Primary pituitary abscess in an adolescent boy: a rare occurrence

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SUMMARY

Primary pituitary abscess is a rare clinical condition at a young age. It is characterised by atypical clinical features which makes the diagnosis difficult. Correct diagnosis and therapy are mandatory due to the potentially lethal outcome. We report the case of a 14-year-old healthy boy. The onset was acute with fever, gait imbalance, slurring of speech and amnesia. MRI brain revealed a hyperintense lesion in T2-weighted image (T2WI) in sellar and suprasellar region, which was hypointense in T1WI. After administration of contrast, there was peripheral rim enhancement suggesting pituitary abscess. The diagnosis was confirmed following evacuation of purulent material, during surgery, through a trans-sphenoidal approach. Postoperatively, the boy needed prolonged intensive care support. He was discharged on day 42 after surgery. Follow-up at 3 months revealed complete motor recovery. He required hormone replacement and was intermittently showing features suggestive of frontal lobe syndrome.

BACKGROUND

Pituitary abscess is a challenging diagnosis for the clinician. This is mainly due to the peculiarities of signs and symptoms associated with this clinical condition.¹

Bacterial or fungal infection of the pituitary gland is a rare pathology. Only about 200 cases are reported in the literature so far. The scarce epidemiological data that are available suggest that pituitary abscesses represent not more than 0.2–1% of all pituitary lesions.^{2–3}

In children, pituitary abscess is an exceptionally rare clinical condition. Review of existing medical literature shows about 20 cases of pituitary abscess in children. The majority of these are due to infection of a Rathke's cleft cyst. Rarely, other underlying lesions are demonstrated. Only a few cases were of primary pituitary origin.^{4–5}

Several of these cases are characterised by the lack of classical signs of infection like fever. Many are not associated with laboratory markers of inflammation such as increased erythrocyte sedimentation rate, leucocytosis, increased C reactive protein and fibrinogen levels.^{6–7}

In our case, an adolescent boy presented acutely with varying neurological manifestation. An MRI at the time of admission suggested collection in the pituitary fossa. Thick purulent material was evacuated by a difficult neuro surgical intervention. So the diagnosis of pituitary abscess was confirmed. Because of the absence of any predisposing factors in a previously healthy child, a diagnosis of primary pituitary abscess was made. Following surgical

drainage, the child had a difficult postoperative period. He needed prolonged ventilation and intensive care support. In this case, an early and accurate diagnosis was made. This was followed up with appropriate surgical intervention and supportive care. The boy had complete motor recovery within 3 months. However, during follow-up, some symptoms of frontal lobe syndrome were noticed. This was probably due to the prolonged neurosurgery and difficult postoperative period.

The rare incidence of this condition in a young boy, its early diagnosis and successful management make it a case worth reporting.

INVESTIGATIONS

Initial investigations showed raised total leucocyte count (15 600/mm³), negative blood culture but a positive C reactive protein (4.2, normal range <1.0). Cerebrospinal fluid study was non-contributory.

Preoperative hormonal assay revealed

- ▶ Serum cortisol (8:00)—166 mmol/l (normal range 123–626 mmol/l)
- ▶ Serum prolactin 6.8 ng/ml (normal range 3.7–17.9 ng/ml)
- ▶ Free T₃ 4.19 pmol/l (normal 4.26–8.10 pmol/l)
- ▶ Free T₄ 16.2 pmol/l (N 10–28.2 pmol/l)
- ▶ Thyroid-stimulating hormone (TSH) 2.48 µIU/ml (N 0.5–4.5 µIU/ml)
- ▶ Serum insulin-like growth factor (IGF) 1 87 µg/l (N 101–267 µg/l)

Preoperative MRI brain showed a hyper intense lesion in T2-weighted image in sella and suprasellar region which is hypointense in T1WI. After contrast, there was peripheral rim enhancement suggestive of pituitary abscess measuring 1.2×1.8×1.7 cm. There were also associated inflammatory changes in sphenoidal and ethmoidal sinuses.

During surgery, purulent material was drained which grew Gram-positive cocci in cultures which was identified as *staphylococcus aureus*.

Postoperative pituitary functional assessment showed serum-free T₃ of 2.67, free T₄ of 21.8, TSH 0.044 and Serum Cortisol level of 294. Serum IGF1 value was 100 immediately following surgery.

MRI, a week after the surgical evacuation, revealed drained abscess with mild residual peripheral enhancement. There were altered signals in both cerebral hemispheres owing to probable associated inflammatory demyelination.

Both preoperative and postoperative serum and urinary osmolality were normal without any evidence of diabetes insipidus.

DIFFERENTIAL DIAGNOSIS

Although imaging findings can suggest the preoperative diagnosis, a pituitary abscess can also simulate

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other conditions because of its locally invasive nature. The differential diagnosis of a fluid sellar lesion is wide and includes cystic pituitary adenoma, craniopharyngioma, Rathke's cleft cyst infection, epidermoid tumour, status after apoplexy and, rarely, arachnoid of the colloid cyst.⁸⁹ Literature review suggests that most cases are due to infection in the Rathke's cleft cyst and only a handful of primary origin. Craniopharyngioma typically appears as a lobular, partially calcified and predominantly multicystic lesion, with a prominent suprasellar component and heterogeneous contrast medium enhancement of its solid portion. Differentiation of pituitary abscess from cystic adenoma or pituitary apoplexy is often difficult because the radiological features of these lesions may be almost indistinguishable.¹⁰ Although cyst formation may occasionally occur in infiltrative tumours, such as germinoma, an enhancing solid component is invariably present. Necrotising hypophysitis may also pose a diagnostic challenge if a non-enhancing necrotic centre is identified.

