OSTEOID OSTEOMA

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OSTEOID osteoma is a most interesting and unusual lesion of bone. Although relatively uncommon, this lesion must always be considered as a possible cause of pain in the extremities or in the backs of patients between the ages of 10 and 30 years. The disease occurs most frequently in the second decade of life and is encountered twice as often in males as in females. Since the clinical signs and the clinical course of an osteoid osteoma are unique, being unlike those of any other lesion of bone, a diagnosis of the condition can often be made even without histologic verification.

Osteoid osteoma was first described as a clinical entity by Jaffe[1] in 1935. However, five years earlier in 1930, Bergstrand[2] published a detailed summary of two cases of a rare benign osteoblastic lesion—one in a metatarsal and one in the phalanx of a finger. Preoperatively, Bergstrand had believed that these lesions were osteogenic sarcomas, but, after examining the involved areas of the resected bones, he concluded that the lesions were neither inflammatory nor neoplastic but probably were due to embryonal rests. These two cases of Bergstrand's were osteoid osteomas, but Jaffe receives the credit for initially describing the clinical and pathologic features of this most unusual lesion.

Jaffe applied the term "osteoid osteoma" to this lesion of bone because he believed it to be a true, benign, osteoblastic tumor consisting of osteoid and atypical bone. The exact nature of osteoid osteomas is still a matter of debate among pathologists, radiologists, and orthopedists. Brailsford,[3] MacKenzie,[4] and Hellner[5] believe that these lesions represent only a low-grade, cortical or subcortical, nonsuppurative inflammation in bone. The absence of any clinical signs of infection, together with the absence of any bacteriologic or microscopic evidence of inflammation in surgical specimens, has led most investigators to the conclusion that osteoid osteoma should be classified as a tumor of bone.

However, if this condition is a true neoplasm of bone, it is unlike any other benign tumor of bone. The extensive sclerosis of bone which is so often associated with an osteoid osteoma, is not seen with any other benign tumor of bone. The apparently spontaneous healing of the lesion makes it seem unlikely that this condition could represent a true tumor of bone. Only a very few cases of osteoid osteoma have been reported in patients more than 40 years of age. Sherman[6] and Moberg[7] have reported a few cases with clinical and roentgenographic findings typical of osteoid osteoma, in which the disease healed over a period

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of years, and the clinical symptoms disappeared with cessation of the progressive development of the nidus or circumscribed core of the lesion. In these cases, the roentgenographic changes persisted for many years, but gradually diminished. None of these cases, of course, was histologically verified, since biopsy would alter the natural course of the disease.

Trauma is believed to play only a small role or none in the production of an osteoid osteoma. In only a few of the reported cases does there seem to be any definite relationship between an injury and the development of the lesion. Furthermore, the majority of these lesions are located in portions of the skeleton not readily exposed to trauma.

Other authors have suggested that an osteoid osteoma may be the result of an embryonic rest, a healing infarct of bone, or a healing giant-cell tumor. Careful histologic studies have failed to substantiate any of these etiologic possibilities.

DIAGNOSIS

No matter what the exact nature or origin of an osteoid osteoma may be, this condition is certainly a definite clinical entity. Pain is the most important symptom of the condition. The pain is usually fleeting at the onset but gradually becomes more constant and more severe. The pain may be aggravated by exercise and is very commonly worse at night. A child with an osteoid osteoma will often be awakened from a sound sleep at night by a severe aching pain in or near the region of the lesion. In lesions about the hip, the pain may be referred to the knee; but, as a rule, the pain of an osteoid osteoma is localized directly over the site of the lesion. Small doses of aspirin almost always give prompt temporary relief from the pain. Pain may antedate, by several weeks, any other clinical or roentgenographic evidence of the disease.

On physical examination there is usually acute tenderness immediately over the site of the lesion. Some swelling may be present, caused either by actual thickening of the bone or by associated edema of the soft tissues adjacent to the lesion. However, there are no other evidences of inflammation, such as local redness, increased warmth, or local adenopathy; and there are no systemic evidences of infection, such as elevated temperature or leukocytosis. A limp may be present when there is a lesion involving one of the bones in a lower extremity; in fact, occasionally a limp may be noticed before pain has become the primary complaint. When the osteoid osteoma lies in close proximity to a joint, there may be associated inflammatory changes within the joint. These changes subside spontaneously after removal of the lesion. When the disease involves the cervical spine, a torticollis may be present; when the dorsal or lumbar spine is involved, a scoliosis may result.

