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SYNOVIAL OSTEOCHONDROMATOSIS OF THE HIP

(*Report of 8 Cases Including 2 in Brothers*)

By

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Synovial osteochondromatosis is a fairly uncommon condition, and is seldom seen in the hip. It was therefore thought that a report on the following 8 cases would be justified, especially since 2 of them were seen in brothers and at least one of the other cases had been precipitated by rheumatic fever.

It appears that *Reichel* (1900) was the first to describe the condition, which he saw in an operated knee (16), where it is 10 times more common than in the hip. So far, some 40 cases of synovial osteochondromatosis of the hip are on record (17). None of the series described, however, consisted of more than 5 cases. The process consists of cartilage and bone formation in a lobulated and multicentric fashion within a synovial tissue. The bodies thereby formed become calcified, pedunculated and even detached and released into the joint as the disease progresses (8). The condition is conceived as neoplastic (Lexer) or metaplastic. All attempts to ascertain whether infection or trauma can cause the condition have hitherto failed. Apart from trivial everyday trauma, the onset is rarely antedated by any known traumatising accident. Histologically inflammation, most likely abacterial, is invariably seen in fresh cases (8, 9). Most recent authors believe the bodies probably formed from embryonic rests in the synovial tissue. Such rests have been shown to occur (8, 15, 19).

The possibility of the lesions being neoplastic is supported by a case reported by *Nixon, Frank, Chambers* in 1960 (13). Their patient, a man, was operated upon for microscopically confirmed osteochondromatosis. One year later the site of the operation was found to harbour a sarcoma, for which the patient was subjected to hindquarter amputation. This patient might have had a sarcoma and not

osteochondromatosis from the very beginning, for it is the only case on record that turned out to be malignant. As a rule, the process is self-limiting (3, 21), non-recurrent (17), non-invasive and monoarticular. It has been shown roentgenographically that single calcified bodies occasionally disappear spontaneously. One polyarticular case affecting the knee, ankle and elbow has been reported (5).

Osteochondromatosis of the hip is considerably more common in males than in females and has been seen in patients of all ages above 13 years.

During the last 20 years synovial osteochondromatosis of the hip has been seen at the orthopedic department in Lund in 8 patients, including 2 brothers.

CASE REPORTS

Case I. A girl, aged 15, complained of 3 to 4 years' aching pain in the left thigh with progressive loss of range of motion of the joint. No known trauma. In the years before she had had *scarlet fever twice.* Roentgenographic examination revealed osteochondromatosis of the hip joint. E. S. R. was normal. Operation with excision of the bodies and partial synovectomy confirmed the diagnosis. When last seen, at 21 6 years after the operation the patient was still free from pains in spite of slightly reduced range of motion. The X-ray picture showed no recurrence.

Case 2. A man, aged 31, had for 4 years complained of disabling aching pain and stiffness of the right hip. Infection or trauma not known. Repeated roentgenographic examination and biopsy to check the possibility of tuberculosis revealed unspecific synovitis only. He was then 29. Repeated determinations of the E. S. R. were invariably normal. When he was 31 another biopsy revealed the nature of the condition and the findings at the later synovectomy confirmed the diagnosis. Two free chondromas were also found in the joint. At 32 arthrodesis was performed because of the continuing pains and at 39 the roentgen picture showed no recurrence of the synovial calcifications.

Case 3. The patient was a man, aged 33. At 13 he had otitis. Three weeks later he was admitted to hospital because of fever with peaks of 41° C, increased E. S. R. (100 mm/1 hr.), and pain in several joints, especially in the left hip. He spent 2 months in hospital, after which he was symptomfree apart from a feeling of slight stiffness of the left hip. Roentgen examination one month after he had left hospital showed some slight skeletal rarefaction of the hip but not the picture of osteochondromatosis. At examination one month after he had left hospital the E. S. R. and the range of mobility of the hip were judged as normal but the patient says that he has never been free from stiffness of that hip since then. The condition had been diagnosed as *rheumatic fever.* The patient returned to his work on a farm and did not seek a doctor again until the age of 33, when an examination including roentgenography of the left hip for military service revealed the picture of a longstanding sclerotic, multicentric, typical synovial osteochondromatosis (Figure 1).

