Original article

The role of the periosteal sleeve in the reconstruction of bone defects using a non-vascularised fibula graft in the pediatric population

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A R T I C L E   I N F O

Article history:
Received 31 July 2016
Accepted 10 May 2017

Keywords:
Non-vascularized fibula graft
Long bone defects
Aneurismal bone cysts
Fibrous dysplasia
Bone grafting
Periosteal sleeve

A B S T R A C T

Background: Following resection of large benign bone tumors surgeons are confronted with bone defects severely affecting the stability of a limb. To restore the mechanical continuity of the bone different treatment methods using bone grafts have been described. In pediatric patients the thick periosteal sleeve is thought to contribute to bone formation.

Hypothesis: An intact periosteal sleeve is crucial in bone remodelling around a non-vascularised fibular graft used to bridge large bone defects.

Methods: We present a treatment technique applied in 6 cases comprising of subperiosteal tumor resection at the diaphyseal or metaphyseal level of long bones followed by defect bridging with a non-vascularised fibula graft inserted into the periosteal sleeve of the resection zone. Elastic intramedullary nails or plates were used for stabilisation.

Results: Due to the intact periosteum at the resection site bone integration occurred quickly and full remodelling was seen in all but one case. Tumor location in this case was at the metaphyseal level resulting in tumor resection at the growth plate. Although bone healing at the distal resection site was seen after a few weeks proximal consolidation was only partial. Full reconstitution of the fibula in the remaining periosteal sleeve was seen in 5 cases, partial reconstitution in 1 case.

Discussion: In the pediatric patient, the described technique is an effective and reliable treatment method for large benign bone tumors requiring resection. However, great diameter discrepancy of the donor and recipient site and a thin periosteum can be a limiting factor for its application.

Level of evidence: Level IV clinical study.

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1. Introduction

Benign bone tumors of the dia- and metaphysis of long bones in children and adolescents usually neither cause pain nor functional restrictions. Diagnosis is often made after a pathological fracture or due to an incidental finding on an X-ray. Resection of a benign tumor is rarely indicated but when it becomes necessary the reconstruction of a large bone defect can be challenging. Surgeons have to decide between bone allografts [1,2], vascularised and non-vascularised fibula grafts [3–6], a combination of bone allograft and vascularised fibula graft as described by Capanna [7,8] or the Masquelet’s induced membrane technique [9,10]. When fibula grafts are used vascularised grafts are often preferred over non-vascularised grafts due to their rapid healing potential and superior strength [11]. However, while massive allografts and the Capanna technique can be used to bridge large bone defects of the lower extremity in adults, single vascularised and non-vascularised fibular grafts alone fail in these weight bearing bones. Additionally, harvesting a vascularised graft demands surgical experience and is not possible in every institution. In pediatric patients, different tissue conditions and healing potentials exist. A thick periosteal layer allows for subperiosteal tumor resection and the remaining periosteum is known to contribute not only to bone healing but also to new bone formation. The role of the periosteum in bone regeneration has recently been discussed in detail in two publications [12,13] and several experimental and clinical studies have shown its potential in bone reconstruction with or without further grafts [14–18]. In children, surgeons can exploit this reconstructive potential combining the periosteal sleeve with a structural non-vascularised bone graft. While the non-vascularised bone graft...
2. Materials and methods

Between July 2006 and November 2013 six patients (4 boys, 2 girls) with an average age of 12.6 years (range: 7–15 years) were treated for large benign bone tumors of either the humerus or the tibia. The lesions comprised of four aneurismal bone cysts (ABC), twice located in the diaphysis and once in the proximal metaphysis of the humerus. The forth lesion was found in the proximal tibial metaphysis. The remaining two tumors were fibrous dysplasias (FD) of the tibial shaft (Table 1, Fig. 1). All lesions were extensive measuring between 6.3 × 3.2 cm and 12.5 × 2.5 cm, which was on average 28.7% (range: 23.3–38.5%) of the length of the affected bone (Table 2). Cortical thinning at the tumor site was found in all patients leading to fractures in 3 cases (2 humerai and 1 tibial fractures). The tibia fracture was stabilised using a flexible intramedullary nail (FIN) whereas humeral fractures were treated conservatively with immobilsation of the arm. At the time of tumor resection all fractures had healed.

