Pituitary function tests in black patients with pseudocyesis

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Summary

Pituitary function was evaluated in a group of 10 patients with pseudocyesis. One patient was postmenopausal; the remainder demonstrated normal basal prolactin, luteinising hormone (LH) and follicle-stimulating hormone (FSH) levels and also normal pituitary-adrenal, pituitary-thyroid axes. Oestradiol deficiency was present in 6 patients, while 2 patients demonstrated elevated serum progesterone values, suggestive of a luteal phase. Gonadotrophin-releasing hormone administration resulted in exaggerated stimulation of LH and FSH in 4 and 2 patients, respectively. Impaired pituitary-adrenal function was present in 6 patients after insulin-induced hypoglycaemia and L-dopa administration. GH impairment is probably a consequence of the oestrogen deficiency that commonly occurs in this condition. It thus appears that there are aberrations in specific pituitary hormone responses after provocation in pseudocyesis.

The aetiology of pseudocyesis, or phantom pregnancy, is as yet unknown, despite the recognition of its existence since 300 BC (by Hippocrates). While psychological stress may play a major role in this condition, altered hypothalamic-pituitary axis function unrelated to this stress has also been incriminated. Most of the work on the endocrine manifestations of this disorder has been limited by lack of sufficient cases, probably as a result of the steady decline of the number of subjects diagnosed in the Western world over the past 50 years. This decline has been attributed to the increasing diagnostic accuracy of conditions such as hyperprolactinaemia and pregnancy, and also to the changing status of women in the community. In southern Africa, however, this condition appears to be common among black women as a result of a strong cultural background where fertility is often a prerequisite for marriage or for a stable relationship. A study was undertaken in order to analyse pituitary function in black patients with a diagnosis of pseudocyesis in an attempt to determine if there was any aberration in this function.

Patients and methods

Ten non-pregnant black patients who attended the antenatal and gynaecological clinics of King Edward VIII Hospital, Durban, over a 7-month period, and who satisfied the following criteria were entered into the study: (i) amenorrhoea for a period > 3 months; (ii) abdominal distension; and (iii) a firm belief that they were pregnant. In every patient pregnancy was excluded by the demonstration of undetectable serum human chorionic gonadotrophin (hCG) levels and normal pelvic ultrasonography.

History-taking and a complete clinical examination were performed with particular attention to social background, onset of fetal movements, presence of breast secretions, abdominal girth > 1 cm below the umbilicus, and cervical softening. Plain abdominal radiography was also carried out. The following pituitary function tests were performed on different days after an overnight fast: thyrotrophin-releasing hormone (TRH) (200 mg intravenously), gonadotrophin-releasing hormone (GNRH) (100 μg intravenously); insulin-induced hypoglycaemia (0.1 J/L/kg body weight); and L-dopa (500 mg orally). Venous blood was withdrawn at timed intervals (usually 0, 20, 30, 45, 60 and 90 minutes) via an indwelling catheter inserted into the antecubital fossa. The protocols of the various tests and assay methods have been described previously. The hormonal responses were compared with reference values established in this laboratory by testing normal premenopausal volunteers during the follicular phase of their cycles.

Statistical data. All results are expressed as mean and standard error of the mean.

Results

The 10 patients were aged 24.9 ± 2.2 years; 2 were widowed and 8 unmarried. "Lobola" (dowry) was being paid off for 7 patients and, although a pregnancy was desirable, these patients denied that fertility was a prerequisite for marriage. Parity was as follows: nulliparous (4 patients); para 1 (3), para 2 (2) and para 4 (1). Four patients were employed as domestic servants, 2 were state registered staff nurses, and 4 were unemployed.

Abdominal radiography and pelvic ultrasonography demonstrated no abnormalities. Galactorrhoea was present in 8 of the 10 patients and cervical softening was assessed to be a feature in only 1 case. Table I shows the period of amenorrhoea, onset of fetal movements and abdominal distension and girth at the time of examination.

**TABLE 1. CLINICAL FEATURES OF PSEUDOCYESIS PATIENTS**

<table>
<thead>
<tr>
<th>Features</th>
<th>Mean ± SEM</th>
<th>Range</th>
</tr>
</thead>
<tbody>
<tr>
<td>Amenorrhoea (mo.)</td>
<td>8 ± 1.1</td>
<td>3 - 12</td>
</tr>
<tr>
<td>Onset of fetal movements* (mo.)</td>
<td>7.3 ± 1.0</td>
<td>3 - 12</td>
</tr>
<tr>
<td>Onset of abdominal distension (mo.)</td>
<td>7.3 ± 1.2</td>
<td>4 - 12</td>
</tr>
<tr>
<td>Height (cm)</td>
<td>160 ± 2.0</td>
<td>152 - 168</td>
</tr>
<tr>
<td>Weight (kg)</td>
<td>81.2 ± 3.9</td>
<td>68 - 86</td>
</tr>
<tr>
<td>Abdominal girth (cm)</td>
<td>100.4 ± 2.8</td>
<td>88 - 111</td>
</tr>
</tbody>
</table>

*Onset of fetal movements and abdominal distension was measured from the cessation of menses.

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Hormonal profile

One patient was postmenopausal, as evidenced by elevated gonadotrophins, and was excluded from the statistics relating to hormonal studies. Tables II, III and IV summarise the basal and peak hormone responses of the respective releasing hormones.

