Summary  Tibial aplasias are difficult entities to manage and sometimes require significant limb lengthening. Preserving the joint is the preferred option in these cases. However, when function is not ensured, lengthening beginning in the joint can be attempted followed by arthrodesis. This report presents the clinical and radiological results of a patient treated with intra-articular lengthening (or arthrodiastasis) associated with talocrural arthrodesis. Although this original technique provided satisfactory results, this surgical program remains complicated.

Introduction

Bone lengthening can be achieved by various techniques. By endomedullary nailing or external fixation with periosteal callus distraction. In certain situations which result in significant limb length discrepancies such as tibial agenesis, the main therapeutic option is often external fixation which nevertheless remains technically difficult if significant lengthening is required. (Courvoisier et al. [1]). In these clinical situations, successful bone consolidation is a real therapeutic challenge comparable to that during traumatic bone loss. In the case of pseudarthrosis, bone substance can be reconstituted by fibular transfer or a vascularized iliac crest graft (El-Gammal et al. [2], Legre et al. [3]), by the induced membrane technique (Masquelet et al. [4]), by the elevator technique (Paley et al. [5], Trigui et al. [6]) or by associating both (Uzel et al. [7]).

To preserve limb function, preservation of under- and overlying joints is the goal in all cases. Nevertheless, in certain complex malformations the joint is pathological or non-functioning. In these cases, sacrificing the joints may be considered and lengthening may be performed in the joint...
Limb lengthening using ankle joint distraction followed by arthrodesis

Figure 1 Preoperative X-ray of the lower limbs showing the 9 cm length discrepancy on the left tibia (a) with a type 2 tibial agenesis (b and c).

space to correct discrepancies then arthrodesis for correct positioning may be performed during the same procedure. This report describes an intra-articular lengthening technique by progressive distraction or arthrodiastasis followed by talocrural arthrodesis.

Clinical case

The patient was a young 20-year-old man with severe tibial Jones type 2 aplasia [8]. During initial management, tibialisation of the fibula was performed resulting in an increase in the length of the left tibia by several centimetres.

Radiological assessment on the patient’s arrival showed that the left side was predominantly affected with severe tibial agenesis associated with overall deformation of the foot resulting in a 9 cm leg length discrepancy. Femurs were of equal length on both sides. (Fig. 1) and the left tibia was 12.4 cm long. Clinical assessment showed no range of motion in the left ankle and range of motion in the knee of 130° flexion and 0° extension.

Because of the bone damage, lengthening was proposed, with sacrifice of the talocrural joint since it was incongruent and non-functioning.

The initial surgical procedure included tibial debridement and abrasion of the talocrural joint articular surface and placement of a circular external fixator bridging the knee, including a pin in the forefoot to prevent the development of an equinus deformity. At the same time, proximal screws were removed and the soft tissues and the fibular head were completely freed so that it could descend as the length discrepancy was corrected. A progressive lengthening program of 1 mm per day was begun for 90 days to obtain 9 cm of distraction (Fig. 2).

In the second stage of the program, bone reconstruction by the induced membrane technique described by Masquelet et al. [4] was performed by inserting a cement spacer and performing a massive cancellous iliac graft with the external fixator in place (associated with repositioning of the transtibial nail because of migration) (Fig. 3).

During the postoperative course, the scar opened because of staphylococcus aureus infection. This was treated medically by antibiotics and wound healing management.

During the lengthening and consolidation phase, external fixation was maintained for 13 months with dynamisation in the last month. Full weight was applied progressively for three more months in a thermoformed cast.

After a postoperative period of stiffness due to bridging of the joint by the external fixator, the patient’s walking gradually improved during clinical follow-up, with satisfactory knee stability and full articular range of motion at the last follow-up assessment (flexion 130°, extension 0°). Two years after the end of surgical management the patient can walk easily and without pain without help or a brace (Fig. 4).

Discussion

Management of congenital diseases of the lower limbs is often complex because significant lengthening is required. In these cases, as in cases of substantial traumatic bone loss, the difficulty of surgery is satisfactory restoration of bone stock. The induced membrane technique described by Masquelet et al. [4] is a reference in the management of bone loss and provides good clinical and radiological results. For example, Biau et al. [9] rebuilt 16 cm of femoral diaphysis in two stages after resection of an Ewing’s sarcoma using this technique. In these clinical situations, it is essential to preserve the joint so that function remains as normal as possible.

However in congenital abnormalities, limb length discrepancies can also be accompanied by articular anomalies due to incongruence resulting in loss of function. In these cases preserving the joint is not an absolute necessity and the joint can be the point of departure of lengthening. This technique, which is called arthrostasis, is similar to temporary articular distraction or arthrodiastasis in the management of severe osteoarthritis of the ankle (Kluesner et al. [10]). The aim of this therapeutic approach is dual; to simultaneously correct length inequalities and to obtain solid, well-positioned arthrodesis at the end of the therapeutic program.

In type 2 forms of tibial agenesis, the traditional surgical steps include tibialisation of the fibula, repositioning of the foot, then correction of length discrepancies during one or several lengthening programs. Courvoisier et al. [1] reported good clinical results and mean lengthening of 9 cm in their series with the Ilizarov fixator after a mean follow-up of 18.3 years. We agree with these authors that it is important to bridge the knee during the lengthening stage to
**Figure 2** Postoperative X-ray after the 90-day lengthening program (a) and insertion of the cement spacer (b).

**Figure 3** Postoperative X-ray after realization of the cancellous bone graft (a) and after bony consolidation 13 months after beginning of the surgical program (b).
preserve stability which is the determining factor for future function.

The next goal is to obtain consolidation of the lengthened segment, which can be achieved by several methods. For example, Courvoisier [1] planned short sessions of lengthening to limit the risk of pseudarthrosis, fracture of the regenerating tissue and malalignment. To favor union in a program with one session of lengthening, we decided to use the induced membrane technique followed by a cancellous bone graft. Whatever the technique chosen, all involve difficult surgical programs associated with well-known risks, in particular infection, during progressive lengthening by external fixation.

The technique described in this report, based on simultaneous talocrural arthrodesis and progressive lengthening starting intra-articularly provided good clinical and radiological results more than two years after the end of the surgical program. We feel that this technique is indicated in specific forms of type 2 tibial aplasia with a preoperatively stable knee to ensure satisfactory postoperative function. Nevertheless, it remains a difficult surgical program, which can be a therapeutic alternative in the management of complex limb malformations.

**Disclosure of interest**

The authors declare that they have no conflicts of interest concerning this article.

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