RECURRING HEMARTHROSIS OF THE KNEE DUE TO SYNOVIAL HEMANGIOMA : REPORT OF TWO CASES OF WHICH ONE WAS TREATED SUCCESSFULLY BY ARTHROSCOPIC EXCISION

F. DE SCHRIJVER, S. GEENS

Although many reports have pointed out that recurrent pain and swelling with nontraumatic hemarthrosis of the knee in a young child suggests the diagnosis of synovial hemangioma, the diagnosis is often not made for several years. Too often, the patient with recurrent knee effusions is not diagnosed because symptoms abate between episodes, and each episode is often attributed to minor trauma. Aspiration of the hemarthrosis should alert the physician to a more severe underlying cause.
Because a clinical diagnosis will be made only if the clinician is aware of the condition, we reviewed the 195 cases previously reported to emphasize the common features of the condition. The two cases reported here show once more that preoperative diagnosis of a synovial hemangioma of the knee is a challenge. We add our experience to the few cases of successful arthroscopic removal of synovial hemangioma reported in the literature.

Keywords : synovial hemangioma ; nontraumatic hemarthrosis ; knee joint.
Mots-clés : hémangiome synovial ; hémarthrose non traumatique ; genou.

INTRODUCTION

As noted by Moon (7), Bouchut first described synovial hemangioma of the knee in 1856. Since then it has been widely reported, and the current number of cases in the international literature is 195 (1-16). The cases reported here make it 197.

Synovial hemangioma of the knee is a rare, although well-known and benign lesion. Often though, several years elapse between onset of symptoms and treatment. A long history of recurrent hemarthrosis and delay in appropriate diagnosis and treatment can cause hemophilia-like joint destruction (11, 15). This makes early and accurate diagnosis important.

Using plain films, angiography (3, 9, 11, 14), arthrography (2, 3, 6, 8), ultrasound, or computed tomography (CT) (3, 7, 9, 12) for preoperative diagnosis has limited success. More recently some authors have become interested in using magnetic resonance imaging (MRI) to diagnose this lesion (3, 5, 14). The role of arthroscopy as a means of diagnosing a synovial hemangioma is emphasized by many publications (1, 4, 6, 8, 9, 14, 16). Arthroscopy surely leads to greater accuracy in diagnostic technique. However, successful arthroscopic treatment is only reported in a few cases (6 in total; 6, 9, 10, 12, 13, 14). We add our experience by reporting two cases where a synovial hemangioma of the knee was diagnosed, of which one was treated successfully by arthroscopic removal; the second had to be removed by open surgery because of its diffuse character.

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REPORT OF CASE 1

A 26-year-old man suffered from anterior knee pain in his right knee for about 19 years. Symptoms started after a fall on his knee which caused a deep wound over the tibial tuberosity at age 7. The wound was closed over a drain, and the boy was hospitalized for 8 days.

He was first seen at our clinic one year after this trauma. His mother reported weakness in his right leg together with some limitation of function. Clinical examination revealed slight swelling of the joint together with some tenderness on patellofemoral grinding. At that time some internal patello-femoral derangement was diagnosed.

He was then lost to follow-up for several years. During this period he had recurrent effusions of his right knee every 3 to 4 months. For this he was treated elsewhere by multiple bloody joint aspirations, followed by long leg casting and immobilization for several months to no avail.

We saw him again at 22 years of age, 4 years before the final diagnosis. He was still complaining of anterior knee pain, and he was still having recurrent effusions. On physical examination there was tenderness over the anteromedial aspect of the knee, medial to the superior pole of the patella, where a plica could be felt. And again he was tender on patellofemoral grinding. Diagnostic measures were directed towards patellofemoral maltracking or a hypertrophic suprapatellar plica which became symptomatic after trauma.

He was again lost to follow-up and four years later, at 26 years of age, he appeared again with the same complaints. Physical examination at this time revealed a doughy mass on the medial side of his right knee. The skin overlying the mass appeared normal, and no vascular lesion was noted. Very slight quadriceps atrophy was detected, with limb lengths being equal. There was some effusion present. Knee movement was restricted with a range from 0-100° compared to 0-135° on his left side. The rest of the physical examination was totally negative. Results of blood studies were all normal.

