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A case of synovial chondromatosis of the knee with 87 free bodies and review of literature

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Abstract. – BACKGROUND: Synovial chondromatosis is a non-malignant synovial disorder characterized by the presence of cartilage formation within the synovial membrane, leading to the emergence of multiple cartilaginous nodules that may be either attached or unattached. The presence of this anatomical feature is frequently observed in articulations such as the knee, hip, elbow, and ankle.

CASE REPORT: In this study, we present a case of synovial chondromatosis in the knee joint of a healthy male in his early 60s. Notably, the patient exhibited the simultaneous presence of 87 large loose bodies. The occurrence of a substantial quantity of unattached entities of notable dimensions within the joint is highly uncommon.

CONCLUSIONS: The patient had several synovial chondromas, a rare disease. Synovial chondromatosis is a benign disorder; however, growing synovium can cause pyogenic cartilage nodules. Most loose bodies in joints can abrade and degenerate articular cartilage, causing long-term discomfort. Thus, an early-stage procedure to remove loose bodies and carefully excise synovial tissue is necessary to treat this condition.

Key Words:
Synovial chondromatosis, Case report, Multiple, Self-limiting proliferative disorder, Open surgery.

Background

Synovial chondromatosis, first proposed by Ambrose Paré in 1558, was further described by Lannec in 1813, and classified as a tumor by Brodie in 1846[1,2]. It is a benign monoarticular disease of unknown etiology characterized by benign proliferation of synovium, leading to the formation of numerous cartilaginous nodules. When these nodules increase and become localized within the joint space, they cause extensive erosion of adjacent tissues, potentially resulting in intra-articular and extra-articular damage. The knee joint is the most commonly affected large joint[1,2]. The pathogenesis of synovial chondromatosis is not yet clear. Currently, a study[3] on the pathophysiology suggests that it is the result of abnormal transformation of macrophage-like synovial cells into anti-inflammatory cells and induces continuous excessive differentiation of mesenchymal stem cells into chondrocyte-like synovial cells. Synovial chondromatosis is a synovial disease where cartilaginous nodules form as outgrowths from the synovium within the joint in the early stages of the disease. These cartilaginous nodules float freely in the joint cavity and eventually detach from the synovium, known as "loose bodies". We hereby report a case of extremely rare knee joint synovial chondromatosis treated with open surgery, accompanied by 87 loose bodies.

Case Presentation

Clinical Present Illness Data

The patient under consideration was a male individual in his early sixties who was engaged in the occupation of farming. The patient presented with unexplained swelling approximately 13 years ago, which subsequently progressed in severity over time. Nevertheless, the individual did not seek any form of medical intervention. A decade ago, the individual received a diagnosis of synovial chondromatosis in his right knee at a nearby healthcare facility, subsequently undergoing excision surgery. A reoccurrence of the condition was observed three years after the surgical intervention. The patient underwent knee joint aspiration and ozone therapy; however, treatment was subsequently discontinued. The patient experienced a progressive exacerbation of pain and swelling, leading them to pursue a thorough evaluation and intervention. He started encountering discomfort and inflammation in the right knee. The act of walking presented considerable difficulty, while the act of squatting posed

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a notable challenge. The individual exhibited a typical level of hunger and a regular sleep-wake cycle, and there were no issues with urination or bowel movements.

Specialist Examination and Imaging Data

The presence of a right knee valgus deformity and swelling was apparent based on a comprehensive clinical assessment. The surgical scar was visible on the anterior lateral aspect of the knee joint. The knee joint demonstrated audible snapping sounds during the processes of flexion and extension, encompassing a range of motion spanning from 0 to 90°. The pain was a consequence of excessive flexion. An absence of tenderness was observed in both the medial and lateral compartments. However, the patellar grind test yielded a positive outcome. According to the radiographic observations, the right knee exhibited numerous opacities resembling masses characterized by irregular calcifications (Figure 1). Notably, a distinct periosteal reaction was absent. The computed tomography (CT) scan revealed the presence of several rounded to oval opacities of bony density within the right knee. These opacities exhibited heterogeneous internal density and were characterized by distinct and well-defined borders (Figure 2 a-d).

Diagnosis, Treatment, and Surgical Procedure

The obtained diagnosis was synovial chondromatosis of the right knee. Based on the patient's condition, it was decided to perform open surgical excision for the patient.

