CONGENITAL ARTERIOVENOUS FISTULA WITHIN THE TIBIA
Report of a Case

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An arteriovenous fistula entirely within a long bone is rare, and a successful result from treatment is even less common. This paper records an example in the tibia of a girl treated three years ago by removal and replacement of a large part of the affected bone. A review of the literature has not revealed any report of successful treatment of arteriovenous fistula by this technique.

CASE REPORT

An eight-year-old girl was found at a school medical examination in 1962 to have the right leg shorter than the left. The discrepancy was ascribed to undiagnosed poliomyelitis and raising of the shoe was prescribed. When she was nine and three-quarters it was decided to perform femoral and tibial stapling on the left (longer) leg. There were local problems from the staples, and after three months two were removed and one was reinserted. At that time
radiographs showed decalcification of the diaphysis of the left tibia, thought possibly to be due to chronic osteomyelitis. All the staples were removed and the bone was drilled, but no pus was found. When the patient was discharged from hospital five months after the last operation the left leg was four centimetres longer than the right.

Two years later, when she was aged twelve, she was readmitted with an ulcer of the shin. This was explored without a tourniquet and the profuse haemorrhage which ensued was controlled only with difficulty, by bone wax. Two units of blood were transfused. It was at this time that the possibility of an arteriovenous fistula was considered (Fig. 1), and shortly afterwards the child came under my care. Arteriography confirmed the existence of a large arteriovenous fistula within the tibia (Figs. 2 and 3). On auscultation a loud “machinery” murmur was heard. In consultation with Mr Martin Birnstingl it was agreed that a major attempt to deal with this vascular anomaly was justified. His suggestion was to explore the popliteal fossa, and to follow this if necessary by stripping the periosteum from the tibia.

Operation—A combined operation was undertaken in March 1966 when the child was twelve and a quarter (Fig. 4). Exploration of the popliteal fossa proved uneventful and the tibia was therefore exposed under tourniquet control. After the periosteum had been stripped from most of the shaft and the tourniquet had been released, it was apparent that the fistula lay within the bone itself; with a sterilised stethoscope the loud machinery murmur could still be heard. After re-inflation of the tourniquet most of the shaft of the tibia between the upper epiphysis and the junction of the middle and lowest thirds was removed. The specimen contained large vascular spaces. As many as possible of the large periosteal vessels were under-sewn and the tourniquet deflated. Brisk bleeding was eventually controlled by further sutures. The tibia was then replaced, the upper end being fixed with two Vitalium staples and the lower end wedged into a slot. Reasonable stability was provided by the intact fibula.

Progress—The skin healed slowly and for several months there were discharging sinuses. Three months after the bone replacement radiographs showed subperiosteal new bone and a very “moth eaten” appearance of the original shaft (Fig. 5). Sequestra formed (Figs. 6 and 7) and were twice removed, five months and eight months after the replacement operation. By nine months the wounds of the leg had healed. Further operation was undertaken a month later to reinforce the slender tibial shaft (Fig. 8) with iliac slivers. There was no evidence of pseudarthrosis.

A year after the operation the femora were equal in length but the left tibia was still two centimetres longer than the right. In August 1968, nearly two and a half years after the main operation, further sequestrectomy was required. This was followed by rapid skin healing. The appearance of the bone at that time is shown in Figure 9.

When the girl was last seen, in April 1969, the leg lengths were almost equal. She was walking well, and the joints had regained good mobility (Figs. 10 and 11). There was a definite increase in warmth over the middle part of the tibia, but no audible murmur.
DISCUSSION

There are several descriptions in the literature of bones involved by arteriovenous fistulae, although in many of these most of the fistula was in the soft tissues. Malan and Puglioni (1965) in a very complete review of congenital angiodysplasia of the extremities described the intraosseous appearance as “confluent regions of osteolysis giving the appearance of worm-eaten wood”. Other examples were described by Lewis (1930), Coursley, Ivins and Barker (1956), Howard (1959), Martorell and Palou (1959), Marin (1960), Castro-Farinas and River Lopez (1961), Delaney (1961), Wojnerowicz (1961), Malan and Puglioni (1964) and Sautot and Duquesnel (1966).

Clay and Blalock (1950), Borszewski and Stadnicki (1957), Wojnerowicz (1961) and Cook and Zbar (1962) all recorded examples in the jaws; these may give trouble during dental procedures. It is of course possible that some of these conditions were aneurysmal bone cysts rather than arteriovenous fistulae.

Nisbet (1954) described a fistula which was found after amputation to lie within the tibia, and Reid’s (1925) series contained an illustration of a fistula in the tibia of a leg which came to amputation after a local operation. Birnstingl (1962) recorded an example in the medulla of a femur which had produced overgrowth of the limb, and was being managed conservatively. In some of these cases local operation or amputation had been indicated, but there are no records of any successful attempts at treatment by removal and replacement of a section of bone containing the entire fistula, although a very similar technique was suggested by Nisbet in 1954.
Figure 8—Ten months after operation. There was a suspicion of pseudarthrosis but at exploration this was found not to be so.

Figure 9—Two and a half years after operation. There is a small sequestrum but reformation of the tibial diaphysis is excellent.

Figure 10

Three and a half years after the major operation. The legs are of equal length and there is good mobility of the joints. Note the scarring.
SUMMARY

1. A case of arteriovenous fistula within the tibia of a girl is described.
2. Removal and replacement of a large part of the tibial diaphysis was followed by "re-formation" of the bone.
3. The literature of the condition is reviewed.

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This patient was shown at the clinical meeting of the Orthopaedic Section of the Royal Society of Medicine in October 1969, and I am grateful to the Editorial Department of the Royal Society of Medicine for permitting me to publish the present report of the case.

REFERENCES