Radiographs of the right knee revealed a soft tissue mass in Hoffa's fat pad, some tilting of the patella and a Maldague sign. No bony lesion or periosteal reaction was noted. CT could not demonstrate patellar maltracking or any soft tissue swelling. Differential diagnosis centered around patellar maltracking and synovial hemangioma. Because of his long history of recurrent internal derangement of this knee and in order to make a definite diagnosis, the patient underwent arthroscopy.

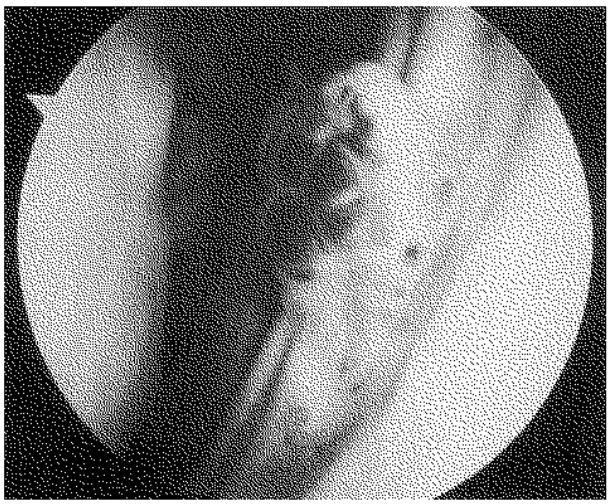
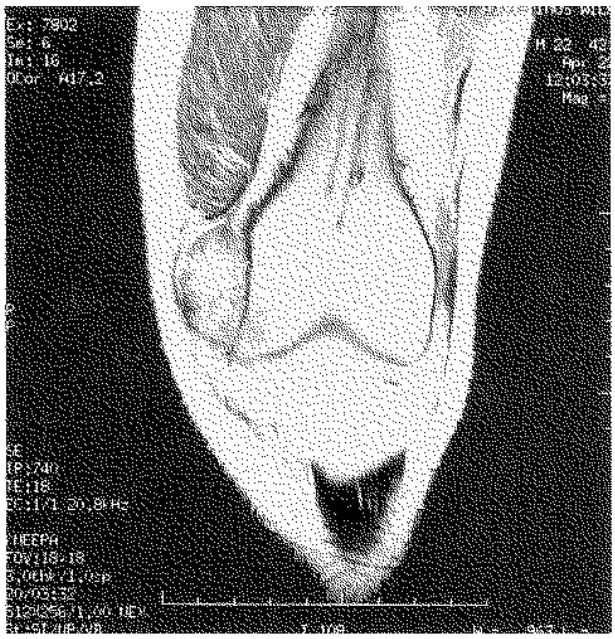


Fig. 1. – Arthroscopic appearance of the synovial hemangioma in the medial gutter.

A bluish strawberry-like pedunculated nodule, suggestive of a synovial hemangioma, was found at the level of the medial gutter (fig.1). This nodule measured 1 x 0.5 cm and was attached to the joint capsule. The nodule was excised and the surrounding synovium and capsule were electrocoagulated to control bleeding.

Initial pathological findings showed chronic synovitis, but after immunohistological preparation of the tissue with CD3 1 and CD34 (specific markers of endothelial cells), a cavernous synovial hemangioma was diagnosed.

We followed the patient on a regular basis in the outpatient clinic, and 18 months postoperatively he was still free of complaints. Never did a hemarthrosis recur, although he played football on a very competitive level. MRI done at this time confirmed healing of the lesion.