After successful lumbar epidural anesthesia, the patient was placed in a supine position. The area was prepared with routine disinfection and draping. An inflatable tourniquet (ATS-I, Shanghai, China) was applied. A midline incision of approximately 16 cm was made in the anterior aspect of the right knee joint. The incision sequentially went through the skin, subcutaneous tissue, and deep fascia, and the joint capsule was opened medially to the patella. The patella was dislocated laterally to expose the joint. During the surgery, a large number of porcelain-white, variably sized, pebble-like, and separated cartilaginous nodules are observed in the suprapatellar pouch and joint cavity (Figure 3 a-d). There were 87 nodules, with the smallest one having a diameter of approximately 1 cm and the largest one having a diameter of approximately 1 cm (Figure 4 a-d). All the cartilaginous bodies were removed, and the proliferative and congested synovial tissue was cleared. The excised tissue was sent for pathological examination. The postoperative pathological report indicated synovial chondromatosis (Figure 5).

Discussion

Synovial chondromatosis is a benign joint lesion; the chance of malignancy is small, and it is mainly characterized by the synovial membrane. Joint capsule, tendon sheath cartilage nodule hy-

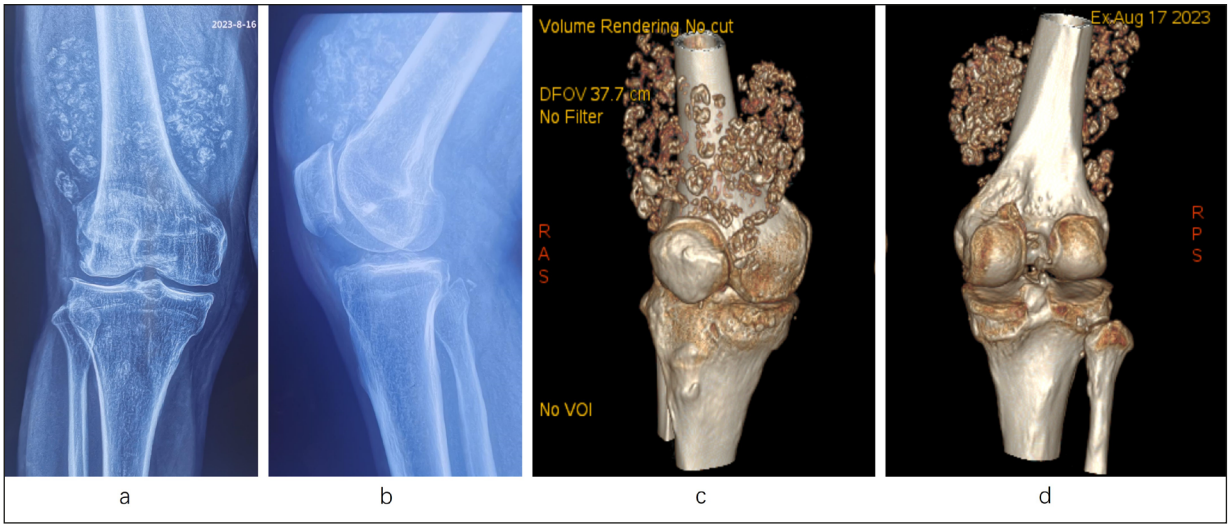

Figure 1. a, AP view of the knee joint; (**b**) lateral view of the knee joint; (**c**) 3D anterior view of the knee joint CT; (**d**) 3D posterior view of the knee joint CT.

