Case Report

Brain abscess as the initial presentation of a macroprolactinoma: Case report

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A R T I C L E   I N F O

Article history:
Received 15 August 2014
Accepted 12 October 2014
Available online 6 December 2014

Keywords:
Brain abscess
Meningitis
Pituitary adenoma
Prolactinoma
Rectus sinus thrombosis
Transphenoidal surgery

A B S T R A C T

Macroprolactinomas may behave invasively and infiltrate the skull base, causing a subsequent thinning that can also lead to a bone defect and a direct route of entry for pathogens. We describe the case of a 34-year-old male admitted to hospital with fever (38 °C), headache, stiffness in the neck, diplopia and neurological impairment. Brain magnetic resonance imaging showed two bilateral abscesses in the fronto-parietal areas with intracranial venous sinus thrombosis and a pituitary adenoma that extended from the suprasellar region, eroding the sellar floor into the sphenoid sinus. Laboratory hormone measurements showed increased levels of prolactin and low levels of FSH, LH and testosterone. The patient received antibiotic treatment and surgery was performed. The patient developed central deafness as a neurological deficit. It is advisable to include pituitary adenoma in the differential diagnosis of meningitis even though its onset as intracranial abscess and rectus sinus thrombosis is extremely rare.

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Absceso cerebral como presentación inicial de un macroprolactinoma: caso clínico

Resumen

Los macroprolactinomas pueden ser invasivos e infiltrar la base del cráneo causando el posterior adelgazamiento que puede conducir a un defecto del hueso y una vía de entrada para patógenos. Describimos un varón de 34 años que ingresó en el hospital con cefalea, rigidez de nuca, diplopia y deterioro neurológico. Las imágenes de resonancia magnética nuclear mostraron dos abscesos bilaterales frontoparietales con trombosis venosa del seno y un
1. **Introduction**

Prolactinomas are usually asymptomatic but sometimes they can give symptoms such as galactorrhea, hypogonadism among others. As the tumor grows larger, it can lead to visual field defects and hypopituitarism.

Some macroprolactinomas, on the other hand, have an aggressive and invasive behavior which can lead to a disruption of the skull base.\(^1\) Anyways a clinical debut with infection of cerebrospinal fluid (CSF) is an infrequent presentation.\(^2,3\)

Meningitis is a serious infectious disease associated with high morbidity and mortality rates. This disease has been described in association with lymphocytic hypophysitis,\(^7\) pituitary abscess\(^2\) and adenomas.\(^4\) In this report, we describe a case of invasive macroprolactinoma that was diagnosed due to symptoms caused by meningitis complicated with brain abscesses and intracranial sinus thrombosis.

2. **Case presentation**

A 34-year-old male patient was admitted to his local hospital with a history of fever (≥ 38°C), occipital headache and diplopia in the previous two days, associated in the last hours with an episode of aphasia and delirium. Neurological examination revealed marked stiffness in the neck and agitation. Shortly afterwards the patient became drowsy and finally he deteriorated to stupor and coma, so orotracheal intubation was performed. Laboratory evaluation, CSF and blood cultures are showed in **Table 1**. Brain computed tomography (CT) scan was performed and revealed signs of osteolysis and enlargement of the sellar region. Also diffuse brain edema and contrast enhanced masses in the sphenoid sinus and into both brain hemispheres were found. It was consider to be an inflammatory-infectious disease. Despite antibiotic treatment and right sphenoidotomy with aspiration of the purulent material, the patient condition did not improve. Magnetic resonance (MR) images demonstrated the existence of a tumoral sellar lesion with suprasellar extension and the two bilateral known brain abscesses with rectus sinus and Galen vein thrombosis (Fig. 1). Serum hormone levels are shown in **Table 1**. Prolactin serum level was >2000 ng/mL (normal range: 2–17 ng/mL). Neurosurgeons carried out an endoscopic endonasal surgery performing a subtotal resection of the pituitary tumor and the sealing of the sellar floor with a nasoseptal vascularized flap. Histological evaluation showed a prolactin-producing pituitary adenoma and inflammatory tissue, without purulent material. Ki 67 proliferation rate was 4%. After surgery, the patient received antibiotic treatment for 8 weeks and anticoagulant therapy for three months to avoid ischemia secondary to the cerebral venous thrombosis. Besides this, an oral treatment with Cabergoline (3 mg per week) was added.

MRI was performed every two weeks within the first two months, as a way of monitoring the evolution of the abscess and venous sinus thrombosis. Brain abscess shrank in size to finally disappeared and there was a reexpansion of the pituitary gland. Moreover, the rectus sinus and Galen vein thrombosis got resolved (Fig. 1). The remnant tumor was stabilized in size and prolactin serum levels normalized.

Despite the progressive improvement of level of consciousness, sensory dysphasia and central deafness persisted. Three months later his performance status was 50 and progressively he ameliorated to a performance status of 70 one year after surgery.

