



## PSAMMOMATOUS PSEUDOTUMOURAL INTRACEREBRAL CALCIFICATION: A CASE REPORT AND REVIEW OF THE LITERATURE

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### Objective and importance:

We present the case of a patient with a very unusual intracerebral pseudotumoural calcification and describe the pathological findings, speculate on possible aetiology and review the related literature.

### Case presentation:

A thirty-one year old man with epilepsy had presented with left hemiparesis and an increase in the frequency of his seizures. Blood tests did not reveal any electrolyte or metabolic abnormality. The patient had a magnetic resonance imaging (MRI) scan of the head which showed a cortical-subcortical sharply circumscribed hypo-intense lesion on both the T1 and T2 weighted scans, in the medial third of the right inferior frontal gyrus (F3 gyrus). Following a right frontal craniotomy the lesion was excised completely. On microscopy the lesion exhibited a lamellar concentric (psammoma-like) pattern of calcium deposits in brain parenchyma.

### Discussion:

There has been no previous report in the literature of the type of pseudotumoural calcification we have reported. The aetiology or pathogenesis of this type of lesion is not known; however, in our experience surgical removal of it was associated with a very favourable outcome.

*Psammoma • Cerebral • Pseudotumoural calcification • Good prognosis*

We present the case of a patient with a very unusual intracerebral pseudotumoural calcification. and describe the pathological findings, speculate on the possible aetiology and review the related literature.

### Clinical presentation

A 31 years old man presented to our neurosurgical clinic with a left hemiparesis and an increase in frequency of his generalised tonic-clonic seizures. The patient was 15 years old when he had his first seizure and this had been kept under control with phenytoin. Prior to this admission no imaging studies of his head had been obtained. The patient's current problems

had developed after the family physician had noted gingival hyperplasia and replaced phenytoin with carbamazepine.

The T1 weighted MRI scans of the head revealed a small (2-3 cm in diameter), sharply circumscribed cortical and subcortical hypointense mass with no contrast enhancement, located in the right frontal lobe under the F3 gyrus; however there was enhancement of surrounding area (fig. 1a, b). The lesion was also hypointense on T2 weighted MRI scan with hyperintensity of the surrounding brain. As the MRI findings were suggestive of the lesion being probably a tumour with calcification, we did not obtain a further

computed tomography (CT) scan of the head, even though the calcified lesions are better demonstrated by this latter method.

The patient was managed surgically with right frontal craniotomy and complete excision of the lesion.

On macroscopic examination the lesion was a whitish, circumscribed, 1.5 cm in diameter hard mass (fig. 2). It was necessary to decalcify it in a 5% aqueous solution of hydrochloric acid for 5 days prior to histological processing.

On microscopic examination there were multiple concentric lamellar structures, akin to psammoma bodies ranging from 3-5 to over 300 micrometers in diameter. These rounded structures were separated by thin cords of nervous tissue showing only patchy, very discrete gliosis. No association of calcium deposits to the vessels was noted. The calcifications appeared basophilic on routine hematoxylin–eosin staining (fig. 3). On Masson's trichrome staining a matrix of collagen and various glycoproteins were visible (fig. 4). There was positive reaction to Periodic acid-Schiff (PAS) staining also (not shown). At the periphery of the psammomatous agglomeration a denser astrocytic gliosis was conspicuous. A histochemical stain for detecting iron [Mallory's modified reaction, as described elsewhere (23)] was performed, which was negative.

A year later the patient was well and a CT scan performed for follow-up showed no local recurrence or other pathological changes. Since the operation the patient has been free of seizures and has not required anticonvulsant therapy.

## Discussion

On literature search with PubMed (19) we were unable to find any previous report of a similar lesion to that we have described.

The findings of the preoperative MRI studies and some of the intra-operative macroscopic appearances of the lesion were suggestive of several types of intra-axial calcified neoplastic tumours such as astrocytoma, pleomorphic xanthoastrocytoma, oligodendroglioma, ganglioglioma, dysembryoplastic neuroepithelial tumour, angioma (8) and even lymphoma (4, 18); however, on microscopy there was no evidence of any neoplastic cells.

