AN ATYPICAL CASE OF INFANTILE CORTICAL HYPEROSTOSES

IAN D. KITCHIN, LANCASTER, ENGLAND

Caffey and Silverman (1945) gave the name of infantile cortical hyperostoses to the bony changes resembling syphilitic osteoperiostitis (McLean 1931) which Caffey (1939) had observed in non-syphilitic infants.

In a recent article by MacGregor and Davies (1949) twenty-eight cases by various authors were collected and the clinical details tabulated. In this group the age of onset varied from two to 120 weeks; the average age was just over twenty-two weeks, but the median was much less, only four patients being older than eighteen weeks. In only three instances, those described by Van Zeben (1948) in which two brothers and a cousin were affected, were more cases than one found in a family.

The case about to be described presents several unusual features: the family history, the early onset, the very marked anterior tibial bowing, and the absence of evident malaise, fever or tenderness.

The patient was a girl. The delivery was spontaneous and uneventful. The birth weight was 7 pounds. Otherwise normal and apparently healthy, she had gross deformities of both legs, which were radiographed a few hours after birth (Fig. 1). The films were of poor quality, but they showed gross thickening of both tibiae and, in the lateral views, well marked anterior bowing and subperiosteal masses. She was kept in hospital for ten days, and during that time was breast-fed. She gained weight and was apparently free from pain. There was no pyrexia.

She was first seen by me when ten days old. Then there was gross anterior and slight lateral bowing of both tibiae. The legs below the knee were thickened and brawny, and pitted slightly on pressure, but were not tender. Knee and ankle movement was full and painless, and no other abnormalities were found. The Wassermann reaction was known to

Fig. 1
A few hours after birth. The quality of the radiographs is poor, but the gross thickening and bowing of the tibia are clearly shown.
At eight weeks. Remodelling of the anteriorly bowed tibiae has begun. There is also some thickening of the fibulae, particularly in their middle thirds.

At one year. Figure 3—Right leg. Figure 4—Left leg. The tibiae have remained bowed both anteriorly and laterally, but the fibulae have remained practically straight throughout.
be negative in each parent, and there were no other clinical signs of scurvy. A diagnosis of subperiosteal haemorrhages of unknown origin was suggested. At this point the child's maternal grandmother volunteered the information that the child's mother (aged twenty-two years) had had, at birth, exactly similar deformities and had been referred to Professor McMurray, who after investigation ascribed the condition to subperiosteal haemorrhages of unknown origin and gave a hopeful prognosis. The deformities disappeared entirely in the first few years. Unfortunately the records of the examination had been destroyed by enemy action.

In view of the unusual nature of the case the patient's films were submitted to Sir Thomas Fairbank who made the diagnosis of infantile cortical hyperostoses. The child's progress was uneventful. She gained weight at normal rate and at all times looked fit and well. The whole skeleton was examined radiographically but apart from some possible irregularities in the femora the condition was confined to the tibiae. Radiographs taken eight weeks after birth showed the condition very clearly (Fig. 2). Anterior bowing was pronounced, but remodelling of the tibiae was already well advanced. At the age of one year there was still marked anterior bowing and slight lateral bowing of both legs, and they still felt rather brawny, but there was no pitting and no pain. She was just beginning to stand. Radiographs showed persistent bowing, further remodelling with buttressing along the concave sides of the bones, and transverse dense bands seen most clearly in the lateral views (Figs. 3 and 4).

Comment—The unusual features of this case of infantile cortical hyperostoses have been mentioned. No previous report of changes at birth has been found, and the initial radiographs (Fig. 1) strongly suggested that the condition had been present for some time in utero. Anterior tibial bowing has been reported previously, but it is very uncommon and had not been noted by Caffey in 100 cases (quoted by Smitham and Palmer 1950). The bowing was not due to post-natal fractures. It is most unfortunate that the mother's records have been destroyed, but the history given by the grandmother of her daughter's deformities and progress and her immediate recognition of the words "subperiosteal haemorrhages" suggests that the mother suffered from the same condition.

REFERENCES