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Synovial chondromatosis of the hip, a case report and literature review

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ABSTRACT

A case of giant synovial chondromatosis of the hip with extra-articular localisation in the ileopectineal bursa is presented and the literature concerning this condition is reviewed. Synovial chondromatosis is a rare condition of unknown aetiology. A literature search was performed to identify studies describing outcome after surgical treatment of synovial chondromatosis of the hip. Thirteen studies including two hundred-ninety patients could be included.

Mean follow-up was fifty-six months. Seven out of two hundred-ninety developed a complication after surgical resection of the lesion. Recurrence rate of synovial chondromatosis is about 19%. Malignant transformation of synovial chondromatosis to chondrosarcoma is extremely rare. No wound infections were reported and avascular necrosis of the femoral head occurred in one patient who was treated with dislocation of the hip during surgery. After resection of synovial chondromatosis excellent functional outcome can be expected.

1. Introduction

A palpable inguinal swelling has an extensive differential diagnosis, consisting of inguinal hernia, lipoma, benign lymph node enlargement, abscess or cyst, swelling of a bursa, aneurysm of the great saphenous vein and soft tissue tumour. Differentiation between these diagnoses solely based on history and physical examination is difficult, therefore further investigation by means of imaging is often required to distinguish between the different diagnoses.[1]

Synovial chondromatosis (SC) is a rare benign condition, of which the aetiology has yet to be determined.[2] The cartilaginous chondromas or ossified chondromas (osteochondromas) may detach from the synovium and become loose bodies in the joint, which can subsequently cause damage to the articular surface and cause osteoarthritis.[3]

SC is most often found in a single joint, which is most frequently the knee. Other joints susceptible to SC are the hip, elbow, shoulder, ankle, wrist and temporomandibular joint.[2] Synovial chondromatosis usually occurs solely intra-articular, but has also rarely been described in extra-articular tissues such as tendon sheaths or bursae. In the majority of cases the condition is monoarticular.[2]

Patients with SC can present with various symptoms including pain, swelling and mechanical complaints.[2] The diagnosis can be made based on imaging alone, starting with radiography. Radiographs can show the calcified nodules in about 70% of the patients with SC.[2] In early stages, calcification may not yet be present and the nodules may not be visible.[4] Signs of osteoarthritis secondary to the loose bodies can be present. A CT-scan or MRI-scan is indicated if diagnosis remains unclear based on the radiograph or it can be used for preoperative planning. An MRI can show the non-calcified mass with chondromas or osteochondromas and the potential connection to extra-articular structures such as bursae can be determined.[2]

Synovial chondromatosis in the bursa has been described in several joints, such as the ankle,[2] the subacromial bursa in the shoulder,[5] bursae in the knee[6] and around the hip joint.[2] The histology is the same as the intra-articular variant. Bursal synovial chondromatosis may originate primarily from the bursa, but can also extend from the intra-articular space to the bursal sac.[6]

Although the vast majority of SC cases are benign, malignant transformation to synovial chondrosarcoma has been described.[5] The differentiation of SC from synovial chondrosarcoma can be problematic for both the pathologist and radiologist.[5]

Giant solitary synovial chondromatosis is first described by Edeiken et al. They set the definition as a mass larger than 1 cm in size. The masses are formed either by clumping of multiple chondromas, forming one large mass, or by growth of a single chondroma. On radiographs, these lesions can be difficult to distinguish from a low-grade malignancy. Few cases of giant synovial chondromatosis have been described, occurring in the ankle, hip and knee.[7]

Giant synovial chondromatosis of the hip mimicking an inguinal hernia has not been reported in literature before. We report the case of

Abbreviation: SC, synovial chondromatosis

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a patient presenting with an inguinal mass based on giant synovial chondromatosis op the hip and ileopectineal bursa and we give a comprehensive review of the available literature on synovial chondromatosis and its treatment.

2. Case report

In October 2017, a 54-year old male was referred to the outpatient clinic by the general surgeon with a tumour in his right inguinal region. The swelling had been present for years, but had lately increased in size. Main complaint was mechanical obstruction during gait. When sitting with the hip in flexion the patient experienced numbness of his right leg.

During physical examination a solid mass was palpated in the right inguinal region. The function of the hip was unaffected, except for a limited internal rotation of 20°. The neurologic and vascular status of the leg were unremarkable.

