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The use of vascularised bone capping to prevent and treat amputation stump spiking in the paediatric population.

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The use of vascularised bone capping to prevent and treat amputation stump spiking in the paediatric population.

Abstract:

Background

Overgrowth of the stump skeleton is a major complication seen in children after an amputation. In advanced cases, perforation of the bone spike through the skin can occur. Many surgical treatments have been employed to treat and prevent this, with best results seen when non-vascularised osteo-chondral bone grafts are placed to try to mimic a trans-articular amputation. We reviewed our outcomes using vascularized bone flaps to prevent and treat spiking.

Patients and Methods

Between 2000 and 2016 we carried out 6 vascularised osteo-cartilaginous bone capping procedures. Five patients underwent the procedure as an adjunct to primary amputation and in a single patient it was used to treat established bone spiking. Trauma accounted for three cases, with the other three being tumour, vascular malformation and ischemia. Three patients had pedicled bone flaps placed on the amputation stump and three underwent free tissue transfer (free calcaneus, free scapular angle, and free proximal tibia). Five cases involved lower limb amputations, with one in the upper limb.

Results

One patient had an early post-operative complication in the form of partial skin flap necrosis that required debridement and skin grafting. All bone flaps survived. Mean follow-up was 6.5 years. All patients had bony union with no development of stump spiking. Two patients required further procedures unrelated to the bone flaps.

Conclusion

Vascularised bone flaps to cap amputation stumps may be a safe and effective method of preventing and treating long-bone stump spiking following amputation in children

Keywords: amputation, children, overgrowth, stump, capping, epiphysis, bone transfer

Introduction:

Overgrowth of the stump skeleton is the major complication seen in children after an amputation. Prevalence varies widely in different series, ranging from 4% to 86%, with the prevalence decreasing as children get older, and after skeletal maturity it is exceptionally rare. It appears most frequently in the humerus, fibula, tibia and femur [1–3].

This bony overgrowth is characterized by swelling, warmth, and tenderness at the amputation stump, and in many cases by the formation of a bursa over the bony spike. In advanced cases, perforation of the bone spike through the skin can occur. When this occurs there is difficulty with using an end-bearing prosthesis. Clinical diagnosis may be confirmed radiologically with a characteristic distal tapering of the bone to a narrow tip seen on x-ray. Overgrowth is often recurrent, and may require multiple revision operations [4–6].

Surgical strategies aimed at correcting this problem are multiple and may be broadly divided into those correcting established overgrowth and those attempting to prevent it at the time of primary amputation. Amputation overgrowth is considered to be a malfunction of normal fracture healing and growth mechanisms occurring at the amputated end of the bone [4]. Occlusion or plugging of the medullary canal appears to significantly retard these processes.

Initial clinical management of stump spiking includes prosthetic or lifestyle adjustments, followed by operative revision if signs or symptoms persist. Reported surgical strategies used to treat terminal osseous overgrowth once it has occurred include simple excision [2–5]; the use of silicone [7], polyethylene or Teflon caps [8]; modifications of the Ertl tibiofibular osteomyoplasty [9]; calcaneo-cutaneous pedicle flaps [10] ; as well as vascularised and non-vascularised bone capping [4,11–13].

Stump capping with osteocartilaginous grafts appears to be the most effective treatment for stump spiking. Vascularized bone flaps have been shown to be superior to non-vascularized grafts in the setting of scaphoid[14], mandible[15] and hip reconstruction[16]. For this reason we elected to use vascularized bone flaps in our series. This technique has previously only been reported in two cases of congenital pseudarthrosis of the tibia[10], without long-term follow-up. We describe a case series of 6 patients treated with vascularized autologous bone to cap amputation stumps. Long-term function and radiological stump appearance is reported.

Patients and Methods:

This report was a retrospective review of a case series and was reviewed and approved by our institution's research review board. We reviewed the medical records of all children who underwent placement of a vascularised autologous osteo-cartilaginous cap to prevent or treat osseous overgrowth of an amputation stump at a single pediatric tertiary care hospital from 2000 – 2016.

We carried out 6 vascularised osteo-cartilaginous bone capping procedures. Demographics are outlined in Table 1. Five patients underwent the procedure as an adjunct to primary amputation and in a single patient it was used to treat established stump spiking. The mean age of the patients at the time of the capping procedure was 9 years 10 months (range 7 years to 13 years 2 months). Trauma accounted for 3 cases, with the other 3 being tumour, vascular malformation and ischemia.

Surgical techniques:

Three patients had pedicled bone flaps placed on the amputation stump and three underwent free tissue transfer. Five cases involved lower limb amputations, with one in the upper limb.

Of the two lower limb pedicled vascularised osteo-cartilaginous cap procedures one was in the setting of trauma, the other in a patient with Factor V Leiden deficiency with non-salvageable distal ischemia. In the trauma setting a patient with a Symes amputation, a fillet foot flap was used to provide soft tissue cover to the amputation stump and the vascularised talus was used to cap the fibula. In a patient with amputation at mid-radius and ulna level, the distal radius epiphysis and metaphysis supplied by the anterior interosseous and radial artery was used to cap the end of the radial osteotomy.

