Genu recurvatum following below knee amputation for congenital tibial pseudarthrosis

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Abstract
We report a case of premature arrest of the proximal tibial apophysis leading to genu recurvatum deformity following below knee amputation for congenital tibial pseudarthrosis. In the literature there are reported cases of genu recurvatum following early growth arrest at the proximal tibial apophysis, these typically followed fracture, skeletal traction or surgery and we believe this to be the only such case. We postulate that this case of genu recurvatum is a complication of prolonged below knee patella weight bearing prosthetic limb use in the young child and patients should be made aware of this.

INTRODUCTION
We report a case of premature arrest of the proximal tibial apophysis leading to genu recurvatum deformity following below knee amputation. The growth of any physis can be impaired in many ways but most commonly it follows a fracture. In the literature there are reported cases of genu recurvatum following early arrest at the proximal tibial apophysis, but these typically followed fracture, skeletal traction or surgery. We believe this to be the only reported case of genu recurvatum resulting from the use of a below knee prosthetic limb.

CASE REPORT
The patient presented to soon after birth in 1990 with a deformity of his right lower leg. Radiographs confirmed an abnormality throughout the tibia with pseudarthrosis in the distal third of the shaft as well as in the distal fibula. A biopsy did not identify neurofibromatosis. By the time the patient was three years of age, a severe angulated deformity has developed, he therefore underwent intramedullary stabilization with a steinmann pin and a tibial onlay graft. Post-operatively he was treated in a hip spica and then broomstick plasters for a total of four months. At this stage the metalwork was removed and the leg held in a long leg plaster for a further three months. The leg was intermittently examined under anaesthetic to assess bony union. Treatment with an ilizarov frame was attempted twice and finally union was achieved, however, whilst in plaster he refractured (Figure 1).

A below knee amputation was performed in 1996 aged six. A conventional below knee patella weight bearing prosthesis was fitted. At age 15, he represented having suffered two right patella dislocations. Clinical examination revealed patella mal-tracking and genu recurvatum. Radiographs (Figure 2) showed growth arrest of the anterior tibial apophysis causing the recurvatum. CT studies of the knee confirmed patella mal-tracking and lateral tilt.
DISCUSSION

Genu recurvatum is the uncommon skeletal deformity resulting from premature closure of the proximal tibial apophysis. This growth discrepancy between the anterior and posterior portions of the physis causes the anterior inclination of the tibial plateau.

The tibial tuberosity does not appear until 12-15 foetal weeks, and it does not have a recognisable growth plate until several months after birth[1]. At birth, the tubercle lies proximal to its eventual position. During postnatal development, the tubercle gradually assumes a position distal to the horizontal portion of the proximal tibial growth plate. Ehrenborg[2] described four characteristic stages of tibial tuberosity development: A-The cartilaginous phase, there is no ossification centre in the cartilaginous anlage of the tibial tubercle. This persists in girls until the age of 11 and in boys until 13 years. B-The apophyseal phase, the ossification centre commences formation in the distal portion of the tuberosity. This occurs between 8 and 12 years in girls and between 9 and 14 years in boys. C-The epiphyseal stage, there is a coalescence of the ossification centres of the tuberosity and proximal tibia forming a tongue of bone continuous with the proximal tibial epiphysis. This occurs in girls between 10 to 15 years of age and in boys from 11 to 17 years. D-The bony stage, the proximal tibial growth plate closes (See Figure 3).

According to Ogden[,], the tuberosity growth plate develops three histologically different regions, although there is no distinct demarcation. Proximally it is analogous to the proximal tibial growth plate and enchondral ossification process except the cell columns are short. This region then gives rise to a fibrocartilagenous zone with layers of fibrocartilage, hyaline cartilage and bone formed by membranous ossification. The third and most distal region shows a change from hyaline cartilage to fibrous tissue, and then to bone again. During growth and distal extension of the secondary ossification centre of the proximal tibial epiphysis into the tuberosity, the columnar region also extends more distally. When the proximal tibial and tuberosity ossification centres fused, the columnar plate extends nearly to the end of the tuberosity, and fibrocartilagenous growth becomes less evident. The fibrous portion remains at the distal end.

There are numerous reported cases of genu recurvatum, but most commonly as a complication of prolonged skeletal traction. Bjerkreim and Benum[3] published a series of seven cases in 1975. All of the patients had sustained an ipsilateral femoral fracture managed with skeletal traction through the proximal tibial metaphysis. They postulated that genu recurvatum resulted from upward migration of the wire traction, damaging the epiphyseal plate. Similarly Van Meter and Branick[4] reported a bilateral case following similar treatment for bilateral femoral fractures. Ishikawa et al[5] report a case following distal femoral traction for a femoral fracture. Morton and Starr[6] reported two cases following tibial traction for proximal tibial fractures and not a femoral fracture. Bowler et al[7] presented two cases after femoral fracture, which had not been treated with tibial skeletal traction. Other published cases include Stirling[8], who in 1952 reported a case associated with Osgood-Schlatter's disease, a case following tibial tubercle transfer[9] and tibial physeal injury following fracture[10].
We believe that our case of genu recurvatum resulted from increased pressure on the anterior tibial tubercle. There are two other similar cases presented in the literature, one following treatment with plaster cast after correction of an angular deformity in a tibial fracture and in the second case after prolonged treatment with patellar tendon bearing brace\[11\]. It was recognised as early as 1862 by Hueter\[12\] and Volkmann\[13\] that pressures parallel to the axis of epiphyseal growth affect the rate of such growth. Increased pressure in this direction will inhibit growth; decreased pressure will favour and accelerate it. It has been shown that the forces required to completely inhibit growth of a physis were at least 400 pounds\[14\], however, Appleton\[15\] in 1932 proved that even relatively small forces upon the growth plate led to definite limb deformities. Trueta\[16\] concluded that compression of the growth plate led to interference of the blood flow. This was found to be proportionate to the damage caused by compression to the epiphyseal side of the growth plate and the duration of compression.

We postulate that our case of genu recurvatum resulted from increased pressure on the apophysis. The pressures were not high enough to completely arrest growth but did cause growth retardation and early apophyseal fusion.

In conclusion, we feel that genu recurvatum is a complication of prolonged below knee patella weight bearing prosthetic limb use in the young child and patients should be made aware of this.

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