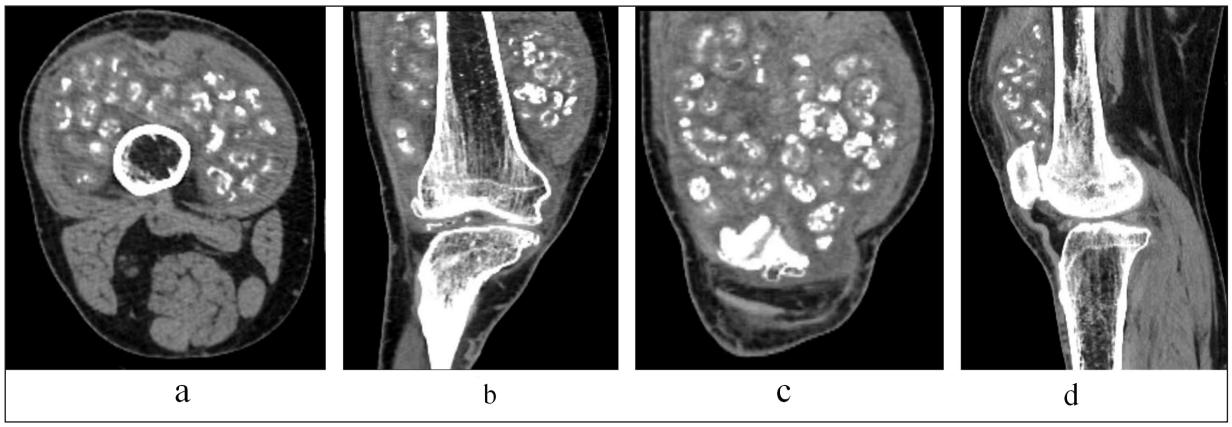

Figure 2. **a**, Axial view of the knee joint CT; (**b-c**) coronal view of the knee joint CT; (**d**) sagittal view of the knee joint CT.

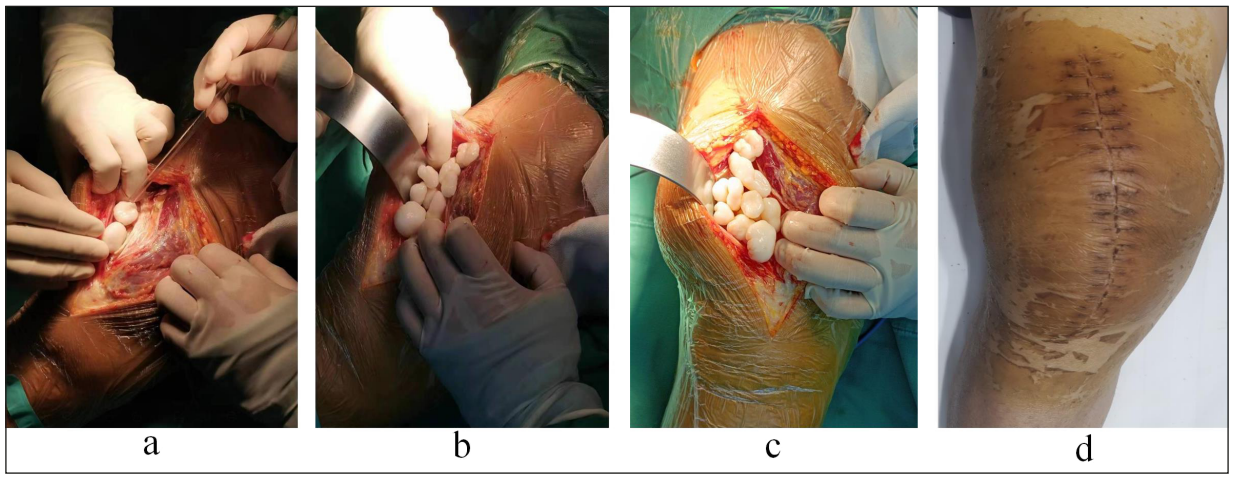

Figure 3. Intraoperative and postoperative incision data. **a-c**, intraoperative incision and detached body images; (**d**) postoperative incision image

