Clinical Case Report

Medicine®

OPEN

Synovial chondromatosis of the hip joint in childhood

A case report and literature review

Jie Wen, MD, Hong Liu, MS, Sheng Xiao, MD*, Xin Li, MD, Ke Fang, MD, Zhongwen Tang, MD, Shu Cao, MS, Fanling Li, MS, Guangqing Cai, MS, Chen Hu, MS, BS

Abstract

Rationale: Pain in the hip joint is a common symptom in children. The common diseases leading to pain in the hip joint in children include transient synovitis of the hip, septic arthritis of the hip, and Legg–Calve–Perthes disease.

Patient concerns: A 7-year-old boy was admitted due to pain in the right hip joint and limping for more than 1 month.

Diagnosis: Synovial chondromatosis.

Interventions: The patient underwent a hip open surgery, all the loose bodies in articular capsule were removed.

Outcomes: At the 6-month follow-up, pain and limping disappeared, and the range of activity of the hip joint was restored to a normal level.

Conclusions: Synovial chondromatosis is an uncommon disease which can cause pain of hip joint in children.

Lessons: When the pediatric orthopedic surgeon treats the children suffered with hip pain the surgeon should be aware of this rare disease.

Abbreviations: CT = computed tomography, MRI = magnetic resonance imaging, SC = synovial chondromatosis.

Keywords: children, hip pain, synovial chondromatosis

1. Introduction

Clinically, pain in the hip joint is a common symptom in children. The common diseases leading to pain in the hip joint in children include transient synovitis of the hip, septic arthritis of the hip, and Legg–Calve–Perthes disease.[1] It can be usually diagnosed by a combination of disease history, characteristics of the pain, signs, as well as x-ray, B-ultrasound, and other auxiliary examinations. However, the pain in the hip joint due to unusual reasons in children should not be ignored.

Synovial chondromatosis (SC) is a chronic joint disease with cartilaginous nodes and joint bodies in the synovium caused by self-restricted hyperplasia of joint synovium. It is a benign lesion and can occur in any synovial joint. SC is more commonly found in 30- to 50-year-old male adults and has an incidence rate of 1/100,000, with a male-to-female ratio of about 1.8:1[2] or, more recently, thought to be 3:1.[3] The most common joints involved are knee joints (70%), hip joints (20%), and elbow joints (7.9%); 33 parts have been reported to be possibly involved in SC.[4]

This study reported the case of a 7-year-old child who mainly complained of pain in the hip joint, and was surgically and pathologically confirmed to be suffering from SC. It is the youngest child with SC of the hip joint reported in the English literature.

2. Case report

The patient was a 7-year-old boy who was admitted due to pain in the right hip joint and limping for more than 1 month. He did not have a history of obvious trauma before the onset of the disease. The physical examination revealed a limp in the right lower limb; swelling in the right hip joint; groin folds asymmetry; moderate skin temperature; local tenderness; limited intorsion, extorsion, and flexion in the right hip joint; and Allis sign (+), Thomas sign (+), and Patrick sign (+) on the right side. B-ultrasound revealed effusion in the right hip joint, x-ray film revealed the widening of the right hip-joint spacing (Fig. 1), computed tomography (CT) showed the destruction of the right acetabulum inner wall (Fig. 2), and magnetic resonance imaging (MRI) showed a large amount of effusion in the right hip joint with abnormal signals (Fig. 3). For the surgery, the patient was placed in a supine position; a Bikini incision was made on the right side; and the rectus femoris was separated, cut, and flipped down to expose the articular capsule, which was then cut open to reveal the overflow of a number of white semitranslucent circular loose

Editor: N/A.

Ethics approval and consent to participate: The study was done after agreement from the local ethics committee and with the patients' informed consent.

Written informed consent for publication of their clinical details and/or clinical images was obtained from the parent of the patient.

The authors have no conflicts of interest to disclose.

Department of Pediatric Orthopedics, Hunan Provincial People's Hospital, the First Affiliated Hospital of Hunan Normal University, Changsha, Hunan, China.

