A case of plurihormonal TSHoma presenting as meningitis

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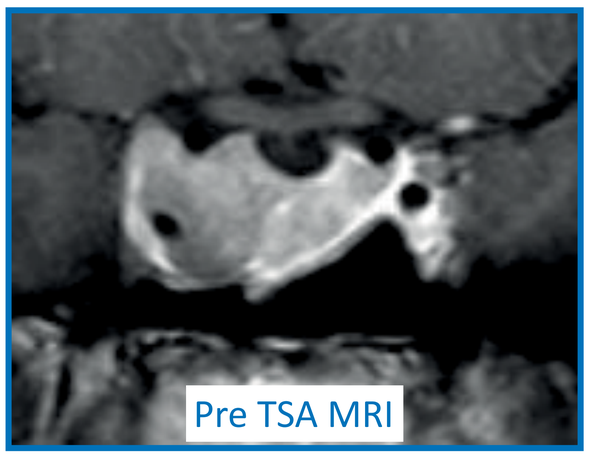
Introduction

TSHomas are rare ,accounting for 0.5-3% of pituitary tumours [1,2]. 70-80% secrete only TSH, while 20-25% co-secrete predominantly prolactin and GH [1,3]. Almost all express somatostatin receptors , hence somatostatin analogues are routinely used as 2nd line treatment after surgery [4]. Post operative cure is expected in more than half of macroadenomas & almost all microadenomas [6]. We describe a recent interesting case of plurihormonal TSHoma from Oxford.

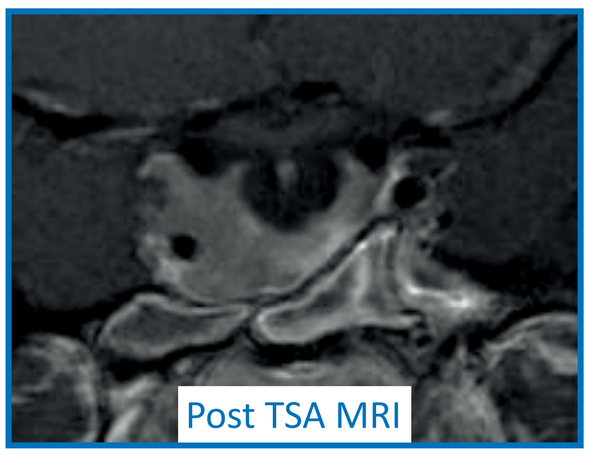
Presentation

A 22 year old lady with previous gestational diabetes presented with Haemophilus Influenza meningitis. MRI revealed an incidental pituitary macroadenoma extending into the right cavernous sinus and breaching the anteroinferior wall of the pituitary fossa with CSF leak. Clinically, she was mildly thyrotoxic but not obviously acromegalic or cushingoid.
Examination and visual fields were normal. Thyroid function showed raised T4 [24.7 pmol/L (normal range 9-19 pmol/L)] and T3 [8.3 pmol/L (normal range 2.6–5.7 pmol/L)] with unsuppressed TSH [1.75 munit/L (normal range 0.3-4.2 munit/L)]. IGF-1 was also raised at 56.7 nmol/L (normal 12-50.1 nmol/L). OGTT (nadir GH 0.88 μg/L) and GH day curve (mean GH 7.2 μg/L) confirmed acromegaly along with possible TSH co-secretion.



Treatment

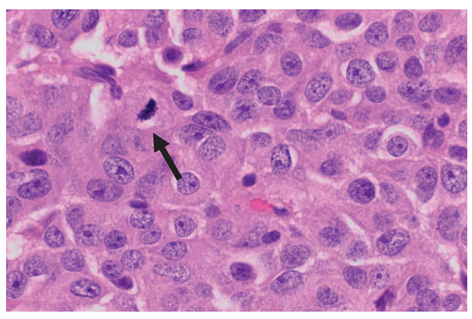
Pending trans sphenoidal surgery, Lanreotide was started which normalised thyroid function and IGF-1. However after 2 months treatment she developed a CSF leak and a second episode of meningitis.
After treatment and resolution of meningitis she had trans sphenoidal adenomectomy. A patch repair was fashioned to prevent further CSF leak on SSA treatment. Histology showed a plurihormonal atypical tumour with 30% expression for GH, 1% expression for TSH and Prolactin and MIB-1 index of 5-10%. After surgery, T3, T4 and TSH remained in normal range at 5.1 pmol/L, 14.7 pmol/L and 1.68 munit/L respectively, however, IGF-1 level started to rise above the normal range at 56.4 nmol/L. Hence she has been started on Lanreotide Autogel 120mg SC four weekly. This has lead to normalisation of IGF-1.

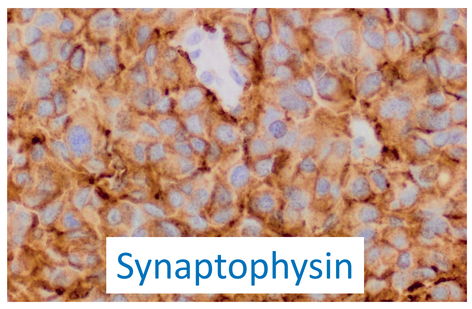


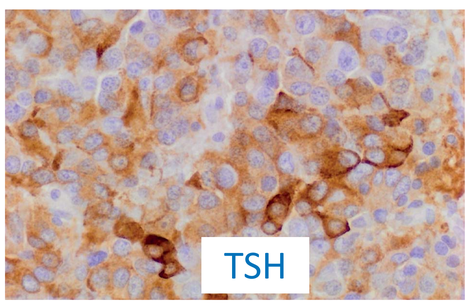
Biochemistry

	Pre TSA	6 weeks Post TSA	12 weeks post TSA	On SSA
FT3 pmol/L	8.3	5.1	5.3	1.9
FT4 pmol/L	23.4	14.7	13.5	10.7
TSH mU/L	1.1	1.68	1.07	0.57
IGF-1 nmol/L (12-50.1)	56.7	44.4	56.4	29.9

Histology







Histology demonstrated a neuroendocrine neoplasm with mitoses (arrow; usually absent in pituitary adenomas), diffuse synaptophysin expression (indicating presence of neurosecretory granules) and strong TSH expression.

Conclusion & clinical message

This interesting case of TSHoma demonstrates an atypical presentation. It is noteworthy that this patient also developed CSF leak after pre-op somatostatin analogues which was probably due to tumour shrinkage. Histology confirmed the plurihormonal nature of the tumour as suspected from initial biochemistry. Clinically she was mildly thyrotoxic and did not have any acromegalic features, however histology showed 30% GH expression compared to only 1% TSH.
Due to cavernous sinus extension, cure was not achieved after surgery and second line treatment with somatostatin analogues is being used to maintain biochemical control and hopefully prevent further growth. Accurate diagnosis of TSHoma is important & inappropriate treatments such as thyroid ablation & surgery in cases of misdiagnosis are not uncommon [5,6]. Thyroid function tests should be analysed carefully and diagnosis should be questioned when response to treatment is not as predicted. A high index of suspicion should be maintained & any instances of “inappropriate TSH” should be carefully tested further to rule out TSHomas [6,7].

References

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BES2017 EP-067

Neuroendocrinology and pituitary

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Poster presented at: SfE BES 2017 6-8 November HARROGATE

PosterSessionOnline.com