OSTEOGENIC FIBROMA OF BONE

Report of a Case

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In this paper we shall record a case of a tumour-like process affecting the vertebral column which, in its clinical, radiological and pathological characters resembles the lesion Lichtenstein (1952) designated osteogenic fibroma of bone. This is an uncommon condition which is liable to give rise to difficulties in diagnosis. Osteogenic fibroma is one type of a group of benign tumour-like processes of bone which are composed of essentially the same constituents, osteoid tissue and more or less calcified atypical bone in a matrix of highly vascular osteoblastic connective tissue. The members of the group present certain differences in respect of growth capacity and the reaction of adjacent tissues, and of their clinical and radiological characters (Jaffe 1935, Jaffe and Lichtenstein 1940, Lichtenstein 1952, Golding 1954, Golding and Sissons 1954). The group includes the lesion known as osteoid osteoma (Jaffe 1935, Jaffe and Lichtenstein 1940, Golding 1954), osteogenic fibroma of bone (Lichtenstein 1952, Golding and Sissons 1954), and certain rarer tumours occurring in children and young adults which may become more aggressive in the course of their development without becoming frankly malignant (Lichtenstein 1952).

CASE REPORT

In 1948 a boy of thirteen was admitted to the hospital with paraplegia of recent onset, and complaining of pain in his back. About six months previously he had fallen from the roof of a shed, but was not aware that he had injured his back, and did not complain of pain until three months later. Two days before admission his legs suddenly became weak, and this weakness became rapidly worse.

On examination he was found to have a tender lower thoracic kyphos with soft-tissue thickening on both sides, especially the right. Both lower limbs were paralysed and there was loss of sphincter control, but no loss of sensation was demonstrated. Radiographs of the spine (Fig. 1) showed a semi-translucent area in the right side of the ninth thoracic vertebra, with a suggestion of new bone formation and expansion laterally. The lesion was apparently confined to the lamina, pedicle and transverse process, without involving the vertebral body. The chest was clear. The Wassermann reaction was negative, and the only abnormality found in the blood was an erythrocyte sedimentation rate of 43 millimetres in the first hour (Wintrobe's method). This was ascribed to a urinary infection, which was already present.

Three days after admission sensory impairment was noted in the lower limbs, and this rapidly progressed in the space of two days to complete sensory loss below the level of the twelfth thoracic segment.

Operation—Laminectomy was undertaken. While the muscles were being stripped from the spinous processes and laminae, excessive bleeding was encountered on the right side, and a soft haemorrhagic tumour-like mass was found to be protruding between the laminae of the eighth and ninth thoracic vertebrae. Closer inspection showed actual invasion of these laminae. Laminectomy was proceeded with and as much of the tumour as possible was removed. The dura, which was seriously compressed but not actually invaded so far as could be seen, was not opened. The origin of the tumour was not ascertained, but it was thought
Radiograph on admission showing lesion in right side of the ninth thoracic vertebra. The vertebral body is not involved. The arrow points to a calcified mass above pedicle of T9.

Figure 2—Radiograph ten weeks after laminectomy, showing expansion and ossification of tumour.
Figure 3—Radiograph two and a half years later.
that it possibly arose in the pedicle, and from its naked-eye appearance the tumour was considered to be almost certainly malignant. After operation the boy was nursed on a plaster bed, and the spinal lesion was treated by x-ray therapy.

Progress—Ten weeks after operation radiographs showed lateral expansion of the tumour and well marked ossification throughout (Fig. 2). The clinical condition remained unchanged, and with the gloomiest of prognosis he was allowed home, where his mother continued to nurse him on the plaster bed.

After a few months he began to show signs of recovery of cord function, and as his general condition continued to be good he was readmitted to hospital. At that time both lower limbs were very spastic, but he had regained a little voluntary control of feet and knees, sensory loss had diminished in extent and degree, and he was no longer incontinent of faeces. Radiographs now showed the spinal lesion to be sclerosed with sharply outlined borders, and bulging quite far to the right of the vertebral body.

![Figure 4](image1.png) ![Figure 5](image2.png)

Figure 4—Cellular connective tissue showing active differentiation into irregular bands of osteoid tissue. (× 55.) Figure 5—Cellular connective tissue showing differentiation into irregular bands of osteoid tissue. No notable degree of cytological irregularity. (× 140.)

After a second course of radiotherapy to the spine, every effort was made to rehabilitate him so far as possible by physiotherapy, neurectomies and braces, and after three years he was able to walk with elbow crutches and without calipers. Radiographs showed progressive ossification of the spinal lesion (Fig. 3). When he was last seen seven years after the onset of symptoms he remained in good general health, and there was no clinical or radiological evidence of metastases.

Pathological examination of excised material—The material consisted of tumour tissue and pieces of adjacent bone. Macroscopically, the tumour tissue was soft, haemorrhagic and "gritty" to the touch. A little soft tissue, similar in character, was attached to the pieces of bone.

