PSEUDARTHROSIS OF THE TIBIA IN CHILDHOOD*

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The purpose of this paper is to report the late results of treatment by the use of the "by-pass" graft, to give a brief account of all the patients treated, and to discuss the possible causes of inception and maintenance of the condition.

It is now fourteen years since the by-pass graft was first introduced. It was used for two patients in the same year and both cases were described in my original paper in 1940. As far as can be ascertained these were the first recorded successes in the treatment of pseudarthrosis of the tibia in childhood. In that paper also is reported the successful use of a double onlay graft (later described by Boyd, 1941) but this has been discarded in favour of the by-pass operation because the latter was more certain in its effect. In all cases the graft has been taken from the sound leg and has been taken from as far up the tibia as practicable in order to obtain a graft with the highest possible proportion of cancellous to cortical bone. Although obviously the cortex is essential for success, previous experience had shown that bone grafts for ordinary non-union became incorporated more quickly and with greater certainty if taken from the part of the tibia where the cortex is thin and cancellous bone preponderates. The method of insertion described in the original paper has been employed in each operation (Fig. 1). It is similar in principle to that of the bolt-graft used in ordinary inlay grafting.

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impacting immediate stability to the limb. That is to say stability sufficient for easy handling though not of course for bearing weight.

RESULTS

The first patient, N. K., operated upon in 1936 at the age of eight years, is now twenty-two years of age: he is married and works as a clerk in a timber merchant's office. The condition of the leg before the by-pass grafting operation, eighteen months after operation, and fourteen years after operation is shown in Figures 2 to 6. Before the insertion of the by-pass many attempts had been made to secure union of his tibia. The limb is now two and three-quarter inches short. This is about the same as in 1939. The bowing then present has largely disappeared although nothing has been done to correct it. Correction has occurred naturally as it has in the other cases.

The second patient, W. P., operated upon at the age of three and a half years, is now nineteen years of age. Figure 7 shows the condition before operation. Radiographs of the leg six months after operation, and fourteen years after operation are shown in Figures 8 and 9. The limb is three-quarters of an inch short, just as it was in 1939.

During the last fourteen years seven further patients with this type of pseudarthrosis have been treated, making nine in all. This is not a large series and although allowance must be made for the interruption of war it is clear that the condition is not a very common one. To my own series are now added two patients treated successfully by other surgeons. Mr Pulvertaft's case (Fig. 10) illustrates the use of the by-pass when the bowing is posterior. Although the employment of screw fixation is not part of my own technique, no harm has resulted. I do not favour screws because I have seen instances of fracture across the bone in the plane of a screw hole some time after resolution of the original lesion. Mr Birkett's patient (Figs. 11 to 20) is extensively illustrated because the radiographic record he has preserved is so delightfully instructive.

Of my own nine patients, sustained success has been achieved in seven. The two failures occurred last year during the first three post-operative months. Although it is generally felt in orthopaedic circles that results ought not to be assessed in less than three to five years, one may presumably claim a failure more quickly than a success. And in any case, however long one were to wait, these two attempts would have been initially unsuccessful. In neither patient was the principle or technique of the operation at fault. Rather was the surgeon's judgment to blame in the way he dealt with the somewhat unusual features. The first patient, P. J. (Figs. 21 to 23), was operated upon at the age of eleven months. Stored rib was used for the graft and was found to be very suitable. Although the post-operative plaster included the flexed knee, the child, who was distinctly chubby, worked the plaster down the leg and cracked the graft across the "popliteal angle" of the plaster (Fig. 22). This uncooperative tendency was overcome by leaving the sole of the foot free of plaster, but neither the graft nor the tibia united (Fig. 23). Although there should be no difficulty in holding such a leg in plaster (we do so for hundreds of children with congenital club foot) it seems that in a similar case it would be well to defer operation until the age of about eighteen months when the child might reasonably be expected to be more manageable. Moreover the recipient bone would then be larger and the operation would be technically simpler. The second failure was in a child of three years who had received no previous treatment (Figs. 24 and 25). A large part of the tibia above the pseudarthrosis was defective, and although the graft was as long as could be obtained from the sound tibia it was not long enough adequately to clear the area of bone absorption. Consequently its lower end shared the fate of any other graft placed in that area and disappeared. It is unlikely that a double onlay graft could have been clamped to such an attenuated bone as was present above the lesion, but the surgeon might reasonably have risked placing the by-pass lower down. A second by-pass graft has since been undertaken in this child and union has now occurred.
Case 1. Figure 2—Appearance of the limb at the age of eight years.
Figure 3—Radiograph at the age of eight years.

