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CASE REPORT

Pediatric Sellar-Suprasellar Tubercular Abscess: A Case Report and Literature Review

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ABSTRACT

Sellar-suprasellar tuberculoma represents 1% of all intracranial tuberculomas, which can convert into a pituitary abscess.

Objectives: This paper aims to present a case of a common lesion in an uncommon site, discuss the challenges in diagnosis in terms of clinical manifestations, imaging and histologic findings, discuss a holistic approach to treatment, and enumerate identified clinical outcomes of reported cases in literature.

Methodology: This paper presents a case report of a sellar-suprasellar tubercular abscess, and reviews similar cases reported in literature.

Result: This is a case of a 16-year old female, Filipino, presenting with a chronic history of intermittent headache, fever, blurring of peripheral vision, polyuria, and increased sleeping time. On neurologic examination, the patient had bitemporal hemianopsia and decreased visual acuity on the right eye. Neuroimaging revealed a hypodense lesion at the sellar-suprasellar area with rim-enhancement on CT and MRI, and laboratory findings suggestive of panhypopituitarism. She underwent Right Pterional Craniotomy and intraoperatively there was note of a firm, yellowish capsule with intracapsular yellowish viscous fluid, which was positive for acid fast bacilli. Marsupialization of abscess was performed and hormonal replacement and anti-tubercular medications were given.

Conclusion: Tuberculoma in the sellar-suprasellar area, can impinge on the optic chiasm, producing bitemporal hemianopsia and pituitary dysfunction. It presents like other sellar-suprasellar masses with non-specific symptoms and these masses share similar features on cranial CT scan. Due to the complexity of the disease, treatment of sellar-suprasellar TB requires integrated management of an infectious disease expert, neurologist, neurosurgeon, endocrinologist, and adolescent medicine specialist. Outcomes of four other cases found in literature were generally good after aspiration or drainage of the abscess followed by TB treatment for 15 to 18 months with resulting improvement in vision, marked reduction in the size or complete resolution of the mass, but with one case having loss of pituitary function.

KEYWORDS: *sellar-suprasellar mass, tubercular abscess, pituitary abscess, pediatric sellar abscess*

INTRODUCTION

Pituitary abscess is a rare potentially life-threatening disease, occurring in all age groups, and is estimated to account for less than 1% of clinically apparent pituitary diseases.¹ It can have a chronic course, mimicking a slow-growing tumor that produces non-specific symptoms due to mass effect, such as headache, panhypopituitarism, and visual field defects. While neuroimaging plays a crucial role in confirming the location and in characterizing the lesion, it can also mislead clinicians because other disease processes can also present similarly as cystic masses on the sellar area.

This case was initially diagnosed as craniopharyngioma but was later on found to be an abscess intraoperatively. On further work-up, the abscess was positive for acid fast bacilli. This case report aims to present a case of a common lesion in an uncommon site; discuss the challenges in diagnosis in terms of clinical manifestations, imaging, and histologic findings; discuss the holistic approach to the treatment of such a disease; and enumerate identified clinical outcomes of similar reported cases in literature.

This case report not only presents as a rare presentation of a common disease, it also highlights the importance of a high index of suspicion, timely intervention, and holistic approach to management as these can improve the prognosis of patients with tubercular abscess.

CASE

A 16-year old female presented with a 3-month history of intermittent headache, episodic esotropia, and “clumsiness” described as frequent bumping of bilateral shoulders onto walls when walking. She was subsequently noted to have increased sleeping time with accompanying polyuria, prompting consultation at a local hospital in Gerona, Tarlac, where cranial CT scan was done with note of a hypodense lesion about 3x3 cm in the sellar-suprasellar area.

Cranial MRI done at the same institution showed a lobulated heterogeneous suprasellar mass with rim enhancement measuring 3.0x2.9x3.5 cm with no parenchymal signal abnormalities. Hormonal work-up showed the following results: FSH 0.59 mIU/mL (NV 3.5-12.5mIU/mL), LH <0.100 mIU/mL (NV 2.4-12.6mIU/mL), cortisol 54.53nmol/L (NV 171-536nmol/L). She was then referred to our institution for further management.

