

Long-term Growth, Functional, and Aesthetic Outcomes after Fibula Free Flap Reconstruction for Mandibulectomy Performed in Children

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Background: The long-term outcome of mandible reconstruction when performed in children has not been well documented.

Methods: This is a retrospective case series of patients who underwent immediate oncologic mandible reconstruction with a fibula free flap at younger than 18 years of age over a 20-year period, by a single surgeon, who had long-term follow-up.

Results: A total of 10 patients met inclusion criteria. Patient age ranged from 3 years and 8 months to 17 years and 9 months (mean 11 years). The etiology was malignant tumors in seven patients and benign locally aggressive tumors in three patients. All fibula flaps survived. All donor sites were closed primarily. The mean follow-up duration was 10 years and 5 months (range 3–20 years). The mean patient age at follow-up was 21 years and 10 months (range 8 years and 9 months to 30 years and 9 months). All patients achieved a regular diet and normal speech. Final occlusion was normal in seven of 10 patients. The aesthetic outcome, as evaluated by clinical examination, was a symmetric mandible in eight patients (in the other two the aesthetic asymmetry and malocclusion was minor and did not require operative intervention). Dental implants were ultimately placed in three patients. Leg function was normal in eight patients. Achilles lengthening and tendon transfer was required in one patient, and one patient developed ankle pain associated with running.

Conclusion: Mandible reconstruction in children with the fibula free flap provides excellent aesthetic and functional outcomes that are durable over time. (*Plast Reconstr Surg Glob Open 2022;10:e4449; doi: 10.1097/GOX.00000000004449; Published online 28 July 2022.*)

INTRODUCTION

Mandibular tumors are rare in the pediatric population.¹ Tumor ablation creates a bony defect (with or without additional soft-tissue deficiency) that is ideally reconstructed in the immediate setting. Reconstruction is challenging because of the small size of facial structures resulting in a tight working space, limited donor sites and small vessels that are prone to spasm.² However, success

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Copyright © 2022 The Authors. Published by Wolters Kluwer Health, Inc. on behalf of The American Society of Plastic Surgeons. This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal. DOI: 10.1097/GOX.00000000004449 rates of pediatric free tissue transfer are comparable to, or even better than, those in adults due to fewer comorbidities, healthy vessels, absence of smoking-related vascular damage and superior ability to heal.³

Mandible reconstruction in skeletally immature patients has an additional variable to consider: growth of the jaw. The residual growth potential has important implications for the final aesthetic and functional result. A good immediate outcome may not remain after either continued jaw growth or impairment of normal growth. As a result, some authors do not advocate for immediate reconstruction of the mandible in pediatric patients.⁴ Furthermore, the consequences of bone and soft-tissue harvest from the growing leg need to be carefully considered. There are very few reports on the long-term outcomes of mandible reconstruction in patients who have not completed skeletal growth.^{5–7} The current report

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Related Digital Media are available in the full-text version of the article on www.PRSGlobalOpen.com. intends to shed light on this important topic by presenting a longitudinal experience with management of this patient population.

MATERIALS AND METHODS

This is a retrospective case series of patients who underwent immediate oncologic mandible reconstruction with a fibula free flap at younger than 18 years of age at the Memorial Sloan Kettering Cancer Center from January 1993 to December 2012 by the senior author (P.C.). The study period was chosen so that sufficiently long follow-up could be obtained to assess for long-term outcomes. Data about patient demographics, tumor pathology, adjuvant therapies, extent of mandibular resection, details of reconstruction, and postoperative course were obtained from patient charts. Assessment of aesthetics, speech, and occlusion were obtained by in-person evaluations by the treating physicians. Orthopantomograms were also reviewed.

The fibula free flap is our method of choice for mandibular reconstruction. The technique has been well described.⁸ The flexor hallucis longus muscle is included in the flap for filling of the dead space in the submental area that is created after neck dissection. Rigid fixation of osteotomized segments is performed with miniplates on the anterior and inferior surfaces of the fibula to provide stability in two planes. Miniplates are easier to contour, may have less restrictive effects on mandibular growth, and are easier to remove for dental implant placement. Defects are classified using our previously published classification scheme into anterior, hemimandible, and lateral defects (Fig. 1).⁹ Anterior defects include the symphysis; hemimandible defects include the body, angle and ascending ramus; and lateral defects include one or two of the body, angle or ascending ramus segments.

Dental implants are placed once the flap has healed and all adjuvant therapies are completed. The miniplates are removed to facilitate unhindered implant placement.

Takeaways

Question: What are the long-term outcomes of mandible reconstruction with fibula free flap in skeletally immature patients?

