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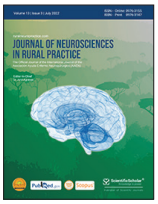

Case Report

Rathke's cleft cyst abscess from *Klebsiella*

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ABSTRACT

Pituitary abscesses are uncommon lesions accounting for <1% of all pituitary diseases. We report a case of a female microbiology technician with a rare congenital heart disease who developed an Rathke's Cleft Cyst abscess from *Klebsiella*. A 26-year-old female biotechnician with a history of congenital heart disease and subclinical immunosuppression presented with a 10-month history of weight loss, amenorrhea, and visual deterioration. There was a history of previous unsuccessful transsphenoidal surgery. Radiology revealed a cystic lesion in the sellar region. The patient underwent an endoscopic endonasal intervention and the cystic cavity was washed with gentamicin, and the patient received meropenem postoperatively. The patient was followed up and had gradual improvement in her overall health, complete normality of her menstrual cycle, her visual field recovering to near normal and improving, no recurrence, and a stable cyst on magnetic resonance imaging.

Keywords: Rathke's cleft cyst, Abscess, *Klebsiella*, Pituitary abscess

INTRODUCTION

Pituitary abscesses are rare lethal conditions accounting for <1% of all pituitary diseases.[1-3] Approximately 70% are primary abscesses (occurring in normal pituitary gland);[1-3] latter 30% being secondary abscesses which are infections in pre-existing pituitary adenomas (most common underlying pathology), craniopharyngioma, or Rathke's Cleft Cyst (RCC).[1-3] RCC abscesses are exceedingly rare, with <60 reported cases in the literature.[1] We report the first case of a RCC abscess from *Klebsiella*.

CASE REPORT

A 26-year-old female biotechnician presented with a 10-month history of weight loss, visual blurring, amenorrhea (all progressive), and finger clubbing. She appeared cachectic and weighing 44.4 kg (body mass index, BMI 18). Her medical history included doublet outlet left ventricle, ventricular septal defect, and pulmonary artery stenosis for which she had undergone a conduit replacement post-Rastelli procedure in 2006 (aged 9) and revision surgery in 2009. Magnetic resonance imaging (MRI) brain [Figure 1] confirmed a cystic lesion in the sellar/suprasellar region with thick peripheral walls enhancing with contrast. The normal pituitary gland could not be visualized and the cyst compressed the optic chiasm. The mass had signals hyperintense on T2 and hypointense on T1 with facilitated diffusion and no susceptibility artifacts. Automated perimetry confirmed bitemporal hemianopia and complete right temporal field loss.

The patient's previous neurosurgeon attempted an endoscopic endonasal approach but failed to proceed beyond the nasal passage. When referred to us, the senior authors utilized an endoscopic endonasal approach to approach the lesion. The previous surgery was evidently visible along the nasal passage, with the mucosa over the anterior wall of the sphenoid sinus and keel intact. The sellar floor was opened, following durotomy a gush of pus emerged after the lesion was nicked. Cavity lining was thin, friable, and moderately vascular capsule was noted, of which only biopsy samples were obtained from the wall. No excision was attempted. Pus samples were collected, then the cavity was thoroughly cleansed/washed with gentamicin solution and left open. Nasal packing was not done. The post-operative course was uneventful, and a 24-h MRI revealed near complete drainage of the cyst cavity with a decompressed optic chiasm but suspicion of small residual fluid volume present on fluid-attenuated inversion recovery (FLAIR) but not appreciable on T2 [Figure 2].

Klebsiella pneumoniae sensitive to meropenem was cultured. The patient was discharged on the 5th postoperative day after

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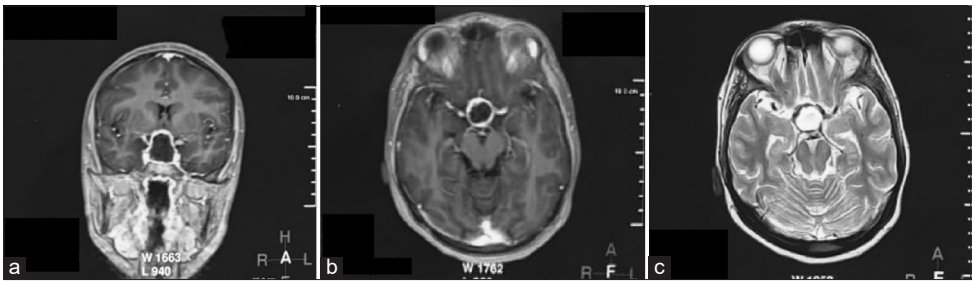

