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Case report

Adult coeliac disease presenting with infertility

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Coeliac disease in adults usually presents with gastrointestinal symptoms,[1] but has rarely been recognised as a cause of infertility.[2, 3] We report a case in which the institution of a gluten-free diet was followed by successful pregnancy.

CASE HISTORY

A 21-year-old female presented to the Mater Infirmorum Hospital in 1983. She had been married for two years and had been attempting to conceive for one year. She had taken a combined oral contraceptive for nine months previously. No abnormal clinical history was obtained, and in particular there was no history of menstrual irregularity. On examination, she showed normal secondary sexual development and there were no physical signs of systemic disease. Her sex hormone profile was normal — serum FSH was 4.4 U/l (normal 0.6 – 7.5), serum LH 11.5 U/l (0.8 – 16), oestradiol (luteal phase) 450 pmol/l (165 – 700), progesterone (luteal phase) 17.7 nmol/l (6 – 80). Serum testosterone was 0.85 nmol/l (0.5 – 2.8) and serum prolactin 310 mU/l (< 360).

Laparoscopy and dilatation and curettage were performed. The Fallopian tubes were patent and no ovarian abnormalities were noted. Endometrium obtained was in the secretory phase of the menstrual cycle. At this time she had a microcytic hypochromic anaemia (Hb 8.4 g/dl) which was treated by transfusion without further investigation. Following this, despite restoration of haemoglobin levels to normal, and also therapeutic trial of clomiphene and cyclofenil, conception failed to occur.

In November 1985 she presented to the Royal Victoria Hospital with a profound recurrent microcytic hypochromic anaemia (Hb 6.5 g/dl). She gave no history of overt blood loss. Her diet was satisfactory and she denied having steatorrhoea. Apart from pallor, no abnormality was found on clinical examination. Her height was 156cm and weight 48.1kg (93% of predicted). Blood films showed a dimorphic red cell pattern. Serum iron was reduced to 4.7 µmol/l (normal 15 – 30), with an iron binding capacity of 96.7 µmol/l (normal 45 – 72) and a serum ferritin of 15 µg/l (normal 40 – 70). Faecal occult bloods were negative.

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Serum vitamin B_{12} and folate levels were normal. Serum carotene was 0.4 μmol/l (normal 1.1 – 3.7) and she excreted only 9% of a 25 g oral D-Xylose load. There was a flat response to a lactose tolerance test. Her fasting breath hydrogen concentration was elevated at 76 ppm (normal less than 20) and no further elevation occurred in response to 25 g lactose. Small bowel barium studies showed dilatation of the jejunal mucosal fold pattern with flocculation, in keeping with a malabsorption state. Jejunal biopsy revealed the presence of subtotal villous atrophy and anti-gliadin antibodies were positive (titre 1:20). Family screening for the presence of anaemia or antigliadin antibodies was negative.

A diagnosis of coeliac disease was made and she was commenced on a gluten-free diet, haemoglobin levels being restored by transfusion. This led to a rapid improvement in her well-being, and within two months she had conceived. She was delivered of a healthy girl in September 1986. Her present haemoglobin is 13.8 g/dl on no supplements.

DISCUSSION

Adult coeliac disease usually presents with gastrointestinal symptoms.[1] In our patient, the lack of such symptoms obscured the diagnosis, which was suspected only after recurrent iron deficiency anaemia without excessive blood loss had been observed. Although infertility is a known complication of coeliac disease,[4] it is rarely a presenting feature.[2, 3] Furthermore, the presence of infertility does not correlate with the severity of the coeliac disease in either males or females.[5]

Conception within two months of commencing on a gluten-free diet, after three years of infertility, strongly suggests a relationship between the two. The infertility is not likely to have been due to anaemia alone as correction of this in the past had not been followed by conception, and likewise her body weight was normal. Similar restoration of fertility has been previously reported in both female[2] and male[6] patients. In male coeliac patients, abnormal sperm motility and morphology have been described[7] which may account for impaired fertility. In female patients the reason for infertility remains unknown, although it has been suggested that it may be due to zinc deficiency.[3] The zinc status of our patient was not determined.

We would suggest that coeliac disease be considered as a cause for unexplained infertility. Although the association is uncommon, treatment is simple and may lead to conception.

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