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Persistent monoarthritis and occult coeliac disease

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Summary: Arthritis is a recognized complication of untreated coeliac disease. Symptoms and signs usually respond to the institution of a gluten-free diet. We report a case of occult coeliac disease presenting as a monoarthritis. Severe and progressive erosive damage has occurred in his talo-navicular joint despite a response to the institution of a gluten-free diet.

Introduction

The association between non-erosive synovitis and coeliac disease has been well documented.[1] In most cases, the arthritis improves following the institution of a gluten-free diet. We report a further case of coeliac disease this time presenting as an acute monoarthritis and intestinal malabsorption. Arthritis of the talo-navicular joint has persisted despite strict adherence to a gluten-free diet for over 12 months.

Case report

A 42 year old male tool setter presented in March 1991 with a 3 week history of swelling of the right ankle. The swelling failed to respond to a course of prednisolone and naproxen instituted by the general practitioner. He recalled having intermittent pain in his right ankle for up to 12 months previously.

He gave no history of psoriasis or inflammatory bowel disease. The only relevant points in his past history were surgery for a left club foot in childhood and diagnosis of Wolf–Parkinson–White syndrome in 1987 for which he was being treated with disopyramide.

On examination, he appeared to be pale but generally well. The right ankle joint was swollen, warm, tender and erythematous. The clinical impression was one of septic arthritis of the ankle. All other joints were clinically normal.

Initial laboratory investigations revealed a haemoglobin of 8.3 g/dl with an mean corpuscular volume (MCV) of 56 fl and a white cell count of 9.7×10^9/l with a normal differential white cell count. Erythrocyte sedimentation rate (ESR) was elevated at 58 mm/hour as was the C-reactive protein (CRP) at 54 mg/l (normal <6 mg/l). Liver function tests revealed an elevated alkaline phosphatase of 126 U/l. Synovial fluid analysis revealed a white cell count of 4.9×10^9/l (75% polymorphs). No crystals were seen and both Gram staining and culture were negative. A Mantoux test was negative.

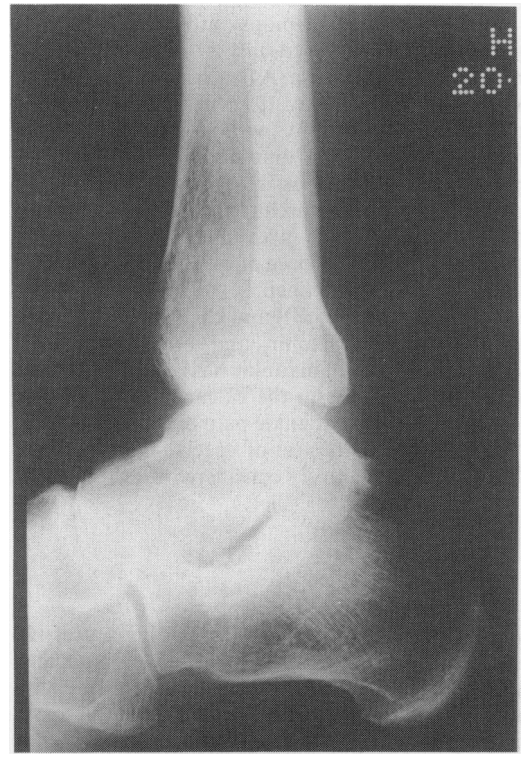

Figure 1 Normal X-ray of ankle at presentation (April 1991).

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Serum IgA was elevated at 418 IU/ml (normal range 80–220) while serum IgG and IgM were normal. He was HLA-A1, A3, B7, B8, DR2 and DR3 positive. Rheumatoid factor (Latex) was negative. Serological examination was negative for yersinia, rubella, parvovirus and chlamydia. Autoantibody profile including anti-reticulin antibodies was persistently negative. Radiological examination of the right ankle revealed marked soft tissue swelling with marginal osteoporosis (Figure 1). X-rays of his hands were normal.

Investigation of the anaemia revealed a vitamin B_{12} level of 148 ng/l (normal 150–800), a serum folate level of 4.8 μg/l (normal 3.5–15), and a serum ferritin of only 5 ng/ml (normal 30–400).

A barium meal examination revealed flocculation of the barium consistent with malabsorption. Duodenal biopsy revealed sub-total villous atrophy.

He was commenced on a gluten-free diet, vitamin B_{12} injections, and oral iron and folate supplements. A few weeks later, examination revealed that the ankle was still quite painful despite the absence of any objective swelling and further attempts at aspiration of the ankle joint were unsuccessful. X-rays at this time revealed severe erosive change and widening of the right talo-navicular joint together with pronounced osteoporosis.

