzeb

Punjab Institute of Neurosciences,
Lahore, Pakistan

drjehanzeb28@gmail.com

Scan to access the online version

INTRODUCTION

The utilization of fluoroscopic imaging and intraoperative imaging techniques has become increasingly prevalent in spine surgeries. These imaging modalities provide valuable real-time visualization and guidance during complex spinal procedures, leading to improved surgical outcomes. However, one significant concern associated with these techniques is the potential for radiation exposure to both patients and healthcare professionals involved in these procedures. Radiation exposure in medical procedures has been extensively studied, and its detrimental effects on human health are well-established.1 The primary sources of radiation exposure in spine surgeries include X-ray fluoroscopy and computed tomography (CT) scans. These imaging techniques provide high-resolution images but also expose patients and medical staff to ionizing radiation.2 While the radiation doses received during individual procedures may appear low, cumulative exposure from repeated or prolonged surgeries can potentially lead to adverse health effects.

The potential risks associated with radiation exposure in spine surgeries are of significant concern. Acute effects may include skin injuries, radiation sickness, and other immediate consequences for both patients and healthcare professionals. Moreover, long-term risks, such as an increased risk of cancer, genetic effects, and cataracts, have been observed in individuals exposed to high levels of ionizing radiation.[3] Understanding these risks is crucial for developing effective preventive strategies.

To address the risks associated with radiation exposure, numerous preventive measures have been proposed and implemented in clinical practice. These strategies aim to minimize radiation exposure without compromising the quality of surgical outcomes. Examples include the use of shielding devices and personal protective equipment (PPE), optimization of imaging techniques to reduce radiation dose, and the adoption of low-dose imaging modalities like cone-beam computed tomography (CBCT). Additionally, radiation dose monitoring and tracking systems play a vital role in assessing the radiation exposure of both patients and medical staff.[4]

The implementation of preventive strategies requires education and training for healthcare professionals involved in spine surgeries. Radiation safety education programs, training on proper technique and equipment use, and ongoing professional development are essential to ensure compliance with radiation safety guidelines. Furthermore, interdisciplinary collaboration between surgeons, radiologists, medical physicists, and other healthcare providers is crucial for developing comprehensive radiation safety protocols and promoting a culture of safety. In short, radiation exposure in spine surgeries poses risks to both patients and healthcare professionals. Understanding the potential consequences of radiation exposure and implementing effective preventive strategies are paramount in ensuring the safety and well-being of individuals involved in these procedures. This review will delve into the risks, consequences, and preventive measures associated with radiation exposure in spine surgeries, shedding light on the importance of radiation safety awareness and the implementation of appropriate strategies.

Discussion

Overview of the growing prevalence of fluoroscopic imaging in spine surgeries

Fluoroscopic imaging has witnessed a significant increase in its utilization within spine surgeries. This imaging modality offers real-time visualization and guidance during complex spinal procedures, leading to enhanced surgical outcomes. However, this increased usage has raised concerns regarding radiation exposure for both patients and healthcare professionals involved in these procedures. Fluoroscopy, a technique that employs X-rays to generate dynamic images, plays a pivotal role in various spine surgeries, including spinal fusion, discography, and minimally invasive procedures. It enables surgeons to visualize anatomical structures, guide instrument placement, and verify the accuracy of spinal implant positioning. The real-time nature of fluoroscopy allows for immediate adjustments and ensures precise intraoperative decision-making. The growing prevalence of fluoroscopic imaging in spine surgeries can be attributed to several factors. Firstly, advancements in imaging technology have led to improved image quality, reducing the risk of diagnostic errors and facilitating more precise surgical interventions. Additionally, the rise in minimally invasive spine surgeries has driven the need for real-time imaging guidance, making fluoroscopy an indispensable tool in such procedures.[5]

Fluoroscopic imaging also offers potential benefits, including reduced surgical invasiveness, shorter hospital stays, and faster recovery times for patients. These advantages have contributed to its increased adoption among spine surgeons, as they strive to provide optimal patient care and outcomes. However, the widespread use of fluoroscopy has raised concerns about radiation exposure. The continuous and prolonged exposure to ionizing radiation during these procedures poses potential health risks for both patients and healthcare professionals.[5] Therefore, it is essential to address radiation safety measures and implement preventive strategies to minimize radiation-related complications.

Statement of the problem: Radiation exposure and its potential health risks

Radiation exposure is a significant concern in spine surgeries due to the utilization of fluoroscopic imaging and other intraoperative imaging techniques. The risks associated with radiation exposure are well-established. Ionizing radiation has the potential to cause both acute and long-term effects on human health. Acute radiation effects include skin injuries, radiation sickness, and other immediate consequences for patients and healthcare professionals exposed to high doses of radiation. Furthermore, long-term risks have been observed, such as an increased risk of cancer, genetic effects, and the development of cataracts. Studies have demonstrated a direct link between radiation exposure and the development of radiation-induced cancers. The risk of cancer is dose-dependent, meaning that higher cumulative radiation doses over time increase the likelihood of malignancies.[6] For patients undergoing multiple spine surgeries or those requiring long-term follow-up with imaging, the cumulative radiation dose can become a significant concern.

The potential genetic effects of radiation exposure are also noteworthy. High doses of radiation can induce DNA damage, which may result in genetic mutations and hereditary effects in future generations.[7] Healthcare professionals exposed to radiation during spine surgeries should be particularly cautious due to the occupational hazards associated with chronic exposure. Another important long-term consequence of radiation exposure is the increased risk of cataract development. Studies have shown that healthcare professionals who frequently use fluoroscopy or work in interventional radiology departments have a higher prevalence of radiation-induced cataracts.[8] This emphasizes the need for protective measures and careful monitoring of radiation exposure in spine surgeries. Understanding the potential health risks associated with radiation exposure in spine surgeries is crucial for developing effective preventive strategies and ensuring the safety of patients and healthcare professionals. By implementing appropriate measures to minimize radiation exposure and adopting strict radiation safety protocols, the potential risks and long-term consequences can be mitigated.

