Carpopedal spasm in an elderly man: an unusual presentation of coeliac disease

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Coeliac disease typically presents in a young person with diarrhoea, abdominal pain, weight loss, anaemia and malabsorption. Increasingly atypical presentations of this condition are recognized.

CASE HISTORY

A 68-year-old single Caucasian man was admitted to the hospital with a 24-hour history of carpopedal spasm of both hands. Apart from generalized weakness, he reported no other symptoms. Physical examination revealed carpopedal spasm, clubbing of fingers and cachexia (body mass index 14 kg/m²).

Blood tests showed severe hypocalcaemia, with a total serum calcium of 1.06 mmol/L (normal range [NR] 2.05–2.55 mmol/L). He also had low serum potassium (2.8 mmol/L; NR 3.5–5.5 mmol/L) and magnesium (0.36 mmol/L; NR 0.65–1.05 mmol/L). Other significant results included haemoglobin 10.6 g/dL (NR 13–18 g/dL), mean corpuscular volume 98.1 fl (NR 82–98 fl), vitamin B12 157 ng/L (NR >165 ng/L), folate 2.8 g/L (NR 3.1–17.5 µg/L), ferritin 252 µg/L (NR 30–250 µg/L), prothrombin time 20 s (NR 11–14 s), thyroid stimulating hormone 0.87 µU/L (NR 0.35–4.5 µU/L), phosphate 0.57 mmol/L (NR 0.8–1.45 mmol/L), albumin 32 g/L (NR 34–48 g/L) and alkaline phosphatase 313 IU/L (NR 47–141 IU/L). His serum total bilirubin, alanine aminotransferase, urea and creatinine levels were normal. Subsequent results revealed vitamin D deficiency with a low serum 25-OH vitamin D of <7 µg/L (NR 7–40 µg/L), a low 24-hour urinary calcium excretion of 0.9 mmol (NR 2.5–7.5 mmol) and a raised serum parathyroid hormone of 22.7 pmol/L (NR 1.6–6.9 pmol/L). Serology for tissue transglutaminase (tTG) antibodies was negative, and a serum IgA level of 4.95 g/L (NR 0.8–4.0 g/L) excluded selective IgA deficiency. Electrocardiograph at admission

Figure 1 Barium follow-through X ray showing dilated proximal bowel loops and absence of normal jejunal mucosal fold pattern

showed prolonged QT interval (corrected QT interval 0.54 s; NR <0.44 s). In view of cachexia, clubbing and negative tTG-antibodies, he was further investigated for an occult malignancy. Chest X ray was unremarkable. Computerized tomographic scan of abdomen and pelvis showed simple liver cysts but no evidence of malignancy. Barium meal and follow through showed dilated proximal bowel loops and absence of normal feathery pattern of the jejunum, features suggestive of a malabsorptive state (Figure 1). Upper gastroscopic examination was normal; however, the duodenal biopsy showed partial and subtotal villous atrophy with increased intra-epithelial lymphocyte infiltration, consistent with the diagnosis of coeliac disease (Figure 2).

He was initially treated with intravenous infusions of calcium, potassium and magnesium, followed by oral supplements of calcium, potassium, vitamin D and multivitamins. Following the diagnosis of coeliac disease, he received dietary advice regarding the gluten-free diet and was discharged from hospital. On review four months after discharge, he was feeling much better, and he had no further episodes of carpopedal spasm. His weight had

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Figure 2 Photomicrograph showing small intestinal mucosa with loss of villi, crypt hyperplasia and increase in intra-epithelial lymphocytes. (H&E $\times 100$)

improved from 38.4 kg at discharge to 43 kg, prothrombin time normalized, and all electrolytes had remained within the normal range.

COMMENT

Our case illustrates several unusual features of coeliac disease, including the advanced age at presentation, severe hypocalcaemia and electrolyte disturbances as the initial manifestations, minimal gastrointestinal symptoms, and negative tTG-antibodies. Although coeliac disease classically presents in a young person with gastrointestinal symptoms and malabsorption, increasingly adults and those presenting with non-gastrointestinal symptoms are being diagnosed with this disease.¹ An increased awareness of the illness and a reduced threshold for investigation, together with better access to serological testing, have contributed to the changing presentation of coeliac disease in recent years.

Although hypocalcaemia is a common finding in coeliac disease, occurring in about 10% of cases at diagnosis, hypocalcaemic carpopedal spasm is a rare initial manifestation of the disease.^{2–5} Hypocalcaemia in coeliac disease is caused either by the reduced intestinal absorption of calcium secondary to malabsorption of fat-soluble vitamin D or by primary malabsorption of calcium.⁴ Furthermore, as magnesium deficiency can lead to defective parathyroid hormone action,⁶ hypomagnesaemia is likely to have contributed to severe hypocalcaemia in our case.

Serological testing for tTG antibodies is usually carried out as an initial investigation of coeliac disease. The test has over 90% sensitivity and 95% specificity.¹ Selective IgA deficiency, which is more prevalent in patients with coeliac disease compared with the general population, will show negative serology for IgA isotype tTG antibodies. IgA deficiency was excluded in our case. This case highlights the importance of considering duodenal biopsy in all patients with a strong clinical suspicion of coeliac disease even if the serology for tTG-antibodies is negative. In a recent study, seven of 585 patients (1.2%) who were negative for tTG antibodies but clinically considered to be at a high risk for having coeliac disease because of the presence of weight loss, anaemia or diarrhoea were found to have the disease on duodenal biopsy.⁷

In conclusion, coeliac disease should be considered in all cases of unexplained hypocalcaemia, particularly in the presence of other electrolyte abnormalities. Advanced age, lack of gastrointestinal symptoms and negative antibodies do not exclude the diagnosis.

Competing interests None declared.

Contributorship KS collated the clinical data, reviewed the literature and wrote the first draft of the article. BV and TS were supervising consultants during the patient's hospital admission. MP carried out histological examination of the duodenal biopsy. All authors contributed to the final version of the article. BV is the guarantor for the article.

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