Roentgenographic findings are to some degree dependent upon the age and the location of the lesion. The typical roentgenogram reveals an oval-to-round area of decreased density surrounded by an area of sclerotic bone. This inner area, or so-called "nidus," usually measures from 0.5 to 2.5 cm. in diameter. The nidus may be obscured by the overlying sclerotic bone, and overexposed films taken at different angles may be necessary before the lesion can be seen.

The regional sclerosis or hypertrophy of bone is greatest when the nidus is located near the cortex of a tubular bone; there is much less reaction if the lesion is more superficially located or if it occurs in cancellous bone. The nidus itself may appear uniformly translucent or may be mottled with irregular, sclerotic spicules of bone. Moberg[8] has recently reported a case of osteoid osteoma of the fourth metatarsal in which reactive formation of bone was not restricted to the bone in which the nidus was situated but was also present on two adjacent bones—the third metatarsal and the proximal phalanx of the fourth toe. The periosteal thickening was noted only on the lateral side of the third metatarsal, that is, on the side of the bone immediately adjacent to the metatarsal containing the osteoid osteoma.

Osteoid osteoma apparently is always a single lesion. There may be a recurrence of the lesion after incomplete removal of the nidus, but there is no evidence that more than one such lesion occurs in any one person.

Grossly, the central nidus is usually reddish in color and sharply demarcated from the surrounding bone. In more mature lesions, the color becomes less red and the cut surface shows reddish-brown flecks mixed with a pearl-gray matrix; the flecks represent calcified osteoid. The nidus may be soft and friable in the early stages, or firm and gritty as the tumor matures. When the lesion is within or near the cortex, the periosteum is thickened and edematous. There is no evidence that the lesion ever has invaded or broken through the periosteum or ever has metastasized.

The histologic appearance of an osteoid osteoma is characteristic: the nidus presents a stroma of vascular, richly cellular, immature, connective tissue containing all the elements necessary to the development of membranous bone, from primitive connective-tissue cells to osteoblasts. Interspersed throughout the stroma are islands and trabeculae of osteoid, mature or immature, depending upon the stage of development of the lesion. In some areas, this osteoid may be partially calcified. No inflammatory cells are seen and there are no signs of vascular necrosis or of old hemorrhage. About the osteoid are arranged large numbers of osteoblasts and a few osteoclasts. As the periphery is approached, the osteoid is more uniformly calcified. It is the osteoid tissue of the nidus itself which is characteristic; the peripheral heavy trabeculae represent normal sclerotic bone.

REPORT OF CASES

Since 1940, 17 cases of osteoid osteoma have been seen here. Eleven of these patients were males, and six were females. The youngest patient was six years of age, and the oldest was 40. Eleven patients, or 65 per cent, were less than 30 years of age.

In this group the distribution of the lesions according to their locations was typical of that in other reported series. The table lists the sites of these 17 lesions; 75 per cent of this group of osteoid osteomas was located either in the femur or in the tibia.

TABLE

Sites of Osteoid Osteoma in 17 Cases

Location	No. of Cases
Neck of femur	3
Shaft of femur	5
Tibia	5
Pubis	2
Carpus	1
Metacarpus	1

Four cases from this group are described here to illustrate the clinical features of osteoid osteoma and to emphasize certain aspects of the treatment of this condition.

Case 1. A 33 year old housewife was first seen on September 22, 1948. She stated that two years previously she had noticed the gradual onset of pain in the right hip with some radiation down the lateral aspect of the thigh. The pain had gradually increased in severity since its onset, and at the time of initial examination the pain was fairly constant but was always worse at night. A single aspirin tablet would give temporary relief from the pain. It was not aggravated by activity and was not relieved by rest. There was no history of any antecedent illness or injury. In January 1948 an exploratory operation had been performed to determine whether there was any evidence of inflammation in the lateral femoral cutaneous nerve. The patient was told by her surgeon that this nerve was not severed because no inflammation was found about it.