Case 1. Figure 4—Condition eighteen months after by-pass grafting. Figure 5—Fourteen years after grafting. Figure 6—Present condition at the age of twenty-two years.
Case 2. Figure 7—Condition of leg before operation, at the age of three and a half years.
Figure 8—Six months after operation. Figure 9—Fourteen years after operation.

Case 3—A successfully treated case in which the upper end of the graft was fixed by screws.
(Reproduced by courtesy of Mr R. G. Pulvertaft.)
Case 4. Figure 11—Child aged one year. Marked anterior bowing but no fracture. Figure 12—Condition at the age of five years. (These and the nine succeeding illustrations are reproduced by courtesy of Mr N. Birkett.

Case 4. Figure 13—Aged six years. Inlay grafting has been unsuccessful. Figure 14—Six months later the graft has been almost completely absorbed. Figure 15—Aged nine years. Massive double onlay graft.
FIG. 16
Case 4. Figure 16—Four months later, union appears satisfactory. Figure 17—Three years after double onlay graft. Union appears consolidated.

FIG. 18
Case 4. Figure 18—Three and three-quarter years after double onlay graft. Absorption of bone around the lowermost screw. Figure 19—Three months later, recurrent pseudarthrosis. Figure 20—Age fourteen years. Seven months after by-pass graft. The graft is incorporated at its upper and lower ends. Hypertrophy of bone is taking place along the line of the graft.
Case 5. Figure 21—Condition at age of eleven months, immediately before operation. Figure 22—Two months after operation, showing break in graft caused by the child working the plaster off the leg. Figure 23—Ten months after operation. The lower end of the graft has been absorbed.

Case 6. Figure 24—Condition at the age of three years. Note the extensive area of tibia affected. Figure 25—Four months after operation. Failure of lower end of graft.
FIG. 27

Case 7. Figure 26—Radiograph showing by-pass operation after failure of inlay graft. The strut consisted partly of the original inlay graft and partly of tibial bone to which it was fused. Figure 27 shows sound bone union six months later.

One of the successful cases is of special interest. A boy aged twelve years had previously been treated elsewhere, first by an inlay graft and later by a by-pass. His parents were unwilling for another piece of bone to be taken from his sound tibia. The affected leg was opened with the intention of packing iliac chips into the angle at the lower end of the graft, which had been placed too high and too medially. But on inspection at operation this was thought to have a poor chance of success. The existing graft therefore was first removed together with a three-inch length of tibia to which its upper end was fused; and the composite bone—part graft, part tibia—was reversed so that the upper end became the lower and was implanted in the usual way. Sound union ensued (Figs. 26 and 27).

PATHOGENESIS

In dealing with these patients the question naturally arose as to why the pseudarthrosis develops, and why it persists in spite of all efforts which would ordinarily produce union. Complete and prolonged immobilisation has failed, step grafts and inlay grafts have failed, and even sliding grafts of half tibial thickness have failed. At one time (about 1932) the author had the impression that in the lesion there were cells possessing a destructive quality equivalent to that present in conditions of local malignancy such as, for example, osteoclastoma. But the evidence is against this for three reasons: 1) In one case the whole area of the pseudarthrosis was excised together with two inches of tibia above and below the lesion: length was maintained by traction for some weeks and the gap was subsequently bridged by an implanted section of fibula: this shared the same fate as any other graft. 2) No destructive cells have been seen in the many sections that have been taken. 3) When fixation is adequate the destructive process is arrested and the bone re-forms. The cause of persistence of the pseudarthrosis must therefore be mechanical. The author's present view is that angulation
and fracture are coincident, and that the bending stress continually maintained by the muscles stimulates bone resorption to a greater degree than the body can tolerate. Under the bending stress the woven bone with which the body attempts to heal the fracture breaks down as quickly as it is formed. This struggle between repair and destruction gives rise to the typical appearance of these fractures.