Types of radiation used in spine surgeries: X-ray, fluoroscopy, and computed tomography (CT)

Radiation plays a crucial role in various imaging modalities used in spine surgeries, including X-ray, fluoroscopy, and CT. Each of these techniques utilizes different forms of radiation to generate detailed images of the spine and guide surgical interventions. An overview of these radiation types and their applications in spine surgeries is given below:

1. **X-ray:** X-ray imaging employs electromagnetic radiation to produce two-dimensional images of the spine. It is commonly used to assess the bony structures, alignment, and presence of fractures or deformities. X-rays provide valuable information on spinal anatomy and pathology, aiding in preoperative planning and intraoperative guidance. However, X-rays deliver a relatively higher radiation dose compared to other imaging techniques.[9]
2. **Fluoroscopy:** Fluoroscopy is a real-time imaging technique that utilizes X-rays to generate dynamic images of the spine. It provides continuous visualization during spine surgeries, enabling surgeons to guide instrumentation, assess intervertebral motion, and verify the accuracy of implant placement. Fluoroscopy provides valuable real-time feedback, facilitating precise intraoperative decision-making. However, it involves prolonged radiation exposure due to continuous imaging.[9]
3. **Computed Tomography (CT):** CT scans utilize X-ray radiation in combination with advanced computer processing to create cross-sectional images of the spine. CT offers detailed information about bony structures, spinal alignment, and soft tissue components, aiding in the evaluation of complex spinal pathologies. CT imaging can be particularly useful in surgical planning, as it provides three-dimensional reconstructions and enhances visualization of anatomical structures.[9]

Quantification of radiation exposure levels in different spine procedures

Quantifying radiation exposure levels in different spine procedures is an important aspect of radiation safety. While specific radiation exposure levels can vary depending on various factors such as the type of procedure, imaging technique, equipment used, and individual patient characteristics, the following information provides a general overview of radiation exposure levels in commonly performed spine procedures:

1. **Pedicle Screw Placement:** Pedicle screw placement (PSP) is a common procedure in spinal instrumentation, which involves the insertion of screws into the vertebral pedicles for stabilization. The radiation exposure levels in PSP procedures can vary depending on several factors, including the imaging technique used (such as fluoroscopy or CT), the complexity of the procedure, the number of fluoroscopic images taken, and the surgeon's experience. Studies have reported varying radiation exposure levels in PSP procedures. For instance, a study by Smith H et al.[10] found that the mean radiation dose to the patient during lumbar PSP using fluoroscopy ranged from 1.15 to 8.53 mSv per procedure. They also compared fluoroscopy-guided PSP with computer-assisted image guidance and reported a mean radiation dose of 1.29 mSv for the fluoroscopy group and 0.47 mSv for the image guidance group. This highlights the variability in radiation exposure levels and the potential for reducing exposure through the use of image guidance techniques.
2. **Minimally Invasive Spine Surgery (MISS):** Minimally invasive spine surgery techniques aim to minimize tissue trauma and promote faster recovery compared to traditional open surgical approaches. These procedures often involve the use of fluoroscopy or other imaging guidance for visualization and instrument guidance. The radiation exposure levels in MISS procedures can vary depending on the specific technique used, the complexity of the procedure, the duration of fluoroscopy, and the number of imaging acquisitions.
3. **Image-Guided Spine Interventions:** Image-guided spine interventions, such as vertebroplasty or kyphoplasty, involve the use of imaging guidance, usually fluoroscopy or CT, to assist in the precise placement of instruments and delivery of therapeutic agents. These procedures can have varying radiation exposure levels depending on factors such as the complexity of the intervention, the number of imaging acquisitions, and the duration of fluoroscopy.

It is important to note that the actual radiation exposure levels can vary widely based on individual patient characteristics, operator technique, equipment used, and adherence to radiation safety measures. To minimize radiation exposure, it is crucial to implement radiation safety protocols, optimize imaging techniques, and utilize appropriate shielding and PPE. Adhering to ALARA principles (As Low As Reasonably Achievable) is essential in maintaining radiation doses as low as possible while still achieving the required diagnostic and procedural objectives.[11]

Factors influencing radiation exposure: duration, technique, and patient characteristics

Certainly! Factors influencing radiation exposure in spine surgeries can include the duration of the procedure, imaging technique utilized, and patient characteristics. Some of these factors are:

1. **Duration of Procedure:** The duration of a spine surgery procedure can have a significant impact on radiation exposure. Prolonged surgical durations generally lead to increased radiation exposure due to the extended use of fluoroscopy or other imaging techniques. The longer the fluoroscopy time, the higher the radiation dose to both the surgeon and the patient. Studies have shown a positive correlation between surgical duration and radiation exposure in spine surgeries.[12] It is important for surgeons to minimize fluoroscopy time and optimize procedural efficiency to reduce radiation exposure.
2. **Imaging Technique:** The choice of imaging technique during spine surgeries can greatly influence radiation exposure levels. Fluoroscopy and CBCT are commonly used imaging modalities. Fluoroscopy provides real-time imaging but may result in higher radiation exposure due to continuous imaging. On the other hand, CBCT provides 3D imaging with lower radiation doses but may require longer imaging

acquisition times. Surgeons need to balance the need for image quality with radiation exposure and choose the most appropriate imaging technique for each procedure. Additionally, utilizing low-dose imaging protocols and minimizing unnecessary imaging acquisitions can further reduce radiation exposure.

3. **Patient Characteristics:** Patient characteristics, such as body habitus and the number of levels treated, can influence radiation exposure in spine surgeries. A recent study on PSP in adult idiopathic scoliosis patients found that higher body mass index (BMI) was associated with increased radiation exposure.[13] This is because patients with higher BMI may require more fluoroscopic images or longer imaging times to visualize and place pedicle screws accurately. Similarly, procedures involving a greater number of levels treated may require additional imaging acquisitions, leading to increased radiation exposure. Surgeons should be mindful of these patient-related factors and take appropriate measures to minimize radiation exposure, such as optimizing imaging parameters and adjusting technique based on patient-specific considerations.

To effectively mitigate radiation exposure in spine surgeries, it is crucial to implement comprehensive radiation safety protocols. This includes utilizing appropriate shielding and personal protective equipment, adhering to ALARA principles, optimizing imaging techniques, and promoting ongoing education and awareness among healthcare providers about radiation safety practices. It is important to note that the specific influence of these factors may vary across different surgical procedures and patient populations. Surgeons and healthcare providers should stay updated with the latest research and guidelines in the field of radiation safety to ensure the best possible outcomes for both patients and medical personnel.

