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Isolated Pituitary Abscess with Postoperative PRES Syndrome, Rare Combination Leading to Dismal Outcome

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Abbreviations: PA: Pituitary Abscess; PRES: Posterior Reversible Encephalopathy Syndrome and TSS: Transsphenoidal excision.

Introduction

Pituitary abscess (PA) is very rare accounting for 0.2%–0.6% of all pituitary lesions [1]. Only around 270 case reports have been described so far since the first case was described by Heslop in 1848 [2]. Pituitary abscess is an uncommon disease that usually presents with vague symptoms. To reach a diagnosis in such cases is usually difficult. The condition is considered a life-threatening one, however, fortunately the major number of cases have a rather chronic course. Posterior Reversible Encephalopathy Syndrome (PRES) is a rare condition in which parts of the brain are affected by swelling, usually as a result of an underlying cause. It is usually associated with chronic renal disease, hypertension, and connective tissue disorders. It is rarely associated with pituitary abscess. Preoperative diagnosis of PA is a dilemma as symptoms such as pituitary abnormal function are non-specific, radiological findings are not distinctive of an abscess and symptoms like headache or visual disturbances are vague. In addition, infectious symptoms such as fever, meningism and high white cell count (WBC) are not seen in around 70–80% of cases [3]. Therefore, majority of the cases are diagnosed either post mortem or postoperatively [3]. Pituitary abscess can develop in a normal pituitary gland (about 70%) or in pre-existing pituitary pathology (about 30%). Of the pre-existing lesions, a pituitary adenoma is most common, followed by a Rathke's cleft cyst, craniopharyngioma and lymphoma [4]. There is still some controversy regarding the pathogenesis and treatment of PA, such as infection sources, imaging features, and mode of neurosurgical interventions. To further understand and provide a reference basis for these issues, more data are needed. Here, we present a case of Pituitary Abscess that was diagnosed and treated at our institution.

Case Report

54 year old female patient presented to us with progressive visual loss since last eight months, beginning first in the left eye and gradually progressing to involve both eyes. She also had associated headache since last 6 months. She was investigated for it with MRI brain which showed a sellar/ suprasellar lesion? Pituitary adenoma with hydrocephalus. She underwent CSF diversion for the same in other hospital and then referred to us for definitive management. It was not associated with fever, vomiting, seizures. On neurological examination, her vision was Finger counting + close to face on the left side and was 06/60 on the right side. Perimetry was done for right side and it was s/o generalized field defect. On the left side, perimetry was not possible due to poor vision. Contrast Enhanced CT scan was obtained and it was also suggestive of sellar / suprasellar region.

Hence she was evaluated further as a pituitary adenoma and was taken for transsphenoidal endoscopic excision of pituitary tumor. Intraoperatively there was frank pituitary abscess, containing yellowish pus, which was drained completely. Postoperatively patient was put on injectable

antibiotics. On post -operative day 07 patient developed seizures initially focal then progressing to generalized seizures. Her sensorium gradually deteriorated due to status epilepticus. Repeat MRI scan was done which showed multiple infarcts s/o Posterior Reversible Encephalopathy Syndrome (PRES). CSF studies were done at this time and it was having negative culture and not having picture of meningitis. Then she also went into severe hypernatremia. Despite all efforts of correction, her hypernatremia was refractory. She gradually deteriorated further despite all efforts and expired.

Discussion

Pituitary abscess (PA) is a rare intracranial mass with high mortality [5]. PAs are uncommon, with only about 270 cases globally reported to date [6]. PA may occur in de novo previously healthy normal pituitary gland in 70% of cases [7,8]. Of the pre-existing lesions, a pituitary adenoma is most common, followed by a Rathke's cleft cyst, craniopharyngioma and lymphoma [4]. Moreover, immunosuppression, previous irradiation or surgical procedures to the pituitary gland are also known risk factors for PA [9,10]. In most cases of PA, no apparent causes are generally found [9].

