CASE REPORT

Abscess inside craniopharyngioma: diagnostic and management implications

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SUMMARY

Abscess inside the pituitary fossa is very rare. Such abscess can be primary, occurring in an otherwise healthy pituitary gland, or secondary, developing inside a diseased gland (ie, harbouring craniopharyngioma, Rathke's cleft cyst, etc). Secondary pituitary abscess inside a craniopharyngioma remains an extremely rare occurrence. Our literature search revealed only six such cases reported so far. In this report, we present the seventh case of craniopharyngioma with abscess in a 38-year-old woman. We describe the uniqueness of the clinical presentation of our case, the radiological pointers to the possible diagnosis and the management issues in our patient. A review of literature is also included to provide a comprehensive picture of this rare condition to the readers.

BACKGROUND

Pituitary abscesses are very rare. These can be either primary, developing de novo inside a normal pituitary gland, or secondary that forms within a diseased gland like microadenoma/macroadenoma, craniopharyngioma or Rathke's cyst.¹ The association of abscess with craniopharyngioma is extremely rare, and only six cases have been described previously.^{1–6} In this report, we describe the seventh case of a craniopharyngioma which housed within it a pyogenic abscess. The clinical, radiological and operative findings are discussed. We also discuss the pathogenesis, clinical features, radiological pointers and the management implications involved in these cases. This extremely rare clinical scenario mandates accurate diagnosis and specific management plan to ensure a satisfactory clinical outcome.

CASE PRESENTATION

A 38-year-old woman presented with amenorrhoea for 9 years accompanied by headache and loss of vision in her left eye for 15 days. On examination, she was conscious, oriented with a visual acuity of 6/12 in the right eye and absence of light perception on the left side. Right temporal field cut was revealed on confrontation test. Examination of the fundi revealed findings consistent with primary optic atrophy bilaterally, indicating that the compression on optic nerves had been there for some time.

INVESTIGATIONS

MRI of the brain showed a sellar–suprasellar mass with an open sella. The mass was hyperintense

on T1-weighted images and predominantly darkly hypointense on T2-weighted images. On contrast examination, the mass showed a uniform and thick rim-like peripheral enhancement. The mass was extending superiorly up to the floor of the third ventricle, and no parasellar extension was noted. Axial sections of the MRI showed enhancement of the choroid plexus of the temporal horns; however, no enhancement of the basal leptomeninges was seen. Sphenoid sinus appeared sellar in type without any evidence of mucositis (figure 1A–E).

Endocrinological evaluation revealed low levels of serum thyroid hormone, serum cortisol, follicular stimulating hormone (FSH) and leutinizing hormone (LH) suggesting panhypopituitarism.

TREATMENT

Considering the clinical picture of acute headache, vomiting with vision loss in one eye and MRI features suggesting possible bleed inside a sellar–suprasellar tumour, we made a diagnosis of pituitary macroadenoma with apoplexy. Hence, under hormonal cover (oral thyroxine and injectable hydrocortisone), we decided to go ahead with urgent tumour decompression. During endoscopic endonasal transsphenoidal tumour decompression, we encountered a greyish, moderately vascular, soft and freely flowing mass with small pockets of purulent material inside it. We did not find any evidence of bleed, whatsoever, inside the tumour. Calcium flakes were also seen inside, indicating the likelihood of craniopharyngioma. Once the purulent material was encountered, cottonoid patties were placed circumferentially around the durotomy site, and gentle suction was applied over them to avoid inadvertent spillage onto the sphenoid sinus and nasal cavity. Thereafter, the purulent material was aspirated using a wide-bore needle mounted on a 2 mL syringe directly from the intratumoral abscess under endoscopic vision. This ensured that the collected specimen represented the abscess per se, not the purulent material contaminated with nasal secretions. Thereafter, further bits of tumour were cleared meticulously. We ensured that the suprasellar arachnoid was not breached in any way to avoid dissemination of the purulent material into the cranial subarachnoid space. Postoperative CT of the head showed gross total excision of the tumour (figure 1F).

