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BENIGN GIANT CELL TUMOUR OR LOCALISED OSTEITIS FIBROSA CYSTICA

BY

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The name applied to this case may be disputed, since a variety of names has been used in the past for the condition now called *Giant cell tumour,* viz.: *benign giant cell sarcoma, giant cell tumour, primary giant cell tumour, endosteal myeloid sarcoma, myeloid disease, myeloid sarcoma,* and *osteoclastoma.* According to Fleischer-Hansen the conceptions of benign giant cell tumour and of localised osteitis fibrosa have varied widely. However, thanks to improved radiographic technique in the last few years, certain, fairly well-defined, clinical types have been recognised. At the outer limits are bone cysts, which may be regarded as a form of localised osteitis fibrosa, on the one hand, and the true benign giant cell tumours on the other. Also at certain sites transitional conditions, e.g. localised osteitis fibrosa, which are difficult to differentiate from a benign giant cell tumour containing cysts, do occur and make a definite diagnosis difficult.

Benign giant cell tumours usually occur at the end of one of the long bones, particularly at the distal end of the femur and the proximal end of the tibia. This case is only reported because of its site—which has been reported 21 times in the literature.

E.G.O. A woman, born 28.11.17. Admitted to hospital 12.11.45. Complained of pain in the R. knee anteriorly and of occasional instability in the knee on walking. The trouble began in June 1945 when the patient was kicked on the R. knee by a cow. The knee swelled rapidly,

but the swelling disappeared in a few days with hot compresses. The knee had given no previous trouble.

O.E. Palpable, but not measurable, atrophy of the thigh muscles. Circumference at 15 cm. above the upper patella border was 44 cm. on both sides. Reduced elasticity of the muscles. No swelling or heat. No deformity. Slight tenderness over the medial border of the patella. Full active movement. Small retropatellar crepitations. Pain felt under the patella when it was compressed against the condyle.

Left knee. Small retropatellar crepitations, otherwise nothing abnormal.

Blood: Serum calcium 11.7 mg. per cent.
Phosphatase 3.5 units.
Hb 82 %.
R.B.C. 4,740,000.
Sed. Rate 8 mm.
Blood Wasserman Reaction: positive.

31.11.45. "Radiography shows thinning at the medial border of the patella; the cause cannot be determined. It is most suggestive of localised osteitis fibrosa". (signed) R. M. (Fig. 1).

26.11.45. "Radiography of pelvis, hips, femora, tibiae and fibulae, and of the upper limbs shows no definite signs of osteitis fibrosa". (signed) R. M.

These radiographs were taken to exclude the possibility that the changes in the patella were part of a generalised osteitis fibrosa. The calcium concentration and the phosphate values were also against this diagnosis. The other possible diagnosis was a syphilitic infection, in view of the positive WR. The differential diagnosis could not be carried further without a biopsy.

16.11.45. *Curettage of the bone tumour in the patella.* As soon as the tendon fibres on the patella were freed, a bluish colour could be seen; the anterior layer of bone came away when an attempt was made to separate off the tendon. Almost immediately one came onto a cavity filled with a grey-brown mass. The opening in the patella was widened, and the tissues filling the hole, which was about the size of a hazel-nut, were scraped out. The walls of the hole were sclerotic and firm; here and there there were ridge-like projections. The only opening was near the ligamentum patella. The soft parts were sutured.