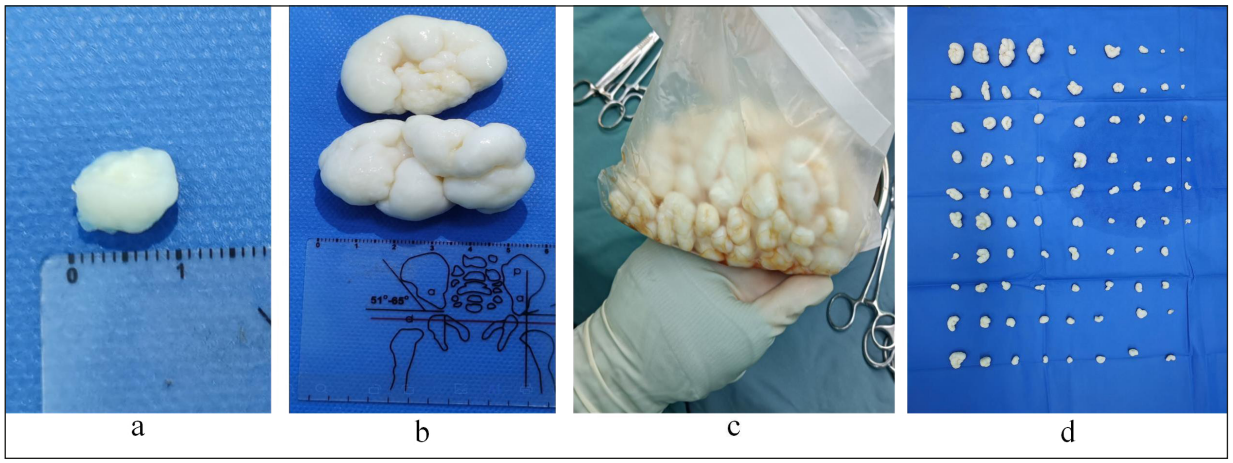

Figure 4. Detached body data. **a**, The minimum diameter of the detached body is approximately 1 cm; (**b**) the maximum diameter of the detached body is approximately 5 cm; (**c-d**) 87 images of detached bodies.

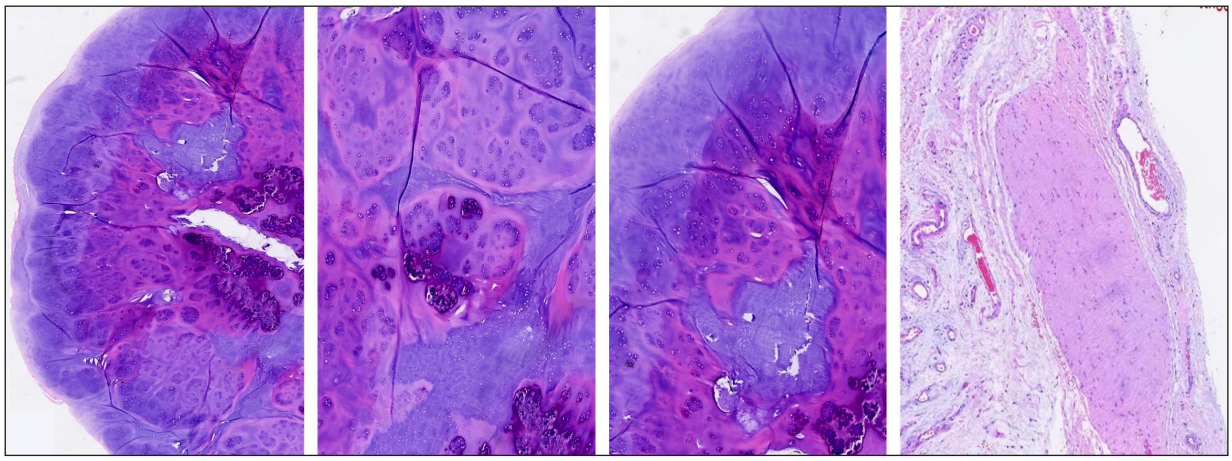

Figure 5. Pathological data. Pathology report: chronic inflammation of the synovial tissue of the knee joint, circular cartilage islands visible in the connective tissue, some nodules protruding from the synovial surface, and the presence of detached bodies. Considering the clinical context, consistent with synovial chondromatosis. From left to right, the magnification of the four images is, respectively, 2x, 10x, 4x, and 2x.

perplasia, cartilage nodule size and shape similar to the synovial membrane can be attached to the synovial membrane or detached from it and free from the joints; they can be single or multiple, and the number can be up to several thousand, mostly occurring in the single joints (occurrence in multiple joints is rare), with the knee joints having the highest incidence, followed by the hip, shoulder, elbow, and other parts of the body[4-6]. It is a rare benign tumor, with an incidence rate of approximately 1.8 cases per million person-years. It mainly occurs in adults aged 30-50, with a male-to-female ratio of about 2:1[7].

The etiology and pathogenesis of the disease are still not well understood. Synovial chondromatosis is a type of synovial connective tissue disorder with an uncertain cause. The primary contributing variables are typically trauma, dysfunction, inflammation, and infection. The etiology of synovial chondroma primarily stems from the following four theories:

1. Trauma theory refers to the excessive growth of synovial cartilage following an injury.
2. The synovial chemotaxis theory suggests that synovial chondrocytes migrate towards cartilage or clusters of cartilage.
3. The aberrant theory suggests that the synovium undergoes a process called 'adult differentiation' when embryonic cells transform into cartilage.
4. The theory of tumors: the tumor is currently considered benign and self-limiting. However, the notion of tumors has been increasingly discarded[8,9].

