

Case Report

Spontaneous rupture of a secondary pituitary abscess causing acute meningoencephalitis: Case report and literature review

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Abstract

Background: Pituitary abscess (PA) is an uncommon finding that is rarely diagnosed preoperatively. If not properly treated it is associated with high morbidity and mortality rates. Nowadays standard diagnostic procedures allow early detection and successful treatment of this lesion in a high number of cases and mortality has been significantly reduced in recent years. PA arising de novo in a healthy gland are defined as primary, whereas those complicating a pre-existing disease of the hypophysis are called secondary abscesses.

Case Description: We present a case of a secondary PA mimicking a large pituitary adenoma extending in the nasal cavity, which was wrongly diagnosed as such. The abscess showed an unexpected evolution in 48 h from presentation due to a sudden, extensive intracranial leakage of pus.

Conclusions: To our knowledge, it is rare to find PA showing a rapid evolution like this, and in the literature only one previous case of a PA not reaching medical or surgical therapy was reported. In that case, hypothalamus involvement was identified as the cause of death. This should be the first case reported of a spontaneous PA rupture causing acute meningoencephalitis. Along with a short review of the literature on the major features of PA, we also tried to identify some features which could be supportive of a diagnosis of secondary PA.

Key Words: Adenoma, brain abscess, meningoencephalitis, pituitary abscess, pituitary neoplasm

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INTRODUCTION

Pituitary abscess (PA) represents 0.2–1.1% of all surgically treated pituitary lesions.^[1] Since its first description in 1848,^[3] almost 250 cases have been described,^[2–5] mostly as single case reports, but some series have been published.^[3,6,7]

PA can grow on a normal gland (70%) or complicate diseases of the hypophysis (pituitary adenomas, Rathke's cleft cysts, craniopharyngiomas).^[5,14,15] The latter are called secondary PA, in contrast primary abscesses, i.e., those arising in a healthy pituitary gland.^[1,14,15]

In many cases no microorganism can be isolated from cultures.^[1,9,10,14] Mortality in untreated PA is reported at 30% and need for long-term hormonal replacements at approximately 70%.^[1,8,15,17]

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PA has unspecific clinical and radiological features^[4,11,12,16,19] and 70–80% of cases don't show symptoms of infection.^[4,8] Preoperative diagnosis of PA is therefore challenging.^[1,4,8,15–17] Nevertheless, standard diagnostic procedures [computed tomography (CT) and magnetic resonance imaging (MRI)] usually make early identification and administration of successful therapy possible, even in cases of a wrong preoperative diagnosis. Mortality has been reduced to 4.5%.^[1]

Here, we present a case of unexpected evolution with fatal outcome of a PA mimicking a partially cystic pituitary adenoma. The reason for this evolution was identified during surgery, when we were able to visualize a large subdural collection of pus.

According to a recent systematic review of the literature,^[1] only few cases of PA showed a fatal outcome, and among them only in one case no therapy could be administered due to the rapid progression of the disease.

In none of them a spontaneous abscess rupture with a large subdural pus diffusion causing a meningoencephalitis was identified as the cause of death.

CASE DESCRIPTION

A 47-year-old woman with a 2-day history of headache and blurred vision was referred to our department from a peripheral hospital. A CT scan of the skull had showed a large mass arising from the sella, eroding the bony structures and invading the nasal cavity.

On admission she presented Glasgow coma scale (GCS) 15, afebrile. Neurological examination showed no alterations. Visual field was intact. Body mass index (BMI) was 28.2, medical history included mild psoriasis, gastroesophageal reflux disease (GERD), cholelithiasis, no relevant infections except in her infancy, and no previous surgeries. She did not take any routine medications. She was the mother of two children and had regular menses. Blood pressure and heart rate were within normal limits. Blood test showed microcromic microcytic anemia, reticulocytosis [hemoglobin (Hb): 8.6g/dL; hematocrit (HCT): 27.8%; mean cell volume (MCV): 64.5; mean corpuscular hemoglobin (MCH): 20 pg; mean corpuscular hemoglobin concentration (MCHC): 30.9 g/dL; red blood cell distribution width (RDW): 20.2 %]. Leucocytes count and inflammatory indexes were within normal limits. Hormone blood concentrations were normal except for a slight decrease in thyroid-stimulating hormone (TSH) and follicle-stimulating hormone (FSH) (0.238 μ U/mL and 0.6 mU/mL); β -17-estradiol was 35.8 pg/mL. Urine osmolality was normal.

