RECURRENT OSTEOID OSTEOMA
Report of a Case with a Review of the Literature

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Osteoid osteoma has been accepted as a distinct clinical and pathological entity since it was described by Heine (1927), Bergstrand (1930) and most clearly by Jaffe (1935). Its features are that of a central nidus of vascular connective tissue stroma in which is scattered osteoid and atypical bone. Curettage will often cure the lesion. The present article reports a patient in whom osteoid osteoma recurred after en bloc excision on two occasions and reviews the literature.

CASE REPORT
A man aged forty-five years complained of pain and swelling of his right hand for a few days after an injury at work. During the next eighteen months he complained of continued swelling over the dorsum of the hand aggravated by shaking hands. Radiographs showed relative density of the second metacarpal shaft (Fig. 1). Examination showed a soft mass which was excised, and a diagnosis of ganglion was made. Six years later, in May 1965, he was readmitted to hospital with recurrent pain deep to the site of the previous operation. Radiographs showed an area of transradiancy with a central nidus of punctate radiodensity in the base of the right second metacarpal bone (Fig. 2). An en bloc excision was done (Fig. 3), the defect being filled by an autogenous cancellous bone graft. There was a cavity in the second metacarpal base with a central firm red nidus and surrounding loose vascular tissue, strongly suggesting a diagnosis of osteoid osteoma. Microscopically this diagnosis was confirmed (Fig. 4).
The operation relieved pain temporarily, but a year and a half later he returned with more pain and swelling in the same part of the hand. Radiographs showed what appeared to be a recurrence (Fig. 5). A second wide resection was done in November 1966 (Fig. 6), with removal of a block of bone two by one and a half centimetres from the bases of the second and third metacarpal bones. A central nidus was again identified, which microscopically was formed by a woven mesh of osteoid inside a shell of denser reactive bone (Fig. 7).

He was again relieved of symptoms initially but returned in June 1968 with a recurrence of the aching pain with weakness and difficulty in using the thumb and index finger. He now
reported relief of pain by aspirin. On examination there was local tenderness at the base of the second and third metacarpal bones, soft-tissue thickening extending into the palm, and wasting of the thenar muscles. The extent of the third excision done at that time is shown in Figure 8.

Figure 5—Radiographic appearance in October 1966, a year and a half after the first excision. Note resolution of bone graft and remarkable resemblance to earlier appearance. Figure 6—Radiograph showing extent of second excision.

Microscopic appearance of the second specimen. Dense osteoid trabeculae in sparse vascular stroma. ($\times$ 254.)

Microscopically the appearance was no longer typical of osteoid osteoma, but the essential features were nevertheless present (Fig. 9). Reactive bone and fibrosis surrounded foci or nodules of osteoid trabeculae, each of which appeared identical to the usual nidus of osteoid osteoma (Fig. 10).
A year later he continued to complain of pain, relieved by activity and by aspirin. His grip was weak and there was marked restriction of flexion at the second and third metacarpophalangeal joints; these and the interphalangeal joints extended fully. There was a small exquisitely tender point in the region of the dorsum of the lunate bone. Thenar muscle tone was good and sensation was intact. Radiographs (Fig. 11) showed residual thickening of the second metacarpal bone, shortening of the third and reactive scarring of bone in the region of the previous three surgical excisions. The findings were interpreted as giving no certain indication of further recurrence of the osteoid osteoma and no further treatment was undertaken.
Recurrences of osteoid osteoma have been recorded usually after curettage or drilling and rarely after en bloc excision. The need for careful radiological and microscopic control at the time of operation was stressed by Mayer (1953), who recorded a case in which the nidus was missed completely at the first excision and was discovered only on subsequent radiographic examination. It is worthy of note that Mayer’s case could have been thought to represent a recurrence.

Recurrence is seldom reported, but when it does occur it is usually ascribed to incomplete removal of the nidus by curettage, a factor stressed by Golding (1954). It seems, however, that recurrence is also possible after attempted en bloc excision unless suitable radiographic control is employed. Even so, the reactive sclerosis encircling the circumference of the long bone in fusiform fashion may mask the nidus radiographically, except on tomography.

We have found reports of thirteen recurrences. Ponseti and Barta (1947) reported three after curettage. In Golding’s (1954) series of twenty patients, five recurred. Freiberger, Loitman, Helpern and Thompson (1959) reported eighty cases, with four patients who needed two operations and one patient who needed three for complete removal of the nidus and permanent relief of symptoms.

Certainly many osteoid osteomata are treated successfully by curettage alone. If incomplete removal of the nidus explains recurrence, then it is difficult to understand why curettage alone is successful in so many cases. It seems inconceivable that curettage could remove a nidus in its microscopical entirety. Golding’s (1954) observation that the radiating trabeculae of the nidus are almost without fusion to the surrounding normal bone lamellae may provide an explanation of the high rate of cure from curettage. Carroll (1953) also called attention to this feature of osteoid osteoma. These observations suggest that the nidus may have a central arterial supply and that it is the interference with this supply by curettage or drilling that leads to infarction with subsequent permanent disappearance of the nidus. On the other hand, the natural history of the lesion is towards spontaneous regression, as noted by Sherman (1947) and Moberg (1951), and it may be that this proceeds despite the operation and leads to cure.

Finally, the relationship of recurrent osteoid osteoma to so-called giant osteoid osteoma (benign osteoblastoma) should be considered. Lichtenstein (1956) regarded this lesion as an osteoid osteoma of unusual size, with osteoid tissue somewhat coarser but nevertheless histologically indistinguishable from that of osteoid osteoma. He believed that a limited growth potential of giant osteoid osteoma (benign osteoblastoma) could be convincingly demonstrated. Dahlin (1967), in contrast, believed that benign osteoblastoma did not share the markedly limited growth potential of the average osteoid osteoma. If in fact an osteoblastoma has unlimited growth potential, it may be that it is this lesion—rather than osteoid osteoma—that is not cured by curettage or excision. In this context, our case is of particular interest in that the florid recurrence after the second excision suggested the greater growth potential of benign osteoblastoma though the histological appearance was more typical of osteoid osteoma.

From our experience with this case and from the review of the literature on recurrence, it seems that en bloc excision is the best treatment. It must encompass the whole tumour and
radiographic control is of the utmost importance. In our case it appeared, from radiographs after each operation, that the excision had been adequate but nevertheless the lesion recurred. Perhaps radiographic examination of the block of bone resected at operation, a technique recommended by both Aegerter and Kirkpatrick (1963) and by Lichtenstein (1965), might have indicated that excision was incomplete.

SUMMARY AND CONCLUSIONS

1. A case of osteoid osteoma which recurred twice after block excision is reported.
2. It is postulated that recurrence is almost certainly caused by incomplete removal of the nidus, either by curettage or by incomplete block excision.
3. Why curettage is successful in most cases but not in others is obscure, but it may be that the arterial supply to the tumour is interrupted.
4. Block excision with adequate radiographic control to ensure its completeness is the treatment of choice.

REFERENCES


