Acta orthop. Scandinav. *40*, 198–204, 1969

From the Orthopaedic Hospital, Aarhus, Denmark.
(Head: Professor Eivind Thomasen, M.D.).

OSTEOID OSTEOMA

By

JENS O. POULSEN

Received 9.i.68

Osteoid osteoma is a not entirely uncommon benign skeletal lesion. Nevertheless, it is striking that the diagnosis often gives rise to difficulties. *Bergstrand* (1930) reported two cases which according to the description have no doubt been osteoid osteomas. *Jaffe* (1935) was the first to describe the disease as a pathological and clinical entity. In his opinion, the lesion represented a benign tumour. This theory has been supported by practically all subsequent authors, although a few have interpreted it as chronic inflammation (*Brown & Ghormley* 1943). According to *Dahlin* (1957) 10 per cent of all benign tumours of bone are osteoid osteomas. The disease is more common in males than in females.

CLINICAL AND X-RAY FINDINGS

Constant, aching pain of increasing intensity is characteristic. The pain is worst at rest and may disappear entirely when the patient is moving about. Frequently salicylates afford relief. As a rule, the pain is localized, but may be radiating, and this causes differential-diagnostic difficulties.

Clinical examination reveals localized tenderness, and swelling may be present also. When affecting a limb, the disease may give rise to muscular atrophy and when affecting the spine to postural abnormalities (*Rushton et al.* 1955, *Maclellan et al.* 1967). Effusion in an adjacent joint has been reported (*Sherman* 1947), and neurological signs in the form of sensibility disturbances and lacking tendon reflexes may occur (*Rushton et al.* 1955). Deformities of a limb may occur in children because of involvement of the epiphysis (*Ponseti* 1947, *Flaherty et al.* 1956). Because of the disproportion between the severe pain and the

usually slight objective changes the patient may risk being labelled as a neurotic.

The most valuable diagnostic aid is X-ray examination, although the findings may be negative at the time of appearance of the first symptoms. *Sherman* (1947), therefore, has stressed the importance of repeated X-ray examinations which should always be done in several projections. The typical X-ray film presents a small translucency surrounded by sclerosed bone. This is the so-called nidus, which seldom exceeds 1 cm in diameter. In some cases the sclerosis is so marked that the nidus cannot be distinguished. In that event tomography is of great value. At times, the entire painful limb, and in some cases also the spine, has to be X-rayed to arrive at the diagnosis. *Lindbom et al.* (1960) have called attention to angiography as a method for visualizing the nidus because of the high vascularization of the lesion.

PATHOLOGY

To render a definite histological diagnosis possible, the nidus has to be removed with a block of the surrounding, sclerosed bone. The nidus is round or oval, of a greyish or brownish colour. In consistency it may range from soft, granulation-like to firm, brittle tissue. The characteristic microscopic finding is a highly vascularized connective tissue lined with osteoblasts and surrounded by well-defined sclerosis of the bone, consisting of a network of anastomosing, osteoid trabeculae. Although this appearance is typical, *Brown & Ghormley* (1943) have published 14 operated cases 10 of which aroused a suspicion of chronic inflammation on microscopic examination. These authors deduced that osteoid osteoma is a variant of inflammation. *Golding* (1954) explained the violent pain as a consequence of increased tension in the nidus due to the high vascularization.

The prognosis is favourable. There have been no reports of malignant degeneration. Spontaneous regression is said to occur (*Moberg* 1951, *Vickers et al.* 1959). Operative treatment is curative and affords immediate relief of pain. X-radiation has no effect upon the lesion.

PRESENT MATERIAL

In the Orthopaedic Hospital, Aarhus, 13 cases of osteoid osteoma have been treated by operation. All were confirmed microscopically. Moreover, there has been a case in which the diagnosis could not be con-

firmed microscopically because of too small a specimen, but the clinical and radiological findings were typical of osteoid osteoma. The sites of the tumours were as follows: Femur (6), tibia (2), spine (1), talus (2), radius (1), cuneiform bone (1), and a phalanx of a finger (1). Of the 14 patients 4 were females and 10 males. The youngest patient was 5 years and the oldest 60 years of age. The oldest patient had a sclerosed osteoid osteoma. The two youngest patients were 5 and 6 years, but otherwise the age ranged from 15 to 38. The predominant symptom was pain. All cases showed positive X-ray findings, although in 2 cases, affecting the neck of the talus and the spine, tomography was required to visualize the lesions. At operation, the nidus was demonstrated macroscopically in 9 cases. In 2 cases the lesion looked like inflammation and in 1 case like a haematoma at operation, but in all cases the microscopic findings were typical.

The duration of symptoms from the first examination in the Orthopaedic Out-patient Department until operation was performed ranged from 2 months to 1 year. 12 patients were relieved of pain immediately after the operation. One patient developed a recurrence, but was relieved of pain by re-operation. In the oldest patient it is difficult to assess the condition. The lesion affected the radius, and the pain in this bone yielded, but the patient also has osteoarthritis of the elbow joint which is still causing pain.