Upon initial consultation, she was drowsy with the following vital signs: blood pressure 100/70 mmHg, heart rate 89 beats per minute, respiratory rate 19 cycles per minute, temperature 36.5⁰ Celsius. On further physical examination, sexual maturity rating of breasts is at 3, and genitalia at 2. The rest of the physical examination was normal. On neurologic exam, patient was drowsy, followed commands with prodding, pupils were 3mm equally and briskly reactive to light with primary gaze at midline. There was decreased visual threat bitemporally. No cranial nerve deficits were noted. She was normoreflexive, with supple neck, no nystagmus. Assessment was Sellar-suprasellar mass probably Craniopharyngioma vs Pituitary macroadenoma. Other hormonal work-ups included TSH 0.20 mIU/mL (NV 0.3-3.8 mIU/mL), FT4 5.7 pM (NV 11-24 pM), cortisol 86.6 nmol/L (NV 138-690 nmol/L), prolactin 59.5 mIU/mL (NV 92-868 mIU/mL). Due to findings of panhypopituitarism, she was started on hormonal replacement with Hydrocortisone and Levothyroxine. There was subsequent improvement in sensorium—and later on became awake and was able to follow commands. Visual field testing showed bitemporal hemianopsia. The plan of management was craniotomy with excision of tumor. While securing funds for surgery, she was discharged improved on Hydrocortisone 20mg/tab 2 tabs TID and Levothyroxine 50mcg/tab 1 tab OD.

