SPINA BIFIDA AND EPIPHYSIAL DISPLACEMENT

Report of Two Cases

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It has been known for at least twenty years that spontaneous and painless fractures in patients with spina bifida show rather bizarre radiological features. Gillies and Hartung (1938) described two fractures of the upper tibia with sclerosis, fragmentation and excessive callus formation as well as widening of the epiphyseal line. Carr (1956) reported that two out of four patients with spontaneous fractures showed excessive callus, one of which also showed the other features of sclerosis, fragmentation and widening of the epiphyseal line. Golding (1960) reviewed the radiological features.

Despite these descriptions diagnostic difficulties still occur, as illustrated by the two cases to be described.

CASE REPORTS

Case 1—This boy, aged twelve, had spina bifida with complete paralysis of both legs, dislocation of the right hip and double incontinence. He had good intelligence and had managed for some years to use leg calipers with a pelvic band with moderate success despite a persistent meningocele.

FIG. 1

FIG. 2

Case 1. Figure 1—Initial radiograph of right knee showing posterior slip of lower femoral epiphysis. The left knee (Fig. 2) is shown for comparison.

First lesion—He was admitted to hospital in June 1956 at the age of seven with a lesion of the lower end of the right femur. It was recorded that he had developed “a septic sore on the right thigh. The thigh and knee were swollen and a bit inflamed, especially the thigh.” His doctor had prescribed penicillin, but the swelling remained. Examination showed a small, clean sore, with a soft-tissue swelling about the knee. There was no pyrexia and the Mantoux test was negative. The erythrocyte sedimentation rate was 25 millimetres in the first hour, and the right knee remained hot and rather red.

On the assumption that the condition was subacute osteomyelitis the femur was drilled and a biopsy specimen taken; pus was not obtained and bacteriological examination showed

no growth on culture. Histologically the specimen consisted of fragments of normal living bone. The marrow space was occupied by delicate connective tissue and a small amount of resolving granulation tissue. The paucity of inflammatory cells suggested that the reaction

FIG. 3

FIG. 4

Case 1. Figure 3—Right knee one month after first examination. There is abundant callus and the epiphysial slip appears to have been corrected. Figure 4—Four months after first examination, showing consolidation of the profuse callus.

FIG. 5

FIG. 6

Case 1. Figure 5—Right tibia and fibula, showing greenstick fractures of both bones with massive callus and periosteal elevation over whole shaft of tibia. Figure 6—Two years later.

was a response to mild irritation or aseptic trauma rather than to bacterial infection. Later, the true pathology became clear. Figure 1 shows posterior slipping of the lower femoral epiphysis on the right side (Figure 2 shows the normal left leg for comparison), which later appeared

to have been reduced spontaneously (Figs. 3 and 4), and to be associated with massive callus formation and some periosteal elevation. There was no history of injury or of pain, and the elevated erythrocyte sedimentation rate was ascribed to a chronic urinary infection.

Second lesion—About two months later, in September 1956, a swelling of the right ankle appeared without a history of injury. There was no pyrexia, but osteomyelitis was suspected. The redness began to subside after two weeks, but there was still some thickening of the lower end of the tibia. Radiographs then showed greenstick fractures of the lower tibia and fibula with massive callus formation and periosteal elevation throughout the length of the tibial shaft (Fig. 5). Figure 6 shows the appearance two years later.

Again it appears that subacute osteomyelitis was the first diagnosis suggested.

Third lesion—No further trouble occurred for over three years. But in February 1960 a warm swelling appeared over the upper end of the right tibia and knee. The swelling prevented the application of his caliper, but otherwise did not affect the boy, and again there was no history of injury. He was afebrile, healthy and eating well. Radiographs (Fig. 7) showed widening of the tibial epiphysal line and irregularity of the bone, suggestive of a bone sarcoma. The erythrocyte sedimentation rate was 41 millimetres in the first hour. The Mantoux test was negative, the white cell count was 5,500 per cubic millimetre. Exploratory operation showed a yellow, thick, adherent periosteum; the underlying bone could be cut with a knife and appeared cancellous. Histologically a specimen from the affected area showed a mass of osteoid tissue and bone which in some places was well orientated in parallel struts;

FIG. 7

FIG. 8

FIG. 9

Case 1. Figure 7—Right tibia in 1960 showing widening of the epiphysal line, irregularity of the bone and periosteal elevation. Figure 8—Four weeks later. Consolidation is occurring. Figure 9—Four months later still.

some cartilage was mixed with the bone. There was evidence of active new bone formation and much remodelling, with active osteoclasia. The appearances were suggestive of fracture callus, and there was nothing to suggest infection or tumour.

When the swelling subsided and the caliper was reapplied it was immediately apparent that it had become too short. The knee joint was at the level of the upper tibial epiphysis, suggesting that the cause of the lesion might have been a recurrent strain at the epiphysal line. Follow-up radiographs (Figs. 8 and 9) have confirmed the benign nature of the pathology.

Case 2—A boy aged ten had high spina bifida, was of rather low intelligence, was totally paralysed from the waist down, had double incontinence and was confined to bed or a wheel chair.

He was admitted to hospital in June 1960 with a hot painless swelling of the left lower leg. There was no history of injury. He was afebrile and in good general health, though with a mild chronic urinary infection. Radiographs showed slight postero-lateral slipping of the left lower tibial epiphysis (Fig. 10). Rest and splintage were advised. The swelling was slow to

FIG. 10

FIG. 11

Case 2. Figure 10—Initial radiograph, showing early postero-lateral slipping of the lower tibial epiphysis.
Figure 11—Six weeks later. Increased deformity and much periosteal elevation.

subside and radiographs taken six weeks later (Fig. 11) showed a considerable increase in the displacement, as well as the extensive callus formation and periosteal elevation reminiscent of that in Case 1 (Fig. 9). It is worth recording that this boy was also admitted with a provisional diagnosis of acute osteomyelitis, and that the possible trauma here was lifting the foot on to the foot rest of a wheel chair.

DISCUSSION

We believe that the four lesions described here, presenting on three occasions as possible osteomyelitis and on one occasion as a possible malignant tumour, were all caused by relatively mild trauma. This could have occurred while the patients were being moved or while they were wearing faulty calipers. None of the lesions showed the typical features of a bone injury, and bruising was notably absent. Local warmth and swelling were the presenting features, and they suggested inflammation rather than the effects of injury. The abnormal radiographic appearance may represent repeated movement of the fracture or epiphysial slip, because such movement must occur easily in the absence of sensation.

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