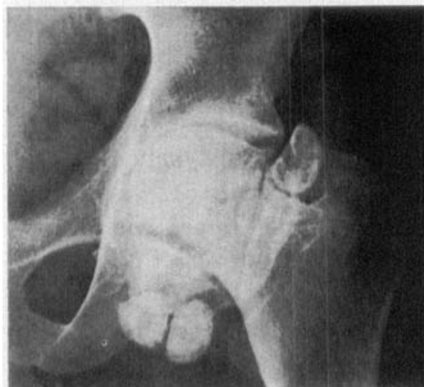

Figure 1. Synovial osteochondromatosis as seen 20 years after the first symptom during rheumatic fever in a 13 years old boy (case 3).

The range of mobility of the hip was only 50 per cent of normal and the quadriceps was severely atrophied. A lump was felt in the groin. Deforming osteoarthritis of the femoral head and acetabulum was also noted (Figure 1). Operation confirmed the diagnosis as did microscopy.

Case 4. The patient was a man, aged 35, who had for 9 years had progressive weakness, stiffness and aching pain of the left hip. No specific trauma, no infection known. Repeated roentgenographic examination had failed to reveal any calcified bodies. Repeated determinations of the E. S. R. had invariably been normal. Arthrography had not been done. Roentgenographic examination 9 years after the onset of symptoms revealed the characteristics of synovial osteochondromatosis. The patient had moved to another hospital region.

Case 5. The patient was a man, aged 40. At 15 he had been diagnosed as having coxa plana. Ten years later he sought treatment for progressive stiffness of that hip. The E. S. R. was normal. Infection or trauma was noted. Roentgenographic examination revealed typical synovial osteochondromatosis and a slight coxa plana deformity of the femoral head. When last seen another 14 years later, he was then 40 and the roentgen appearance of the hip was unchanged. The patient is still at work without pain and operation is not accepted.

Case 6. The patient was a man, aged 81. Suspected osteochondromatosis of the hip was discovered incidentally at roentgen examination after a minor traffic accident. The hip was not painful but its range of motion was slightly decreased. The E. S. R. was normal. Operation is not indicated, the diagnosis is not confirmed.

Case 7. The patient was a man, aged 45. At 26 he had had *tonsillitis with glomerulonephritis* treated in hospital, and at 40 he had a ureteric stone, which passed spontaneously. He reported that for 5 years he had had aching pain and stiffness of the right hip. He could not remember any trauma. During these years he had been examined repeatedly and had received various courses of physiotherapy. His working capacity was considerably reduced. The E. S. R. had been determined on 7 occasions and had always been normal. AST and ASTA were

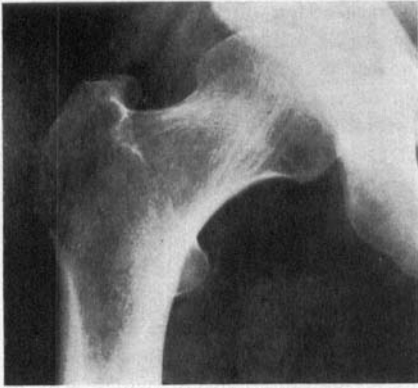

Figure 2. Case 7. Continuing hip pain in a 43 years old man as seen two years after the first symptom, still no calcification in the synovia.

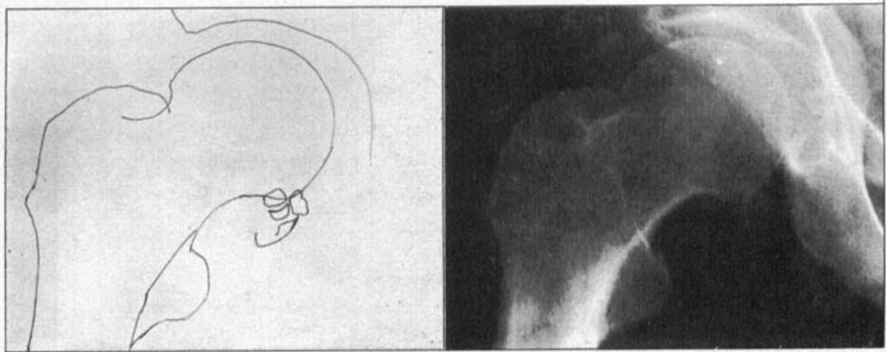

Figure 3. Case 7. Seven months later, two and a half years after the first symptom, non-diagnostic calcifications are visible.

