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Incidental intraoperative finding of meningioma in spontaneous acute subdural hematoma: A case report from Saudi Arabia and review of literature☆

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ABSTRACT

INTRODUCTION: An association of meningioma with spontaneous acute subdural hematoma has been rarely reported in the literature. Up to date, 38 cases have been reported.
PRESENTATION OF CASE: A 74-year-old Saudi female known case of hypertension presented suddenly with dizziness, headache, and left lower limb weakness for 6 h. No history of trauma or anticoagulant medication.

Computed tomography scan showed acute subdural hematoma. Pre-operative images were negative for any vascular pathology or lesional tissue. The patient underwent surgery for evacuation of the subdural collection. The presence of abnormal soft tissues within the hematoma was discovered during the surgery and sent for analysis. Histopathological results showed meningothelial subtype grade I meningioma. The patient recovered well, with no obvious neurological deficit or immediate complication.
DISCUSSION: Spontaneous acute subdural hematoma without a predisposing factor is a rare occurrence; consequently, a thorough investigation is mandatory in such case to reach the hidden aetiology.
CONCLUSION: In this report a rare association of meningioma with an acute subdural hematoma described. Our case was the first one reported where meningioma incidentally discovered during procedure without preoperative suspicion. A small size intracranial lesion may not be detected by preoperative radiological assessment in the presence of a hematoma. Systematic inspection of the operative field is an important surgical step despite negative preoperative radiological images. Our case supports the mechanism of rupture of abnormal vascular structure. More cases needed to understand the mechanism of such a rare association.

1. Introduction

Meningioma is the most common benign brain tumor accounting for 20% of all primary intracranial tumors [1]. However, reporting their association with hemorrhage is considered to be a rare incidence [2]. Most of these hemorrhages were either intracerebral or intramural hematomas, and their manifestation as an acute subdural hematoma is rarely reported in the literature [2–5]. Up to date, only 38 cases have been reported as meningiomas associated with acute subdural hematoma [7,15,16]. All of them were either diagnosed or suspected preoperatively. The current case was the only one reported as an intraoperative incidental finding of meningioma in acute subdural hematoma. The pathophysiological mechanisms of their hemorrhagic association are not yet fully understood. Therefore, a possible mechanism of such an association was discussed along with reviewing the relevant literature. This is the first reported case from Saudi Arabia. That work has been reported in line with the SCARE criteria [17].

2. Presentation of case

A 74-year-old Saudi female patient, a house wife, known case of hypertension and endometrial cancer who underwent total hysterectomy followed by chemotherapy and radiotherapy four years

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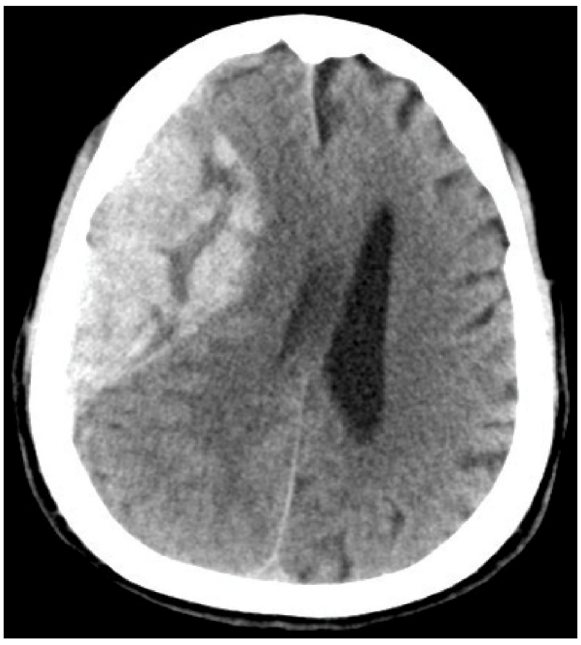

Fig. 1. Pre-operative CT scan showing large right frontotemporal acute subdural hematoma.

