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Sellar space-occupying lesion: not always a pituitary tumour!

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ABSTRACT

Background and objective: Hypophysitis is an inflammatory disease of the pituitary gland that is clinically and radiologically similar to pituitary tumours. We are reporting a case of xanthogranulomatous hypophysitis which was confused as a pituitary neoplasm preoperatively.

Materials and methods: A 56-year-old woman presented with extreme tiredness and visual disturbances for 4 months and had visited multiple doctors for the same. NCCT head and CEMRI done preoperatively were suggestive of sellar SOL. The patient was optimised preoperatively and underwent trans-nasal transsphenoidal surgery was performed. Histologic examination of the tissue was s/o xanthogranulomatous hypophisitis

Conclusion: We have described an unusual inflammatory lesion of the pituitary in the sellar region that was mimicking neoplasm. A high level of clinical suspicion and knowledge regarding the differential diagnosis of the sellar region is necessary for correct diagnosis and management.

Key message: All the sellar space occupying lesions are not always pituitary adenomas. Rare entities like xa thogranulomatous hypophisitis should be thought of when we encounter patients with hypocortisolism.

Keywords
sellar SOL, pituitary, xanthogranulomatous hypophisitis

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INTRODUCTION

Pituitary hypophysitis is one of the rare, inflammatory condition that may present both clinically and radiologically similar to a pituitary tumor (1). Clinically, it might present with f/o headaches and/ or visual disturbance due to the local mass effect and compression of the optic chiasm. Inflammatory infiltration can lead to pituitary dysfunction in these patients.

Xanthogranulomatous hypophysitis, is an inflammatory disease showing granulomas with foamy macrophages, multinucleated giantcells, epithelioidcells, and infiltration of lymphocytes (1). Hypophysitis can be histologically divided into the five distinct categories: lymphocytic, granulomatous, xanthomatous, necrotising and xanthogranulomatous hypophysitis (XGH). Xanthogranulomatous

type is a mixture of the xanthomatous and granulomatous subtypes (1,2). Based on their anatomical location and areas of infiltration, they can also be classified as either adenohypophysitis or infundibuloneurohypophysitis (1,2). Most common type of hypophisitis is lymphocytic hypophysitis (1) and XGH is rare type. On histopathological evaluation of Xanthogranulomatous hypophysitis, cholesterol clefts, haemosiderin deposits, multinucleated giant cells, macrophage accumulation and fibrous proliferation can be seen (1,3). Currently the pathogenesis of this condition is not very well described and aetiologically it can either be a primary (most common) or a secondary hypophysitis. Primary hypophysitis is autoimmune in origin. Primary hypophisitis can occur in isolation or as a part of systemic disease such as polyglandular autoimmune syndromes or IgG4 systemic disease (1)(4). Secondary hypophysitis might occur as a consequence of local inflammation caused by lesions including craniopharyngioma (CP)/Rathke's cleft cyst or as a part of systemic diseases such as tuberculosis, sarcoidosis, Wegener's granulomatosis or syphilis (1) (4) (5).

We report here a case of xanthogranulomatous hypophysitis in a 56-yr-old woman, who presented with symptoms mimicking pituitary neoplasms.

CASE REPORT

A 56-yr-old woman came with c/o extreme tiredness and visual disturbances in the form of reduced peripheral vision since 4 months associated with headache and on and off vomiting. There was h/o significant weight loss. Patient had visited a physician with these complaints and underwent a thorough examination by which she was found to have hypothyroidism was started on replacement Thyroxine 25mcg OD and . Patient visual distubances continued to worsen for which she consulted an ophthalmologist who evaluated and advised a MRI orbit which showed an incidental pituitary lesion. On further hormonal work up by an endocrinologist she was found to have hypocortisolism with baseline hormone profile values being as follows: Replacement hydrocortisone of 10mg in the morning and 5 mg in the evening was started with which patient improved significantly with reduced apathy and regained appetite with weight gain.

On further revaluation with CEMRI brain, there was a heterogeneously enancingT2/T1 isointense lesion seen in the sella and suprasellar region, measuring 2.3x1.5x2.0cm. It was causing widening of the sella(Fig 1a) . Anterior pituitary gland couldnot be identified separately from the lesion and pituitary stalk was involved. Superiorly, lesion was extending upto the floor of 3rd ventricle causing displacement and compression of optic chiasm. On right lateral side, lesion extends beyond the lateral tangent line into the superior cavernous sinus compartment, partially encasing the right cavernous ICA(KNOSP grade 3A). On left side, it is extending between intercarotid line and lateral tangent (KNOSP grade 2) (Fig 1b). The minimum diameter between the cavernous segment of both ICA was 14mm

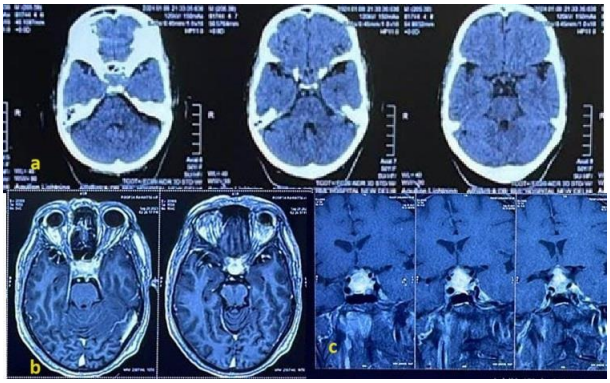

Figure 1. (**a**) NCCT head showing isodense sellar space occupying lesion. (**b**) MRI axial image of Isointensense sellar lesion. (**c**) MRI coronal image of Knosp grade 3A on the right and grade 2 on the left.