Findings: This is a retrospective case series of 10 patients. At long-term follow-up, patients were found to have good occlusion, normal oral intake and excellent facial symmetry.

Meaning: Mandible reconstruction with fibula free flap in skeletally immature patients has very good long-term aesthetic and functional outcomes.

RESULTS

A total of 10 patients met inclusion criteria over the 20-year study period. Table 1 provides details about patient demographics, mandibular pathology, neoadjuvant and adjuvant therapy, type and extent of resection, and reconstruction. Patient age at surgery ranged from 3 years and 8 months to 17 years and 9 months (mean 11 years). Tumor pathology was benign in three patients and malignant in seven patients. The mandibular defect was a hemimandible in four patients, lateral mandible in three patients, anterior mandible in two patients and subtotal mandible in one patient. The ascending ramus was resected in five patients due to tumor involvement; in four of these patients, the condylar segment was taken off the specimen ex vivo and placed on the fibula as an autograft. (See figure, Supplemental Digital Content 1, which displays patient #7, who underwent subtotal mandibular reconstruction at the age of 16 years. A, Specimen comprising the left hemimandible and extending to the right angle. B, Fibula flap shaped and plated. The condylar segment has been autotransplanted. C, Follow-up at 3 years demonstrating a symmetric mandible. D, Orthopantomogram showing successful dental implant placement. http://links.lww.com/PRSGO/C117.)

In one patient, the condyle was involved with the tumor and thus could not be autotransplanted; the

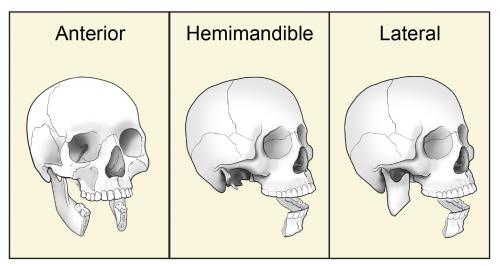


Fig. 1. Classification of mandible defects.9

		ζ		Neoadjuvant			Condyle	Condyle Recon-	Recipient	Recipient
rauent	Age	Gender	Diagnosis	ınerapy	Adjuvant 1 nerapy	Derect	Removed	suruction	Artery	vein
1	5 y 3 mo	н	Ewing's Sarcoma	Chemotherapy	Chemotherapy	Hemimandible (right canine to	Yes	Autograft	Superior	Facial
5	17 y 5 mo	Ы	Cement-ossifying	Ι	I	subcondylar) Lateral (left angle to midline)	No	I	thyroid Lingual	Internal
60	12 y 2 mo	Μ	nbroma Fibrous dysplasia	Ι	Ι	Anterior (right first molarto left	No	I	Facial	Jugular Facial
4	9 y	Г	Ewing's Sarcoma	Chemotherapy	Chemotherapy	second molar) Hemimandible (left canine to	Yes	Autograft	Lingual	Facial
10	9 y 2 mo	Ч	Metastatic neuroblastoma	Chemotherapy and immunotherapy	Chemotherapy and immuno-	Hemimandible (right canine to condyle)	Yes	Burred fibula	Superior thyroid	Internal jugular
9	9 y 8 mo	Μ	Ewing's Sarcoma	and radiation Chemotherapy	therapy Chemotherapy	Lateral (left first premolar to left	No	I	Lingual	Facial
7	$16 \mathrm{y} 10 \mathrm{mo}$	Μ	Chondrosarcomatous Chemotherapy	Chemotherapy	Chemotherapy	angle) Subtotal (right condyle to left	Yes	Autograft	Superior	External
86	$\stackrel{3}{_{2}}$ y 8 mo 17 y 9 mo	MM	osteogenic sarcoma Desmoplastic fibroma Osteogenic sarcoma	 Chemotherapy	 Chemotherapy	angle) Lateral (right canine to angle) Hemimandible (left central	No Yes	Autograft	(Unknown) Superior	Jugular (Unknown) Internal ·
10	12 y 11 mo	Μ	Ameloblastic fibrosarcoma	I	and radiation Radiation	incisor to right subcondylar) Anterior (left angle to right second molar)	No	I	tnyroid Facial	Jugular Facial

Table 1. Demographics and Surgical Information

end of the fibula was burred to a round shape to form the neo-condyle. Osteomyocutaneous flaps were used in seven patients and myo-osseous flaps in three patients. The width of the skin paddle ranged from 2 to 4 cm (mean 3 cm). All donor sites were closed primarily. Microvascular arterial anastomosis was performed to the superior thyroid artery in four patients, lingual artery in three patients and facial artery in two patients. A single venous anastomosis was performed to the common facial vein (end-toend) in five patients, external jugular vein (end-to-end) in one patient, and internal jugular vein (end to side) in three patients. One patient did not have microsurgical information in the medical record.

