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Case Study

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CENTRAL DIABETES INSIPIDUS AS PRESENTING MANIFESTATION OF SUPRACELLAR EPIDERMOID CYST: A CASE REPORT AND LITERATURE REVIEW

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ABSTRACT

Epidermoid cysts (ECs) are rare intracranial brain tumors. They compromise approximately 1% of intracranial tumors They are slowly growing benign tumors, clinically present with head ache, visual symptoms and anterior pituitary hormonal deficiencies due to mass effect. Polyuria due to central diabetes insipidus(DI) as a presenting clinical manifestation is rare .Only 2 cases was reported in the literature. Here we present a 49 year male presenting with polyuria and visual disturbance. The investigations confirmed presence of Central DI and the MRI brain showed cystic supracellular lesion. He underwent trans sphenoidal surgery and the histopathology confirmed epidermoid cyst. He did well post operatively but the DI persisted after surgery. The epidermoid cysts of sellar region vary in their in presentation depending upon their location and extension into surrounding areas producing mass effect. DI is rare presentation of these rare tumors.

KEYWORDS: Supracellar tumor. Epidermoid cyst, Central diabetes insipidus.

INTRODUCTION

Epidermoid cysts(ECs) result from the inclusion of squamous epithelial elements during neural tube closure. ECs are tumors constituting 02-1.8% of all brain tumors. ECs are typically found in cerebello-pontine angle, but occasionally develop in sellar region. ECs are usually clinically silent, but may produce signs of mass effect as headaches, visual field defects. ECs presenting with Central Diabetes insipidus(DI) is reported but rare. Only two cases were reported in literature(Ref:1).Here we report a case of sellar Epidermoid cyst presenting with DI.

Case Presentation

49 year male presented with one month history of polyuria, polydipsia and weight loss and visual disturbance. The clinical examination showed blood pressure 132/85 mm/Hg with pulse rate of 80/ minute with regular sinus rhythm, normal examination of respiratory, cardiovascular and gastrointestinal systems. His neurological examination was normal apart from bi-temporal hemianopia. The initial work up identified normal blood sugars, serum calcium and renal function. The other biochemical and hormonal values are shown in Table1 The water deprivation test confirmed the diagnosis of DI(Table no:2). Patient had panhypopituitarism and was started on appropriate hormonal treatment. MRI pituitary showed sellar and supra sellar mass(Fig no:1).

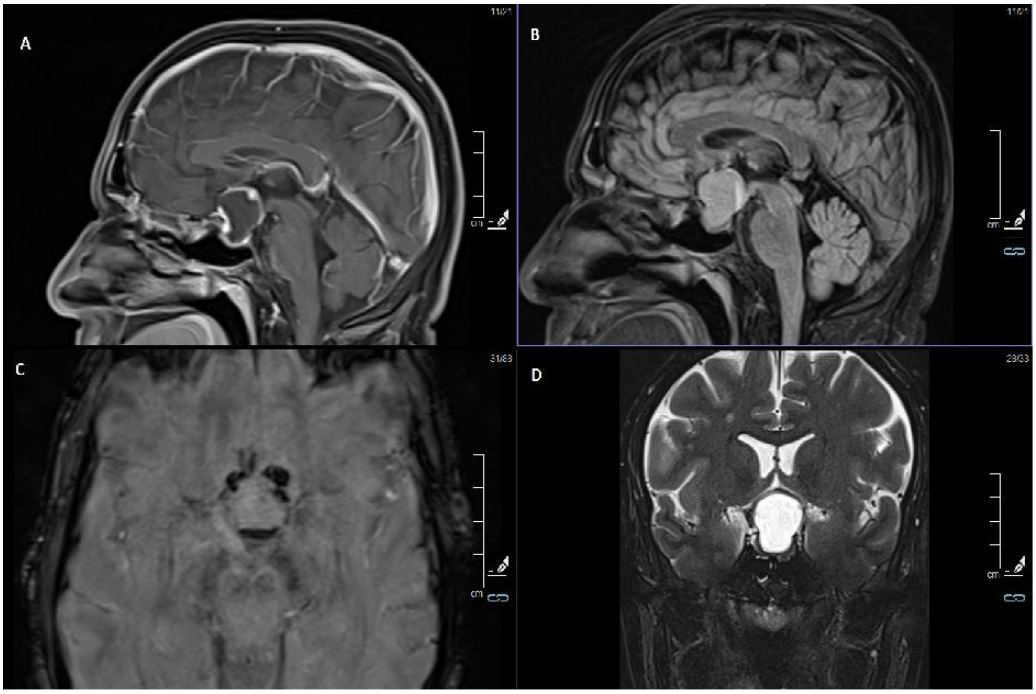
Trans-sphinoidal surgery was performed to remove the tumor. The histo- pathological report confirmed the tumor as epidermoid cyst.

