CASE REPORT

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Unusual proximal tibiofibular synostosis

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Abstract Proximal tibiofibular synostosis without multiple hereditary exostosis is extremely rare and only 7 cases have been reported in the literature. All of the previously reported cases accompanied deformities such as distal positioning of the proximal tibiofibular joint, leg length discrepancy, bowing of the fibula, and valgus deformity of the knee. The present case of a 24-year-old man had neither a history of trauma nor deformity around the knee. Therefore, it was suggested that this type of synostosis occurred after epiphyseal plate closure.

Résumé Une synostose tibio-fibulaire proximale sans une maladie exostosante héréditaire multiple est extrêmement rare et seulement sept cas ont été mentionnés dans la bibliographie. Tous les cas précédemment rapportés s'accompagnaient de déformations telles qu'un positionnement distal de l'articulation tibio-fibulaire proximale, une anomalie dans la longueur de la jambe, une courbure de la fibula avec une distance interosseuse augmentée, et/ou une déformation en valgus du genou. Le cas présent concerne un homme âgé de 24 ans ne présentant pas l'antécédent d'un trauma ni d'un déformation du genou. Par conséquent, il est suggéré que ce type de synostose s'est produit après une obturation de la plaque épiphysaire.

Introduction

Although synostosis of the distal tibiofibular joint has occasionally been described in the literature, proximal tibiofibular synostosis is extremely rare. Only 7 cases

[2,4,5,7,9,11], not associated with a known generalized disease such as multiple hereditary exostosis [10], have been previously reported. O'Dwyer [7] in 1991 radiologically classified proximal tibiofibular synostosis into 3 types. All 3 types include at least one deformity such as distal positioning of the proximal tibiofibular joint, leg length discrepancy, bowing of the fibula with increased interosseous distance, and valgus deformity of the knee. Therefore, the anomalies were congenital or occurred before closure of the epiphyseal plate. The present case had none of these deformities and no history of trauma.

Case report

A 24-year-old man, who had been involved in a volleyball club for 6 years from 13 years of age, was referred to our department in February 1992 with a 3-year history of pain at the lateral side of his right knee while standing up or squatting. There was neither a significant family history nor a past history. Examination revealed tenderness at the proximal tibiofibular joint and slight quadriceps muscle atrophy. There was no evidence of any neurological defect, the leg lengths were equal, and no valgus or varus deformity was observed. The pain was induced on flexion of the knee with dorsiflexion of the ankle during weight-bearing. Roentgenograms of the knee demonstrated absence of the proximal tibiofibular joint space and proliferative bone formation around this joint. There was neither dislocation nor subluxation of the proximal tibiofibular joint (Fig. 1). Bone scan revealed intense accumulation at the right proximal tibiofibular joint (Fig. 2). Computed tomography showed bony continuity of this joint without joint space (Fig. 3). Magnetic resonance imaging (MRI) demonstrated no cartilaginous intensity in proximal tibiofibular joints (Fig. 4). Biopsy of the joint was performed to exclude tumors and synovial disease. The histopathology of the specimen revealed only osseous or fibrous tissue without hyaline cartilage, inflammatory reaction, or atypism of the nucleus (Fig. 5). We diagnosed the patient with idiopathic proximal tibiofibular synostosis and rec-

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Fig. 1A–C Radiographs of the right knee taken in February 1992. **A** Anteroposterior view; **B** Lateral view; **C** Oblique view

Fig. 2 Bone scintigram taken in February 1992 showing high uptake at the proximal tibiofibular joint in the right knee

Fig. 3 Computed tomogram in February 1992 showing osseous continuity between tibia and fibula

Fig. 4 MRI showing no cartilaginous intensity in proximal tibiofibular joints

Fig. 5 Histopathology obtained from the biopsy of the proximal tibiofibular joint in March 1992 (haematoxylin and eosin stain)

ommended resection of the fibular shaft to reduce mechanical stress to the proximal tibiofibular joint from the ankle joint. However, he refused further treatment and was closely followed. The patient still complained of pain at the lateral side of his right knee while standing up or squatting and dorsiflexion of his right ankle was limited when he was last seen in April 1998.

Discussion

Reports of proximal tibiofibular synostosis are extremely rare and only 7 cases have been previously reported [2,4,5,7,9,11]. Rahm [9] in 1924 described the first case of synostosis in a 43-year-old patient and O'Dwyer [7], in 1991, reviewed 7 cases, including his case, and classified them into 3 types. Type 1 (12-year-old patient [11]) had a straight fibula with synostosis arising 2 cm distal to the fibular growth plate and extending to the junction of the middle and distal thirds of the tibia. Type 2 (6- [7], 9- [11], and 13- [4]-year-old patients) had a fibula of almost normal length with mild bowing and widening of the interosseous distance in the proximal half. The oldest type 2 patient [4] also demonstrated valgus deformity of the knee. Synostosis in type 2 occurred at the proximal end of the fibula. In type 3 (17- [2], 43- [9], and 50- [5]-year-old patients), synostosis occurred in a more distal level than type 2, and marked bowing of the fibula and increased interosseous distance occurred throughout its length. The patients with type 3 accompanied valgus deformity of the knee. Synostosis in type 3 occurred at the proximal end of the fibula. Two of 3 cases had bilateral synostosis.

The cases with types 1 and 2 were considered to occur before the growth spurt and those with type 3 were considered to occur after the growth spurt. In both types 2 and 3, synostosis occurred at the ordinal proximal tibiofibular joint and so this synostosis situated distal to the growth plate of the proximal tibia and proximal to the growth plate of the fibula. Therefore, the site of synostosis locates more distal accompanying growth of the lower leg, and type 2 is supposed to transfer to type 3. The present case had a fibula of normal length and shape, normal position of the proximal tibiofibular joint, and normal alignment of the knee. These findings suggested that this synostosis occurred after closure of the growth plate. We advocate that classification of proximal tibiofibular synostosis should include our case as type 4.

Inclination of this joint surface varies considerably from a nearly horizontal plane to a steep incline with the articular surface of the fibula facing anteriorly. According to Ogden [8], there were two basic types of this joint, namely the horizontal type and the oblique type that were divided by an arbitrary definition of 20°. He also found that as the obliqueness of the incline increased, the surface area and the mobility of this joint decreased. Barnett and Napier [1] found a significant correlation between the anatomical shape of the joint and progressive dorsiflexion of the ankle. The primary functions of the joint are dissipation of torsional loading applied at the ankle joint and absorption of lateral tibial bending moments [6,8,10]. Eichenblat and Nathan [3] found the absence of articular cartilage in 1.9% at the tibial side and 2.2% at the fibular side in this joint and reported the possibility of syndesmosis of this joint as found in the distal tibiofibular joint.

In the present patient, who exhibited a narrow joint surface with little mobility of the joint and possible absence of joint cartilage, namely a syndesmotic joint, repeated mechanical stress to this joint caused by dorsiflexion of the ankle during volleyball resulted in ossification of this joint.

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