The most probable pathogenic basis for this condition is currently synovial metaplasia, as synoviocytes possess robust proliferative capacity and the potential to exhibit osteoblastic function. When exposed to various stimuli, the synovial membrane, synovial bursa, or tendon sheath undergo a transformation of their connective tissue, resulting in the thickening of the synovial membrane and the formation of nodules. These nodules continue to grow or detach within the joint cavity, gradually developing into chondrocytes or bone tissues. The pathological manifestation of synovial membrane transformation is observed in the articular capsule, synovial bursa, and tendon sheath. Fibroblasts create hyaline cartilaginous nodules, which are connected to the synovial membrane through broad bases or tips. Fibroblasts generate hyaline cartilaginous nodules that are linked to the synovium through a wide base or tip, known as the suspensory body. When the tip of an object breaks off and the nodule is separated from the space within a joint, it becomes a free body directly supplied with nutrients by the synovial fluid. The shape and number of free bodies, as well as the degree of calcification and ossification, are closely related to the progression of the disease. As the disease progresses, the number and size of the free bodies increase, along with the severity of calcification or ossification[10]. The pathophysiology involves hyperplastic synovium-coated lobulated, bluish-white hyaline cartilaginous nodules on the whole articular surface. A lot of these nodules provide a "cobblestone" effect. Lobulated cartilag-

inous nodules in sub-synovial tissue may damage soft tissues, bursa, or bone. Intra-articular focal lesions are rare and controversial. The extra-articular kind resembles the intra-articular type but can spread to the bursa and tendon sheath. Subsynovial hyaline cartilage nodules can exist independently from the synovium in the joint cavity, bursa, or tendon sheath. Nodules can reattach to synovium, resorb, or refloat. The cartilage nodule grows because synovial fluid feeds hyaline cartilage. Consistency in cartilage nodule size and shape, from a few millimeters to several centimeters, often suggests synchronous lesions. From few to many, cartilaginous nodules vary in number. A huge globular sign, a 20-cm-diameter spherical structure, can occur when many cartilaginous nodules fuse. According to Milgram's study[11], the course of synovial chondromatosis can be divided into three stages: Stage I – the active phase of synovial lesion, where synovial chondral bodies are formed within the synovium, without the production of loose bodies; Stage II – the transitional phase of synovial lesion, where synovial proliferation and metaplasia gradually evolve into loose bodies; Stage III – the static phase of synovial lesion, where the tumor detaches to form a large number of loose bodies and gradually undergoes calcification. If the loose body mechanically impinges or wears down the joint for a prolonged period, it can lead to the clinical manifestations of arthritis. The typical clinical manifestation is the presence of a hard, loose body moving within the joint cavity, accompanied by joint locking. Other clinical manifestations include joint swelling, pain, tenderness, crepitus, giving way of the leg, limited range of motion in the knee joint, and sometimes palpable mobile hard masses within the joint[12]. The specialized examination of this case effectively demonstrated these characteristics.

In this study, it was found that the specific manifestations and severity of patients are related to the location, number, and size of synovial nodules and loose bodies, combined with patient symptoms. Patients often seek medical attention due to knee joint pain, locking, and limited mobility caused by loose bodies. Most patients are classified as Milgram III stage when they seek medical attention. X-ray is the most commonly used diagnostic tool for this disease. Calcified loose bodies or synovial nodules in the joint can appear as round or oval shapes with lighter density in the center and higher density in the surrounding area on the lateral view of the knee joint X-ray. The nearby bone may even show signs of destruction due to compression from the tumor. X-ray findings are significant for the diagnosis of Milgram III stage patients, and they are related to the degree of calcification of the cartilaginous tumor. From simple joint effusion to multiple high-density joint bodies, they are usually small in size and consistent in size. Due to incomplete calcification of the tumor, X-rays can only show irregular high-density shadows, making it difficult to detect and diagnose the disease early. In the later stages of the disease, the increased size of the abnormal growth can make it difficult to distinguish the boundary between the bone and the new formation on X-rays. This can lead to confusion between synovial chondromatosis and malignant soft tissue tumors like synovial sarcoma or peripheral chondrosarcoma. As a result, additional CT or MRI scans are necessary[13-16]. CT is superior to regular X-rays as it provides a clearer observation of the position, size, morphology, joint effusion, and early changes in bone and joint diseases of free bodies. It can provide a definite diagnosis for typical cases. On the other hand, MRI has high soft tissue signal resolution and better displays thickening of synovium, effusion, and early changes in bone and joint diseases caused by cartilaginous free bodies[17,18]. MRI can display knee joint effusion, prominent synovial hyperplasia, extensive cystic low-density shadows inside the joint cavity, irregular morphology, uneven high-density shadows surrounding the outside, and calcifications visible within the cyst. The lesions appear as low-density shadows in T1 images and high-density shadows in T2 images. Therefore, MRI can detect knee joint synovial chondromas early and accurately observe the location and quantity of the tumors, providing guidance for thorough removal of the lesions during surgery and reducing the postoperative recurrence rate[19]. The preferred examination method should be chosen if the patient's financial conditions allow.