Anterior pituitary hypo function is the most common symptom of PA, such as anorexia, weakness, amenorrhea, vomiting, and hypogonadism [9]. Headache is the second-most common symptom. In a series of 66 patients, 46 reported a headache [6]. Visual disorders, such as hemianopsia, are common, in one series [11] 57% of patients with PA presented with visual disorders. Central diabetes insipidus has an incidence of 41.4%–69.7% among PA patients [9,12] . However, it is uncommon for pituitary adenomas to present with diabetes insipidus [13]. Heary reported that diabetes insipidus was only present in 10% of pituitary adenoma cases [14]. Therefore, diabetes insipidus in the diagnosis of PA is of great significance. Fever with signs of meningeal irritation was inspected in only 25% of patients [10].

MRI is one of the most effective imaging approaches for pituitary lesions. Typical features of PA include a single cystic or partially cystic mass that appears hypo intense in T1-weighted and hyper intense in T2-weighted imaging, and shows rim enhancement after gadolinium injection. However, in one series only 66.7% of the patients exhibited such typical features on MRI [6]. Even our patient had not presented with such typical features, she had presented with a rim enhancing mass in the sellar/suprasellar region. Hence, it can be concluded that MRI is not that very specific for recognising Pituitary Abscess from other pathologies of the supra-sellar region. Hence confirmative diagnosis of PA can be made only after intraoperative drainage of pus as done in our case.

The pus was drained and sent for investigations. The gram staining showed pus cells with few gram-positive cocci in chains and few gram-positive bacilli seen. However, on final culture, no growth was obtained. Microorganisms such as Staphylococcus and Streptococcus are the most common isolated pathogens from pituitary abscess [10]. Additionally, plenty of different microorganisms have been seen in surgical cultures from PA such as *Neisseria spp. Micrococcus, Citrobacter spp., Escherichia coli, Brucella, Salmonella, Corynebacterium*, and *Mycobacterium* [10]. However, *Aspergillus fumigatus* was most frequently noted in cases of secondary PA [6]. In our patient, there were plenty pus cells with few bacteria but however, there was no organism isolated on culture. This could be because the patient had received antibiotics preoperatively. Or it could be due to the insufficient volume of pus prevented culturing of the bacteria [15]. 50% of cases can culture the pathogens [16].

Our patient underwent trans sphenoidal endoscopic drainage of abscess followed with antibiotic therapy (Inj Piperacillin+Tazobactam iv for 07 days , Inj Amikacin for 05 days , Inj Metronidazole for 03 days) with hormonal replacement therapy , patient had improved very well with this protocol. Transsphenoidal excision (TSS) and antibiotic therapy are the main treatment for PA [6,9,12]. Transsphenoidal resection is preferred over craniotomy, as TSS has the advantages of efficacy, safety, and minimal invasiveness and craniotomy can cause infectious substances to spill into cerebrospinal fluid [12,17]. Antibiotic therapy should be instituted for approximately 4–6 weeks [17]. Hormone replacement therapy is needed based on pituitary hormone deficiency [17,18].

Then on 7th post-operative day , she started getting focal myoclonic seizures involving left upper limb and it was persistent despite upgrading to two anticonvulsants – status epilepticus. The incidence of status epilepticus in post-operated pituitary abscess is 0.2% [17]. She was diagnosed as having developed Posterior Reversible Encephalopathy Syndrome (PRES).

PRES is a rare condition in which parts of the brain are affected by swelling, usually as a result of an underlying cause. The name of the condition includes the word "posterior" because it predominantly though not exclusively affects the back of the brain (the parietal and occipital lobes). It is often—but by no means always—associated with acute hypertension [19,20]. Chronic kidney disease and acute kidney injury are both commonly present in patients with PRES [21], and PRES is strongly associated with conditions that co-exist in patients with renal disease, such as hypertension, vascular and autoimmune diseases, exposure to immunosuppressive drugs, and organ transplantation. There is very rare association of PRES with brain abscess (0.1%) [22]. There is

no association reported with pituitary abscess seen however our patient was a known case of hypertension and Type 2 DM. Brain abscess is rare cause of PRES so it could be a rare complication of pituitary abscess that was seen in this patient. 1A

PRES evolves over a matter of hours, with the most common presenting symptoms being seizures, disturbed vision, headache, and altered mental state [23] (Figure 1). In our patient also there was myoclonic seizure involving the left upper limb which gradually became status epilepticus.