Many hypotheses have been put forward to explain the initial cause of this peculiar pseudarthrosis. That it is not a "birth" fracture is abundantly clear. If a fracture of a normal tibia at birth occurs at all it does so very seldom indeed. Not only is such a fracture not included among the large number of birth fractures the writer has treated; it is also absent from the list of birth defects recorded in two maternity hospitals whose combined total of births in the last ten years was 48,000. Moreover the author has knowledge of only one case in which pseudarthrosis was encountered within a short time after birth. This was in a patient seen by Mr Gorony Thomas of Liverpool (Fig. 28). The radiograph shows that the tibia was not normal before it broke: part of the shaft was thin and dense. Reference will be made later to this typical defect.

![Fig. 28](image1)
Radiograph showing tibia fractured soon after birth. Pseudarthrosis developed but it is clear that the tibia was defective before it broke. (Reproduced by courtesy of Mr G. E. Thomas.)

![Fig. 29](image2)
Wedge defect of tibia, suggesting the possibility of development from multiple ossific centres. Pseudarthrosis did not develop.

The possibility of a separate centre of ossification was suggested by the case illustrated in Figure 29, but this child did not develop pseudarthrosis and no evidence of multiple ossific centres has been present in the established cases.

An attractive theory is postulated that in utero a foot in extreme calcaneus would press on the tibia and render it defective in such a way that it would subsequently break very readily. This theory would also explain the laxity of the calf muscles and the consequent tendency to calcaneus which is such a constant feature of the clinical picture of pseudarthrosis. But children born with calcaneus deformity do not develop pseudarthrosis: the tibia is often normal and even if it is bowed posteriorly it does not break and then fail to unite. Even if a tibia with congenital posterior bowing is corrected by osteotomy it unites quite readily (Fig. 30).

The explanation most satisfying to the author is that a fatigue fracture occurs in a congenitally defective tibia. The fracture occurs usually within about the first year of walking. There is no doubt that in all cases of pseudarthrosis a defect exists in the shaft of the tibia in addition to the fracture itself. The defective area varies in length but may involve nearly
Figure 30—Uneventful union of the tibia and fibula after corrective osteotomy for congenital posterior bowing.

Figure 31—Cystic changes in shaft of tibia. Figure 32—Same case three months after by-pass operation.

Section of limb of chicken showing tibial kyphosis. Note the bilateral deformity shown in radiograph (inset). (P. K. Duraiswami.)
the whole shaft. Usually the bone is attenuated and dense but occasionally it is cystic (Figs. 31 and 32). This hypothesis does not rest entirely on clinical evidence. Duraiswami (1950), working in the Department of Orthopaedic Surgery of the University of Liverpool, has, by injecting insulin into the yolks of hens' eggs, caused various congenital defects in the chickens. By appropriate dosage and timing of the injection he has been able to determine the defect which will be present at birth. Figure 33 shows one of the chickens in which defects in the tibia closely resembling the pseudarthrosis in children were produced. It is not unreasonable to suppose that some influence may similarly affect the human embryo and give rise to a similar dysplasia of the mesenchyme which later results in a defect of the tibia.

SUMMARY

1. The results of the by-pass grafting operation in eleven cases of pseudarthrosis of the tibia in childhood are presented.
2. The operation was successful in securing sound bone union in nine cases, and unsuccessful in two.
3. The causes of failure in the two unsuccessful cases are analysed.
4. The possible causes of occurrence and persistence of tibial pseudarthrosis in childhood are discussed.
5. It is suggested that the pseudarthrosis results from a fatigue fracture of a congenitally abnormal tibia, but that its persistence depends entirely on mechanical factors.
6. There is evidence to suggest that the underlying congenital abnormality may arise from dysplasia of the mesenchyme.

REFERENCES