



Iliac crest apophysis transfer to treat stump overgrowth after limb amputation in children: case series and literature review

Rami Jahmani¹ · Craig Robbins² · Dror Paley²

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Abstract

Purpose Stump overgrowth is the main problem of limb amputation in children. Many surgical procedures have been developed to overcome the problem, but all have shown inconsistent results. The only surgical procedure that has been successful in preventing overgrowth is capping of the amputated limb with a cartilaginous cap taken from the amputated limb, usually from the head of fibula. A donor site is not available in revision cases. Iliac crest apophysis transfer was suggested to treat the condition, but has never been previously reported. The purpose of this study is to review the results of iliac crest apophysis transfer to prevent stump overgrowth.

Method Five children with amputation stump overgrowth underwent iliac apophyseal transfer to cap the resection site of the overgrowth.

Results Retrospective review showed that three of the five suffered recurrent stump overgrowth two to four years after the index surgery. All three were revised again. One patient was lost to follow-up.

Conclusion Although many studies showed capping of the stump with cartilaginous cap to be successful in preventing stump overgrowth, iliac crest apophysis transfer was only successful in one of four cases available to follow-up (25%). This is not a reliable enough method to be used routinely.

Keywords Children amputation · Stump overgrowth · Marquardt · Iliac crest bone grafting

Introduction

Stump overgrowth is the most common complication after limb amputation in children with a prevalence of 4 to 86% [1]. The exact pathogenesis is unknown. The younger the patient, the greater the incidence of stump overgrowth. Overgrowth (heterotopic ossification) in adults rarely requires revision surgery. The incidence is higher with post-traumatic

amputation and lowest in cases of limb agenesis. It has never been reported in disarticulation amputation [2]. Incidence is twice as high in boys than in girls [3].

The chief complaint relates to prosthetic fitting, skin perforation may occur. The initial treatment is prosthetic modification. Revision of the stump has been associated with a high incidence of recurrence [2, 4, 5]. Many surgical procedures have been suggested to reduce the incidence of recurrence. Prophylactic capping of the stump with a cartilaginous cap taken from the amputated limb, usually the head of the fibula (Marquardt 1974), is associated with successful results and has reduced the incidence of recurrence to zero [6, 7]. Donor site availability is the main obstacle in Marquardt stump plasty for revision cases. Many synthetic materials have been suggested to cap the end of the bone. These results are not as favourable as with the Marquardt procedure. Marquardt suggested using the cartilaginous apophyseal cap taken from iliac crest to treat stump overgrowth but never tested this idea. The senior author began using the iliac apophysis to cap the residual limb in 2011. The purpose of this study is to report the results of this procedure.

✉ Rami Jahmani
dr.jahmany@yahoo.com

Craig Robbins
crobbins@paleyinstitute.org

Dror Paley
dpaley@paleyinstitute.org

¹ Department of Special Surgery, Faculty of Medicine, Jordan University of Science and Technology, P.O Box 3030, Irbid 22110, Jordan

² Paley Orthopedic and Spine Institute, St. Mary's Hospital, West Palm Beach, FL 33407, USA

Method

After obtaining Institutional review board and ethical committee clearance, we retrospectively reviewed the medical reports of patients treated for stump overgrowth at Paley Orthopedic and Spine Institute through the period of time between 2011 and 2017. Those who had been treated by apophyseal iliac crest capping were included in the study. Data include age at the time of apophyseal capping, cause of amputation, site of amputation, gender, and patients' follow up X-rays. Failure of treatment is considered when the case needed revision surgery for stump overgrowth.

Surgical procedure

A transverse skin incision is made over the ends of the bones. The skin, fascia, and muscle are reflected back as a flap exposing the sharp ends of the tibia and or fibula for the lower leg and humerus for the arm. The bone spike is resected with the periosteum. A separate incision is made over the iliac apophysis. Two cuts are made parallel to each other to cut out the graft. The apophysis is cut with a knife while the bone is cut with a saw. The two tables of the ilium are split, hinging on the apophysis. This graft is now mounted on the ends of the tibia and fibula to join the two bones together (much like the Ertl procedure). The graft is secured with two retrograde cannulated screws, one in the tibia and one in the fibula. In the humerus, it requires one screw (Figs. 1 and 2).