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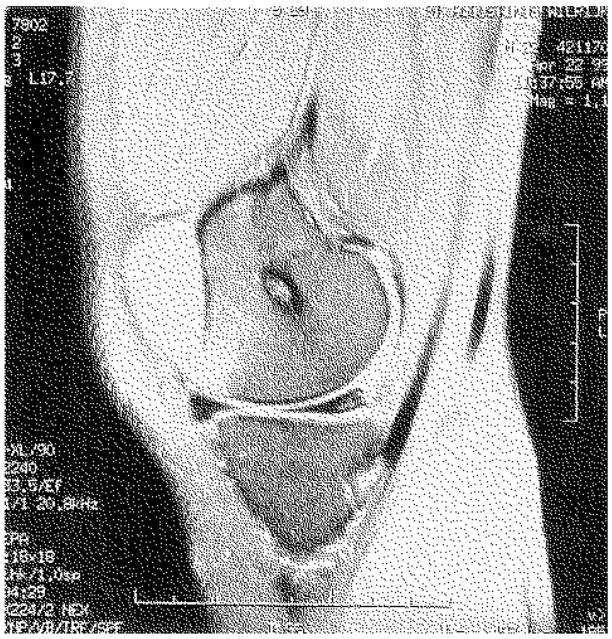

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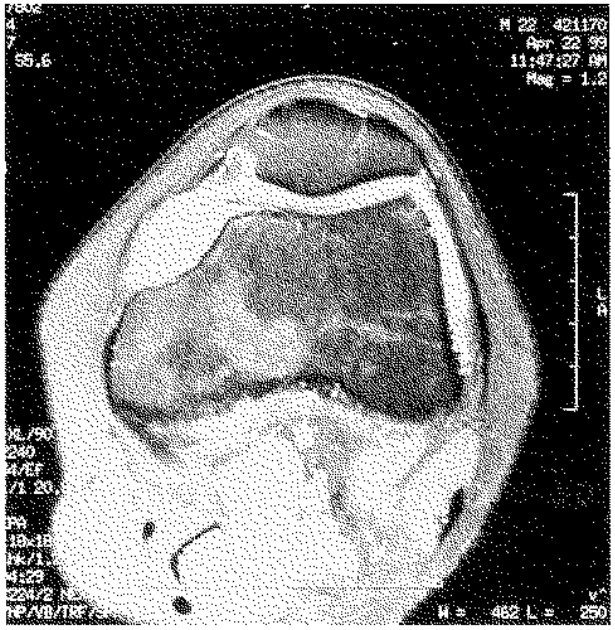

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Fig. 2-4. – MRI appearance of the synovial hemangioma in the medial capsule and retinaculum.

REPORT OF CASE 2

The history of the second patient, a 22-year-old male, started eight years before he first attended our clinic.

After a skiing accident he presented elsewhere with a swollen left knee, and he was treated with aspiration of the hemarthrosis followed by three weeks of immobilization in a long leg cast, with good results. Six weeks later he had fully recovered. The following two years however he was treated four more times by cast immobilization for recurrent hemarthrosis after minor trauma.

Two years after his first episode, he still had complaints about his left knee, and arthroscopy was done. A suprapatellar plica was resected. For two years he remained free of symptoms but he then relapsed. At rearthroscopy some scar tissue at the initial site of the suprapatellar plica was resected. Again he had no symptoms for four years.

He was first seen by us at this time. He presented with hemarthrosis of his left knee, after he had hyperflexion trauma of this knee at work.

Clinical examination revealed a hypersensitive, doughy and pulsating mass, the size of an olive, at the anteromedial capsule and retinaculum. The overlying skin was normal, and there was no muscle atrophy. Examination of the knee showed some resistance on flexion, but normal extension. There were no signs of instability, nor meniscal or patellofemoral involvement. Radiographs showed no bony lesions.

The history and clinical examination were very suggestive for a synovial hemangioma. To obtain an objective document, an MRI was done, which confirmed this diagnosis (fig. 2-4). A soft tissue mass in the medial retinaculum was shown, large in size and diffuse in nature, compatible with a synovial hemangioma. Diagnostic arthroscopy was done which showed some scar tissue at the site of the plica resection, but did not locate the lesion precisely. The lesion, size 3 × 2 cm., was removed *in toto* by means of anteromedial arthrotomy (medial parapatellar incision) and open resection and coagulation of the bed.