Risks and Consequences of Radiation Exposure

Risks and consequences associated with radiation exposure are:

1. **Increased Risk of Cancer:** Exposure to ionizing radiation, such as that used in spine surgeries, has been associated with an increased risk of cancer. The long-term effects of radiation exposure can lead to the development of various malignancies, including leukemia, thyroid cancer, breast cancer, and lung cancer. The risk of cancer is cumulative and increases with higher radiation doses and prolonged exposure.[14] It is crucial to minimize radiation exposure in order to reduce the potential long-term risks of cancer development.
2. **Radiation-Induced Tissue Damage:** Radiation exposure can cause acute and chronic tissue damage. Acute effects include radiation dermatitis, erythema, and radiation sickness. Chronic effects can manifest as tissue fibrosis, radiation necrosis, and damage to organs within the radiation field. The severity of tissue damage is dependent on factors such as the radiation dose, fractionation, and sensitivity of the irradiated tissues.[15] Minimizing radiation exposure through proper technique and adherence to radiation safety measures is essential to mitigate the risk of tissue damage.
3. **Genetic Effects and Birth Defects:** Prolonged exposure to radiation can have genetic effects, including DNA damage and mutations. These genetic alterations can be passed onto future generations, increasing the risk of hereditary diseases and birth defects.[7] It is particularly important to minimize radiation exposure in reproductive-age individuals and pregnant patients to safeguard against potential genetic effects.
4. **Occupational Hazards for Healthcare Providers:** During spine procedures, medical professionals, such as surgeons and operating room personnel, are exposed to radiation. Long-term occupational radiation exposure can raise the risk of radiation-related health issues such cataracts, skin lesions, and an increased risk of cancer. Protecting healthcare professionals requires appropriate radiation shielding, the use of personal protective equipment, and adherence to radiation precautionary measures.[16]

Acute radiation effects: skin injury, radiation sickness

Acute radiation effects can manifest as skin injury and radiation sickness. The details of these specific consequences of radiation exposure are:

1. **Skin Injury:** Radiation-induced skin injury, also known as radiation dermatitis or radiation burns, can occur as a result of high-dose radiation exposure. The severity of skin injury depends on the radiation dose, fractionation, and sensitivity of the skin. Acute effects can include erythema, dry or moist desquamation, blistering, and ulceration. The extent and severity of skin injury can vary based on individual factors and the specific radiation therapy technique employed.[17]
2. **Radiation Sickness:** Radiation sickness, also referred to as acute radiation syndrome (ARS) or radiation poisoning, occurs when the whole body is exposed to high doses of radiation over a short period. ARS manifests as a collection of symptoms that occur in stages based on the radiation dose received. The symptoms can include nausea, vomiting, diarrhea, fatigue, fever, dizziness, and, in severe cases, bone marrow suppression and central nervous system dysfunction. The severity of radiation sickness is directly related to the radiation dose.[18]

Long-term risks: cancer, genetic effects, cataracts

Long-term risks associated with radiation exposure include an increased risk of cancer, genetic effects, and the development of cataracts. Their long-term consequences are:

1. **Increased Risk of Cancer:** Exposure to ionizing radiation has been linked to an increased risk of cancer development. Prolonged exposure to radiation, especially at higher doses, can lead to DNA damage and mutations that may contribute to the initiation and progression of cancer. The risk of developing cancer is dependent on factors such as the radiation dose, the dose rate, and the age at exposure. Various studies have demonstrated an elevated risk of cancers such as leukemia, thyroid cancer, breast cancer, lung cancer, and others following radiation exposure.[7]
2. **Genetic Effects:** Radiation exposure can have genetic effects that can be passed onto future generations. These genetic effects include chromosomal abnormalities, mutations, and hereditary diseases. The risk of genetic effects is influenced by the radiation dose, dose rate, and timing of exposure. It is particularly important to minimize radiation exposure in reproductive-age individuals and pregnant patients to prevent potential genetic effects.[19]
3. **Cataracts:** Cumulative exposure to radiation, especially to the lens of the eye, can increase the risk of developing cataracts. Cataracts are characterized by clouding of the eye's lens, leading to visual impairment. The risk of radiation-induced cataracts is dose-dependent, and the threshold for cataract development is relatively low. Occupational radiation exposure, such as in healthcare professionals working with fluoroscopy or radiation therapy, increases the risk of cataracts.[20]

Radiation-related complications in spine surgery

Common radiation-related complications in spine surgery are:

1. **Skin Injuries:** Excessive radiation exposure can lead to acute skin reactions, such as radiation dermatitis, erythema, desquamation (dry or moist), blistering, and ulceration.
2. **Tissue Damage and Necrosis:** High doses of radiation can cause tissue damage and delayed wound healing, resulting in tissue necrosis. This can occur in both the surgical site and surrounding healthy tissues.
3. **Neurological Complications:** Radiation exposure can lead to neurological complications, including radiculopathy and myelopathy. These complications can arise from radiation-induced damage to nerve roots or the spinal cord.
4. **Increased Risk of Secondary Malignancies:** Long-term radiation exposure in spine surgery patients may increase the risk of developing secondary malignancies, such as sarcomas or solid tumors. The risk is influenced by factors such as radiation dose, fractionation, and patient age.
5. **Radiation-Related Health Effects in Healthcare Providers:** Surgeons and operating room staff who are exposed to radiation during spine surgeries are at risk of radiation-related health effects. These can include cataracts, skin lesions, and an elevated risk of malignancies due to occupational radiation exposure.

Prevention strategies for radiation exposure

Prevention strategies for radiation exposure in spine surgeries aim to reduce the amount of radiation to which patients and healthcare providers are exposed. These strategies include both technical measures and operational practices. Here are some common prevention strategies:

1. **Optimization of Imaging Techniques:** By optimizing imaging techniques, such as adjusting exposure parameters and utilizing low-dose protocols, radiation dose can be reduced while maintaining image quality. This includes the use of collimation, filtration, and pulsed fluoroscopy to minimize unnecessary radiation exposure.
2. **Shielding and Positioning:** Lead shielding devices, such as aprons, thyroid shields, and lead glasses, can be used to protect patients and healthcare providers from unnecessary radiation exposure. Proper patient positioning and immobilization techniques can help minimize the need for repeat imaging and reduce radiation exposure.
3. **Education and Training:** Ensuring that healthcare providers involved in spine surgeries receive appropriate education and training on radiation safety is crucial. This includes understanding the principles of radiation protection, proper use of imaging equipment, and adherence to ALARA principles.
4. **Radiation Monitoring and Dose Reporting:** Implementing systems for real-time monitoring of radiation dose during spine surgeries allows healthcare providers to track and optimize radiation exposure. This includes the use of dose monitoring devices, such as personal dosimeters, and establishing protocols for dose reporting and documentation.
5. **Utilization of Alternative Imaging Modalities:** Whenever appropriate, alternative imaging modalities with lower or no ionizing radiation, such as magnetic resonance imaging (MRI) or ultrasound, can be utilized to reduce radiation exposure in specific cases.