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Figure 1 (A) MRI axial T1-weighted image (T1WI) shows thick-walled suprasellar mass with central hyperintensity. (B) Mass is predominantly hypointense on T2WI with few T2 hyperintense pockets along the right lateral border of the lesion. Postcontrast T1WI in axial (C), sagittal (D) and coronal (E) planes show uniform and thick rim-like peripheral enhancement of the mass with central non-enhancing areas. (F) Postoperative CT sagittal image shows no residual mass.

OUTCOME AND FOLLOW-UP

The patient had an uneventful postoperative recovery. There was no meningitis or diabetes insipidus during the postoperative period. At the time of discharge, the patient had significant improvement of the vision in her left eye (FC at 3 m). Histopathology of the tumour was suggestive of adamantinomatous variety of craniopharyngioma. The culture and sensitivity of the aspirated purulent material showed exuberant growth of methicillin-sensitive *Staphylococcus aureus* that was sensitive to levofloxacin and clindamycin. The patient was administered intravenous form of these antibiotics for 1 week, and oral antibiotics were continued for another 6 weeks after discharge.

At 3 months' follow-up, the patient was doing well. The vision in her left eye improved further (6/36). She was on hormone replacements for panhypopituitarism.

DISCUSSION

The first case of pituitary abscess was described by Simmonds in 1914.⁷ Primary pituitary abscesses developing de novo inside a normal pituitary gland are more common than their secondary counterparts occurring in the setting of pathological conditions like craniopharyngioma, Rathke's cleft cyst or pituitary adenoma.^{8–10} Till now, 20 cases of secondary abscesses associated with pituitary adenoma,⁹ 13 cases associated with Rathke's cleft cyst¹¹ and 6 cases associated with craniopharyngioma have been described in literature.^{1–6} To the best of our knowledge, this is the seventh case of secondary abscess associated with craniopharyngioma to be reported to date (table 1).

The exact pathophysiology of such abscesses is not clearly known. Various factors like tumour mass effect leading to an impaired blood circulation, central necrosis and local immunological impairment have been incriminated by various authors.¹⁰

In the presence of these predisposing factors, localised sphenoid sinus infection may spread locally or haematogenously and can give rise to secondary pituitary abscess.¹ Craniopharyngioma is also known to provoke reactive inflammatory changes in the surrounding pituitary tissue producing changes akin to abscesses.¹

In our patient, there was no evidence of sphenoid sinusitis either on imaging or during surgery.

As far as the clinical presentation is concerned, all the patients reported earlier presented with headache, loss of vision and pituitary insufficiency. Five out of the six reported patients had endocrine abnormalities. Panhypopituitarism, coupled with impaired local immunological response, has also been incriminated in the abscess formation.¹ History of amenorrhoea in our patient suggested the presence of long-standing hypopituitarism, a fact

Table 1 Reported cases of abscess inside craniopharyngioma

S No	Author and year	Presentation	Culture
1	Riser <i>et al</i> , ⁸ 1956	HA, vision loss	
2	Montrieul <i>et al</i> , ⁶ 1965	HP, vision loss	Negative
3	Obrador <i>et al</i> , ⁷ 1972	HP, vision loss, cranial nerve palsy	Negative
4	Arseni <i>et al</i> , ¹ 1975	HP, vision loss	<i>Escherichia coli</i>
5	Shanley <i>et al</i> , ⁹ 1994	HA, vision loss	<i>Salmonella typhi</i>
6	Qi <i>et al</i> , ¹¹ 2009	HA, vision loss, HP	<i>Staphylococcus aureus</i>
7	Present case, 2017	HA, HP, vision loss	Methicillin-sensitive coagulase-negative <i>S. aureus</i>

HA, headache; HP, hypopituitarism.

that was later documented on investigation. Hence, we agree with Qi *et al*¹ that long-standing hypopituitarism may play a role in the causation of these rare abscesses. The clinical presentation in our patient was unusual of a case of secondary pituitary abscess. Such symptoms usually indicate apoplexy inside an existing adenoma.