Three characteristic case histories will be reported below:

(1) A 27-year-old joiner (Case No. 147412) was referred for pain in the left knee of 1 year's duration. The pain present during work as well as at rest.

Objective findings: 2 cm atrophy of the left thigh. No other abnormalities. X-rays of both knee joints in 2 views + special view of the patella, AP view of both hips, lower leg, and left femur failed to reveal any abnormalities. Tomography of the lower end of the left femur gave a suspicion, in the AP view, of a central translucency distally in the femur. However, this translucency could not be demonstrated on the lateral tomograms. The patient was kept under observation, and at a new follow-up visit a lateral X-ray film showed a 4 cm long cortical thickening posteriorly on the femur with a distinct nidus 25 cm above the joint line of the knee (Figure 1).

Operation was performed, chiselling off the nidus in a block, and microscopic examination revealed osteoid osteoma. Free of pain immediately after the operation.

(2) A boy, aged 5 years (Case No. 130646). Six weeks before he was seen in the Out-patient Department, he had complained of pain in the back and had ever since winced at the slightest touch of the loin.

Objective findings: Tenderness on a level with the spinous process of L 2 and slight lumbar scoliosis. X-rays showed a small translucency in the lamina of L 2. This translucency was also observed on pyelography which showed no other

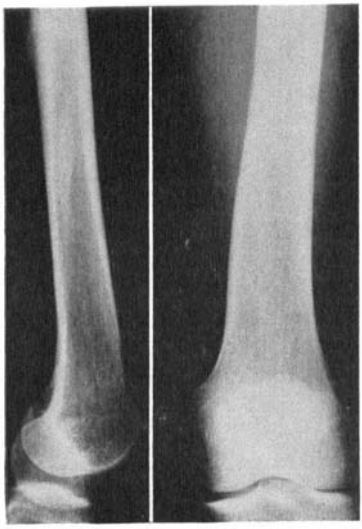

Figure 1. X-rays of distal end of right femur in 2 projections. The lateral shows cortical thickening with a nidus.

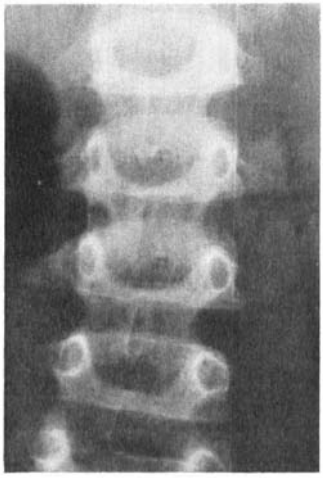

Figure 2. Tomography of the spine, showing in the arch on the right a translucency surrounded by sclerosed bony tissue.

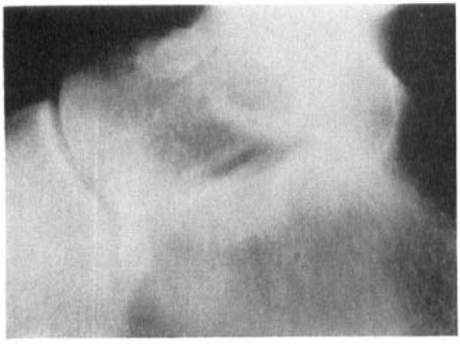

Figure 3. Tomography of the right talus showing an area of irregular sclerosis superiorly in the neck of the talus.

abnormalities. After the examination had been supplemented by tomography, there was no longer any doubt that this lesion was a nidus (Figure 2).

Operation showed thickening of the right arch of L 2. Chiselling disclosed a greyish-red tumour, somewhate larger than a pea. Microscopic examination: Osteoid osteoma. The patient has been fit ever since.

(3) A mechanic's apprentice, aged 19 (Case No. 118327). Referred to the Outpatient Department because of pain in the right ankle joint, most severe at rest. No complaints when walking. Acetylsalicylic acid alleviated the pain. The examination showed no abnormalities.

11 months later he was seen again because of pain, of a radicular type, in the entire right leg. There was 2 cm atrophy of the right lower leg and 1 cm atrophy of the thigh. The patient was admitted, and X-ray examination of the right foot disclosed a lesion on the anterior aspect of the neck of the talus (Figure 3). Tomography showed an irregular configuration of the bone at this site. Operation revealed a raised area at the anterior edge of the talar joint surface from which a round piece of bone could be lifted. Beneath, there was a pea-sized area of dry, brittle bony tissue. Micr. exam.: Osteoid osteoma. The patient has been free of pain since the operation.

SUMMARY

Persistent, deep, aching pain, as a rule worst at rest, and responding to acetylsalicylic acid should make one think of osteoid osteoma. The pain may be radiating. Repeated X-ray examinations may be needed to disclose the typical nidus surrounded by sclerosed bony tissue. The characteristic microscopic appearances are described. Operative removal of the lesion gives immediate relief of pain. At operation, the nidus should be removed together with a block of bony tissue, and an opening should be made to the medullary cavity. Curettement is not sufficient, as it may be followed by recurrence.