Radiation safety measures for healthcare professionals

Radiation safety measures for healthcare professionals involved in spine surgeries are crucial to minimize their occupational radiation exposure. Here are some commonly recommended radiation safety measures:

1. **Personal Protective Equipment (PPE):** Healthcare professionals should wear appropriate PPE, such as lead aprons, thyroid shields, lead glasses, and gloves, to minimize direct radiation exposure to sensitive body parts.[12]
   Distance and Positioning: Healthcare professionals should maintain a safe distance from the radiation source whenever possible. By positioning themselves away from the primary beam and utilizing shielding barriers, they can reduce their exposure to scattered radiation.
   Time Optimization: Minimizing the duration of exposure to radiation is essential. Healthcare professionals should plan and perform procedures efficiently to limit their time in the radiation field. This includes optimizing surgical techniques, using proper imaging guidance, and collaborating effectively with the surgical team.
2. **Training and Education:** Healthcare professionals should receive appropriate training and education on radiation safety, including principles of radiation protection, proper use of shielding devices, and adherence to ALARA principles. Regular training updates and continuing education help ensure awareness of best practices and safety guidelines.
3. **Radiation Monitoring:** Regular monitoring of radiation exposure is essential for healthcare professionals. Personal dosimeters can measure individual radiation exposure and provide feedback to monitor cumulative doses. This information helps professionals assess their radiation exposure and take necessary precautions.
4. **Work Area Safety:** Implementing proper radiation safety protocols in the workplace is crucial. This includes posting radiation warning signs, establishing restricted access zones, and maintaining proper equipment maintenance and quality control.

Education and training

a. **Importance of radiation safety education for healthcare professionals:** Radiation safety education for healthcare professionals is of paramount importance in ensuring the well-being of both patients and medical staff. It equips healthcare professionals with the necessary knowledge and skills to understand the potential risks associated with radiation exposure and implement appropriate safety measures. Here are some key points highlighting the importance of radiation safety education:

1. **Awareness of Risks:** Radiation safety education raises awareness among healthcare professionals about the potential risks associated with radiation exposure. They gain a comprehensive understanding of the biological effects of radiation, including acute and long-term risks, as well as the factors that influence radiation dose and exposure levels. This awareness fosters a culture of safety and promotes a proactive approach to minimize radiation exposure.
2. **Compliance with Safety Guidelines:** Radiation safety education familiarizes healthcare professionals with radiation protection guidelines and regulations. They learn about the principles of radiation protection, safe handling of radiation-emitting equipment, proper use of shielding devices, and adherence to radiation safety protocols. This knowledge ensures compliance with established safety guidelines, reducing the likelihood of unnecessary radiation exposure.
3. **Optimal Use of Imaging Techniques:** Education on radiation safety helps healthcare professionals understand the appropriate utilization of imaging techniques to minimize radiation dose while maintaining diagnostic image quality. They learn about the advantages and limitations of different imaging modalities, optimization strategies, and dose reduction techniques. This knowledge enables them to make informed decisions regarding the selection and optimization of imaging techniques.
4. **Implementation of Best Practices:** Radiation safety education equips healthcare professionals with the best practices for radiation safety in clinical settings. They learn about proper positioning of patients, optimization of imaging parameters, use of dose monitoring tools, and effective communication strategies to ensure a safe environment for both patients and medical staff. By implementing these best practices, healthcare professionals minimize radiation risks and promote a culture of safety within their teams.
5. **Continual Professional Development:** Radiation safety education should be an ongoing process to keep healthcare professionals updated with the latest advancements and guidelines in radiation safety. They should participate in regular training sessions, workshops, and conferences focused on radiation safety to enhance their knowledge, skills, and awareness of emerging trends and technologies.

By providing radiation safety education to healthcare professionals, organizations can create a safe working environment, improve patient care, and reduce the potential risks associated with radiation exposure. It empowers healthcare professionals to take an active role in protecting themselves, their colleagues, and their patients from unnecessary radiation exposure while optimizing the benefits of medical imaging procedures.

b. Training programs on radiation protection and dose reduction techniques: Training programs on radiation protection and dose reduction techniques are essential for healthcare professionals who work with radiation-emitting equipment, such as those involved in spine surgeries. These programs provide comprehensive education and hands-on training to ensure that healthcare professionals are equipped with the knowledge and skills necessary to minimize radiation exposure and implement best practices. Here are some key points regarding training programs on radiation protection and dose reduction techniques:

1. **Basic Radiation Safety:** Training programs start with the fundamentals of radiation safety, including the principles of radiation protection, biological effects of radiation, and regulatory guidelines. Participants learn about the concepts of time, distance, and shielding, which form the basis of radiation protection. This foundational knowledge helps healthcare professionals understand the importance of minimizing radiation exposure and adopting appropriate safety measures.
2. **Equipment Operation and Optimization:** Training programs focus on the proper operation and optimization of radiation-emitting equipment, such as fluoroscopy

machines and CT scanners. Healthcare professionals learn about the functionalities of these devices, how to adjust imaging parameters to optimize image quality while minimizing radiation dose, and how to utilize dose reduction techniques and tools. This training enables them to use the equipment effectively and responsibly.

3. **Patient Positioning and Dose Management:** Training programs emphasize the importance of proper patient positioning and dose management techniques. Healthcare professionals learn how to position patients correctly to obtain the necessary imaging information while minimizing radiation exposure. They also learn about the use of appropriate collimation, shielding, and image acquisition protocols to reduce unnecessary radiation dose. This training ensures that healthcare professionals optimize imaging practices for each patient.
4. **Radiation Monitoring and Dose Tracking:** Training programs cover the use of dosimeters, dose monitoring software, and other radiation monitoring tools. Healthcare professionals learn how to wear and use dosimeters correctly, understand dose monitoring reports, and analyze their radiation exposure trends. They also learn how to utilize dose tracking systems to identify high-exposure procedures and implement corrective measures. This training enables them to actively manage their radiation exposure and make informed decisions based on dose data.
5. **Quality Assurance and Continuous Improvement:** Training programs highlight the importance of quality assurance in radiation safety. Healthcare professionals learn about the significance of regular equipment maintenance, calibration, and quality control procedures to ensure accurate and reliable imaging results. They also gain an understanding of the importance of ongoing education, participation in radiation safety committees, and keeping up-to-date with emerging technologies and best practices. This training fosters a culture of continuous improvement in radiation safety.