An enhanced MRI [Figures 1a-c] confirmed a solid-cystic lesion arising from the sella turcica, abutting both cavernous sinuses and wrapping both carotid siphons.

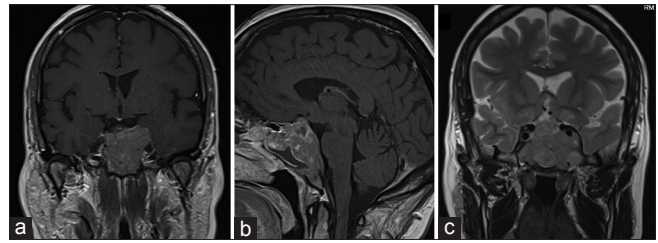


Figure 1: (a) Pre-operative magnetic resonance imaging (MRI) with gadolinium. T1-weighted coronal section showing a large enhancing sellar lesion with suprasellar extension, impinging the chiasm and abutting both cavernous sinuses. (b) T1-weighted sagittal section with gadolinium showing a mixed solid-cystic component of the tumor. The chiasm is dislocated upwards (c) T2-weighted coronal scan showing a mixed solid-cystic components of the lesion

According to the clinical and radiological data, a large pituitary adenoma with a mixed solid/cystic consistence was diagnosed. The patient was admitted and scheduled for surgery the same week. Her headache was treated with pain medications. She complained of no vision problems.

Forty eight hours after onset of symptoms, her headache suddenly worsened and she rapidly developed gaze palsy and nuchal rigidity. GCS fell from 15 to 6 (E:1, V:2; M:3). After an emergency CT showed no parenchymal infarction, extra-axial bleeding, fluid collection, or ischemia [Figure 2], an urgent craniotomy was performed. A right frontotemporal approach was done.

After opening the dura mater, the brain was swelling, with a considerable amount of dense pus covering the brain surface [Figure 3a and b]. More purulent material was found within the tumor located in the sella and drained [Figure 4].

The patient was subsequently taken to the intensive care unit (ICU), but she did not recover and died a few days later.

The pathological analysis of the intraoperative material confirmed a nonfunctioning pituitary adenoma with chronic inflammation and necrosis [Figure 5a-c]. Cultures of the intraoperative material were negative.

DISCUSSION

In this case a secondary PA was misdiagnosed as a cystic pituitary adenoma. The patient had no history of infections or metabolic diseases, or any signs or symptoms of infections. Her clinical condition worsened rapidly and unexpectedly in less than 2 days. During surgery a large amount of pus was found intradurally. An abscess was found within the tumor, which was confirmed to be an adenoma. Death due to PA is considered rare nowadays, with a mortality of 4.5%.^[1]

We explained this finding as the consequence of a sudden spontaneous PA rupture. Pus leakage from a PA has been hypothesized as a possible cause of recurrent

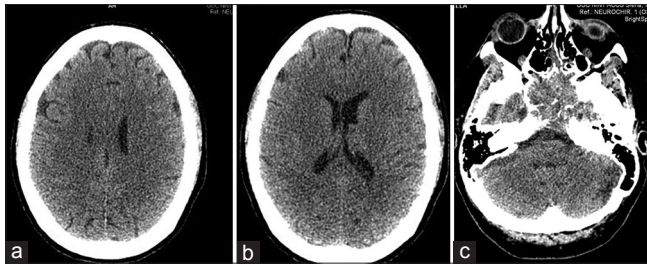


Figure 2: (a and b) CT scan performed after sudden clinical deterioration showed no clear intra- or extra-axial bleeding or fluid collection. No ischemia could be detected. (c) The lesion of the sella had eroded the skull base