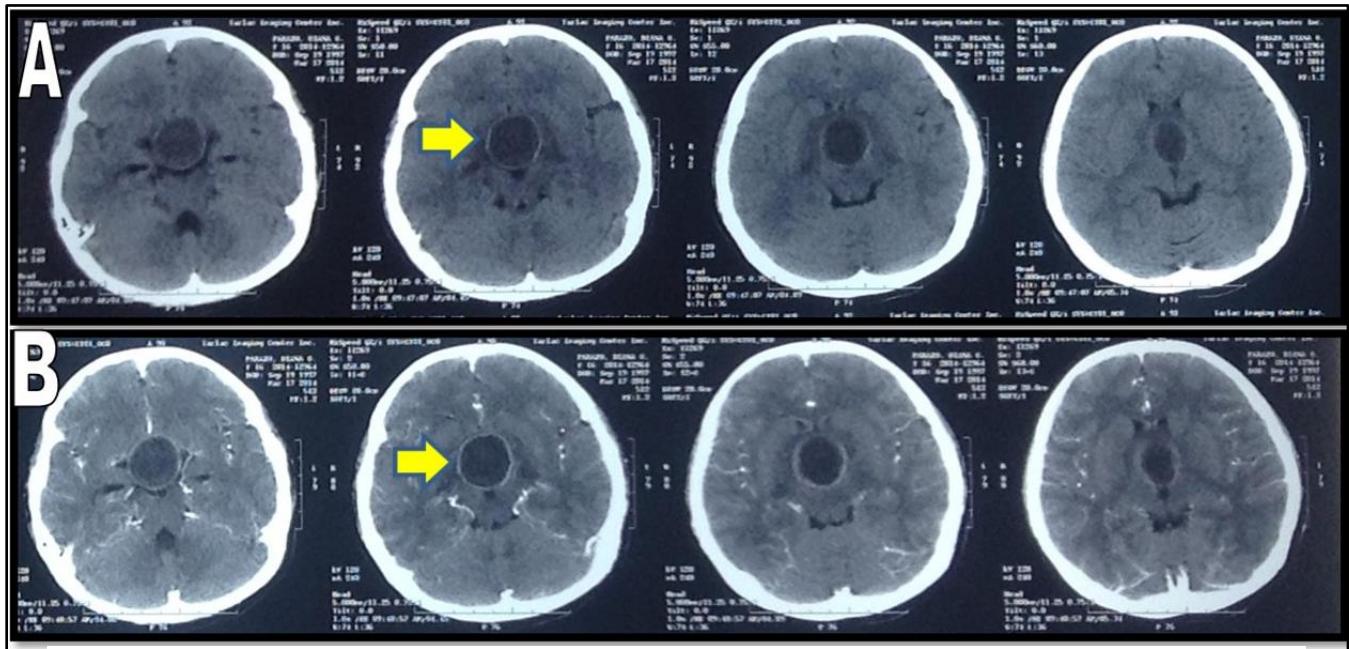


Figure 1. A) Plain cranial CT scan showing hypodense lesion (yellow arrow) measuring 3x3cm at the sellar-suprasellar area. B) Contrast study showing marginal enhancement of the lesion.

She was on regular follow-up with her neurologist, endocrinologist, neurosurgeon, with good compliance to medications.

In the interim, she was apparently well until three days prior to admission, when she developed high-grade fever. This was accompanied by increased sleeping time, prompting her second admission at our institution two months after the first admission. She was seen at the emergency room awake, conversant, not in cardiorespiratory distress with stable vital signs. Optic nerve examination showed no evidence of papilledema. There was bitemporal hemianopsia elicited on confrontational visual field testing. No other cranial nerve or sensorimotor deficits were noted. At the ward, she had episodes of headache, fever, and blood pressure spikes as high as 130/80. Fluid balance was negative. Serial serum electrolytes with simultaneous urine electrolytes were determined. The patient consistently had hypernatremia and hypokalemia with urine specific gravity ranging from 1.001 to 1.003. Oral fluids were given *ad libitum* and hydrocortisone as well as levothyroxine were

continued. Desmopressin (Minirin) was eventually started and free water deficit correction was done. The neurosurgical plan was to proceed with contemplated procedure.

She had intermittent low-grade fever with occasional spikes reaching 39°C. Blood count showed leukocytosis with neutrophilic predominance. Cefuroxime was started for urinary tract infection. Due to persistence of fever, Cefuroxime was shifted to Ceftazidime and Amikacin for a possible nosocomial infection. Acute phase reactants were elevated, but blood cultures were negative. The patient was cleared for surgery after 10 days of IV Ceftazidime.

On her 13th hospital day, she underwent Right pterional craniotomy. Intraoperative finding was a firm yellowish capsule at the sellar-suprasellar area with intracapsular yellowish viscous fluid. On frozen section, the specimen was consistent with fibrocollagenous tissue in acute inflammation. Marsupialization of abscess was done. Cerebrospinal fluid studies were sent for analysis revealing leukocytosis with lymphocytic

predominance, with increased levels of glucose and total protein. On Gram stain, no organisms were seen.

Post-operatively, the patient reported improved peripheral vision. There were no recurrences of headache, blurring of vision, episodes of vomiting, or nausea. However, the patient still had febrile episodes despite nine days of antibiotics. Abscess AFB smear revealed +2/30 fields. Further TB work-up showed an unremarkable chest x-ray but sputum AFB was +1. The patient was started on quadruple anti-TB medications. On the 4th day of treatment, the fever began to lyse.

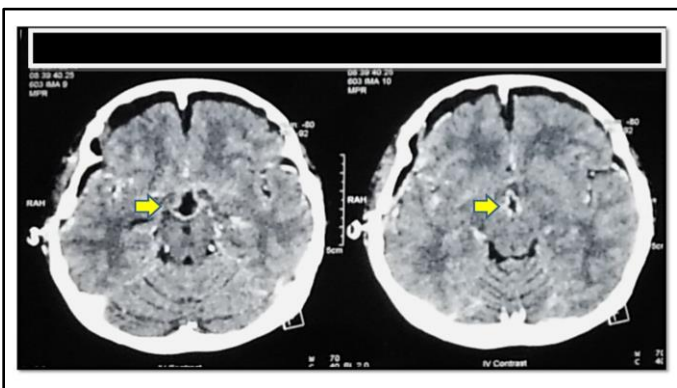


Figure 2. Post-operative cranial CT scan with contrast showing a decreased size of the mass.