Prolactin levels measured daily on 4 occasions in each patient were within the reference range of our laboratory. The prolactin responses to thyroid-stimulating hormone (TRH) were within the reference range in all except 1 patient, who had an exaggerated peak response. L-dopa induced a decrease in prolactin values in all patients. A mean decrease of 75.4 ± 20.4% (range 53.4 - 100%) was recorded.

The luteinising hormone:follicle-stimulating hormone (LH:FSH) ratio computed for each patient ranged between 0.73 and 1.74 (mean 1.14 ± 0.40). Basal LH values (range 7.1 - 18.9 IU/l) were normal in all cases. Peak LH responses to GNRH ranged from 25.0 IU/l to 229.0 IU/l; 4 of the 9 patients demonstrated an exaggerated response. Basal FSH ranged between 6.2 IU/l and 15.6 IU/l and the peak responses to GNRH were between 12.9 IU/l and 38.5 IU/l. Two patients, who demonstrated exaggerated LH responses to GNRH, also had exaggerated FSH responses.

Oestradiol levels were low in 6 of 9 patients and 2 patients had progesterone levels similar to the luteal phase of the menstrual cycle.

Six patients had blunted growth hormone (GH) responses to one or other stimuli. Following adequate insulin-induced hypoglycaemia (glucose range 0.5 - 2.0 mmol/l; suppression to 27.3 ± 10.8% of basal level), 5 patients had impaired GH responses. Four patients demonstrated impaired responses to L-dopa; 3 of these patients also had impaired GH responses to hypoglycaemia. Five of the 6 patients with impaired GH secretion also demonstrated oestradiol deficiency.

One patient, who had a paradoxical GH response to TRH, also showed exaggerated responses of prolactin and LH to TRH and GNRH, respectively. Cortisol responses to hypoglycaemia were adequate in all patients studied (peak responses exceeded 600 nmol/l). Thyroid stimulating hormone (TSH) responses to TRH ranged between 5.3 mU/l and 36.8 mU/l, with only 2 patients showing exaggerated responses.

Discussion

This report of 10 patients is, to date, one of the largest series of patients with pseudocyesis in whom evaluation of pituitary function was undertaken. Previous studies were largely case reports, each based on 2 - 3 patients. The patients in this study constituted a fairly homogeneous group; all were young subjects of child-bearing age, normoprolactinaemic and had no biochemical evidence of polycystic ovarian disease. Only 1 patient had elevated basal gonadotrophins suggestive of a menopausal status. She was excluded from the statistics in order to maintain homogeneity in the remaining group. Menopausal patients can present with pseudocyesis as evidenced by a 22% incidence in a study by Burin and Klinger. We also confirmed in some of our patients the findings of Burin and Klinger of an earlier perception of fetal movements in pseudocyesis compared with real pregnancies (Table I). This was also
true of abdominal distension in this study. In some patients, however, these features became apparent later than in normal pregnancy. Galactorrhea, which was present in 8 of the 10 patients, is a common presenting feature; however in a study of the clinical features of 12 patients with pseudocyesis in Nigeria, absence of galactorrhea appears to be more common. The patients there were older and menopausal and probably did not represent true pseudocyesis. Other case reports also confirm the frequent occurrence of galactorrhea.4,10

The hormonal profiles in this study were very interesting. None of the patients had hyperprolactinaemia although sampling was undertaken on 4 separate occasions. Although elevated basal prolactin levels have been reported in a few cases, normoprolactinaemia is more common. Basal gonadotrophins were normal in all patients and the LH:FSH ratio was less than 2 in each case. Exaggerated LH responses were present in 4 patients, 2 of whom also had exaggerated FSH responses. Other reports have demonstrated normal12,14,15 and elevated basal gonadotrophins11 and only exaggerated LH responses to luteinising hormone-releasing hormone.12,14 The present study also demonstrated exaggerated FSH responses.

GH responses to hypoglycaemia and i-dopa administration proved to be inadequate in 6 of the 9 patients, despite their normal stature. Only 1 patient had an exaggerated GH response to TRH, a finding that has previously been recorded in pseudocyesis.13,14 While no previous reports have documented impaired GH secretions in pseudocyesis, it is not unreasonable to speculate from the data in the present study that patients with pseudocyesis might have a defect in GH secretion. The normal cortisol responses to insulin-induced hypoglycaemia suggest that these patients have a normal pituitary-adrenal axis. Furthermore, normal thyroxine levels and adequate TSH responses to TRH indicates that these patients also have intact pituitary-thyroid axes. Oestradiol deficiency was demonstrated in 6 of the 9 patients. Persistent corpora lutea have been cited as a possible aetiological factor in pseudocyesis; Moulton16 reported 3 cases of pseudocyesis with corpora lutea at laparotomy, while in Zarate et al.'s12 report of 2 cases laparoscopy revealed absent luteal cysts. Only 2 of our patients had progesterone levels compatible with the luteal phase of the menstrual cycle. However, we did not subject them to laparoscopic examination to ascertain the presence of cysts in the ovaries.

In this study we demonstrated normal basal prolactin, gonadotrophins and TSH levels in patients with pseudocyesis. Abnormalities recorded in this study include oestrogen deficiency, exaggerated LH responses to GnRH and impaired GH secretion, despite normal stature. We propose that the impaired GH secretion in pseudocyesis is probably a consequence of oestrogen deficiency, since definite correlations between oestrogen and GH have been demonstrated.17 The normal gonadotrophin levels and responses to GnRH suggest a hypothalamic defect in this disorder; this defect probably relates to deficient pulsatility of GnRH and subsequently LH, which in turn is responsible for the hypogonadism.

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REFERENCES