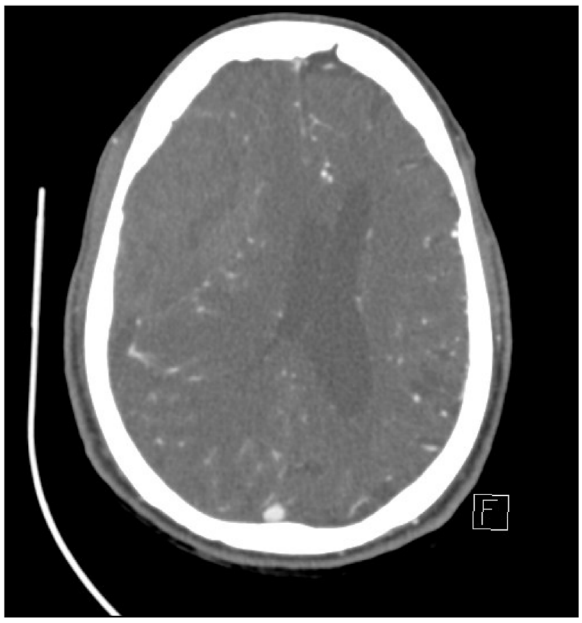

Fig. 2. Pre-operative CT Angiogram doesn't show any vascular or enhancing lesion.

ago. No history of drug abuse or smoking. No psychosocial or family history of medical significance. She presented with a history of sudden dizziness, headache, and left lower limb weakness for 6 h duration. No history of seizure, nausea, or vomiting and no trauma were reported by her or by the relatives.

On clinical examination, she was drowsy with Glasgow Coma Scale of 14/15. There was no external evidence of trauma to her head. Pupils were equal and reactive to light. There was left- side hemiparesis with power of 3/5. Laboratory studies including coagulation profile were within normal limits. Non-enhanced computed tomography scan (CT scan) brain showed large right frontotemporal acute subdural hematoma measuring 4.4 cm in its maximum axial thickness and 7 cm in sagittal plan (Fig. 1). No abnormal masses or tissues were noted in the radiological report. Further investigation to exclude any vascular abnormality was realized. Neither vascular lesions nor enhancing tissues were detected by CT angiography (Fig. 2). Preoperative MRI was not conducted, as the patient was an urgent case.

The patient underwent standard right frontal craniotomy surgery for evacuation of the subdural collection. Incidental abnormal soft tissue was discovered during surgery and sent for histopathology. The lesion size was 2 × 1.5 × 1 cm in dimension, soft in consistency, avascular, grayish in color with small brownish stained regions that might be related to the presence of hemosiderin. A small tortuous vessel was noticed intraoperatively with dual attachments to both the lesion and the cortex that looks like an enlarged feeding vessel. It was hard, sclerotic, grayish to yellowish in color, and no transparency of blood. Histopathology showed a syncytial neoplastic meningothelial meningioma (WHO grade I) with cells that have been arranged in nests and whorls with hemorrhage (Fig. 3). Unfortunately, pictures were not taken during the procedure as it wasn't expected to be a meningioma in the first place.

The patient recovered well, with no obvious neurological deficit or immediate complication from the surgery. Postoperative brain MRI showed no evidence of abnormal soft tissue mass lesion at the resection cavity. Patient has been followed up closely at 3, 6 and 12 months in the outpatient neurosurgery clinic with no reported issues ever since.

3. Discussion

Only 38 cases in the literature reported an association of a spontaneous acute subdural hematoma with meningioma [7,15,16]. The mean age among the reported cases was 61 years (85 years maximum and 32 years minimum), with a female predominance of 57% [15]. Despite our case being a female and falling in the higher age range, however some authors reported that gender and age have no significant correlation to the hemorrhagic event [4,6,10]. Several other predisposing factors like Trauma, hypertension, anticoagulation therapy, Valsalva maneuver, and pregnancy/postpartum were reported [9–11,15]. Hypertension was a suspected predisposing factor in our case, but it has not been reported as the only associated factor in the previous cases. Probably, the lack of documentation in the reported cases made it difficult to link this factor to such a phenomenon.

The role of meningioma location in hemorrhage is considered to be controversial. According to the majority of the authors, the convexity was the most frequent location representing 65% of the cases [7,15]. The convexity was the site of our case as well. Chakis et al. [12] disagreed and believed that it has no influence on the hemorrhagic event. meningothelial meningioma was the most common type representing 47%, including our case [15].

Pre-operative diagnosis of intracranial extra-axial lesions in all of the previously reported cases were recognized or at least suspected depending on radiological signs. However, the discovery of the meningioma in the current case was incidental through the recognition of an abnormal intraoperative tissue appearance. The presence of a hemorrhagic event on CT scan may obscure the existence of an extra-axial lesion. Our case was systematically approached, starting with clinical finding, CT, then CTA, our suspicion was strong so we carefully inspect the operative field. In our case, CTA was conducted preoperatively and showed no enhancing lesion or vascular malformation.

