CASE REPORT

Cystic meningioma: diagnostic difficulties and utility of MRI in diagnosis and management

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Accepted 26 February 2015

SUMMARY

We report the case of a patient presenting with convulsions in the emergency department. A CT examination revealed a mixed solid and cystic frontal peripheral mass. The intra-axial or extra-axial location and the differentiation between primary or secondary origin were not evident so MRI was performed. The most probable diagnosis was cystic meningioma with intratumoural cysts. Based on MRI findings, the neurosurgeon resected the mass together with its cystic components. The histological report verified the diagnosis and the patient had an excellent outcome.

Because of the acute angle formed by the lesion and the meninges, the relation of this peripheral mass to the meninges was not evident. To better characterise the lesion, MRI (GE Discovery MR 750 3.0T) was carried out (figure 2A–G), which showed an extra-axial location of the mass. Treatment with antiepileptic drugs was started without adverse reaction.

DIFFERENTIAL DIAGNOSIS

Cystic meningioma was the first hypothesis; cerebral metastasis was also included in the differential diagnosis.

TREATMENT

The patient underwent general anaesthesia. A large left frontal craniotomy was performed and the dural plane was opened around the implantation of the lesion, which was bleeding due to arterial feeding branches of the tumour. Once deafferented, the tumour volume was reduced using an ultrasonic surgical aspirator (CUSA) and then dissected near the cerebral parenchyma respecting its pial plane. A complete excision of the lesion was carried out. A dural plasty was finally performed to cover the meningeal defect of the tumour implantation.

OUTCOME AND FOLLOW-UP

The postoperative outcome was favourable with no neurological deficit and the control CT scan excluded surgical complication such as haemorrhage. Three days after surgery, treatment with levetiracetam (Keppra) of 1.5 g/day was started because of an absence type seizure episode. The patient returned home 1 week after surgery without neurological impairment.

DISCUSSION

Cystic meningiomas are rare, accounting for 4–7% of all meningiomas.^{5 6} They can be easily misdiagnosed as metastases, gliomas and hemangioblastomas both macroscopically and by CT or MRI. Analysis of the cyst content cannot help in distinguishing gliomas from meningiomas.² Although there is no significant correlation between cyst formation and any particular histological type,⁷ Weber *et al*⁸ found a correlation between peritumoural cystic meningiomas and atypical histological types.

The imaging features of an extra-axial meningioma are a broad dural base with a tail, the presence of cerebrospinal fluid (CSF) or vascular cleft between meningioma and normal parenchyma, white matter buckling and cortical thinning.⁹ In a study by Goldsher *et al*,¹⁰ three criteria were

BACKGROUND

Meningiomas are the most common extra-axial tumours, accounting for 15% of all intracranial tumours. Although imaging is typical for these lesions, there are many atypical forms, such as cystic meningioma, lipoblastic meningioma and meningioma in atypical positions.¹ Cystic meningioma is rare and more frequently affects men.² It can easily be misdiagnosed as glial tumour, metastasis, neuroblastoma or haemangioblastoma.³ The diagnosis is problematic in cases based exclusively on CT imaging. Although there are less dilemmas with MRI, diagnosis is not obvious in 20% of cases.⁴ The purpose of this article is to review the classification of cystic meningiomas, to discuss the diagnostic tools that can help to differentiate the various types and to emphasise the importance of preoperative diagnosis.

CASE PRESENTATION

A 48-year-old woman was brought to the emergency department after an epileptic crisis. Her medical history was unremarkable. Clinical examination and laboratory tests were within normal limits.

INVESTIGATIONS

A CT scan was performed (GE Healthcare 64-slice, dual-source scanner). On unenhanced images, a large, complex, peripheral mass was found, located in the left frontoparietal region (figure 1A–F). Hypodense cystic components were depicted in an otherwise isodense mass. After iodinated contrast injection, an intense enhancement of the solid component as well as a perilesional oedema was noted. Cerebral perfusion showed elevated cerebral blood flow and cerebral blood volume in the lesion. Glioma, metastasis and meningioma were retained in the differential diagnosis.

