CONGENITAL PSEUDARTHROSIS AND BOWING OF THE FIBULA

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The purpose of this paper is to describe experience with four children who presented with bowing or pseudarthrosis of the fibula. These children demonstrate a gradation in the severity and significance of this condition. There may be: 1) fibular bowing without fibular pseudarthrosis (Case 1); 2) fibular pseudarthrosis without ankle deformity (Case 2); 3) fibular pseudarthrosis with ankle deformity but without the late development of tibial pseudarthrosis (Case 3); or 4) fibular pseudarthrosis with the late development of pseudarthrosis of the tibia (Case 4).

The aetiology and pathology of bowing and pseudarthrosis of the fibula are similar to those of antero-lateral bowing and pseudarthrosis of the tibia. Similar conditions affect other bones developed by endochondral ossification—the first rib, clavicle, femur, radius and ulna. Thus pseudarthrosis in the fibula may occur after fracture through a congenital cyst or an area of fibrous dysplasia, or occasionally in neurofibromatosis. Fracture may also occur through an area of mesodermal maldevelopment in which the diaphysis is tapered, sclerotic and often bowed and the cortex thickened, with a narrowed or obliterated medullary cavity. Pseudarthrosis or bowing may involve the fibula alone, but more commonly it is associated with abnormalities in one or both tibiae.

Langenskiöld (1967) described three cases of progressive valgus deformity of the ankle with persisting pseudarthrosis of the fibula following successful bone grafting for a pseudarthrosis of the tibia. Carrell (1938), Boyd (1941) and Boyd and Sage (1958) also mentioned similar cases. Boyd and Sage described a case of congenital pseudarthrosis of the fibula with a normal tibia. The opposite leg was affected by pseudarthrosis of both bones.

CASE REPORTS

Case 1—A girl was first seen in July 1969 at the age of 11 months, because of bowing of the lower half of the left leg with prominence of the fibula just above the ankle. There was no history of injury. Radiographs showed lateral bowing and sclerosis of the lower shaft of the fibula, without obvious abnormality in the tibia (Fig. 1). The bowing diminished slightly in the ensuing three years (Fig. 2). At review late in 1972 it was noted that she had developed soft swellings on the forehead and right side of the back, and also some pigmented hairy areas on the back.

Case 2—A girl was brought for advice at the age of 15 months in 1966. It had been noted at birth that there was mild bowing of the left leg. The child had begun to walk at the age of one year. At the initial examination the left leg was bowed below the knee, the convexity of the bowing being antero-lateral at the junction of the middle and lower thirds of the tibia and fibula. There was also mild torsion of the left tibia of approximately 30 degrees, compared with approximately 10 degrees of medial torsion on the right side. There were no stigmata of neurofibromatosis. Radiographs showed a pseudarthrosis of the left fibula (Fig. 3). There was a mesodermal defect of mild degree at the mid-section of the tibia, with increased density of the cortex, narrowing of the medullary cavity and antero-lateral bowing.

It was feared that the abnormality of the tibia might precede pseudarthrosis of that bone. For this reason a half ring-top caliper was fitted and worn for a year. Since then the leg has gradually straightened spontaneously. At the most recent review the child was aged eight years; she was free from symptoms and leading an active and normal life. Radiographs at that time showed a persisting pseudarthrosis of the fibula with improved appearances of the tibia (Fig. 4).

Case 3—A boy aged twelve years attended in April 1971 with severe valgus of the left ankle. The deformity had been noticed by the parents for six months. There were no symptoms, and there was no history of any injury. Radiographs showed a valgus ankle with a pseudarthrosis of the lower fibula (Fig. 5). The remainder of that bone was tapered and sclerotic and the tibia showed slight thickening of the medial cortex. Presumably this thickening was due to increased medial stress in the weight-bearing bone associated with the valgus deformity. Supramalleolar closed-wedge osteotomy was performed in October 1971. Two screws were placed across the fibula into the tibia, one above the osteotomy site and one below. The tibial bone removed in the osteotomy was used as a graft to join the lower fibula to the
Case 1. Figure 1—Antero-posterior radiographs of both legs at presentation at the age of 11 months in July 1969. Figure 2—Three years and one month later the bowing of the fibula has lessened; the tibia remains normal.

Case 2. Figure 3—Antero-posterior and lateral radiographs at the age of 15 months. Figure 4—Antero-posterior radiographs of both legs at the age of 7 years. Both leg bones are less bowed—the pseudarthrosis persists.
Case 3. Figure 5—The radiological appearance at presentation with pseudarthrosis of the fibula and a valgus deformity of the ankle. Figure 6—The appearance following union of the osteotomy. Figure 7—Antero-posterior radiograph following removal of the screws.

