BENIGN OSTEOLASTIC CHANGE RESEMBLING OSTEOLID OSTEOMA

Three Cases with Unusual Radiological Features

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We have recently encountered three patients with benign osteoblastic lesions which closely resembled osteoid osteomata but in whom the radiological features were unusual. This led to difficulty in diagnosis before operation although, in retrospect, the clinical features of each lesion fitted the description given by Jaffe in 1935.

CASE REPORTS

Case 1—A boy aged fifteen had pain and swelling in the region of the ulnar border of his right wrist for nine months. In 1956 he had lacerated this wrist, and had divided several flexor tendons and the median nerve. Surgical repair was carried out and the injuries healed uneventfully with subsequent full function.

On examination the temperature was normal and he appeared well. There was a firm fusiform swelling over the lowest third of the right ulna. The swelling was tender but not red or hot. Radiographs (Fig. 1) showed marked hyperostosis of the lower end of the ulna with sclerosis of bone and a radiotranslucent lesion one by one and a half centimetres in size. At its centre was an indefinite area of calcification. There was also considerable periosteal new bone on the adjacent border of the radius.

The diagnosis of osteomyelitis was made and he was given antibiotics for two weeks, at the end of which time he was admitted to hospital. The erythrocyte sedimentation rate was 9 millimetres in the first hour, the haemoglobin was 15.1 grammes, he had 8,100 white blood cells and the differential count showed 41 per cent of polymorphonuclear cells, 1 per cent of staff cells, 50 per cent of lymphocytes, 1 per cent of monocytes and 7 per cent of eosinophils. At operation part of the involved ulna was excised, which included the central nidus or sequestrum. A culture taken at the time of operation produced no growth.

Histological examination showed atypical, dense, irregular trabeculae of primitive bone with very sparse acellular stroma (Fig. 3). The surrounding sclerotic bone was more regular in its trabeculation (Fig. 4) with varying amounts of intertrabecular fibrous stroma.

The operation wound healed by first intention and when seen ten months after operation he had no complaint although supination was limited by 20 degrees. A year and a half after operation he continued to be symptom-free and radiographs at that time (Fig. 2) showed almost complete resolution of the bone changes.

Comment—The radiological appearances of the severe reaction not only in the bone of origin, the ulna, but also in the adjacent radius and the atypical histology prevented an immediate diagnosis of osteoid osteoma, although the normal laboratory tests, the gross appearance at operation, a negative culture and the subsequent course were in favour of that diagnosis.

Case 2—A boy aged sixteen had noticed a swelling on the dorsum of his right wrist for three months, with pain for two and a half months. There was diffuse thickening at the wrist with severe local tenderness at the radio-ulnar joint, but no redness or heat. There was full wrist movement and no cubital or axillary adenopathy. Investigations were normal with a haemoglobin of 15.6 grammes, and erythrocyte sedimentation rate of 3 millimetres in the

FIG. 1

FIG. 2

Case 1. Figure 1—The lesion in the ulna resembling an osteoid osteoma with periosteal reaction of the radius.
Figure 2—One and a half years after operation the bones are nearly normal.

FIG. 3

FIG. 4

Case 1. Figure 3—The atypical dense trabecular bone of the nidus. (Haematoxylin and eosin, $\times 41$.)
Figure 4—The sclerotic bone surrounding the nidus. (Haematoxylin and eosin, $\times 41$.)

Case 2. Figure 5—The lesion in the radius, with a small area of periosteal new bone to be seen on the ulna. Figure 6—Seventeen months after operation the ulna is almost normal but the radius is still thickened.

first hour, a white blood count of 4,600 per cubic millimetre and a differential count of 45 per cent of polymorphonuclear cells, 7 per cent of staff cells, 5 per cent of eosinophils, 2 per cent of basophils, 39 per cent of lymphocytes and 2 per cent of monocytes.

Radiographs (Fig. 5) showed an oval transradiant area half by one centimetre in size with surrounding sclerosis involving the ulnar aspect of the lower end of the radius. The

FIG. 7
Case 2—The typical osteoblast-lined osteoid trabeculae in vascular fibrous stroma. (Haematoxylin and eosin, $\times 41$.)

appearance was thought to be typical of osteoid osteoma, except for a small area of periosteal new bone which was evident on the adjacent ulna.

At operation the lesion was excised. The radius was enlarged and hard, with thick, oedematous soft tissues which were adherent. A red and gritty nidus, half by one centimetre

in size, was found deep to the cortex and lying in a bed of dense cancellous bone. A culture of the lesion was negative.

Histological examination of the nidus showed a typical picture of an osteod osteoma with heavily calcified, irregular, osteoblast-lined osteod trabeculae lying in a vascular fibrous stroma in which there were also a few giant cells (Fig. 7). The operation wound healed well and seventeen months after operation he had no complaint and wrist movements were full. Radiographs (Fig. 6) at this time showed persistent thickening of the distal radial metaphysis with distortion of the distal radio-ulnar joint. The lesion had healed and the periosteal reaction on the ulna had subsided.