Physical examination revealed that the patient walked with a limp on the right leg. Internal rotation of the right hip was restricted to 15 degrees; other motions of the hip were normal. There was some deep tenderness over the anterior aspect of the right hip joint. The remainder of the findings of the general physical examination was essentially normal.

Laboratory studies, including urinalysis, blood count, hemoglobin determination, blood sugar content, Wassermann and Kahn tests, were all within normal limits. Roentgenograms of the pelvis and right hip, which the patient had had taken two months previously, showed no evidence of any bony abnormality except for slight cortical thickening along the inferior aspect of the femur with a small, oval, radiolucent area within this zone of sclerotic bone (fig. 1).

On November 2, 1948, the right hip joint was exposed through an anterior iliofemoral incision. The synovial lining of the hip joint was thickened, and there was an increased amount of fluid within the joint. On the anterior inferior aspect of the femoral neck was an area of roughened cortex. A marker was placed at this site and a roentgenogram was made which showed that this area coincided with the site of the nidus. A block of bone was then removed, measuring 15 by 10 by 6 mm., which included a small area of soft, reddish tissue corresponding in size with the small area of radiolucency seen on the roentgenogram. The walls of the cavity were then curetted, and another roentgenogram was made to be sure that the entire nidus had been removed.

Pathologic examination of the specimen revealed the typical histologic pattern of an osteoid osteoma. There was a well-defined layer of dense trabecular bone surrounding a small area in which the normal bony architecture was completely lost. This area was formed by thin, irregular, interlacing trabeculae of osteoid tissue, separated by relatively abundant, highly vascular, fibrous tissue containing osteoblasts and numerous multi-

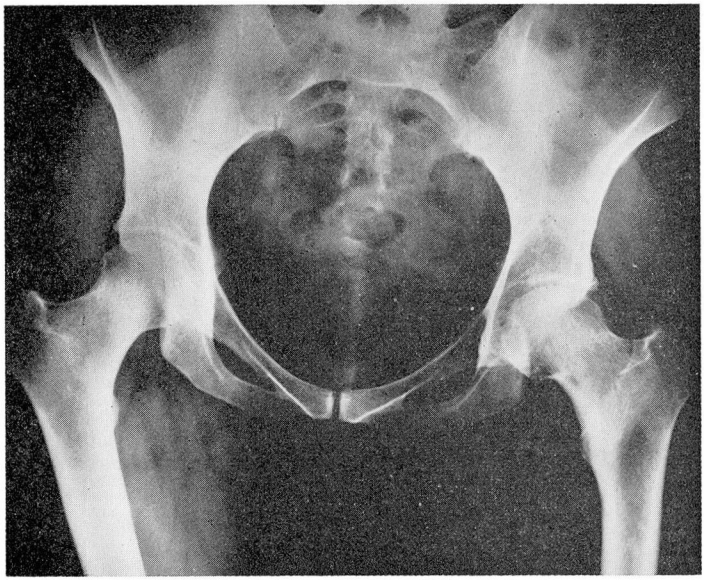

Fig. 1. (Case 1) Preoperative roentgenogram of pelvis. On the right there is a small radiolucent area along the inferior aspect of the femoral neck with some sclerosis of bone extending down to the lesser trochanter.

