ANEURYSMAL BONE CYST

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Aneurysmal bone cyst is a benign lesion of bone. It was first described by Jaffe and Lichtenstein in 1942 and consists essentially of fibrous tissue honeycombed by vascular spaces. The lesion has been observed in many parts of the skeleton but it occurs especially in the shafts of long bones and in the vertebral column. It causes localised distension and destruction of the affected bone, limited peripherally by a thin bony shell (Fig. 1).

In the past the lesion has usually been regarded as an atypical giant-cell tumour or a cavernous haemangioma of bone. The purpose of this paper is to support the contention of Lichtenstein (1950) that aneurysmal bone cyst is a clinical and pathological entity, which deserves wider recognition than it has had hitherto.

The case reports that follow illustrate some of the more important features of the lesion.

CASE REPORTS
Case 1 (Reported by the courtesy of Mr D. Lloyd Griffiths)—A girl of thirteen complained of a painful swelling of the upper end of the right ulna of three months duration, which had increased rapidly in size. There was no history of injury. Examination revealed a hard swelling arising from the upper end of the ulna, which had a nodular surface. The swelling was warmer than the surrounding tissues, but there was no pulsation. Radiography disclosed a cyst arising from the anterior surface of the upper third of the ulna. It extended from the base of the coronoid process for 6.5 centimetres down the shaft of the bone (Fig. 1). The cyst had destroyed the anterior cortex of the ulna and invaded the medullary canal; it was bounded anteriorly by a thin bony shell.

Operation (22.3.49)—The cyst was excised together with the upper end of the ulna.

Histology—Pathological examination revealed numerous large cystic spaces containing fresh blood (Fig. 2). The walls of the cyst were lined by cellular connective tissue in which there were numerous giant cells.

Progress—The head of the radius dislocated anteriorly, and an attempt to retain the reduced position was unsuccessful. Later the upper end of the ulna was replaced by an acrylic prosthesis. The patient has now good function in the arm and is training as a nurse. There has been no recurrence of the cyst.

Case 2—A girl of seventeen felt a sharp pain in the lower part of the back while pushing a door against resistance. The pain persisted until her admission to hospital two years later; it sometimes radiated down the posterior aspect of the thigh to the leg, and was aggravated by coughing, sneezing and exercise. Clinical examination revealed restriction of flexion and right lateral bending in the lumbar spine and tenderness to the right of the lumbo-sacral joint. Radiographs disclosed destruction of the spinous process, lamina and right superior and inferior articular processes of the fifth lumbar vertebra. There was also erosion of the transverse process and the adjacent part of the body of this vertebra, and of the upper part of the ala of the sacrum on the right side (Figs. 3 and 4).

Biopsy revealed what was thought to be a vascular "tumour" lying deep to erector spinae, and later reported to have the histological features of an aneurysmal bone cyst.

Operation (19.3.54)—The "cyst" was exposed; it was deep reddish brown in colour, and well demarcated from the surrounding tissues. Almost the whole of the neural arch of the fifth lumbar vertebra was destroyed, and only the tip of the spinous process remained, loosely attached to its surface (Fig. 5). The cyst contained friable vascular tissue which was removed partly by curettage and partly by suction. The dura was now exposed and the fifth lumbar nerve root could be seen emerging from it on either side. The bone in the floor of the cyst was smooth; its appearance suggested erosion by pressure rather than destruction by tumour (Fig. 6).

Histology—The specimen consisted of many fragments of soft, highly vascular, reddish stranded connective tissue. Microscopy showed fibrous tissue, in some areas dense and collagenous, but for the most part cellular and with strands of osseous metaplasia. Running throughout were wide vascular channels containing fresh blood, except for a few where newly formed thrombi were found. Giant
Case 1. Figure 1—Radiograph of a typical lesion in the upper end of the ulna (Case 1). The cyst has caused marked "ballooning" of the bone, and is bounded anteriorly by a thin shell of subperiosteal bone. Figure 2—Section through the lesion showing gross appearance.

Cells were numerous, especially at the borders of the trabeculae, and tended to be smaller in size than those in a giant-cell tumour. The appearances were those of aneurysmal bone cyst.

Progress—The patient was examined fifteen months after operation. She then complained of slight aching pain in the lower part of the back after exercise. There was free movement in the lumbar spine. The radiograph showed normal density of the ala of the sacrum, and partial regeneration of the neural arch. The right transverse process and body of the fifth lumbar vertebra had reossified (Fig. 7).