Pathological Investigation. The ground substance consisted of numer-

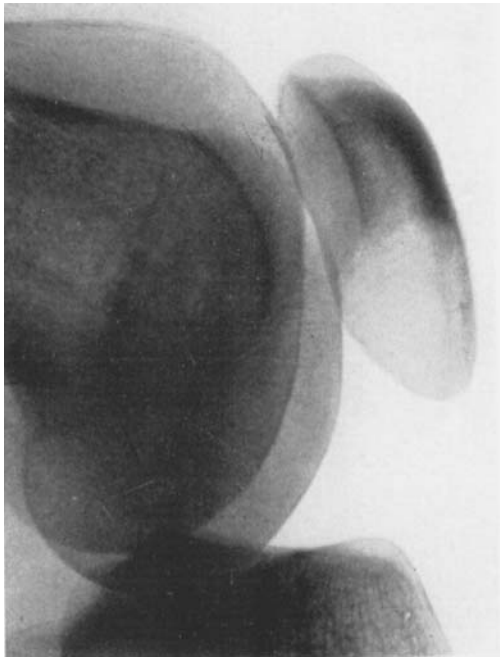

Fig. 1.
Lateral view of patella before operation.

ous regular multinucleate giant cells of epulis type. There was also plentiful blood pigment, and individual fragments of necrotic bone. The picture appeared benign.

Diagnosis: Localised osteitis fibrosa, or benign giant cell tumour (A. L. Fig. 2).

28.11.45. The patient returned home, walking quite well; able to bend the knee to 90°.

5 of the 21 cases in the literature were treated by curettage; in 2 the condition recurred. The present case has been followed for 3½ years.

17.6.46: Reports continued improvement. Walks well on flat ground; can walk for over a mile. Has difficulty in kneeling on the knee. Pain

across the R. knee when she sits for a long time with the knee flexed. No stiffness in the morning. Sometimes pain on walking upstairs. No spontaneous pain; no swelling.

O.E. R. knee. Palpable, but not measurable, atrophy of the thigh muscles. Circumference 15 cm. above the upper border of the patella

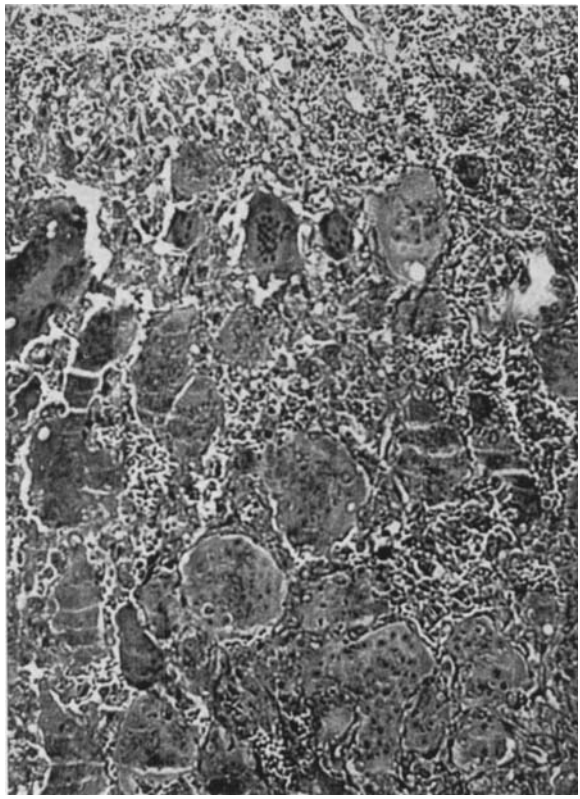

Fig. 2.
Microscopic picture of resected material.

47 cm. on both sides. No swelling. Full active extension and flexion. Bilateral small, retropatellar crepitations on extension. Normal gait.

Radiography: "The operation cavity has been considerably filled in with bone tissue" (signed) O. O. (Fig. 3).

5.6.47: Reports she is better than at the last examination. Walks

unchanged. No swelling. Full active flexion and extension. Small bilateral retropatellar crepitations. Gait N. A. D.