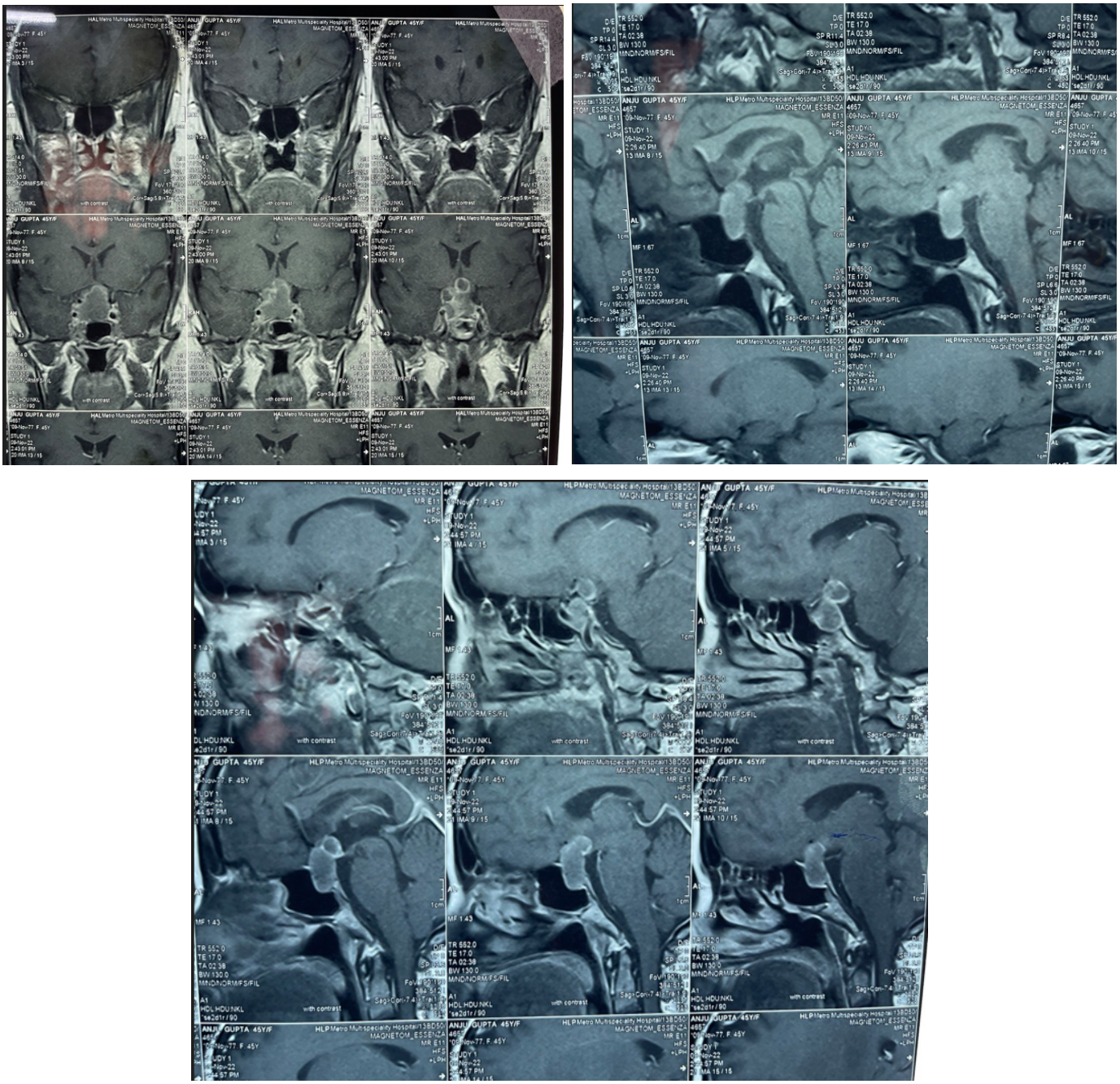

Figure 1a-c: Pre-operative MRI images showing the lesion.