The mean follow-up duration was 10 years and 5 months (range 3-20 years) (Table 2). The mean patient age at follow-up was 21 years and 10 months (range 8 years and 9 months to 30 years and 9 months). There was no change in speech quality in any of the patients. All patients eventually resumed a regular diet. Final occlusion was normal in seven patients. Patient #4, who underwent hemimandible reconstruction at the age of 9 years, developed a slight lateral shift of the mandible to the reconstructed side on jaw opening. (See figure, Supplemental Digital Content 2, which displays patient #4, who underwent reconstruction at the age of 9 years. A, Seven years after reconstruction, showing a symmetric mandible, but soft-tissue hollowing of the lateral lower face due to extensive soft-tissue resection. B, Jaw opening resulted in a slight drift of the mandible to the reconstructed side on the left with clicking. She also had a marginal mandibular nerve palsy. C, Orthopantomogram demonstrating insufficient bone stock for dental implant placement. http:// links.lww.com/PRSGO/C118.)

Patient #6, who underwent lateral mandible reconstruction at age 9 years 8 months, developed a slight anterior open bite. (See figure, Supplemental Digital Content 3, which displays patient #6, who underwent reconstruction at the age of 10 years. A, 5.5 years after reconstruction demonstrating a symmetric mandible. B, Occlusion demonstrates an open bite. C, Orthopantomogram demonstrates well healed bone segments. http://links.lww.com/PRSGO/C119.)

Patient #8, who underwent lateral mandible reconstruction at 3 years of age, developed an occlusal cant due to nongrowth of the reconstructed mandible which caused growth restriction of the maxilla. Marginal mandibular nerve palsy was observed in four patients; in one of those patients, the nerve was deliberately divided as it was a part of the tumor specimen. The aesthetic outcome was assessed to be a symmetric mandible in eight patients (Fig. 2). Patient #1, who underwent hemimandible reconstruction at age 5 years and 3 months, developed chin point deviation due to nongrowth of the reconstructed lateral segment. Patient #8, who underwent hemimandible reconstruction at age 3 years and 8 months, developed an upward occlusal cant due to suboptimal growth of the reconstructed mandible.

Hardware removal was performed in seven patients at 3–20 months after reconstruction (mean 10 months). This was done to facilitate dental implant placement, as fixation screws can be in the way of implants. Implants

					Recipient Site	t Site			Donor Site	
Patient		Follow-up Age at Last Duration Follow-up	Aesthetics/ Symmetry	Occlusion	Speech	Diet	Dental Implants	Marginal Mandibular Nerve Palsy	Leg Function	Secondary Operations
-	10 y 6 mo	15 y 9 mo	Slight asymmetry	Normal	Perfect	Regular Yes	Yes	Yes	Normal	Hardware removal for
00 10	$\frac{10 \text{ y } 10 \text{ mo}}{18 \text{ y } 7 \text{ mo}}$	$ \begin{array}{c} 10 \text{ y } 10 \text{ mo } 28 \text{ y } 3 \text{ mo} \\ 18 \text{ y } 7 \text{ mo } 30 \text{ y } 9 \text{ mo} \end{array} $	or cum pourt Symmetric Symmetric	Normal Normal	Perfect Perfect	Regular No Regular Yes	No Yes	No No	Normal Ankle pain when	Hardware removal for dental
4	6 y 10 mo	6 y 10 mo 15 y 10 mo Symmetric	Symmetric	Lateral jaw drift on mouth opening, Popping sound in	Perfect	Regular	Perfect Regular No (thin bone stock)	Yes	running Achilles contracture requiring length- ening and partial	implant placement Hardware removal for dental implant placement
ы	20 y	29 y 2 mo	Symmetric	right TMJ Normal	Perfect	Regular	Perfect Regular No (wearing removable	No	AT transfer Normal	Hardware removal for dental implant placement
9	6 y 5 mo	16 y 1 mo	Symmetric	Anterior open bite	di Perfect Regular No	Regular	dentures) No	No	Normal	Hardware removal for dental
1	3 y	19 y 10 mo	19 y 10 mo Symmetric	Normal	Perfect	Perfect Regular Yes	Yes	No	Normal	Implant placement -Re-exploration (negative) for flap congestion
×	5 y 1 mo	8 y 9 mo	Mild right lower	d maxillary	Perfect	Regular	No (thin bone	Perfect Regular No (thin bone Yes (nerve sacrificed Normal	Normal	-Hardware removal for dental implant placement Keloid excision from neck
6	$10 \mathrm{~y} \mathrm{~3~mo}$	28 y	tace hypoplasia Symmetric	cant Normal	Perfect	st Perfect Regular No	stock) No	during surgery) Yes	Normal	and leg Hardware removal for dental
10	13 y	25 y 11 mo	Symmetric	Normal	Perfect	Regular No	No	No	Normal	implant placement Tracheostomy scar revision