Radiography: "Comparison with the radiograph of 17.6.46 shows further bone formation in the cavity. Only a small cavity, the size

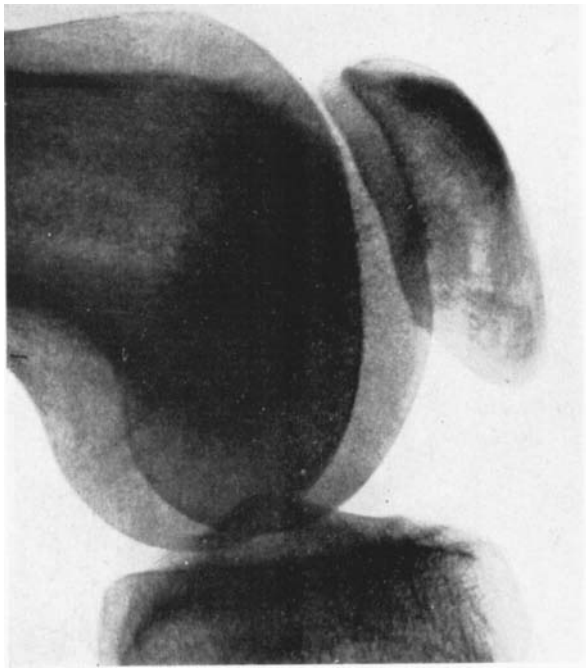

Fig. 3.
Lateral view of patella immediately after operation.

well in rough country for over a mile. No difficulty in going up stairs. May still have some pain from the knee after sitting for a long time with the knee flexed (e.g. at a cinema). No stiffness in the mornings. No spontaneous pain. No swelling.

O. E. R. knee. No atrophy of the thigh muscles, and circumference of a pea, remains in the medial part of the patella. Otherwise the bone outline of the patella is well-defined". (signed) O. O.

12.7.49: Reports that she feels almost cured. No difficulty in walking on rough ground or on stairs. Occasionally she has slight discomfort after sitting for a long time with the knee flexed. No stiffness in the morning. No swelling; no spontaneous pain.

O. E. No atrophy of the thigh muscles. Measurements unchanged. Full active extension and flexion. No swelling. Small bilateral retropatellar crepitations. Gait: N. A. D.

Radiography: "Comparison with previous picture shows further bone formation in the cavity, of which only an insignificant remainder persists. There also appears to be slight periosteal bone formation". (signed) O. O. (Fig. 4).

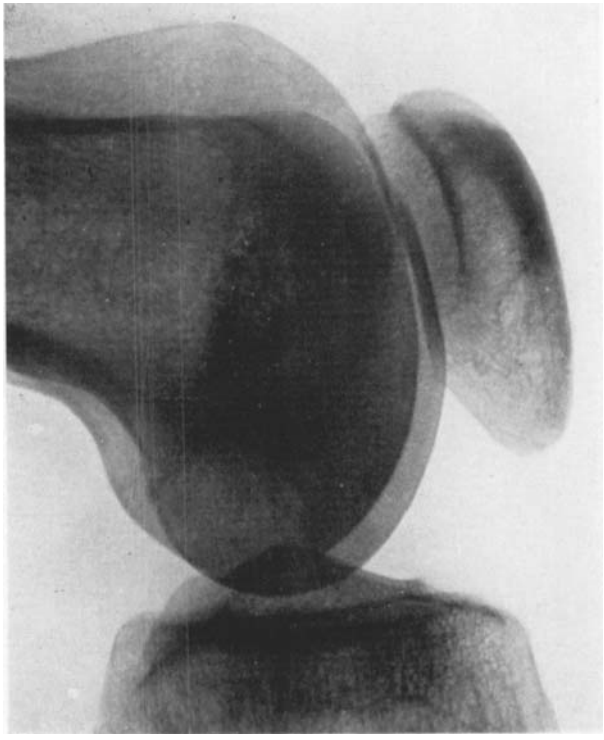

Fig. 4.
Lateral view of patella 3½ years after operation.

