Case Report

Capsula Eburnea

A CURIOUS CASE OF MENINGIOMA WITH A CONCURRENT ABSCESS IN A DISTANT BRAIN AREA

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SUMMARY

Brain abscess continues to be a life-threatening disease. On rare occasions, this pathology can be concurrent with an intracranial neoplasia; to date, only 27 cases have been reported in literature. Intratumoral bleeding/necrosis or gaps in the blood brain barrier are supposed to facilitate a bacterial/fungal superimposition by haematogenous spread from a distant infective focus. In this report, we describe a patient with a subclinical meningioma who developed a brain abscess in a distant cerebral region. We discuss the influence of his multiple risk factors, and whether or not the neoplasia might be considered co-responsible in the pathogenesis of the abscess.

Introduction

Brain abscesses represent a relatively rare disease, accounting for 2% of intracranial masses (8% in developing countries). Male/Female ratio is 1.3/1 to 3/1 [1, 2]. Clinical findings are nonspecific. The most frequent signs and symptoms are headache, nausea, vomiting, fever, consciousness alteration, seizures, and motor weakness. Classical triad (headache, fever and neurological deficit) is present only in 15-30% of patients [3]. Evolution is generally faster than observed in brain tumours.

Lumbar puncture is contraindicated because increased intracranial pressure can cause cerebral herniation [3]. Since there are no pathognomonic signs, CT and MR images are mandatory for the right diagnosis. Imaging features depend on the stage of the lesion: after an initial phase of cerebritis, in mature 10-14 day-old abscesses (suppurative inside with a well-defined capsule) CT and T_1-weighted MRI scans show uniform ring enhancement after administration of contrast medium; while T_2-weighted MR images show hyperintense signal in the cavity with a hypointense rim surrounded by hyperintense perilesional oedema. Diffusion weighted MR images help differentiate abscesses from tumoral lesions: usually, the former have an increased signal diffusion image with a reduced apparent diffusion coefficient (ADC), whereas the latter have a decreased signal diffusion with high ADC [4, 5].

There continues to be a lack of consensus regarding the best management of cerebral abscesses, in respect to surgical indications, type of surgical procedure and antibiotic of choice [2, 6, 7]. Conservative treatment is effective whenever the etiological agent is known and antibiotic therapy is specific, but, unfortunately, this is not always the case. The definitive microbiological diagnosis, which is necessary to set up a specific antibiotic therapy, often arises from the direct analysis of the specimens obtained by surgical aspiration, excision of the lesion, or from a biopsy of its wall [7, 8, 9].

Indeed, advances in neuroradiology, surgery, and pharmacology have improved outcome, decreasing mortality from 40-60% in the pre CT era to 5-25% nowadays [10].

In our report, we describe a patient with a subclinical meningioma, who developed a

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brain abscess in a different cerebral region. The influence of his multiple risk factors is discussed, as well as arguments in favour or against the theoretical co-responsibility ascribed to the tumour in the formation of the abscess.

Case presentation

A 72-year-old man was referred to the Emergency Department of our hospital because of confusion (arisen 5 days before) along with a 24-hour history of loss of movement coordination and weakness of his left arm.

In anamnesis, the patient reported a history of diabetes, recurrent sinusitis, and left lumbosciatalgia chronically treated with corticosteroids. His surgical history was negative, except for multiple minor surgical procedures, such as bilateral turbinectomy, excision of dorsal cutaneous nevi, and extraction of the lower wisdom teeth in the past. Less than a month before admission, he had undergone an uneventful extraction of the upper left premolar, preceded by routine administration of antibiotic prophylaxis. One week before hospitalization, a sense of mental confusion and difficulty concentrating had appeared, along with progressive troubles in handling objects with his left hand.

On admission, the patient was found alert with a Glasgow Coma Scale score of 15. A 7^{th} left cranial nerve deficit and a slight weakness of his left hand were present. The remainder of the neurological examination was normal.

A cerebral CT revealed the presence of two concurrent and distinct lesions: the first one was 37 mm in diameter, round, deep median frontal, inhomogeneous, with a slightly hypodense central area and an homogeneous post-contrastographic enhancement; the second one was 25 mm in diameter, superficial right frontoparietal, much more hypodense than the first one, with a defined isodense rim and surrounded by perilesional oedema. MRI scans showed that the deep median frontal lesion was isointense with the cerebral cortex, with a inhomogeneous central area and a homogeneous enhancement after administration of gadolinium (Fig. 1); while the superficial right frontoparietal lesion was hyperintense on diffusion weighted image, with a post-contrastographic ring enhancement (Fig. 2). The first lesion was certainly a sphenoidal ridge meningioma, while the second one had features compatible with a brain abscess.

