Pediatric Synovial Osteochondromatosis of the Knee with Leg Length **Discrepancy: A Case Report**

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Learning Point of the Article:

We should not forget a possibility of synovial osteochondromatosis accompanied by leg length discrepancy in the case of children.

Introduction: Synovial chondromatosis (SC) is very rare among children. We are aware of no reports of patients with SC accompanied by leg length discrepancy (LLD).

Case Report: We describe a case of synovial osteochondromatosis of a 7-year-old boy complicated by LLD. We performed epiphysiodesis of the distal femur and arthroscopic resection of loose bodies and total synovectomy. Three years after surgery, LLD had been corrected and there was no sign of recurrence.

Conclusion: Physicians should be aware of synovial osteochondromatosis complicated by LLD in childhood and take radiographs of the whole length of lower legs when this condition is suspected.

Keywords: Synovial osteochondromatosis, leg length discrepancy, knee joint, child.

Introduction

Synovial chondromatosis (SC) is an unusual condition characterized by the formation of multiple hyaline cartilage nodules in joints, tendon sheaths, and bursae [1]. It is reported that the knee is the most involved joint. X-ray shows no radiopaque bodies in 5-30% of SC cases [2]. In cases when calcification cannot be seen on radiographs, MRI is useful and typically shows hyperintensity masses on T2-weighted images in affected joints, which reflect chondroid tissues. SC usually occurs between the third and fifth decades with a minor male predominance and it is unusual among children [2]. This report describes a rare case of a 7-year-old boy with SC of the knee, accompanied by leg length discrepancy (LLD).

Case Report

Case: 7-year-old boy

Chief complaint: Limitation of knee joint range of motion

A 7-year-old boy was admitted to our hospital complaining of limited right knee flexion over a period of 6 months. He had swelling in his right knee, but no limping gait or tenderness. The right knee range of motion was limited to 0-120°. Radiographs revealed juxta-articular calcified or ossified loose bodies (Fig. 1). MRI demonstrated joint effusion and small loose bodies in the anterior part of the knee joint (Fig. 2). Although we were concerned about radiation exposure, we conducted computed tomography (CT) scans to examine if the masses were calcified

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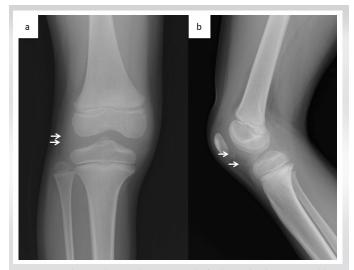


Figure 1: Plain radiographs. Some calcific loose bodies are visible in the lateral anterior knee joint (arrow). (a) Anteroposterior view and (b) lateral view.

and determine their exact location. CT scan revealed multiple subtle calcifications in the joint (Fig. 3). We diagnosed him with SC of the knee. We opted for monitoring due to his lack of severe pain.

When he was 10 years old, he returned to our hospital and radiographs showed a LLD of 2.2 cm (Fig. 4). The loose bodies had grown in both size and number. One year later, he developed severe right knee pain in addition to the 20° limitation in the right knee flexion. We decided to perform epiphysiodesis to treat the LLD and arthroscopic resection of the tumors.

We first performed the epiphysiodesis of his distal right femur using an eight-Plate® (Japan Medicalnext Co., Ltd., Osaka, Japan). Four months after the epiphysiodesis, we performed the



Figure 2: Magnetic resonance imaging (MRI). MRI reveals many loose bodies that signaled low in T1-weighted and T2-weighted images. (a) T1-weighted sagittal view and (b) T2-weighted sagittal view.

arthroscopic removal of the loose bodies. We found dozens of loose bodies in the affected joint. We performed total synovectomy and resection of the loose bodies with arthroscopy. Histopathological examinations of synovial tissues confirmed the diagnosis of SC (Fig. 5).

About 1 week postoperatively, the patient's knee pain was greatly improved. Right knee range of motion was improved to 0–130°. Two years postoperatively, his LLD had been reduced to only 0.4 cm (right: 74.6 cm, left: 74.2 cm). We, thus, decided to remove the plate. At final follow-up, there was no clinically significant LLD, and he showed no signs of recurrence (Fig. 6).