**Table 1 – Complementary tests.**

<table>
<thead>
<tr>
<th>Test</th>
<th>Results</th>
</tr>
</thead>
<tbody>
<tr>
<td>Blood test</td>
<td>Hemoglobin: 10.3 g/dL (normal range: 14–18 g/dL)</td>
</tr>
<tr>
<td></td>
<td>Hematocrit: 32% (normal: 42–54%)</td>
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<tr>
<td></td>
<td>Leucocytes 19,800 (normal range: 4500–10,800)</td>
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<tr>
<td></td>
<td>82% neutrophils</td>
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<tr>
<td></td>
<td>2% lymphocytes</td>
</tr>
<tr>
<td></td>
<td>C Reactive protein (CRP): 185 mg/L (normal: Glucose: 148 mg/dL (normal range: 76–110 mg/dL))</td>
</tr>
<tr>
<td></td>
<td>1850 cells/mm(^3)</td>
</tr>
<tr>
<td></td>
<td>83% polymorphonuclear</td>
</tr>
<tr>
<td>Cerebrospinal fluid</td>
<td>Total proteins: 3.65 mg/dL</td>
</tr>
<tr>
<td></td>
<td>Glucose: 1 mg/dL</td>
</tr>
<tr>
<td></td>
<td>Gram’s stain: negative</td>
</tr>
<tr>
<td>Blood culture</td>
<td>Streptococcus pneumoniae</td>
</tr>
<tr>
<td>Hormonal serum levels</td>
<td>Prolactin &gt; 2000 ng/mL (normal range: 2–17 ng/mL)</td>
</tr>
<tr>
<td></td>
<td>FSH 0.1 mU/mL (normal range: 2–10 mU/mL)</td>
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<tr>
<td></td>
<td>LH &lt; 0.1 mU/mL (normal range: 1.5–9.3 mU/mL)</td>
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<tr>
<td></td>
<td>Testosterone 0.36 ng/mL (normal range: 3–10 ng/mL)</td>
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<tr>
<td></td>
<td>Cortisol 21.80 µg/dL (normal range: 8–25 µg/dL)</td>
</tr>
</tbody>
</table>

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Trombosis del seno recto Cirugía transesfenoidal

adenoma hipofisario que se extendía desde la región supraselar, erosionando el suelo de la silla hacia el seno esfenoidal. El análisis hormonal mostró aumento del nivel de prolactina y descenso de los niveles de FSH, LH y testosterona. El paciente recibió tratamiento antibiótico y fue intervenido quirúrgicamente. El paciente desarrolló sordera central como déficit neurológico. Es recomendable incluir el adenoma hipofisario en el diagnóstico diferencial de la meningitis aunque su debut como abscesos intracraneales y trombosis del seno recto es extraordinario.

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3. Discussion

Meningitis has been described in literature after transsphenoidal surgery or due to pituitary invasive adenomas, being the Streptococcus pneumoniae the most common microorganism implicated. However it is infrequent to be associated with an indolent pituitary adenoma as first symptom at presentation.

Moreover CSF rhinorrhea may occur as a complication of surgery, radiotherapy or even dopamine-agonist therapy and its presence is going to facilitate the diagnosis of CSF leak, but it is important to bear in mind that its absence, does not exclude a skull base defect.

In this report, we describe an extraordinarily rare case of bacterial meningitis associated with brain abscesses and rectus sinus thrombosis as the first diagnosis signs of a pituitary adenoma. The absence of previous headache, visual field defects, rhinorrhea or other symptomatology related to pituitary dysfunction led to a delay in diagnosis and also to a development of sequelae such as central deafness and dysphasia. To our knowledge this case report is the first one ever published with such characteristics.

On the other hand, the first-line treatment of prolactinomas is medical therapy with dopamine agonist, and surgery is reserved as a second-line therapy. However, surgery was chosen in this case, as first-line treatment, for three main reasons: Firstly, to start as soon as possible anticoagulant treatment at therapeutic doses to avoid progression of sinus thrombosis. On the second hand, to repair the skull base defect in order to prevent the infectious progression toward the central nervous system (CNS) and finally to reduce tumoral mass. An oral treatment with Cabergoline was added posteriorly to control prolactin levels.

We want to highlight the importance of considering the diagnosis of pituitary adenomas in patients presenting with clinical features suggestive of meningitis and where there is evidence of an intrasellar lesion on cerebral imaging. It is crucial to early recognize the existence of a skull base defect so measures can be taken to prevent infection from spreading into the CNS and furthermore to treat the patient disease. Endocrine management could be needed, but surgical treatment is essential to avoid new episodes of meningitis and facilitate the early onset of other therapies such as anticoagulants.
4. Conclusion

Aggressive pituitary adenomas may facilitate the entry of pathogens into the CNS by causing bone erosion. This involvement should be suspected in patients with pituitary adenomas and meningitis, although CSF leak signs are not present in the clinical course. Although the onset as sphenoidal and brain abscesses with rectus sinus thrombosis results extraordinary, it is necessary to keep in mind the suspicion of pituitary adenoma in the differential diagnosis of meningitis. Early diagnosis and surgical treatment may improve prognosis in these patients.

Conflict of interest

The authors declare no conflict of interest.

REFERENCES