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Transitional Meningioma with Intratumoural Bleed and Associated Subdural Haemorrhage: Case Report and a Review of Literature.

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Abstract

Haemorrhage in intracranial tumours is usually associated with malignant primary brain tumours or metastases and rarely with grade 1 meningiomas. We present the case of a 64-year-old woman, who presented acutely and was found to have a convexity meningioma with intratumoral and subdural haemorrhage. A review of the literature has been performed to look at all published cases of meningiomas with haemorrhages, particularly in relation to their subtypes.The association of meningiomas with different types of intracranial haemorrhage has been discussed. Various factors potentially responsible for haemorrhage and association with subtypes has also been discussed.

Introduction

Meningiomas are the commonest benign intracranial tumours and vast majority of these are categorized as Grade 1 by WHO classification. Haemorrhage associated with tumours is usually a phenomenon associated with aggressive tumours such as glioblastomas and metastases, besides some of the grade 2/3 tumours such as oligodendrogliomas [1-5]. Those meningiomas presenting with haemorrhage can be associated with different types of haemorrhage, such as intratumoral, subarachnoid, subdural etc [6,7]. We present a rare case of a Grade 1 meningioma of transitional type, presenting with intratumoral and subdural haemorrhage.

Case Report

A 64-year-old woman, presented with a 2 week history of sudden, severe intermittent headache, mainly over the top of her head and her right temple. The headache was triggered by standing up from a sitting position. Her husband had also noted a recent decline in her short term memory. The patient had a long history of hypertension. Neurological examination and blood tests were normal on admission. No visual disturbance was reported or identified. CT scan revealed a mass with haemorrhage over right sided cerebral convexity, which appeared peripheral and probably dural based. There was some associated subdural haemorrhage over the right sided cerebral convexity Figure 1. MRI scan showed a heterogenous extra-axial lesion measuring 4 x 2.8 cm over the right fronto-parietal convexity with small fluid-fluid levels, again raising the possibility of underlying haemorrhage. On contrast, there was enhancement within the tumour and also of the adjoining meninges, raising the possibility that it could be a meningioma, although due to haemorrhage, other possibilities such as dural based metastasis were considered more likely. Subdural haemorrhage was again confirmed over right frontal and parietal convexity. Due to concerns about metastases, a CT of Chest/Abdomen/Pelvis was performed which was normal. A right frontoparietal craniotomy was performed. The subdural haematoma was evacuated, revealing a grayish-white tumour which had significant adhesion to the dural surface. No large feeding artery was noted. All macroscopic tumour were gradually resected, along with its adherent dura. Histological diagnosis was transitional meningioma WHO grade I, with increased proliferation index. Hence, closer monitoring of the patient was recommended. There were also foci of haemosiderin deposition indicating previous intratumoral haemorrhage, but no evidence of true tumour necrosis was noted. The patient recovered well, with no obvious neurological deficit or immediate complication from the surgery. The patient will be followed up closely in the outpatient neurosurgery clinic post-op.

Review of Literature

Meningiomas are slow-growing, extra-axial tumours which arise from the arachnoid cap cells in the meninges [1]. They are the commonest benign intracranial tumour and

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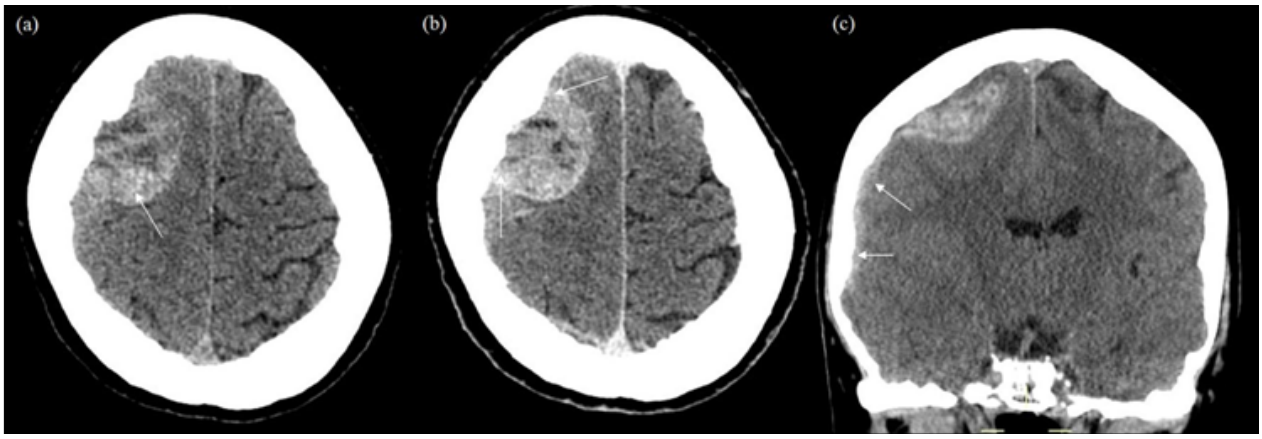