Two patients underwent free vascularised osteo-cartilaginous flaps for prevention of overgrowth in primary amputations; one elective and on in the trauma setting. The trauma patient had the amputation stump covered by a free calcaneus and sole fillet composite flap that was fixed to the end of the tibia with a cancellous screw. The flap was vascularised using the anterior tibial vessels end to end on to the posterior tibial vessels. In the case of an osteosarcoma of the distal metaphysis of the femur, we used the tibia, distal to the tumour to provide an amputation stump of adequate length to facilitate prosthesis wear and ambulation (Fig. 1).

In a patient where the tibia had grown through the skin envelope and developed chronic osteomyelitis at 9 years after a below knee amputation, we managed with an osseomusculocutaneous latissimus dorsi with the scapula angle free flap.

Results:

Five patients had an uncomplicated post-operative course with no acute wound healing problems. One trauma patient who underwent a free calcaneus and fillet of sole flap required operative debridement and skin grafting for partial skin flap necrosis.

The bone flaps survived in all patients, and all 6 achieved clinical and radiological bony union facilitating prosthesis use.

One patient, 16 years after a free fillet foot flap with calcaneus, had radiological evidence of fibular overgrowth, but this was not clinically evident, and did not cause pain or discomfort or interfere with the ability to wear a prosthesis. A second patient required further surgery in the form of removal of metalwork and hemi-epiphyseodesis for medial malleolar overgrowth. There was no clinical or radiological evidence of fibular overgrowth however, where the vascularized bone flap had been placed.

The single case where a free scapular flap was used to treat established stump spiking at 4 years of follow up had no further stump problems or signs of overgrowth.

Mean follow-up period was 6.5 years (range 3.5 years to 16 years). All 6 patients had stable amputation stumps with no clinically evident stump spiking. All could wear prosthesis and had good function.

Case Reports:

Case 1:

The single upper limb case was an 8-year-old boy who had underwent multiple previous debulking procedures for a large lympho-venous malformation of the left forearm and hand (Fig. 2a). Despite these procedures the patient had poor function in the hand, persistent pain and recurrent infections and the decision was taken to proceed to amputation. Amputation was carried out at mid-radius and ulna level. The distal radius epiphysis and metaphysis, including the growth plate, was maintained on the anterior interosseous and radial artery blood supply and used to cap the end of the radial osteotomy (Fig. 2b). The patient's immediate post-operative course was uneventful. Clinically and radiologically the flap has survived. At 4 years follow up the patient has had no stump problems, no clinical or radiological evidence of overgrowth. X-ray at one year post-operatively shows growth from the distal radial epiphysis with 6mm of growth seen (Fig. 2c).

Case 2:

A 9-year-old girl presented with an osteosarcoma of the distal metaphysis of the femur. Resection would result in a high level above knee amputation and the family deemed rotationplasty unacceptable. As an alternative we proposed using an inverted, reversed proximal tibial free flap.

Skin and muscle flaps were raised though a transverse incision at the knee with distal medial and lateral extensions. The long saphenous vein and the femoral and popliteal vessels were preserved. The distal popliteal, posterior tibial, and anterior tibial vessels were identified and preserved (Fig. 3a). The popliteal artery was divided proximal to its trifurcation, allowing removal of the leg distal to the knee joint.

Tumor resection was completed 15 cm from the distal end of the femur. The tibial bone flap vessels were prepared on a side table. The tibia was transected distal to the nutrient vessel at 14 cm from the proximal joint surface. The tibia was then inverted, reversed, and fixed with an intramedullary nail. The posterior tibial artery was anastomosed end to side to the femoral artery due to size mismatch. The popliteal vein was anastomosed end to end, to a similar sized tributary of the long saphenous vein (Fig. 3b).

The patient's immediate post-operative course was uneventful. Clinically and radiologically the flap has survived. At 3.5 years follow up the patient had no stump problems, wore prosthesis comfortably and competed at sports (Fig. 4a & 4b).

Discussion:

Overgrowth is one of the most frequent complications following amputation in children, occurring in 4% to 35% depending on the age of the patient and location of amputation. In a series of 109 children who underwent transmetaphyseal or transdiaphyseal amputation, 46% underwent at least one revision for overgrowth, and 18% required multiple revisions [6]. Overgrowth is more common in younger children, but can occur at any age prior to skeletal maturity. Abraham *et al.* found that patients who underwent amputation after the age of 12 did not require revision surgery[5]. Other series, however, have documented overgrowth requiring revision in children 12 and over, albeit less commonly[3,17]. For the two adolescents in our series undergoing primary amputation, aged 10 and 12, we elected to cap the amputation stump because of this risk.