Table 1: DI along with pan-hypopituitarism persisted post operatively and managed with appropriate management.

Parameter	On presentation	Normal Ranges
TSH	< 0.0083	0.4-4 microUI/ml
Free T4	9.47	9-19 pmol/l
Prolactin	854	73-407 nmol/l
FSH	0.81	1.2-9 U/L
LH	<0.12	2-20U/L
Morning Plasma cortisol	<27	138-635 nmol/l
Water deprivation test	Confirmed the diagnosis of Diabetes insipidus (Table:2)	

Table 2: Water deprivation test results.

Time	Urine osmolality mOsm/kg H2O	Serum osmolality mOsm/kg H2O	Urine output ml/hr
Base line	234	298	120
1 hour	269	305	100
2 hour	325	310	80
Post Vasopressin			
1 hour	573	297	10
2 hour	564	290	15



MRI brain shows sella and suprasella mass.
A image sagittal T1 WI C+ (gd) shows thin enhancement around the periphery .
B image sagittal FLAIR shows bright ssignal higher than CSF with layering.
C image axial SWI shows blooming artifact of calcfications and dependent pat hemorrhage.
D image coronal T2 WI shows bright signal mass isointense to the CSF.

Fig. 1: MRI Pitutary.

DISCUSSION

Epidermoid cysts (ECs) are rare. They comprise 1%-2% of all intracranial tumors.[1] They can occur in and around the region of the pituitary gland and supracellar cisterns. They are basically embryonic rests of skin cells and make all sort of skin related gum that can collect

within them. The differential diagnosis for these tumors include ECs, Craniopharyngiomas, and Ratkys cleft cysts.[2] In view of their common embryological origin of the Hypothalamic- pituitary axis, the pathology of theses lesions are similar and overlap. The ECS are filled with keratin debris, lipid and water and appear hypodense on CT scan.[3,4] The slow growth of these tumors often results in them remaining asymptomatic, until their size is large enough to compress surrounding structures.[5] With regard to clinical presentation the ECs remain silent in 15-20% of cases, while most ECs present with head ache and visual symptoms as their linear growth expands. The head ache associated with ECs has not demonstrated correlation with the size of the tumor.[6] the most common symptoms are head ache and visual disturbance. Anterior pituitary endocrinal dysfunction was uncommon and reported in only 4 cases.[7,8,9] ECs affecting the posterior pituitary and presenting with polyuria and polydipsia due central Diabetes insipidus(DI) is extremely rare and only two cases reported in the review of 29 published cases.[10,11] Our patient presented with polyuria and plolydipsia and the water deprivation test performed confirmed the diagnosis of DI(Table:2).Surgical resection is the standard of treatment for ECs with gross total resection including the cyst and necessary to prevent recurrence.[12] Our patient underwent Transhenoidal surgery and the cyst was removed partially. Of note Samii et al observed that only 50-80% of patients undergo surgical resection for intracranial ECs obtain complete removal. The complication that can occur with subtotal resection are severe with 40% of the patients acquiring chemical meningitis due spillage of the cyst contents[14] Our patient did not develop any post operative complication and had uneventful recovery. The DI persisted after the surgery and patient was started on desmopressin.

CONCLUSION

ECs of sellar region vary in clinical presentation depending upon their location, and extension into surrounding areas producing mass effects. DI is a rare presentation of these rare tumors

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