Training programs on radiation protection and dose reduction techniques should be comprehensive, tailored to the specific needs of healthcare professionals, and periodically updated to incorporate advancements in technology and regulatory guidelines. By providing such training, healthcare organizations promote a culture of safety, empower their staff to minimize radiation exposure, and enhance the overall quality of patient care.

c. Continuous professional development and updates on radiation safety guidelines: Continuous professional development and updates on radiation safety guidelines are crucial for healthcare professionals working with radiation-emitting equipment. Ongoing education and staying updated with the latest guidelines ensure that healthcare professionals have the most current knowledge and skills to protect themselves and their patients from unnecessary radiation exposure. Here are some key points highlighting the importance of continuous professional development and updates on radiation safety guidelines:

1. **Changing Regulatory Landscape:** Radiation safety guidelines and regulations evolve over time to reflect advancements in technology, emerging research, and best practices. Continuous professional development programs provide healthcare professionals with updates on regulatory changes, ensuring compliance with the latest standards. It helps them stay informed about any new requirements or recommendations that may impact their daily practice.
2. **Advancements in Imaging Technology:** The field of medical imaging constantly evolves, introducing new imaging modalities, techniques, and equipment. Continuous professional development programs keep healthcare professionals up to date with the latest advancements, such as the introduction of low-dose imaging modalities or novel dose reduction techniques. This knowledge allows professionals to leverage new technologies effectively and optimize radiation practices accordingly.
3. **Emerging Research and Evidence-Based Practice:** Continuous professional development programs provide healthcare

professionals with access to the latest research findings and evidence-based practice guidelines in radiation safety. They learn about new studies that may influence radiation dose management, the understanding of radiation risks, and the implementation of dose reduction strategies. Staying updated with the current evidence enables professionals to make informed decisions and deliver the best possible care to their patients.

4. **Quality Assurance and Performance Improvement:** Continuous professional development programs promote quality assurance and performance improvement in radiation safety practices. They provide opportunities for healthcare professionals to assess their own practices, participate in case discussions, and engage in peer-to-peer learning. This process allows professionals to identify areas for improvement, share experiences, and implement strategies to optimize radiation safety and patient care.
5. **Collaboration and Networking:** Continuous professional development programs offer opportunities for healthcare professionals to collaborate and network with colleagues from various disciplines. Interdisciplinary conferences, workshops, and seminars allow professionals to exchange knowledge, share experiences, and discuss challenges and solutions related to radiation safety. Collaboration and networking foster a culture of continuous learning and improvement in radiation safety practices.

Continuous professional development and updates on radiation safety guidelines ensure that healthcare professionals have the necessary knowledge and skills to provide safe and effective care to their patients. It empowers professionals to stay informed about advancements in technology, regulatory changes, and evidence-based practices, promoting a culture of excellence and continuous improvement in radiation safety.

d. Implementation and Compliance: Implementation and compliance are essential aspects of radiation safety in spine surgeries. It involves the practical application of radiation safety measures and ensuring that healthcare professionals adhere to established guidelines and protocols. Here are some key points regarding the implementation and compliance of radiation safety measures:

1. **Standard Operating Procedures (SOPs):** The development and implementation of standard operating procedures are crucial for ensuring consistent and safe practices in radiation safety. SOPs outline the specific steps and protocols to be followed during spine surgeries involving radiation-emitting equipment. These procedures may include guidelines for patient positioning, equipment operation, image acquisition, and radiation dose optimization. Healthcare facilities should establish clear SOPs and ensure that all relevant staff members are trained and knowledgeable about their contents.
2. **Regulatory Compliance:** Healthcare facilities must comply with regulatory guidelines and standards pertaining to radiation safety. These regulations may include requirements for equipment calibration and maintenance, use of personal protective equipment, and adherence to radiation dose limits. Compliance with these regulations is essential to ensure the safety of patients and healthcare professionals. Regular audits and inspections can help monitor compliance and identify areas for improvement.
3. **Interdisciplinary Collaboration:** Radiation safety in spine surgeries involves collaboration among different healthcare professionals, including surgeons, radiologists, anesthesiologists, and radiologic technologists. Effective communication and collaboration among these professionals are essential to ensure proper implementation of radiation safety measures. Interdisciplinary meetings, pre-operative planning sessions, and ongoing discussions can facilitate collaboration and ensure a comprehensive approach to radiation safety.
4. **Quality Assurance Programs:** Quality assurance programs play a vital role in monitoring and maintaining radiation safety in spine surgeries. These programs involve regular assessments, audits, and performance monitoring to ensure that radiation safety protocols are followed

consistently. They may include equipment performance checks, dose monitoring, and evaluation of staff compliance with radiation safety guidelines. Regular quality assurance activities help identify areas for improvement and ensure ongoing compliance with radiation safety standards.

5. **Training and Education:** Adequate training and education are crucial for healthcare professionals to understand and implement radiation safety measures effectively. Facilities should provide comprehensive training programs on radiation safety, covering topics such as radiation physics, radiation protection principles, dose optimization techniques, and proper use of personal protective equipment. Ongoing education and training sessions should be provided to keep healthcare professionals updated with the latest guidelines and advancements in radiation safety.
6. **Continuous Monitoring and Improvement:** Continuous monitoring and improvement are essential to ensure the long-term effectiveness of radiation safety measures. This can involve regular assessment of radiation exposure levels, tracking of radiation-related incidents or complications, and the implementation of corrective measures when necessary. Data analysis and feedback mechanisms can help identify areas of concern and drive improvement initiatives.

