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Case Report

Rapid Development of Severe Osteoarthritis Associated with Osteoid Osteoma in a Young Girl

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Abstract: In a 14-year-old girl with a 5 month history of left sided thigh pain, X-rays demonstrating osteoarthritis and a bone scintigraphy showing increased focal uptake in the femoral neck led to the diagnosis of an osteoid osteoma. The protruberant bone at the femoral neck was removed en-bloc and the diagnosis histologically confirmed. The synovium was extensively infiltrated with lymphocytes. Postoperatively the girl experienced neither the expected pain relief nor improvement of her hip function over the next 4 months. MRI and CT results indicated development of a severe osteoarthritis even though no residual lesion could be found.

Keywords: Juvenile rheumatoid arthritis; Osteoarthritis; Osteoid osteoma

Introduction

Osteoid osteoma was first described by Jaffe in 1935 as a 'benign neoplasm of bone' [1]. Since then it has been found to respond well to total surgical excision although an intracapsular or juxta-articular localisation causes diagnostic and operative difficulties. In this case report of a 14-year-old girl, severe osteoarthritis developed as a result of osteoid osteoma only 5 months after the onset of symptoms, and progressed despite total excision.

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Case Report

A 14-year-old Caucasian girl presented with a 5-month history of left-sided thigh pain, initially felt only after activity but eventually also at rest and increasingly at night. The pain initially responded to aspirin. The range of movement of the left hip continued to decrease. The girl did not complain of morning stiffness. A family history of osteoarthritis could not be determined.

Physical examination revealed a limited range of internal and external rotation, a flexion contracture of 10° and tenderness in the groin. All other joints were normal. All microbiological investigations of joint fluid, including tests for *Mycobacterium tuberculosis* and other laboratory sera tests such as antinuclear antibodies (ANA) and HLA-B27, were negative. A reduced joint space was seen on radiography (Fig. 1). On scintigraphy the femoral head and neck had an increased focal uptake. The soft-tissue phase indicated an enhancement of the entire hip joint. A computed tomographic (CT) scan demonstrated a reduced joint space, subchondral cysts in the acetabulum and irregular cortical bone formation at the lateral femoral neck (Fig. 2). Magnetic resonance imaging (MRI) showed thickening of the joint capsule and soft-tissue formation at the lateral femoral neck with enhancement of gadolinium (Fig. 3). Based on CT and scintigraphic findings, osteoid osteoma was diagnosed.

The osteoid osteoma was excised en block. Intraoperatively, thickening of the joint capsule and synovitis were observed. Histological examination showed a typical osteoid osteoma. The synovium of the joint capsule and the perivascular region were extensively infiltrated with lymphocytes.

Despite total tumour resection (Fig. 4), neither the pain nor the hip function improved, even 4 months after

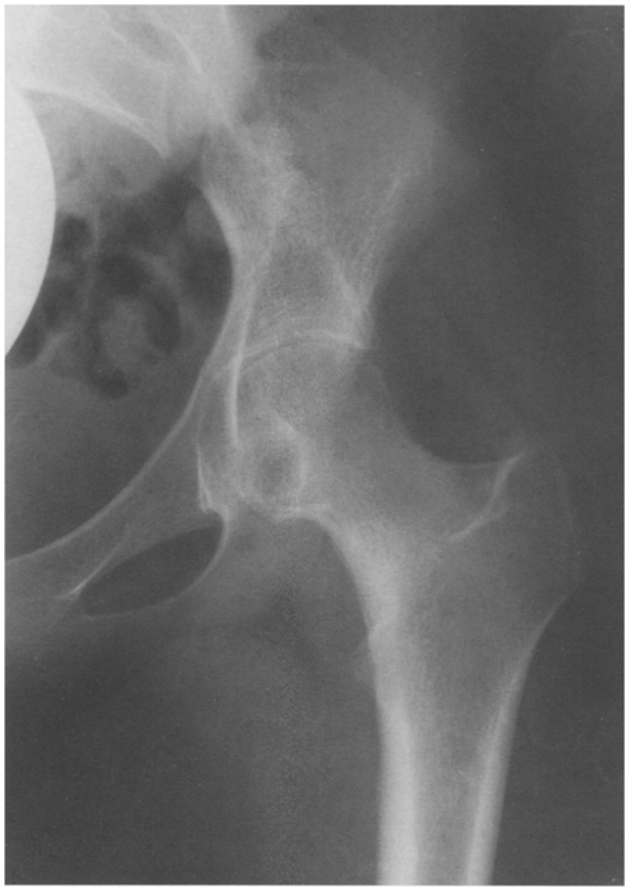

Fig. 1. Radiograph of the left hip revealing a reduced joint space and no sign of osteoid osteoma.