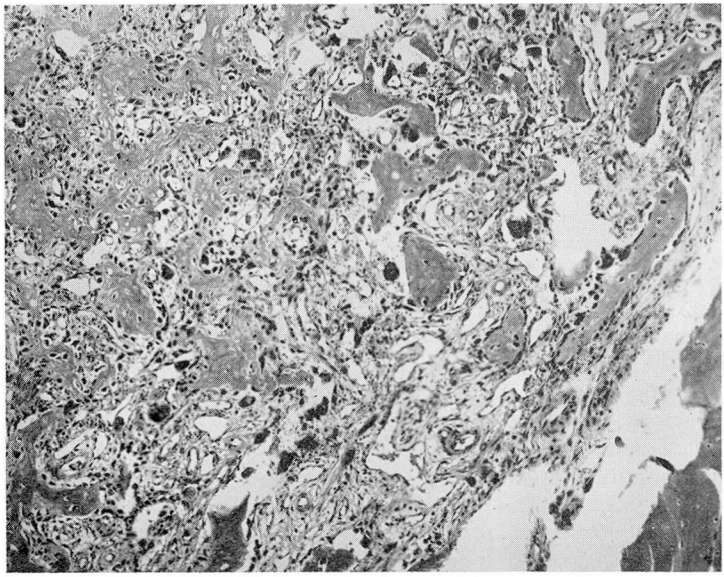

Fig. 2. (Case 1) Photomicrograph (X 90) of a portion of the osteoid osteoma removed from patient. Numerous small trabeculae of osteoid tissue are separated by a loose vascular connective-tissue stroma containing osteoblasts and osteoclasts.

nucleated giant cells of the osteoclastic type (fig. 2). A biopsy of the synovial lining showed evidence of a nonspecific type of inflammation, characterized by small foci of hemorrhage and diffuse infiltration of the synovial membrane by lymphocytes and plasma cells.

On the first day postoperatively, the patient was certain that she no longer had the pain in the hip which she had had prior to the operation. She has remained well, with no symptoms referable to the right hip for the five and one-half years since the operation.

Case 2. A 38 year old shoe-salesman was first seen on May 19, 1948, with the complaint of constantly aching pain in his right wrist of about 15 months' duration. There was no history of any injury to the wrist, although the patient believed the condition might have been aggravated by playing tennis. Previous treatment had consisted of roentgenologic therapy, physical therapy, splints and casts, and had not produced even temporary relief of pain. Injections of procaine into the tender area, and aspirin gave temporary relief.

Examination revealed an area of marked tenderness over the dorsal aspect of the second carpometacarpal joint. There was slight restriction of motion in the right wrist but no swelling in the joint. Findings of a general physical examination were essentially normal, and routine laboratory studies, including urinalysis, blood count, blood sugar content and blood serology, were within normal limits. Roentgenograms of the wrist and hand failed to reveal any abnormality.

On September 10, 1948, approximately four months after initial examination, the tender area was surgically explored under local anesthesia. On the dorsal proximal aspect of the lesser multangular bone there was found a small osseous defect, approximately 0.5 cm. in diameter, which was filled with a strawberry-colored, gritty, fibrous tissue. This tissue was removed with a curet and sent to the pathology department for examination; histologic diagnosis of osteoid osteoma was made.

The patient was relieved promptly of the pain in the wrist and has remained well for almost six years after operation.

Case 3. A nine year old boy was first seen on December 7, 1951, with the complaint of pain in the right knee. The pain had been present intermittently for 18 months prior to examination and had first occurred when the patient had injured the knee while playing football. The pain would persist for several weeks at a time, and then would disappear almost entirely for several months. The boy would be awakened at night by pain in the knee which could be relieved by local heat and massage. The boy's parents noticed recently that he limped slightly on the right leg.

Physical examination revealed a well-developed, well-nourished boy, who walked with a slight limp on the right leg. There were 2.5 cm. of atrophy of the right thigh and 1.5 cm. of atrophy of the right calf, as compared with the left. There was tenderness over the medial femoral condyle. The right knee lacked the few degrees of hyperextension present in the left knee; otherwise, motion in the right knee was complete. There was no swelling within the joint. There was slight soft-tissue swelling over the medial femoral condyle.

Roentgenograms of the right knee showed a radiolucent area measuring 1 cm. in diameter in the medial condyle of the femur, midway between the epiphysial line and the joint cartilage. Within this radiolucent area the normal bony architecture had been destroyed and some irregular sclerotic bone was present (fig. 3).

Surgical treatment was postponed because the lesion was located within the femoral epiphysis and because, at the time of the initial examination, the patient's symptoms were not severe. The symptoms increased in severity during the next year, yet roentgeno-

grams taken every three or four months failed to reveal any change in the appearance of the lesion in the medial femoral condyle.

At operation on December 4, 1952, approximately one year after initial examination, the anterolateral aspect of the medial femoral condyle was exposed; a small window was made in the bone, and through this window the nidus was removed with a curet. Roentgenograms taken at the time of the operation showed that the entire lesion had been removed. Pathologic examination of the removed tissue disclosed a typical osteoid osteoma.

