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## International Journal of Surgery Case Reports

journal homepage: [www.casereports.com](http://www.casereports.com)

## Endoscopic management of Atypical sellar cavernous hemangioma: A case report and review of the literature

A.M. Al-Sharydah<sup>a</sup>, S.S. Al-Suhibani<sup>a</sup>, S.A. Al-Jubran<sup>a</sup>, A.H. Al-Abdulwahhab<sup>a</sup>, M. Al-Bar<sup>b</sup>, H.M. Al-Jehani<sup>c</sup>, W.M. Al-Issawi<sup>c,\*</sup><sup>a</sup> Radiology Department, Imam Abdulrahman Bin Faisal University, King Fahd Hospital of the University, Saudi Arabia<sup>b</sup> Otolaryngology Department, Imam Abdulrahman Bin Faisal University, King Fahd Hospital of the University, Saudi Arabia<sup>c</sup> Neurosurgery Department, Imam Abdulrahman Bin Faisal University, King Fahd Hospital of the University, Saudi Arabia

## ARTICLE INFO

## Article history:

Received 30 October 2017

Received in revised form

29 November 2017

Accepted 2 December 2017

Available online 8 December 2017

## Keywords:

Cavernous hemangioma

Magnetic resonance imaging

Pituitary tumor

Sellar/parasellar tumor

Vascular malformation

## ABSTRACT

**INTRODUCTION:** supratentorial cavernous hemangiomas, particularly those found in the sellar region, are extremely rare. We present a case of sellar cavernous hemangioma with radiological characteristics that have never been reported. Due to the difficulty diagnosing these lesions, misdiagnosis might occur. Thus, briefing surgeons about the clinico-radiological features of such rare lesions is crucial for better understanding the enigmatic features of such rare lesions and to develop early management approaches that could result in better surgical excision with a lower tendency for complications.

**PRESENTATION OF CASE:** A 43-year-old male presented with headache, blurred vision, and impotence for the last 2 years. Brain magnetic resonance imaging showed an atypical sellar mass displaying signals of heterogeneous intensity on T1- and T2-weighted imaging. The mass exhibited heterogeneous enhancement after gadolinium injection. Endoscopic endonasal surgery was subsequently performed, during which an uneventful subtotal resection of the mass was achieved. Histopathological analysis confirmed the diagnosis of intrasellar cavernous hemangioma.

**DISCUSSION:** Many questions regarding how best to manage such lesions remain unanswered. Hence, we summarize the relevant surgical techniques and discuss misconceptions.

**CONCLUSION:** Sellar cavernous hemangioma (SCH) is an extremely rare lesion that can be misdiagnosed. It is characterized by clinico-radiological features similar to those of other lesions such as pituitary macroadenoma and should be included in the differential diagnosis. The endoscopic endonasal transsphenoidal (EET) approach with subtotal/total resection appears to be a feasible option for debulking, with less surgical complications. Nonetheless, combining stereotactic radiosurgery will reduce postsurgical morbidities.

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## 1. Introduction

Cerebral cavernous malformations (CCMs) are benign vascular lesions with an incidence of 0.5% among all individuals [1]. However, recent advances in neuroradiology techniques have improved the ability to detect vascular malformations [2]. Clinically, affected patients usually present with various signs and symptoms, including headaches, epileptic seizures, intracranial hemorrhage, and focal neurological deficits [3]. However, approximately 50–80% of CCMs are asymptomatic and are usually found incidentally on mag-

netic resonance imaging (MRI) [4]. CCMs were once considered inoperable because their treatment was associated with a high risk of complications; however, various surgical procedures are now performed to treat CCMs. Stereotactic radiosurgery is currently widely used in combination with surgery as a standard of care and has a low incidence of surgical complications [5]. Sellar cavernous hemangioma (SCH) is a very rare lesion, as only a few cases of SCH have been reported in the literature [6–12]. In this report, we describe a case with a CCM located in the sellar region in line with the SCARE criteria [13] that exhibited atypical radiological features that have not been reported previously. We also describe the histopathological characteristics and the surgical approach used to treat this lesion in our academic institution. It considers the first case from a Middle Eastern country.