Figure 1: Initial CT scan. (a) Non contrast axial images, (b) Post contrast axial images and (c) Non contrast coronal images. White arrows in (a) show the mass with high areas of attenuation suggesting haemorrhage. White arrows in (b) show enhancement within tumour following contrast. White arrows in (c) show subdural haemorrhage

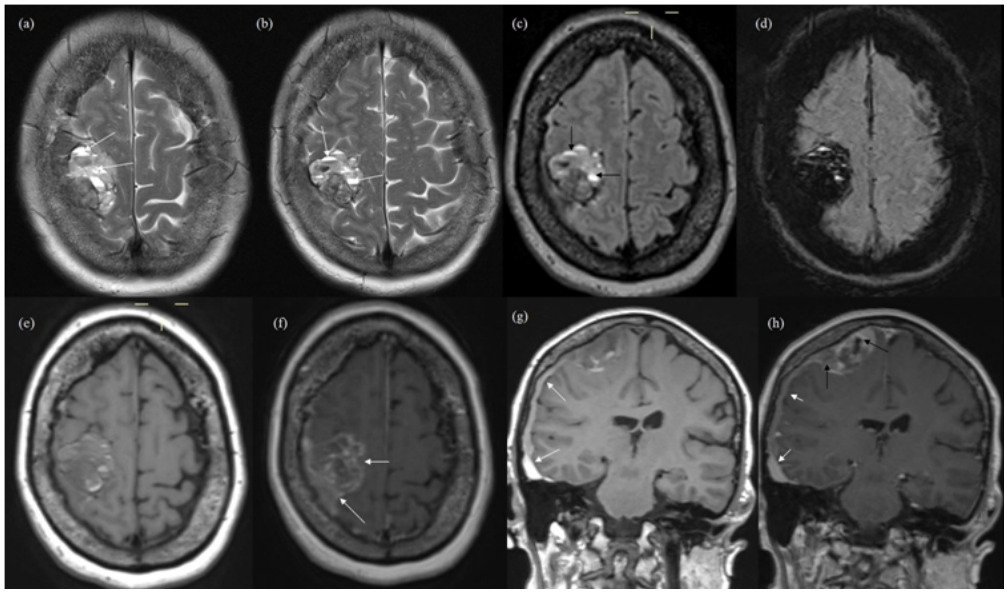

Figure 2: MRI scan. T2-weighted images (a,b) show the mass with several small fluid-fluid levels suggesting areas of haemorrhage (white arrows). FLAIR images (c) also show fluid-fluid levels (black arrows). SWI(susceptibility weighted images) (d) show blooming artefact further suggesting haemorrhage. T1-weighted axial images (e-non contrast and f-post contrast), show areas of enhancement within tumour in (f) as white arrows. T1-weighted coronal images (g-non contrast and h-post contrast) show subdural haemorrhage in (g,h) as white arrows. Enhancement within the tumour is again noted in post contrast images (h) as black arrows.

they constitute about 20% of all primary intracranial tumours [1-3]. Meningiomas are classified into several subtypes in the World Health Organization (WHO) 2016 Classification, with meningothelial being the most common, followed by transitional and fibrous histologies [1,4,5]. Meningiomas are typically benign (90% WHO grade I); however some subtypes with malignant histology and/or rapid growth have also been described (5-7% WHO grade II – atypical, 1-3% WHO grade III – anaplastic) [1,3,5].

Spontaneous intracranial haemorrhage associated with brain tumours is more commonly seen in malignant tumours such as glioblastoma, mixed oligodendroglioma/astrocytoma and metastatic brain tumours [6,7]. No such haemorrhage was reported in large studies of meningiomas done by Cushing and Eisenhardt and Hoessly et al., who examined 313 and 280 cases respectively [9].

More recent studies by Wakai et al., have reported 4 haemorrhages out of 310 meningiomas (1.3%) [6]. Niiro et al., reported 2.0% of meningiomas with haemorrhage and in the series by Martinez-Lage et al., 2.4% incidence of haemorrhage was detected [10]. Helle and Conley described higher bleeding tendencies in angioblastic, fibrous and malignant meningiomas and this trend was equally noted via the Bleeding Propensity Index (BPI) measured by Bosnjak et al., [11,12]. In their review of 145 cases, Bosnjak et al, also found an increased bleeding tendency in two age groups (< 30 years old and > 70 years old) and two tumour locations (convexity and intraventricular) [12].