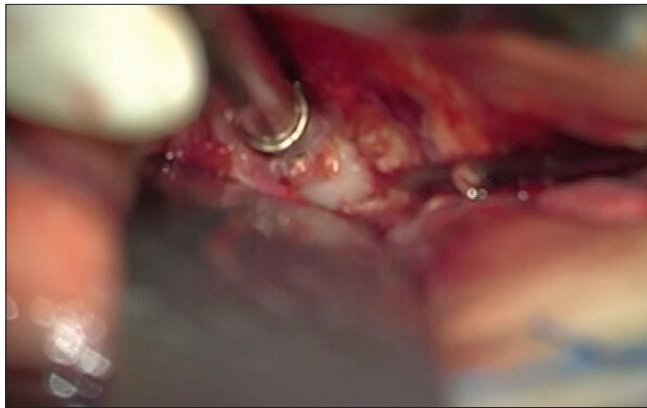


Figure 4: Intra-operative picture: Inside the sellar lesion, more pus was found and drained

meningitis^[18] but, to our knowledge, this was never documented.

Since its first descriptions (Heslop, 1848),^[3] PA has represented a diagnostic challenge, due to its unspecific clinical features,^[13,17] radiologic signs,^[6] and to its rarity.^[1,10] PAs have largely been reported as single cases, but recently some series have been published.^[12,17,19]

Predisposing factors are infections (sinusitis, cavernous sinus thrombophlebitis, tooth infections, osteomyelitis, endocarditis), cerebrospinal fluid (CSF) leakage, previous surgeries or local radiotherapies, and immunocompromise.^[1,11,14,17,18]

A proposed classification based on the etiology identified four groups:^[3] 1. Secondary PAs; 2. Direct extension or hematogenous diffusion to a normal pituitary gland from an infective focus; 3. Complications from previous surgeries; 4. No clear etiology.

In secondary cases without previous infections the origin can be caused by direct tumor invasion of extracranial structures or by tumor necrosis with impairment of local immunity.^[1,7]

No microorganism can be isolated in approximately 50% of cases (so called primary abscess).^[4,6,18] In the rest of cases, commonly found agents are *Staphylococcus spp.*,

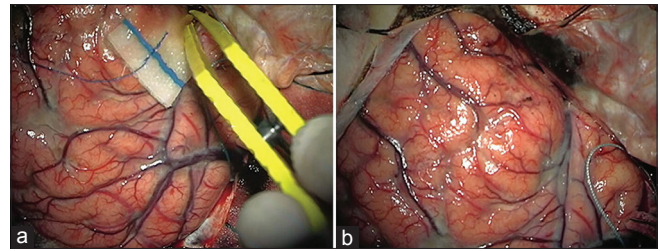


Figure 3: (a and b) Intra-operative pictures, right fronto-temporal craniotomy: The brain was swelling, with evident pus in the sulci and on the brain surface

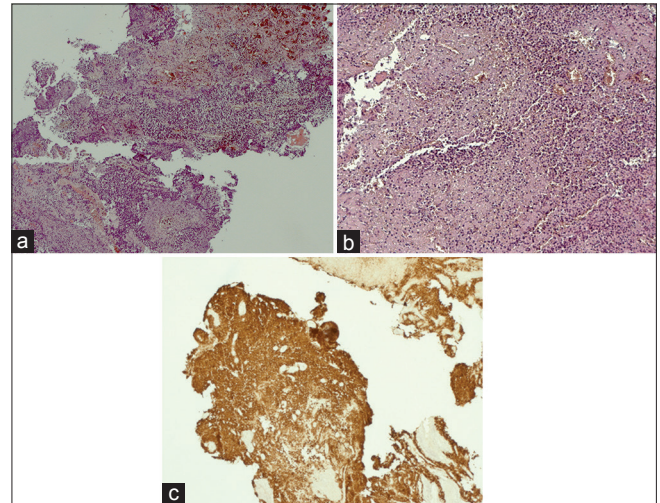


Figure 5: (a) Haematoxylin-eosin stained section of the adenoma at 4x magnification showing neoplastic cells admixed with a robust inflammatory infiltrate with neutrophils. (b) The same section at 10x magnification. (c) Synaptophysin positive immunostained section(4x)