** Correspondence: Sheng Xiao, Department of Pediatric Orthopedics, Hunan Provincial People's Hospital, the First Affiliated Hospital of Hunan Normal University, Changsha 410005, Hunan, China (e-mail: 153816333@qq.com).*

Medicine (2018) 97:51(e13199)

Received: 9 July 2018 / Accepted: 16 October 2018

http://dx.doi.org/10.1097/MD.0000000000013199

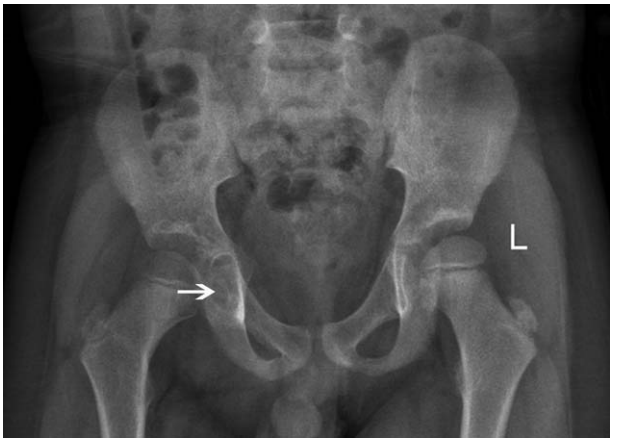

Figure 1. X-ray film revealed widening of the right hip-joint space.

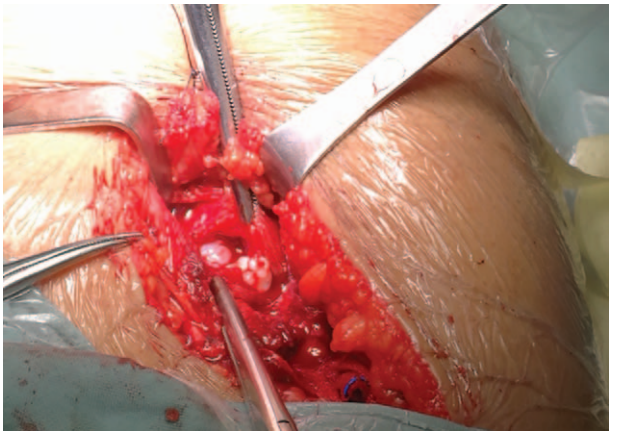

Figure 4. Overflow of a number of white semitranslucent loose bodies was visible when the articular capsule was cut open intraoperatively.

bodies (Fig. 4). Also, the articular cavity was filled with loose bodies. A bone defect of about $2 \times 2\,cm^2$ was detected in the inner acetabular wall, the lower part of the articular capsule was ruptured, and the synovial tissue was hyperplastic. The loose bodies were of different sizes, white or milky white, smooth and semitranslucent (Fig. 5), tough, cartilage-like, and elastic, a few of which were pedicled and connected with the synovium. A translucent and clear fluid was visible after the loose bodies were cut open (Fig. 6). Postoperative pathology confirmed SC (Figs. 7 and 8) and the patient was finally diagnosed as primary SC. At the 6-month follow-up, pain and limping disappeared, and the range of activity of the hip joint was restored to a normal level.

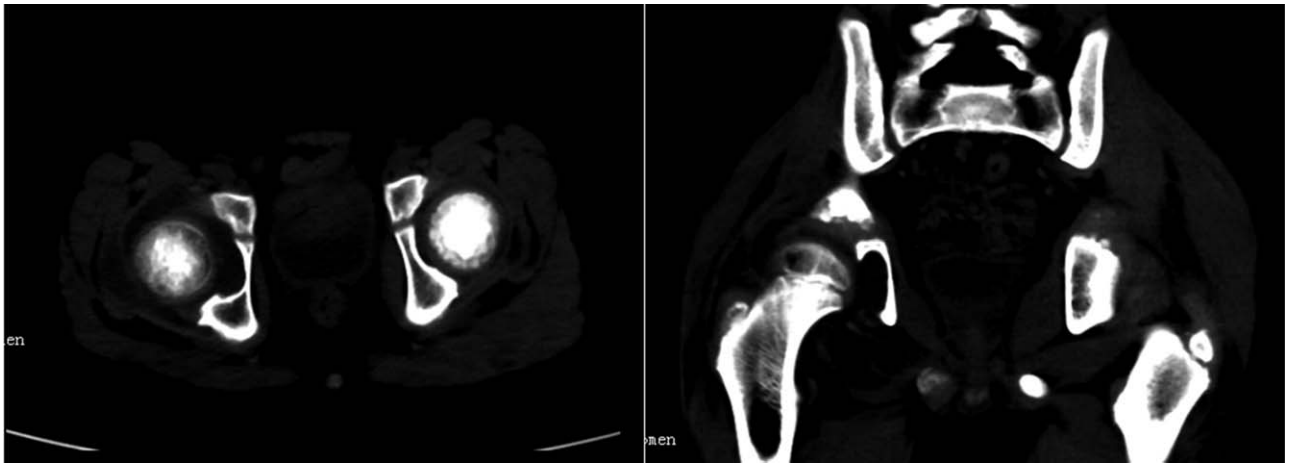

Figure 2. CT showed destruction of the right hip-joint bone. CT = computed tomography.