A previously performed ultrasound showed a calcified mass anterior to the femoral neck with a diameter of 5 cm. Conventional radiography of the pelvis and right hip showed an inhomogeneous partially calcified tumour of the soft tissues anterior to the femoral neck, appearing like a chondroid lesion, suspect for synovial chondromatosis (Fig. 1a). A CT-scan showed a partly lytic and partly sclerotic lesion of suspected chondroid origin, located intra-articular in the right acetabulum and extending anteriorly into the ileopectineal bursa (Fig. 1b). An additional MRI-scan was performed which showed localization of the bulk of the lesion in the ileopectineal bursa and it's communication with the joint (Fig. 1c). The largest diameter of the tumorous mass was 5.4 cm. Due to the mass the psoas muscle was deviated laterally, the communal femoral artery was deviated anteriorly and the femoral nerve was deviated medially and anteriorly (Fig. 1d).

Based on radiologic findings the diagnosis synovial chondromatosis was acknowledged.

Because of the localization close to the communal femoral artery, surgery was performed by an orthopaedic surgeon and a vascular surgeon. A longitudinal anterior incision was made and the communal femoral artery overlying the mass was moved aside (Fig. 2). The mass extended from the right hip joint into the ileopectineal bursa and was partially adhesive to the psoas major muscle. A limited anterior arthrotomy of the hip was performed to remove the intra-articular chondromas. To prevent vascular damage the hip was not dislocated and the acetabular chondromas were left in situ. Pathologic examination of the resected tissue confirmed the suspicion of synovial chondromatosis.

The postoperative period was complicated by a wound infection which was treated with surgical debridement and oral antibiotics, after which further recovery was uneventful.

3. Review of the literature

3.1. Methods

PubMed/MEDLINE, EMBASE, and the Cochrane Library were searched up to January 1st, 2018 for articles describing diagnosis or treatment of synovial chondromatosis in general and of the hip in particular. Studies describing synovial chondromatosis in adult patients were included. Exclusion criterium was minor age. Case reports were excluded to limit report bias.

We collected all information regarding level of evidence, baseline patient characteristics and mean period of follow-up. Data regarding SC location, type of surgery (arthroscopy or arthrotomy, with or without dislocation of the joint), surgical approach, complications, recurrence rate, functional and radiologic outcome and patient satisfaction were extracted.

3.2. Results

The search resulted in 178 articles, of which 13 studies including 290 patients could be included in the review.[3,8–19] Two studies did not report outcomes, but did analyse the extra-articular extent of synovial chondromatosis.[3,14] General patient characteristics can be found in Table 1. All reported numbers are sample-size weighted.

The average age was 41 years, 164 males and 126 females were included. The mean follow-up was 56 months. Location of synovial chondromatosis was equally distributed over the left and right extremities. Synovial chondromatosis was removed by arthroscopy in six studies[8–11] and by hip arthrotomy with dislocation of the hip in three studies.[9,11,18–20,21] One study compared resection using arthroscopy to an arthotomy[10] and one study compared treatment with an arthrotomy with or without dislocation dependent on patient characteristics.[15] The

Fig. 1a. The lesion on conventional radiography. The anteroposterior radiograph of the pelvis showing a partially ossified tumour anterior to the right femoral neck, appearing like a chondroid lesion.

Fig. 1b. Chondroid lesions intra-articular and in the ileopectineal bursa on CT-scan. CT-image showing a partly lytic and partly sclerotic lesion of suspected chondroid origin, located intra-articular in the right acetabulum and extending anteriorly into the ileopectineal bursa.

overall average local recurrence rate after resection of SC of the hip was 19%. In cases in which arthroscopy was performed a mean recurrence rate of 23% was reported.[8,10–19] No recurrence was reported after arthrotomy in combination with surgical dislocation of the hip.[9–12,15,17–20,21]. Lim and colleagues compared arthrotomy without dislocation to arthrotomy with dislocation. Choice of treatment was made based on the location and extent of the lesion. They reported a significantly higher complication rate in patients treated with dislocation in a population of 21 patients ($p = 0.042$). In patients treated with arthrotomy with dislocation, three complications were described; one neuropraxia of the femoral nerve, one avulsion fracture of the lesser trochanter and one post-operative osteonecrosis of the femoral head. In patients who underwent arthrotomy without dislocation no complications were reported.[15]

Several complications were described in patients treated with arthroscopy, including two patients with transient paraesthesia[16] and two patients with a reflex sympathetic dystrophy.[12] Functional outcomes were measured by Harris Hip scores in six studies. Average score improved from 53 preoperatively to 86 postoperatively. Type of treatment was not related to postoperative Harris Hip scores (Table 1).[8–11,13,15,17,19]

Seven studies described whether patients presented with extra-articular SC localisation, in 41 out of 103 patients extra-articular localisation of SC was reported.[3,8,9,13,15,18,20] Robinson and colleagues analysed the extra-articular localisation SC around the hip and reported that SC either spreads into the iliopsoas bursa or the bursa of the obturator externus.[3] Osteonecrosis of the femoral head was observed in two patients, of which one was diagnosed 12 years after surgery and

Fig. 1c. Communication of the lesion with the hip joint in MRI. MRI image showing localization of the bulk of the lesion in the right ileopectineal bursa and it's communication with the hip joint.