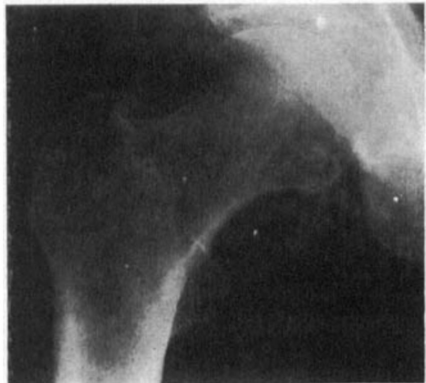

Figure 4. Case 7. Another 11 months later, three years after the beginning of continuing hip pain. Synovial osteochondromatosis probable and confirmed by operation.

negative. Roentgenography had shown some rarefaction but not the picture of osteochondromatosis (Figure 2). Tuberculosis was considered. Three years after the onset of the symptoms calcification was observed for the first time (Figure 3). The stiffness of the joint had not progressed during the last 2 years. Operation was performed one year later (Figure 4) with subtotal synovectomy. The excised tissue weighed 130 gm. The microscopic picture of the tissue was typical of osteochondromatosis.

Case 8. The patient was a man, aged 48. He was the brother of the patient in case 7. At 38 the right hip became painful. The pain had not been preceeded by any known trauma or infection. Three months later, by which time the pain was incessent, he was admitted to hospital for examination. Roentgenographic examination revealed nothing remarkable. The E. S. R. was normal. A biopsy specimen was removed via a short Langenbeck incision. The joint capsule, which was of

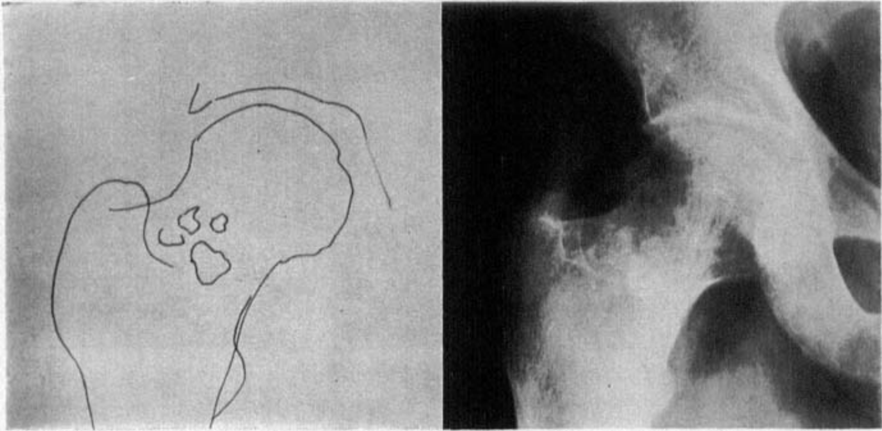

Figure 5. Case 8. A 38 years old man with hip pain since half a year. Non-diagnostic calcifications in the soft tissue, tuberculous was suspected.

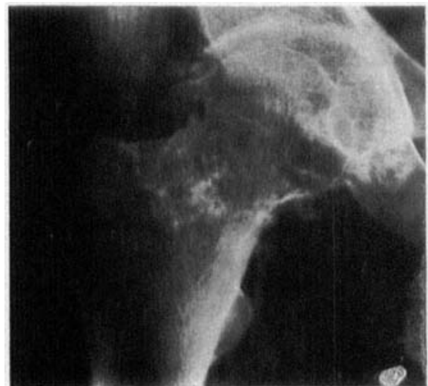

Figure 6. Case 8. The same patient as Figure 5, 8 years later. Picture typical of synovial osteochondromatosis. Brother of case 7, Figures 2–4.

normal gross appearance, showed histological evidence of villous hyperplasia with perivascular lymphocytes and histiocytes, but no specific elements. The pathologist's report was: non-specific inflammation. Roentgenographic examination 3 weeks later revealed for the first time a typical calcification (Figure 5). Arthrography was not performed. Body temperature, E. S. R. were normal and the clinical picture was unchanged. One year later the patient returned to work: the hip was then less painful but still somewhat stiff. Seven years later the patient's brother was found to have osteochondromatosis and then the patient was re-examined again, and this time roentgenography revealed the synovial osteochondromatosis (Figure 6). Deforming osteoarthrosis, which is sometimes seen in synovial osteochondromatosis, was not seen. Operation is planned mainly because of the severe limitation of mobility of the joint.