Comment—This case was typical of an osteod osteoma were it not for the unusual radiological feature of periosteal reaction in the adjacent ulna.

Case 3—A girl aged eighteen had complained of a painful, stiff, swollen ankle for three years after a slight injury. Immobilisation, physiotherapy and antituberculous drugs had failed to improve her condition.

On examination there was marked thickening at the left ankle with exquisite tenderness at its antero-medial aspect. Both ankle and subtalar joints appeared ankylosed. There was no redness or heat but she had a fever of 99 degrees Fahrenheit.

Laboratory investigation revealed 12.6 grammes of haemoglobin, an erythrocyte sedimentation rate of 21 millimetres in the first hour, a white blood count of 5,800 per cubic millimetre and a differential count of 60 per cent of polymorphonuclear cells, 4 per cent of staff cells, 3 per cent of eosinophils, 28 per cent of lymphocytes and 4 per cent of monocytes.

Radiographs (Fig. 8) revealed an ill-defined transradiant area about a centimetre in diameter in the medial part of the talus at the junction of body and neck. There was decreased bone density surrounding the lesion, narrowing of the joint space and periosteal new bone in the region of the medial malleolus. More unusual, however, was the appearance of the tibial and fibular shafts proximal to the lesion (Fig. 9). Here the bone had almost the appearance of melorheostosis, with hyperostosis of the adjacent borders of the shafts; the opposite leg was normal.

Though the lungs showed no evidence of tuberculosis, the tuberculin test was positive and before operation a diagnosis of tuberculosis was made. At operation the soft tissues were found to be thickened and oedematous and the bone porotic. The periosteum had been raised from an area on the neck of the talus where the bone was discoloured and soft. A curette easily removed granular, gritty material from the lesion in the talus. A typical nidus was not seen. Synovial tissue from the ankle was hyperplastic, vascular and friable and there was excess synovial fluid which yielded no bacterial growth. Soft hyperostotic fibular bone was also taken; it appeared grossly and histologically to be simply sclerotic bone.

Histological examination showed a remarkably hyperplastic synovial tissue (Fig. 10). The bony lesion consisted of osteod trabeculae lined by osteoblasts and enclosing areas of vascular fibrous stroma with prominent giant cells with great cellularity (Fig. 11).

Within two weeks of operation the pain had gone, measurable movements of ankle and subtalar joints had appeared and the gait had greatly improved. Examination two months later showed further improvement with normal gait and freedom from pain.

Comment—This patient illustrates such widespread involvement of the limb bones that the detection of the primary focus and an accurate diagnosis before operation were virtually impossible. In addition to the radiographic changes in the adjacent medial malleolus, there were changes in the fibula and tibia far removed from the talar lesion, with degenerative or destructive changes in the adjacent ankle joint. This has been emphasised by Sherman (1947a) and mentioned by Dahlin and Johnson (1954). The differential diagnosis here was between osteod osteoma and benign osteoblastoma, but the prominent clinical feature of pain, and the small size of the lesion, is most usually found with an osteod osteoma.

FIG. 8

FIG. 9

Case 3. Figure 8—There is an ill-defined transradiant area in the talus with a periosteal reaction of the medial malleolus. Figure 9—Hyperostosis of the interosseous borders of the tibia and fibula above the lesion of the talus.

FIG. 10

FIG. 11

Case 3. Figure 10—Hyperplastic synovial tissue from the ankle joint. (Haematoxylin and eosin, $\times 41$.) Figure 11—The cellular fibrous stroma with irregular osteoid trabeculae lined with osteoblasts. (Haematoxylin and eosin, $\times 41$.)

DISCUSSION

Osteoid osteoma has become a well established lesion since Jaffe first defined it in 1935, and its radiological appearance in both cancellous and cortical bone was described by Jaffe and Lichtenstein (1940). The round or oval transradiancy, perhaps with a central calcified nidus, in cancellous bone and the often massive surrounding sclerosis in cortical bone are readily recognised.

The three cases reported, however, illustrate two radiological features which have not previously been widely recognised or associated with osteoid osteoma: changes in an adjacent joint and in an adjacent bone. Sherman (1947*a*) discussed two cases involving the neck of the femur and the olecranon fossa in which the adjacent hip and elbow joints showed mild degenerative changes. Dahlin and Johnson (1954), in reporting eleven cases, included one osteoid osteoma in which the pre-operative diagnosis was destructive arthritis of the ankle and which is strikingly similar to our Case 3. Freiburger, Loitman, Helpert and Thompson (1959) in a report of eighty cases, included two involving the femoral neck and lower fibula in which there was synovitis of the adjacent joint.

Sherman (1947*b*) also reported one case of osteoid osteoma in the distal metaphysis of the radius and "the shadow of periosteal new bone on the adjacent ulna." Her illustration resembles our Figure 5. Moberg (1952) described a remarkable case of osteoid osteoma of the head of the fourth metatarsal with periosteal thickening of the adjacent third metatarsal and proximal phalanx of the fourth digit. We have been able to locate only one other pertinent case, that of Sankaran (1954). His statement that "there is also sometimes a reactive proliferation of the neighbouring bone with thickening of the fibula with a distinct lesion in the tibia" is poorly illustrated.