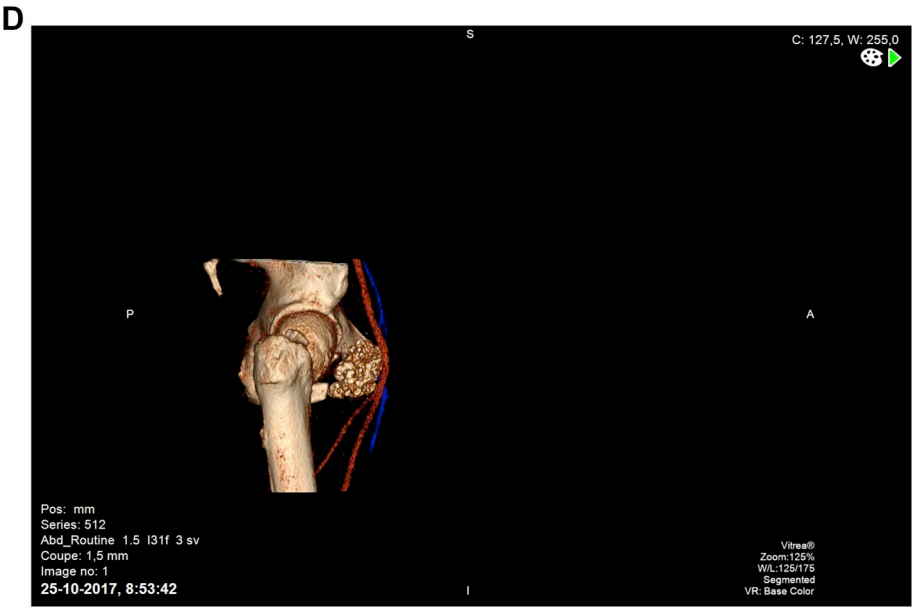

Fig. 1d. CT-reconstruction of the lesion and adjacent vessels. CT reconstruction image showing deviation of the psoas muscle laterally, the communal femoral artery (red) anteriorly and the femoral nerve (blue) medially and anteriorly. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

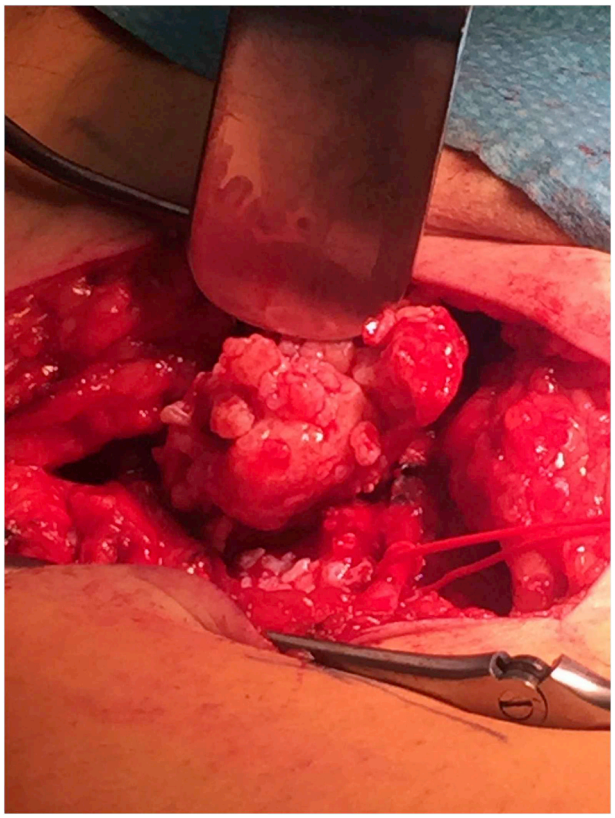

Fig. 2. The lesion during surgery. Photograph showing the synovial chondromatosis lesion with typical white cartilaginous aspect. The communal femoral artery is held aside.

was not reported as complication of surgery.[12,15] None of the 268 patients experienced malignant progression of the synovial chondromatosis to chondrosarcoma during the follow-up period.