The surgical procedure consisted of a subperiosteal tumor resection leaving an intact periosteal sleeve at the resection site. Tumors located in the diaphysis were excised with a distal and proximal safety margin of about 1 cm. The metaphyseal ABC of the humerus had a reduced safety margin proximally in order to preserve the growth plate. The ABC located in the metaphysis of the tibia extended to the growth plate, which was thus sacrificed during tumor resection. Following this primary step of the operation a non-vascularised ipsilateral fibula graft, slightly longer than the resected tumor, was harvested in a subperiosteal fashion allowing for new bone formation in the remaining periosteal sleeve. In two cases stabilisation of the tibiofibular syndesmosis using a position screw was required as the distal resection end of the fibula graft was in proximity of the ankle syndesmosis. The resected graft was then introduced into the periosteal sleeve at the tumor resection site and due to the slight over-length impacted distally and proximally. Fixation of the graft was achieved with FINs (T2 kids 2.5 mm, Styker, Switzerland or France) in the humerus (Fig. 2) and with angular stable plates (3.5 mm LCP plate, Synthes, France) for diaphyseal reconstruction of the tibia (Fig. 1). At the metaphyseal resection site of the tibia, an external ring fixator (taylor spatial frame [TSF], Smith and Nephew, France) was applied as stabilisation with a plate was deemed insufficient (Table 1). In one case, a small amount of bone allograft chips (Tutoplast®, 1–5 mm) was added into the periosteal sleeve.

Postoperatively immobilisation was either in an arm sling (humeral reconstruction) or in a non-weight bearing lower leg cast (diaphyseal tibial reconstruction). No additional immobilisation was necessary in the external fixator case. Clinical and radiological follow-ups were carried out in regular intervals of 6–8 weeks till consolidation, thereafter at intervals of 6–12 months until the end of growth. Consolidation was defined as circumferential callus formation with corticalisation on biplane radiographs.

The functional outcome was evaluated with respect to the range of knee/ankle and elbow/shoulder motion in comparison to the unaffected side, the pain status (numeric rating scale), the wound status and complications. Radiographic data was analyzed for tumor size, total bone length, distance of tumor from the growth plate, bone diameter distally and proximally of the tumor, graft length and diameter. Using this data the following ratios were calculated: (1) percentage of graft diameter (graft diameter divided by recipient site diameter × 100), (2) normalised tumour length in percent (length of tumor divided by total bone length × 100) (Table 2).

Preoperative tumor assessment was done by radiographic and MRI evaluation and biopsy and confirmed by postoperative histopathology of the resected specimen.

All patients/parents consented to the data being used for publication.

3. Results

Consolidation was found in five of the six cases, namely all three humeral- and both tibial shaft reconstructions. Average time of healing in these cases was 5.3 months (range: 3.4–10.2 months). There were no intra- or postoperative complications. No complications (ankle or knee instability, peroneal nerve palsy, weakness of the long toe extensors or extensors) at the donor site were encountered. In the case of a metaphyseal ABC of the tibia, consolidation was only found at the distal but not at the proximal tumor resection site. Healing was eventually achieved after additional bone grafting and application of an angular stable plate (proximal tibial LCP plate, Synthes, France). In no case, graft atrophy or immediate remodelling was observed. On the contrary, the transplanted fibula graft was clearly visible throughout the consolidation process (Figs 1 and 2). Newly formed bone was taking up the space between the graft and the remaining periosteal sleeve, resulting in equal diameter of the bone before and after reconstruction. Full fibula regeneration was encountered in 5 cases after an average of 3.9 months (range: 2.9–5.4 months) (Fig. 3). In one case, we observed only partial fibula reconstruction. Equal limb length on both sides was found in the five cases in which the growth plate was preserved. In the patient who underwent resection of the metaphyseal ABC bordering, the growth plate length difference was negligible as the growth potential at the time of surgery was minimal.

Analysis of the radiographic data showed no correlation between the normalised tumour length and the observed healing time. A trend towards quicker consolidation was however noticed in the upper extremity. Diameter ratios of the resection surface of the fibula graft and the recipient bone were calculated and averaged for the five healed cases at 50% proximally and 59% distally. A correlation between consolidation time and diameter was found, indicating that bigger diameter differences correspond to longer healing times. In the metaphyseal ABC case where a non-union developed at the proximal junction, the proximal diameter of the fibula graft was 30% of the tibia. Distally, where consolidation was encountered after a few months, the ratio was calculated as 61%.

Clinical evaluation at the time of consolidation showed no limitation of range of motion in the adjacent articulations and pain-free patients.