The description of our three cases, together with the possibly six pertinent cases quoted from the literature, gives rise to two questions: first, if such reaction is possible with osteoid osteoma, why does it not occur more often, particularly in carpal or tarsal bones or in the tibia with its adjacent fibula? Wallace (1947) and Carroll (1953) gave details of some twenty-eight cases of osteoid osteoma in the hand bones, but made no mention of involvement of adjacent bones. Moberg (1952) accounted for this lack of sclerotic response by the absence of adjacent periosteal surfaces.

The second question which comes to mind is whether the appearance of remote sclerosis has any pathogenetic significance. Is there cause to re-examine the two widely accepted theories of its pathogenesis, neoplastic and infective, and perhaps add a third? The localised nature of the essential lesion, the nidus, does not suggest the possibility of a more remote infiltrative or suppurative process. Rather, spread of a toxic humoral material by diffusion or by lymphatic or vascular channels seems more appropriate. Both the neoplastic and the infective processes could conceivably give rise to this phenomenon. Certainly the extremely localised nature of the nidus is difficult to correlate with this more remote, less localised but equally dramatic change in adjacent bones and joints. In this regard the work of Lindbom, Lindvall, Söderberg and Spjut (1960) with angiography in osteoid osteoma is of considerable interest. By this technique they were able, before operation, to distinguish osteoid osteoma from sclerosing osteomyelitis. Its application in cases such as we report and in benign osteoblastoma might well be instructive.

Whether or not these lesions should be named osteoid osteoma in preference to benign osteoblastoma (Jaffe 1956, Lichtenstein 1956) is not easily decided. Benign osteoblastoma was considered particularly in Case 3, but the clinical, radiological and pathological features were those of an osteoid osteoma. In Case 1 the relatively acellular nature of the lesion was against the diagnosis of benign osteoblastoma, while in Cases 2 and 3 the marked local tenderness favoured osteoid osteoma. Pain, not usually an important feature of an osteoblastoma, was present in all three patients and particularly in Case 3. Though larger than many osteoid osteomata, all were under one and a half centimetres in size and therefore

not approaching the two to ten centimetres size described by Jaffe (1956). In spite of the very cellular appearance of the lesion in Case 3 and the atypical histology of Case 1 we believe that these three cases most closely resembled osteoid osteomata.

SUMMARY AND CONCLUSIONS

1. Three cases of a benign osteoblastic lesion of bone are described. An outstanding feature of each was the hyperostosis of adjacent bones or synovitis in an adjacent joint.
2. The clinical, radiological and histological features resembled osteoid osteomata more than benign osteoblastoma in each case.
3. The significance of this observation is questioned in relation to the pathogenesis of osteoid osteoma.

REFERENCES

- CARROLL, R. E. (1953): Osteoid Osteoma in the Hand. *Journal of Bone and Joint Surgery*, **35-A**, 888.
- DAHLIN, D. C., and JOHNSON, E. W., Jun. (1954): Giant Osteoid Osteoma. *Journal of Bone and Joint Surgery*, **36-A**, 559.
- FREIBERGER, R. H., LOITMAN, B. S., HELPERN, M., and THOMPSON, T. C. (1959): Osteoid Osteoma: a Report on 80 Cases. *The American Journal of Roentgenology, Radium Therapy and Nuclear Medicine*, **82**, 194.
- JAFFE, H. L. (1935): "Osteoid-Osteoma": a Benign Osteoblastic Tumor Composed of Osteoid and Atypical Bone. *Archives of Surgery*, **31**, 709.
- JAFFE, H. L. (1956): Benign Osteoblastoma. *Bulletin of the Hospital for Joint Diseases*, **17**, 141.
- JAFFE, H. L., and LICHTENSTEIN, L. (1940): Osteoid-osteoma: Further Experience with This Benign Tumor of Bone. *Journal of Bone and Joint Surgery*, **22**, 645.
- LICHTENSTEIN, L. (1956): Benign Osteoblastoma. *Cancer*, **9**, 1044.
- LINDBOM, A., LINDVALL, N., SÖDERBERG, G., and SPJUT, H. (1960): Angiography in Osteoid Osteoma. *Acta Radiologica*, **54**, 327.
- MOBERG, E. (1952): Further Observations on "Corticalisosteoid" or "Osteoid Osteoma." *Acta Radiologica*, **38**, 279.
- SANKARAN, B. (1954): Osteoid Osteoma. *Surgery, Gynecology and Obstetrics*, **99**, 193.
- SHERMAN, M. S. (1947a): Osteoid Osteoma Associated with Changes in Adjacent Joint: Report of Two Cases. *Journal of Bone and Joint Surgery*, **29**, 483.
- SHERMAN, M. S. (1947b): Osteoid Osteoma: Review of the Literature and Report of Thirty Cases. *Journal of Bone and Joint Surgery*, **29**, 918.
- WALLACE, G. T. (1947): Some Surgical Aspects of Osteoid Osteoma. *Journal of Bone and Joint Surgery*, **29**, 777.