

Recurrent Primary Pituitary Abscess

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ABSTRACT

Pituitary abscesses are rare and potentially life-threatening. Recurrent pituitary abscesses are rarer still. We present a case of primary pituitary abscess with recurrence 11 years after drainage of the initial abscess. We could trace only five previously reported cases of recurrent pituitary abscesses and, in these, the duration for recurrence varied from 2 days to 4 years.

Keywords: Primary pituitary abscess, Hypophysitis, Recurrent pituitary abscess.

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INTRODUCTION

Hypophysitis (inflammatory involvement of hypophysis) is a rare phenomenon and accounts for less than 0.5% of all the diseases of anterior pituitary.¹ Pituitary abscesses *per se* form a very rare subgroup of this rare affliction. In Cushing's classical treatise on pituitary lesions² no discussion of pituitary abscess is found. It was first described by Simmonds in 1914³ and since then about 210 cases of pituitary abscesses have been reported.⁴ The rarity can be gauged from the fact that Scanarini et al⁵ recorded only two cases in their series of 500 expansive pituitary lesions over a 27-year period. For some reason, recurrence of the abscess after drainage seems very rare. There are only five reported cases of recurrent primary pituitary abscesses.^{7,9} Here, we report a case of recurrence of pituitary abscess 11 years after initial drainage, who had developed meningitis and septicemia postoperatively, leading to fatal outcome.

CASE REPORT

A 53-year-old male who was on regular follow-up in our endocrinology department was reported with headache and diminishing vision of both eyes. The patient was initially presented in 1999 with headache and diminishing vision. Magnetic resonance imaging (MRI) was suggestive of a sellar lesion which was thought to be a pituitary adenoma and the patient was taken up for transcranial excision of the lesion. A subfrontal approach was used and intraoperatively an encapsulated lesion filled with pus was encountered which was completely drained. No organism could be grown from the pus. Postoperatively his vision improved but he

developed panhypopituitarism for which he was on replacement therapy and was on regular follow-up. In July 2010 (11 years after the initial surgery), he again started having headache and noticed diminishing vision. There was no history of fever, cough, night sweats, vomiting, seizure or other neurological deficit. He was not immunocompromised. He had no history of tuberculosis and his chest X-ray was normal. Contrast MRI showed a sellar lesion suggestive of pituitary macroadenoma (Fig. 1). He had no evidence of diabetes mellitus, hypertension and was clinically euthyroid with normal FT4 levels. He was taken up for transsphenoidal excision of the mass. Intraoperatively, an encapsulated pus-filled lesion was encountered (Fig. 2) and was drained (Fig. 3). The capsule was excised completely and sent for histopathological examination. The pus was negative for bacteria (Gram's stain), acid-fast bacilli (Ziehl Nelson stain) and fungus (periodic acid-Schiff stain) and pus culture was sterile.