The pathophysiology of terminal osseous overgrowth remains poorly understood. It has been suggested that periosteal and/or endosteal abnormalities, endochondral ossification, heterotopic ossification, and metaplasia of fibrous tissue are responsible [4,5]. Initial attempts at preventing overgrowth focused on proximal epiphysiodesis in the belief that this growth centre was a driver of distal overgrowth [18]. Later work in humans and animals has recognized that the distal osteotomy site is the primary cause of overgrowth due to woven bone being deposited on the original cortical bone, leading to the described soft tissue problems [2,19,20].

Many approaches have been taken in treating this problem, with some form of "capping" of the bone end after debridement of the overgrowth proving most efficacious. It is thought that by occluding the medullary canal and distal aspect of the bone, overgrowth is inhibited by preventing communication between external mediators and bone marrow elements and endosteum [4]. This approach was first described in the late 1960's by Swanson who used a heat-moulded, intramedullary stemmed and collared silicone implant which is sutured to the bone. Swanson described a cohort of fourteen children who had undergone multiple revision procedures for terminal bony overgrowth. Although the series showed a reduction in the rates of overgrowth, the number of complications was high with infection, skin breakdown and implant fracture seen in 50% of cases [7,21].

Later work by Meyer *et al.* looked at using a porous polyethylene cap, which they postulated would allow for bony ingrowth and therefore better retention. In 6 children with a mean of 14 months follow up they showed good short-term results, but longer term had the same complications as silicon with fracture of the implant, extrusion and infection [22]. Tenholder *et al.* described their experience using PTFE felt caps in 17 cases of established overgrowth. Mean follow-up was 4 years 9 months. They compared their results with historical control groups undergoing simple excision and showed a revision rate of 29% with PTFE caps compared to 86% in the control group [8].

Despite the obvious advantages of synthetic caps, the complication rates led to investigation of the use of autologous materials. In 1974 Marquardt introduced cap-plasty of the stump using an autologous cartilage-bone graft to bear weight and to prevent overgrowth and skin penetration [23,24]. The goal being conversion of a transosseous amputation into a stump resembling that seen in a

disarticulation. A number of different sources of autologous graft have subsequently been described including iliac crest wedges, head of fibula, and various bone segments from the amputated limbs [4,10–12]. The majority of described treatments use non-vascularised bone grafts despite the incidence of bony union being higher, with less need for revision surgery, utilising vascularised bone flaps.

Bernd *et al*. had a revision rate of 12% (6 of 50 limbs) with autologous bone "cap-plasties" performed for the treatment of established overgrowth following amputations. Follow up was for a mean of 7 years. A variety of graft sources were used including iliac crest, fibular head and talus [12]. Davids *et al.* looked at their results over a 31-year period where treatment of overgrowth evolved from simple excision to synthetic capping to autologous capping with non-vascularised iliac crest. They showed a decrease from 84% revision rate in resection group, to 77% in the synthetic cap group and ultimately a 30% revision rate in the iliac crest group [4].

Using a rabbit model of amputation Wang *et al.* showed that capping with a metatarsal epiphysis reduced the rate of overgrowth from 80% in the control group to 0% in the epiphyseal transfer group [20]. Subsequent clinical work has also shown good results with non-vascularised epiphyseal transfer. Benevenia *et al.* showed an overgrowth rate of 10% (1/10) of primary amputations for tumor with use of a prophylactic epiphyseal transplant from the amputated segment [11]. They compared these results with a control group in their institution who had an 86% overgrowth rate. In most patients the first metatarsal epiphysis was used, with the distal fibula also being used in a few cases. A larger study by Fedorak *et al.* also showed a bony overgrowth rate of 10% following proximal fibular epiphysis grafting to treat overgrowth of the tibia.

Free epiphyseal transfer to maintain growth was first attempted in the late 19th century. Results with this technique remained disappointing, with limited growth and early closure of the growth plate until the advent of microvascular transfer with early studies demonstrating that growth could be maintained when the blood supply to the growth plate was preserved [25–28]. Weber described a new technique of stump capping of the lower leg in two patients with congenital pseudoarthrosis of the tibia where all neurovascular structures supplying the calcaneus are carefully maintained and subsequently, the tibia and fibula are inserted into the calcaneus. With 5 years of follow up no overgrowth was seen [10].

Using this knowledge we elected, when possible, to provide paediatric patients undergoing transosseous amputations with vascularised epiphyseal stump capping. Using a vascularized flap rather than a non-vascularized graft allowed us to use larger bone segments, when indicated, to preserve length, to include vascularized soft tissue cover in the form of fillet of sole flap as well as talus in one patient, and to potentially preserve growth with a vascularized epiphysis. In all the prophylactic cases the concept of "spare parts" surgery was used, therefore limiting donor site morbidity, as what was used would have otherwise been discarded with the amputation specimen. Only one patient required any revision surgery with a mean follow-up of 6.5 years. In this case the distal tibial epiphysis had been preserved and he underwent medial distal tibial hemi-epiphyseodesis as the medial malleous was becoming prominent. He also underwent removal of prominent screws at the same time.

We believe this to be the first reported case series describing the use of vascularized bone flaps to prevent stump spiking with medium and long term follow up.