The striking feature on microscopy was of the lamel-

lar concentric pattern of calcium deposits. These were similar in appearance to psammoma bodies, mostly encountered in meningioma. The pathogenesis of calcium deposition in the typical pattern of psammoma bodies, in any location, is not known. Osteopontin, a major phosphorylated glycoprotein normally present in bone and produced by CD68-positive macrophages found in meningiomas may play a significant role in the development of psammoma bodies in these tumours (9). Psammoma bodies contain calcium phosphate deposits (9). However, we had not chemically analysed the nature of the calcium deposits in our lesion.

Certain psammomatous meningiomas can mostly be composed of psammomatous bodies with only a few cellular elements (12). However, this is unlikely to be the nature of the lesion in our patient as on microscopy no meningothelial cells or any other features suggestive for such a tumour were noted; besides, the lesion was intraparenchymal [though very rare cases of intraparenchymal meningiomas have been reported (11, 16, 25) in the literature, they are not of the psammomatous type].

Psammoma bodies do occur extracranially in relation to certain neoplasms but, not likely to be relevant in our case. Extracranial psammoma bodies can be rarely be found in association with uterine serous carcinoma, serous carcinoma of the ovary or peritoneum (3).

Deposition of calcium in abnormal tissue, defined as dystrophic calcification, can also exhibit scattered calcium pyrophosphate deposits. Such changes at the site of a previous traumatic or vascular event may result in a cerebral scar that can undergo calcification even after many years from the initial injury. However, no records of such injury were found in the past medical history of the patient; furthermore the surrounding parenchyma did not show any features of previous rich glial proliferation.

Intracranial calcifications could be due to vascular malformations. However, no evidence of such a lesion was noted on microscopy.

In our patient, the pseudotumoral calcification could be due to mineralization of some previous infectious process located in the frontal lobe. Rarely, some parasitic (neurocysticercosis), bacterial (abscesses) or fungal lesions may accumulate mineral salts. However, the clinical data and histological findings in our

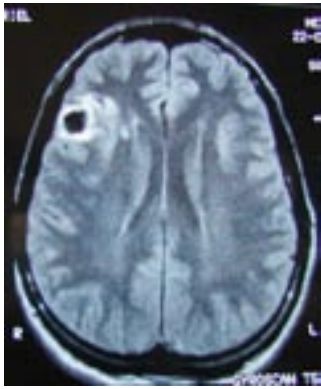


Figure 1a

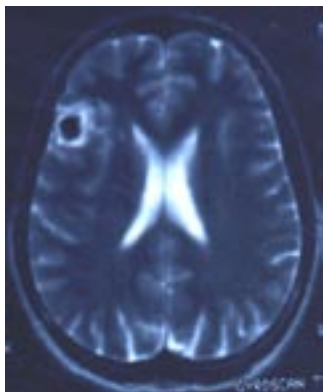
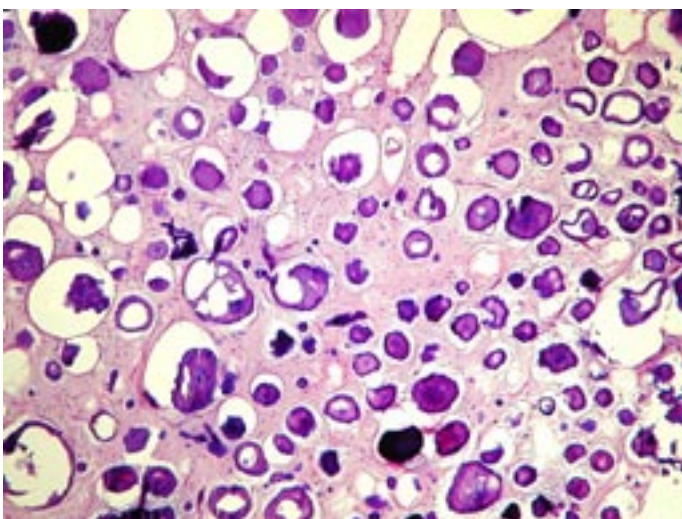