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To cite: Sotiriadis C, Vo QD, Ciarpaglini R, *et al*. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2014-208274

Figure 1 (A) On non-contrast CT imaging, a heterogeneous, well circumscribed, peripheral frontal lesion is visible. (B and C) After intravenous contrast injection, intense enhancement of the mass is noted except for some well-delimited regions, which are hypodense. There is always an acute angle between the mass and the meninges and the intra-axial or extra-axial origin of the lesion is not obvious. (D) CT-perfusion imaging shows obvious elevation of cerebral blood flow. (E) CT-perfusion imaging shows obvious elevation of cerebral blood volume. (F) No adjacent bone reaction is visible.

proposed for the definition of the tail: presence on at least two consecutive images, greatest thickness next to the tumour with tapering away from it and enhancement greater than the

tumour itself. The 'mother-in-law' type of enhancement in conventional angiography is characteristic (early homogeneous enhancement with persistence in late phases).

Figure 2 (A) MRI, sagittal T2-weighted image, reveals an extra-axial, heterogeneous, frontal lesion associated with adjacent meningeal alterations indicating meningioma. No obvious vasogenic oedema of the adjacent parenchyma is reported. Multiple isointense to cerebrospinal fluid (CSF) components are found inside the tumour. (B–D) Gadolinium injection reveals two components, one with intense enhancement and the other without. The non-enhancing regions are well-delimited cystic components. Note tapering of the meninges away from the tumour. (E) Diffusion-weighted imaging shows high signal of the enhancing component. (F and G) Apparent diffusion coefficient (ADC) maps show slightly lower ADC value of the intratumoural cysts compared with the CSF.

Although multiplanar reconstruction on CT or MRI may differentiate extra-axial from intra-axial tumours, a meningioma can mimic an intra-axial lesion when invaginating in the brain.¹¹ Cysts related to meningiomas can be located inside the tumour or in its vicinity. Rengachary *et al*¹² classified

meningioma cysts as intratumoural or extratumoural based on whether the cyst is lined by meningotheial cells. Various authors have proposed several pathophysiological mechanisms for cyst formation. For intratumoural cysts, the following mechanisms were proposed: ischaemia with necrosis and cystic

degeneration, haemorrhage with cystic changes and active secretion of fluid by tumour cells. Concerning extratumoural cysts, glial proliferation, parenchymal oedema with evolution into cyst formation and CSF entrapment were described.^{4 13 14}

Nauta classified cystic meningiomas into four types (table 1). In type 1, the cystic component is entirely contained within the tumour and centrally located. In type 2, the cyst is wholly intratumoural but peripherally located and encapsulated by tumour cells. In type 3, the cyst is peripherally located and lies in the adjacent parenchyma. In type 4, the cyst is limited to the interface between the tumour and the brain as loculation of the CSF in the subarachnoid space, so it is not located in the tumour nor in the cerebral parenchyma.¹⁵

Jung *et al*⁵ added type V as a mixed type of meningioma combining types I and III.

El-Fiki *et al* correlated the location of the cyst and its content and also described four types (table 2).

In type A, the cyst is extratumoural and contains clear water or CSF-like fluid. It can be subdivided into type A1 when entrapped CSF is found and the cyst is surrounded by an arachnoid membrane, and into A2 when the content is the result of an excretory process of the tumour cell or an exudation without the presence of arachnoid separation. In type B, the content is a xanthochromic fluid and in B1 the cyst is extratumoural without a definite wall, while in B2 the fluid is surrounded by a thin rim of enhancing tumour or enhancing nodule. An intermediate degree of oedema accompanies the cyst. In type C, dark brown or yellow dirty intratumoural fluid was found accompanied by marked peritumoural oedema. Type D includes meningiomas with the presence of both clear extratumoural or peritumoural cysts and small dark brown intratumoural cysts.¹⁶

Moreover, El-Fiki correlated his classification system with Nauta's and noted that type 1 corresponds to type C, type 2 to type B2, type 3 to B1 or A2 and type 4 to A1 (table 3).

The classification of cystic meningiomas is very important as it affects the surgical strategy. Preoperative identification, especially of type II and III cysts, is necessary as removal of the whole capsule with the cyst is mandatory in type II while tumour resection only without the cyst is the method of choice in type III.¹⁷ Many authors have noted tumour recurrence due to incomplete resection of the cyst capsule in type II cystic meningiomas. However, differentiation of the two types is not always obvious on the basis of MRI and so microsurgical inspection or histological examination of the cyst wall is mandatory.¹⁷ It is worthy to mention that the cyst wall is not identifiable on microsurgical inspection in type III, whereas a thin, easily detachable capsule is compatible with type II cystic meningioma. According to Jung *et al*,⁵ biopsy of the cyst wall should always be carried out even if there is no contrast enhancement in preoperative MRI. An imaging feature that may help in differentiating types II and III is that the margins of the solid tumour towards the cyst are well defined when the cyst is extratumoural (type III), whereas the margins are usually irregular with the intratumoural location of the cyst.⁷