Haemorrhagic locations commonly associated with meningiomas include intratumoral, intracerebral, subarachnoid and subdural haemorrhages, with many patients presenting with more than one type of haemorrhage [11,13]. Helle and Conley reported that the most frequently seen haemorrhage associated with meningioma was subarachnoid haemorrhage (35%) [11]. The location of meningiomas could also be associated with the type of intracranial haemorrhage. Bosnjak et al, reported that convexity, parasagittal and falcine

tumours mostly produced subdural, intracerebral and intratumoral haemorrhages whereas tumours at other locations mainly resulted in subarachnoid haemorrhage [12].

In the context of radiological findings, Bosnjak et al. described better early identification of haemorrhagic meningioma after the advent of CT scans and this was reflected by the reduced mortality and morbidity rates in CT scanning era [12]. Haemorrhagic meningiomas have been reported by Niiro et al. to characteristically show hyperintensity on T2-weighted imaging on MRI [10]. This was not a feature in our case.

Several predisposing factors have been identified in the literature and these include trauma, hypertension, anticoagulation therapy, Valsalva manoeuvre and pregnancy/post-partum [14-20]. The mechanisms causing haemorrhage in meningioma are still not fully understood; however many hypotheses have been proposed:

Several histopathological findings have shown evidence of abnormally thin-walled intratumoral vessels, intravenous thrombosis and intratumoral necrosis leading to neovascularisation which might have contributed to intratumoral haemorrhages [15,21,22]. Angiographic findings had also revealed enlarged, tortuous feeding arteries which could rupture due to reduced resistance to blood pressure fluctuations, resulting in subarachnoid haemorrhage as described by Askenasy et al [22,23]. Kim et al, described a subdural vein seen directly attached to the tumour in their surgical findings and the stretching of this vein due to tumour growth could have led to subdural haemorrhage[15]. They have also suggested the possibility of subdural haemorrhage from peritumoural vessel rupture due to increase in intratumoral pressure secondary to tumour infarction [24].

Subdural haemorrhage is less commonly seen with meningiomas; making up only 18% and 25% of meningiomas which bled, as reported by Helle and Conley and Martinez-Lage et al., respectively [11,13]. Only 7% of the cases reviewed by Helle and Conley had a combination of intratumoral and subdural haemorrhages [11]. In the review by Bosnjak et al., 42 out of 145 haemorrhagic intracranial meningioma cases (29%) between 1961-2001 were found to present with subdural haemorrhage [12]. A literature review done by Hambra et al, outlined 18 cases of non-traumatic acute subdural haemorrhage between 1988-2013 [25].

We have done a further review on more recent meningioma cases with subdural haemorrhage between 2001-2017; data displayed in Table 1. The average age of patients included in our dataset is 66 years and our data showed an almost equal distribution between males and females (F:M=16:14). Out of the 31 cases, 48% (15 cases) were acute, 13% (4 cases) sub-acute and 35% (11 cases) chronic subdural haemorrhage. Many cases had a combination of acute, sub-acute and/or chronic subdural haemorrhages. However, only less than 10% (3 cases) had bilateral haemorrhages at presentation. 12 out of the 31 cases (38.7%) also reported concurrent intratumoral bleed alongside subdural haemorrhage and only 1 case had a concomitant intraventricular haemorrhage. As shown in Table 2, we found that the majority of meningiomas associated with subdural haemorrhage are located along the convexity (44.8%). This is comparable to the data of Bosnjak et al., which showed 31 out of 42 meningiomas with subdural