Under the indications of a specialist in infective diseases, a third generation cephalosporins and metronidazole therapy was started. An echocardiogram and a chest x-ray were performed to exclude other possible origins of haematogenous bacterial spread: both were negative. Three hemocultures were also negative. A neuronavigation-guided aspiration of the frontoparietal lesion was performed, collecting purulent material from which a few colonies of an anaerobic Streptococcus strain grew on microbiological cultures . Pharmacological therapy was then optimized according to the indications obtained by the antibiotic essay, and scheduled to be continued for the following three months. Three weeks after the neuronavigation-guided aspiration the patient was completely asymptomatic. A cerebral CT confirmed the disappearance of the abscess with slight residual oedema in the right frontoparietal area, and, as expected, the deep median frontal meningioma was

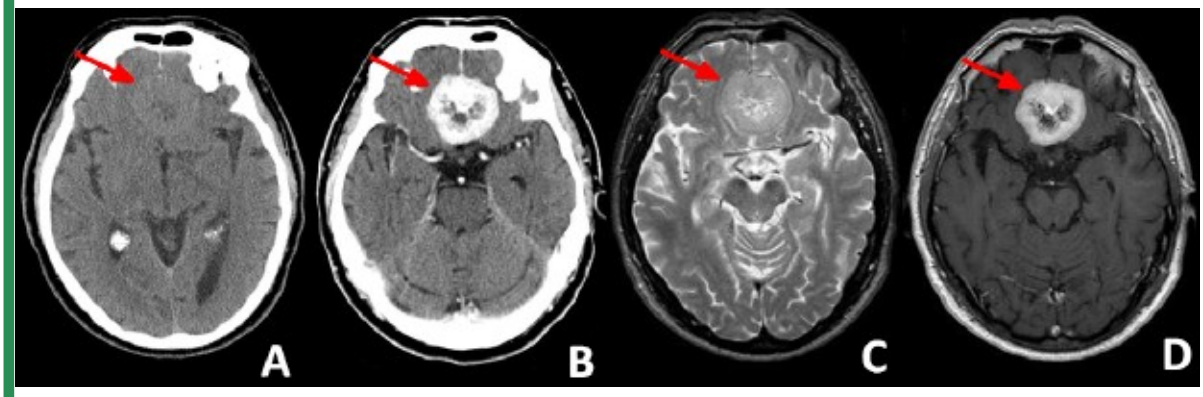

Figure 1 Images of the meningioma on CT scan before (A) and after (B) administration of contrast medium, on T_2-weighted MRI scan (C) and on T_1-weighted MRI scan after administration of contrast medium (D). *Red arrows indicate the tumour.*

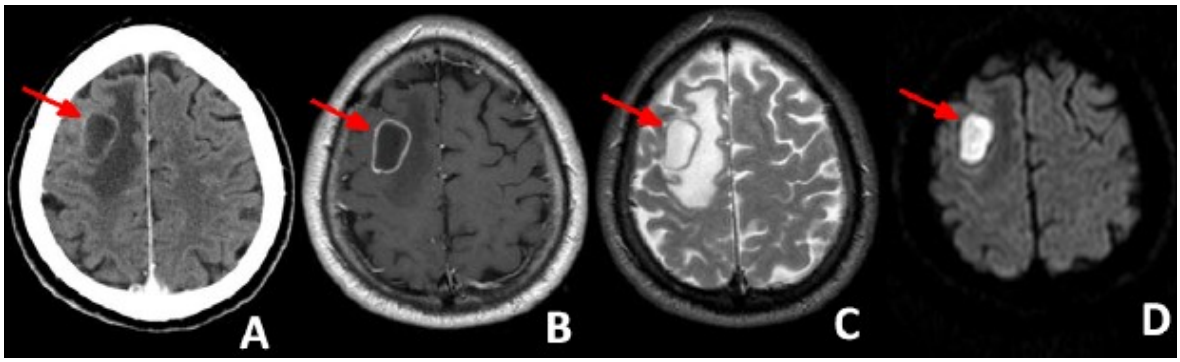

Figure 2: Images of the abscess on CT scan after administration of contrast medium (A), on T_1-weighted MRI scan after administration of contrast medium (B), on T_2-weighted MRI scan (C) and on Diffusion weighted MRI scan (D). *Red arrows indicate the abscess.*

unchanged. Two months later a cerebral MRI scan showed a minimal area of residual right frontoparietal gliosis (Fig. 3), and surgical excision of the meningioma was then planned upon conclusion of the antibiotic therapy.