Fig. 1 Overgrowth before surgery



Fig. 2 Stump with apophyseal cap fixed using two screws, one in the tibia and one in the fibula

Results

There were five patients, aged one to eight years old at the time of apophyseal capping; four transtibial and one transhumeral amputees. Two tibias are post-traumatic, two tibias and the one humerus were due to congenital amniotic bands. Three transtibial patients showed recurrent overgrowth and needed revision surgery; the one (humerus) did not overgrow. One tibial patient was lost to follow-up (Table 1).

Discussion

Stump overgrowth is one of the major complications of limb amputation in children. It may happen in any bone, most commonly in the tibia and humerus and less common in the femur [3]. Treatment has been always a challenge due to a high recurrence propensity. Lack of understanding of the pathogenesis has led to a wide variety of treatment recommendations. Multiple revisions (more than one revision) are reported to be as high as 18% of cases [8]. One case of six revisions has been reported [9]. Pellicore et al. observed proximal growth stimulation and proposed that stump overgrowth is because the soft

Table 1 Patients and results

Patient	Age at time of surgery	Site	Cause	Follow-up duration	Revision surgery, time ^a
1	1 year	Humerus	ABS ^b	2 years	No
2	3 years	Tibia	ABS	4 years	4 years
3	8 years	Tibia	Trauma	4 years	2 years
4	8 years	Tibia	Trauma	3 years	3 years
5	2 years	Tibia	ABS	Missing	

^a Time from capping surgery until revision

^b ABS: amniotic band syndrome

tissues cannot keep up with the rapid bone growth [10]. Attempts to treat the condition by proximal epiphysiodesis and leaving a redundant soft tissue envelope failed to stop overgrowth [11].

The high incidence of stump overgrowth in surgical amputation and post-trauma patients [5], compared with the absence of stump overgrowth in disarticulation amputation, and congenital agenesis [12] suggested that stump overgrowth might be a result of surgical or injury trauma rather than a phenomenon of growth stimulation of the stump. This would mean that it is related to a local process of bone formation and wound healing occurring in the distal stump. Aitken, by implanting a radiographic

marker, confirmed that overgrowth occurs distal to the marker and proved that overgrowth does not represent an epiphyseal contribution, but rather a local phenomenon of bone healing [13, 14]. This explains why overgrowth does not occur in disarticulation where there is intact articular cartilage rather than transected bone. If stump overgrowth is a local phenomenon, it is unclear why it is not seen following adult amputations. Speer described the pathogenesis of stump overgrowth in the immature skeleton of rabbits and explained why it does not occur in mature skeleton [15]. His study indicates that an amputation stump responds by wound healing and intramembranous bone formation. In the immature