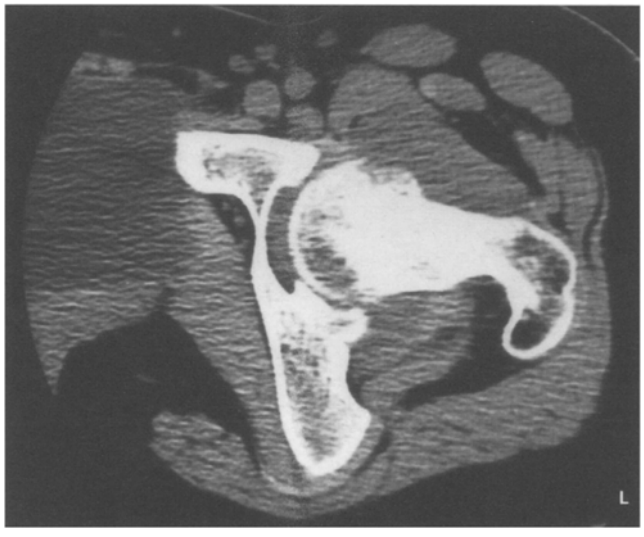

Fig. 2. CT scan of the involved hip showing a reduced joint space, subchondral cysts in the acetabulum and irregular cortical bone formation at the lateral femoral neck.

surgery. Physical examination then showed an unchanged range of movement with pain during the entire range of movement. The radiograph showed

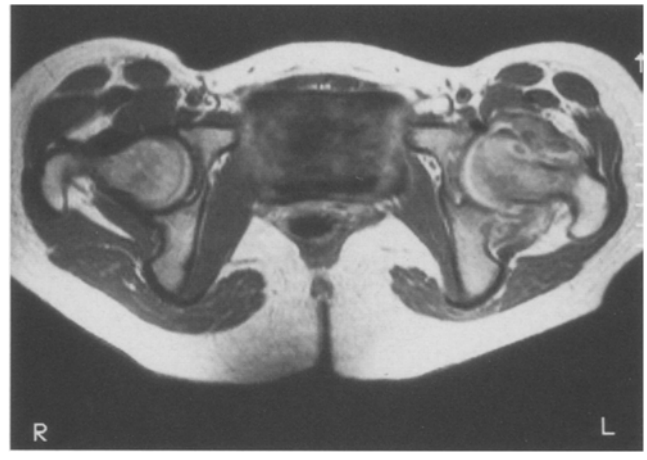

Fig. 3. Transverse MRI scan of both hips, T1 sequence. The nidus and the swelling and thickening of the joint capsule are seen in the left hip.

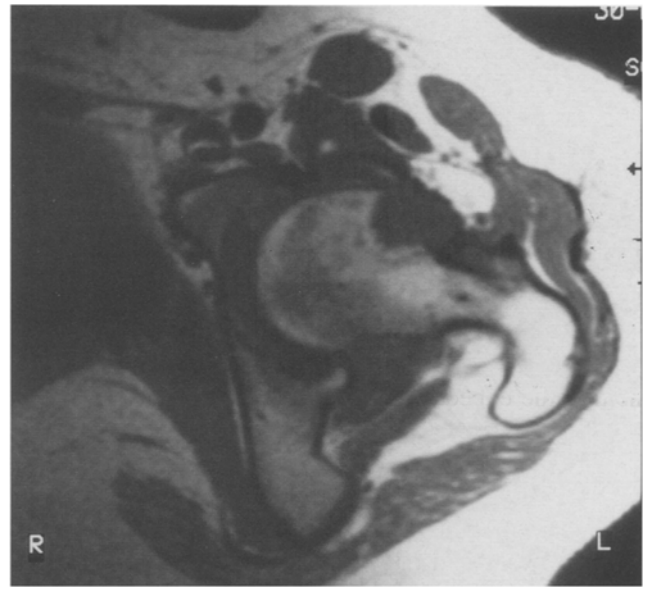

Fig. 4. Transverse MRI scan of the left hip, T1 sequence. The nidus has been totally removed.