Fourteen operated cases are reported and 3 characteristic case histories given. The disease is most common in the age range 15 to 30 years, but may also occur in children; the present material includes two children, of 5 and 6 years.

RESUME

Douleur profonde, persistante, d'une manière générale plus accentuée au repos et réagissant à l'acide acétylsalicylique est un des traits caractéristiques de l'ostéome ostéoïde. La douleur peut rayonner. Des examens radiologiques répétés peuvent être nécessaires pour révéler le foyer typique entouré de tissu osseux sclérosé. Les aspects microscopiques caractéristiques sont décrits. L'enlèvement opératoire de la lésion apporte immédiatement un soulagement aux douleurs. A l'opération le

foyer doit être extirpé avec un bloc du tissu osseux et il doit être pratiqué une ouverture jusqu'à la cavité médullaire. Un curetage n'est pas suffisant, il peut être suivi d'une récidive.

Quatorze cas opérés sont rapportés et il est donné l'histoire de 3 cas caractéristiques. La maladie se produit surtout dans la période d'âge entre 15 et 30 ans, mais peut aussi être observée chez des enfants; les présentes observations comptent deux enfants de 5 et 6 ans.

ZUSAMMENFASSUNG

Konstanter, tiefsitzender Schmerz, der in der Regel am stärksten in der Ruhe ausgesprochen ist und auf Acetylsalicylsäure günstig reagiert, sollte an osteoid Osteoma denken lassen. Die Schmerzen können ausstrahlend sein. Wiederholte Röntgenuntersuchungen können notwendig sein um den typischen Nidus, der von sklerosiertem Knochengewebe umgeben ist, zu entdecken. Der charakteristische, mikroskopische Befund wird beschrieben. Operative Entfernung der Erkrankung giebt sofortige Befreiung von den Schmerzen. Bei der Operation sollte der Nidus zusammen mit einem Block von Knochengewebe entfernt werden und eine Öffnung zur Markhöhle sollte gemacht werden. Curettement ist nicht genügend, da es von Rückfall gefolgt werden kann.

Vierzehn operierte Fälle werden berichtet und drei charakteristische Krankengeschichten werden gegeben. Die Erkrankung findet man zumeist im Alter von 15 bis 30 Jahren, kann aber auch bei Kindern vorkommen. Das vorliegende Material schliesst zwei Kinder im Alter von 5 bis 6 Jahren ein.

REFERENCES

1. Bergstrand, H. (1930) Über eine eigenartige, wahrscheinlich bisher nicht beschriebene osteoblastische Krankheit in den langen Knochen der Hand und des Fusses. *Acta radiol.* **11**, 596–613.
2. Brown, R. C. & Ghormley, R. K. (1943) Solitary eccentric (cortical) abscess in bone. *Surgery* **14**, 541–553.
3. Dahlin, D. C. (1957) Bone tumors. Thomas, Springfield.
4. Flaherty, R. A., Pugh, D. C. & Dockerty, M. B. (1956) Osteoid Osteoma. *Amer. J. Roentgenol.* **76**, 1041–1051.
5. Golding, J. S. R. (1954) The Natural History of Osteoid Osteoma. With a Report of Twenty Cases. *J. Bone Jt Surg.* **36** B, 218–229.
6. Jaffe, H. L. (1958) Tumors and Tumorous Conditions of the Bones and Joint. Lea & Fibiger. Philadelphia.
7. Jaffe, H. L. (1935) "Osteoid-Osteoma". A Benign Osteoblastic Tumor Composed of Osteoid and Atypical Bone. *Arch. Surg.* **31**, 709–728.

8. Lindbom, A., Lindwall, N., Söderberg, G. & Spjut, H. (1960) Angiography in Osteoid Osteoma. *Acta radiol.* **54**, 327–333.
9. Löfgren, I. (1953) "Osteoid-Osteoma". *Acta chir. scand.* **104**, 383–404.
10. Maclellan, D. I. & Wilson, F. C. (1967) Osteoid Osteoma of the spine. *J. Bone Jt Surg.* **49** A, 111–121.
11. Moberg, F. (1951) The natural course of osteoid osteoma. *J. Bone Jt Surg.* **33** A, 166–170.
12. Ponseti, I. & Barta, C. K. (1947) Osteoid Osteoma. *J. Bone Jt Surg.* **29**, 767–776.
13. Rushton, J. C., Mulder, D. W. & Lipscomb, P. R. (1955) Neurologic Symptoms with Osteoid Osteoma. *Neurology* **5**, 794–797.
14. Sherman, Mary S. (1947) Osteoid osteoma associated with changes in adjacent joints; report of two cases. *J. Bone Jt Surg.* **29**, 483–490.
15. Sherman, Mary S. (1947) Osteoid osteoma; review of literature and report of thirty cases. *J. Bone Jt Surg.* **29**, 918–930.
16. Vickers, C. W., Pugh, D. C. & Ivins, J. C. (1959) Osteoid Osteoma. A Fifteen-Year Follow-Up of an untreated Patient. *J. Bone Jt Surg.* **41** A, 357–358.