Repeat CT scan showed a decrease in the size of the previously described predominantly fluid-attenuating mass lesion with well-defined hyperdense enhancing borders and a punctate internal calcification in the sellar-suprasellar region. It now measured approximately 2.8 x 2.2 x 1.7cm from a previous 4.3 x 2.7 x 3.0cm. The patient was discharged within a week after initiating anti-TB medications. The final diagnosis was Disseminated Tuberculosis (sellar-suprasellar tubercular abscess, PTB) with Panhypopituitarism (hypocortisolism, hypoprolactinemia, hypothyroidism, diabetes insipidus) S/P Pterional Craniotomy, Right; Marsupialization of Abscess.

DISCUSSION

The pituitary gland is situated in the sella turcica which is a depression within the sphenoid bone. It is connected to the hypothalamus by the infundibulum or pituitary stalk, which closely lies posterior to the optic chiasm. A disease process localized at the sellar-suprasellar area may exert a mass effect and impinge on the optic chiasm, producing bitemporal hemianopsia. The same mass effect will also produce pituitary dysfunction.

There is a multitude of conditions that can exert a mass effect and produce the clinical manifestations seen in our patient hence, performing imaging studies is of paramount importance to confirm the location of the lesion as well as to gain clues to the type of the disease process involved. In our patient, imaging revealed a hypodense mass with rim-enhancement, which can be any of the following: craniopharyngioma, Rathke's cleft cyst, or pituitary abscess.¹

Craniopharyngiomas are benign tumors that grow near the pituitary gland. They can be solid tumors or cysts. Approximately 10-15% of pituitary tumors are craniopharyngiomas. They are most commonly found in children, teenagers, and adults older than 50. The MR appearance of craniopharyngiomas depends on the proportion of the solid and cystic components, the content of the cyst(s), and the amount of calcification.² The solid portions of the tumor appear as iso- or hypointense relative to the brain on pre-contrast T1-weighted images, but can also have a mottled appearance owing to calcific regions. They are usually of mixed hypo- or hyperintensity on T2-weighted sequences, and heterogeneously enhance following Gd administration.^{3,4}

Rathke's cleft cysts are non-neoplastic cysts arising along the craniopharyngeal duct from the remnant of squamous epithelium of Rathke's pouch. They consist of a single layer of cuboidal or columnar epithelial cells with mucoid, cellular or serous components in the cyst fluid. They are often

discovered incidentally, but may become symptomatic in a minority (5-9% of all surgically-resected sellar lesions) when intracystic bleeding or infection occurs, leading to symptoms similar to those of our patient. On MRI, Rathke's cleft cysts appear well-circumscribed, centrally located spherical or ovoid, non-calcified cyst lesions of the sellar region. The lesion is fairly isointense to CSF.

Pituitary abscess is a rare potentially life-threatening disease, occurring in all age groups, and is estimated to account for less than 1% of clinically apparent pituitary diseases.¹ The typical MR features of an abscess are the presence of a round cystic or partially cystic sellar mass that appears as hypo- or isointense on T1 and hyper- or isointense on T2, with an enhanced rim after Gd injection and a central cavity that is isointense to the brain.^{5,6} The sella may be enlarged and, occasionally, extensively eroded.

Only 200 cases of primary pituitary abscesses have been reported in literature, primarily in adults. Diagnosis may be difficult, as symptoms can be nonspecific, and without signs of an infectious process. Median time from the onset of symptoms to diagnosis is 6 months.² Most patients present with a chronic and indolent course with few manifestations thus mimicking a pituitary tumor.⁵ The most common symptoms are neurological (headache, visual impairment and cranial nerve palsy), but manifestations of hypopituitarism are frequent. Diabetes insipidus and headache are the most common presenting complaint (70%)⁷ and over half of the patients complain of visual disturbances. Most patients (85%) have partial or total hypopituitarism (including prolactin deficiency). The following are the identified etiologic agents of pituitary abscesses: Gram-positive cocci, fungi, and other organisms including *Mycobacterium tuberculosis*, *Toxoplasma*, *Clostridium difficile*, and *Pseudomonas aeruginosa*. In our patient, the abscess was positive for acid-fast bacilli.