progressive osteoarthritic changes of the joint (Fig. 5). A repeat MRI scan revealed progressive reduction of the jont space and the development of more subchondral cysts in the acetabulum. There was no evidence for persistence or recurrence of the osteoid osteoma.

Discussion

This case illustrates the development of severe osteoarthritis in the hip of a 14-year-old girl only 5 months after the onset of initial symptoms; the process developed despite total resection of the osteoid osteoma. Histologically the diagnosis of osteoid osteoma was confirmed. Radiologically the removal of the tumour was complete. Nevertheless, the osteoarthritis progressed.

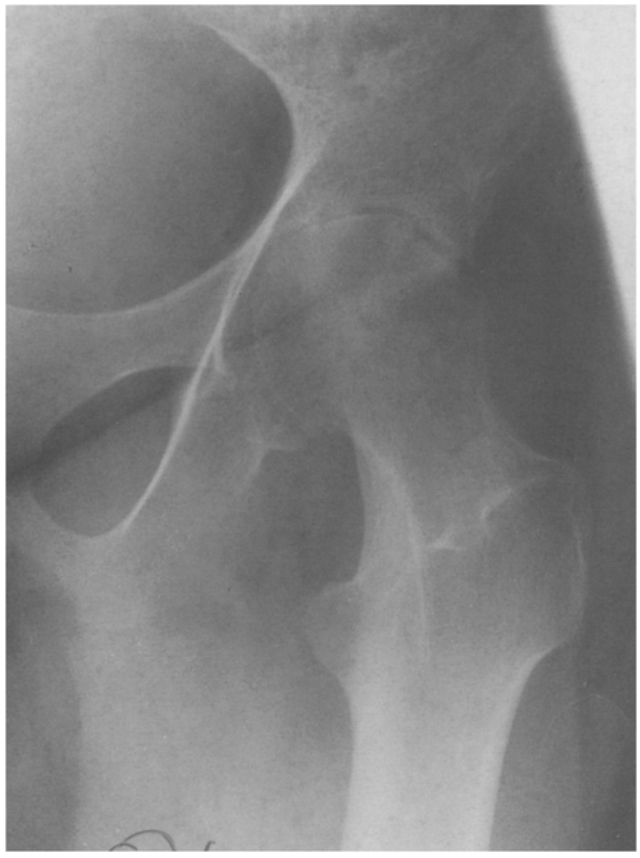

Fig. 5. Radiograph 5 months after the operation showing severe reduction of the joint space.

Such an intracapsular osteoid ostoema is known to be a diagnostic challenge because of its association with reactive synovitis, effusion and capsular thickening [2–4]. According to the published case reports, the diagnosis of intra-articular osteoid osteoma of the hip has always been delayed (6 months to 7 years) [5,6]. This delay could be reduced through routine use of newer diagnostic methods such as technetium-99m bone scintigraphy, CT imaging and MRI. In this case, the plain radiograph did not show the tumour, as could be expected according to the literature.

The development of synovitis has been postulated to be caused by an increased production of arachidonic acid by the tumour cells [7]. The persisting synovitis and capsular thickening can explain the restricted movement and the pattern of pain, which improved with mobilisation, as in any form of idiopathic childhood arthritis [8]. The diagnosis of idiopathic arthritis can be supported by histological inflammatory infiltration in the synovium.

Osteoid osteoma, but also to an extent idiopathic inflammatory arthritis, responds to non-steroidal anti-inflammatory treatment. In cases of no response, excision of the lesion is recommended [1,6,9]. Total surgical excision of the nidus, as in the presented case, should lead to relief of symptoms within a mean of 4.1 days [3]. Numerous paediatric case reports on intra-articular osteoid osteoma have described a total remission after tumour resection [3,5,6,10–13].