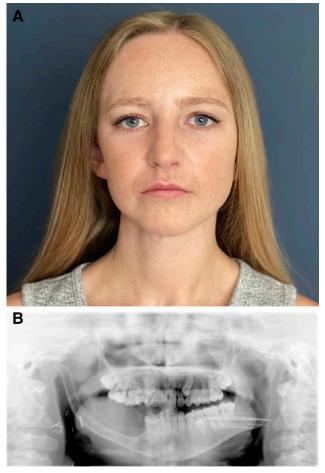


Fig. 2. Patient #5 underwent reconstruction at the age of 9 years. A, Twenty years after reconstruction, showing a symmetric mandible. B, Orthopantomogram obtained 3 years postoperative demonstrates well healed bone segments. The end of the fibula was burred and has remodeled into a neo-condyle.

were eventually placed in three patients (Fig. 3). The bone stock was deemed inadequate for implant placement by the dental service in two patients. Removable partial dentures were used in one patient. Five patients did not undergo dental implant placement due to either personal choice or lack of financial resources. One patient underwent keloid excision from the neck and leg and one patient underwent tracheostomy scar revision. Leg function was reported to be normal in eight patients. Patient #4 developed an Achilles tendon contracture for which she underwent Achilles tendon lengthening and partial tibialis anterior tendon transfer. Patient #3 reported ankle pain while running but not with walking.

DISCUSSION

Mandible reconstruction with the free fibula flap was first described by Hidalgo⁸ at Memorial Sloan Kettering Cancer Center in 1989. Since then, the procedure has undergone several refinements including virtual surgical planning and immediate dental rehabilitation.^{10,11} It is now the most popular technique for microvascular mandible reconstruction.¹² The consistently good outcomes reported in the literature are due to the many anatomic features of the fibula bone that are conducive to mandible reconstruction.¹³ The fibula is a long segment of cortical bone that can withstand multiple osteotomies, allows for osseointegration, and permits skin and muscle harvesting, with acceptable donor site morbidity.

Microsurgical head and neck reconstruction in pediatric patients may be challenging due to small vessel size, limited working space, and spasm-prone arteries. Mandible reconstruction with free fibula flap in children has been shown to have success rates comparable to the adult population.^{7,14,15} However, there are very few reports of long-term functional and aesthetic outcomes.⁵⁻⁷ Most of the concerns with pediatric jaw reconstruction arise from continued growth of the facial skeleton. The majority of jaw growth is completed by the age of 16 years in girls and 18 years in boys, with a small amount of growth continuing till 20 years of age.¹⁶ Since the bone flap itself does not intrinsically grow, a continued development of the jaw relies on the growth of the remaining native mandible and stress remodeling of the fibula.¹⁷⁻¹⁹ Transfer of proximal fibular epiphyseal growth plate has been described for preserving growth potential in long bone reconstruction, but there is a paucity of reports in mandible reconstruction.^{20,21} Resection of the condyle, radiation, and age younger than 8 years at surgery are factors that have been shown to adversely affect growth potential of the residual mandible.²² In our series, eight of 10 patients had a symmetric mandible on long-term follow-up as judged by clinical examination, and seven of 10 patients had perfect occlusion. Objective measurement of mandibular growth is very challenging. Orthopantomograms have some degree of distortion depending on how the image is taken, and therefore, studies cannot be compared 1:1. Our evaluations are thus based on clinical examinations performed by the plastic surgeon, otolaryngologist, and dentist. In patients who did not have a perfect result, the degree of jaw asymmetry and malocclusion was minor enough that patients did not wish to have surgical correction. In our series, the patients who had asymmetry or malocclusion tended to be younger (<10 years at surgery) and had lateral or hemimandible defects. Radiation did not affect growth in our patients, although the number is too small to draw definitive conclusions.