Two days postoperatively the child stated he no longer had any pain in the knee, and he has remained symptom free for one and a half years after operation. A roentgenogram of the knee in July 1953, seven months after operation, showed that the surgical defect in the medial femoral condyle had become completely filled in with normal cancellous bone.

Case 4. An 11 year old boy was first seen in the neurosurgical section on May 12, 1950. He had been referred to that department by his local physician because of a suspected neurologic condition producing pain in the child's left groin and hip. This pain had begun insidiously in June 1949, about one month following an uncomplicated

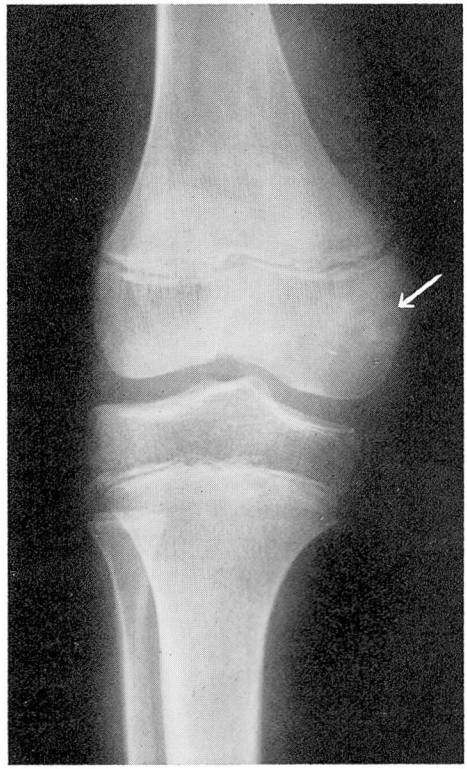

Fig. 3. (Case 3) Preoperative roentgenogram of knee, showing osteoid osteoma as a radiolucent area in medial condyle of femur, midway between the epiphysial line and the joint cartilage. The lesion being located in cancellous bone, there is very little surrounding sclerosis of bone. The arrow points to the lesion.

appendectomy. There was no history of any injury. The pain was described as "a toothache in the hip." The pain was aggravated by excessive exercise and relieved to some extent by rest and aspirin. At first the pain had been present only at night, awakening the child from a sound sleep. During the few months prior to examination, the pain had been present also during the day. The child was observed to limp on the left leg when the pain was present. He had been hospitalized elsewhere for two months because of a tentative diagnosis of tuberculosis of the hip, but at the time of his discharge the physician told the boy's parents that no disease had been found.

Results of the physical examination were essentially negative, except for definite tenderness to deep pressure in the left groin and 1 cm. of atrophy of the left thigh as compared with the right. There was no restriction of movement in the left hip joint.

Roentgenograms of the hip showed a somewhat ill-defined radiolucent area in the superior ramus of the left pubis. There was a shallow zone of sclerotic bone about this area (fig. 4).

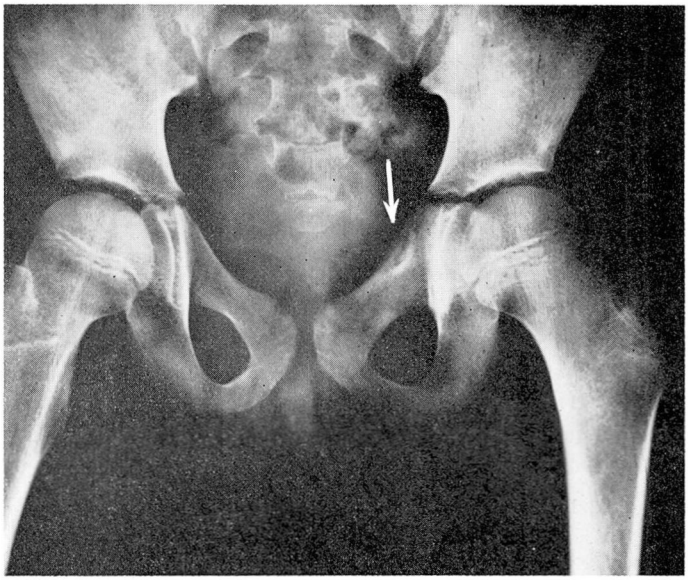

Fig. 4. (Case 4) Preoperative roentgenogram of hip. The arrow points to an irregular area of decreased density in the superior ramus of the pubis near the acetabulum. The nidus is not sharply defined and is surrounded by a narrow zone of dense sclerotic bone.