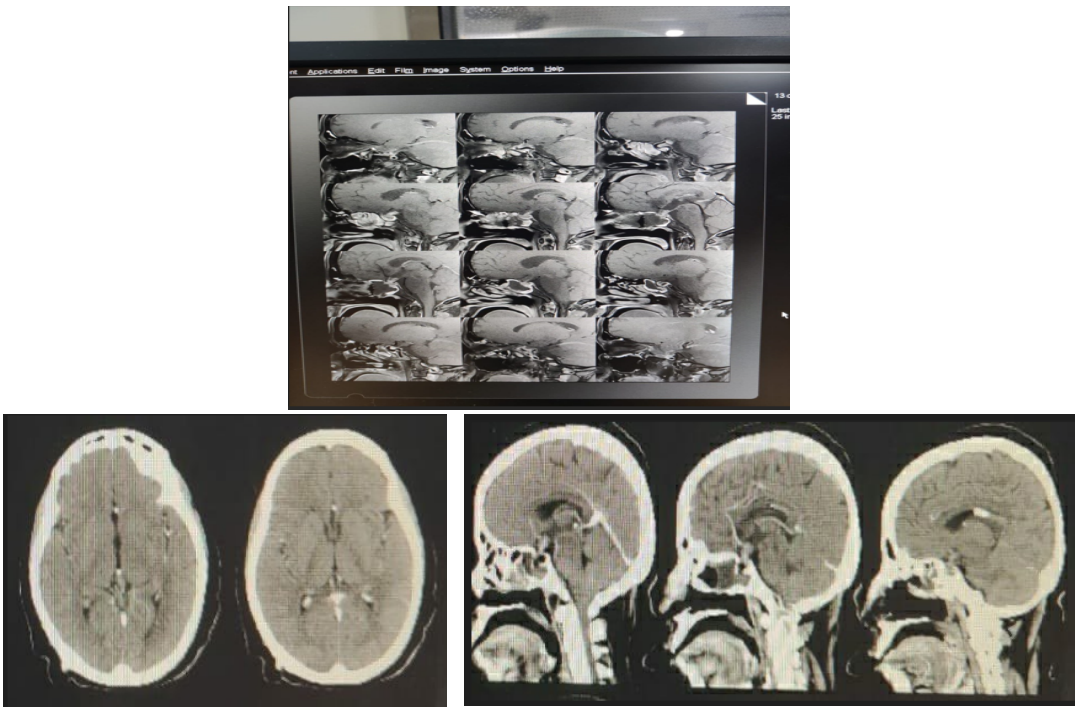

Figure 2a-c: Post Operative images.