x	Authors, Year	Patient (age, gender)	Type of haemorrhage	Location	Tumour histology
1	Nery et al, 2017 [26]	85, F	Chronic SDH	Convexity	Microcystic
2	Ravindran et al, 2017 [27]	36, F	Acute SDH, ITH	Sphenoid	NS
3	Kim et al, 2015 [15]	61, F	Sub-acute SDH, ITH	Frontal	Fibrous
4	Krishnan et al, 2015 [28]	62, M	Sub-acute SDH	Falx	Fibroblastic
5	Hambra et al, 2014 [25]	59, M	Acute SDH, ITH	Sphenoid	Angiomatous and papillary
6	Munivenkatappa et al, 2014 [29]	65, F	Acute SDH, ITH	Convexity	Atypical
7	Levine et al, 2014 [30]	69, M	Sub-acute SDH	Convexity	Grade I
8	Eljebbouri et al, 2014 [31]	51, M	Chronic SDH	Convexity	Meningothelial
9	Rocha et al, 2013 [32]	52, M	Acute SDH	Convexity	Meningothelial
10	Chonan et al, 2013 [33]	67, F	Acute SDH	Convexity	Meningothelial
11	Deprez et al, 2012 [34]	66, M	Acute and sub-acute SDH, ITH	Convexity	Grade I
12	Eom et al, 2012 [14]	75, F	Acute SDH, IVH, ITH	Convexity	Meningothelial
13	Czyż et al, 2011 [35]	69, F	Bilateral chronic SDH	Parasagittal	Fibrous
14	Lakshmi et al, 2010 [36]	73, M	Acute SDH, ITH	Sphenoid	NS
15	Worm et al, 2009 [18]	64, M	Acute SDH	Falx	NS
16	Kashimura et al, 2008 [22]	50, M	Acute SDH	Convexity	Lipomatous
17	Di Rocco et al, 2006 [21]	72, M	Chronic SDH	Convexity	Meningothelial
18	Di Rocco et al, 2006 [21]	74, M	Chronic SDH, ITH	NS	Transitional
19	Mitsuhara et al, 2006 [37]	60, F	Acute SDH, ITH	Petrotentorial	Meningothelial
20	De Silva et al, 2004 [38]	61, F	Chronic SDH	Sphenoid	Meningothelial
21	Bruno et al, 2003 [39]	NS	Bilateral chronic SDH	Convexity	NS
22	Goyal et al, 2003 [40]	66, M	Acute SDH	Falx	Transitional
23	Dallocchio et al, 2003 [41]	73, F	SDH, acute ICH	Parasagittal	Meningothelial
24	Rabinstein et al, 2002 [42]	84, F	Chronic SDH	NS	Meningothelial
25	Lefranc et al, 2001 43]	59, F	Acute and chronic SDH	Frontal	Angioblastic
26	Lefranc et al, 2001 [43]	62, M	Acute SDH, ITH	Frontal	Syncytial/meningothelial
27	Lefranc et al, 2001 [43]	68, F	Acute SDH, ITH	Parietal	Transitional
28	Lefranc et al, 2001 [43]	85, F	SDH, ITH	Frontal	Atypical
29	Taraszewska et al, 2001 [44]	79, F	Bilateral SDH	Convexity	Angiomatous
30	Sinha et al, 2001 [45]	68, M	Chronic SDH	Frontotemporal	NS
31	Sinha et al, 2001 [45]	70, F	Chronic SDH	Convexity	NS

Table 1: Summary of 31 meningioma cases with subdural haemorrhages between 2001-2017
SDH: Subdural Haemorrhage, ITH: intratumoral haemorrhage, IVH: intraventricular haemorrhage, NS: not specified

haemorrhage (73.8%) located along the convexity [12]. In Table 3, our review showed that most meningiomas with subdural haemorrhage are of the meningothelial subtype (43.5%). This is similar to the prevalence reported by Bosnjak et al., (48.6% of meningiomas with subdural haemorrhage), however the Bleeding Propensity Index (BPI) could not be calculated based on our review alone as there is no data on non-haemorrhagic meningiomas to compare [12].

Tumour location	Number of cases (Total = 29)	Percentage of cases (%)
Convexity	13	44.8%
Falx/parasagittal	5	17.2%
Sphenoid	4	13.8%
Petrotentorial	1	3.4%
Others	6	20.7%

Table 2: Distribution of meningioma based on tumour location. *2 cases did not specify location of tumour

Tumour histology	Number of cases (Total = 23)	Percentage of cases (%)
Meningothelial	10	43.5%
Transitional	3	13%
Fibrous	3	13%
Angiomatous/angioblastic	3	13%
Atypical	2	8.7%
Lipomatous	1	4.3%
Microcystic	1	4.3%

Table 3: Distribution of meningioma based on histological subtypes. *8 cases did not specify histological subtype of meningioma.

Due to the rarity of meningiomas with haemorrhage, dural based tumours with a haemorrhagic component are often viewed with suspicion, since it is more commonly seen in aggressive tumours. Such patients are often treated aggressively rather than being followed up with serial imaging only. Preoperatively, it is often difficult to raise the possibility of meningiomas which have haemorrhaged since the odds are heavily in favour of aggressive tumours; however, the possibility can be raised if the tumour is clearly seen as dural based, something that can be difficult to say with confidence in the presence of a surrounding haemorrhage.

Conclusion

Our review has shown a higher prevalence of subdural haemorrhage in meningiomas of meningothelial subtype and those located along the convexity. Our case is more unusual since it is a transitional meningioma with histological and radiological evidence of intratumoral haemorrhage and further radiological evidence of subdural haemorrhage, which is also less often seen. Due to the rarity of such tumours to present with haemorrhage, these are often treated aggressively since preoperative imaging diagnosis of meningioma is difficult to make in this situation. This case report is meant to serve as a reminder of this rare association.

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