Case 3 (Reported by the courtesy of Mr J. E. Paterson)—A man of twenty complained of left-sided sciatica. The pain commenced after an injury at rugby nine months earlier, and for the past month had been severe enough to compel rest in bed. Clinical examination revealed guarded movement of the lumbar spine and tenderness over the left sacro-iliac joint. Laségue's sign was positive at
Case 2—A lesion of the fifth lumbar vertebra and sacrum as seen from behind. Almost the whole of the neural arch, and part of the body and right transverse process of the fifth lumbar vertebra, are destroyed.

Case 2—An oblique radiograph to demonstrate the destruction of the neural arch of the fifth lumbar vertebra and the cyst in the right ala of the sacrum.
Case 2—Diagrams showing the appearance of the cyst at operation (Fig. 5) and the degree of bone destruction (Fig. 6). The dura is exposed in the floor of the cyst.

Case 2—A radiograph three months after operation, for comparison with Figure 2. There is evidence of healing. The lesion in the transverse process of the fifth lumbar vertebra is now more obvious due to the calcification of its bony shell.
5 degrees on the left side and at 45 degrees on the right. The left ankle jerk was depressed. There was no other abnormal neurological sign. No lesion was observed in the radiographs, and the pre-operative diagnosis was a lumbo-sacral disc protrusion.

Operation (27.10.54)—When the muscles were being stripped from the spinous processes a large cyst arising from the upper part of the sacrum was opened. Its posterior wall was no thicker than an egg shell, and it was lined by a thin layer of vascular tissue which peeled easily from the sides of the cavity. The contents of the cyst were not removed completely, but its deeper part probably involved the dura. Progress—The patient complained of severe pain in the leg for the first few days after the operation, but thereafter his convalescence was uneventful. When discharged from hospital three weeks later he was free from pain and walking in comfort. A further study of the pre-operative radiographs disclosed destruction of the upper part of the sacrum on the left side.

![Fig. 8]

Case 3—Cyst in the left ala of the sacrum eight months after operation. Its outline is much clearer than in the pre-operative radiograph, due to the calcification of the bony shell.

The patient was re-examined eight months after the operation. He then complained of a little weakness of the back when lifting heavy weights. There was a good range of movement in the spine, and Lasègue's sign was negative. A radiograph showed commencing reossification of the cyst in the left ala of the sacrum (Fig. 8).

Case 4—A boy of eight received a kick on the left leg three weeks before admission to hospital. A swelling appeared, but was not painful. Examination revealed a firm non-tender fusiform swelling of the lower third of the tibia. A radiograph disclosed a cystic lesion of the lower part of the shaft of the tibia, which had produced some expansion of the bone (Fig. 9). The cyst was finely trabeculated and had a thin bony shell.

Operation and progress—The cyst was opened and contained bloodstained fluid, and a thin lining membrane which was not removed completely. The patient was traced fourteen years later; he had no complaints. A radiograph showed a small cyst in the lower part of the tibia but the rest of the bone was normal in appearance. The sections were re-examined recently and are typical of an aneurysmal bone cyst.
Case 4—Radiograph of a lesion of the left tibia. The uniform expansion of bone and the degree of involvement of the medullary canal are unusual.

Case 5—(Reported by the courtesy of Mr. E. W. Somerville)—A boy of fourteen complained of pain in the right leg of six months duration, and of a swelling which had appeared some time later. There was no history of injury. Examination revealed a tender swelling of the posteromedial aspect of the upper third of the tibia. Radiographs disclosed a cyst which had caused a localised expansion of the posterior cortex of the tibia, bounded posteriorly by a thin shell of subperiosteal bone (Fig. 10). The cyst extended within the medulla to the upper tibial epiphysis.

Biopsy—The cyst was lined by vascular osseous tissue. Histological examination showed connective tissue with numerous haemorrhages, multinuclear giant cells, and fragments of bone. The fluid from the centre of the cyst contained red blood cells and a few leucocytes. The diagnosis was a haemorrhagic bone cyst of tibia.

Treatment and progress—The cyst was re-explored on 21.8.55 and the lining carefully removed by curettage. The cavity was packed as far as possible with slivers of bone taken from the opposite tibia. The post-operative progress was satisfactory. Radiographs showed progressive consolidation of the grafts, and no extension of the cyst.

Apart from the contributions of Jaffe (1950) and Lichtenstein (1950, 1953) there are as yet only a few isolated case reports of aneurysmal bone cyst in the surgical literature. This may be explained partly by lack of familiarity with the clinical and radiographic features of the disease, and partly by the rarity of the lesion. The account which follows is based on a
review of the relevant literature, and reports of atypical giant-cell tumours which may now be reclassified as aneurysmal bone cysts.

**CLINICAL FEATURES**

*Age and sex incidence*—Most of the patients are children, adolescents, or young adults. Both sexes are affected, males more frequently than females.