Operation was performed on June 8, 1950, approximately one month after initial examination; the superior ramus of the left pubis was exposed subperiosteally. Roentgenographic examination accurately localized the diseased area of the bone, which was then excised piecemeal with osteotome and rongeur. Many small pieces of bone were sent to the pathology laboratory for examination. Microscopic examination showed that the surgical specimen had the characteristic appearance of an osteoid osteoma. Another roentgenogram of the surgical area, in the operating room, revealed that the entire radiolucent area had been removed.

The child was relieved of his pain by the second postoperative day. Four years after the operation there had been no recurrence of any discomfort in or about the left hip.

DISCUSSION

Once a physician has seen a patient with osteoid osteoma, he will always remember to consider this lesion as a possible cause of pain in the bones of a child or young adult. The dramatic and permanent relief from pain which is obtained by surgical removal of the nidus will leave little doubt in the clinician's mind that this condition represents a real pathologic and clinical entity. Although it has been observed that osteoid osteomas will eventually heal spontaneously over a period of several years, surgical excision of the lesion is the treatment of choice. The risk of the operation is slight when compared with the years of continued suffering that the patient will have to endure if the nidus is not removed.

All of the patients in our series were successfully treated by surgical removal of the lesions. Two of the patients operated upon here and one of the patients operated upon elsewhere, each required two operations to remove the nidus completely. The entire nidus must be removed or the patient will continue to have pain. We have learned that roentgenographic control is imperative in the operating room: to locate the lesion and, before the incision is closed, to ascertain whether the nidus has been completely removed.

Occasionally, as in case 2, the nidus at the time of operation will be recognized as a well-encapsulated, reddish mass of fibrous tissue. In most of our cases, however, the central lesion has not been well defined, and only by studying roentgenograms made at the time of the operation could we be certain that the lesion had been completely removed. In order to cure osteoid osteoma, it is necessary to remove only the nidus—not the adjoining sclerotic bone. A block dissection of the nidus may be performed, provided that this does not entail removal of too great an amount of bone. A few cases[9] have been reported of fracture and of nonunion following too radical excision of an osteoid osteoma. The majority of our cases were not treated by block excision.

Pathologic verification of the nature of the lesion was not obtained in seven of our cases. In three of these, the surgical specimen was sent to the bacteriology laboratory for culture in the belief that the condition represented an inflammatory lesion of bone. But these cultures, as well as cultures taken from most of the other cases, were negative. In four cases the pathologist was unable to demonstrate the typical microscopic picture of an osteoid osteoma in the fragments of bone removed at the time of the operation. In these seven cases without pathologic confirmation of the diagnosis, the clinical course and the roentgenograms were so characteristic of osteoid osteoma that we believe we are justified in including them in this series.

Sherman[10] has pointed out that osteoid osteoma may be associated with changes in the adjacent joint. In case 1 of our report there was noted marked thickening of the synovial lining of the hip joint, as well as considerable increase in the amount of fluid within the joint. Microscopic examination of the synovial lining, however, failed to reveal any evidence of active inflammation. In case 3 there were also some arthritic symptoms noted in the knee joint, although the

joint itself at the time of operation showed no evidence of arthritic changes. Case 2 also presented some arthritic symptoms, referable to the wrist joint.

In case 3 the osteoid osteoma was located within the distal femoral epiphysis. The presence of an osteoid osteoma within an epiphysis is most unusual.

SUMMARY

Although the etiology and exact nature of osteoid osteoma have not yet been determined, the roentgenographic and physical findings in patients with this unusual lesion of bone are so typical that this condition must be accepted as a clinical entity.

The findings in a series of 17 cases of osteoid osteoma seen here since 1940 are presented. Four of these cases are described in detail to illustrate the clinical course of the disease.

Complete surgical removal of only the nidus, or central core of the lesion, is all that is necessary to obtain an immediate permanent cure.

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