

Giant Intra-Articular Extrasynovial Osteochondroma of the Knee: A Report of Two Cases

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Abstract: We report two cases of a giant extrasynovial osteochondroma of the knee located in the infrapatellar fat pad region, in two females who were 58 and 71 years old respectively. Both patients had noticed the mass many years before our first clinical observation. In both patients, at physical examination a solid, firm and hard mass was palpable in the anterior part of the knee in Hoffa's fat pad region, and the range of motion of the knee was severely restricted and painful. CT scan examination with 3D-reconstruction showed two large, calcified neoformations behind the patellar tendon, between the apex of the patella and the proximal third of the tibia. In both cases, the mass was completely resected surgically through an anterior longitudinal approach. At histological examination, the excised masses consisted of an outer layer of hyaline cartilage without significant chondrocyte atypia and an inner region of bone trabeculae formed by endochondral ossification. At follow-up, 8 and 4 years after the operation, both patients were pain-free, with complete recovery of the range of motion of the knee and without any clinical or radiographic evidence of recurrence. The authors believe that intra-articular extrasynovial osteochondroma of the knee is a primary metaplasia of Hoffa's fat pad. Usually, the tumor develops slowly and asymptotically over many years. The treatment of choice is a marginal resection of the mass, although a biopsy should be considered in some cases. Recurrences are extremely rare.

Keywords: Hoffa's fat pad, Intra-articular osteochondroma, Knee tumors.

INTRODUCTION

Intra-articular or para-articular osteochondroma of the knee is an uncommon benign lesion located in the region of the infrapatellar fat pad. Many different terms have been used to describe this lesion that in some cases can become quite large, causing pain and restriction of the range of motion of the knee, especially of flexion [1-10]. In these cases, surgical treatment is indicated: it consists of complete excision or marginal resection with an open anterior approach [3, 5, 10]. Differential diagnosis should always be performed for chondrosarcoma and synovial chondromatosis. Some authors believe that intra-articular or para-articular osteochondroma of the knee is the end-stage of Hoffa's disease [9].

The authors report two rare cases of a giant intra-articular extrasynovial osteochondroma of the knee which was marginally resected; they describe the clinical, CT scan and histological aspects of the lesion, and discuss the possible pathogenesis, the differential diagnosis and the indications for treatment.

CASE 1

A 58-year-old woman, who was a teacher, presented with a painful subcutaneous mass on the medial side of the patellar tendon of the right knee, which she had first noticed about 10 years earlier. She had had no pain in the affected knee until one year previously, but she reported a slow,

progressive enlargement of the mass. She had no history of trauma. During the previous year, the mass continued to increase in size and became painful, with a progressive restriction of the range of motion of the knee. At physical examination a tender, round, firm and hard mass was palpable in the anterior part of the right knee; flexion was restricted and painful. She was able to walk short distances, with a limp. X-rays and CT scan examination with 3D-reconstruction showed a large, calcified neoformation, lying between the inferior margin of the patella and the proximal third of the right tibia. The mass was unique and replaced Hoffa's fat pad. Laboratory tests were normal. The mass was interpreted as a benign tumor; therefore, surgical treatment was planned. At surgery, the joint was exposed by an anterior longitudinal approach; the mass, which almost completely occupied the infrapatellar fat pad, was isolated and marginally resected. At histological examination, the tumor consisted of a bone mass formed by endochondral ossification which was surrounded by an outer layer of hyaline cartilage without significant chondrocyte atypia. The intertrabecular spaces were occupied by adipose tissue. These findings were diagnostic for osteochondroma of the infrapatellar fat pad. At follow-up, 8 years after the operation, the patient was pain-free and her walking was unrestricted; her knee had fully regained its range of motion, although she complained of mild discomfort when kneeling. No clinical or radiographic evidence of recurrence was present (Fig. 1A-D).

