Case report

Osteomalacia and coeliac disease presenting as isolated dactylitis

A S M JAWAD,* S B SQUIRE, AND J D PERRY

From the Department of Rheumatology, The London Hospital, Whitechapel, London

SUMMARY A 30 year old Pakistani female patient with osteomalacia and coeliac disease presenting as an isolated dactylitis is reported.

Dactylitis is a term used to describe swelling of the short tubular bones of hands and feet, usually without joint involvement. Causes of dactylitis include tuberculosis, congenital syphilis, sarcoidosis, and sickle cell anaemia. We describe the first reported case of a patient with osteomalacia and coeliac disease (gluten sensitive enteropathy) presenting as an isolated dactylitis.

Case report

A 30 year old Pakistani woman presented with pain in the right middle finger. She was otherwise well and had no past history of or contact with tuberculosis. She had, however, been treated in Pakistan 10 years previously for osteomalacia and five years previously for iron deficiency anaemia. She was receiving no regular drugs at the time of referral to the rheumatology department.

Examination was normal apart from a diffuse bony swelling of the middle phalanx of the right middle finger. This was neither tender nor inflamed.

INVESTIGATIONS

Investigations showed the following: serum calcium 2.19 mmol/l, serum phosphate 0.57 mmol/l, total protein 83 g/l, albumin 46 g/l, alkaline phosphatase 451 IU/l (normal range (NR) 25-90), haemoglobin 107 g/l, white blood cell count $8.6 \times 10^9/l$ (neut-

rophils 38%, lymphocytes 57%), and erythrocyte sedimentation rate 52 mm/h.

An x ray examination of the hands showed local osteoporosis and a lytic process involving the middle phalanx of the right middle finger with subperiosteal resorption on the radial side (Fig. 1). The chest x ray was normal.

With a possible diagnosis of tuberculous dactylitis, she was admitted for bone biopsy of the affected area. She had a very weakly positive Mantoux test at 1/10 000, and the histology of the bone biopsy specimen was non-specific. No acid fast bacilli were cultured from the specimen. The subsequent vitamin D level was $<1.8 \mu\text{g/l}$, and she developed symptoms of weakness and abdominal pains. Looser's zones were noted on x ray examination of her pelvis. An iliac crest bone biopsy specimen showed no uptake of labelled tetracycline, which confirmed the diagnosis of osteomalacia. Her dietary assessment showed her to have a low overall intake, normal calcium intake, low vitamin D intake, and minimal exposure to sunlight. Further investigations indicated a more generalised malabsorption as follows: vitamin B₁₂ assay 207 ng/l (NR 210-920), serum folate $0.6 \mu\text{g/l}$ (NR 2-14), red cell folate $152 \mu\text{g/l}$ (NR 125-600), plasma iron $5 \mu\text{mol/l}$ (NR 13-31), and total iron binding capacity ($74 \mu\text{mol/l}$) (NR 45-73).

A jejunal biopsy specimen taken by Crosby capsule confirmed a flat jejunal mucosa with a prominent lymphocytic infiltrate in the lamina propria, suggesting gluten sensitive enteropathy. Examination of duodenal aspirate showed no parasites.

After treatment with a gluten free diet and

Accepted for publication 8 January 1988.

Correspondence to Dr J D Perry, Department of Rheumatology, The London Hospital, Whitechapel, London E1 1BB.

*Present address: Department of Rheumatology, Norfolk and Norwich Hospital, Brunswick Road, Norwich NR1 3SR.

Fig. 1 Local osteoporosis and a lytic process involving the middle phalanx of the right middle finger with subperiosteal resorption on the radial side.

Fig. 2 After treatment with a gluten free diet and calciferol the x ray appearance of the middle phalanx of the right middle finger resolved.

calciferol the x ray appearance of the middle phalanx of the right middle finger resolved (Fig. 2), as did the classical radiological features of osteomalacia in her pelvis.

Discussion

Well recognised rheumatic manifestations of coeliac disease include osteomalacia, muscle weakness (due to vitamin D deficiency or hypokalaemia, or both),

and paraesthesiae (due to hypocalcaemia).¹ Other reported features include polymyositis,² recurrent pericarditis,³ and cutaneous vasculitis.⁴ A symmetrical, seronegative, non-deforming polyarthritis or oligoarthritis involving mainly large joints may be the presenting features of coeliac disease and joint pain may precede the bowel symptoms by many years.⁵⁻⁷ In all reported cases the joint symptoms responded to a gluten free diet.

The tendency towards hypocalcaemia in osteomalacia almost invariably leads to secondary hyperparathyroidism.⁸ The radiological bone changes in our patient's hands were confined to the middle phalanx and though they may well have been caused directly by the osteomalacia, it seems more likely that they were a manifestation of secondary hyperparathyroidism. Unfortunately, we have no measurement of the patient's parathyroid hormone concentration at the time of presentation.

In our patient the apparent dactylitis and the radiological bone changes resolved after treatment with a gluten free diet and calciferol, as did the radiological features of osteomalacia in the pelvis.

References

- 1 Falchuk Z M. Gluten-sensitive enteropathy. *Clin Gastroenterol* 1983; **12**: 475-93.
- 2 Henrikssen K G, Hallert C, Norrby K, Walan A. Polymyositis and adult coeliac disease. *Acta Neurol Scand* 1982; **65**: 301-19.
- 3 Dawes P T, Atherton S T. Coeliac disease presenting as recurrent pericarditis. *Lancet* 1981; **i**: 1021-2.
- 4 Meyers S, Dikman S, Spiera H, Schultz N, Janowitz H D. Cutaneous vasculitis complicating coeliac disease. *Gut* 1981; **22**: 61-4.
- 5 Bourne J T, Kumar P, Huskisson E C, Mageed R, Unsworth D J, Wojtulewski J A. Arthritis and coeliac disease. *Ann Rheum Dis* 1985; **44**: 592-8.
- 6 Adelizzi R A, Pecora A A, Chiesa J C. Coeliac disease: case report with an associated arthropathy. *Am J Gastroenterol* 1982; **77**: 481-5.
- 7 Prier A, Le Quintrec Y, Camus J P, Gendre J P, Pradel B. Polyarthritides inflammatoires associées à une atrophie villositaire du grêle. *Rev Rhum Mal Osteoartic* 1980; **47**: 661-5.
- 8 Bikle D D. Osteomalacia and rickets. In: Wyngaarden J B, Smith L H, eds. *Cecil textbook of medicine*. 17th ed. Philadelphia: Saunders, 1985: 1425-31.