Case 4. Figure 8—Antero-posterior radiographs of both legs at presentation in June 1960. The fracture of the fibula is difficult to discern at this stage; however, the abnormality of both tibia and fibula is obvious. Figure 9—The fracture is obvious in radiographs five months after presentation and there is no evidence of callus formation.
tibia. After three months (Fig. 6) there was solid union of the osteotomy in good position. The screws were removed in December 1972 (Fig. 7). Histological section of the pseudarthrosis showed fibrous tissue only.

**Case 4**—A boy aged thirteen months was brought in June 1960 with a history of having caught his left leg in the wheel of a bicycle. As a result, there was a fracture of the lower shaft of the fibula through abnormal bone and an abnormal tibia with narrowing and dense cortex and lateral bowing (Fig. 8). A pseudarthrosis developed in the fibula (Fig. 9). Five months after the fracture, in December 1960, the pseudarthrosis was explored and a biopsy showed fibrous tissue only. Figure 10 shows the appearance five months after biopsy. In August 1962 an attempt was made to secure union of the fibular fragments by medullary nailing combined with an inlay iliac bone graft, after excision of the pseudarthrosis (Fig. 11). The leg was kept in plaster for four months. The pseudarthrosis persisted.

In January 1966 the left leg was 1.25 centimetres shorter than the right. The ankle showed valgus deformity. Radiographs taken soon after this showed a partial fracture of the lower third of the left tibia, which had remained dense and rather bowed while the fibula remained ununited. The leg was immobilised in a caliper. By August 1966 the boy was complaining of pain in his left leg. Radiographs in October of that year showed a definite fracture of the tibia (Fig. 12).

In October 1966 a double onlay cortical bone graft from the right tibia was applied by the Boyd technique, with insertion of a block of cancellous bone between the two main fragments. The operation was successful in securing union. The boy has continued to wear the caliper; at the age of thirteen he remains well (Fig. 13).

**DISCUSSION**

Congenital pseudarthrosis and bowing of the fibula are similar in aetiology and pathology to those of the tibia, but not so serious because it is not a weight-bearing bone.

Severe bowing of a congenitally abnormal fibula may occur and persist without obvious abnormality in the tibia and without the development of valgus deformity at the ankle (Case 1). Treatment is not necessary. The condition is analogous to bowing of the tibia without the development of pseudarthrosis.

A related condition is pseudarthrosis of the fibula alone (Case 2). Unlike congenital pseudarthrosis of the tibia, progressive deformity and disability do not necessarily ensue. The bowing of the leg may improve and deformity of the ankle may not develop. Pseudarthrosis of the fibula should not be treated by grafting because failure is as likely to occur as in pseudarthrosis of the tibia (Case 4; see also Langenskiöld's (1967) experience). Furthermore, because the fibula is not a weight-bearing bone, its integrity is not so necessary as is the case with the tibia.

Pseudarthrosis of the lower fibula may lead to a progressive valgus deformity of the ankle (Case 3). If the child is young and the deformity uncontrolled by splinting, the operation of synostosis of the fibula to the tibia is advised (Langenskiöld 1967). The operation involves excision of the pseudarthrosis and grafting of the distal fibular metaphysis to the tibia. Progressive deformity is thus prevented. Osteotomy of the tibia is necessary, in addition, if the valgus deformity is severe enough to warrant correction. If the patient presents with a valgus ankle after maturity, supramalleolar osteotomy of the tibia is all that is required (Boyd and Sage 1958).
Case 4. Figure 12—In October 1966 there is a clearly defined fracture of the tibia. Figure 13—The present radiological appearance. There is sound union of the tibia and persistent pseudarthrosis of the fibula.

Should pseudarthrosis of the fibula be associated with obvious abnormality of the tibia (Case 4), there is a high likelihood of the subsequent development of pseudarthrosis of the tibia. Protective splinting should be worn until skeletal maturity.

SUMMARY
1. The cases of four children who presented with bowing or pseudarthrosis of the fibula only, are described.
2. There is a gradation in the severity and significance of this condition. There may be fibular bowing without fibular pseudarthrosis; fibular pseudarthrosis without ankle deformity; fibular pseudarthrosis with deformity but without the late development of tibial pseudarthrosis; or fibular pseudarthrosis with the late development of tibial pseudarthrosis.
3. Proper management is dependent on a knowledge of this range of conditions.

REFERENCES