Bilateral sabre-like tibial deformity in secondary syphilis: case report

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SUMMARY A girl aged 12 years having early acquired syphilis with a rare bilateral sabre-like tibial deformity is reported.

Bone involvement, in particular sabre-tibia is a well known complication of congenital and late acquired syphilis.1 In early syphilis, bone invasion is much less obvious and has been infrequently reported, partly due to the variations in the methods of evaluation.2,3 The reported incidence of bony and joint symptoms in acquired syphilis is approximately 4%.4 Syphilis is to be suspected in a patient presenting with bone pains, worse at night.1,5 We report a case of early acquired syphilis presenting with bony swelling of both tibiae (sabre-like) and bone pains.

Case report

A 12 year female child was referred to the STD clinic from the orthopaedic department with complaints of swelling and nocturnal pain in both lower legs of 4 months duration. Pain was insidious in onset, dull aching and came in bouts. It progressively increased in intensity, frequency and duration. There was history of a genital ulcer 9 months previously, which had healed with topical medication after 2 weeks. There were no other constitutional or systemic symptoms. History of sexual assault of the girl 10 months previously by her mother’s paramour, in an attempt to get rid of his genital ulcer, was revealed. Both the parents had genital ulcers 21 months before, the mother was affected first, and subsequently the father.

General physical examination revealed an afebrile, emaciated, pale child with significant generalised lymphadenopathy involving axillary, inguinal, cervical and epitrochlear groups of lymph nodes. They were not tender, discrete, firm and rubbery in consistency. There was no jaundice, cyanosis or pedal oedema. Systemic examination of the respiratory, cardiovascular, neurological and gastrointestinal systems was normal. Examination of musculoskeletal system revealed tender tibiae with “sabre-like” swelling. There was no evidence of joint involvement. Gynaecological examination revealed only a depigmented scar on the vulva. Serological tests for syphilis demonstrated Venereal Disease Research Laboratory positivity.

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Fig 1 Anteroposterior and lateral radiograph of left leg, showing increased thickness of the cortex of tibia.
Laboratory (VDRL) to be 1:64 and Treponema pallidum haemagglutination (TPHA) to be 1:160. CSF biochemistry and cytology were within normal limits and both VDRL and TPHA were non-reactive in CSF. A complete blood count, urine, blood biochemistry, chest radiograph, and ECG were within normal limits.

Radiological examination of bones of both legs revealed thickening of the diaphyses only, metaphyses and epiphyses being normal (fig 1). Tibial bone biopsy revealed extensive new bone formation without much evidence of inflammation. Radiography of rest of the bones was essentially normal. A similar finding was obtained on CT as well (fig 2).

Examination of both parents revealed old healed atrophic scars over the genitalia with generalised nontender, discrete, significantly enlarged lymph nodes. In addition, the mother also had significant, non-tender hepatosplenomegaly. Blood VDRL and TPHA were highly reactive, being 1:64 and 1:160 in the mother and 1:4 and 1:80 in the father. CSF cytology and biochemistry were normal. CSF-VDRL and TPHA were also non-reactive in both parents. ESR was high in both parents but other routine investigations were normal. Radiological skeletal survey in both parents revealed no abnormalities.

The patient and her parents were treated as early syphilis with penicillin in accordance with the recommendations of the WHO. Bone pain disappeared in the patient within 2 days and she was completely asymptomatic when seen after 4 weeks. Bony prominence was, however, present.

Discussion

The secondary stage of acquired syphilis usually starts six to eight weeks after the primary stage, with manifestations such as cutaneous and mucosal lesions along with generalised lymphadenopathy. Occasionally internal organs are involved leading to manifestations such as hepatitis, splenomegaly, nephrosis, iritis, meningitis, myositis, arthrosis, bursitis, and tenosynovitis.

Bone involvement appears to be unusual in the secondary stage, in contrast to its involvement in the tertiary stage of acquired syphilis and congenital syphilis. The exact incidence of bone involvement is not known in early acquired syphilis. Wile and Senear reported bone and joint invasion in 36% of their 165 patients with early acquired syphilis. Reynolds and Wasserman, however, reported an incidence of only 0-15% of destructive bone disease in early acquired syphilis. There is also some evidence that bone involvement is not uncommon, but perhaps only rarely appreciated because radiological findings may be normal at this stage.

The bone involvement may be in the form of proliferative periostitis or osteolytic lesion or both, former being the commoner. The bones most often affected are the skull and the long bones of the limbs, the legs more frequently than the arms, followed by sternum and ribs. The highly cellular syphilitic granulation tissue formed around blood vessels extends into the Haversian canals leading to thickened, expanded and elevated periosteum resulting in the local pain followed by the osteoblastic process which contributes to the exostosis. The principal clinical manifestations are nocturnal pain affecting the lower limbs and exaggerated by heat. There may be localised swelling, erythema and tenderness, and pathological fractures may also occur. Our patient had swelling of both tibiae with nocturnal pain and tenderness. Radiography and CT showed diffuse thickening of the cortex and periosteum consistent with proliferative periostitis and ostitis as described earlier in the literature. More recent reports have also described invasion of skull and the limbs in the form of osteolytic lesions.

Though sabre tibia is a well known complication of late and congenital syphilis, it can occur in secondary syphilis as well. Thus, a thorough examination should be done in all young patients with bone pains to rule out early acquired syphilis.

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