An Unusual Presentation of an Ovarian Teratoma

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INTRODUCTION

Hormone-secreting teratomas are well described. However teratomas secreting pancreatic hormones are rare, with even fewer cases producing clinically significant effects. We describe possibly the first documented case of hyperinsulinaemic hypoglycaemia due to an insulin-secreting ovarian teratoma

INVESTIGATIONS

Investigation	Result
Cortisol	624 nmol/L
Free T_4	18.2 pmol/L
TSH	1.4 mU/L
FSH	3.1 mIU/ml
LH	3.0 mIU/ml
Oestradiol	480 pg/ml
Prolactin	144 ng/ml
IFG-1	14 nmol/L (age adjusted reference range 13-50)

Trans abdominal US: 10 x 7cm right sided pelvic mass containing fat, soft tissue and calcification

CT CAP: Right 10cm intact cystic structure. Arising from right ovary, most likely teratoma. Left ovary normal. Rest of pelvis and abdomen normal. Normal pancreas and adrenals

CASE PRESENTATION

A twenty three year old woman with no past medical history presented with transient symptoms of lethargy, weakness and mild abdominal discomfort over a period of six months. The symptoms could occur at any time of the day and were progressively worsening in severity. She had used her father's capillary glucose meter to measure her own blood glucose, recording values of 2.1 mmol/L and 2.8 mmol/L.

Her clinical examination was unremarkable other than a right lower abdominal fullness and tenderness. Further investigation confirmed a fasting glucose of 2.7 mmol/L, elevated insulin (40.4 mIU/L) and C-peptide (5.6 ng/ml), with normal IGF-2/IGF-1 ratio (<10).

Abdominal ultrasound and CT scans revealed a 10 cm x 7cm intact cystic structure arising from right ovary, with normal appearances of the pancreas and adrenal glands.

The ovarian mass was removed laparoscopically. Histopathology findings were of a mature cystic teratoma containing teeth, hair, skin, sebaceous material, cartilage and a large, mature, pancreatic tissue component. Within this, immunohistochemistry showed differential expression of insulin, glucagon and somatostatin in islet cells.

Hypoglycaemia did not recur post-operatively. The resolution of symptoms following surgical excision suggests that the patient's hyperinsulinaemic hypoglycaemia was due to ectopic insulin secretion from apparently mature pancreatic tissue within the teratoma

DISCUSSION

There are few case reports of pancreatic hormone secreting teratomas. A literature search using PubMed revealed several cases of mediastinal teratomas secreting varying amounts of somatostatin, insulin and glucagon but without evidence of pre-operative hypoglycaemia (1,2,3).

There was one case of asymptomatic hyperinsulinaemic hypoglycaemia in a five year old male secondary to a mediastinal teratoma with fetal pancreatic features (4). The authors of this case felt that persistent insulin secretion at low glucose concentrations provided a potential mechanism for the hypoglycemia. In a second similar case report, a fifty four year old lady was found to have symptomatic hypoglycaemia secondary to an ovarian teratoma but in this case the insulin concentration in the peripheral blood was low whilst the hypoglycaemia was felt to be secondary to high somatostatin secretion from the teratoma (5). However there were no case reports of hyperinsulinaemic hypoglycaemia secondary to an insulin-secreting ovarian teratoma.

CONCLUSIONS

Ectopic hormone release from teratomas is well described, including AFP from yolk sac-containing teratomas, hCG from pineal teratomas, and, most notably, thyroid hormones from cystic struma ovarii, which may rarely cause thyrotoxicosis. Nevertheless there are few cases of pancreatic hormone-secreting teratomas in the literature, with only a handful causing clinically significant effects. We could not find any other cases of hypoglycaemia secondary to an insulin-secreting ovarian teratoma. In summary, this unusual case demonstrates failure of normal glucose homeostasis secondary to ectopic insulin secretion from an ovarian teratoma, resulting in recurrent hypoglycaemia.

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