Figure 1b

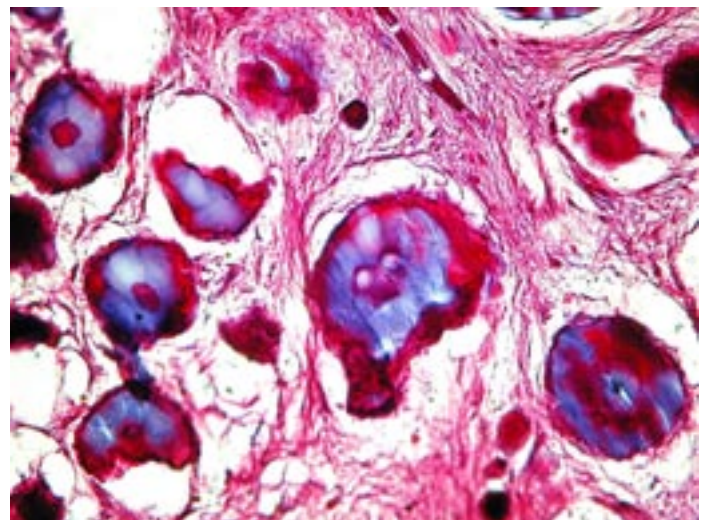
**Figure 1a.** T1 weighted MRI scan with contrast; right frontal hypo-intense lesion with surrounding contrast enhancement. **Figure 1b.** T2 weighted MRI scan: right frontal hypodense lesion with surrounding hyper-intensity.



**Figure 2.** Macroscopic view of the lesion; it had a diameter about 1.5 cm.



**Figure 3.** On microscopy (H&E stain;  $\times 10$ ) the lesion was composed of abundant calcified, psammoma-like structures of variable diameter which were intensely basophilic and surrounded by normal brain parenchyma.



**Figure 4.** On additional microscopy ( $\times 40$ ; Masson's Trichrome), the intermingling parenchyma appears normal. There is some oedema evident but no astrocytic gliosis.

case do not offer any support for such a hypothesis.

There can be non-neoplastic intracranial calcifications in neurofibromatosis type 2 (17); however, this type of calcification usually occurs in cerebellar cortex, choroid plexus or on the surface of cerebrum (17); furthermore, the patient did not exhibit any features consistent with criteria for neurofibromatosis type 2 (7).

Systemic non-neoplastic pseudotumoral calcification could occur in disturbances of calcium or phosphate metabolism (22). These apparently *de novo* lesions are defined as metastatic calcification. The serum calcium and phosphate levels were within normal range in our patient. In the condition known as 'tumoral calcinosis' there is extra-skeletal calcification but the deposits do not exhibit psammoma bodies on microscopy (14, 15, 20, 21, 26).

The common location of metastatic calcification is usually in the soft subcutaneous periarticular tissues (13). Some unusual presentations can be found in various sites and, in regards to the nervous system is frequently seen in the spine, mostly as extradural masses resulting in spinal cord compression (5, 6, 10), some rare cases in the meninges (27) and rarely even in the vertebrae (2).

There have been reports of tumoural calcinosis related to spinal meninges (5, 6, 10, 27) and in an unusual case of a calcifying tumoural lesion in the brain (1), described as a "pseudotumor calcificans". In this latter case the lesion was complex with fibro-osseous proliferation but containing no psammoma-like structures.

Fahr disease is a very rare condition where there is intraparenchymal calcification in the basal ganglia and dentate nucleus or the choroid plexuses (14).

Singh et. al, (24) have reported focal cortical-subcortical calcification in patients with epilepsy; however, they did not report of any pathological features; their study was based on EEG and radiological findings; cortical dysgenesis and heterotopias they report can become calcified without becoming a tumoural mass.

## Conclusion

The histological type of psammoma-like pseudotumoral calcinosis we report is unusual and previously not reported. Imaging and macroscopic studies were suggestive of a neoplastic entity; however, microscop-

py revealed it to be a non-neoplastic lesion. The cause or pathogenesis of this type of lesion is not known. Surgeons must be aware of this type of lesion because of its rarity and of the good prognosis following its radical removal, which is uncomplicated.

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