The underlying mechanisms of such an association are still not fully understood; however, many theories have been proposed to explain spontaneous hemorrhage in meningioma. The most common mechanism involves the rupture of the abnormal vascular networks of the tumor based on histological findings such as weak thin-walled vessels and direct erosion of the adjacent vessels by the

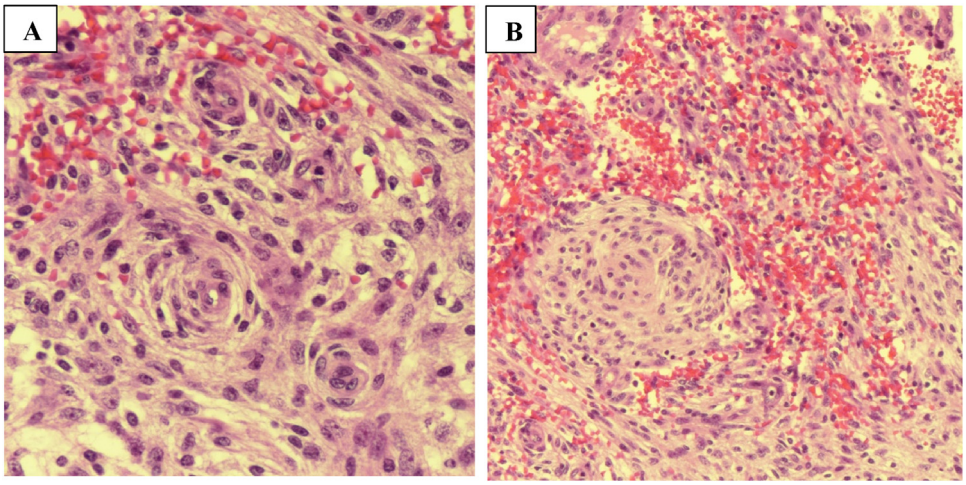

Fig. 3. Histopathological result showing a tumor composed of syncytial neoplastic meningiothelial cells arranged in nests and whorls with hemorrhage. Tumer cells have oval nucleoli, eosinophilic cytomplasm and indistinct cell borders (A) (B).

tumor [12]. Many authors recognized their cases as having similar vascular features and believed that it might have contributed to the hemorrhagic event [8–11]. In our case, the presence of the attached tortuous sclerotic vessel along with its chronicity may lead to an increased pressure inside the vessel which might contribute to its rupture and the occurrence of such a hemorrhagic association. In addition, patient age and hypertension are considered to be risk factors for the development of such a tortuous vessel [13]. Askenasy et al. proposed a theory of enlarged tortuous feeding arteries that become less resistant to blood pressure changes and ultimately prone to rupture under stress which may be the reason in our case [14].

Low number of cases have been reported in the literature is the main limitation of our study. Only 38 cases reported worldwide including our case as a first case from Saudi Arabia. Thus, further case series, to extract more understanding of the pathophysiology of such a rare association, are needed.

4. Conclusion

Presentation of spontaneous nontraumatic acute subdural hematoma in meningiomas is a rare association. Current case is the only one discovered intraoperatively without any preoperative clues. Intraoperative surgical field should be inspected carefully, especially in the absence of a radiological finding suggesting an extra axial lesion. Postoperative Images should be examined thoroughly looking for any abnormal hyperdense lesions that may explain the bleeding tendency. The presence of only a few reported cases challenges authors to extract deep understandable hypotheses regarding the above association. Our case supports the mechanism of rupture of abnormal vascular structure and the presence of hypertension might increase the risk. To have more comprehensible pathophysiological explanation, more cases and larger series are needed.

Declaration of Competing Interest

The authors declare that there is no conflict of interests regarding the publication of this article.

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Ethical

As per the “King Abdullah Medical City in holy Capital, Saudi Arabia,” case reports do not require an ethical approval; provided that there were no patient identifiers appear in the report. However, informed consent was received from the patient to participate in the report.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editor-in-chief of this journal on request.

Author contribution

Management of the case clinically, operatively and follow up: Abdoh. Suggestion of publication: Alhamss. Conception, design and literature review: Abdoh and Alghabban. Acquisition of data: Abdoh, Alghabban. Drafting the article: Abdoh and Alghabban. Analysis and interpretation of data: Abdoh, Alghabban and Farag. Critically revising the article: Abdoh and Farag. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript: all authors.

Registration of research studies

N/A.

Guarantor

The corresponding author Mohammad Ghazi Abdoh, is the Guarantor for this work.

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