Primary Intra-articular and Extra-articular Synovial Chondromatosis in a Child: A Rare Cause of Shoulder Pain in Children

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ABSTRACT
Synovial chondromatosis is a rare condition with chondroid metaplasia of the synovial membrane and multinodular proliferation of the synovial membrane of the joints, tendons and bursae without any certain etiology. The disease generally presents in monoarticular form; and the most commonly involved joint is the knee. It commonly occurs in third and fifth decades of life. Synovial chondromatosis of the shoulder in children is extremely rare; and up to now, only two cases have been reported in the literature. Herein, we report synovial chondromatosis of the shoulder joint in a child with both intraarticular and extraarticular involvement. To the best of the authors' knowledge, this is the first case in the literature with both intra- and extra-articular involvement of synovial chondromatosis of the shoulder joint in children.

Key Words: Synovial chondromatosis, Metaplasia, Children.

INTRODUCTION
Synovial chondromatosis is a rare condition with chondroid metaplasia of the synovial membrane and multinodular proliferation of the synovial membrane of the joints, tendons and bursae without any certain etiology. The nodules may enlarge and detach from the synovium. The disease generally presents in monoarticular form and the most commonly involved joint is the knee (50-65% of the cases), followed by hip. Shoulder involvement is rare with 5% occurrence rate. It commonly occurs in third and fifth decades of life and twice common in male population. Synovial chondromatosis of the shoulder in children is extremely rare; and up to now, only two cases have been reported in the literature.

Herein, we report synovial chondromatosis located in the shoulder joint of a child with both intraarticular and extraarticular involvement. To the best of the authors’ knowledge, this is the first case in the literature of synovial chondromatosis with both intra- and extra-articular involvement in a child.

CASE REPORT
A 17-year boy was admitted to orthopaedic outpatient clinic with a 6-month history of the left shoulder pain and restriction in the shoulder movements. In history taking, the patient stated that the pain increased gradually and no shoulder trauma and swelling or bruising of the shoulder has happened as he remembered. The pain was localised to superior and anterior aspects of the glenohumeral joint. There was no remarkable deformity around the left shoulder on inspection. On physical examination, there was decreased range of motion with limitation in active and passive abduction over 90° and internal rotation. The blood tests were normal and the family history was not remarkable. X-Ray of the shoulder showed multiple well defined osseous intra- and extra-articular loose bodies (Figure 1a). Computerised tomography (CT) revealed multiple calcified fragments in the same location (Figure 1b). There was no remarkable extrinsic bone erosion. Magnetic resonance imaging (MRI) showed multiple loose bodies within the glenohumeral joint and also the subacromial bursa, around the long tendon of the biceps, and around the rotator cuff muscle tendons. The fragments were hypointense on T1-weighted MRI and hyperintense on T2-weighted MRI with hypointense areas inside that reflected the high water content of the cartilaginous lesions with accompanied inner calcific areas (Figure 2). The radiologic findings were pathognomonic for primary synovial chondromatosis and the patient was referred to orthopedic tumor surgeons for arthroscopy to remove the loose bodies with concomitant synovectomy.
Milgram divided the disease into three phases. In phase one, typically gradually increase during the course of the disease. include pain, swelling, and restriction of the range of motion and point was the distinctive feature of our case. Clinical symptoms in intra- and extraarticular involvement of the shoulder joint. This particular synovial chondromatosis of the biceps tendon sheath in a 12-year-old boy.

The treatment of choice for synovial chondromatosis is surgical resection. Traditional treatment for synovial chondromatosis is open arthrotomy, synovectomy and complete removal of the free fragments. However, recent advances in arthroscopic techniques and methods has led to widening of the indications for arthroscopic treatment. Open and closed arthroscopic methods can be used in the treatment of synovial chondromatosis. Synovectomy and excision of the chondral bodies are regarded as the optimal treatment. Extra-articular involvement is important to detect in imaging during preoperative evaluation because it cannot be treated through arthroscopic techniques. In recurrent disease, which has been noted in 3-23% of cases because it cannot be treated through arthroscopic techniques.

In conclusion, it is important that radiologists and orthopedic surgeons keep PSC in mind while approaching synovial joint pain in children. Preoperative detailed radiological evaluation and detection of the any intra-articular, bursal and peritendineal chondroid bodies are crucial to direct the orthopedic surgeon during operation. Additionally, patients should be informed well about the possible recurrence and malignant transformation risks and should be followed accordingly.

**PATIENT’S CONSENT:**
Written informed consent was obtained from the patient and the family to report this case study.

**CONFLICT OF INTEREST:**
The authors declared no conflict of interest.

**AUTHORS’ CONTRIBUTION:**
BAU, CS: Involved in the conception, design and interpretation, wrote the manuscript and collected data, reviewed relevant published reports and provided the images. Read and approved the final manuscript.
REFERENCES