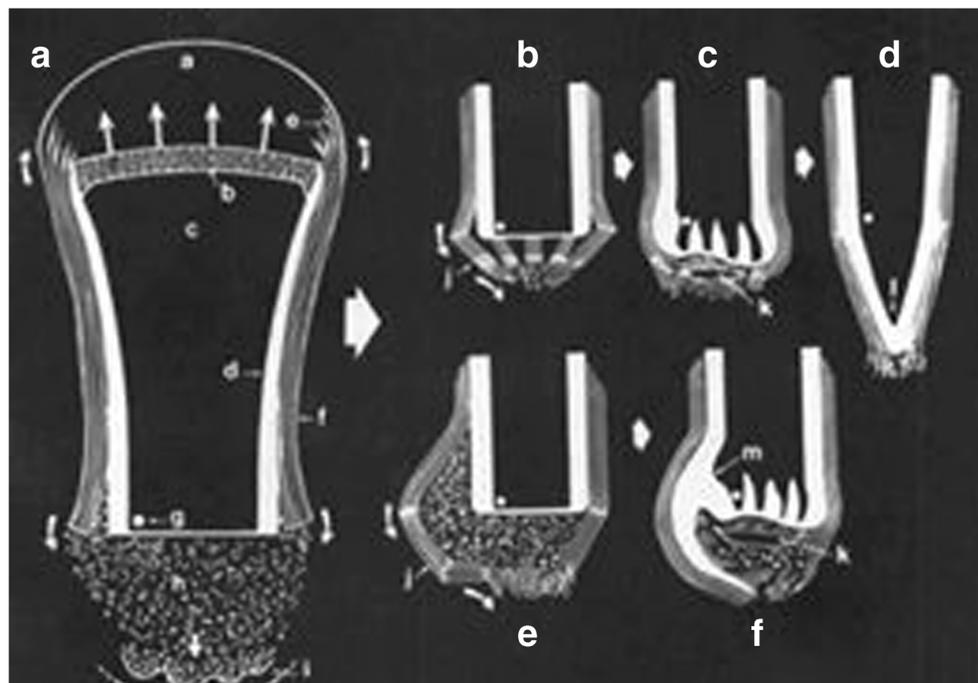


Fig. 3 Pathogenesis of amputation stump overgrowth showing growth mechanisms and evolution of stump shapes as seen by polarized light microscopy [15]. (A) Initial amputation and illustration of the normal mechanism of longitudinal growth of cortical bone at the growth plate. (B) Organization of collagen fibre groups of scar and periosteum as a continuous mass. (C) Progressive orientation of collagen fiber groups longitudinally about the apex by scar contraction with deposition of new bone at periosteal, endosteal, and apical aspects of the original bone. (D) Conical shape of mature overgrown stump. (E) More extensive

periosteal elevation and haematoma than in A. (F) Formation of bulbous shape stump. [Small case letters designate the following: (a) epiphysis, (b) physis, (c) metaphysis, (d) cortical bone of intramembranous origin, (e) insertion of perichondrial-periosteal collagen fibres into epiphysis, (f) periosteum, (g) metal marker placed in bone at the time of amputation (note in all figures), (h) fibrin clot and haematoma, (i) skin, (j) oriented scar collagen fibers, (k) bursa, (l) closure of medullary canal by appositional new bone, and (m) spur formation combined with overgrowth]

skeleton, the tension on the periosteum elasticity allows it to pull away from the end of the amputee stump and leads to local bone formation (Fig. 3).

Many surgical procedures have been developed to overcome this phenomenon including: de-innervation of the stump, distal stump synostosis, and capping the stump with artificial or biological cap. De-innervation of bone in animals showed reduction of bone formation, but sacrificing of innervations in amputated stump may not be applicable in humans [4, 6, 9, 16, 17].

The idea of capping the stump aims to interrupt the interaction between the endosteum and periosteum to reduce the intensity of healing process. Swanson in 1969 was the first who used this method where he used silicon as a cap [16]. Different capping materials were tried including: cancellous and cortical bone cap from iliac crest, cartilaginous cap from the amputated stump, and polyethylene cap [4, 6, 9, 17]. A comparison study between capping with polyethylene and simple bone graft harvested from iliac crest showed a higher incidence of failure in polyethylene-capped stump; 70% of cases needed revision, mainly due to failure of fixation and infection. Although capping with bone grafting showed relatively good results, it still has 30% of failure rate mainly due to continuation of overgrowth [4]. Marquardt in 1974 has been credited as being the first to propose application of a biological cap to prevent overgrowth of the bone in children. He described his procedure of using an epiphysis taken from the amputated limb (usually the proximal fibula) as a cap to prevent overgrowth of a distal tibia amputation [6]. The goal was to convert a diaphyseal amputation into a stump resembling a disarticulation type. Animal studies on rabbits showed epiphyseal capping to be a very successful procedure to prevent overgrowth [18]. The result of epiphyseal capping of stump in human was tested later, in 1992; recurrent overgrowth requiring reoperation has not occurred in any limbs [7]. Recent

publication of capping the stump with fibular head was associated with 10% of revision for tibia and 30% of revision for humerus; relative high revision rate in humerus was attributed to loose interference fit between humeral shaft and head of fibula [19, 20].

Review of the literature recommends the cartilaginous cap as the best way to treat and prevent stump overgrowth. The cartilaginous cap interrupts the interaction between bone marrow and outside periosteum and pushes the end of the stump away from the transected periosteum cutting off any intramembranous bone formation. Donor site availability is the main challenge in Marquardt stump plasty for revision cases. Using apophyseal iliac crest was first suggested by Marquardt, but has never tested [9]. Unfortunately, our report did not prove this to be an effective solution. The cause of failure could be related to the use of an apophysis instead of an epiphysis for the capping. The role of screw fixation vs smooth K-wires is also not known even though the screws were removed. Another concern of this technique is donor site morbidity. Even after four years of follow-up, a bone defect in the iliac crest is still evident on the pelvic x-ray (Fig. 4). This was not a cause of any clinical symptoms. The limitations of our study are: it is retrospective and small number of patients. In conclusion, apophyseal iliac crest capping failed to treat stump overgrowth phenomenon in children.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

For this type of study formal consent is not required.

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Fig. 4 Iliac crest defect

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