Table 1 Nauta's classification of cystic meningioma

Type I	Cyst entirely contained within the tumour, centrally located
Type II	Cyst entirely contained within the tumour, peripherally located
Type III	Cyst peripherally located relative to the tumour but lying in the adjacent parenchyma
Type IV	Cyst peripherally located but lying between the tumour and the adjacent parenchyma

Table 2 El-Fiki's classification of cystic meningioma

Type	Content	Location
Type A	Clear water or CSF-like fluid	A1 Extratumoural, CSF surrounded by arachnoid membrane
		A2 Extratumoural, clear water without the presence of arachnoid membrane
Type B	Xanthochromic fluid	B1 Extratumoural, no wall
		B2 Intratumoural, thin rim of enhancing tumour surrounding the cyst or enhancing nodule
Type C	Dark brown or yellow dirty fluid	Intratumoural
Type D	Contains both clear-water, extratumoural or peritumoural cysts and dark brown intratumoural small cysts	
CSF, cerebrospinal fluid.		

As mentioned previously, MRI appearance of meningeal cyst is not indicative of the type of meningioma.¹⁷ Chen *et al*⁹ tried to correlate the MRI and especially diffusion-weighted imaging (DWI) features of cystic meningiomas with the histopathological findings. They found a lower Apparent diffusion coefficient (ADC) ratio in two cysts of type I and a higher ratio in one type III, and they believe that DWI may play an important role in preoperative classification of these cysts.

In our case, histological analysis revealed a microcystic meningioma World Health Organization grade I (figure 3A, B). Tumour cells focally express epithelial membrane antigen (figure 3C). They do not express receptors for progesterone. Mitoses are rare (between 1–2 mitoses per 10 fields) and proliferation index is less than 5% (figure 3D).

On CT scan, the diagnosis of a meningioma was quite difficult due to the presence of the cysts, which changed the morphology of the tumour. In coronal and sagittal reconstructions, acute angles with meninges were noted. The buckling of the white matter was not obvious and no CSF-cleft was found. The formation of the cyst gives the lesion a mushroom shape, which is responsible for the invagination in the cerebral parenchyma. The only finding suggesting a meningioma was the intense contrast enhancement and the contact of the mass with the meninges. In contrast, on MRI, all the characteristics of a meningioma were found: dural tail, CSF cleft, white matter buckling. Also, there was no doubt as to the intratumoural location of the cysts (type II), which had a slightly lower ADC value ($2.82 \times 10^{-3} \text{ mm}^2/\text{s}$) than the CSF itself ($3.32 \times 10^{-3} \text{ mm}^2/\text{s}$). The enhancing component showed restriction on DWI.

The differential diagnosis of these lesions depends on the location. For supratentorial masses, high-grade gliomas and metastases have to be included. In infratentorial lesions, schwannomas with cystic degeneration, metastases and hemangioblastoma in adults and pilocytic astrocytomas in children have to be considered.

Table 3 Correlation of Nauta's and El-Fiki's classification

Nauta's	I	II	III	IV
M El-Fiki's	C	B2	B1 or A2	A1
Necessitating surgical excision of the cystic component	Yes	Yes	No	No

Figure 3 (A and B) Hypocellular tumour tissue with a conspicuous loose-textured quality (glioma-like), vascular hyalinisation, and scattered vacuolated cells with distinct cell borders and large hyperchromatic nuclei. These findings were compatible with cystic meningioma. (C) Epithelial membrane antigen (EMA) staining showing weak and diffuse membranous expression for EMA. (D) MIB-1 was less than 5%.

Learning points

- ▶ CT is insufficient for the diagnosis of cystic meningioma.
- ▶ MRI is necessary not only for the diagnosis but also for the classification of the cysts.
- ▶ Differentiation between type II and III cystic meningioma is not always obvious in preoperative imaging. In such cases, neurosurgeons have to perform a biopsy in order to decide whether the cyst should be resected with the tumour or not.
- ▶ In cases of known primary tumour, cerebral metastasis has to be included in the differential diagnosis.

Contributors CS conceived the idea, performed the literature research, noted the patient's history and wrote the article. QDV wrote the article. RC performed the surgery and wrote the article. HMH made corrections and suggestions, and wrote the article.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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