Sellar-suprasellar tuberculosis may present as a tuberculoma,⁸ which are inflammatory granuloma cells surrounded by a rich network of collagen and reticulin. Sellar-suprasellar tuberculoma represents 1% of all intracranial tuberculoma. The tuberculoma can undergo caseation resulting in a pituitary abscess. A TB abscess is an encapsulated collection of pus containing viable tubercle bacilli and without typical tubercular granuloma and epithelioid cells. A review of literature on pituitary or sellar tubercular abscess yielded only five cases.^{9,10,11,12,13}

The first report of a TB abscess by Dutta⁹ and colleagues was in 2006, on a 13-year old Indian boy who presented with meningitis, deteriorating vision and panhypopituitarism. Cranial MRI showed a solid-cystic lesion with suprasellar extension. The patient underwent stereotactic aspiration and biopsy of abscess which was positive for AFB. He was treated with anti-TB medications and steroids. The patient had improved vision after 2 weeks, and follow-up imaging showed a marked reduction in the size of the lesion, 6 months into treatment. This patient, however, was lost to follow-up.

The second case was reported in 2008¹⁰ on a 45-year old man from India with holocranial headache for 6 months, A sellar mass with peripheral rim enhancement was seen on cranial MRI. Trans-sphenoidal excision was done and the specimen was positive on mycobacterial culture. He was given anti-TB medications and hormonal replacement. The patient eventually had complete loss of pituitary function and required permanent hormonal replacement therapy.

The third report in 2011¹¹ presented similarly: a 27-year old from India with progressive headache, occasional vomiting, hypothyroidism, and hyperprolactinemia. Cranial MRI revealed a cystic sellar lesion with suprasellar extension. Drainage and decompression of the cyst was done. The cyst cavity contained creamy pus which was positive for mycobacterial colonies after six weeks. She was also given anti-TB medications. The patient

had no residual cyst on post-operative imaging, had normal thyroid function after three months with pituitary hormone status recovered to normal after six months, and as of the time of writing of the report, was doing well at 12 months.

The fourth report¹² in 2012 was a case of a 50-year old Filipina, diabetic with gradual loss of bitemporal vision, headache, polyuria and polydipsia. Trans-sphenoidal surgery was done which yielded AFB from the abscess. She was treated with anti-TB medications, vancomycin and cefepime. Outcomes reported only included improved vision and subsequent development of central diabetes insipidus.

Finally, the last report¹³ in 2016 was on a 44-year old Indian female, a known case of tuberculous meningitis for one year who now presented with behavioral changes and increased frequency of micturition. Cranial MRI revealed a hypointense lesion on T1 images in the hypothalamic region, iso-intense on T2, and restricted diffusion on diffusion-weighted images. The lesion was rim-enhancing on contrast study. Pterional craniotomy and sylvian fissure opening were performed with total excision of the lesion. Post-surgery, the patient had diabetes insipidus which eventually resolved.

Abscesses resulting from hematogenous spread can localize in any part of the brain, but most commonly occur in the distribution of the middle cerebral artery at the junction of the gray and white matter of the cerebral hemispheres. In contrast, abscesses derived from contiguous sources tend to be superficial and close to the infected bone or dura.

