

Dilemma of Distinction: Synovial Chondrosarcoma and Synovial Chondromatosis - A Rare Entity with Different Therapeutic Modalities

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Abstract

Case Report

Synovial chondrosarcoma, whether primary or secondary to synovial chondromatosis, is a very rare entity. These two entities share a rather similar profile, not only epidemiologically and clinically, but also radiologically and anatomopathologically. Thus, the distinction between synovial chondromatosis and synovial chondrosarcoma is a real dilemma. However, this distinction is of great interest, given the very different evolutionary and therapeutic modalities. In this work, we will report a case of synovial chondrosarcoma in a 62 years old woman, whose diagnosis was confirmed by a histological test. We will then discuss the contribution of the different authors, as well as all the scientific trials, aiming at establishing concrete and objective criteria, allowing to evoke the aggressiveness of the chondrosarcoma, hidden behind the benignity of the synovial chondromatosis.

Keywords : Synovial Chondrosarcoma ; Synovial Chondromatosis ; Malignant transformation ; MRI ; Histology.

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INTRODUCTION

Synovial chondrosarcoma of the knee is a rare malignancy that develops in the cartilage cells of the knee joint. This tumor is characterized by its ability to involve surrounding tissues, including muscle, bone and nerves. The most common symptoms include persistent pain, swelling and stiffness in the knee [1,2].

The diagnosis of synovial chondrosarcoma of the knee is often difficult to make because of its rarity and similarity to other common joint conditions. Imaging tests such as X-rays, MRI and CT scans can help confirm the diagnosis. A biopsy is often necessary to determine if the tumor is malignant or benign [3,6]. Treatment for synovial chondrosarcoma of the knee depends on the size and location of the tumor, as well as the extent of its spread. In many cases, surgery to remove the tumor is necessary. Surgical options include wide excision, joint resection or amputation of the affected limb. In cases where the tumor has spread to other parts of the body, chemotherapy and/or radiation therapy may be necessary [3,9]. This case report has been reported in line with the SCARE Criteria [14].

CASE PRESENTATION

A 62 years old woman, married with three children and no profession, presented to the hospital with pain and swelling of her right knee. She has had high blood pressure for 10 years, for which she takes β -blockers and diuretics, but has no known diabetes, tuberculosis, gout, metabolic disease, or systemic disease. She underwent a cholecystectomy in February 1995 and had a right shoulder dislocation in 1999, but no right knee trauma was reported. She had a spontaneous miscarriage in 1980 and has been menopausal for 20 years. She has no known smoking, alcoholism, or other toxic habits. Her mother died of breast cancer and there is high blood pressure in her family.

In October 2020, the patient presented with a five years history of right knee pain. The pain began intermittently but became persistent over time, with no reported trauma. The last three months were marked by a significant increase in pain intensity, with no particular radiation. Joint swelling occurred repeatedly, requiring several visits to the emergency room, where anti-inflammatory treatment was initiated without success, without adequate secondary exploration. The patient also reported increasingly frequent episodes of slippage. The pain became extremely severe, with marked functional

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impotence, and the patient was referred to orthopedics. It should be noted that the history was not accompanied by fever or altered general condition.

On clinical examination, the patient was conscious, oriented in time and space, and had normally colored conjunctivae. Although she had difficulty walking, she did not use crutches or supports. The orthopedic examination, comparative standing, walking and lying on the back, showed a genu varum morphotype with an inter-condylar distance of 1.5 cm measured in the standing position. The pitch angle was open out, estimated at 10°. Monopodal support aroused moderate pain but without rocking. In the supine position, the inspection revealed a positive patellar shock and a moderate effusion, as well as a discreet flexum of a few degrees. Active flexion was 120° and passive flexion was 130°, while flexum was 5° and there was no recurvatum. The extensor apparatus showed neither convergent nor divergent strabismus of the patellae, no pain point in the patella or patellar tendon and a positive patellar test on the right knee. The meniscal testing was performed without abnormalities, and the laxity study showed that the lachman-trillat test was negative, as was the direct anterior and posterior drawer, while the posterior drawer at 90° flexion was also negative. The rest of the exam was normal.

The patient's paraclinical exploration report shows that standard radiographic findings revealed significant diffuse bone demineralization of the femoral condyles, tibial plates, and patella, as well as pinching of the patellofemoral joint space with suprarotational calcifications and osteophytosis of the femoral condyles (figure 1). MRI revealed a large, heterogeneously enhancing tumor process after contrast, with tissue and cystic components, arising in the subquadricepsal recesses and extending laterally from the inferior femoral metaphysis, especially on the medial side. There is no

femoral invasion or intra-articular extension, and the lesion is relatively well delineated, also respecting the patella, the quadriceps tendon, as well as the adjacent muscle planes, notably the vastus lateralis of the quadriceps (figure 2). The CT scan did not reveal any suspicious lesions in the thoracic or abdominal regions (figure 3). Bone scan showed very moderate intensity hyperfixation of the distal third of the left femur, extended beyond the bony contour of the medial side, associated with heterogeneous hyperfixation of the left knee. These fixation abnormalities are related to the known tumor process. There is also a slight asymmetry of shoulder fixation in favor of the right side, without any pejorative character. In conclusion, the patient has a suspicious tumor process with synovial origin without invasion of the femur and the femorotibial joint.