Conclusion:

Vascularised bone flaps to cap amputation stumps may be a safe and effective method of preventing and treating long-bone stump spiking following amputation in children. They may provide a well vascularised stump cap with good bony healing, prevent overgrowth, and have no donor site morbidity when spare parts can be used.

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Figure Legends

Fig. 1 Illustration of the reconstruction with preoperative anatomy, and technicalities of the procedure with emphasis on the microvascular anastomoses, intramedullary fixation. Of note the venous drainage of the flap was in an antegrade direction but arterial flow was in a retrograde direction via the distal end of the posterior tibial artery.

Fig. 2a, 2b & 2c. (a) Lympho-venous malformation of the hand prior to amputation. (b) Immediate post-operative x-ray of the distal radius used to cap the amputation stump. (c) X-ray 1 year post-operatively showed 6mm growth of the radius from the distal epiphysis with no signs of bony overgrowth.

Fig. 3a & 3b. Photograph of (a) the prepared flap. The ruler shows 14cm of tibia with green arrows depicting the popliteal vessels just proximal to trifurcation and the posterior tibia vessels at the level of the tibial osteotomy. (b) the inset flap after intra-meduallary nail fixation. The arterial (cranial) and venous (caudal) anastomoses are indicated by the tips of the surgical instruments.

Fig. 4a & 4b. Immediate post-operative x-ray of the proximal tibia used to cap the femoral amputation stump. X-ray 3.5 year post-operatively showed 25mm growth of the distal bone and no signs of bony overgrowth.

Table 1. Patient demographics

Patient	Age at Index Procedure (years)	Sex	Site	Diagnosis	Flap Used	Flap size	Donor Vessels / Pedicle	Post-Op Complication	Length of Follow Up (years)	Final Outcome
1	12	Female	Tibia/Fibula	Trauma	Free vascularised calcaneus	2 x 2 cm	Posterior Tibial artery + venae commitans	Nil – radiological posterior spike clinically undetectable	16	Good function, no stump problems
2	10	Male	Tibia/Fibula	Arterial Thrombosis	Pedicled distal tibial epiphysis + metaphysis	5 x 2 cm	Posterior Tibial artery + venae commitans	Nil	7	Good function, no stump problems
3	7	Male	Fibula	Trauma	Pedicled Talus	3 x 1.5 cm	Posterior Tibial artery + venae commitans	Removal screws + epiphyseodesis	5	Good function, no stump problems
4	9	Female	Femur	Osteosarcoma	Free Proximal Tibial Epiphysis + Metaphysis	14 x 2 cm	Femoral artery + Popliteal vein	Nil	3.5	Good function, no stump problems
5	8	Male	Radius/Ulna	Lymphatic-Venous Malformation	Pedicled Radius Epiphysis + Metaphysis	2.5 x 1.2 cm	Radial + Anterior Interosseous arteries + venae commitans	Nil	4	Good function, no stump problems
6	13	Male	Tibia/Fibula	Trauma	Free Scapular angle + Latisimus Dorsi flap	2 x 1.5 cm	Posterior Tibial artery + venae commitans	Nil	4	Good function, no stump problems

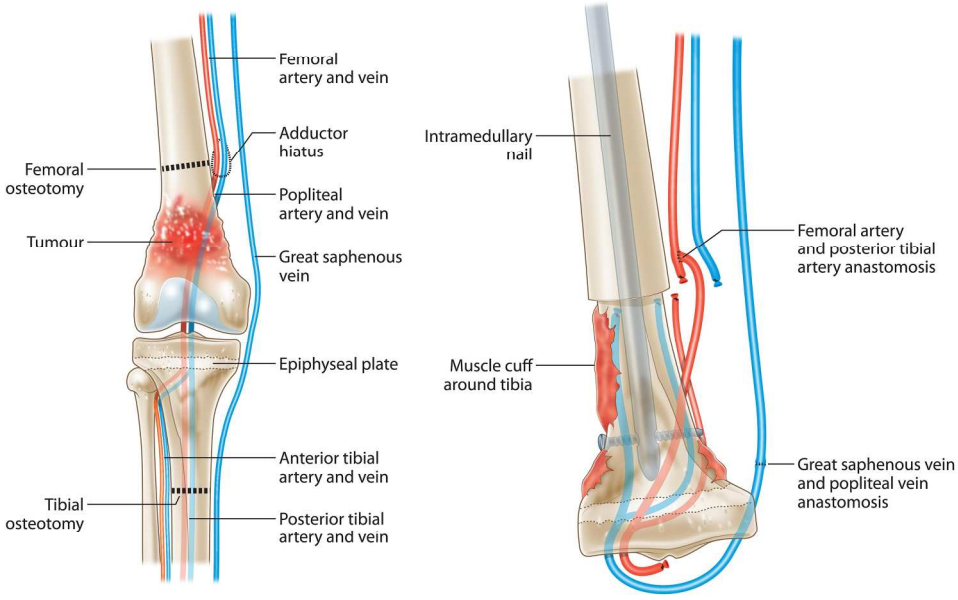

Fig. 1 Illustration of the reconstruction with preoperative anatomy, and technicalities of the procedure with emphasis on the microvascular anastomoses & intramedullary fixation. Of note the venous drainage of the flap was in an antegrade direction but arterial flow was in a retrograde direction via the distal end of the posterior tibial artery.