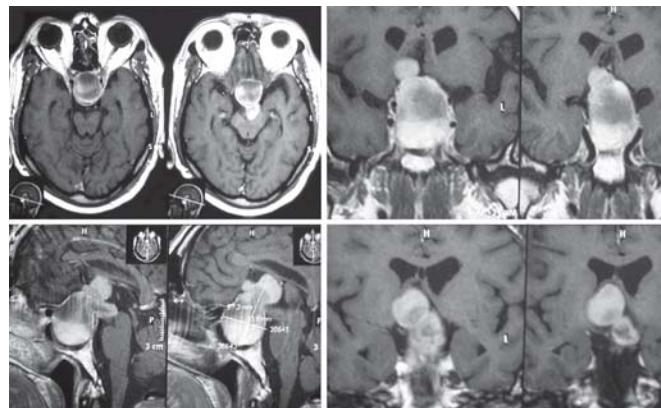


Fig. 1: Postcontrast MRI

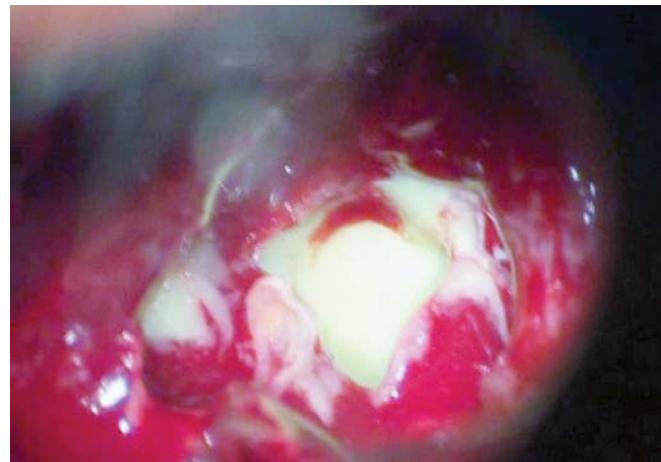


Fig. 2: Intraoperative photograph showing copious pus in the pituitary abscess



Fig. 3: Sella at conclusion of surgery

Histopathological examination of extracted material revealed only necrotic material without evidence of granuloma or caseation. Postoperatively, he was given hydrocortisone infusion (10 mg/h for first day and 7.5 mg/h on second day) and the patient did well for 2 days. Thereafter, he developed features of meningitis. A diagnostic cerebrospinal fluid was indicative of meningitis but there was no growth on culture. He was treated with empirical antibiotics (ceftazidime and vancomycin) and supportive management. Repeat radiology did not reveal any residual abscess. However, he continued to deteriorate and finally succumbed to meningitis and septicemia.

DISCUSSION

Pituitary abscesses are very rare and in recent times, Hanel et al⁶ report an incidence of 0.2% (single case of pituitary abscess in their series of 503 sellar lesions). The largest series is of 24 cases reported from University of California, San Francisco by Vates et al.⁷ For some reason, recurrence of the abscess after drainage seems very rare. After the first drainage, the patients may have recurrent aseptic meningitis and even a frontal lobe abscess is reported but recurrent pituitary abscess seems very rare.⁷ We have been able to find only five more reported cases of recurrent primary pituitary abscesses.⁷⁻⁹ The time duration of presenting with a recurrent abscess in these patients varied from 2 days to 4 years after drainage of the first abscess (Table 1). We have not found a case like ours where the abscess recurred after such a long period of 11 years. Though tubercular abscess of pituitary has been reported from India,¹⁰ our case had no history or clinical evidence of tuberculosis. Moreover, a symptom-free interval of 11 years without antitubercular treatment strongly argues against this possibility.

In about 50% of primary abscesses, the source of the infection remains unknown and often called ‘primitive’ pituitary abscesses.¹¹ Present case was immunocompetent and had no evidence of systemic source of infection, hence, was a case of primitive pituitary abscess. About one-third of pituitary abscesses are secondary, i.e. they arise in

Table 1: Patients with recurrent pituitary abscess as reported in the literature

Case no.	Author	Age and Sex	Time after primary surgery for recurrence	Presenting complaints	Endocrine status	Imaging	Treatment	Bacteriological examination	Outcome
1	Case 3 of Vates et al ⁷	69/M	4 years	Headache, hemianopia	Panhypo	Skull film, arteriography	TS drainage	Gram-positive cocci	Fatal
2	Case 5 of Vates et al ⁷	12/M	2nd post-operative day	Headache, hemianopia	Panhypo	Skull film, arteriography	TS drainage	Negative	Recovered
3	Case 12 of Vates et al ⁷	69/M	15 months	Headache, quadrantanopia	Panhypo	MRI: Intrasellar mass with heterogeneous signal and rim enhancement	TS drainage	Staph aureus	Recovered
4	Case 4 of Jain et al ⁸	24/M	1.5 years	Headache, visual deterioration	Not mentioned	CT: Ring-enhancing lesion in sellar and suprasellar region	TS drainage	Aspergillus	Recovered
5	Case 4 of Dutta et al ⁹	12/F	1 year	Fever, headache, visual deterioration	No deficiency	MRI: Sellar mass hypointense on T1 and hyperintense on T2WI with variable enhancement	TS drainage	Acinetobacter and Staphylococcal species	Recovered
6	Present case	53/M	11 years	Headache, visual deterioration	Panhypo	MRI: Sellar and suprasellar lesion with variable signal and rim enhancement	TS drainage	Negative	Fatal