DISCUSSION

The occurrence of synovial osteochondromatosis in two brothers suggests the possibility of some familial factor in the causation of the disease. These two patients belonged to a sibship of 7 members (5 males and 2 females). Physical and roentgenological examination of the brothers and sisters revealed nothing remarkable apart from recurrent spells of pain in the left hip of one of the males. At the examination this man was treated with penicillin and had a registrable limitation of motion range of one hip although neither plain roentgenography nor contrast arthrography (Urografin) showed anything abnormal. Inquiry into the familial history of the remaining 6 cases in the series revealed nothing of interest.

Figure 7. The authors' material from the Orthopaedic Clinic in Lund 1945–65.

Case No.	1	2	3	4	5	6	7	8
Age at diagnosis	15	31	33	35	40	81	45	48
Male	—	+	+	+	+	+	+	+
Female	+	—	—	—	—	—	—	—
Durations of symptoms before diagnosis in years	3.5	4	20	9	14	—	5	10

Although examination did not reveal osteochondromatosis in a third member of the sibship, and though familial occurrence has never before been described, the possibility of some familial factor in the causation of osteochondromatosis cannot be excluded. The members of this family showed a tendency to obesity but were all healthy. There was no heredity of allergy or collagenosis, nor was any other joint disease known in the family.

In all the 8 cases the diagnosis of synovial osteochondromatosis had

been preceded by several years of hip trouble, sometimes very disabling (Figure 7). All of the cases had finally been diagnosed by roentgenography. As is known, in suspected cases the diagnosis can be demonstrated earlier by contrast arthrography (18). It was noteworthy that in seven of the eight patients the E. S. R. was invariably found normal. At the time of roentgenographic diagnosis all were normal. This argues against an active infectious coxitis and rheumatoid arthritis in a patient seeking attention for hip pains. If plain roentgenography shows no evidence of deforming osteoarthritis and the unexplained pain continues for many months the cause may be a synovial chondromatosis and arthrography should be considered.

Finally, by documented patient history, case n: o 3 shows that the hip affection of a rheumatic fever may lead to synovial osteochondromatosis. Further, one other patient (case 7) had had glomerulonephritis several years before the diagnosis of osteochondromatosis and case 1 had a record of scarlet fever twice. It all indicated that non-suppurative complications of streptococcal infection might be the start of a process which years later is diagnosed as synovial osteochondromatosis in a period when the E. S. R. is normal. Maybe the weak familial factor in question is a tendency for this immunological reaction to a streptococcal infection.

SUMMARY

A short review of the literature is given, the characteristics of the disease are described. Eight new cases are presented including two brothers, whose brothers and sisters were examined. In one of the eight cases described the disease seems to have started as a rheumatic fever, and non-suppurative complications of streptococcal infections are discussed as a causative mechanism in certain cases.

RESUME

Il est donné un aperçu de la littérature. Les tratis caractéristiques de la maladie sont décrits. Huit nouveaux cas sont présentés y compris deux frères dont les frères et soeurs furent examinés. Dans l'un des huit cas décrits, la maladie semble avoir débuté par une fièvre rhumatismale. Il est discuté de complications non-suppuratives d'infections streptococciques comme d'un mécanisme causatif dans certains cas.

ZUSAMMENFASSUNG

Eine kurze Übersicht über die Litterature wird gegeben und die Kennzeichen der Erkrankung werden beschrieben. Acht neue Fälle werden vorgestellt, einschliesslich zweier Brüder, deren Brüder und Schwestern untersucht wurden. In einem der beschriebenen Fälle hat die Krankheit scheinbar als rheumatisches Fieber begonnen. Nicht suppurative Komplikationen von Infektionen mit Streptokokken werden als Ursache in gewissen Fällen besprochen.

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