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CASE REPORT VI

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TSHoma:A Rare cause of Pitutary Adenoma

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Abstract

Background: The TSH-producing adenoma (TSHoma) prevalence in the general population is 1 to 2 cases per million, accounting for less than 2% of all pituitary adenomas. It was in 1960s where the first case of TSHoma was reported with the inappropriate secretion of TSH, could only be recognized after the introduction of the TSH RIAs. The recent development of ultrasensitive TSH assays facilitates earlier diagnosis by detecting TSH in the presence of elevated free thyroid hormones, thus ruling out primary hyperthyroidism, primarily Graves' disease. Failure to detect the presence of TSHoma may result in dramatic consequences. This study is a review of our experience in the management of TSHoma at National academy of Medical sciences, Endocrine Unit. Here we report a 36 years old male presented with weight loss, increased frequency in stool ,hand tremors and loss of libido, who was found to have TShoma in further investigation and was managed.

Key words: Pituitary adenoma, TSHoma, Hyperthyroidism

Introduction

Case presentation

A 36 years old boy was referred to our center for further management of TShoma from pokhara superspeciality Health clinic. He presented with weight loss of 20 kg in 1 year, with both hand tremors, increased frequency of stool and loss of libido since 4-6 months. He also gave history of frequent clear water like secretions from nose and have visited local pharmacy for the same, which was treated as allergic rhinitis. Suspecting thyrotoxicosis on further investigation was found to have Raised TSH, Free T3 and Free T4 as TSH-10.20(0.31-4.31).FT3-5.75(2.1-3.8) FT4-2.52(0.8-1.63). Further Hormonal evaluation with Dynamic MRI study of Pituitary gland was done. Which revels 25*30*42 mm lobulated lesion is seen expanding the sella with suprasellar extension, with mass effect on optic chaisma,suggesting of Pituitary macroadenoma (Knosp grade II on both sides). Fig 1

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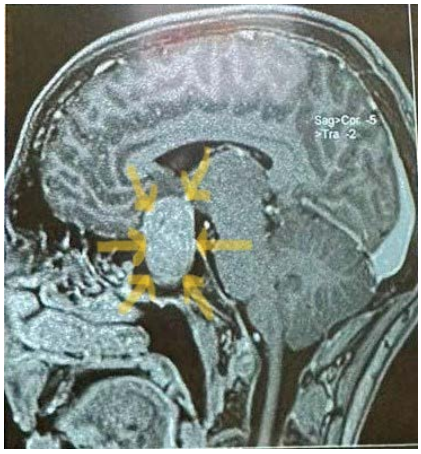
Fig 1-Pitutary mass before surgery



He was then referred to Neurosurgery department for surgical management of TSHoma. Endoscopic Endonasal trans-sphenoidal pituitary adenomectomy was done. He was discharged with Cortilone 10mg at 8am and 5mg at 4pm,Tablet Thyroxine 50mcg once daily in empty stomach.Histopathology report shows-sellar mass.Biopsy-Pitutary adenoma with no Necrosis and mitotic figures. Immuno histopathology cytology examination shows-pituitary adenoma with tumor cell express on Focal TSh and negative for Prolactin, Growth hormone, Adrenocorticotrophic hormone, Leutinising Hormone,
Follicular stimulating hormone. Postoperatively, normalization of thyroid hormones was achieved with no deficiency of other pituitary hormones. The patient was discharged from the hospital on the 12th postoperative day. A followup MRI was performed 3 months after surgery (Fig 2) ; no regrowth of the tumor was noted, and the patient's thyroid function tests were within normal limits.

Table-1 : Various results before and after surgery is given below in table.

	Test	Results	unit	Ref range
Before surgery	TSH	10.2	µIU/l	0.38-4.31
	Free T3	5.75	µIU/l	2.1-3.8
	Free T4	2.52	µIU/l	0.8-1.63
	Serum cortisol	2.89	µIU/dl	6.4-21
	serum prolactin	28.9	ng/dl	3.6-16.3
	serum testosterone	230.7	ng/dl	262-870
Post Surgery	TSH	1.09	µIU/l	0.46-4.68
	FT3	2.24	pg/ml	2.77-5.27
	FT4	1.35	ng/dl	0.78-2.19
	serum cortisol	2.23	µg/dl	4.46-22.7
	serum testosterone	388.53	ng/dl	262-870
Recent reports	TSH	1.4	µIU/l	0.38-4.31
	FT3	2.4	pg/ml	2.1-3.8
	FT4	0.94	ng/dl	0.8-1.63
	serum cortisol	1.3	µg/dl	4.46-22.7

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Among all functional pituitary adenomas, thyroid-stimulating hormone (TSH)-secreting pituitary adenomas (TSHomas) are rare and account for only 0.5%–3% of all pituitary tumors [1, 2]. In this situation, TSH secretion is autonomous and refractory to the negative feedback of thyroid hormones (inappropriate TSH secretion) and TSH itself is responsible for the hyperstimulation of the thyroid gland and the consequent hypersecretion of T4 and T3 [1, 8]. They occur in all age group and exhibit no gender predilection[2] The presence of a TSH-oma has been reported at ages ranging from 8 to 84 years [8,9], the mean age at diagnosis being 46 ± 6 years [9].TSHomas are defined as serum hormonal levels suggestive of central hypothyroidism as in our case where we found raised TSH, raised FT3 and raised FT4,low level serum cortisol,raised prolactin,low testosterone which can present with clinical features of Hyperthyroidism.[3,4] . The diagnosis of TSHomas is challenging. A correct and early diagnosis of TSHomas is essential to avoid misdiagnosis, and thereby, proper treatment could be taken immediately [10]. Signs and symptoms of hyperthyroidism along with values of thyroid function tests similar to those found in TSH-oma may be recorded also among patients affected with resistance to thyroid hormones [10-11]. This form of resistance to thyroid hormones is called pituitary resistance to thyroid hormones (PRTH), as the resistance to thyroid hormone action appears more severe at the pituitary than at the peripheral tissue level. The clinical importance of these rare entities is based on the diagnostic and therapeutic challenges they present.

Failure to recognize these different diseases may result in dramatic consequences, such as improper thyroid ablation in patients with central hyperthyroidism or unnecessary pituitary surgery in those with PRTH

As was the case with our patient, TSHomas are often reported as macroadenomas after a delayed period, because the clinical features are varied and it often takes a long time to be diagnosed [6.7]. Patients with TSH-omas present with signs and symptoms of hyperthyroidism that are frequently associated with those related to the pressure effects of the pituitary adenomas, causing loss of vision, visual field defects, headache, and/or loss of anterior pituitary function [12,13] The enlarged mass seen in the MRI may cause visual field defects (40%), headaches (20%), and mild or severe loss of pituitary functions. Partial hypogonadism occurs in about 1/3 of patients; it is mainly identified through menstrual irregularities in females and through central hypogonadism, and decreased libido in males[6] as we see in our patient too. Surgical resection is considered the first-line therapy for TSHomas [1,5], aiming at removing tumor mass and normalizing thyroid function.

Conclusion:

TSHoma is a rare type of functional pituitary adenoma and often presents with hyperthyroidism. A correct and early diagnosis of TSHomas is challenging.

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