Rathke cleft cyst has a similar radiological appearance to pituitary abscess, including a variable signal intensity of the cystic contents. However, no contrast medium enhancement is typical.

Instances of super-infected Rathke cleft cyst should be of particular concern to physicians and radiologists, although the diagnosis is frequently made histopathologically after surgical drainage.¹⁰

TREATMENT

The abscess was drained by right Tran nasal trans-meatal evacuation under endoscopy guidance. Intraoperatively, the lesion contained purulent and necrotic material and no tumour, or other associated lesion, was detected.

The boy remained in a deeply comatose state for about a week completely ventilator dependent. Immediately following the surgery, there was a significant drop in blood pressure requiring maximum inotrope support which was gradually tapered off within a week. The antibiotics were escalated to cover both Gram-positive and Gram-negative organisms. There were associated episodes of seizure that required multiple anticonvulsants. Attempts of extubation, on days 7 and 13, following operation were unsuccessful. Pre-empting the need for

prolonged ventilatory requirement, a tracheostomy was performed on day 17. Finally, the child was extubated on day 29 following surgery. Thereafter, he made a satisfactory recovery and was discharged on day 42 after surgery. He was discharged on anticonvulsion medication and hormone supplementation. He was advised physiotherapy and appropriate occupational therapy.

OUTCOME AND FOLLOW-UP

We arranged a multidisciplinary follow-up of the child, weekly for the first month and monthly thereafter. The child was on regular hormone supplementation and under appropriate physical rehabilitation programme. He responded very well and made a complete motor recovery within 3 months. However, the parents reported some features suggestive of frontal lobe syndrome. The manifestations included aggressive outbursts, increased libido and tendency to harm himself. Once he even tried to jump off the roof top. These manifestations sometimes appear following trauma or prolonged neurosurgical procedures. We referred the child for neuropsychiatric follow-up. Now, he is on medications suggested by the neuropsychiatric team. During our latest follow-up, he appeared more settled. Parents have reported that he is feeling happy and has started attending special school.

DISCUSSION

Here, we report a case of primary pituitary abscess in a previously healthy 14-year-old boy. He had an acute presentation with fever, gait imbalance, slurring of speech and amnesia. An MRI at presentation revealed characteristic features of pituitary abscess. This was confirmed by drainage of purulent material during surgery. Following a prolonged and difficult neurosurgery, the boy needed maximal intensive care support. The post-operative period was so difficult, probably because of an inflammatory demyelination of the surrounding brain tissue. An element of hypoxia which might have occurred during the prolonged surgery also cannot be ruled out. He needed ventilator support for about 4 weeks and was subsequently sent home

Figure 1 Abscess location.

Figure 2 Abscess location.

after 6 weeks of hospital admission. On follow-up, we found that the child has almost completely regained gross motor function. However, he was showing certain behavioural changes suggestive of frontal lobe syndrome. This was possibly due to the difficult neurosurgery following which such manifestations are sometimes evident.

Primary pituitary abscess is an extremely rare clinical diagnosis. Although less probable, an undiagnosed, self-limited, asymptomatic regional infection contributing to the development of the abscess cannot be completely excluded. Headache, visual impairment, occasionally meningeal irritation¹¹ and endocrine disturbances are the most frequent symptoms of a pituitary abscess.

Figure 3 Abscess location.

Various endocrine disorders are reported to be associated with pituitary abscess. Between 30% and 50% of patients have anterior pituitary hormone deficiencies. Most frequently, they present with amenorrhoea and panhypopituitarism. Often they show signs of central diabetes insipidus at the onset.^{12 13} Secondary extension of the process into the suprasellar region may lead to compromise of the optic chiasm and proximal portions of optic nerves. This may cause visual disturbances such as changes in visual fields, visual acuity or diplopia in up to 50%.^{13 14}

Simmonds described the first case of pituitary abscess in 1914. This disorder is so rare that only a few hundred cases have been reported in the literature in the last century. Majority of these were published as case reports.¹³ Vates *et al*¹⁴ reported 24 cases drawn from a series of more than 3500 patients who underwent pituitary surgery. Jain *et al*² reported a case series of 6 cases of 1000 pituitary surgeries. Dutta *et al*¹² reported a series of 4 cases among 1060 cases of pituitary pathologies at their centre. Therefore, pituitary abscess represents less than 1% of all cases of pituitary disease in a busy referral practice. In children and adolescents pituitary abscess is an exceptionally uncommon clinical condition. Dalan *et al*¹³ reported 25 cases of pituitary abscess in last 5 years of which only two were in this age group. Review of existing medical literature on this issue showed about 20 cases of pituitary abscess at a young age (figures 1–3).

Learning points

- ▶ In children and adolescence primary pituitary abscess is a rare disease.
- ▶ A high index of suspicion is required for an early and accurate diagnosis that is absolutely essential for a favourable prognosis.
- ▶ Appropriate surgical intervention backed up by sound supportive intensive care can ultimately lead to near normal outcome.
- ▶ Hormonal profile should be checked pre operatively and followed up postoperatively to look for resolution.

Competing interests None.

Patient consent Obtained.

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