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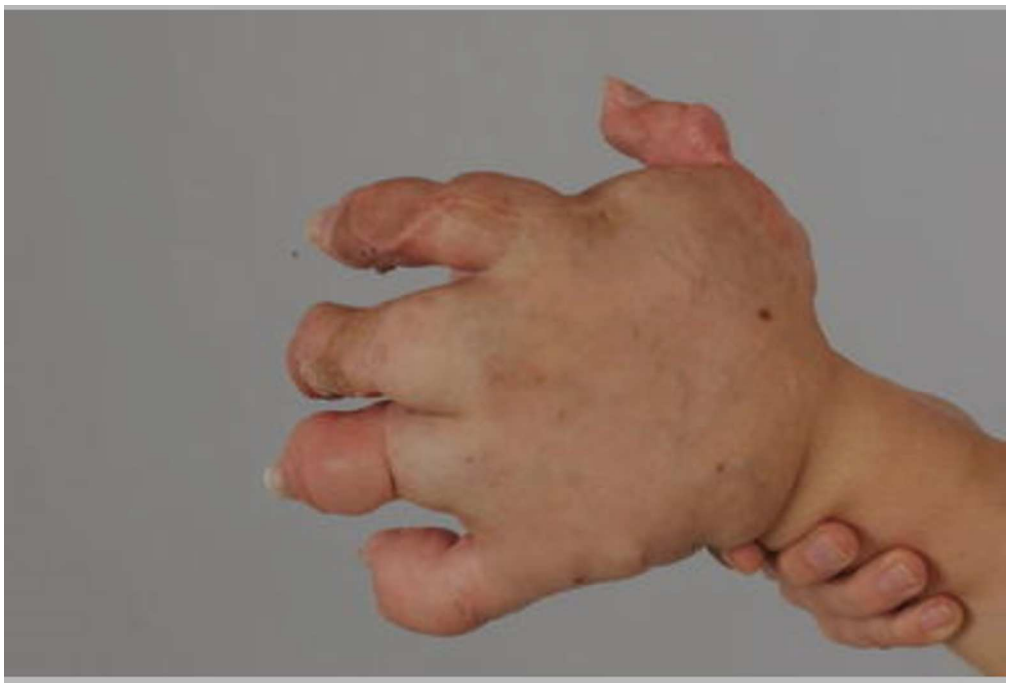

Fig. 2a, 2b & 2c. (a) Lympho-venous malformation of the hand prior to amputation. (b) Immediate post-operative x-ray of the distal radius used to cap the amputation stump. (c) X-ray 1 year post-operatively showed 6mm growth of the radius from the distal epiphysis with no signs of bony overgrowth.

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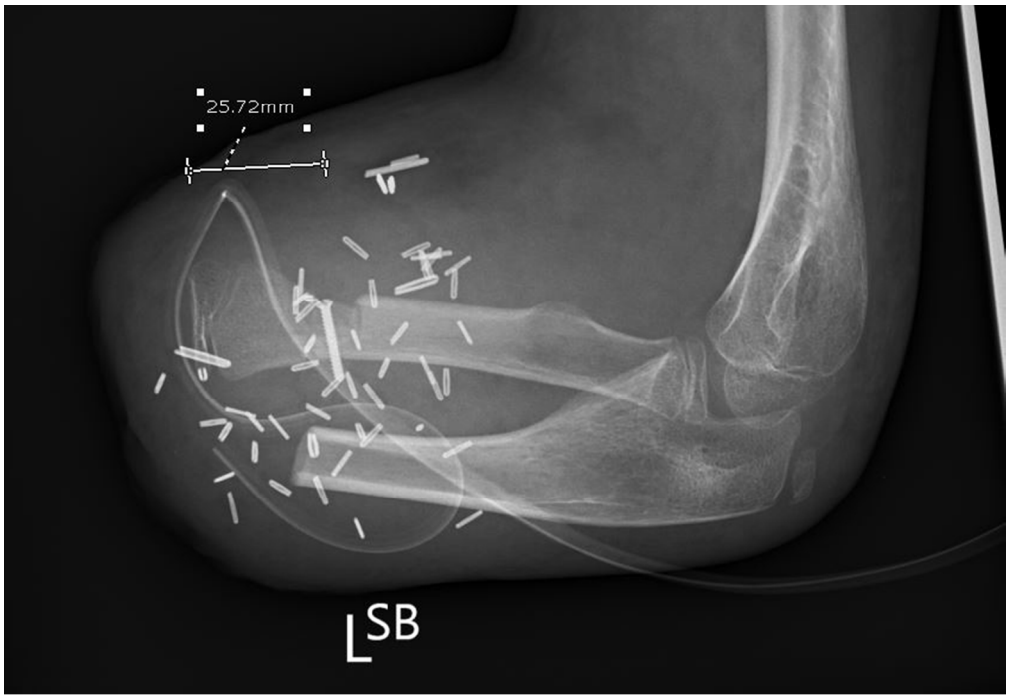