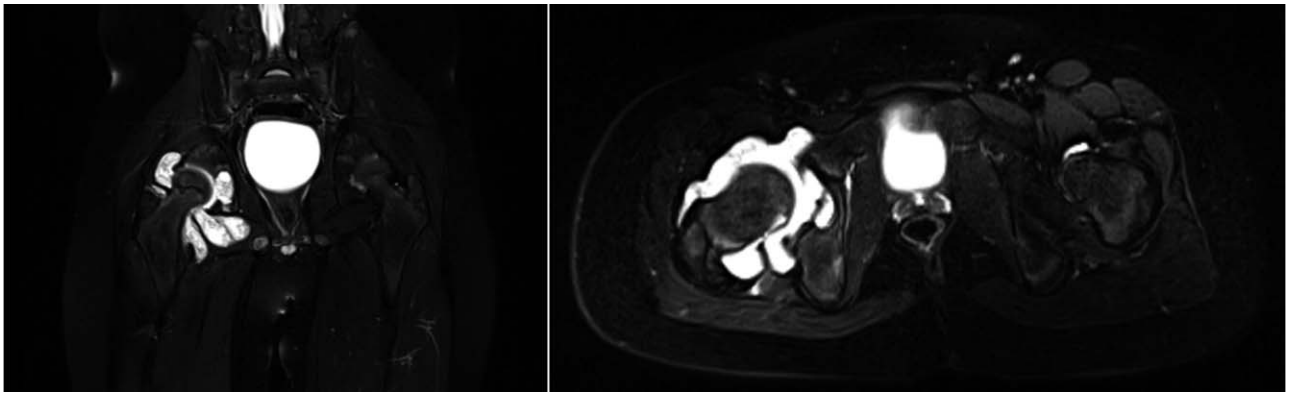

Figure 3. MRI showed a large amount of effusion in the right hip joint and abnormal signals. MRI = magnetic resonance imaging.

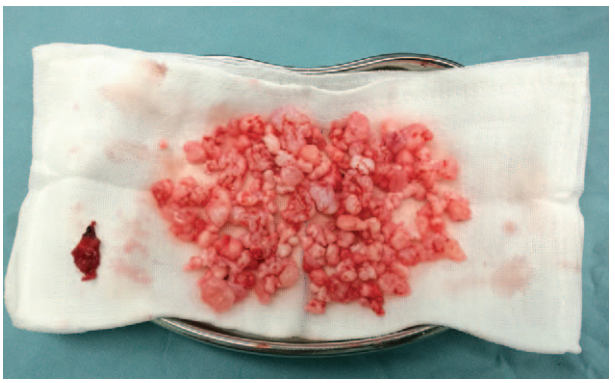

Figure 5. Intraoperatively, loose bodies were found to be of different sizes, white or milky white, smooth and semitranslucent, tough, cartilage-like, and elastic.

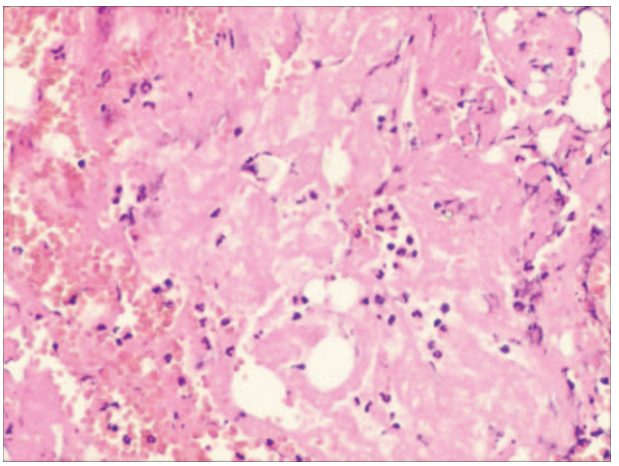

Figure 7. Hematoxylin and eosin staining showed a large number of chondrocytes. ×100.

