MASSIVE OSTEOLYSIS OF THE HUMERUS TREATED BY RESECTION AND PROSTHETIC REPLACEMENT

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Massive osteolysis or vanishing bone disease was described by Jackson in 1838. Since then over fifty cases have been reported. The etiology, however, is still in dispute and the treatment presents many problems. In the instance recorded here prosthetic replacement of diseased bone was carried out with preservation of a useful limb.

The patient was a thirty-seven year old man born in British Guinea. He first noticed pain in the region of his right shoulder in January 1962. There was no history of injury, though he was at that time doing heavy work involving much lifting. The pain gradually increased so that in January 1963 he reported to a hospital. Radiography at that time showed no abnormality and he was treated by radiant heat and exercises. Seen in October 1963 he was still complaining of pain. The radiographs showed an irregular circumferential osteolytic lesion (Fig. 1).

A few weeks later he fell and sustained a pathological fracture of the right humerus (Fig. 2). A biopsy specimen of bone from above and below the site of the pathological fracture, including adjacent soft tissue and curettings of the medullary cavity, did not show any evidence of tumour; there was some vascular fibrous tissue and there were numerous large vascular spaces. Osteoclastic resorption was seen on the endosteal surface of the bone. There was some developing bony callus consistent with a reparative process at the site of fracture. A diagnosis of massive osteolysis or vanishing bone disease was made.

Skeletal survey did not reveal any other lesion. The blood chemistry was normal. Arteriography was normal (Fig. 3).

Operation (February 1964)—The upper 25 centimetres (9½ inches) of the right humerus were resected by extraperiosteal dissection and replaced by a hollow Titanium 160* prosthesis that had been sealed by welding (Fig. 4). The prosthesis was bonded into the lower humeral stump by an acrylic dental polymer, Simplex P.† Bleeding was controlled by a tape placed around the subclavian artery. The operation was done by Mr H. Jackson Burrows, Mr M. A. Birnstingl and Dr J. T. Scales.

* Titanium 160 is manufactured by Imperial Metal Industries (Kynoch) Ltd., New Metals Group, complying with B.S.3531, 1962, amendment 1, 1964.
† Simplex P is manufactured by Northill Plastics Ltd., London.
Pathology—Examination of the resected specimen showed the same large vascular spaces seen in the biopsy. These spaces extended down to the line of resection, and presumably extended into the lower humeral stump. Large cavities were present in the cortical bone, causing an irregular osteoporosis.

Progress—The patient made an excellent recovery from the operation and when he was last seen in November 1967, three years and nine months after operation, the prosthesis was secure and no extension of the disease was evident on radiography. Elbow movements were full but there was little active movement of the shoulder, combined abduction being only 30 degrees. He was working as a clerical assistant, using his right hand for writing and filing.

DISCUSSION

The diagnosis of “massive osteolysis” is made by negative deduction. The radiological appearance, though quite often striking, shows only a lytic appearance of bone. The microscopic appearance is mainly characterised by lack of any tumour or inflammatory material to account for the osteolysis. Numerous large vascular spaces are constantly found but the excessive osteoclastic activity is not always evident.
The etiology remains in dispute. Gorham and Stout (1955) and Johnson and McClure (1958) like many other authorities postulated a vascular lesion. Coste and Gaucher (1943) considered a neurotropic factor of prime importance. Lagier and Rutishauser (1965) suggested that the disease was associated with a disorder of proline metabolism. Crasselt (1961) distinguished several types: post-traumatic, neurogenic and inflammatory, and mentioned the relationship of the disease to haemangiomatosis. This case has helped little in elucidating the etiology.

The prognosis of the untreated disease is poor. The lesion spreads across joints to adjoining bones until the patient succumbs to some intercurrent disease.

Many forms of treatment have been described. Johnson and McClure (1958), on the supposition that the disease was primarily of vascular origin, made a plea for the use of radiotherapy. Others have favoured amputation or resection. Butler, McCance and Barrett (1958) described a patient in whom a lesion of the femur was resected and replaced by a homogenous cortical graft and autogenous cancellous bone. Aston (1958) reported a patient of Dr J. T. Scales and Mr H. Jackson Burrows who had had an extraperiosteal resection of a femur with replacement of the upper two-thirds of the bone with a cobalt-chrome-molybdenum alloy prosthesis; this converted what would have been an amputation through the hip into an above-knee amputation. Branco and da Silva Horta (1958) described a large number of treatments tried on a boy who had involvement of the right lower limb; all failed and amputation was eventually necessary; Vitali (1962) described the external prosthesis eventually used.

The present case is the first reported in which the lesion was resected and replaced by a prosthesis, preserving the limb. The use of a prosthesis seems more logical than replacement by bone, itself presumably susceptible to erosion.

**SUMMARY**

1. A case of massive osteolysis of the right humerus is described. Diseased bone was resected leaving only the lower part of the humerus. The resected bone was replaced by a titanium prosthesis.
2. The diagnosis, etiology, prognosis and treatment of the condition are briefly discussed.

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**REFERENCES**


JACKSON, J. B. S. (1838): Absorption of Bone (Boneless Arm). *Boston Medical and Surgical Journal*, 18, 368.

