

Isolated congenital anterolateral bowing of the fibula : A case report with 24 years follow-up

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Only very few cases of congenital anterolateral bowing of the fibula have been described in literature. We report a girl with congenital anterolateral bowing of the fibula with a 24 year follow-up. Asymptomatic bowing of her left leg below the knee was noted at the age of three years. Radiographs showed an isolated anterolateral bowing of the fibula with a normal ipsilateral tibia. Supervised neglect was advised. A spontaneous fracture occurred one year later. A shortening osteotomy resulted in solid consolidation of the fracture. At the age of 27 years the deformity had progressed into a two-level fracture of the fibular diaphysis with pseudoarthrosis. The segment in-between the fractures was resected. Following this, the patient returned to a normal level of activities in just a matter of weeks. She could even participate in recreational tennis six weeks after surgery and was then discharged from the outpatient clinic. Isolated congenital anterolateral bowing of the fibula is a very rare condition and there is no consensus on its treatment. In this case, removal of the painful pseudarthrotic fragment of the fibula led to a good result.

Keywords : congenital bowing fibula ; congenital pseudoarthrosis fibula.

INTRODUCTION

Congenital anterolateral bowing of both tibia and fibula is a well recognized diagnosis in children. On the other hand, isolated congenital bowing of the fibula is very rare and only a few cases have been reported. We report on a 24 years follow-up of a young girl with asymptomatic congenital anterolateral bowing of the fibula that eventually progressed into a symptomatic two-level pseudoarthrosis in adult life.

CASE REPORT

A girl was presented to the outpatient clinic at the age of three years with an asymptomatic bowing of the lower leg. There was no history of injury. The family history reported neither skeletal congenital malformations nor neurofibromatosis. Physical examination showed bowing of the left lower leg at the lateral side. The ankle joint was in normal alignment and there were no other abnormalities. Radiographic examination of the left and right lower legs showed an isolated anterolateral bowing of the left fibula with a normal straight tibia (fig 1).

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Fig. 1. — Antero-posterior (AP) (a) and lateral (b) radiograph of the left lower leg at the age of 3 years, showing bowing of the fibula, a straight tibia and normal ankle alignment.

There were no abnormalities in the right lower leg. Supervised neglect was advised, without any surgical attempts to achieve correction. One year later a spontaneous fracture of the fibula occurred (fig 2). To treat this fracture a shortening osteotomy of the fibula was performed, leading to fracture healing (fig 3). At the age of twelve years she complained of lower leg pain on weight-bearing. Radiographs showed an obliterated medullary cavity in the distal end of the left fibula (fig 4). Supervised neglect and analgesia was advised. During routine screening, a minimal leg length difference was found at the age of 18, which caused no complaints. As an adult of 27 years of age the patient once again presented spontaneously, complaining of pain at the lateral side of her left lower leg. Physical examination revealed a significant swelling at the distal end of the fibula. Radiographic examination showed a two-level fracture of the distal one-third of the fibula with pseudoarthrosis (fig 5). Radioisotope scan



Fig. 2. — AP radiograph of the left lower leg showing a diaphyseal fracture of the fibula at the age of 4 years.

showed increased uptake at the fracture sites. Magnetic Resonance Imaging did not reveal any malignant abnormalities. Due to the sclerotic appearance of the bone and the narrowed marrow cavity, chances of good consolidation seemed to be very poor, so an invasive reconstruction with a bone graft was not attempted to treat the pseudarthrosis of the fibula. Instead, the bone fragment was excised (fig 6). At the first follow-up, two weeks after surgery, the wound healed nicely and she was asymptomatic. Her gait was normal ; even playing recreational tennis six weeks after surgery at second follow-up did not cause any symptoms, therefore she was discharged from the outpatient clinic

DISCUSSION

Bowing of the lower leg can be anterior or posterior. Whereas posterior bowing has a tendency to remodel, anterolateral bowing is the more serious



Fig. 3. — AP radiograph of the left lower leg showing a solid consolidation of the fibular fracture at age 5 years.