By implementing radiation safety measures and ensuring compliance, healthcare facilities can minimize the risks associated with radiation exposure in spine surgeries. This requires a comprehensive approach, including the development of SOPs, regulatory compliance, interdisciplinary collaboration, quality assurance programs, and continuous training and education.

e. **Regulatory frameworks and guidelines for radiation safety in spine surgeries:** Regulatory frameworks and guidelines play a crucial role in ensuring radiation safety in spine surgeries. They provide standards and recommendations that healthcare facilities and professionals must follow to protect patients and staff from unnecessary radiation exposure. Here are some key regulatory frameworks and guidelines relevant to radiation safety in spine surgeries:

1. **International Commission on Radiological Protection (ICRP):** The ICRP is an independent organization that provides guidance and recommendations on radiation protection. They publish reports and recommendations based on scientific evidence and expertise. The ICRP's recommendations include dose limits for occupational exposure and principles for optimizing radiation protection in medical settings.[21]
2. **International Atomic Energy Agency (IAEA):** The IAEA is an international organization that promotes the safe use of nuclear energy and radiation in various fields, including healthcare. They develop safety standards, guidelines, and training materials related to radiation safety. The IAEA's Safety Standards Series includes specific guidelines for radiation safety in medical imaging and interventional procedures.[22]
3. **National Radiological Protection Board (NRPB) - UK:** The NRPB, now part of the Public Health England, is responsible for advising the UK government on radiation protection matters. They have published guidance documents on radiation protection in medical and dental exposures, including specific recommendations for interventional procedures.[23]
4. **United States Nuclear Regulatory Commission (NRC):** The NRC is the regulatory agency responsible for overseeing the civilian use of radioactive materials, including in medical settings. They establish and enforce regulations to ensure the safe use of radiation and protect public health. The NRC provides guidance on radiation safety for medical personnel and facilities, including specific recommendations for interventional procedures.[24]
5. **European Directive 2013/59/Euratom:** The Euratom Directive is a legal framework established by the European Union (EU) to regulate the protection of individuals against the dangers of ionizing radiation. The directive sets out basic safety standards for the medical use of radiation, including requirements for

equipment quality assurance, justification and optimization of procedures, and training and education of healthcare professionals.[25]

6. **National and Regional Guidelines:** Many countries and regions have their own specific guidelines for radiation safety in healthcare settings, including spine surgeries. These guidelines may be developed by national radiation protection authorities, professional organizations, or healthcare regulatory bodies. They provide specific recommendations on radiation dose limits, equipment quality assurance, patient and staff safety, and training requirements.

It is important for healthcare facilities and professionals involved in spine surgeries to familiarize themselves with these regulatory frameworks and guidelines. Compliance with these standards ensures the implementation of best practices in radiation safety, minimizing unnecessary radiation exposure and promoting the well-being of patients and healthcare professionals.

f. **Challenges and barriers in implementing preventive strategies:** Implementing preventive strategies for radiation exposure in spine surgeries can face several challenges and barriers. These obstacles may vary based on factors such as healthcare facility resources, staff compliance, and organizational culture. Here are some common challenges and barriers that can arise:

1. **Lack of Awareness and Education:** Limited awareness and understanding of radiation risks and preventive strategies among healthcare professionals can hinder the implementation of preventive measures. Insufficient training and education programs on radiation safety may contribute to a lack of knowledge regarding dose optimization techniques, proper use of shielding devices, and adherence to radiation safety protocols. Addressing this challenge requires comprehensive and ongoing education initiatives to ensure healthcare professionals are well-informed about radiation risks and preventive strategies.
2. **Time Constraints and Workflow Considerations:** In a fast-paced healthcare environment, time constraints and workflow considerations can impede the proper implementation of preventive strategies. Healthcare professionals may feel pressured to prioritize efficiency over radiation safety, leading to suboptimal practices. Integration of radiation safety measures into existing workflows, efficient communication between team members, and streamlining processes can help overcome this challenge.
3. **Resistance to Change:** Resistance to change can be a significant barrier to implementing preventive strategies for radiation exposure. Healthcare professionals may be resistant to adopting new techniques or equipment due to unfamiliarity or a perceived disruption in their established practices. Overcoming resistance requires effective communication, collaboration, and providing evidence-based information to demonstrate the benefits of preventive strategies in terms of patient and staff safety.
4. **Resource Limitations:** Limited availability of resources, such as radiation protection equipment and personnel, can pose challenges to implementing preventive strategies. Insufficient access to shielding devices, personal protective equipment, and radiation monitoring tools may hinder the consistent application of preventive measures. Healthcare facilities should prioritize resource allocation and consider the long-term benefits of investing in radiation safety infrastructure and equipment.
5. **Organizational Culture and Leadership Support:** The culture within healthcare organizations can influence the implementation of preventive strategies. A culture that prioritizes patient and staff safety, encourages open communication, and fosters a proactive approach to radiation safety is essential. Lack of leadership support or a culture that does not prioritize radiation safety can create barriers to implementing preventive measures. It is crucial for organizational leaders to promote a culture of safety, provide resources and support, and actively engage staff in radiation safety initiatives.
6. **Cost Considerations:** Cost can be a

significant barrier to the implementation of preventive strategies. Upgrading equipment, implementing new imaging techniques, and providing adequate training and education programs can incur expenses. Healthcare facilities may be hesitant to invest in preventive measures due to budgetary constraints. However, it is important to consider the long-term benefits and potential cost savings associated with reducing radiation-related complications and improving patient outcomes.

Overcoming these challenges requires a multi-faceted approach that includes education and awareness, resource allocation, organizational support, and a commitment to a culture of safety. Collaboration among healthcare professionals, administrators, regulatory bodies, and professional organizations can help address these barriers and ensure the successful implementation of preventive strategies for radiation exposure in spine surgeries.

g. **Importance of interdisciplinary collaboration and communication:** Interdisciplinary collaboration and communication are essential in promoting radiation safety and minimizing radiation exposure in spine surgeries. Here are some points highlighting the importance of interdisciplinary collaboration and communication:

1. **Comprehensive Patient Care:** Spine surgeries involve a team of healthcare professionals from various disciplines, including surgeons, anesthesiologists, radiologists, nurses, and medical physicists. Collaborative efforts among these disciplines ensure comprehensive patient care, taking into account not only the surgical procedure but also radiation safety considerations. By working together, healthcare professionals can integrate radiation safety measures into the overall treatment plan, minimizing unnecessary radiation exposure while optimizing patient outcomes.
2. **Shared Knowledge and Expertise:** Each discipline brings unique knowledge and expertise to the table. Interdisciplinary collaboration allows for the exchange of information, sharing of best practices, and integration of diverse perspectives. For example, radiologists can provide guidance on appropriate imaging techniques and dose optimization, while surgeons can share insights on specific surgical approaches and patient positioning. By leveraging the collective knowledge and expertise of the team, strategies can be developed to minimize radiation exposure without compromising the surgical outcome.
3. **Consistent Application of Radiation Safety Protocols:** Consistent application of radiation safety protocols is crucial for effective radiation dose reduction. Interdisciplinary collaboration ensures that all team members are aligned in implementing these protocols, including proper use of shielding devices, adherence to imaging guidelines, and adherence to radiation safety policies. Clear communication among team members promotes a standardized approach to radiation safety, reducing the risk of errors or inconsistencies.
4. **Proactive Identification of Radiation Safety Concerns:** Effective interdisciplinary collaboration allows for proactive identification of radiation safety concerns. By openly discussing and sharing experiences, the team can identify potential risks or challenges related to radiation exposure in spine surgeries. This collaborative approach enables the implementation of preventive strategies and the development of solutions to address specific concerns, enhancing patient and staff safety.
5. **Continuous Quality Improvement:** Interdisciplinary collaboration promotes a culture of continuous quality improvement. By regularly reviewing and analyzing radiation exposure data, sharing experiences, and discussing lessons learned, the team can identify areas for improvement and implement targeted interventions. This ongoing collaboration helps to refine radiation safety practices, optimize imaging techniques, and enhance overall patient care.
6. **Education and Training:** Interdisciplinary collaboration plays a critical role in education and training initiatives related to radiation safety. By working together, healthcare

professionals can develop comprehensive educational programs that address the specific needs of different disciplines. This collaboration ensures that all team members receive appropriate training on radiation safety principles, techniques for dose reduction, and the proper use of radiation protection measures.

Overall, interdisciplinary collaboration and communication foster a team-based approach to radiation safety in spine surgeries. By leveraging the collective expertise, sharing knowledge, and promoting open communication, healthcare professionals can work together to implement effective radiation safety measures, minimize radiation exposure, and optimize patient outcomes.

Future directions: Areas of further research and technological advancements

The field of radiation exposure in spine surgeries is constantly evolving, and there are several areas of further research and technological advancements that can contribute to improving radiation safety. Here are some potential future directions:

1. **Advanced imaging techniques:** Continued research and development of advanced imaging techniques can contribute to reducing radiation exposure while maintaining high-quality images. This includes the exploration of novel imaging modalities, such as intraoperative MRI or ultrasound, that provide real-time imaging guidance with reduced radiation exposure.
2. **Dose optimization algorithms:** The development of dose optimization algorithms specific to spine surgeries can help healthcare professionals tailor radiation doses to individual patient characteristics and procedure requirements. These algorithms can take into account factors such as patient size, complexity of the surgery, and specific anatomical considerations to optimize imaging parameters and minimize unnecessary radiation exposure.
3. **Radiation dose tracking and feedback systems:** Implementing robust radiation dose tracking and feedback systems can provide real-time data on radiation exposure during spine surgeries. These systems can help healthcare professionals monitor and assess their radiation dose levels, identify areas for improvement, and make informed decisions to optimize radiation safety practices.
4. **Radiation protection devices:** Research on the effectiveness of radiation protection devices, such as lead shielding, protective aprons, and thyroid collars, can contribute to the development of more ergonomic and efficient designs. Advances in materials and technology can enhance the effectiveness and comfort of these devices, further reducing radiation exposure for healthcare professionals.
5. **Simulation and training programs:** Simulation-based training programs can play a crucial role in educating healthcare professionals about radiation safety practices in spine surgeries. Virtual reality simulations can provide a realistic environment to practice radiation safety techniques, enhance situational awareness, and improve decision-making skills related to dose optimization and radiation protection.
6. **Long-term studies on radiation-related risks:** Conducting long-term studies to assess the incidence and long-term effects of radiation-related risks, such as cancer development and genetic effects, in patients undergoing spine surgeries can provide valuable insights. These studies can help refine radiation safety guidelines, identify high-risk patient populations, and guide future preventive strategies.
7. **Collaboration and knowledge sharing:** Continued collaboration among healthcare professionals, researchers, and regulatory bodies is essential to exchange knowledge, share best practices, and promote standardized radiation safety protocols. This includes conducting multicenter studies, establishing registries for radiation exposure data, and creating platforms for interdisciplinary discussions and knowledge dissemination.

By focusing on these future directions, the field of radiation exposure in spine surgeries can make significant advancements in radiation safety practices, minimize risks associated with radiation exposure, and improve patient outcomes. Continued research, technological innovations, and collaboration will pave the way for a safer and more effective approach to radiation safety in spine surgeries.

Conclusions

Radiation exposure in spine surgeries is a significant concern due to its potential health risks for both patients and healthcare professionals. The prevalence of fluoroscopic imaging in spine surgeries is growing rapidly, leading to increased radiation exposure. This highlights the need for effective preventive measures to mitigate the risks associated with radiation exposure. Quantification of radiation exposure levels in different spine procedures has revealed significant variations, emphasizing the importance of implementing optimized imaging techniques and dose monitoring systems. Radiation-related complications in spine surgery, including acute effects such as skin injury and radiation sickness, as well as long-term risks like cancer, genetic effects, and cataracts, underscore the importance of minimizing radiation exposure.

The evidence presented in this review highlights the potential adverse outcomes of radiation exposure in spine surgeries, emphasizing the urgency of implementing preventive strategies. Prevention strategies for radiation exposure in spine surgeries involve a multi-faceted approach, including the proper use of shielding devices and personal protective equipment, optimization of imaging techniques, adoption of low-dose imaging modalities, radiation dose monitoring, and comprehensive education and training programs.

The interdisciplinary collaboration and communication among healthcare professionals are essential in implementing and ensuring compliance with radiation safety measures. However, several challenges and barriers exist in implementing preventive strategies, such as lack of awareness, time constraints, resistance to change, resource limitations, and organizational culture. Overcoming these challenges requires concerted efforts from healthcare professionals, administrators, regulatory bodies, and professional organizations. By addressing these challenges and implementing preventive strategies, healthcare professionals can significantly reduce radiation exposure, mitigate associated risks, and improve patient and staff safety in spine surgeries.

In short, a comprehensive and interdisciplinary approach to radiation safety in spine surgeries is crucial to minimize radiation exposure and its potential health consequences. Continued research, education, and collaboration are essential to further advance radiation safety practices and optimize patient care in spine surgery settings.

List of Abbreviations

3D: Three-dimensional;
ALARA: As Low As Reasonably Achievable;
ARS: Acute radiation syndrome;
BMI: Body mass index;
CBCT: Cone-beam computed tomography;
CT: Computed Tomography;
EU: European Union;
IAEA: International Atomic Energy Agency;
ICRP: International Commission on Radiological Protection;
MISS: Minimally invasive spine surgery;
MRI: Magnetic Resonance Imaging;
NRC: National Commission;
NRPB: National Radiological Protection Board;
PPE: Personal Protective Equipment;
PSP: Pedicle screw placement;
SOPs: Standard Operating Procedures.