In a retrospective study of 36 patients with osteoid osteoma of the hip, osteoarthritis developed in 50% of the cases in which the joint was involved [14], the earliest onset of osteoarthritis being 18 months after onset of symptoms. In our case, osteoarthritis had already developed after only a 5-month history of thigh pain and progressed despite complete surgical excision of the osteoid osteoma. Sherman [15] described narrowing of the joint space after 2 years of pain in the joint related to osteoid osteoma, but after surgical removal of the tumour the patient was immediately relieved of her pain, and the limitation of movement was persistent, but not progressive.

The presence of subchondral cysts and rapid joint space narrowing, as described in our patient, resembles symptoms of a rapid progressive osteoarthritis of the hip, as reported by others [16,17]. In contrast to our patient, however, these patients were elderly women, the mean age at onset being 76.4 years and the progressive osteoarthritis was not related to osteoid osteoma.

In our patient no risk factors for early osteoarthritis were found, the family history was negative and no other osteoarthritic changes were found outside the involved hip. We have no explanation for the peculiarly rapid progression of joint destruction, despite the total removal of the osteoid osteoma, except an effect of the intra-articular osteoid osteoma. The case emphasises the need to be aware of possible rapid progression of an intra-articular osteoid osteoma to osteoarthritis in children.

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References

1. Jaffe HL. Ostoid osteoma – a benign osteoblastic tumor composed of osteoid and atypical bone. Arch Surg 1935; 31:709–28.
2. Brabants K, Geens S, van Damme B. Subperiostal juxta-articular osteoid osteoma. J Bone Joint Surg [Br] 1986;68:320–4.
3. Cohen MD, Harrington TM, Ginsburg WW. Osteoid osteoma: 95 cases and review of the literature. Semin Arthritis Rheum 1983;12:265–81.
4. Pikoulas C, Mantzikopoulos G, Thanos L, Passomenos D, Dalamarinis C, Glampedaki-Dagianta K. Unusually located osteoid osteomas. Eur J Radiol 1995;20:120–5.
5. Alani WO, Bartal E. Osteoid osteoma of the femoral neck stimulating an inflammatory synovitis. Clin Orthop 1987; 223:308–12.
6. Gille P, Gross PH, Brax P, Carcopino JM, Aubert D, Giordan H. Osteoid osteoma of the acetabulum: two cases. J Pediatr Orthop 1990;10:416–8.
7. Saville PD. A medical option for the treatment of osteoid osteoma. Arthritis Rheum 1980;23:1409–10.
8. Fink CW. Proposal for the development of classification criteria for idiopathic arthritides of childhood. J Rheumatol 1995; 22:1566–9.
9. Giustra PE, Freiberger RH. Severe growth disturbance with osteoid osteoma. Radiology 1970;96:285–8.
10. Orlowski JP, Mercer RD. Osteoid osteoma in children and young adults. Pediatrics 1977;59:526–32.
11. Rougerie C, Pider O, Cheavalier X, Larget-Piot B, Gouttallier D,

Claudepierre P. Apparent hip osteoarthritis in a 16 year old girl. Ann Rheum Dis 1997;56:343–5.
12. Siebenrock K, Asencio J, Ganz R, Poal-Manresa J. Osteoid osteoma in the femoral head – a report of 3 cases. Acta Orthop Scand 1997;68:70–6.
13. Miller SJ, Smith DW, Beckett WW, Gaines RW. Intra-articular osteoid osteoma of the acetabulum in a 6 year old. Orthopedics 1997;20:654–5.
14. Norman A, Abdelwahab F, Buyon J, Matzkin E. Osteoid osteoma of the hip stimulating an early onset of osteoarthritis. Radiology 1986;158:417–20.
15. Sherman MS. Osteoid osteoma associated with change in adjacent joint. J Bone Joint Surg 1947;29:483–90.
16. Rosenberg ZS, Shankman S, Steiner GC, Kastenbaum DK, Norman A, Lazansky MG. Rapid destructive osteoarthritis: clinical, radiographic, and pathologic features. Radiology 1992;182:213–6.
17. Lequesne M. Les coxopathies rapidement destructrices. Ann Radiol 1993;36:62–4.

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