A CASE OF ADAMANTINOMA OF THE TIBIA

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A twenty-year old Chinese woman came to Orthopaedic 'C' Unit with a 1 year history of pain in the left leg which she noticed particularly when she walked faster than usual. She remembered knocking this leg against a pole while playing net-ball, and a painful swelling had occurred at the site of injury one month later.

On examination, an oval swelling $1\frac{1}{2}$" x $3\frac{1}{4}$" could be seen on the anterior aspect of the middle third of the left leg. The swelling was firm in consistency at the periphery and cystic at the centre.

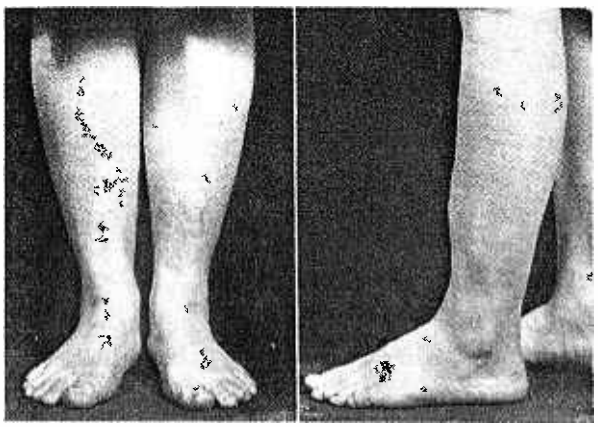

Fig. 1 (a) Fig. 1 (b)

X-ray showed a cystic tumour of the tibia bounded by a zone of sclerotic bone. Part of the tumour involved the whole thickness of the cortex.

A provisional diagnosis of adamantinoma of the tibia was made on clinical and radiological grounds.

The case was discussed at an Orthopaedic Departmental Clinical Meeting, and it was decided that wide re-section of the tumour and biopsy should be carried out. This was done (by Mr. D.R. Gunn) on 23.1.'59. Six inches of the shaft of the tibia including the tumour was excised, and biopsies were taken from each end of the cut surfaces adjacent to the excision. The defect in the tibia was bridged by a vitallium plate, as well as bone chips from the patient's left iliac crest and from ribs from the bone bank.

The biopsy report was as follows:

> A greyish tumour arising deep to the periosteum. There is destruction of bone with blood clots.
>
> Histologically the tissue showed an infiltrating epithelial tumour with microscopic features of a basal cell carcinoma (the so-called adamantinoma) of the tibia. Sections from the upper and lower ends of the tibia show no evidence of tumour involvement.

Post-operatively the patient required a blood transfusion because of anaemia. A skeletal survey on 18.2.'59 showed no secondary deposits. The patient was re-admitted on 21.5.'59 for further bone-grafting. Last seen in November 1959, the patient was well, and could walk normally for "as far as she liked".

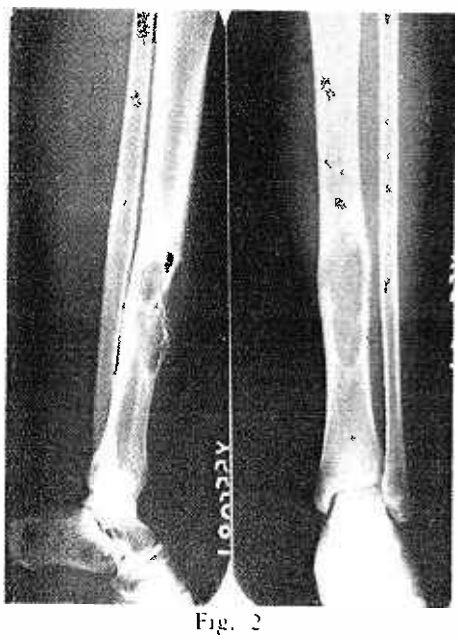

Fig. 2

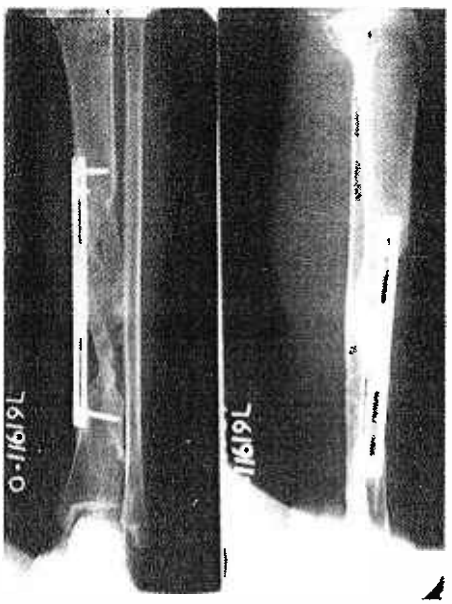

Fig. 3.

REVIEW

In a comprehensive review in 1954 in the Journal of Bone and Joint Surgery, P.L. Baker and others analysed a total of 27 cases including 3 of their own, of Adamantinoma (so-called) of Long Bones.

"Adamantinoma" indicates a tumour with enamel. No tumour with enamel has, in fact, been found in long bone. Of the 27 cases described, 25 were in the tibia and one each in the femur and ulna. Sixteen patients were male, eleven female. Their ages ranged between 12 to 57 years.

Pain was the commonest symptom, reported by 14 patients. Swelling was reported in 7 cases. Duration of symptoms ranged between $1\frac{1}{2}$ months to 17 years.

The gross pathological features vary, and have been reported as grey, white or reddish brown, smooth or lobulated. A capsule may be present. The consistency may be firm, elastic and gritty, or soft and brain-like. They may involve the cortex and periosteum of the bone, or the marrow, or soft tissue. Calcification or bone particles may be present, but no tooth or enamel. Microscopically they appear as:

(1) Epithelial islands in which peripheral cells are columnar and arranged in palisaded fashion with central stellate cells producing reticular formation.

(2) Islands of cells resembling basal cells scattered throughout fibrous stroma reminiscent of basal cell carcinoma.

(3) Islands of squamous epithelium scattered throughout a fibrous stroma. Great variation in size and shape of the islands are found. Pearl formation is a feature.

Recurrence after local excision is common, and 4 proven cases of metastases to inguinal nodes have been reported. Another 4 cases showed radiological evidence of metastases to other bones, lung and brain.

X-ray changes are not diagnostic but generally show cystic or multi-cystic destructive lesions, some with cortical expansion and periosteal reaction.

Much speculation has naturally been aroused by the fact that the lesion arises almost exclusively in limb bones with subcutaneous surfaces or borders. A plausible explanation put forward is that these rare tumours arise from cell rests derived from the skin, the usual overlying intact cortex makes it unlikely that such rests could result from trauma, and in all probability they should be explained as a developmental anomaly.

There is, however, disagreement regarding how epithelial tissue gets into bone and whether the lesion is identical with recognised adamantinoma of the jaw, or basal cell carcinoma with known variants, or squamous cell carcinoma with known variants. Of the 27 cases reported, 13 had a preceding history of contusion or abrasion, 2 had fractures, 1 a puncture wound. Moreover, in 5 cases, the tumour cells were found in areas of fibrous dysplasia.

The frequency of recurrence—17 of the reported 27 cases—demands a very thorough removal of the tumour with all the affected bone by block excision; a very large or recurrent lesion in a limb will call for amputation. The results of radio-therapy have been unsatisfactory.

ACKNOWLEDGEMENT

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