Pathological findings confirmed a venous synovial hemangioma.

Arthroscopy one year later (for a meniscal lesion) did not show any sign of recurrence. The patient himself was very satisfied and had no further symptoms.

DISCUSSION

I. Clinical

Synovial hemangiomas are rare, benign lesions, typically affecting children and young adults and found most frequently in the knee joint (4, 7).

The average age of patients at diagnosis is 13.5 years (mean age for men was 14.3 ranging from 2 to 53 years ; for women it was 10.9 ranging from 5 to 55 years). There is a slight male predominance, comprising 51% of the total.

Almost all patients with synovial hemangioma are symptomatic. Symptoms reported at the time of presentation most frequently included pain and swelling (90.4%), pain alone (4.2%), and swelling or mass without pain (5.4%). Limitation of motion was noted in 95 out of 169 recorded cases (56.2%). The degree of pain and tenderness can vary from mild to excruciating. Pain can also mimick a meniscal injury or a meniscal cyst (12).

Other clinical features are a palpable mass with a doughy consistency (65.9%) and quadriceps atrophy (90.1%). The size of the mass may decrease with elevation of the extremity. Adjacent or distant cutaneous hemangiomas were reported in 33 patients (16.8%). Increased limb length of the involved side has been mentioned in only 17 cases (8.7%). Aspiration of bloody fluid from the affected knee was specifically mentioned in 59 patients (30.1%).

Patients often report a chronic history of recurrent episodes of joint effusions and varying degrees of pain of several years' duration. This recurrent history of pain and swelling is a characteristic feature.

A history of previous trauma was elicited from 55 patients. This means that 28.1% of the entire series had a probable significant trauma to the knee prior to their symptoms.

Because symptoms may be intermittent and there may be no abnormal signs, diagnosis is often not made for several years. The mean interval between onset of symptoms and final diagnosis ranges from 1 month to 30 years, with an average of 6.8 years. It is important to emphasize that recurrent pain and swelling with hemarthrosis in a young child suggest the diagnosis of synovial hemangioma (16).

The site of the lesion may be juxta-articular, i.e. outside the joint capsule but in relation to it, intra-articular, i.e. inside the joint capsule, or intermediate (both intra- and extraarticular). Only intra-articular, and sometimes intermediate, lesions can give rise to hemarthrosis (16), because they generally involve the synovial membrane. Nontraumatic knee effusions can result from many causes, including meniscal tears, discoid menisci, loose bodies, osteochondritis dissecans, hemophilia and coagulopathies, essential thrombocythemia, synovial osteochondromatosis, malignant tumors, hemangiomas, pigmented villonodular synovitis, nonspecific synovitis/bursitis, juvenile rheumatoid arthritis and other inflammatory joint disease (Lyme disease ...).

Synovial hemangioma may be either diffuse or localized. Bilateral involvement of the knees was never reported.

2. Diagnosis

The most salient feature of the histories the authors reviewed was the diagnostic difficulties that this tumor offered the clinician. No definite

diagnosis was made for years in most cases, as in ours. This means that synovial hemangiomas are frequently misdiagnosed. By one estimate a preoperative diagnosis is made in only 22% of cases (7).

Radiological studies often are unrevealing (46.2% ; 68 out of 147 reported cases had normal radiographs). A vague soft tissue mass or effusion may be seen on plain films of the affected joint in some cases (24.5%). Phleboliths and bone erosions adjacent to lesions can be identified in patients whose lesions involve the extra-articular soft tissue (15%). Arthritic changes are also occasionally noted (8%) (3, 7). Other abnormal findings can be grouped as follows : periosteal reaction or thickening of the femur or tibia (3.5%), bone atrophy or lytic lesion (3.5%), vascular tumor or shunt (3.5%), osteolysis of femoral or tibial condyles (1.5%), advanced maturation of the epiphysis.