*Distribution*—There is a preference for the vertebral column and the shafts of long bones. The lesion occurs at all levels in the spine. The neural arch is involved oftener than the body of a vertebra. In the long bones the cyst is usually situated beneath the periosteum of the shaft and rarely invades the epiphysis. Aneurysmal bone cysts have also been described in most of the flat bones, skull, and bones of the hands and feet. Contiguous bones are often affected, especially in lesions of the spine (Figs. 3 and 4).

*Symptoms*—Pain as a rule is not severe, but it may be if the collapse of a vertebra has caused pressure on a nerve root (Case 3). The symptoms often date from a specific injury. This may have drawn attention to a pre-existing lesion, but this is not always so, for sometimes a radiograph taken shortly after injury was negative.

*Clinical signs*—When the cyst is in the spine there is always some restriction of movement. It may be considerable if there is destruction and collapse of a vertebra, when paraplegia is a possible complication. In the long bones the swelling is often palpable; it may become very large and interfere with the function of the limb.

![Fig. 10](image)

Case 5—Radiograph of a typical lesion of the right tibia. Note the "ballooning" of the posterior cortex.

**RADIOGRAPHIC FEATURES**

In a long bone the lesion appears as a cyst arising from the cortex of the shaft, and limited peripherally by a thin shell of subperiosteal bone (Fig. 1). There is usually some destruction of the underlying cortex, but the cyst does not as a rule invade the medulla, and only rarely is there uniform expansion of the affected bone (Fig. 9). The cyst frequently has a mottled or trabeculated appearance, and there may be irregular strands of ossification which are sometimes a feature of older lesions. In the spine the picture is often obscure, because it may be difficult to demonstrate the bony shell, and the osteolytic process is then likely to be attributed to a primary or metastatic tumour (Figs. 3 and 4). These tumours are, however, usually situated in the body of a vertebra, whereas an aneurysmal bone cyst is found more frequently in the neural arch or transverse process.

**PATHOLOGY**

When the bony shell of the cyst is opened one is confronted by strands of deep reddish brown vascular tissue honeycombed by spaces filled with blood (Fig. 2). Blood wells out of the cavity, indicating a free circulation within the cyst, but there is seldom any pulsation.
or brisk arterial bleeding. Small strands of fibre bone may be interspaced throughout the tissue and give it a gritty feel. Sometimes the tissue is less vascular and has a greyish appearance; it is then distinctly more collagenous, suggesting an attempt at repair.

**Fig. 11**
Section showing the topography of the lesion. There are many large vascular spaces separated by strands of fibrovascular tissue. (H. and E., × 45.)

**Fig. 12**
Section showing strands of fibrovascular tissue containing multinucleated giant cells. (Masson stain, × 270.)

In a typical lesion histological examination reveals large vascular spaces separated by strands of fibrous tissue in which there are numerous capillaries (Figs. 11 and 12). Multi-
nucleated giant cells are common around areas of haemorrhage; these giant cells are smaller and have fewer nuclei than those of a giant-cell tumour. Filaments of osteoid tissue and new bone are often present in the fibrous tissue (Fig. 13).

![Image](https://example.com/image13.png)

**Fig. 13**

Two large vascular spaces are seen in the upper part of the field. The tissue lining the space on the left contains numerous giant cells. In the lower part of the field there are filaments of osteoid tissue surrounded by osteoblasts. (H. and E., × 58.)

**DIAGNOSIS**

The lesion which has been described has long been recognised. Ewing (1940) referred to it as an aneurysmal giant-cell tumour, and Coley (1949), Thompson (1954) and others as an atypical or subperiosteal giant-cell tumour. Apart from the presence of giant cells, aneurysmal bone cyst has little resemblance to a typical giant-cell tumour. The main points of difference between them are indicated in Table 1.

**TABLE 1**

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<th>Aneurysmal bone cyst</th>
<th>Giant-cell tumour</th>
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<tr>
<td><strong>Age</strong></td>
<td>Usually children and young adults</td>
<td>Patients usually over thirty years</td>
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<tr>
<td><strong>Site</strong></td>
<td>Shafts of long bones and vertebrae (neural arch). May involve contiguous bones</td>
<td>Epiphysial end of long bones especially upper end of tibia and lower end of femur</td>
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<td><strong>Appearance</strong></td>
<td>In long bones is usually situated eccentrically beneath periosteum</td>
<td>Expands bone from within</td>
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<td><strong>Pathology</strong></td>
<td>Fibrovascular tissue honeycombed by vascular spaces. Giant cells small and not present throughout tissue</td>
<td>Large numbers of multinucleated giant cells in vascular spindle-celled stroma</td>
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<tr>
<td><strong>Behaviour</strong></td>
<td>Strong tendency to spontaneous regression. Incomplete removal often followed by cure</td>
<td>Invasive and aggressive. Sometimes malignant from onset</td>
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A simple or unicameral bone cyst is situated in the centre of the metaphysis, and seldom causes pain unless there is a spontaneous fracture. It contains yellowish fibrous tissue which

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is firmly attached to the walls of the cyst, and amber coloured fluid which is not bloodstained unless there has been a recent fracture.