Discussion

SC usually occurs in a single major joint, most commonly the

Table 1: Past reported cases of knee pediatric synovial chondromatosis					
Authors (year)	Age	Sex	Chief complains	Examination	Management
Carey (1983) [4]	9	M	swelling	Plain radiograph	OS
Carey (1983) [4]	10	F	swelling	Plain radiograph	OS
Kistler (1991) [5]	12	F	Persistent arthritis	Plain radiograph	AS
Tiedjen et al. (2006) [6]	9	F	swelling	Plain radiograph and MRI	AS
Chou et al. (2007) [7]	14	F	Pain	Plain radiograph and MRI	AS
Kukreja (2013) [8]	16	M	pain, swelling	Plain radiograph and MRI	OS
Giancane et al. (2013) [9]	7	M	pain, swelling, limp	Plain radiograph and MRI	AS
Temponi et al. (2016) [10]	2	M	pain, lameness	Plain radiograph and MRI	OS+AS
Cho and Suh (2017) [11]	10	F	limited ROM, limp	Plain radiograph and MRI	AS
Pirimoglu <i>et al</i> . (2021) [12]	2	F	pain, lameness	Plain radiograph, MRI, and US	OS
Yothakol et al. (2022) [13]	12	F	Pain	Plain radiograph and MRI	AS
Present case	7	M	limited ROM, swelling	Plain radiograph, MRI, and CT	AS

ROM: Range of motion, MRI: Magnetic resonance imaging, US: Ultrasonography, CT: Computed tomography, OS: Open synovectomy, AS: Arthroscopic synovectomy



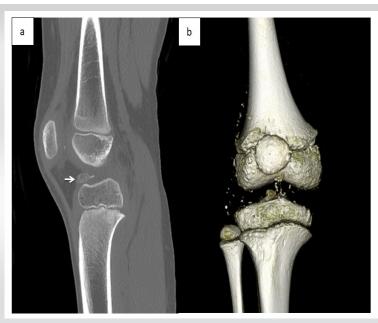


Figure 3: Computed tomography (CT) scan. CT shows numerous loose bodies in the knee joint. (a) Sagittal view and (b) three-dimensional-CT (3D-CT).

knee, followed by the hip and elbow. SC has an incidence of one per 100,000 individuals among the general adult population, and about two in three patients are male [3]. It is extremely rare in children. A search of peer-reviewed papers reviewed only 12 patients with

SC in childhood (Table 1) [4-13]. Open resection was performed in four of those cases, with arthroscopic surgery performed in the other nine. As of this writing, the authors are aware of no reports of SC accompanied by LLD. This is the first case report of SC with LLD in a pediatric patient.

LLD is a clinically important condition when it occurs in growing children. A wide range of causes have been reported, including congenital factors, vascular malformation, infection, tumors, and trauma. Importantly, inflammation often plays a role in LLD.

Simon et al. [14] reported that 35 out of 51 patients (69%) in monoarticular and pauciarticular juvenile rheumatoid arthritis had LLD of 1.5 cm or more. LLD was more frequent among patients who experienced disease onset before the age of nine. Moued et al. [1512] reported 37 patients affected with oligoarticular juvenile idiopathic arthritis (JIA). Of them, 9 patients (24.3%) were reported with LLD. One possible reason for this LLD is stimulation of the adjacent epiphyseal growth plate due to inflammation in the affected joint.

Intra-articular steroid injection has been reported as an acceptable treatment option in JIA [16-19]. Sherry et al. [20] reported that intra-articular steroid in jection (triamcinolone hexacetomide) was effective for the prevention

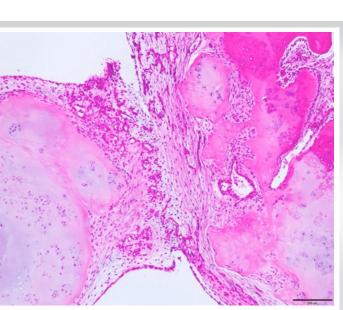


Figure 5: Microscopic image. (Hematoxylin-eosin staining) Pathological analysis shows multiple nodules consisting of hyaline cartilage surrounded by synovial tissues.



Figure 6: Full-leg radiograph at final follow-up. Leg length discrepancy had been largely eliminated 6 months after removal of the eight plates. There is no apparent sign of recurrence.



Figure 4: Full-leg radiograph.

Full-leg radiograph

demonstrated a leg length

discrepancy of 2.2 cm (right:

68.7 cm, left: 66.5 cm).

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of LLD in growing children with JIA. However, steroid injection into joints has some potential adverse effects such as skin atrophy, acute pain, irritation of the joint, and local calcification. Clinicians should be mindful of these potential risks when considering steroid injections.

Conclusion

Clinicians should be aware of SC as a potential, if rare, diagnosis, even among children. Before epiphyseal arrest, full-

leg radiographs should be used when LLD is suspected.

Clinical Message

We should be aware of synovial osteochondromatosis complicated by LLD in childhood and take radiographs of the whole length of lower legs when this condition is suspected.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil Source of support: None

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Consent: The authors confirm that informed consent was obtained from the patient for publication of this case report

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