Angiography usually shows highly vascular lesions (vascular lesions were identified in 16 out of 22 recorded cases (73%)) and can reveal arteriovenous shunts, but arteriography may fail to identify the tumor (7). It is useful in delineating the extent of the lesion in a patient with a synovial hemangioma of the knee, but most physicians have had no experience with the use of this modality in the diagnosis of synovial hemangioma. Never-theless angiography remains an effective initial procedure in cases where physical examination gives the impression of the existence of a more widespread vascular lesion and in cases where arthroscopy showed a diffuse variety of hemangioma.

Arthrography (or pneumoarthrography) may demonstrate a filling defect with a villous configuration in some patients with synovial hemangioma, as it did in 50% of reported cases. Nevertheless this technique is imperfect and findings are nonspecific (3).

Computed tomography can aid in diagnosis when it reveals a heterogenous-appearing soft tissue mass, but it can also show findings that are nonspecific for synovial hemangioma (3). CT failed to show the lesion in 3 out of 8 cases ; it may be helpful to delineate extra-articular extension.

MRI may be of aid in delineating the extent of the lesion. Some authors (3, 14) even consider MRI to be the diagnostic procedure of choice. MRI findings of this soft tissue mass typically exhibit intermediate signal intensity on T1-weighted sequences, appearing isointense with or slightly brighter (higher) in signal intensity than surrounding muscle but much less bright than fat. The mass appears much brighter than subcutaneous fat on T2-weighted images and on fat suppression images and shows thin, often serpentine, low-intensity septa within it.

We believe arthroscopy to be the best means of making an early diagnosis when there is hemarthrosis. The value of arthroscopy is beyond doubt, although it may be difficult to recognize the lesion if this is in or near Hoffa's fat pad (1, 10). Of the 16 reported cases where diagnostic arthroscopy was used, 5 initial arthroscopies failed to show the lesion, these same 5 cases turned into positive arthroscopies at second look. These false negatives have been explained by the proximity of the lesion to the arthroscopic portal (12), by the altered colors of the tumor because of television equipment (1), by tourniquet related ischemia of the limb (8), or by poor visibility due to intraoperative bleeding (4). Other diagnostic tools are venography, sonography or thermography : they are of little use.

Accurate and adequate preoperative assessment assists in the classification of the lesion and guides definitive management with the aim of complete resection.

3. Pathology

Pathologic differential diagnostic considerations include nonspecific bursitis/synovitis, pigmented villonodular synovitis, nodular synovitis, and organizing hemorrhage (2).

As in our case the sectioning of three-dimensional papillary synovial fronds in two dimensions led referring pathologists to overlook the vascular nature of the true synovial hemangiomas and diagnose a nonspecific synovitis. Spaces lined on either side by synovial cells (and thus normally filled with synovial fluid) may be confused with immediately subjacent dilated vascular channels of a cavernous hemangioma. Once the impression of a vascular lesion has been formed, it is incumbent on the pathologist to correlate histologic with clini-

cal/operative findings. Should the confusion persist, positive immunohistochemical staining of the lining cells for factor VIII-related antigen may be of some use in confirming their endothelial nature (2).

The dominant histologic patterns (30 cases mentioned so far), included cavernous hemangioma (58%), lobular capillary hemangioma (14%), mixed arteriovenous hemangioma (21%), and venous hemangioma (7%). Capillary hemangiomas are composed of an endothelial component forming many small blood vessels, while the cavernous variety was characterized by large blood-filled spaces lined with a thin endothelium and lacking the cellularity of the former type (7).

4. Treatment

Treatment methods have varied in the past; radiotherapy, radical synovectomy, partial synovectomy, mass excision, and the use of sclerosing agents, cautery, freezing and injection of boiling water have all been reported (7).

Initial subtotal resections are mentioned in 8 cases; they all developed recurrent disease. One recurrence is reported even after complete resection (7). Total excision is clearly the objective of treatment.

Surgical excision with partial synovectomy has shown consistently good results when adequacy of removal can be assured (6 out of 55 recurred (10.9%)). Radiation therapy should now be reserved for cases in which excision is not possible.