In the embryo the patella passes through pronounced pre-cartilaginous and cartilaginous phases in its development. *Geschichter* and *Copeland* found that giant cell tumours occur exclusively in bone which originates embryologically from cartilage. They believe that this type of tumour is caused by trauma to the cartilaginous bone. The trauma damages

the periosteal blood vessels, and the result is a reduced blood supply to part of the bone. They have also suggested that the tumour represents a local overstimulation of osteoclastic activity, which has caused a revascularisation of the part. They have stressed earlier that benign giant cell tumours occur in places where the spongiosa is relatively soft, as in the proximal part of the tibia. When there is considerable mechanical resistance to its growth, as in the solid cortical bone of the shaft of the femur, it remains as a localised osteitis fibrosa containing many giant cells. This theory agrees well with the finding of these tumours in the patella, which has hardly any cortex and is virtually all spongiosa.

From the point of view of insurance, it is of great interest to know how far trauma can be regarded as influencing the occurrence of the tumour, especially as, according to the literature, it affects mainly those between 28 and 30 years. Trauma seems to have occurred in 19 out of the 21 cases in the literature, but one must not, of course, attribute too much importance to this, since the patella is often injured without any persisting symptoms arising. Further, one would expect the tumour to occur more often, if trauma was the cause. The trauma appears to occur usually from weeks to years before any symptoms appear. In fact, it is doubtful whether trauma can really be regarded as predisposing to this condition.

The Americans insist upon routine resection of the patella in cases of benign giant cell tumour for two reasons. Firstly, relapses do occur; secondly, it is, according to *Geschichter* and *Copeland,* difficult to distinguish it radiographically, or even microscopically, from malignant conditions.

Nevertheless, curettage was used in this case in order to save the patella, if possible. So far, the follow-up examination has given rise to no fear of relapse or malignancy. A prerequisite for the use of this less mutilating operation is, of course, that the curettage is sufficiently extensive and is carried into healthy bone, and that the patient is followed up.

SUMMARY

1. The commonest age for benign giant cell tumour is 28-30 years.

2. In 20 out of the 22 published cases trauma was regarded as a predisposing factor.

3. The occurrence of benign giant cell tumour in the patella agrees with the theory that this tumour develops only in spongiosa bone, which embryologically arises from cartilage.

4. This case, in which the less disabling curetting operation was used, does not give any support for the American view that the resection of the patella should be routine in these cases.

5. If the curetting method is used, the case must be followed up for at least 3 years, in view of the risk of recurrence and malignant degeneration.

RESUME

1. L'âge le plus commun pour l'apparition de tumeur bénigne à cellule géante est 28—30 ans.

2. Dans 20 des 22 cas publiés, un traumatisme était considéré comme un facteur prédisposant.

3. L'apparition de tumeur bénigne à cellule géante dans la rotule concorde avec la théorie que la tumeur ne se développe que dans les os spongieux qui embryologiquement des cartilages.

4. Ce cas, dans lequel on a procédé à un curettage, c'est-à-dire à une opération moins radicale, n'appuie absolument pas la théorie américaine suivant laquelle il convient de procéder à la résection de la rotule.

5. Si l'on recourt au curettage, le cas doit être minutieusement suivi, au moins pendant 3 ans, en raison des risques de rechute ou de dégénération maligne.

ZUSAMMENFASSUNG

1. Das gewöhnlichste Alter für den gutartigen Riesenzellentumor ist das 28.—30. Lebensjahr.

2. In 20 der bisher beschriebenen 22 Fälle wurde das Tauma als prädisponierender Faktor angesehen.

3. Das Vorkommen von gutartigen Riesenzelltumoren in der patella stimmt gut überein mit der Theorie, dass diese tumoren ausschliesslich in spongiösem Knochen vorkommen, der embryologisch vom Knorpel abstammt.

4. Ein Anhaltspunkt für die von Amerikaneren stereotypisch anbefohlene Patellarresektion liegt nicht vor in diesem Fall und man bevorzugt hier die weniger verstümmelnde Curettagetechnik.

5. Anwendet man die Curettagetechnik, so muss man unter Hinblick auf die Gefahr des Rezidivs und der malignen Entartung wenigstens während 3 Jahre dem Pat. folgen.