A number of aneurysmal bone cysts have also been incorrectly diagnosed as haemangiomas. A true angioma is situated within the medulla of a long bone, or the vertebral body. It produces a cystic lesion of the bone with a marked "soap bubble" effect. Microscopic examination reveals a large number of dilated capillaries lined by endothelium in a loose connective tissue stroma; the formation of osteoid tissue and new bone is not a feature.

When the lesion is in the shaft of a long bone it may have some resemblance to an osteolytic osteosarcoma or a malignant bone aneurysm. Both these tumours are intensely painful; the area of bone destruction is not so well defined, nor is there the trabeculation of an aneurysmal bone cyst, and the tumour quickly invades the soft tissues and destroys the bony shell.

**TREATMENT**

All case reports agree that the lesion is benign. Lewis (1924) reviewed seventeen giant-cell tumours of the spine, of which many might now be reclassified as aneurysmal bone cysts. Recovery occurred spontaneously, or after partial removal of the tumour, in thirteen of the seventeen patients. Lewis stated that he had doubt as to the neoplastic nature of some of the tumours because of their clinical behaviour and the tendency to spontaneous ossification. Coley (1949) described an atypical subperiosteal giant-cell tumour which had a uniformly good prognosis. Radiotherapy has also given completely satisfactory results, but sometimes the recalcification of the lesion occurred so soon after treatment that it cannot have made any significant contribution to the cure (Brock and Bogart 1945).

It is evident that many aneurysmal bone cysts have an inherent tendency to spontaneous regression, but some become so large that conservative treatment is no longer practicable (Lichtenstein 1953). For this reason they should be treated by excision or curettage as soon as possible. Cysts in the long bones can usually be excised completely, but curettage is the only method for lesions of the neural arch or of the spinous and transverse processes. Operative treatment may be feasible when the cyst is in the body of a thoracic or lumbar vertebra; and even if the curettage is incomplete the results are usually satisfactory. Radiotherapy has the disadvantage of not providing histological confirmation of the diagnosis and should be reserved for inaccessible lesions.

**DISCUSSION**

Jaffe (1950) was doubtful whether aneurysmal bone cyst was an entity. He suggested that it might follow "a haemorrhagic blow-out in a pre-existing benign bone lesion, in the course of which the original lesion may have been destroyed." There is, however, no evidence in the cases reported here or elsewhere of a parent lesion, and it is unlikely that even a severe haemorrhage could have destroyed all trace of a previous pathological process. Furthermore the common sites for aneurysmal bone cyst are distinctly rare for any other benign lesion of bone. The only feature common to a giant-cell tumour and an aneurysmal bone cyst are the multinucleated giant cells. In all other respects the two conditions are quite distinctive (Table 1).

The remarkable feature of an aneurysmal bone cyst is the tendency to heal after incomplete removal or even without treatment, which suggests that it may not be a tumour. Is it then an abnormal response to injury? There is much to support this suggestion. In many of the reported cases, though not in all, the patient's symptoms date from a precise injury. In some a radiograph taken shortly after injury was negative (Bogart and Imler 1947), refuting the assumption that the injury drew attention to a pre-existing lesion. There is a close resemblance between some of the healing cysts and an ossifying haematoma (Cone 1928), and it is possible that the lesion is a peculiar response to a circumscribed subperiosteal haemorrhage. The presence of osteoid tissue and fibre bone in the lesion is an indication of an attempt at repair.
Although there are good reasons for regarding trauma as an etiological factor, it cannot be the only one, for if it were the lesion would certainly be more common.

Since the pathogenesis of the lesion is obscure, the term aneurysmal bone cyst is as satisfactory as any. It is non-committal and describes with reasonable accuracy the pathological features of a well defined disease process which merits individual recognition and treatment.

SUMMARY

1. The clinical, radiographic and pathological features of aneurysmal bone cyst are described and illustrated by case reports.
2. Reasons are given for accepting the lesion as a clinical and pathological entity.
3. The cyst has a tendency to spontaneous regression and healing may occur after partial excision.

My thanks are due to those surgeons who have given me the opportunity of examining the case records of patients under their care, and to Mr Gabriel Donald and Miss Dorothy Davidson for the illustrations. I am especially indebted to Dr M. E. Catto and Dr W. A. Taylor of the Pathology Department of Glasgow University. They first drew my attention to the lesion and were also responsible for the histological preparations.

REFERENCES