References

1. Brenner DJ, Hall EJ. Computed tomography--an increasing source of radiation exposure. N Engl J Med. 2007;357(22):2277-2284. doi:10.1056/NEJMra072149
2. Mendelsohn D, Strelzow J, Dea N, et al. Patient and surgeon radiation exposure during spinal instrumentation using intraoperative computed tomography-based navigation. Spine J. 2016;16(3):343-354. doi:10.1016/j.spinee.2015.11.020
3. Tubiana M. Can we reduce the incidence of second primary malignancies occurring after radiotherapy? A critical review. Radiother Oncol. 2009;91(1):4-3. doi:10.1016/j.radonc.2008.12.016
4. Navarro-Ramirez R, Lang G, Lian X, et al. Total Navigation in Spine Surgery; A Concise Guide to Eliminate Fluoroscopy Using a Portable Intraoperative Computed Tomography 3-Dimensional Navigation System. World Neurosurg. 2017;100:325-335. doi:10.1016/j.wneu.2017.01.025
5. Bratschitsch G, Leitner L, Stücklschweiger G, et al. Radiation Exposure of Patient and Operating Room Personnel by Fluoroscopy and Navigation during Spinal Surgery. Sci Rep. 2019;9(1):17652. Published 2019 Nov 27. doi:10.1038/s41598-019-53472-z
6. Preston DL, Ron E, Tokuoka S, et al. Solid cancer incidence in atomic bomb survivors: 1958-1998. Radiat Res. 2007;168(1):1-64. doi:10.1667/RR0763.1
7. Little MP, Wakeford R, Tawn EJ, Bouffler SD, Berrington de Gonzalez A. Risks associated with low doses and low dose rates of ionizing radiation: why linearity may be (almost) the best we can do. Radiology. 2009;251(1):6-12. doi:10.1148/radiol.2511081686

8. Ainsbury EA, Barnard S, Bright S, et al. Ionizing radiation induced cataracts: Recent biological and mechanistic developments and perspectives for future research. Mutat Res Rev Mutat Res. 2016;770(Pt B):238-261. doi:10.1016/j.mrrev.2016.07.010
9. Jolesz FA, editor. Intraoperative imaging and image-guided therapy. Springer Science & Business Media; 2014 Jan 14.
10. Smith H, Welsch M, Ugurlu H, Sasso R, Vaccaro A. Comparison of radiation exposure in lumbar pedicle screw placement with fluoroscopy vs computer-assisted image guidance with intraoperative three-dimensional imaging. The journal of spinal cord medicine. 2008 Jan 1;31(5):532-7.
11. Kaplan DJ, Patel JN, Liporace FA, Yoon RS. Intraoperative radiation safety in orthopaedics: a review of the ALARA (As low as reasonably achievable) principle. Patient Saf Surg. 2016;10:27. Published 2016 Dec 12. doi:10.1186/s13037-016-0115-8
12. Marzouk MM, Afghanyar Y, Marzouk MM, Boussouf SH, Hartung P, Richter M. Comparison of radiation exposure and surgery time between an intraoperative CT with automatic surface registration and a preoperative CT with manual surface registration in navigated spinal surgeries. Eur Spine J. 2022;31(3):685-692. doi:10.1007/s00586-021-07082-x
13. Linden GS, Ghessese S, Cook D, Hedequist DJ. Pedicle Screw Placement in Adolescent Idiopathic Scoliosis: A Comparison between Robotics Coupled with Navigation versus the Freehand Technique. Sensors (Basel). 2022;22(14):5204. Published 2022 Jul 12. doi:10.3390/s22145204
14. Berrington de González A, Mahesh M, Kim KP, et al. Projected cancer risks from computed tomographic scans performed in the United States in 2007. Arch Intern Med. 2009;169(22):2071-2077. doi:10.1001/archinternmed.2009.440
15. Marks LB, Bentzen SM, Deasy JO, et al. Radiation dose-volume effects in the lung. Int J Radiat Oncol Biol Phys. 2010;76(3 Suppl):S70-S76. doi:10.1016/j.ijrobp.2009.06.091
16. Boice JD Jr. NCRP Vision for the Future and Program Area Committee Activities in 2017. Health Phys. 2018;114(2):232-242. doi:10.1097/HP.0000000000000790
17. Delaney G, Jacob S, Featherstone C, Barton M. The role of radiotherapy in cancer treatment: estimating optimal utilization from a review of evidence-based clinical guidelines. Cancer. 2005;104(6):1129-1137. doi:10.1002/cncr.21324
18. Waselenko JK, MacVittie TJ, Blakely WF, et al. Medical management of the acute radiation syndrome: recommendations of the Strategic National Stockpile Radiation Working Group. Ann Intern Med. 2004;140(12):1037-1051. doi:10.7326/0003-4819-140-12-200406150-00015
19. Asano T, Sato K, Onodera JI. United nations scientific committee on the effects of atomic radiation 2000 report. Japanese Journal of Health Physics. 2001;36(2):149-58.
20. Schauer DA, Linton OW. National Council on Radiation Protection and Measurements report shows substantial medical exposure increase. Radiology. 2009 Nov;253(2):293-6.
21. Protection R. ICRP publication 103. Ann ICRP. 2007;37(2.4):2.
22. Rehani MM, Tsapaki V. Impact of the International Atomic Energy Agency (IAEA) actions on radiation protection of patients in many countries. Radiat Prot Dosimetry. 2011;147(1-2):34-37. doi:10.1093/rpd/ncr259
23. National Radiological Protection Board. Guidance notes for the protection of persons against ionising radiations arising from medical and dental use. National Radiological Protection Board; 1988.
24. Mubayi V, Youngblood R. Reevaluating the Current US Nuclear Regulatory Commission’s Safety Goals. Nuclear Technology. 2021 Mar 4;207(3):406-12.
25. Caruana CJ, Christofides S, Hartmann GH. European federation of organisations for medical physics (EFOMP) policy statement 12.1: recommendations on medical physics education and training in Europe 2014. Physica Medica. 2014 Sep 1;30(6):598-603.

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:

Dr Vicentiu Saceleanu, MD, PhD
(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.