deformity, with a strong tendency towards progressive bone resorption and fracture, often leading to a pseudoarthrosis (1,6). The aetiology and pathology of congenital anterolateral bowing and pseudoarthrosis of the fibula are similar to those of anterolateral bowing and pseudoarthrosis of the tibia. Treatment of fibular bowing however, is not similar to tibial bowing, because the fibula is not a weight bearing bone (3). It only carries 7-15% of the body weight (4). Grading is based on the amount and functional repercussions of fibular bowing and / or pseudoarthrosis e.g. on the ankle function. There may be a) fibular bowing without fibular pseudoarthrosis ; b) fibular pseudoarthrosis without ankle deformity ; c) fibular pseudoarthrosis with deformity but without the late development of tibial pseudoarthrosis; or d) fibular pseudoarthrosis with the late development of tibial pseudoarthrosis (3). The present case started as type a) and progressed



Fig. 4. — Lateral radiograph of the left lower leg at the age of 18 years shows an obliterated marrow cavity in the distal fibula.

into type b) with a two-level pseudoarthrosis. The treatment of anterolateral bowing without any other abnormalities is usually directed towards management of impending fracture or pseudoarthrosis and its persistence (6). Others favour the idea that treatment is not necessary in case of severe fibular bowing without any abnormality in tibia or ankle alignment (3). Spontaneous pathologic fractures mostly occur in the bowed region. These fractures often do not heal normally, leading to non-union or pseudoarthrosis (5). Our patient underwent a shortening osteotomy at the age of 4 years. Because of her young age, chances of consolidation were good and after one year the fracture healed. The presence of callus usually represents a normal healing response and it indicates a favourable prognosis (6). However, at the age of 27, our patient developed a pseudoarthrosis of a two-level fracture of the distal one-third of the fibula. Various treatments have



Fig. 5. — AP radiograph of the left lower leg showing pseudoarthrosis of a two-level fracture of the fibula, at age 27 years.

been described in literature for this type of fracture or pseudoarthrosis (2). The goal of treatment should not be to just restore the continuity of the fibula, but rather to preserve normal development and alignment of the ankle and prevent late valgus deformity (7). Based on the latter principle, the recommended treatment of patients with fibula pseudoarthrosis without ankle deformity is observation and close monitoring. Pseudoarthrosis of the fibula should not be treated by grafting because failure is as likely to occur as in pseudoarthrosis of the tibia (3). Some authors have advised against simple resection of the pseudoarthrosis (7). In this case however the patient was an adult and we felt that resection of the pseudoarthrosis with an adjacent bone segment was possible without the risk of development of valgus of the ankle. The procedure led to a good clinical



Fig. 6. — AP radiograph of the left lower leg two weeks after resection of the mobile bone fragment of the fibula in between the two-level fracture. The configuration of the ankle joint is normal.

result. Fibular osteosynthesis is only indicated when 1) the ankle alignment is neutral 2) the distal fibular fragment is large and long enough to be internally fixed and 3) the gap at the pseudoarthrosis can be approximated or filled with bone graft at the time of internal fixation (2). In the patient presented, radiographs of the left leg showed an obliterated medullary cavity in the distal part of the fibula. Therefore, osteosynthesis appeared as a poor option, and the fibular fragment was resected. Our patient was already skeletally mature and because the syndesmosis remained intact during surgery the ankle was not affected.

Pseudoarthrosis of the fibula may also occur through a congenital cyst, an area of dysplasia or occasionally in neurofibromatosis (3). Neurofibromatosis has been associated with 50% of patients with pseudoarthrosis of the tibia (6). It is assumed that neurofibromatosis is also associated with pseudoarthrosis of the fibula. Our patient did not have neurofibromatosis. Due to its rare appearance the treatment of isolated fibular bowing should be individually determined. Isolated congenital pseudoarthrosis of the fibula follows a variable clinical course with fibular status, ankle alignment and patient age influencing treatment (2).

This case is of interest because of the success of the simple treatment applied. Moreover, because of the very long follow-up and the detailed description of the pathology and subsequent treatment, it is possible to gain more insight into this rare condition. This case report differs from other case reports in treatment and outcome : surgical intervention at a young age did not prevent the patient from presenting fractures later in life, and subsequent removal of the pseudoarthrotic fragment without bone grafting resulted in durable functional recovery.

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