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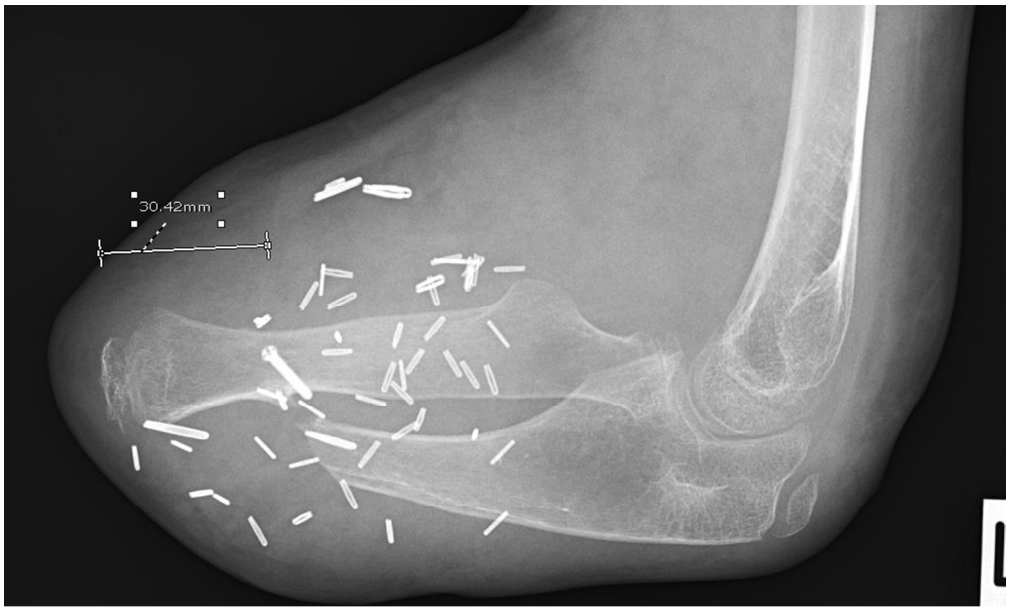

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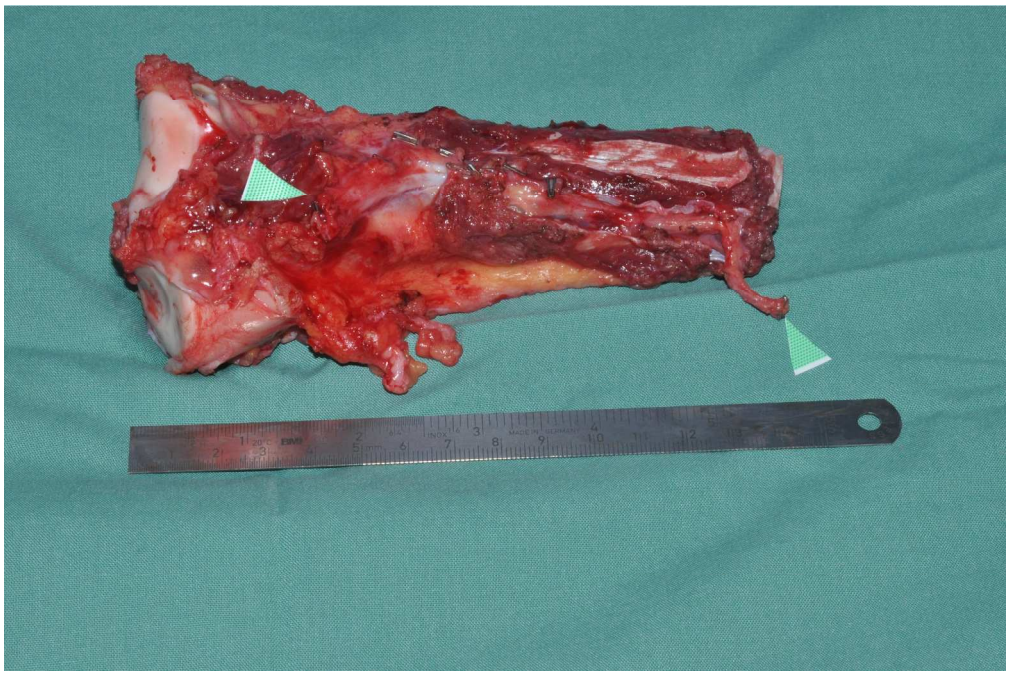

Fig. 3a & 3b. Photograph of (a) the prepared flap. The ruler shows 14cm of tibia with green arrows depicting the popliteal vessels just proximal to trifurcation and the posterior tibia vessels at the level of the tibial osteotomy. (b) the inset flap after intra-meduallary nail fixation. The arterial (cranial) and venous (caudal) anastomoses are indicated by the tips of the surgical instruments.

