PERIOSTEAL GANGLION

PAUL D. BYERS and THOMAS G. WADSWORTH, LONDON, ENGLAND

From the Institute of Orthopaedics, London, and Metropolitan, St Leonard's and Hackney Hospitals, London

We describe here four cases with a cystic lesion over the subcutaneous surface of the tibial shaft, a condition which has been called "periosteal ganglion".

Tillmanns (1895) quoted Ollier (1864) and Poncet (1874) as the first to describe a peculiar form of periostitis which they termed "periostitis albuminosa" or "ganglion periostale": it was stated that a tenacious mucoid fluid lies either beneath the periosteum, or on its outer surface, or within it in the form of a cyst.

Reidinger (1887), quoted by Clarke (1908), aspirated a cyst in the periosteum over the tibia in an old man on two occasions and it refilled for the third time within ten days. He considered it to be the same as a ganglion of the tendon sheath and thought that under certain circumstances the periosteum could secrete the same material as the tendon sheath.

Thin-walled cysts containing synovial-like fluid occurring either in the periosteum or in bones themselves have been described by Schlange (1887), Tixier (1903) and Deutschlander (1905). Fisk (1949) described a ganglion overlying the medial malleolus and lying in a cup-shaped depression in the bone; the floor of the cyst was formed by periosteum and it was thought that the ganglion had arisen from the periosteum and caused erosion of the bone before bursting into the subcutaneous tissue. Woods (1961) and Crabbe (1963, 1966) described several cases of subchondral bone cyst. These lesions, histologically similar to simple soft-tissue ganglia, lie immediately beneath subchondral bone with a surrounding area of sclerosis which may be striking. There was no degeneration of cartilage overlying these, and intact cysts did not show a communication with the joint cavity.

In our cases the lesions have been present over the shaft of the bone and have in no way been connected with ligamentous or joint structures. In two of the cases the bulk of the altered periosteum had been elevated from the cortical bone and the floor has been the almost denuded cortex. There was roughening of the underlying cortical bone in two of the four cases. Recurrence of the lesion followed removal in two patients, a cure being effected in one by a second operation.

CASE REPORTS

Case 1—A man aged thirty-eight years had complained of pain along the length of the left tibia three months previously. A few days after the onset of pain he noticed a swelling, but no erythema. A short course of penicillin given by his general practitioner was associated with cessation of the pain. Nevertheless, the swelling persisted and in fact increased in size for some weeks but he remained free from pain.

On examination there was a painless, fluctuant, firm swelling overlying the subcutaneous surface of the left tibia at the junction of the upper and middle thirds; the swelling measured 3.5 by 2.5 centimetres, and appeared to be attached to bone. There was no erythema or adenopathy and the radiographs showed no significant abnormality.

At operation the swelling was found to be due to a cyst beneath thickened periosteum. This was incised and clear gelatinous fluid escaped. The thickened area of the periosteum was excised and a small amount of soft tissue scraped from the base of the cyst. The exposed cortical bone appeared to be normal; it seemed that the bulk of the periosteum had been raised from the cortex. The post-operative course was uneventful and six years later there had been no further symptoms.

Histological sections showed a densely collagenous membrane with one smooth surface and one surface covered by fibro-fatty tissue. The smooth surface, which formed the lining of the cavity, was not covered by any specialised cells. However, within the wall there was a focus of mucoid degeneration of the collagenous tissue with the early formation of subsidiary cysts. There was a small amount of immature bony tissue incorporated within the ganglion wall at one point.

FIG. 1

Case 2—The swelling over the right tibia before operation.

FIG. 2

FIG. 3

Case 2. Figure 2—The lesion exposed at operation: the opalescent swelling in periosteal tissues is evident. Figure 3—The cup-shaped depression from which the cyst was removed: the edges are formed by thickened fibrous tissue. The base seemed to be bone but this was not the case histologically in Case 3 (Fig. 5).

Case 2—A man aged forty-two years had noticed a swelling over the front of the right tibia for a year. It had varied in size from time to time but he had not been bothered by other symptoms.

On examination there was a fluctuant swelling which appeared to be attached to the lower part of the upper third of the subcutaneous surface and anterior border of the right tibia (Fig. 1); the swelling felt lobulated. The radiographs were normal.

At operation a multiloculated cyst was found, the main part measuring 2.5 centimetres in diameter, lying beneath the periosteum which was thickened particularly at the periphery of the swelling (Fig. 2). The periosteum was incised and gelatinous fluid evacuated. The floor of the cyst was slightly roughened cortical bone; the possibility that there was a tenuous soft-tissue covering was not explored. There was a tail-like extension of the ganglion over the epimysium of the tibialis anterior muscle (Fig. 3). The thickened periosteum was excised.

The patient progressed well at first, but about four months after operation he developed further swelling which he himself attributed to the fact that he had been massaging the scar. The swelling this time was far more diffuse than on the first examination.

