CHRONIC SCLEROSING OSTEOMYELITIS
AN UNUSUAL CASE

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An unusual case of bilateral chronic sclerosing osteomyelitis of the clavicles is reported. A culture of resistant Staphylococcus aureus was obtained. Curettage of the lesions resulted in healing and symptomatic relief. There has been no recurrence on follow-up at one year.

In 1893 Garré described a sclerosing form of osteomyelitis that caused distension and thickening of bone, but no suppuration, sequestration or fistula formation. Hardmeier, Uehlinger and Muggli (1974) introduced the term primary chronic sclerosing osteomyelitis to differentiate the clinical presentation of chronic osteomyelitis from the clinical description given by Garré; they also described the radiological appearances.

Primary chronic sclerosing osteomyelitis is a rare condition often causing diagnostic difficulty (Cabanela et al. 1974; Kopits and Debuskey 1977). Such is the insidious onset and the radiological appearances of expansion and sclerosis of bone (Jacobsson et al. 1978), combined with the absence of signs of acute sepsis, that this condition often gives rise to a preliminary diagnosis of Ewing’s sarcoma, osteosarcoma (Cabanela et al. 1974) or osteoid osteoma (Jaffe and Lichtenstein 1940).

Positive bacteriological culture is uncommon (Anderson 1980; Collert and Isacson 1982) although there may be a history of chronic sepsis at another site (Turlington 1973; Kopits and Debuskey 1977; Goldstein, Byrne and Miller 1979).

The characteristic radiological appearance is one of reactive sclerosis with layers of subperiosteal bone visible. Cystic change may occur within the sclerotic area, but sequestra are uncommon. Radioisotope bone scanning using technetium–99m shows a high uptake at four hours. (Jacobsson et al. 1978).

The histological picture is of low-grade chronic osteomyelitis associated with new bone formation and zones of possible necrosis (Jacobsson and Heyden 1977; Collert and Isacson 1982). It is thought to represent a proliferative response of bone in a patient with a high tissue resistance or a low-grade infection, or both (Goldstein et al. 1979). This appearance is quite different from the bone destruction usually associated with infection.

Histological confirmation and, where possible, bacteriological confirmation of the diagnosis are important in order to avoid confusion with a neoplasm. Curettage is usually diagnostic and often curative (Kopits and Debuskey 1977) although Blockey (1983) described three cases of chronic osteomyelitis (featuring frequent relapses) which were controlled by antibiotics.

CASE REPORT

A 14-year-old girl presented with a three-month history of pain and swelling in the medial end of the right clavicle. There was no history of illness or previous minor sepsis and she had not been taking antibiotics. Examination revealed a swelling over the medial end of the right clavicle; the overlying skin appeared normal and there was no lymphangitis or lymphadenopathy.

Radiological examination showed expansion of the medial end of the right clavicle with layers of sclerotic new bone suggestive of a Ewing’s sarcoma. Tomograms revealed cystic changes within this sclerotic area which were more suggestive of chronic sepsis.

The erythrocyte sedimentation rate was 6 mm in the first hour. The white cell count was 7.8 x 10⁹/μl, with a normal differential count. Salmonella and brucella titres were normal and the Venereal Disease Research Laboratories test also was normal. The serum alkaline phosphatase level was elevated at 180 (normal range 21–91) IU/l. Calcium and phosphorus levels and an intravenous pyelogram were all normal.

A biopsy of the right clavicle was performed. Histological examination showed broad trabeculae of
Histological appearance of biopsy specimen from the right clavicle showing dense osteosclerosis with bone trabeculae forming an interlacing pattern. Osteoblasts and occasional osteoclasts are seen applied to the trabecular surfaces. The intertrabecular spaces are filled with bland reactive fibrous tissue (haematoxylin and eosin, × 50).

Bone surrounded by osteoblasts and occasional osteoclasts (Figs 1 and 2). The intertrabecular spaces were filled with loose fibrovascular material containing foci of acute and chronic inflammatory cells suggesting a diagnosis of chronic osteomyelitis.

A radioisotope bone scan four days after biopsy showed intense uptake over the medial end of the right clavicle, and also, increased uptake over the medial end of the left clavicle (Fig. 3). The left clavicle had been asymptomatic. Further tomographic examination of both clavicles confirmed sclerosis and cyst formation which also affected the left clavicle (Fig. 4). Curettage of both clavicles was then performed two months after presentation and soft "amorphous" material removed. Cysts were entered in the right clavicle. No frank pus was present.

From the specimen, coagulase positive Staphylococcus aureus with multiple antibiotic resistance was grown. Cultures for acid-fast bacilli were negative. The girl was given flucloxacillin and fusidic acid for six weeks. Wound healing was slow but uneventful and she had no further pain. Radiological examination at review (one year from onset) showed reduction in the extent of the sclerosis and cystic change.

DISCUSSION
A search of the literature did not reveal any similar report of chronic sclerosing osteomyelitis affecting both clavicles symmetrically and concomitantly. The majority of reports refer to maxillary or mandibular foci subsequent to chronic low-grade oral or dental sepsis. (Turlington 1973;
Jacobsson and Heyden 1977; Jacobsson et al. 1978). Collert and Isacson (1982) described the involvement of the clavicle in one of their eight cases and reported a 50% occurrence at a second site within 5.5 years.

The isolation of Staphylococcus aureus, coagulase positive, is an unusual feature in this case. Anderson (1980) states that bacteriological culture is always negative while Turlington (1973), reporting on oral cases, states that culture is always positive, with Staphylococcus aureus most commonly isolated. Blockey (1983) describing three cases of chronic sclerosing osteomyelitis reported no positive bacteriological cultures. He excluded the possibility of a compromised immunological system.

Collert and Isacson (1982) described the clinical course as characterised by the insidious onset of local pain and distension of bone, in children or young adults.

The symptoms would return at intervals for some years and then subside. Chronic fistulae did not occur in their series. Blockey (1983) described a fluctuating course with acute exacerbations controlled by antibiotics.

In the absence of the usual systemic manifestations of infection, (pyrexia, elevation of the erythrocyte sedimentation rate and a leucocytosis) the initial clinical and radiological features in this case were suggestive of a primary bone neoplasm. Jaffe and Lichtenstein (1940) and Cabanela et al. (1974) discussed the clinical problem of osteomyelitis masquerading as neoplasm and presenting in a similar way to osteoid osteoma, Ewing’s sarcoma or osteosarcoma. Thus in chronic sclerosing osteomyelitis biopsy is mandatory, and every effort should be made to culture the organism, which may be resistant to many antibiotics.

REFERENCES