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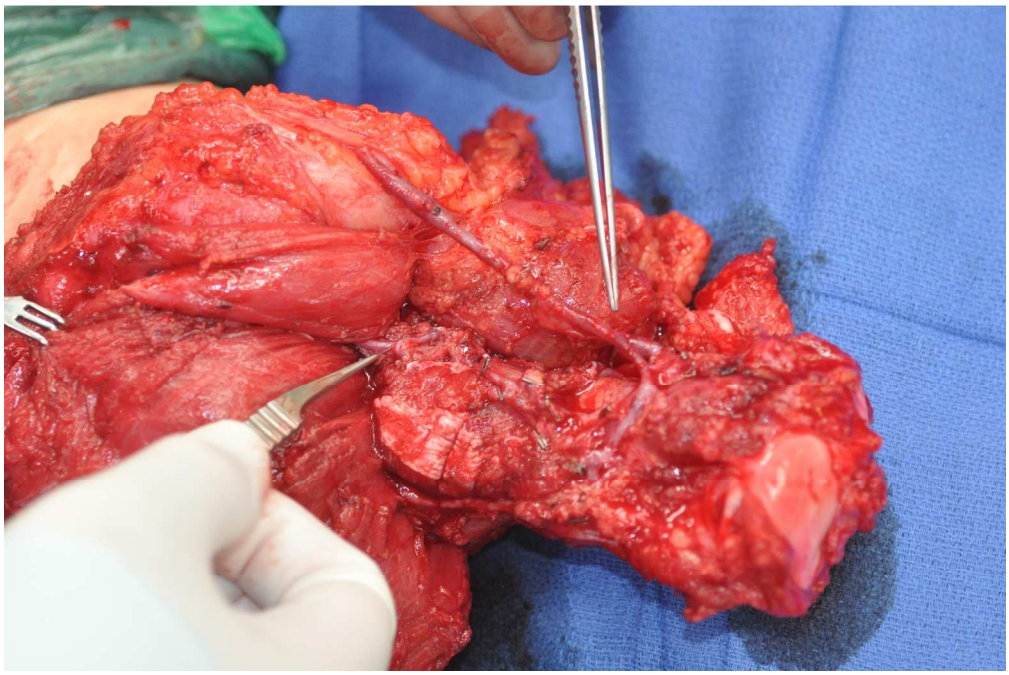

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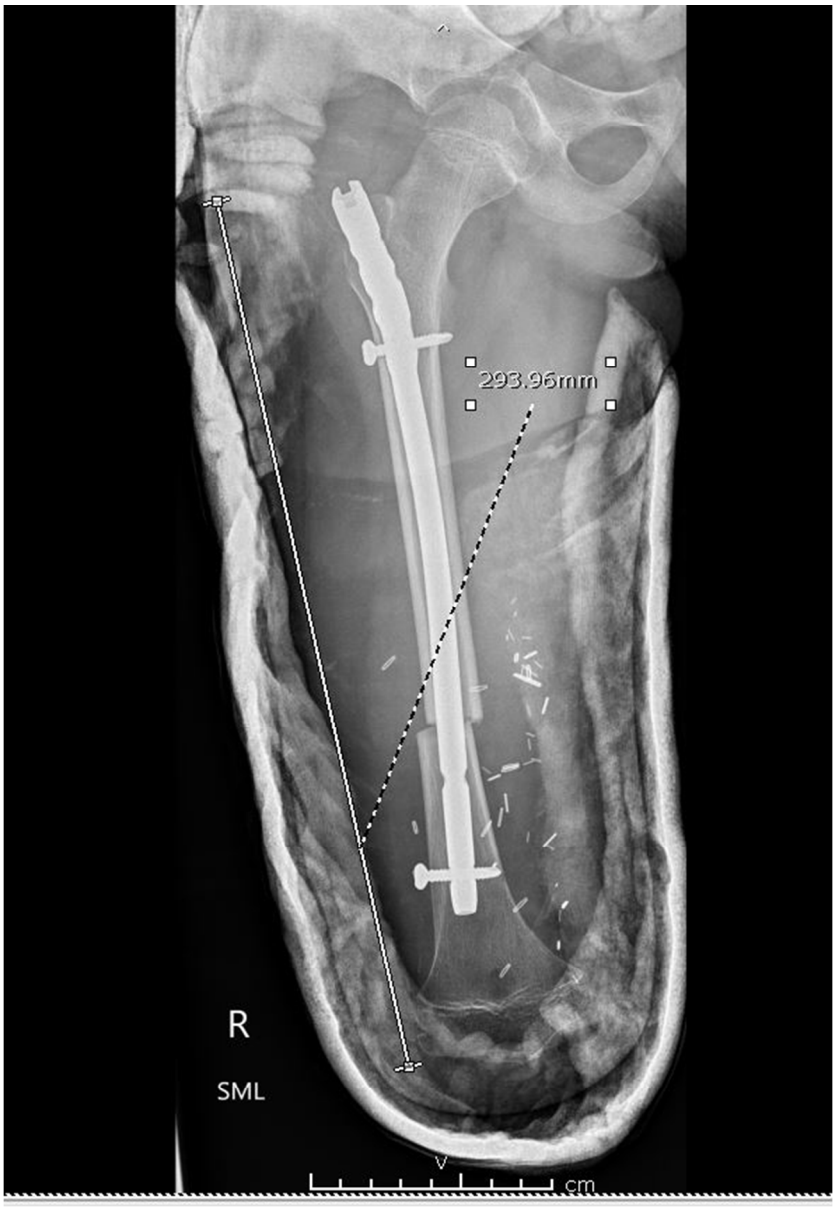