## 2. Presentation of case

A 43-year-old male with no known medical illnesses, surgical intervention or family history of a similar condition was presented

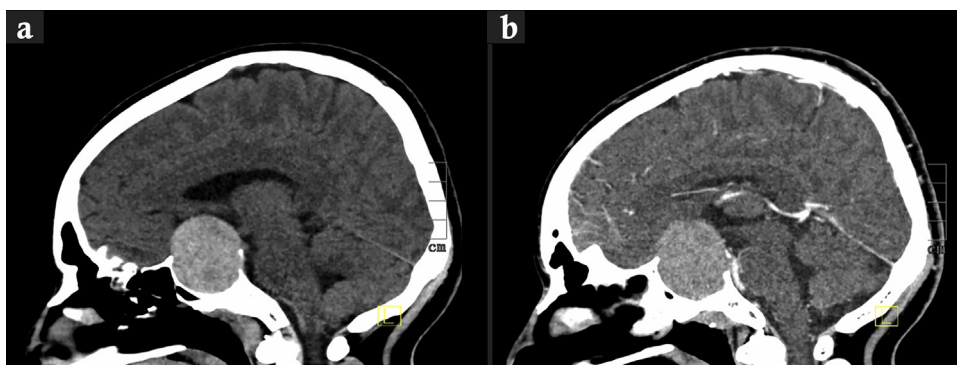
**Abbreviations:** SCH, sellar cavernous hemangioma; EET, endoscopic endonasal transsphenoidal; CCMs, cerebral cavernous malformations.

\* Corresponding author at: Imam Abdulrahman Bin Faisal University (University of Dammam), King Fahd Hospital of the University, Neurosurgery Department, Dammam City, P.O.Box 2998, Eastern Province 31952, Saudi Arabia.

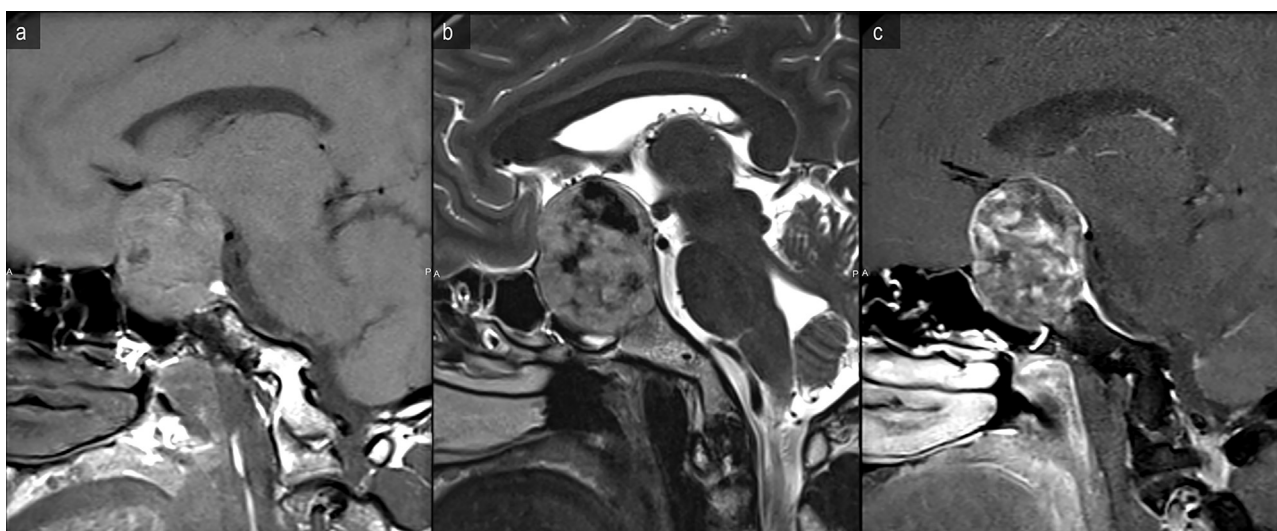
E-mail addresses: [dr.sari@hotmail.com](mailto:dr.sari@hotmail.com) (S.S. Al-Suhibani), [dr.sa3ed@hotmail.com](mailto:dr.sa3ed@hotmail.com) (S.A. Al-Jubran), [ahabdulwahab@iau.edu.sa](mailto:ahabdulwahab@iau.edu.sa) (A.H. Al-Abdulwahhab), [m.albar@hotmail.com](mailto:m.albar@hotmail.com) (M. Al-Bar), [hjehani@iau.edu.sa](mailto:hjehani@iau.edu.sa) (H.M. Al-Jehani), [wmalissawi@iau.edu.sa](mailto:wmalissawi@iau.edu.sa) (W.M. Al-Issawi).