At a second operation a much wider area of thickened periosteum was found over the subcutaneous surface of the tibia and this altered periosteum was excised. There has been no further recurrence three years later.

The specimen from the first operation was a flattened cystic structure 5 by 1 centimetres with a wall about 1 millimetre thick. Histological sections reveal small subsidiary cysts and little foci of mucoid degeneration in fibro-fatty tissue outside the wall of the main cavity. The wall of each cystic space is dense collagenous tissue; the inner layer of cells is sometimes prominent owing to their flattened cuboidal shape. Strips of fibrous tissue 1 to 4 centimetres in length and a few millimetres thick were removed from the second specimen together with a dozen minute fragments of bone from the base of the cyst. Some of the fibrous strips had one smooth surface that formed the cavity wall. The pieces from the base of the lesion were a mixture of reactive and normal bone trabeculae mixed with fibrous tissue. Owing to the fragmented nature of the material definite statements are difficult, but it seems that the bone proper was separated from the cyst cavity by a thin fibrous layer and that this formed the deep wall or "base" of the cyst.

Case 3—A man aged thirty-seven years came with a swelling over the subcutaneous surface of the left tibia which he first noticed some weeks after a motor scooter accident. He was referred for treatment when the swelling had been present for several months and had been aspirated on three or four occasions; the swelling had recurred soon after each aspiration. There was a non-tender tense cystic swelling over the subcutaneous surface of the left tibia.

At operation an ovoid cystic mass 4 by 2 centimetres in size was found to be attached to the tibia; removal of the cyst uncovered rough cortical bone and a portion of this was removed. The patient was not seen again after his discharge.

The specimen contained clear mucinous fluid held in loculated spaces (Fig. 4). Histologically the dense fibrous wall formed one large cavity with only a few small subsidiary cysts in it. The loculated appearance seen on gross examination was due to the presence within the cyst cavity of fibrinous material which contained pools of mucinous substance. Some of the fibrinous material in contact with the wall was being organised. In other places this same material contained quite heavy collections of lymphocytes and plasma cells with a few polymorphs. The sections of the bone that were removed from beneath the cyst showed a thin layer of cortical bone covered by a zone of reactive bone formation which had nodules of ossifying hyaline cartilage at its surface (Fig. 5). These reactive tissues were overlain in their turn by a thin fibrous layer which must have been continuous with the external aspect of the cyst wall.

Case 4—A man aged fifty-eight years presented with a swelling over the left tibia which he had noticed for one week; he had had pain for two weeks before the swelling appeared. Examination showed a firm, smooth swelling, 2 centimetres in diameter, fixed to the left tibia.

At operation the swelling proved to be a cyst in the periosteal tissue filled with clear, thick fluid. The involved periosteum was excised. Gelatinous yellow fluid was aspirated from the

FIG. 4

FIG. 5

FIGS. 4 AND 5

Case 3. Figure 4—The excised cyst showing its ovoid shape and multiloculated structure. Figure 5—Photomicrograph of tissue beneath the periosteal ganglion. The dense cortical bone is covered by actively forming reactive bone and cartilage tissue; the latter is undergoing ossification. (Haematoxylin and eosin, $\times 63$.)

FIG. 6

Case 4—Photomicrograph of the ganglion. Parts of two large spaces are seen, surrounded by dense fibrous tissue and lined by a single layer of low cuboidal cells. In the looser connective tissue three foci of mucoid degeneration are present, giving rise to small spaces surrounded by a condensation of collagen fibres. (Haematoxylin and eosin, $\times 93$.)

site of excision several weeks later. In the ensuing year the swelling appeared and disappeared on two occasions. It reappeared a third time sixteen months after operation and persisted, but as the patient was not bothered no further action was taken.

On gross examination the specimen appeared to be a sheet of tissue measuring 20 by 10 by 5 millimetres. However, the sections reveal several cystic spaces in the fibrous tissue which was undergoing widespread mucoid degeneration. Several of the spaces were lined by prominent flattened cells (Fig. 6).

DISCUSSION

Carp and Stout (1928) reported their experience with 255 ganglia, none of which were located in the periosteum although they mention this as having been reported. Despite the fact that the majority of the lesions recorded by these authors were in the neighbourhood of joints, they had never found evidence that the lesions communicated with joints. Experience of ganglia in the Department of Morbid Anatomy at the Institute of Orthopaedics, where several hundreds have been examined, agrees with this and applies equally to lesions at such sites as the wrist, ankle or knee (mainly cystic meniscus).