Visual loss is a common presenting complaint due to the proximity of the lesion to the optic nerves, chiasm and optic tracts. For our patient, this was manifested as bitemporal hemianopsia. Involvement of the cavernous sinus produces symptoms and signs related to affectation of the cranial nerves (3rd, 4th, and 6th, as well as 1st and 2nd divisions of the 5th cranial nerve) that run in the cavernous sinus. Our patient had episodes of

esotropia. Headache develops either as a consequence of increased intracranial pressure, distortion of the diaphragm or irritation of the parasellar dura.¹⁴

Both tuberculoma and abscess do not present with hypersecretory syndromes but rather with hypopituitarism or symptoms of mass effect due to compression of nearby vital surrounding structures, the severity of which depends on the location, size and growth potential of the lesion⁶. Owing to their compressive effects, varying degrees of anterior pituitary dysfunction can develop later with or without central diabetes insipidus. Hypopituitarism with hyperprolactinemia occurs, that causes galactorrhea and amenorrhea in females and decreased libido in males.^{15,16} In the case of our patient, hypothyroidism and hypocortisolism were noted.

Diagnostic Tests to be done in such cases include neuroimaging to confirm the location of the lesion; endocrine work-up and hormone level determination to distinguish functioning from non-functioning masses; tuberculosis work-up since we are in a TB-endemic setting

The role of neuroimaging in this case cannot be overemphasized. Further differentiating tuberculoma from a TB abscess is important because of differences in management. They may be distinguished through MRI where the stages of tuberculoma evolution are better appreciated, except in cases where central liquefaction has taken place, when the two may become indistinguishable from one another. Sellar tuberculoma may occasionally appear hyperintense on T1-weighted images owing to their high protein content. On T2, they can be hypo or hyperintense. With contrast, the tuberculomas characteristically appear as conglomerate ring-enhancing nodules. On the other hand, sellar tubercular abscess on MRI shows cystic sellar lesion with ring enhancement on contrast. On T2, there is a hyperintense signal in the cyst cavity.^{17,18,19}

Whitner has established the following diagnostic criteria for intracranial TB abscess.²⁰ There has to be (1) macroscopic evidence of a true abscess formation within the brain as confirmed during surgery or autopsy; (2) histological proof of presence of inflammatory cells in the abscess wall and (3) demonstration of Acid Fast bacilli in the pus or abscess wall in immunocompromised patients with or without HIV infection or in an immunocompetent patient from an endemic region with a pulmonary focus of infection. The following are the risk factors for the development of tuberculous brain abscess: immunocompromised condition, endemicity, and a pulmonary focus of infection.

Distinguishing between tuberculoma and a TB abscess is crucial since a tuberculoma would usually resolve with anti-TB medications. In fact, some authors recommend starting anti-TB medications in patients suspected to have sellar tuberculoma by imaging even without histologic diagnosis, especially in endemic areas. On the other hand, a TB abscess needs to be aspirated due to the impenetrability of the abscess wall to drugs.

The procedure of choice is the transphenoidal approach as it allows local cure, provides a histopathological diagnosis, and avoids CSF contamination. Radical excision is not necessary. Based on literature, a four-drug regimen consisting of Isoniazid, Rifampicin, Pyrazinamide, and Ethambutol for 18 months has been given to patients with both tuberculoma and TB abscess. However, there is still no definite consensus as to the duration of treatment for this condition.

As in our patient, hormonal replacement should be initiated if indicated. Family members should also be worked up for TB. Lastly, the patient and her family should be educated on the importance of compliance to therapy and follow-up.

CONCLUSION

Tuberculoma in the sellar-suprasellar area, can impinge on the optic chiasm, producing bitemporal hemianopsia and pituitary dysfunction. It presents like other sellar-suprasellar masses with non-specific symptoms and these masses share similar features on cranial CT scan—hypodense lesions with rim-enhancement. Due to the complexity of the disease, treatment of sellar-suprasellar TB requires integrated management of an infectious disease expert, neurologist, neurosurgeon, endocrinologist, and adolescent medicine specialist. Outcomes of four other cases found in literature were generally good after aspiration or drainage of the abscess followed by TB treatment for 15 to 18 months with resulting improvement in vision, marked reduction in the size or complete resolution of the mass, but with one case having loss of pituitary function.

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