A clinically functioning gonadotroph adenoma presenting with abdominal pain, bilateral multi-cystic ovaries & fibromatosis

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Introduction

- We present the case of a clinically functioning gonadotroph adenoma in a pre-menopausal woman presenting with abdominal pain, multiple large ovarian cysts and fibromatosis
- To our knowledge, this is the first case of fibromatosis associated with a functioning gonadotroph adenoma

Case

- 35y female
- Three emergency admissions with abdominal pain:
  - 1st – bilateral cystectomy for large benign follicular cysts
  - 2nd – right oophorectomy and salpingectomy for ovarian torsion and left ovarian cyst aspiration
  - 3rd – resection of a 4 x 1.7cm rectus abdominis muscle mass. Histology confirmed fibromatosis (desmoid tumour)

Endocrine clinic review

- Persistent abdominal pain
- Irregular periods
- No galactorrhoea
- No headaches
- Normal examination

Investigations

- Pelvic ultrasound images are shown in figures 1 and 2
- Endocrine investigations are shown in table 1
- Pituitary MRI images are shown in figures 3 and 4

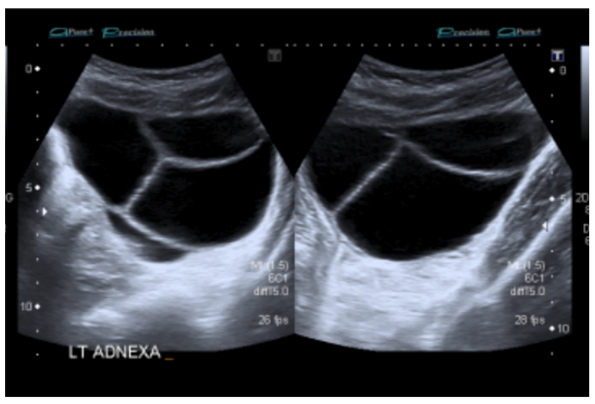

Figure 1

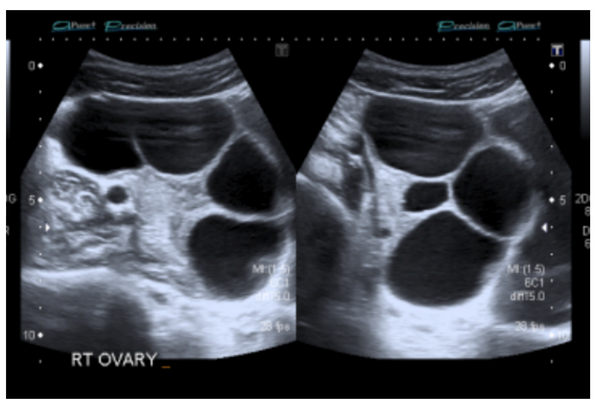

Figure 2

Figures 1&2: Pelvic ultrasound demonstrating large, bilateral ovarian cysts
Figure 3: Pre-operative MRI pituitary showing a 1.5cm, right sided pituitary mass
Figure 4: Post-operative MRI pituitary showing resection of the gonadotroph adenoma

	Pre-operative		Post-operative
	12/5/2017	26/5/2017	27/11/2017
Oestradiol (pmol/L) FP <571 LP 122-1094	2096	1111	< 100
FSH (IU/L) FP & LP 1-9	8.7	9.0	3.2
LH (IU/L) FP 3.2-8.0 2.4-7.2	0.8	0.7	1.8
Prolactin (mIU/L) <700	740		191
Testosterone (nmol/L) 0.2-1.7	< 0.3		
Free T4 (pmol/L) 12-22	19.9		21.5
TSH (mIU/L) 0.27-4.2	1.07		0.60
Cortisol (nmol/L)	396		428
IGF1 (nmol/L) 7.4-31.3	17		

Table 1: Endocrine investigations (FP = follicular phase; LP = luteal phase)

Diagnosis & management

- A diagnosis of an FSH secreting pituitary adenoma was made
- The patient underwent transphenoidal hypophysectomy
- Histology confirmed a pituitary adenoma with FSH immunopositivity in keeping with gonadotroph cell adenoma