3. Discussion

In 1558, SC was first reported by Ambrose Pare.[5] In 1813, Leannac put forward that the pathologic change is due to synovium-derived intra-articular bodies.[6,7] SC can be classified into primary and secondary categories. The primary category is in the majority with no history of osteoarticular basic diseases, while the secondary category is mostly secondary to the already occurred pathological changes, such as osteoarthritis, rheumatoid arthritis, and bone degeneration.[3] It has been reported that the primary SC has a higher risk of recurrence.[8]

Many scholars believe that loose bodies are formed due to synovial metaplasia. The pathological change is metaplastic synovium and the nodular processes on its surface is detached to form loose bodies,[9] which are nourished by the joint fluid and can be integrated. Synovial pathology reveals angiogenesis, lymphocyte infiltration, and metaplasia of synovial fibroblasts into chondrocytes.[10] In 1977, Milgram pathologically classified the disease into 3 phases according to the histological studies of SC: phase I (active intrasynovial disease only, with no loose bodies) referred to synovial metaplasia without loose bodies; phase II (transitional lesions with both active intrasynovial proliferation and free loose bodies) referred to synovial metaplasia with loose bodies; and phase III (multiple free osteochondral bodies with no demonstrable intrasynovial disease) referred to the presence of loose bodies and absence of synovial metaplasia.[11]

Since the incidence of SC is low in adults and even less in children, this report was presented in the form of case study. In 1983, Carey[12] reported 2 cases, aged 9 and 10 years, with SC in the knee joint, both of them underwent synovectomy and recovered well with the disappearance of symptoms postoperatively. In the same year, Pelker et al[13] reported a case of an 11-year-old boy who underwent removal of SC via arthrotomy of hip and showed good results without recurrence during a 3.5-year follow-up. In 1991, Kistler[14] reported the arthroscopic removal of loose bodies from the knee joint of a 12-year-old patient, but did not include the follow-up results. In 2006, Tiedjen et al[15] reported a case of a 9-year-old patient who underwent arthroscopic treatment, but they also did not include the follow-up results. In 2014, Raza et al[16] reported arthroscopic synovectomy in a 12-year-old girl. However, they believed that the excision was not extensive and open synovectomy was not performed because it might lead to avascular necrosis. In 2017, Manesh and Sajeer[17] reported a 7-year-old female child who suffered from SC treated by arthrotomy through an anterolateral approach. In the present study, the patient was a 7-year-old boy, the youngest child with SC of the hip joint reported in the English literature similar to the

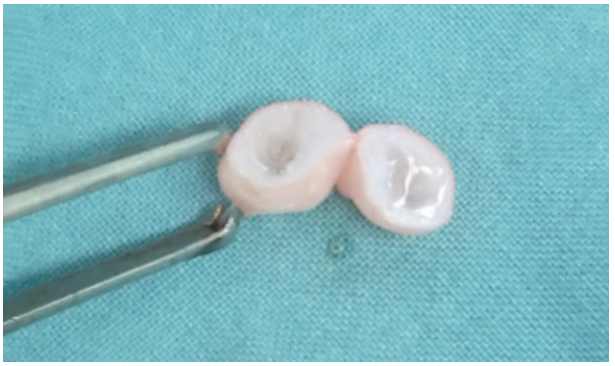

Figure 6. Translucent and clear fluid was visible when a loose body was cut open.

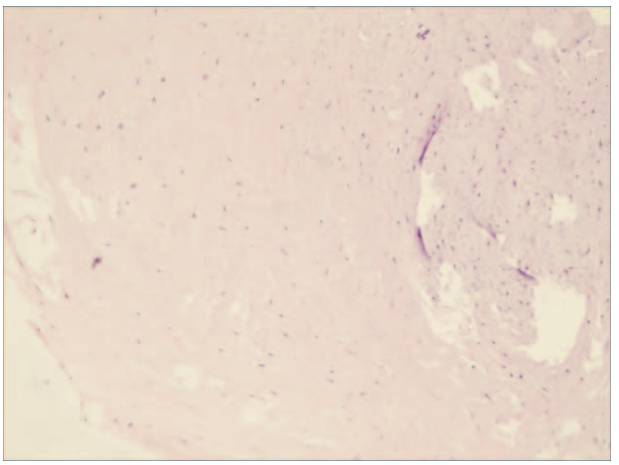

Figure 8. Loose bodies consisted of a large number of chondrocytes. ×100.

Table 1

Concise overview of case reports of synovial chondromatosis of pediatric case in literature.

Author	Year	Location	Case number	Case age
Carey[12]	1983	Knee	2	9,10
Pelker[13]	1983	Knee	1	11
Kistler[14]	1991	Knee	1	12
Tiedjen[15]	2006	Knee	1	9
Raza[16]	2014	Hip	1	12
Manesh[17]	2017	Hip	1	7

case from Manesh and Sajeer. The child underwent the open removal of hip synovium and loose bodies, and avascular necrosis was not reported during a 6-month follow-up. Literature search on PubMed and Medline has revealed only few reported cases of SC in children (Table 1).