Fig. 4a & 4b. Immediate post-operative x-ray of the proximal tibia used to cap the femoral amputation stump. X-ray 3.5 year post-operatively showed 25mm growth of the distal bone and no signs of bony overgrowth.

156x226mm (96 x 96 DPI)

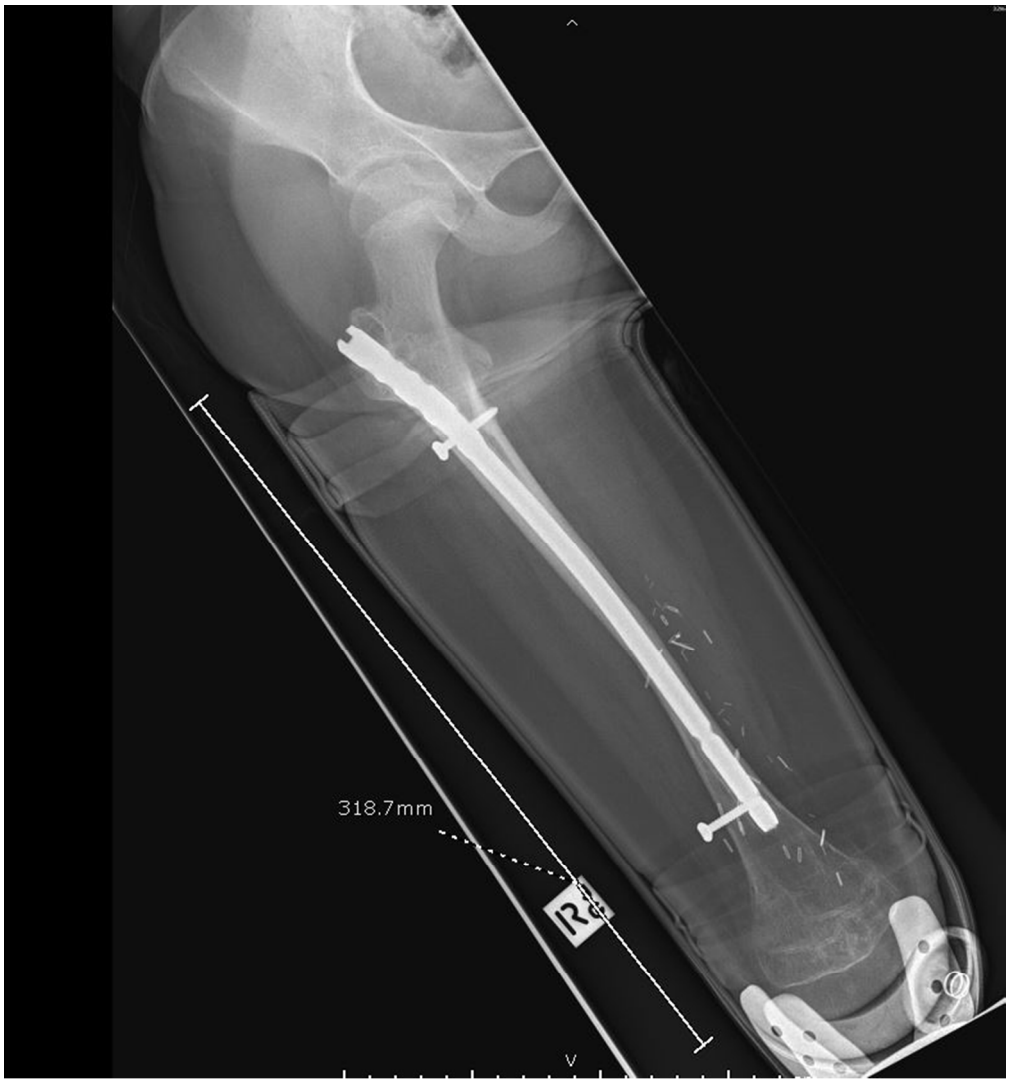

Fig. 4a & 4b. Immediate post-operative x-ray of the proximal tibia used to cap the femoral amputation stump. X-ray 3.5 year post-operatively showed 25mm growth of the distal bone and no signs of bony overgrowth.

206x221mm (96 x 96 DPI)