Figure 1: Pre-operative MRI. (a) coronal T1 with contrast, (b) axial T1 with contrast, (c) T2 sequence. MRI confirmed a 2.1 × 2.5 × 2.8 cm cystic mass in the sellar/suprasellar region with thick peripheral wall enhancement without visualization of normal pituitary. The lesion's maximum thickness was 4 mm anteriorly. T2 signals demonstrate hyperintensity.

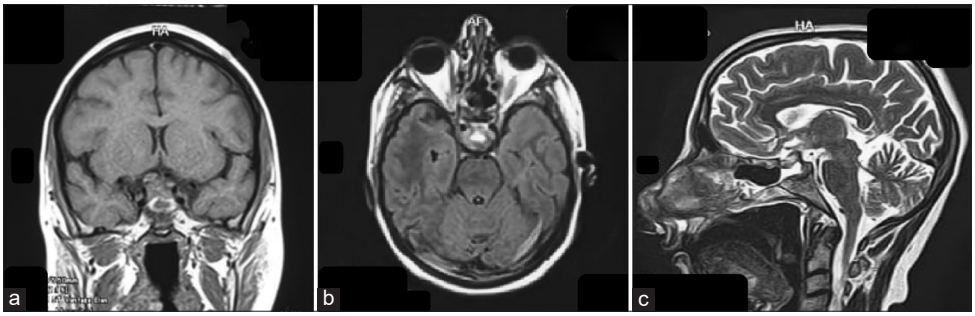

Figure 2: Immediate post-operative (a) T1 coronal, (b) fluid-attenuated inversion recovery (FLAIR) sequence, (c) T2 sequence. There is near complete drainage of the cyst cavity with a decompressed optic chiasm but a suspicion of small residual fluid volume present on FLAIR but not appreciable on T2.

initiating meropenem. Automated perimetry performed 1 week later showed a significant improvement in her vision. Histopathology report confirmed the lesion to be a RCC. Her cardiology consultant later performed a cardiac echo to exclude valvular abnormalities/vegetation. The patient's hormone profile demonstrated impaired pituitary function [Table 1], for which she takes thyroxine and daily prednisolone.

Her last clinic visit (12 months) confirmed her menstrual cycles had normalized. She has returned to work and weighs 52 kg (BMI 21.2). Follow-up MRI showed no further changes, but a resolution of the fluid seen on postoperative FLAIR MRI, and a stable cyst. The patient also reported a further improvement in her vision.

DISCUSSION

Pituitary abscesses present from either the mass effect compressing the surrounding structure (manifested by raised ICP and bitemporal hemianopia) or functional impairment of the pituitary gland with symptoms of hypopituitarism. A recent review on RCC's abscess reported that headaches were present in 70% of patients, visual disturbances in 35%, and hypopituitarism in 80%.[1]

RCC abscesses may manifest with highly unusual radiological findings.[4] Pituitary abscesses are a diagnosis of exclusion, and a clinical suspicion can be strengthened on pus observed intraoperatively, but diagnosis can only be confirmed with histopathology and microbiological cultures.

There are three risk factors in the literature for developing pituitary abscesses: immunosuppression, underlying lesions, and prior surgery of the sellar region.[1] The underlying mechanism is hematogenous dissemination and/or local spreading to/from the sphenoid sinus.[1] In our patient's case, while a valvular/vegetative mechanism for the hematogenous spread was excluded by cardiology, there is a degree of subclinical immunosuppression given her cardiac history. The patient had an underlying RCC and as she underwent a previous (but unsuccessful) transsphenoidal surgery, all these factors likely contributed to her RCC abscess development. As she continued working in a microbiological laboratory, this was likely the source of contamination from *Klebsiella*.