preexisting lesions in the sellar region.¹² Such 'abscessification' of sellar lesions occurs most commonly with a pituitary adenoma (30% of cases).¹¹ The most common cause of 'abscessification' of a pituitary adenoma is infarction of the adenoma.¹³ Histopathological examination of drained pus did not reveal any evidence of tumor cells, hence, this possibility was unlikely.

Pathologically, the pus evacuated at surgery is sterile in almost half of the cases.^{7,14} However, when the culture does yield an organism, the common pathogens are *Staphylococcus sp*, *Streptococcus sp* and *Neisseria species*.⁶ In our case, microscopic examination revealed only necrotic material without evidence of acid-fast bacilli, Gram's stain was negative and bacteriological examination was sterile. There was no evidence of granuloma or caseation.

Clinically, pituitary abscess may be indistinguishable from a pituitary adenoma and this differentiation (clinically and radiologically) prior to surgery remains the biggest significant challenge in the management of these abscesses. These lesions present either with symptoms related to mass effect, hypopituitarism or infection. Headache and visual symptoms are the commonest complaint.^{7,14} Diabetes insipidus is seen in almost half of the patients with pituitary abscesses.^{5,9,14} Our patient had no pre- or postoperative evidence of polyuria; however, plasma and urine osmolality was not measured. Regarding symptomatology pertaining to infective focus, patients may have fever,⁷ recurrent meningoencephalitis or recurrent aseptic, sterile meningitis.¹⁵ Rarely, systemic features of toxemia may also be present.⁹

Radiologically, it remains a challenge to differentiate a pituitary abscess from a liquefied adenoma or pituitary apoplexy. The preoperative diagnosis continues to be difficult in spite of computed tomography (CT) and MRI findings are more or less same in these three situations.^{6,11,12} Shimamura et al¹⁶ report that pituitary abscess can be differentiated from pituitary adenoma as a lesion with a homogenous high uptake on Thallium-201 single photon emission computed tomography (SPECT) on both early and delayed images and no enhancement of the central portion on MRI. An Indium-111 labeled autologous white blood cell scan has also been used in the diagnosis of a pituitary abscess.¹⁷ Quantitative ¹H-MR spectroscopy shows choline peak in pituitary adenoma,¹⁸ whereas patients with pituitary abscess had lactate, amino acids (including valine, alanine and leucine) and acetate peaks.¹⁹

The management of choice remains surgical drainage (by transsphenoidal route) followed by appropriate antibiotics.^{4,6,7,9,11} A transcranial approach may result in intracranial dissemination of infection and should be avoided.^{6,7} Whatever the procedure followed,

these remain formidable challenges for management with high mortality. Dominique et al reported mortality of 28%, which increased to 45% if meningitis was coexistent.¹⁴ Heneger et al reported 51% mortality and in cases not treated surgically mortality was 100%.²⁰ However, the largest series of Vates et al 2001 reports a more modest mortality figure of 8.3%.⁷ Our case developed meningitis with systemic signs of sepsis with septic shock postoperatively, leading to fatal outcome.

CONCLUSION

We have reported case of a recurrent primary pituitary abscess which seems to be only the sixth reported case of recurrent pituitary abscess. Transsphenoidal drainage of the abscess with appropriate antibiotics remains the accepted cornerstone of management. Recurrence after drainage of a pituitary abscess seems very rare.

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