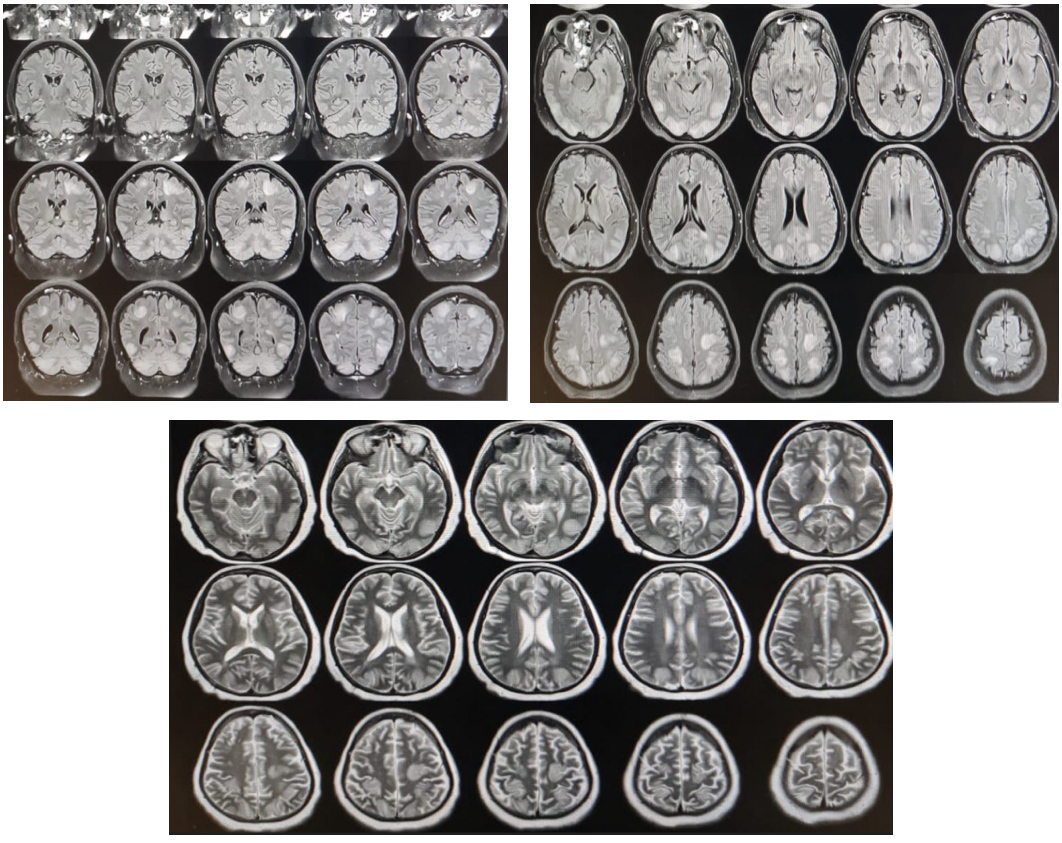

Figure 3a-c: MRI Images showing PRES infarct.

MRI is diagnostic modality of choice. Typical MRI findings in PRES are of bilateral white-matter abnormalities in vascular watershed areas in the posterior regions of both cerebral hemispheres, affecting mostly the occipital and parietal lobes [19,21,24]. Atypical features— including haemorrhage, asymmetrical changes (such as in the case described by Ogawa and colleagues), isolated involvement of the frontal lobes, and cortical lesions [19,21,24] are common. Our patient had ischemic infarcts spread over watershed areas in B/L cerebral hemispheres.

No clinical trials have evaluated the management of PRES, but rapid withdrawal of the trigger appears to hasten recovery and to avoid complications [25].

Antiepileptic drugs should be used to treat seizures, and anaesthesia and ventilation should be instituted in generalized status epilepticus and to protect the airway in obtund patients [20]. Corticosteroids should theoretically improve vasogenic oedema, but there is no evidence for their use in PRES [20]. We gave aggressive antiepileptic treatment and gave mechanical ventilation to our patient. She was already receiving tab. Methyl prednisolone as part of hormone replacement.

With adequate treatment, 70-90% of people with PRES make a full recovery within hours to days. 8–17% of people with PRES die [26]. Non-resolution of MRI abnormalities has been linked with poorer outcomes [27]. Factors that predict poorer prognosis are the person's age, the level of C-reactive protein in the blood (a marker of inflammation), altered mental state at the time of diagnosis, and altered markers of coagulation [26]. Our patient had many of the above proven factors a/w poor prognosis- she had non resolving MRI abnormalities, she was elderly age with raised CRP, had altered mental status at the time of diagnosis of PRES .She was diabetic and also had abnormalities in corpus callosal (infarct) .Hence it may be postulated as to why she did not recover and even succumbed.

Conclusion

Pituitary abscess mimicking pituitary adenoma, without any infective symptoms, focus is rarely reported. Post-operative PRES syndrome in a case of pituitary abscess is not reported in literature. Both these conditions need aggressive management. As prognosis in both conditions is poor independently. Early surgical intervention with complete evacuation of pituitary abscess, endoscopically followed by higher antibiotics for 21 days is needed. Intraoperatively care needs to be taken not to open the arachnoid when abscess is seen. PRES syndrome responds well to medical management.

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