4. Discussion

Bone reconstruction of long bone defects can be a challenge. Free vascularised fibula grafts are promoted by some surgeons as the graft is quickly incorporated and remodels in the same fashion as normal bone [5,6,11,19,20]. The Capanna technique has also been shown to be effective in adult and pediatric patients undergoing tumor resection in the lower extremity [7,21]. However, these techniques are demanding, time consuming and not applicable in every clinical setting. Simple bone allografts and the Masquelet’s induced membrane technique provide a reliable and simple way of bone reconstruction. Although the Masquelet’s technique is associated with an elevated non-union rate and graft resorption [9,22–24],
Table 1

<table>
<thead>
<tr>
<th>Patient</th>
<th>Tumor</th>
<th>Bone</th>
<th>Location</th>
<th>Side</th>
<th>Fracture</th>
<th>Age at surgery, years</th>
<th>Stabilisation</th>
<th>Time to consolidation tumor site in months</th>
<th>Time to full fibula regeneration in months</th>
<th>Complication</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>ABC</td>
<td>Humerus</td>
<td>Diaphysis</td>
<td>Right</td>
<td>Yes</td>
<td>13</td>
<td>FIN</td>
<td>3.4</td>
<td>3.4</td>
<td>No</td>
</tr>
<tr>
<td>2</td>
<td>ABC</td>
<td>Humerus</td>
<td>Diaphysis</td>
<td>Left</td>
<td>Yes</td>
<td>15</td>
<td>FIN</td>
<td>3.7</td>
<td>3.7</td>
<td>No</td>
</tr>
<tr>
<td>3</td>
<td>ABC</td>
<td>Humerus</td>
<td>Metaphysis</td>
<td>Right</td>
<td>No</td>
<td>14</td>
<td>TSF, proximal tibial locking plate and bone grafting</td>
<td>64.6</td>
<td>Only partial consolidation</td>
<td>Pseudarthrose proximal junction</td>
</tr>
<tr>
<td>4</td>
<td>ABC</td>
<td>Tibia</td>
<td>Metaphysis</td>
<td>Left</td>
<td>No</td>
<td>7</td>
<td>LCP plate</td>
<td>10.2</td>
<td>3</td>
<td>No</td>
</tr>
<tr>
<td>5</td>
<td>Fibreuse dysplasia</td>
<td>Tibia</td>
<td>Diaphysis</td>
<td>Left</td>
<td>Yes</td>
<td>12</td>
<td>LCP plate</td>
<td>5.4</td>
<td>5.4</td>
<td>No</td>
</tr>
<tr>
<td>6</td>
<td>Fibreuse dysplasia</td>
<td>Tibia</td>
<td>Diaphysis</td>
<td>Right</td>
<td>No</td>
<td>12</td>
<td>LCP plate</td>
<td>5.4</td>
<td>5.4</td>
<td>No</td>
</tr>
<tr>
<td>Mean</td>
<td></td>
<td></td>
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<td></td>
<td>12.7</td>
<td></td>
<td>5.38*</td>
<td>3.9*</td>
<td></td>
</tr>
</tbody>
</table>

* Without case 4.

Fig. 1. Patient 6: a and b: preoperative magnetic resonance imaging showing the tibial fibrous dysplasia (asterisks); c and d: anterior-posterior radiograph 1 week (c) and 5 months (d) postoperatively. The fibula graft (arrows) remains clearly visible throughout the consolidation process.

it remains one of the reference techniques in bone reconstruction after extensive traumatic bone loss or in bone defects due to post-traumatic osteomyelitis [25,26]. Recent papers have discussed the application of this technique in pediatric long bone reconstruction after trauma and malignant tumor resection with promising results [9,27,28]. One of the relative disadvantages of the Masquelet’s technique in the treatment of benign lesions is the requirement of two surgeries and thus a prolonged treatment period.

Non-vascularised fibula grafts are good structural grafts and relatively easy to harvest. Donor site morbidity, especially in children...
### Table 2
Radiological measurements and calculated ratios.

<table>
<thead>
<tr>
<th>Patient</th>
<th>Tumor length times width (cm)</th>
<th>Total bone length (cm)</th>
<th>Relative tumor length (%)</th>
<th>Distance tumor to proximal physis (cm)</th>
<th>Resection length (cm)</th>
<th>Graft length (cm)</th>
<th>Graft diameter in % at proximal graft recipient junction</th>
<th>Graft diameter in % at distal graft recipient junction</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>10 × 2.5</td>
<td>32.3</td>
<td>31.3</td>
<td>7.8</td>
<td>12.7</td>
<td>13.7</td>
<td>52</td>
<td>45</td>
</tr>
<tr>
<td>2</td>
<td>12.5 × 2.5</td>
<td>36.5</td>
<td>34.2</td>
<td>10.1</td>
<td>14.5</td>
<td>18</td>
<td>70</td>
<td>64</td>
</tr>
<tr>
<td>3</td>
<td>9 × 3.2</td>
<td>36</td>
<td>25</td>
<td>0.8</td>
<td>8.7</td>
<td>11.6</td>
<td>39</td>
<td>68</td>
</tr>
<tr>
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<td>11.4 × 2.5</td>
<td>39</td>
<td>38.5</td>
<td>0</td>
<td>17.9</td>
<td>21.3</td>
<td>30</td>
<td>61</td>
</tr>
<tr>
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<td>6.3 × 3.2</td>
<td>30.8</td>
<td>20.5</td>
<td>7.74</td>
<td>12.9</td>
<td>14.7</td>
<td>38</td>
<td>54</td>
</tr>
<tr>
<td>6</td>
<td>10.1 × 2.8</td>
<td>43</td>
<td>23.3</td>
<td>12.9</td>
<td>18.6</td>
<td>24.6</td>
<td>51</td>
<td>65</td>
</tr>
<tr>
<td>Mean</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Mean= 28.8</td>
<td>Mean= 59</td>
</tr>
</tbody>
</table>

* Without case 4.