The mainstay treatment of pituitary abscesses is surgical drainage and antibiotic therapy.[1-3] Early surgical drainage aims to decompress mass effect and prevent hypopituitarism.[1-3] The transsphenoidal approach is the most common but transcranial approaches have been reported in cases with extensive suprasellar involvement.[2,3] However, these cases are from 1956

Table 1: Hormonal function test and improvement from baseline to 3-month postoperative.

Parameter	Preoperative	3-month postoperative	Reference range
Serum TSH	0.12 uIU/mL	4.86 uIU/mL	0.4–4.2 uIU/mL
Serum cortisol (early morning)	2.5 uIU/mL	0.96 uIU/mL	3.7–19.4 uIU/mL
Serum prolactin	104.4 ng/mL	5.1 ng/mL	5.1–27 ng/mL
Serum follicle-stimulating hormone	0.67 IU/L	2.07 IU/L	1.38–5.47 IU/L
Serum luteinizing hormone	<0.12 IU/L	0.73 IU/L	0.56–14 IU/L

Red highlights are abnormal values, green highlights are within normal range

and 1998.[2] There is now widespread uptake and refinement of endoscopic/extended endoscopic approaches; thus risk of CSF contamination from a transcranial approach for a pituitary abscess must be considered. Post-operative antibiotics are essential, reducing the recurrence rates from 31% to 13%.[1] Most patients recover well with partial or complete endocrine recovery requiring hormonal replacement as appropriate.[2,3]

Interestingly, RCC abscess are suggested to be associated with a high recurrence rate relative to other causes of secondary pituitary abscesses.[1,5,6] One patient with RCC abscess even underwent transsphenoidal surgery 4 times due to recurrence causing meningitis.[6] However, given the limited information from the type of these studies and overall small sample size, the suggestion of RCC abscess having a higher recurrence rate is merely an observation, and numerous other variables likely contribute to recurrence, independent of the underlying pituitary pathology.

In our case, the excision of the actual RCC was not attempted to prevent an already compromised pituitary function/immunocompromised and deteriorated patient from further decline. The priority was surgical decompression of the chiasm and clearing the abscess, which proved to be the correct decision as the patient's BMI improved from an anorexic range to normal. She has regained regular menses and has returned to work. The RCC is stable on imaging and is being radiologically monitored for growth.

CONCLUSION

Pituitary abscesses whilst rare should be on the differential for sellar lesions, especially with a high index of clinical suspicion in cases where the clinical presentation and history suggest an infective cause. Known risk factors include prior transsphenoidal surgery, immunosuppression, and radiation. From our case, a history of occupational exposure to microorganisms may be a contributing factor.

Ethical approval

The individual case report was written after obtaining written informed consent from the patient, to report on their clinical case, and use their radiology scans and histopathology images for publication.

Research registration

Not applicable.

Provenance and peer review

Not commissioned and externally peer reviewed.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Aranda F, García R, Guarda F, Nilo F, Cruz JP, Callejas C, *et al*. Rathke's cleft cyst infections and pituitary abscesses: Case series and review of the literature. Pituitary 2021;24:374-83.
2. Zhang X, Yu G, Du Z, Tran V, Zhu W, Hua W. Secondary pituitary abscess inside adenoma: A case report and review of literature. World Neurosurg 2020;137:281-5.
3. Agyei JO, Lipinski LJ, Leonardo J. Case report of a primary pituitary abscess and systematic literature review of pituitary abscess with a focus on patient outcomes. World Neurosurg 2017;101:76-92.
4. Coulden A, Pepper J, Juszczak A, Batra R, Chavda S, Senthil L, *et al*. Rathke's cleft cyst abscess with a very unusual course. Asian J Neurosurg 2022;17:527-31.
5. Jain KC, Varma A, Mahapatra AK. Pituitary abscess: A series of six cases. Br J Neurosurg 1997;11:139-43.
6. Uchiyama T, Sakai K, Asanuma M, Aoyama T, Hongo K. Pituitary abscess manifesting as meningitis and photophobia associated with Rathke's Cleft cyst in a child-case report. Neurol Med Chir 2011;51:455-9.

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