Radiologically, our patient showed T1 hyperintensity and T2 hypointensity inside the tumour, features suggesting blood in the acute stage. On contrast administration, however, we could see thick and uniform rim-like enhancement in the periphery of the lesion. Pituitary stalk could not be visualised. Reports on pituitary abscess show conflicting MRI findings.¹² Most reports agree that the abscess appears hypointense on T1 and hyperintense on T2. However, in our case, the contrary was found. If we consider craniopharyngioma, high proteinaceous content may appear hyperintense on T1; however, it will not become hypointense on T2. On the other hand, it is possible that T2 hypointense area could have been secondary to calcification within craniopharyngioma. The thick and uniform capsule-like enhancement found in our patient has been described by some authors as a pointer towards concomitant abscess.¹² Other features described are evidence of pituitary stalk thickening, leptomeningeal enhancement, evidence of sphenoid sinusitis and so on.¹²

Ideal management of these conditions is not conclusively established. This is because clinical and radiological features hardly allow preoperative diagnosis in the majority of these cases. In their report of 24 cases of pituitary abscess, Vates *et al*¹⁰ noted that majority of the patients were diagnosed at the time of surgery. If, however, one can make a diagnosis preoperatively, decompression via the transsphenoidal route appears to be the procedure of choice.¹² It ensures persistent extracranial decompression without violation of the subarachnoid space (if one remains careful enough to only decompress not radically excise the tumour!) and concomitant management of sphenoid sinusitis. Antibiotics covering Gram positive, Gram negative and anaerobes need to be empirically started after the diagnosis of abscess is made. Once the bacteriogram is available, specific antibiotics must be initiated. It has been recommended that antibiotics must be given for at least 4 weeks.¹² Additionally, these patients require lifelong hormone replacements for hypopituitarism.¹²

Although purulent material can be demonstrated in almost all cases of pituitary abscesses, the causative microbial agents can be isolated in only about 30% of them. A review of the previous 6 cases of craniopharyngioma with abscess reveals that cultures were negative in half of them. In our patient, methicillin-sensitive *S aureus* was isolated. Other organisms that have been isolated from abscess accompanying craniopharyngioma are *Escherichia coli* and *Klebsiella*.²⁻⁶ It is important to subject the infected material to all three correct forms of bacterial cultures (aerobic, anaerobic and tubercular) and fungal stains/culture.

The clinical outcome of such abscesses is generally good as long as there is no evidence of sepsis or meningitis.¹⁰ Transsphenoidal decompression with antibiotic therapy suffices in a large majority of these patients. Subarachnoid dissemination of the pus is a poor prognostic event leading to mortality in 8.3% of the cases.¹⁰ Headache and visual symptoms usually improve, as seen

in our patient as well. Hypopituitarism, however, is irreversible in most cases, requiring lifelong hormone replacements.¹²

Learning points

- ▶ Secondary abscess inside craniopharyngioma is an extremely rare but a potentially serious condition.
- ▶ Symptoms rarely help in their preoperative diagnosis, although headache, vision loss and panhypopituitarism are consistent findings in all reported cases.
- ▶ Contrast-enhanced cranial MRI showing uniform and thick peripheral enhancement within a cystic sellar–suprasellar mass, thickened pituitary stalk, leptomeningeal and sphenoid sinus mucosal enhancement must raise an eyebrow of suspicion, especially if the patient has evidence of panhypopituitarism.
- ▶ Such abscesses may clinicoradiologically simulate pituitary apoplexy, as seen in the present case.
- ▶ Transsphenoidal decompression, antibiotic therapy and lifelong hormone replacement constitute the backbone of their management.

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