A repeat duodenal biopsy after 6 months of a gluten-free diet revealed only a limited and partial regrowth of the mucosa. A further duodenal biopsy was performed in May 1992 and revealed patchy partial villous atrophy with a mild increase in inflammatory cells. A marked improvement compared to the previous biopsy was noted with improvement of the architecture and less inflammation. Rechallenge with gluten as well as a synovial biopsy have been offered to the patient but have so far been declined.

At 12 months, his ESR and CRP are now normal as are his haemoglobin and other haematinic indices. An X-ray of his ankle has revealed progressive erosive change in the calcaneo-cuboid joints (Figure 2). Persistent ankle pain on weight-bearing has necessitated the use of special surgical shoes and he is being actively considered for arthrodesis of the right ankle.

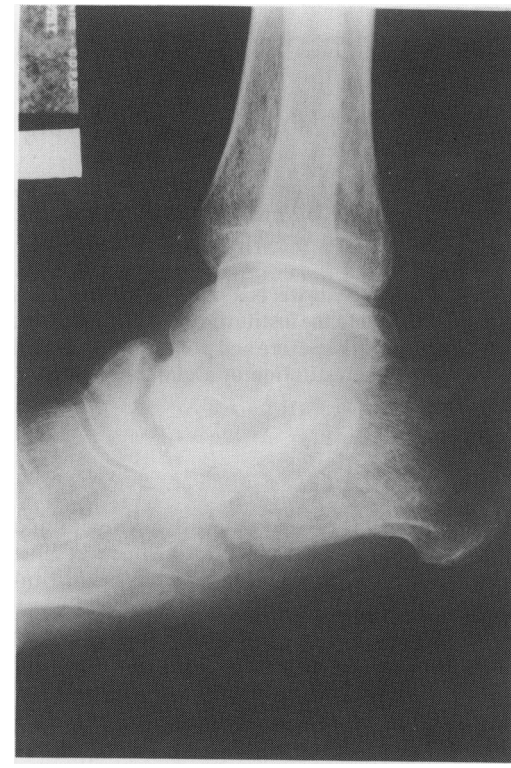

Figure 2 X-ray of ankle showing progressive erosive change with involvement of calcaneo-cuboid joints (April 1992).

Discussion

We have described a case of a rapidly progressive and destructive arthropathy of the talo-navicular joint in a patient with coeliac disease. Although the association may well be incidental in this case, investigation has so far been unsuccessful in identifying a more likely aetiology. Tuberculous arthritis would be such a candidate; however, the Mantoux test is invariably positive in these cases and it was unequivocally negative in our patient making this possibility very unlikely.

Severe joint erosion has not been reported previously with this condition, although a recently described case[2] had mild carpal and metatarsophalangeal erosions.

Early diagnosis of coeliac disease before the onset of radiological change may prevent joint damage, although articular symptoms do not invariably remit with a gluten-free diet.[3] The value of serum folate testing in the diagnosis of occult coeliac disease has been documented,[4] although this is not a universal finding[1] as in this case and should not be used as a screening test. Red cell folate estimation is probably more sensitive in testing for occult coeliac disease.[4]

The diagnosis of coeliac disease requires a positive duodenal biopsy, a clinical response to a gluten-free diet, and ideally clinical and/or haematological relapse on rechallenge with gluten.

Anti-gliadin antibodies can be used as a screening test especially in children. Serum IgA anti-gliadin antibodies decline rapidly on a gluten-free diet and can be used to follow dietary control. Antibodies are also seen to a number of autoanti-

gens including connective tissue reticulin, nuclear antigens, thyroid and parietal cells.[5]

Clinical response to strict gluten withdrawal is usually rapid, especially in children[6] and often in adults.[4,7] The extent of mucosal response, although often complete, varies widely from patient to patient. While it seems likely that most of the variation between patients is directly related to the strictness of gluten exclusion, mucosal response to a strict diet may remain incomplete for several years, especially in older people. Severe gluten-induced damage, associated with severe disease and therefore presumably extensive, may reach an irreversible state.[8]

Occult coeliac disease should be considered in patients with an atypical oligoarthritis even in the presence of joint damage. Bowel symptoms are only present in 50% of patients with coeliac disease.[7] Greater awareness of the association between coeliac disease and arthritis may lead to earlier recognition and treatment.

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Notes