Discussion

Brain abscesses are a rare but serious and life-threatening disease. They are caused by contiguous spread (i.e. from sinusitis, ear infections and mastoiditis), haematogenous spread (from pneumonias, dental procedures, endocarditis, arteriovenous shunts or in case of congenital cyanotic heart disease), or local diffusion after neurosurgical procedures or head traumas. Our patient presented multiple risk factors in his clinical history: chronic sinusitis, recent dental extraction, abuse of corticosteroids, and a history of diabetes. Certainly, as immunosuppression is an important risk factor, abscesses have a higher incidence in patients who have undergone transplants, are in the late stage of AIDS, or are undergoing cancer chemotherapy; moreover, immunocompromised patients have a 2.8-fold risk of poor outcome [1, 2, 3, 6, 9, 11, 12].

Although infrequent, an intracranial neoplasia can be superimposed by an intratumoral or peritumoral abscess. According to Kalita *et al.*, there are only 27 cases described in English from 1950 to 2008 [13]: they are mostly adenomas or craniopharyngiomas grown in the sellar region, but also lobar meningiomas or gliomas. Only three patients were diagnosed prior to surgery: in those cases, sphenoid sinusitis preceded the abscesses; nevertheless, the majority of intratumoral or peritumoral abscesses have been accidental findings.

The case herein described is peculiar since the meningioma was located in a cerebral area distant from the abscess: we have therefore wondered, what was the role of the former in the formation of the latter; even if the two lesions are far apart from each other, can we say that they are totally independent? Noteworthily, in the rare cases of superimposition of gliomas, ependymomas and meningiomas, authors have argued that an haematogenous spread [14] even from a distant infection, could have been facilitated by tumoral necrosis, intratumoral bleeding, or, especially in meningiomas, gaps in the blood brain barrier [15]. This process might also be sustained by a temporary reduction of the neuro-immune modulation, since in

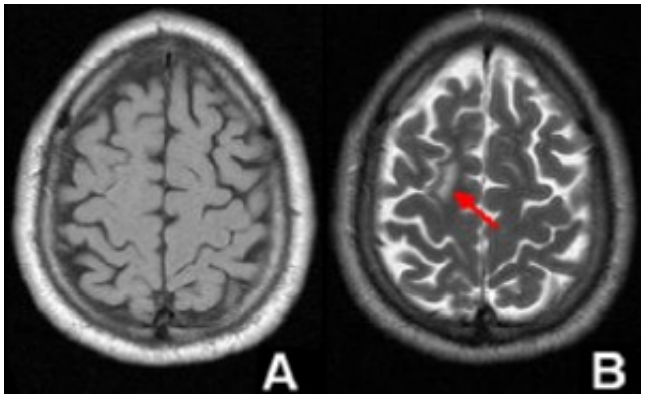

Figure 3: T_1-weighted images (A) and T_2-weighted images (B) MRI scans two months after the neuronavigation-guided surgical aspiration of abscess. *Red arrow indicates the gliosis.*

some cases intratumoral abscesses have evolved during intensive oncological therapy with corticosteroids and/or chemotherapy and/or radiotherapy [13, 16].

There are mainly three reasons to support the theory that the tumour did not affect in any way the growth of the abscess: first, intracranial tumours have thus far never been identified as risk factors in this respect; second, whenever a neoplasia has been superimposed by an abscess, the latter has grown in the intratumoral or peritumoral area and not far from the tumour itself; third, the meningioma probably was not superimposed by the abscess until the dental procedure had taken place. On the other hand, we cannot completely exclude that the meningioma might have played a role in the formation of the abscess for the following reasons: first, the closeness of the tumour to the sphenoidal sinuses might justify a spread of germs by contiguity in a patient with chronic sinusitis [13]; second, while previous surgical procedures had not caused any complication, after the last dental extraction (and despite routine antibiotic prophylaxis) the growth of the meningioma with its probable alteration of the blood-brain barrier apparently facilitated the haematogenous spread of germs [15]; and third, the whole process could have been facilitated by the concomitant underlying reduced immune response due to the chronic uptake of corticosteroids [17].

In light of the above, shouldn't a certain amount of co-responsibility of the tumour in the formation of the abscess be taken into account? Of course a final conclusion cannot be drawn, yet the fact that normal brain defences may fail under the incidental concomitant influence of comorbidities, triggering events, and a predisposing neuropathological background, should not be underestimated.

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