Distinguishing SC from the common pain in the hip joint in children, such as transient synovitis and Legg–Calve–Perthes disease, is important for accurate diagnosis. It is usually difficult to distinguish SC from the transient synovitis in x-ray films because the intra-articular loose bodies are basically cartilage, which contains fluid and is not significantly calcified (Fig. 6), in children with SC. The child in the present case study was admitted to the hospital and diagnosed with transient synovitis, after visiting several hospitals. He did not recover any better after receiving skin traction treatment, and his CT scan revealed that the inner acetabular wall was squeezed by the loose bodies, which appeared as skeletal destruction (Fig. 2). MRI showed a large number of fluid signals (Fig. 3), making it difficult to distinguish the symptoms from the effusion of hip joint. Therefore, it is recommended to treat children with pain in the hip carefully to avoid misdiagnosis and Synovial chondromatosis of hip should be considered a differential diagnosis in a child with hip pain.

Author contributions

Jie Wen, Hong Liu, Sheng Xiao, and Xin Li were the surgeons who did the operation, Ke Fang and Zhongwen Tang did the article search on PubMed, Shu Cao and Fanling Li collected the radiation data and edited the photos. This article was written by Jie Wen and modified by Sheng Xiao. All the authors read and approved the final manuscript.

Conceptualization: Jie Wen.
Data curation: Hong Liu, Fanling Li.
Formal analysis: Guangqing Cai.
Investigation: Sheng Xiao.
Methodology: Xin Li.
Project administration: Ke Fang.
Resources: Zhongwen Tang.
Software: Shu Cao.
Writing – original draft: Chen HU.

References

[1] Cook PC. Transient synovitis, septic hip, and Legg-Calvé-Perthes disease. Pediatr Clin North Am 2014;61:1109–18.
[2] Jimenez-Martin A, Zurera-Carmona M, Santos-Yubero FJ, et al. Arthroscopic treatment of synovial chondromatosis, an unusual cause of shoulder pain. Reumatol Clin 2014;10:416–7.
[3] Ozmeric A, Aydogan NH, Kocadal O, et al. Arthroscopic treatment of synovial chondromatosis in the ankle joint. Int J Surg Case Rep 2014;5:1010–3.
[4] Davis RI, Hamilton A, Biggart JD. Primary synovial chondromatosis: a clinicopathologic review and assessment of malignant potential. Hum Pathol 1998;29:683–8.
[5] Barwell R. Clinical lectures on movable bodies in joints. Br Med J 1876;1:403–5.
[6] Evans S, Boffano M, Chaudhry S, et al. Synovial chondrosarcoma arising in synovial chondromatosis. Sarcoma 2014;2014:647939.
[7] Halstead AEIV. Floating bodies in joints. Ann Surg 1895;22:327–42.
[8] Peh WC, Shek TW, Davies AM, et al. Osteochondroma and secondary synovial osteochondromatosis. Skeletal Radiol 1999;28:169–74.
[9] Alexander JE, Holder JC, McConnell JR, et al. Synovial osteochondromatosis. Am Fam Physician 1987;35:157–61.
[10] Marchie A, Panuncialman I, McCarthy JC. Efficacy of hip arthroscopy in the management of synovial chondromatosis. Am J Sports Med 2011;39 (suppl):126S–31S.
[11] Milgram JW. Synovial osteochondromatosis: a histopathological study of thirty cases. J Bone Joint Surg Am 1977;59:792–801.
[12] Carey RP. Synovial chondromatosis of the knee in childhood. A report of two cases. J Bone Joint Surg Br 1983;65:444–7.
[13] Pelker RR, Drennan JC, Ozonoff MB. Juvenile synovial chondromatosis of the hip. A case report. J Bone Joint Surg Am 1983;65:552–4.
[14] Kistler W. Synovial chondromatosis of the knee joint: a rarity during childhood. Eur J Pediatr Surg 1991;1:237–9.
[15] Tiedjen K, Senge A, Schleberger R, et al. Synovial osteochondromatosis in a 9-year-old girl: clinical and histopathological appearance. Knee Surg Sports Traumatol Arthrosc 2006;14:460–4.
[16] Raza A, Kailash K, Malviya A. Rare cause of hip pain in a young girl. BMJ Case Rep 2014;2014: Pii: Bcr2014203802.
[17] Manesh CP, Sajeer U. Synovial chondromatosis: a rare differential diagnosis of hip pain in a child. J Orthop Case Rep. *2017;7:37–39.*