<https://doi.org/10.1016/j.ijscr.2017.12.006>

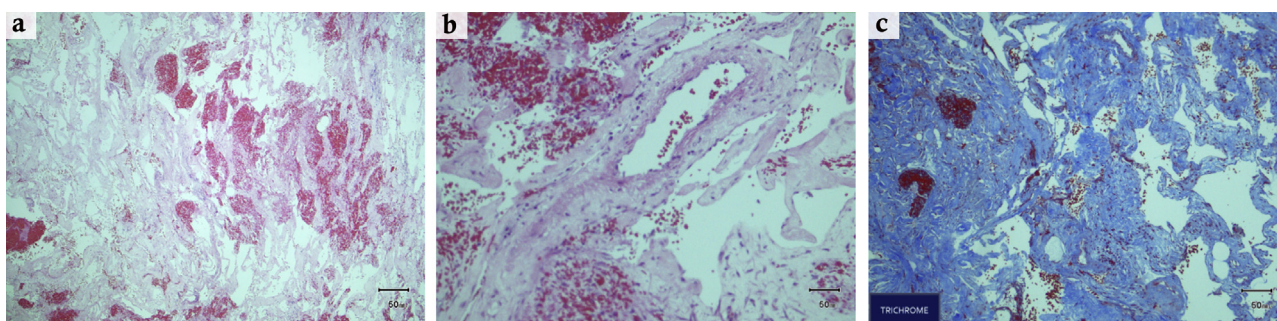
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**Fig. 1.** A) Sagittal non-enhanced CT scan demonstrating a large sellar/suprasellar hyper-dense lesion with superior extension compressing the floor of the 3rd ventricle. B) Post-contrast image showing faint enhancement.



**Fig. 2.** A) Sagittal pre-contrast T1-weighted image showing an iso-dense signal in the gray matter. A bright spot in the neurohypophysis is preserved. B) Sagittal T2-weighted image showing heterogeneous intensity signals and multiple foci of signal voids. C) Post-gadolinium contrast T1-weighted image showing heterogeneous enhancement.



**Fig. 3.** A) Low-magnification image showing dilated blood vessels of varying sizes filled with red blood cells. B) High-magnification image showing thin-walled blood vessels separated by collagen. C) Glial fibrillary acidic protein staining was negative, indicating that no glial tissue was present in the section under examination. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

to the emergency room with headache, blurred vision, decreased libido, and impotence for the last 2 years. The patient reported no history of medications or genetic diseases. Endocrinological and biochemical test results were normal. However, ophthalmological examination revealed decreased visual acuity in the left eye, bitemporal homonymous hemianopsia, and bilateral 6th nerve palsy, and cranial CT showed a large sellar/suprasellar mass (Fig. 1). T1- and T2-weighted imaging demonstrated a large sellar/suprasellar lesion displaying heterogeneous intensity signals and heterogeneous enhancement post-gadolinium injection. The

lesion compressed the brain parenchyma and suprasellar structures (Fig. 2). The patient was treated with an *endoscopic endonasal transsphenoidal (EET)* approach under the supervision of specialized surgeons in the fields of neurosurgery and head & neck surgery. The surgeons encountered a firm and large tumor that was dark red in color and of vascular origin while exposing the sellar floor. A subtotal resection of the lesion was performed without complications. A subsequent histopathological examination confirmed the diagnosis of CCM (Fig. 3). Follow-up examinations during the post-operative period revealed that the patient had hypothyroidism, for

which he was treated with replacement therapy. The patient was satisfied with the surgical operation and discharged from the hospital in good health. He was referred for further management, which included stereotactic radiosurgery and a follow-up evaluation at the endocrinology clinic.