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**Fig. 2.** Patient 2: anteroposterior radiographs after resection of a diaphyseal ABC of the humerus: a: one day postoperative (the periostal sleeve shows cortical bone residues in the distal part); b: 2 weeks postoperative; c: 3 months postoperative; d: almost 6 months postoperative. There is progressive new bone formation in the periosteal sleeve. A small amount of bone allograft (Tutoplast® chips, 1–5 mm) were added to the periosteal sleeve. The fibula graft (arrows) does not change in size and is clearly visible throughout the consolidation process.
is low [16,18]. Large fibula grafts can be harvested as biomechanical studies have demonstrated that 10% of residual distal fibular length is sufficient to prevent ankle instability [29]. However, one of the downsides of non-vascularised fibula grafts is their diminished biological ability. Although in an experimental setting similar hypertrophy of non-vascularised and vascularised grafts was found [30] and confirmed in one clinical study [31], other studies have reported that graft hypertrophy is superior in vascularised grafts [32–34]. As a lack of graft hypertrophy leads to a thinner bone, stability is reduced and the graft is more susceptible to fatigue fractures [31,32]. Thus single non-vascularised fibula grafts are often considered unsuitable to bridge large bone defects in the lower extremity. To avoid these complications, we transferred a non-vascularised fibula strut into a periosteal sleeve, hypothesising that the periosteal sleeve would promote bone formation and graft integration. While no change in the size of the fibula graft could be seen, rapid new bone formation along the periosteum was noted leading not only to consolidation at the graft-recipient junction but also to a reconstitution of bone diameter as determined preoperatively. Similar to previous studies we found no correlation of graft length and healing time [31,35], however, the average time to healing using the above-described technique was slightly below those found in literature [31,35]. A recent publication by Grzegorzewski [4] concerning non-vascularised bone grafting of the humerus using the intraperiosteal technique reported similar healing times to ours indicating that the periosteal sleeve not only promotes reconstitution of bone size but also positively effects consolidation at the graft-recipient junction.

High failure rate due to non-union have been reported for single fibula grafts whereas double or triple grafts achieve union. One possible explanation might be the graft-recipient diameter difference. Analysing our data we determined a substantially bigger diameter ratio in our case of non-union than in the cases of union. Although there was some bone formation in the periosteal sleeve at the metaphyseal region, it did not suffice to result in consolidation. A contributing factor could also be the thickness of the periosteum in older children and the difficulty to separate the periosteal sleeve from the bone at the physis. However, as there was only one case our data is speculative and further cases are needed to either proof or discard this theory. In our study the method of stabilisation did not seem to have an effect on healing and we are not aware of reports showing different consolidation times with different stabilisation methods.

An interesting aspect we were not able to investigate due to low patient numbers was the influence of an intact open growth plate on the healing time and the integration of the fibula graft. As there is a high remodelling potential in immature bones it could be hypothesised that the fast integration and reconstitution of the bone diameter after intraperiosteal graft transfer is due to the growth plate’s activity.

Our results demonstrate the value of an intact periosteum for the reconstruction of bone defects in the pediatric population. Not only does healing time decrease but also new bone formation is found along the entire periosteum surrounding the graft resulting in high stability. The downside of this method is its limited application. In tumor surgery for example it is restricted to benign tumors as the periosteal sleeve needs to be resected in malignant neoplasms. While young children have a thick periosteal layer, it is relatively thin and frequently adherent to the bone in adults thus making subperiosteal resection a challenge. Furthermore, the reduced periosteal thickness and its bone adherence in the metaphyseal region can impede separating it from the underlying bone. Nevertheless, careful separation even if only partially possible does in our opinion promote bone formation.

The main limitation to our study is the low patient number, its retrospective character and its lack of a control group. Furthermore our study reports only on bone reconstruction of the humerus and the tibia. While we believe that this technique will also be applicable in the femoral bone of young children, we have no data to confirm or disproof our assumption. Although we obtained data showing a high reconstruction potential of this operative technique, further research is necessary to confirm our findings.

**Disclosure of interest**

The authors declare that they have no competing interest.

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