Dental rehabilitation is an important component of a functional mandible reconstruction. It enables optimal mastication, facilitates speech, and improves oral competence. Dental rehabilitation can be performed with removable partial dentures or osseointegrated implant borne prostheses. Osseointegrated implants have several advantages. Implants load the fibula, which may improve bone strength and minimize bone resorption.^{25–27} They also ensure better compliance, as patients do not have to apply their prosthesis every day. Dental rehabilitation also restores more normal occlusal relationships, which may promote more normal midface growth. On the other hand, patients need frequent follow-up for prosthesis adjustments so that as the mandible grows, the prosthesis can be modified for changing jaw dimensions and to

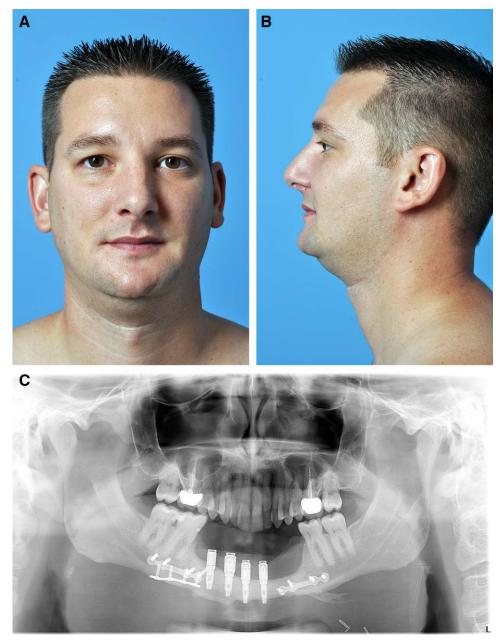


Fig. 3. Patient #3 underwent reconstruction at the age of 12 years. A, Eighteen years after reconstruction, showing a symmetric mandible with good chin projection, frontal view. B, Lateral view. C, Orthopantomogram demonstrates successful dental implant placement.

ensure that growth of the native mandible is not restricted. One of the challenges with dental rehabilitation in very young patients is that the fibula is thin and may not have sufficient height for dental implant placement. Another issue is that dental rehabilitation is frequently not covered by insurance and thus some patients do not have the requisite financial resources. In our series, three patients underwent dental implant placement, five patients were not interested or were unable to afford implants, and two patients had insufficient bone height for implants. We now advocate for immediate placement of osseointegrated dental implants when possible and uncover the implants 4–6 weeks later for dental prosthesis placement.¹¹ This results in earlier loading of bone, avoids an additional operation, and obviates surgery on radiated tissue.

Fibula harvest is well tolerated in terms of donor site morbidity. The incidence of long-term complications is low and include chronic pain, limited range of motion, reduced strength, ankle instability, altered gait, claw toe, and sensory deficits.²⁸ The distal fibula provides stability to the ankle joint. One of the major concerns about harvest of fibula flaps in growing individuals is the development of valgus deformity of the ankle.²⁹ The primary etiology of this is proximal migration of the residual

distal fibula which causes lateral shift of the talus resulting in stress on the lateral tibial growth plate.³⁰ As a result, growth occurs asymmetrically, resulting in ankle valgus. In adults, it has been shown that preservation of 6 cm of the distal fibula is important to preserve ankle stability.³¹ Another study showed that the distal fibular stump should be at least 10% of the total fibular length for ankle stability.³² We do not leave a predetermined length of fibula bone but rather preserve the distal tibiofibular syndesmosis. The syndesmotic ligament is thicker than the interosseous membrane, and their transition point can be palpated. The distal fibular osteotomy is made above this ligament. We do not perform primary ankle stabilization. Patients are followed up on a regular basis, and if there are any signs of ankle deformity or instability, ankle X-rays are obtained (anteroposterior, lateral, and mortise views). If these demonstrate developing valgus deformity, ankle stabilization is performed via a quadricortical syndesmotic screw. None of our patients developed ankle instability or valgus deformity, which we attribute to paying meticulous attention to preserving the distal tibiofibular syndesmotic ligament.

This study demonstrates high success rates of mandible reconstruction with fibula free flaps in children with functional results that are maintained in the long term. We strongly advocate free flap reconstruction for oncologic mandibular defects in the pediatric population since there is typically a long segment bone deficit, significant soft tissue that needs to be replaced, and often, patients receive neoadjuvant or adjuvant therapy which can create unfavorable healing conditions. The major strength of this study is the long follow-up on a cohort of patients who have not yet obtained complete skeletal maturity. The main weaknesses are a small sample size and assessments via clinical examinations rather than strictly objective methods. Furthermore, patient-reported outcomes have not been obtained. Future research is needed to objectively quantify the factors that influence jaw growth after free flap mandible reconstruction in the pediatric population.

CONCLUSION

The free fibula osteocutaneous flap is an excellent option for oncologic mandible reconstruction in children. The donor site morbidity is well tolerated, and the aesthetic and functional results are durable over time.

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