CASE 2

A 71-year-old woman, who worked on a farm, presented with a large mass on the anterior part of the left knee, which had become painful during the preceding 8-12 months. The patient had noticed the mass about 25 years earlier, but it

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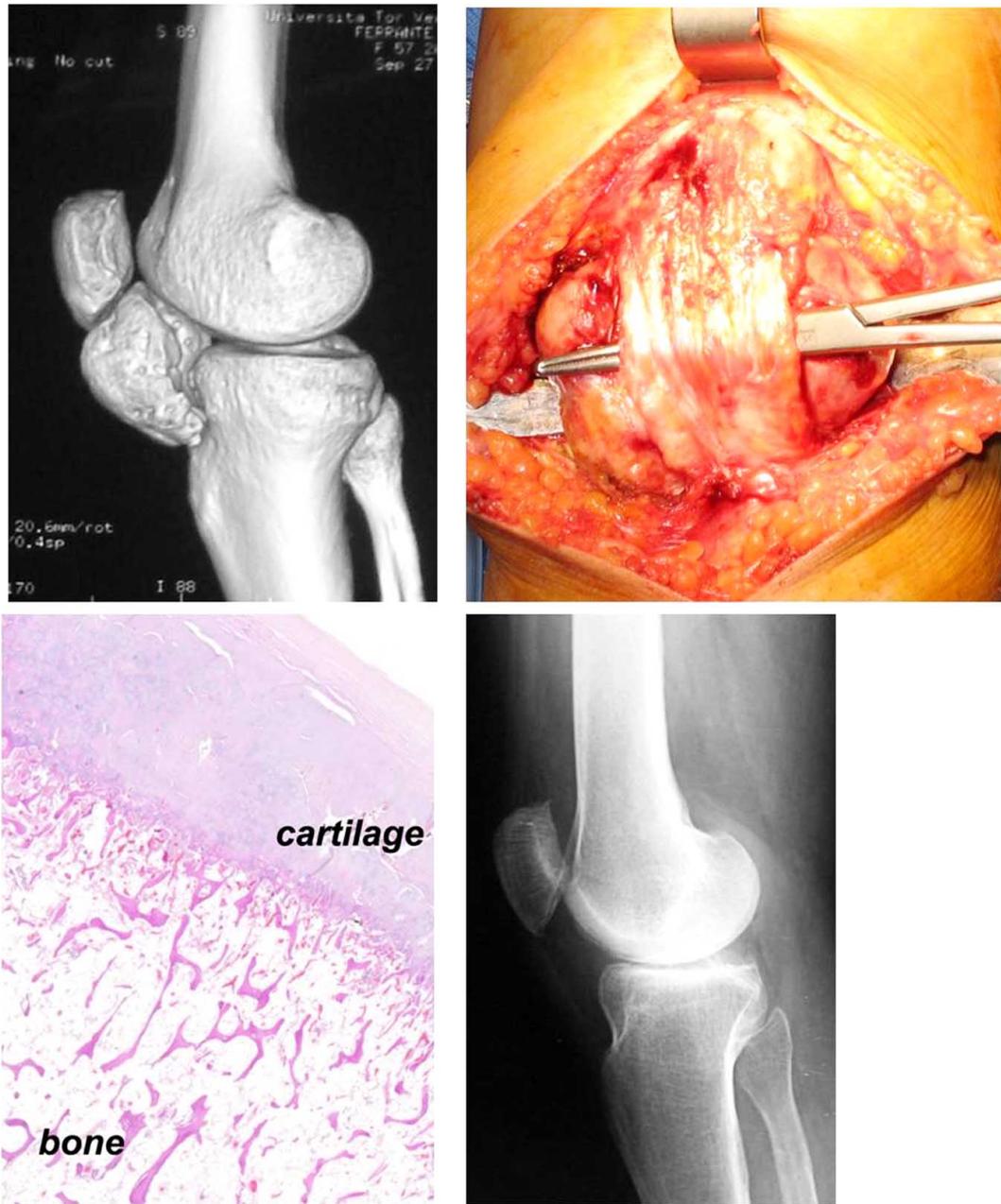


Fig. (1). Three-dimensional CT scan examination of the right knee in a 58-year-old woman affected by a giant osteochondroma located between the apex of the patella and the proximal tibia (A). The tumor was isolated behind the patellar tendon and marginally resected (B). At histological examination, the bone mass formed by endochondral ossification was surrounded by an outer layer of hyaline cartilage without chondrocyte atypia (C). At follow-up, 8 years after surgery, the radiographic examination did not show any evidence of recurrence (D).

was quite small and completely asymptomatic. She reported very slow growth of the mass, which remained painless during the following 15 years. At that time, the mass was still very small, but started to be quite painful; it was treated successfully with four steroid injections. During the following 9 years, the mass continued to grow progressively without pain, but the patient noticed an increasing restriction of the range of motion of her knee, especially of flexion. At our observation, the mass was very large and painful, with severe limitation of knee function. At physical examination a solid, hard neoformation was present on both sides of the patellar tendon. The range of motion was severely restricted and painful. Laboratory tests were normal. X-rays and CT

scan examination with 3D-reconstruction showed a calcified neoformation, located over both the medial and lateral border of the patellar tendon, between the apex of the patella and the proximal tibia. In this case, too, the mass was located in Hoffa's fat pad region, and it was considered as a benign tumor. It was marginally resected en bloc, through an anterior approach: the neoformation was intra-articular but extrasynovial. The histological examination showed the same finding as the previous case. At follow-up, 4 years after the operation, the patient was pain-free and able to perform farm work, with full range of motion of the operated knee. No evidence of recurrence was present.