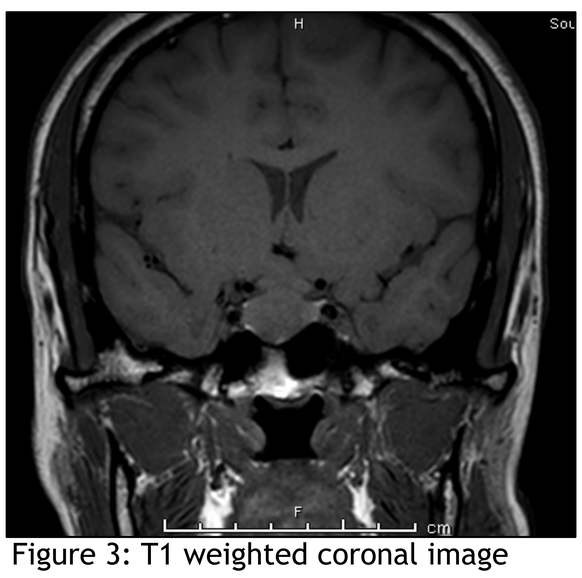

Figure 3: T1 weighted coronal image

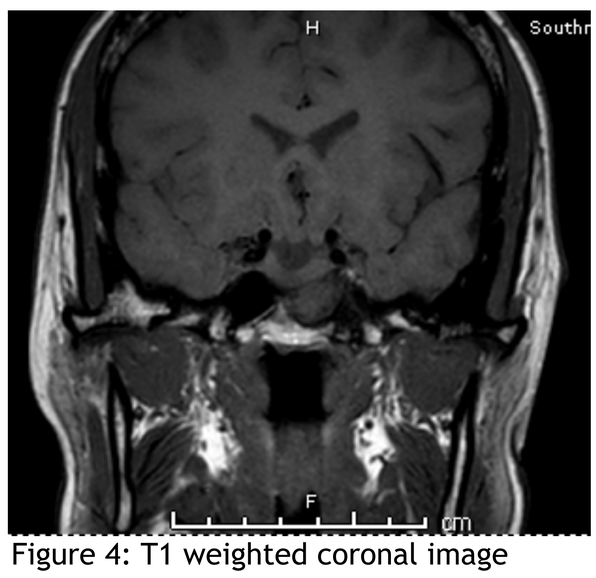

Figure 4: T1 weighted coronal image

Post-operatively

- Abdominal pain resolved and normal menstrual cycle returned
- Oestradiol, FSH and LH levels normalised
- Pelvic ultrasound showed two normal follicles, 2-3cm in size
- MRI pituitary 3 months post-operatively, showed removal of the pituitary tumour

Discussion

- Gonadotroph adenomas are usually clinically non-functioning, but in rare cases can cause symptoms secondary to hormone hypersecretion. More commonly, gonadotroph adenomas present with symptoms secondary to mass effect or are discovered incidentally.
- The prevalence of clinically functioning gonadotroph adenomas is not known, but there are only about 30 reported cases in the literature(1).
- The most common symptoms include menstrual irregularity, spontaneous vaginal bleeding and infertility. Ovarian stimulation is usually mild, but cases of multiple ovarian cysts leading to ovarian torsion have been reported(2).
- Fibromatosis (desmoid tumours) are rare tumours that are locally aggressive but do not have metastatic potential(3). They have been associated with high levels of oestrogen, for example in women during or following pregnancy. However, evidence for this association is limited to retrospective cases(4).
- The main biochemical finding is elevated oestradiol levels, which can be mild to extremely elevated(5). FSH levels can be normal to mildly elevated. LH levels are usually suppressed.
- Pituitary MRI reveals a macroadenoma in the majority of cases(5).
- Transphenoidal surgery is recommended as first line treatment for clinically functioning gonadotroph adenomas.
- Medical treatments have been used in individual cases with inconsistent results(5).
- There is limited data on long term outcomes, however long term follow up is required due to risk of recurrence.

References

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