### 3. Discussion

CCMs were initially classified as rare lesions but have recently become a more common finding in multiple locations, especially with recent advances in imaging technology [2]. They are known to reside anywhere within the neuroaxis and are found in both intra- and extraaxial regions. However, CCMs in the extraaxial sellar region are considered exceedingly rare [14]. The term SCH was first used in 1980 referring to a 72-year-old female whose tumor was discovered incidentally on autopsy after she died from breast cancer [12]. Our patient's MRI showed a large sellar/suprasellar lesion with inside-out extension to the neuroaxis. This lesion showed atypical radiological characteristics that have never been previously reported: it is the first lesion to show signals of heterogeneous intensity on T1- and T2-weighted imaging and to exhibit heterogeneous enhancement on T1-weighted imaging post-gadolinium contrast administration. In addition, it considers the first case to be conducted from a Middle Eastern country.

Physicians should be aware of the possibility of SCH when they identify a mass on MRI whose characteristics are consistent with those of a pituitary lesion, such as a pituitary macro-adenoma. This is particularly true in cases in which a highly vascularized lesion is found within the pituitary region. Many questions regarding how to appropriately manage these vascular lesions remain unanswered. In a previously reported trial, Nagai et al. [10] observed that replacement therapy elicited spontaneous improvement in mass. Multiple surgical approaches, including subfrontal [6], frontal [9], and EET approaches, have reportedly been utilized for the treatment of such lesions and have resulted in various outcomes [7,8,11].

The approach used to treat these lesions depends on whether they exhibit extension. We advocate performing an EET approach, which has recently become a standard of care and allows easier access to and more feasible debulking of the sellar mass, especially in cases in which lesions likely extend to the suprasellar cistern and the cavernous sinus. However, managing residual cavernous hemangioma with adjunctive treatments, such as stereotactic radiosurgery, after confirming the diagnosis with biopsy can achieve an excellent outcome and avoid additional morbidities [5]. Additionally, patients should undergo proper follow-up assessments to ensure that they recover safely from their preoperative symptoms and deficits and do not develop additional morbidities.

Reported risks of SCH treatment with surgical resection and implementing radiosurgery compare favorably with the risks of recurrent hemorrhage from a vascular brain tumor. Therefore, the findings of this study have implications for future research, especially for long-term effects of stereotactic radiosurgery, which have not been fully identified [5]. The lack of high-quality studies with long-term follow-up; randomized, prospective, comparative cohort studies; and independent assessments of functional outcomes to assess the effects of our standardized management justify future research to allow surgeons to answer these questions regarding implementation.

### 4. Conclusion

CCMs are rare lesions that can reside anywhere in the neuroaxis. They cause secondary pathology and display variable imaging features. SCH is an extremely rare vascular mass that exhibits enigmatic clinico-radiological features and can be misdiagnosed

as other types of lesions, thus compromising proper management. SCH should be included in the differential diagnosis of pituitary masses such as pituitary macroadenoma. The EET approach is a standardized option for managing this lesion, with less surgical complications. The desired management outcomes can be ensured with early detection, radiosurgery, and proper postoperative follow-up.

### Conflicts of interest

None.

### Funding

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

### Ethical approval

As per the "Imam Abdulrahman Bin Faisal University (University of Dammam) Institutional Review Board," case reports do not require ethical approval or patient consent, provided that there was no intervention and that no patient identifiers appear in the report. Therefore, neither ethical approval nor patient consent was required for this case report. However, Written informed consent was obtained from the patient. A copy of the consent form is available for review by the Editor of this journal.

### Consent

Written informed consent was obtained from the patient. A copy of the consent form is available for review by the Editor of this journal. Consent for publication of the manuscript and the related patient information has been obtained by King Fahd Hospital of the University, Imam Abdulrahman Bin Faisal University.

### Author contribution

A.M. Al-Sharydah wrote the original manuscript. Radiology image reporting was performed and interpreted by S.S. Al-Suhibani, S.A. Al-Jubran and A.H. Al-Abdulwahhab. Surgical notes and the revision of the final manuscript was performed by M. Al-Bar, H.M. Al-Jehani and W.M. Al-Issawi. A.M. Al-Sharydah drafted the paper, and all authors read and approved the final manuscript.

### Guarantor

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

### Acknowledgements

None.

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