DISCUSSION

Large osteochondromas involving the joint capsule or para-articular soft tissues adjacent to a joint are rare osteocartilaginous tumors, first described by Jaffe in 1958 [11]. Subsequently, these lesions have been reported using various terms such as para-articular osteochondroma [2, 5, 10], intracapsular chondroma [6], para-articular chondroma [7, 8], giant extrasynovial osteochondroma [9], Hoffa's disease [4] and giant intra-articular osteochondroma [1]. In both our cases, the neoformation was interposed between the joint capsule externally and the synovial membrane internally, and at histological examination the mass was characterized by trabecular bone tissue surrounded by hyaline cartilage. For this reason, we believe that the most appropriate term to define our lesions is "giant intra-articular extrasynovial osteochondroma". In most cases, this type of osteochondroma involves the anterior compartment of the knee (76% of the cases), although they may develop in other regions such as the ankle (19% of the cases) or the foot (5% of the cases) [5]. The location in the posterior compartment of the knee is extremely rare [12].

The pathogenesis and classification of this lesion are still controversial. Rizzello *et al.* [10] reported that this tumor seems to originate from a cartilaginous metaplasia of the articular and para-articular connective tissue. Rodriguez-Peralto *et al.*, [6] emphasize a possible metaplastic origin of the tumor, due to a traumatic event of Hoffa's fat pad. Other authors [9, 13] reported a relation between this type of osteochondroma of the knee and a chronic impingement of the infrapatellar fat pad; they conclude that this lesion is the end-stage of Hoffa's disease. Neither of our cases reported any history of significant trauma or symptomatic impingement of the infrapatellar fat pad, although both patients had noticed the mass many years before our first observation. So, in our cases the pathogenesis is not relatable to a mechanical post-traumatic event or to a chronic impingement of Hoffa's fat pad (end-stage of Hoffa's disease), but to a primary cartilaginous metaplasia of the infrapatellar fat pad.

Usually, osteochondromas of the knee develop slowly over many years; however, recently Carmont *et al.*, [14] reported a para-articular osteochondroma of the knee developing in 5 months, following a minor injury. Both our patients had noticed the mass respectively 10 and 25 years before our first observation.

Impingement symptoms related to osteochondromas have rarely been reported, although Ozturan *et al.*, [15] recently described a case of patellar tendinopathy caused by a para-articular extraskeletal osteochondroma located in the infrapatellar region of the knee. In both our cases, the tumor grew slowly without any significant subjective symptoms for many years; it became symptomatic only when the mass was very large.

Several authors reported no clinical or radiographic signs of degenerative joint disease after surgical resection of osteochondroma, although mild osteoarthritis may coexist in some cases [6, 9, 10]. We did not observe significant signs of osteoarthritis in either of our cases, even though one of our patients was 71 years old at surgery.

The most common differential diagnosis for these lesions are synovial chondromatosis, low-grade chondrosarcoma and osteosarcoma, so in some cases a biopsy should be performed before excising the tumor. In both our cases, the clinical history, physical examination and CT scan findings of the tumor pointed to a benign osteochondroma; therefore, we did not perform a biopsy. However, a complete resection might be useful also in osteosarcoma, but postoperative management will be different. We believe that a biopsy is absolutely indicated when a significant diagnostic doubt exists. Magnetic resonance (MR) may be helpful for the diagnosis [8, 13], but in both our patients it was contraindicated, in one case owing to the presence of a pacemaker and in the other case for psychiatric problems.

In their review of the literature on surgical treatment of this lesion, Reith *et al.*, [5] did not report any recurrence of the mass after surgical resection, although the follow-up was in all cases quite short. In agreement with these authors, no clinical or radiographic signs of recurrence were noticed in our cases, followed up 4 and 8 years after surgery; therefore, in these tumors, a marginal resection of the mass represents the treatment of choice.

CONFLICT OF INTEREST

None of the authors has any conflict of interest.

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