Hormone profile was done and showed results as mentioned in the Table 1.

Table 1. Pre operative hormone profile reports

Hormone	IGF-1(ng/ml)	Prolactin (ng/ml)	fT3 (pg/ml)	fT4 (ng/ml)	TSH (mcUI/ml)	ACTH (pg/ml)	Cortisol (mcg/dl)
Levels (N)	44-210	1.8-20.30	2.30-4.20	0.89-1.76	0.55-4.76	<46	2.5-25
Patient Report	35.30	3.40	1.52	0.33	2.15	27.20	0.75

Preoperatively patient was started on Tab. Hisone 10mg in the morning and 5mg in the night and Tab. Thyroxinbe 25mcg OD daily and was optimized and was taken up for surgery

Patient underwent Transsphenoidal tumor decompression and intraoperatively there was a fibrotic firm tumor which was moderately vascular . Near total tumor decompression was done and

tissue was fixed immediately in formalin and sent for histopathological examination. And on Histopathological examination , there was Rathke's cleft in the pituitary tissue (Fig. 2b) highlighted by synaptophysin on IHC surrounded and infiltrated by foamy histiocytic collection (xanthoma cells) and along with both Langerhans type 2 foreign body giant cells surrounded by lymphocytes with no caseous necrosis seen (Fig. 2a).

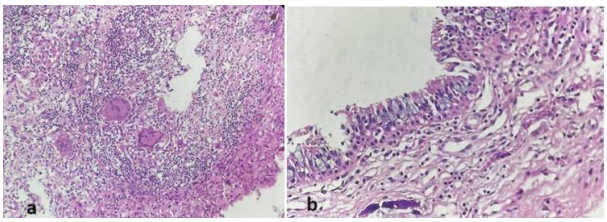

Figure 2. (**a**) Histological features showing xanthogranulomatous lesion (10x). (**b**) Simple cyst lined by single layer of columnar cell supported by fibrocollagenous stroma (40x).

Postoperatively patient developed diabetes insipidus which was managed with the help of endocrinologist. Rest of the postoperative period was uneventful. Replacement steroid and thyroxine is being continued in the postoperative period and the post op hormone profile was as mentioned below (Table 2).

Table 2. Postoperative hormone profile

Hormone	IGF - 1 (ng/ml)	Prolactin (ng/ml)	fT3 (pg/ml)	fT4 (ng/ml)	TSH (mcUI /ml)	ACTH (pg/ ml)	Cortisol (mcg/dl)
Levels (N)	44-210	1.8-20.30	2.30-4.20	0.89-1.76	0.55-4.76	<46	2.5=25
Patient Report	75.4	2.14	1.92	0.49	0.41	3.2	18.5

Discussion

Hypophysitis can be due to number of inflammatory processes that are different in etiology, pathogenesis, and morphology [8]. Idiopathic or primary inflammatory lesions of the hypophysis include lymphocytic hypophysitis, granulomatous hypophysitis, xanthomatous hypophysitis, xanthogranulomatous hypophysitis, and necrotizing hypophysitis [2,4,7].

Granulomatous hypophysitis is histologically characterized by aggregates of multinucleated giant cells, macrophages, and extensive plasma cell infiltration [3,6]. The histologic findings consisted of foamy histiocytic (xanthomatous) infiltration without evidence of associated adenomas, cyst, hemorrhagic infarct, granulomas, Langerhans cells, neutrophilic exudates, or Michaelis– Gutmann bodies.

In our case, the MRI findings showed homogenously isointense lesion in the sellar and suprasellar region causing widening of sella from which anterior pituitary couldn't be visualised separately.

And the histopathological specimen in our case included Rathke's tissue highlighted by synaptophysin on IHC and infiltrated by foamy histiocyte collection (xanthoma cells) and long with both Langhan's giant cells and foreign body type giant cells surrounded by lymphocytes.

The pathological mechanisms underpinning autoimmune pituitary disorders merit some discussion. The present patient did not have a history of autoimmune disorders, but her sex and age may suggest the progression of LH to XGH by the time of presentation. However, infection or systemic disease may also have been involved in the etiology. Systemic conditions such as tuberculosis, sarcoidosis, and other granulomatous diseases were excluded as etiologic factors in the present patients.

Surgical resection is typically the only treatment that is required. However, anti-inflammatory and immunosuppressive drugs may be indicated in patients with residual or recurrent hypophysitis

The overall prognosis for XH is good, but improvement of pituitary function after transsphenoidal surgery has been reported in less than 50% of the cases in the literature. Chronic inflammation may result in destruction and fibrosis of the pituitary gland (6)

In summary, we described an unusual inflammatory lesion of the pituitary mimicking a neoplasm in the sellar region. A high level of clinical suspicion of the inflammatory disorder is necessary to provide the correct diagnosis and choose optimal management. (8)

Conclusion

In the present report, we have described a patient with a rare pituitary pathology who presented with hypopituitarism and visual impairment and was misdiagnosed as having a macroadenoma. Postoperative histological evaluation revealed an XGH of a remodeled RPC. Following surgical treatment, the patient had persistent pituitary

dysfunction. Surgical intervention early in the development of such lesions may have beneficial effects on pituitary function, because chronic inflammation leads to the destruction of the pituitary gland and permanent pituitary dysfunction

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