Arthroscopy surely has gained a place and is even advisable in the treatment of small, circumscribed intra-articular tumors. Arthroscopic resection of diffuse synovial cavernous hemangioma is a submarginal resection with a high risk of recurrence and during which bleeding can be a problem necessitating repeated surgery or open resection. It is probably wise, if the lesion appears more diffuse, to perform arteriography before further intervention. In conditions that suggest a more widespread vascular abnormality, arteriography should also be carried out first. This is in cases with cutaneous hemangiomas, varicose veins of unusual size or location, or leg-length inequality.

In the literature some authors reported on successfully using the Yag laser for arthroscopic coagulation of the lesion (13). Its use is still controversial. As mentioned earlier recurrence is the problem in the diffuse variety.

CONCLUSION

Synovial hemangioma of the knee is a rare although well-known and benign lesion. Despite this, several years often elapse between onset of symptoms and treatment.

A recurrent history of pain and swelling is a characteristic feature. It is important to emphasize that recurrent pain and swelling with hemarthrosis in a young child suggest the diagnosis of synovial hemangioma. A clinical diagnosis will be made only if the clinician is aware of this condition.

We add our experience by reporting a case where a synovial hemangioma of the knee was successfully diagnosed and treated by arthroscopy.

Arthroscopy should be the method of choice for the early diagnosis of synovial hemangioma of the knee, and also to treat localized hemangioma and to plan subsequent treatment of diffuse forms of this tumor.

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SAMENVATTING

F. DE SCHRIJVER, S. GEENS. Recidiverende hemarthose van de knie veroorzaakt door een synoviaal hemangioma. Gevalsbespreking en nazicht van de literatuur.

Synoviale hemangiomen van het kniegewricht zijn zeldzame doch welgekende en goedaardige aandoeningen. Desondanks en spijtig genoeg, nog steeds te vaak gaat er een grote tijd verloren tussen het moment waarop zich de eerste symptomen voordoen en de uiteindelijke behandeling.
Het verhaal van recidiverende pijn en zwelling is nochtans vrij suggestief. Daarom eens te meer is het belangrijk de nadruk hierop te leggen, want men kan pas de klinische diagnose van een synoviaal hemangioom stellen, als men het verhaal en de kliniek kent.
Wij stellen een geval voor waarbij een synoviaal hemangioom van de knie arthroscopisch werd vastgesteld en met succes behandeld. Bij een tweede geval was het hemangioma te diffuus om arthroscopisch te verwijderen, en gingen we over tot open resectie.
Wij zijn van mening dat de arthrosopie zijn plaats heeft verdiend in zowel het stellen van de diagnose als de behandeling van een locaal hemangioom. Daarenboven heeft de arthroscopie haar waarde in de verdere therapieplanning van de meer diffuse vorm van deze aandoening.

RÉSUMÉ

F. DE SCHRIJVER, S. GEENS. Hémarthrose récidivante du genou due à un hémangiome synovial. Présentation de deux cas, dont un traité avec succès par arthroscopie.

L'hémangiome synovial du genou est une tumeur bénigne exceptionnellement rencontrée. Bien que cette tumeur soit connue, il se passe souvent plusieurs années entre l'apparition des symptômes et le traitment.
Une histoire de douleurs et d'épanchements articulaires à répétition doit suggérer le diagnostic. Cela suppose que l' on connaisse l'existence de cette lésion, et aussi que l'on prête attention à l'histoire clinique du patient.
Nous rapportons notre expérience d'un cas où un hémangiome synovial a été diagnostiqué et traîté avec succès par arthroscopie. Dans un autre cas, la lésion a été réséquée classiquement parce qu'elle était trop volumineuse pour se prêter au traitement arthroscopique.
Nous pensons que l'arthroscopie est la méthode de choix pour diagnostiquer et traîter les formes localisées de l'hémangiome synovial. Devant un hémangiome diffus, l'arthroscopie peut aussi aider à déterminer le traitement approprié.