Initially, it was decided to perform a surgical biopsy. For this, a medial approach to the knee was performed, followed by a parapatellar arthrotomy, which revealed a macroscopic appearance of chondromatosis (figure 4). After that, a synovectomy was decided and the surgical specimen (figure 5) was sent for pathological examination.

The surgical specimen was sent for pathological examination in three different laboratories. The results showed different but concordant findings. In the first laboratory, the cartilaginous proliferation was well differentiated and suggested a well-differentiated low-grade soft tissue chondrosarcoma rather than a chondroma, and needed to be compared with the clinical data. In the second laboratory, the tumor proliferation was undecided between a chondroma and a low-grade chondrosarcoma, but given the high cellularity, binucleation and atypia, the diagnosis was in favor of a low-grade chondrosarcoma. Finally, the third laboratory confirmed the diagnosis of low-grade chondrosarcoma.



Figure 1: X-ray of the left knee face and profile



Figure 2: MRI of the left knee and thigh showing a suspicious tumor process with synovial origin without invasion of the femur and the femorotibial joint

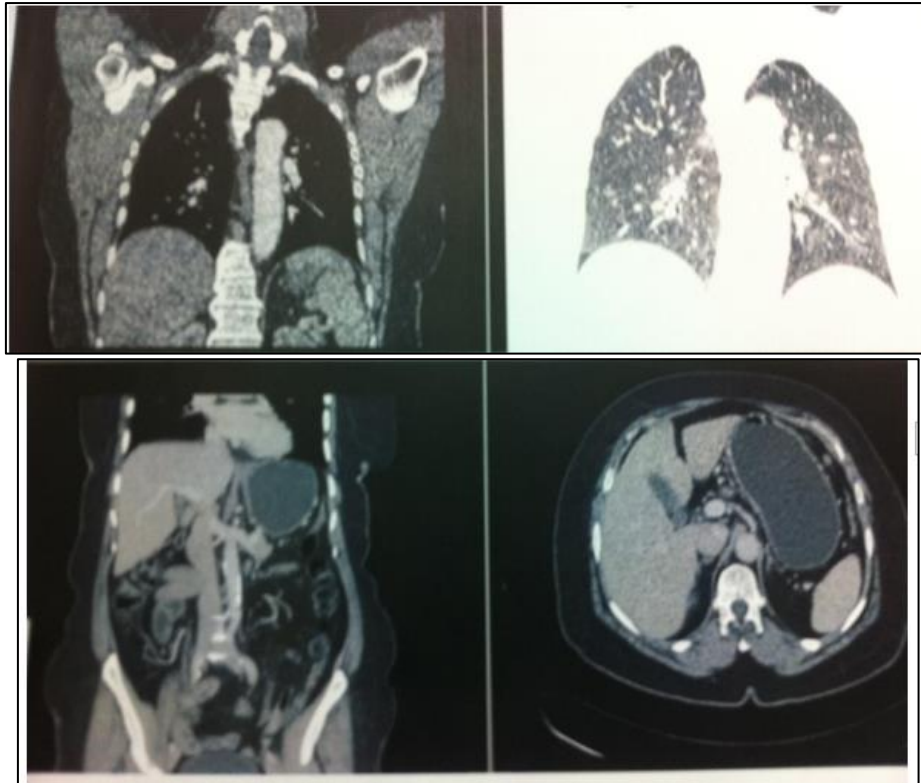


Figure 3: The extension assessment did not reveal any secondary location



Figure 4: Intraoperative image showing a macroscopic presentation of chondromatosis

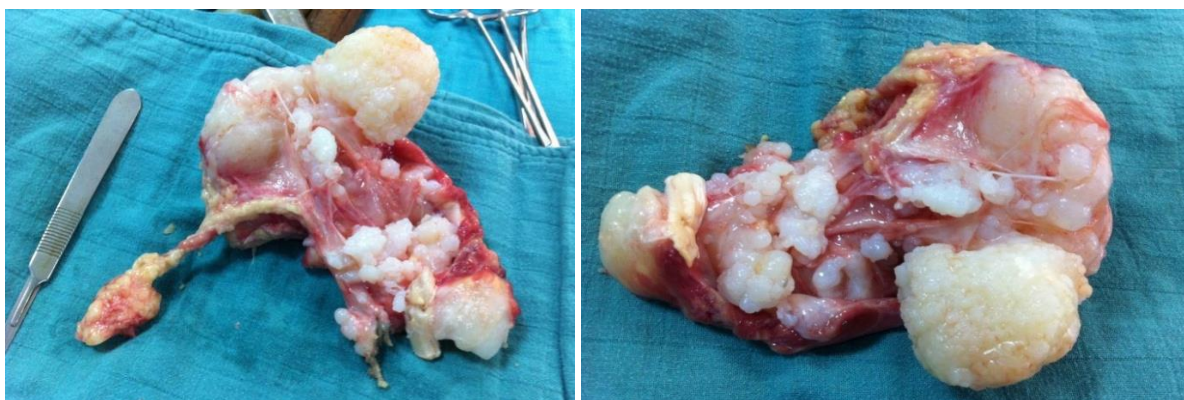


Figure 5: Surgical specimen after tumor resection

DISCUSSION

The aim of this study is to review the literature on synovial chondrosarcoma, a rare synovial tumor pathology. This review will focus on the epidemiological, clinical, paraclinical, evolutionary and

therapeutic aspects of this entity. The objective is to evaluate the persistent diagnostic difficulties despite the scientific advances in imaging and anatomopathology, and to compare our case with the data of the literature to improve our management of this pathology.