This disease is a self-limiting proliferative disorder of the synovial membrane in joints. The diagnosis of this disease should be based on clinical manifestations, imaging examinations, and pathological examinations, with confirmation relying on pathological diagnosis. It should be differentiated from conditions such as osteoarthritis, osteochondritis dissecans, osteochondral fractures, and neuropathic arthropathy. As synovial chondromatosis can cause structural damage to the knee joint and secondary destruction of cartilage and bone, early surgical removal of

loose bodies and excision of pathological synovium should be performed. Once diagnosed with synovial chondromatosis, active surgical treatment is generally recommended, including traditional open surgery and minimally invasive arthroscopic surgery. Removal of loose bodies and excision of pathological synovial tissue are effective treatment methods for this disease, as the removal of loose bodies can prevent joint cartilage wear and degeneration, while complete removal of abnormal synovial tissue can effectively prevent recurrence[20].

Before the advent of arthroscopic technology, synovial chondromatosis was treated using open surgery. Traditional open surgery involves the removal of loose bodies and synovium under epidural anesthesia. While this method ensured more thorough cleaning, it also resulted in larger surgical trauma, higher risks, and slower functional recovery of the affected limb. With the advancement of technology and the development of minimally invasive concepts, arthroscopic techniques have rapidly progressed and have been gradually applied in the treatment of synovial chondromatosis in the knee joint. Arthroscopy allows for full visualization of various joint spaces, especially the posterior compartment of the knee joint, the popliteal hiatus, and the area below the meniscus, where small hidden bodies can be completely removed. During the procedure, arthroscopy can also accurately determine the type and staging of synovial chondromatosis, enabling the selection of the ideal surgical approach to remove the lesions and affected synovial tissues. With minimal surgical trauma and minimal impact on joint function, arthroscopic surgery allows for early postoperative rehabilitation exercise, avoiding joint damage. Additionally, it enables the examination and treatment of other important intra-articular structural injuries. Therefore, arthroscopic minimally invasive treatment of synovial chondromatosis has gradually replaced traditional open surgery[21].

Conclusions

This case report describes a rare condition called synovial chondromatosis. Considering the large number and widespread distribution of loose bodies in the patient, lack of evidence to determine if all loose bodies are within the joint, incomplete clearance during minimally invasive surgery, inability to remove large loose bodies, and the longer duration and higher cost of the minimally invasive procedure, open surgery was performed to achieve better exposure and a greater chance of complete removal. During the procedure, care was taken to protect blood vessels and nerves. After thorough clearance, knee joint flexion and extension exercises were performed to avoid missing smaller loose bodies or other tissue fragments. Due to the extensive range of movement in the knee joint, loose bodies can cause damage to internal structures during joint motion. After handling the tumor, a careful examination of the cartilage and meniscus was conducted, followed by appropriate treatment as needed. Post-surgery, a corresponding rehabilitation plan was developed based on the intraoperative procedure. Normal activities can generally be resumed after at least 2 weeks of rest. Patients should have regular follow-up appointments after discharge to prevent recurrence of synovial chondromatosis.

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Authors' Contributions
Manuscript writing and surgery were done by PQ, and ZX reviewed and revised the manuscript.

Conflict of Interest
The authors declare that there is no conflict of interest.

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Ethics Approval
The study was approved by the Ethics Committee of Liaocheng City Hospital of Traditional Chinese Medicine (approval No. 2023-10-27) on October 27, 2023. The study was conducted in accordance with the Declaration of Helsinki and its later amendments.

Informed Consent
Informed consent was obtained from the patient before the article was published.

Data Availability

Data from this study are available upon request from the corresponding author.

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