Streptococcus spp., *Mycobacterium spp.*, *Neisseria spp.*, *Micrococcus*, *Citrobacter*, *E. Coli*, *Brucella*, *Salmonella*, *Corynebacterium*,^[17] *Aspergillus*, or *Candida*.^[3]

Clinically, they manifest with headache, which is the most common symptom and can be the only presenting, visual disturbances, hormonal disfunctions, and cranial nerve disabilities.^[1,14,17,19] In the literature, fever was present in 18–42% of cases.^[1] Serum leukocytosis with neutrophilia can be the only laboratory alteration.^[1]

On CT, PA appears as a cystic lesions with peripheral ring enhancement.^[17,18] On MRI, hypo- or isointense on T1 and iso-/hyperintense on T2 with rim enhancement are common findings, and diffusion weighted imaging (DWI) sequences can be helpful in diagnosing an abscess.^[1,14]

Notwithstanding this, colliquative or cystic adenomas, pituitary apoplexy,^[4,5] central necrosis,^[1] carcinomas, colloid cysts, Rathke's cleft cysts, epidermoid tumors, craniopharyngiomas, and metastasis^[4,8,11] can appear as cystic lesions as well and share some features with a PA.

PAs are often misdiagnosed preoperatively as cystic pituitary adenomas.^[2,17] A correct diagnosis was possible

in a proportion ranging from 24% to 83% of the reported cases.^[1]

Zhang^[19] suggested that the presence of the following features could help in diagnosing a PA: 1. Cystic sellar lesion; 2. History of sinusitis, CSF leakage, recurrent meningitis, previous surgeries, or local radiotherapy; 3. Rapid onset of diabetes insipidus despite relatively small mass size.

Moreover, the peripheral enhancement observed in normal tissue surrounding an adenoma is usually thicker than dose related to a PA.^[4,5] Thallium-201 single-photon emission computed tomography (SPECT) and MRI spectroscopy can be useful in suspected cases.^[6,17]

The definitive diagnosis is made on direct vision and on histology showing acute or chronic inflammation with neutrophils or monocytes and lymphocytes.^[1,17]

If an abscess is diagnosed or suspected, antibiotic therapy should be started and surgical drainage planned as soon as possible. Prompt therapy can reduce mortality to less than 10%.^[1,10,13,16,17] The transnasal approach is the most used, because it is thought to expose to less risk of CSF contamination.^[1,3,5,11,15] Craniotomy is not contraindicated and is preferred in cases with relevant suprasellar extension and in redo surgeries, though higher recurrence rates are associated with craniotomies.^[1]

There is no consensus on the duration of the antibiotic therapy: Yet most cases were successful after a 4–6-week course of treatment and some patients were treated with antibiotics alone.^[1]

A life-long hormonal replacement can be necessary,^[11,15,17] since only 32.3% of cases apparently reach a complete recovery of hormonal function.^[1] There is a higher risk of permanent pituitary deficits for those patient with a long history of symptoms.^[10] Recurrence is rare, but possible in 10% of cases.^[1,6,12]

PAs can be fatal and, according to the literature, death can be caused by pituitary apoplexy, hypothalamic involvement, complications of sepsis, or unknown causes.^[1]

Our case represented an acute purulent meningoencephalitis caused by the spontaneous rupture of a secondary PA. To our knowledge, this occurrence has never been observed. This patient had showed no fever or signs of infections and her medical history showed no relevant pathologies. The sudden worsening of this case is peculiar and only one previous case with similar evolution was described.^[1]

CONCLUSIONS

We describe a possible complication of PA. As far as we know, this evolution has never been described. PA can undergo spontaneous rupture and cause acute meningoencephalitis.

This case also raises some important questions on the opportunity to consider pituitary abscess as a possible diagnosis in mixed cystic/solid sellar lesions in order to establish proper therapies, avoiding fatal outcomes. A large sellar lesion extending over the anatomical boundaries of the skull base and showing a cystic component might have undergone a superinfection and harbor a secondary PA.

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Conflicts of interest

There are no conflicts of interest.

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