Primary synovial chondromatosis is a benign synovial dystrophy characterized by the formation of cartilaginous nodules (chondromas) that may ossify secondarily (osteochondromas) within a joint or, more rarely, a synovial lining. It corresponds to a metaplasia of the synovial membrane of unknown origin, producing chondromas that can pediculate and detach forming free foreign bodies in the joint cavity. It classically affects young adults between 20 and 50 years of age, and is more likely to be male [1]. The knee joint is the most frequent location, about 50% [2], followed by the hip, shoulder, elbow, ankle and finally the wrist. However, this synovial chondromatosis, which has long been considered benign, may be complicated by malignant degeneration into synovial chondrosarcoma, of which rare cases have been reported [3-6].

In this work, we outline the problem related to the diagnosis and therapeutic management of synovial chondrosarcoma based on the data in the literature. We identified 42 cases of synovial chondrosarcoma reported to date, the first in 1957 [7] and the last in 2011 [4]. All authors had to answer the same problematic question: how to differentiate synovial chondromatosis from synovial chondrosarcoma based on epidemiological, clinical, radiological, and anatomopathological criteria for a better therapeutic management [3-6,8,9,10]. A second concern of the authors was to distinguish primary synovial chondrosarcoma from synovial chondrosarcoma secondary to synovial chondromatosis, based mainly on evolutionary criteria [4,5,8,10]. This diagnostic problem remains a challenge despite scientific progress in imaging and anatomopathology.

The epidemiological study of synovial chondrosarcoma highlights several important elements. Of the 42 cases reported in the literature, the age of the patients ranged from 25 to 82 years, with an average age of 50 years. There is a predominance in individuals in the age range [11,12]. Men are more affected than women, with a sex ratio of 2.23. The most frequent location of this pathology is the knee (50%), followed by the hip (30.9%), ankle, shoulder, elbow and finally the wrist. This observation has been confirmed by several authors, in particular the series by Bertoni [3], which included ten cases and showed that five lesions involved the knee, three lesions the hip, one lesion the elbow and one lesion the ankle. The predominance of the knee location has also been noted by other authors [6].

The clinical study of synovial chondrosarcoma reveals that the initial symptoms vary from patient to patient. Of the 42 cases reported in the literature, 21 patients presented with different initial symptoms, such as pain, painful swelling, masses, and episodes of effusion. In a series of ten patients, Bertoni *et al* [3] observed that five patients complained of pain, three of them also had a mass, the fourth had stiffness and the fifth had swelling with effusion episodes. Two patients had painful swelling, while the other three cases have not

been described in the literature. The evolution of symptoms before the first consultation was noted in 15 cases, with a mean duration of five years, ranging from two months to 15 years [3,9]. However, these data do not allow differentiation between synovial chondromatosis and synovial chondrosarcoma, as these two conditions share a very similar epidemiological and clinical profile [3,6].

The distinction between synovial chondromatosis and synovial chondrosarcoma is of great importance because of their very different evolutionary and therapeutic modalities. Of the ten cases in the series by Bertoni [3], nine patients had tumor recurrence after local excision of the tumor. Eight patients underwent amputation and one patient underwent wide excision with arthroplasty. Despite these aggressive surgical measures, five patients developed lung metastases, four of whom died and only one survived after excision of the metastasis. The authors retained that inadequate surgical excision carries a high risk of local recurrence and subsequent metastasis [3,6]. The incidence of these metastases, especially pulmonary, can be as high as 29% [9,13]. In chondromatosis, treatment consists essentially of excision of the cartilage nodules with or without synovectomy. However, synovial chondrosarcoma hides a very important aggressive potential, requiring large resections, often leading to amputation [3,4,6,9]. It should be noted that 60% of patients with synovial chondrosarcoma eventually undergo amputation [4]. Therefore, most authors suggest a wide excision or even a radical treatment for a better local control of the tumor [3,9].

CONCLUSION

Our clinical case presents the profile of a primary synovial chondrosarcoma, a very rare tumor, which, according to the review of the literature, is the sixth reported case. However, the means of differential diagnosis, between synovial chondromatosis and synovial chondrosarcoma, mark several limits, which leaves the doors wide open for further investigations.

Ethics approval: Not required for case reports at our hospital. Single case reports are exempt from ethical approval in our institution.

Consent to participate: Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Availability of data and materials: The datasets used and analysed during the study are available from the corresponding author.

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