The pathogenesis of these lesions is mucoid degeneration of connective tissues. The stimulus to this is not known. The fibroblasts produce intercellular mucin which forms pools that coalesce to form one or several cysts. The surrounding tissue is compressed by the accumulating fluid but there is also a fibroblastic proliferation and collagen formation which results in a fibrous wall. The cells on the inner aspect of the wall often become somewhat cuboidal; although this appearance suggests a relationship to synovium, clearly this is not the case. The foregoing pathogenetic mechanism occurs in a number of sites in the body and results in mucin-filled cysts. In soft tissues these are ganglia (Carp and Stout 1928) and in bone they have been called subchondral bone cysts (Woods 1961) because this is their usual location. The lesions in bone seem to depend in the first instance on the development of focal marrow fibrosis within which the degenerative process takes place. Nevertheless it is a widely held belief that these osteolytic lesions, particularly in degenerative arthritis, result from the penetration of synovial fluid through microfractures. Landells (1953) purported to establish this by showing cysts communicating with the joint space in osteoarthritis. He assumed that those osteolytic foci filled with solid fibrous tissue arose through filling of the cystic space by fibrosis. With an experience of large numbers of degenerate joints it is not difficult to establish that it is at least equally probable that the fibrous tissue appears first, to be followed by mucoid degeneration, and that the communication with the joint space is established subsequent to this. This is also supported by examination of subchondral cysts such as have been reported by Woods (1961). The purpose in referring to these lesions in arthritic and non-arthritic bones is to emphasise the fact that there are in many parts of the body fluid-filled lesions which share a common pathogenesis and pathology.

Despite the gross appearances at operation, from the pathological examination it is evident that the base of the periosteal lesions is not formed by cortical bone and that the cysts form within rather than beneath the periosteum. So far as can be judged from the one adequate specimen (Case 3) the bone beneath the affected site is apparently not always resorbed (Fisk 1949) so as to produce a cavity, but reacts by the production of new bone. But this is a point which can well be studied in other cases in the future.

Recurrence of these lesions does not necessarily come about through inadequate excision. The mucoid degenerative process could well commence after operation in the connective tissues occupying the surgical defect or lying immediately adjacent to the operative site. The clinical observation that these lesions (and those at other sites) can come and go, together with the pathological observation of organising fibrinous material in Case 3, suggests not only that the fluid within the cyst may be absorbed, but also that the cavity may be closed by organisation of a fibrin content.

SUMMARY

1. Four cases of periosteal ganglion are described. Their relationship to other mucin-filled cysts of connective tissue, both in soft tissues and in bone, is discussed and the fact that they all result from mucoid degeneration of fibrous tissue is emphasised.
2. Treatment is by surgical excision, with removal of a margin of apparently normal periosteum as well as the lesion itself. The lesion may recur, even after apparently adequate excision, by mucoid degeneration taking place in the periosteum immediately adjacent to the operative site or in the connective tissues occupying the surgical defect.

We wish to express our appreciation to Mr Donal Brooks for permission to include one of his patients (Case 3).

REFERENCES

- CARP, L., and STOUT, A. P. (1928): A Study of Ganglion. *Surgery, Gynecology and Obstetrics*, **47**, 460.
- CLARKE, W. C. (1908): The Pathogenesis of Ganglia. *Surgery, Gynecology and Obstetrics*, **7**, 56.
- CRABBE, W. A. (1963): The Diagnosis of Some Cystic Conditions of Bone. *Guy's Hospital Reports*, **112**, 66.
- CRABBE, W. A. (1966): Intra-osseous Ganglia of Bone. *British Journal of Surgery*, **53**, 15.
- DEUTSCHLANDER (1905): Einen Fall von Ostitis aluminosa des rechten Radius. *Deutsche medizinische Wochenschrift*, **31**, 932.
- FISK, G. R. (1949): Bone Concavity Caused by a Ganglion. *Journal of Bone and Joint Surgery*, **31-B**, 220.
- LANDELLS, J. W. (1953): The Bone Cysts of Osteoarthritis. *Journal of Bone and Joint Surgery*, **35-B**, 643.
- OLLIER (1864): Quoted by Poncet, M. A.
- PONCET, M. A. (1874): De la périostite albumineuse. *Gazette Hebdomadaire de Médecine et de Chirurgie*, **11**, 133 and 179.
- REIDINGER (1887): Ueber Ganglien periostale. *Festschrift. F. A. V. Kolliker, Leipzig* (Quoted by Clarke, W. C.).
- SCHLANGE (1887): Ueber einige seltenere Knochenaffectionen. I. Die sogenannte Periostitis und Ostitis aluminosa. *Archiv für klinische Chirurgie*, **36**, 97.
- TILLMANN, R. H. (1895): *Principles of Surgery and Surgical Pathology*, pp. 580 and 643. New York: D. Appleton and Company.
- TIXIER (1903): Périostite Albumineuse du Fémur d'origine probablement syphilitique. *Lyon Médical*, **100**, 619.
- WOODS, C. G. (1961): Subchondral Bone Cysts. *Journal of Bone and Joint Surgery*, **43-B**, 758.