4. Discussion

Synovial chondromatosis is a rare benign condition which can affect joints such as knee, hip, shoulder and elbow. To prevent risk of secondary osteoarthritis of the joint the osteochondromas can be excised if diagnosis is set before signs of osteoarthritis are present. Type of approach used for resection is based on patient characteristics, extent and localisation of the lesion and surgeons' preference.[4]

A rare presentation of synovial chondromatosis is a giant solitary synovial chondromatosis, a single lesion over one centimetre in size. This lesion can be formed by either an enlarged single chondroma or by clustering of several chondromas.[7]

In patients presenting with a palpable localised mass of the groin ultrasonography will often be the first choice of imaging.[1] A conventional radiograph of the joint can also be performed when SC is expected, which may show a partially calcified mass. If more information about location, extent and characteristics of the tumour is desired for diagnosis or preoperative planning, a CT or MRI can be performed. The radiological appearance of synovial chondrosarcoma, a malignant lesion of the synovium can be very similar to SC.[2]

In case of SC of the hip, osteochondromas can be removed by arthroscopy or by arthrotomy either with or without dislocation of the hip.[4,15] Recurrence of SC in the same hip can be expected in about 19% of patients. Rate of recurrence is lower in patients treated with arthrotomy and dislocation, compared to patients treated with arthroscopy (0% vs 23% respectively).

Complications of excision of SC of the hip are related to type of treatment. Wound infection is more likely to occur in open resection compared to arthroscopic treatment. Avascular necrosis of the femoral head occurred in one patient treated with peroperative dislocation of the hip. No avascular necrosis of the femoral head was encountered in patients treated with arthroscopy or with arthrotomy without dislocation of the hip.

This study gives a comprehensive review of the literature concerning synovial chondromatosis in general and of the hip in particular, and its treatment and expected outcome. The literature search shows a lack of quality evidence. High level evidence on treatment and prognosis of synovial chondromatosis is absent due to the rarity of the condition. The weaknesses of the original studies are reflected in our results. Patient specific characteristics such as smoking, obesity and diabetes, which affect the risk of developing avascular necrosis of the femoral head or postoperative wound infection, were not reported in the original studies and therefore not analysed.

Surgical resection of synovial chondromatosis can aid in prevention of secondary osteoarthritis. The type surgical approach depends on localisation and extent of the lesion and surgeon preference. In case of

Table 1
Overview of the literature describing surgical removal of synovial chondromatosis of the hip; patient characteristics, performed surgery and outcomes.

Author	Year	Level of evidence	No. of patients	Follow-up (M)	Age	Female	Type of surgery	Recurrence	Extra-articular involvement	HHS[a] pre-op	HHS[a] post-op
Polesello	2015	IV	6	17	45	4	Arthroscopy	0	x	54	90
Ferro	2015	IV	23	30	44	12	Arthroscopy	1	0%	62	85
Marchie	2011	IV	29	64	41	14	Arthroscopy	7	x	x	x
Boyer	2008	III	111	79	43	57	Arthroscopy	34	x	x	x
Zini	2013	IV	11	22	34	5	Arthroscopy	0	x	59	87
Abolghasemian	2014	IV	5	22	34	1	Arthrotomy with dislocation	x	0	39	81
Yoon	2011	IV	21	71	34	9	Arthrotomy with dislocation	0	x	x	x
Schoeniger	2006	IV	8	78	38	4	Arthrotomy with dislocation	0	0	x	x
Lim	2006	IV	21	53	41	7	All patients	2	8	58	91
			8				*Arthrotomy with dislocation*	*0*	*x*	*55*	*89*
			13				*Arthrotomy without dislocation*	*2*	*x*	*60*	*93*
Lee	2012	IV	24	41	43	4	Arthroscopy and arthrotomy without dislocation	4	22	39	82
Yu	2011	IV	9	25	51	2	Artroscopy and arthrotomy without dislocation	1	x	x	94
Robinson	2004	IV	7	x	29	3	x	x	5	x	x
Kim	2002	IV	15	x	37	4	x	x	6	x	x
Total/Mean			290	46	40	126		49		53	86

[a] HHS: Harris Hip score. x: not mentioned in the article

extra-articular localisation of SC an arthrotomy is more often performed.[17] Malignant progression of SC is extremely rare.[2]

Declarations of interest

None.

Informed consent

The patient involved in this study gave his informed consent authorizing use and disclosure of his protected health information.

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