Histological examination—Tumour tissue: this consisted of highly cellular and vascular connective tissue mixed with recent blood-clot. In many parts of this cellular tissue active differentiation into irregular areas and bands of osteoid tissue was in progress (Figs. 4 and 5). In other parts the cellularity was more marked and differentiation much more irregular, and
in such areas there was often a deposition of calcium salts on highly irregular spicules of bone (Fig. 6). Scattered throughout the tissue osteoclasts were present in moderate numbers (Figs. 4, 8 and 9). Mitoses were present but they were not numerous.

![Figure 6](image1)

**Figure 6** - Markedly cellular and atypical tissue containing irregular spicules of calcified bone. (× 140.)

![Figure 7](image2)

**Figure 7**—Normal bone (left) bounding tumour tissue (right). (× 55.)

![Figure 8](image3)

**Figure 8**

Tumour tissue infiltrating cancellous spaces of normal bone. (× 140.)

![Figure 9](image4)

**Figure 9**

Pieces of adjacent bone: this consisted chiefly of normal bone (Fig. 7), some of which was newly formed. At parts of the inner surface of the bone and extending into the cancellous spaces, there were plaques of cellular osteoblastic tissue similar in character to the tumour tissue (Figs. 8 and 9).
At the time of the surgical exploration diagnosis in this case was difficult. At operation the lesion appeared to be a tumour invading bone and probably malignant. Microscopic examination of the material suggested either an osteogenic sarcoma or active and disorderly osseous granulation tissue. Although there was clear evidence of invasion of bone, the stromal cells did not exhibit the degree of cytological irregularity to be expected in osteogenic sarcoma, and mitoses were not numerous. On the other hand, there was no apparent explanation of the presence of osseous granulation tissue. From the subsequent history of the patient it seems clear that the lesion was not an osteogenic sarcoma. The radiographic appearances did not suggest giant-cell tumour of bone, and the histology of the biopsy material was quite unlike giant-cell tumour.

The diagnosis of osteogenic fibroma in this case depends on the absence of any discoverable lesion to explain the presence of osseous granulation tissue, on the clinical course which excludes osteogenic sarcoma, and on the similarity of the clinical, radiographic and pathological features to those in other recorded cases of osteogenic fibroma (Lichtenstein 1952, Golding and Sissons 1954).

As indicated above, osteogenic fibroma of bone is one type of a group of benign osteoblastic tumour-like processes of bone (Lichtenstein 1952). The scope and status of the whole group, and the relationship, if any, between the members of the group are not yet clearly defined.

Osteoid osteoma was formerly regarded as a chronic inflammatory process of bone, and still is so regarded by some observers (Brailsford 1942). Jaffe (1935), who made a careful study of the lesion, concluded that it is a benign tumour of bone, and this view has now gained fairly wide acceptance (Jaffe and Lichtenstein 1940, Jackson et al. 1949, Golding 1954). As pointed out by Coley (1949) and Willis (1953), however, the status of the lesion is still debatable.

In some respects osteoid osteoma and osteogenic fibroma present a close similarity, but Lichtenstein (1952) and Golding and Sissons (1954) believed that the two processes were different. In two cases classified as osteogenic fibroma by Golding and Sissons (1954), in both of which the lesion occurred in the vertebral column, these authors found that the lesions differed from osteoid osteoma in several respects. They did not cause the severe pain characteristic of osteoid osteoma, and they were considerably larger and showed progressive enlargement with compression of adjacent soft tissues. These features, with the absence of a zone of sclerosis surrounding the lesion, resulted in a totally different radiographic appearance. Pathologically, Golding and Sissons (1954) found in the lesions of osteogenic fibroma the same tissue components as in osteoid osteoma, but the proportions of each and their arrangement were different. In particular, the continuous network of bone and osteoid tissue which make up the "nidus" of osteoid osteoma was absent, and there was much more fibrous tissue. In the case reported here the clinical, radiographic and pathological features resemble those of osteogenic fibroma. Pathologically, the structure of the material was distinctly more irregular and imperfectly differentiated than is usual in the "nidus" of osteoid osteoma, and there was no dense sclerosis of bone adjacent to the tumour-like tissue.

There seems to be a close relationship between osteogenic fibroma of bone and the histologically similar but rare tumours referred to by Lichtenstein (1952) as benign osteoid-tissue-forming tumours of bone. These tumours occur in children and young adults, show a predilection for the bones of the hand and foot, and may become aggressive in the course of their development "and yet fall short of being osteogenic sarcoma."

From our studies of the literature and of the case recorded above, we incline to the view that the lesion described is a benign neoplasm rather than a manifestation of bone dysplasia, and we have accordingly employed the designation osteogenic fibroma of bone. But it is clear that more studies of similar cases are required to establish the true nature of the condition.
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REFERENCES


