Case Report

Local control of pediatric head and neck sarcoma with free flap reconstruction obviating the need for radiotherapy: a case report

Kolapo Dairo[1]^, Ryan Bishop[1]^, Rishabh Sethia[2,3]^, Matthew Old[2], Kris R. Jatana[2,3], Charles A. Elmaraghy[2,3]

[1]College of Medicine, The Ohio State University, Columbus, OH, USA; [2]Department of Otolaryngology, The Ohio State University Wexner Medical Center, Columbus, OH, USA; [3]Department of Pediatric Otolaryngology, Nationwide Children's Hospital, Columbus, OH, USA

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Correspondence to: Charles A. Elmaraghy, MD. Department of Otolaryngology, The Ohio State University Wexner Medical Center, 410 W. 10th Ave., Columbus, OH 43210, USA; Department of Pediatric Otolaryngology, Nationwide Children's Hospital, 700 Children's Drive, Columbus, OH 43205, USA. Email: charles.elmaraghy@nationwidechildrens.org.

Background: Microvascular free-tissue transfer is a widely used technique for surgical site reconstruction following head and neck mass resection. While it is commonly used in adults, the rarity of head and neck cancers in children makes free flap reconstruction relatively rare in this population. Free flap reconstruction allows for coverage of large defects which may result from wide resections performed to avoid exposing pediatric patients to primary radiotherapy.

Case Description: We present two pediatric oncologic cases using free flap reconstruction. The first, a 4-year-old male, presented with a rapidly enlarging tongue lesion that was diagnosed as a biphasic synovial sarcoma. The mass was resected, and a radial forearm free flap was placed. The second, a 9-year-old male, presented with jaw pain and a mandibular mass. Biopsy confirmed a diagnosis of Ewing sarcoma. The patient was treated initially with chemotherapy, followed by surgical resection and placement of an osteocutaneous fibular free flap. Both patients are disease-free 5 years post-operation.

Conclusions: In each patient, complete resection of the primary tumor was performed eliminating the need for radiotherapy with utilization of free flap reconstruction. As advances in medicine enable better evaluation of clear margins resulting in larger resection defects, free tissue transfer provides a useful reconstructive option in the pediatric population.

Keywords: Free flap; radiotherapy; synovial sarcoma; Ewing sarcoma; case report

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Introduction

Microvascular free-tissue transfer is a commonly utilized reconstructive technique following resection of head and neck masses in adults. Its indications include composite oral cavity defects, and extensive skull base and scalp defects, among others. However, the use of free flap reconstruction remains rare in children due to the low incidence of pediatric head and neck malignancies as well as surgeon hesitancy regarding the smaller anatomic structure and the necessity for growth at the recipient and donor sites. In response, there have been numerous case reports in the literature that demonstrate the safety and efficacy of pediatric free flaps, including those utilized in the head and neck region (*Table 1*).

^ ORCID: Kolapo Dairo, 0000-0003-2708-4884; Ryan Bishop, 0000-0003-0747-2652; Rishabh Sethia, 0000-0003-0425-7682.

Pediatric head and neck malignancies have traditionally been managed with radiation with or without chemotherapy, while surgical resection has typically been reserved for smaller lesions in less anatomically complex regions. When deciding between primary radiotherapy and surgery for local control, bulkier and more infiltrating tumors have traditionally been managed with radiation due to significant concerns regarding plausible reconstruction at the affected site and subsequent asymmetry of the face as the child ages (13). Until the recent utilization of free flaps for pediatric oncologic needs, for lack of a better alternative, the choice of primary radiotherapy came with a high likelihood of subjecting children to the array of associated adverse effects (14). Thus, as the literature grows and as surgeons become more adept at performing free tissue transfer, the management of major pediatric head and neck tumors may evolve to include this promising reconstruction option.

In this article, we describe the successful management of head and neck sarcoma in two pediatric patients with surgical resection and free flap reconstruction, thereby avoiding the short and long-term complications of radiation therapy. We present this article in accordance with the CARE reporting checklist (available at https://tp.amegroups.com/article/view/10.21037/tp-24-4/rc).

Case presentation

Case 1

The first patient, a 4-year-old male, presented with a rapidly enlarging tongue lesion with associated bleeding. Ultrasound evaluation revealed a heterogenous solid mass with features concerning for malignancy and subsequent biopsy confirmed a diagnosis of biphasic synovial sarcoma of the lateral tongue.

Tracheostomy was performed, followed by partial anterior glossectomy, resection of the floor of the mouth, neck dissection, retropharyngeal lymph node dissection, and radial forearm free flap placement. Resection began with identification of the lesion, a 2 cm × 2 cm area of induration on the right anterior margin of the tongue, followed by circumferential incision with 1–2 cm margins. Attention was turned to the floor of the mouth, beginning with identification and ligation of the right lingual nerve and submandibular duct. The glenoid tubercle was identified and the muscular attachments of the tongue were divided in order to release the tongue and improve exposure. Dissection continued toward the left aspect of the tongue, and the left lingual nerve was identified and ligated. The incision was extended posteriorly to free the remaining intrinsic muscles of the tongue and genioglossus from their attachments, and the specimen was removed *en bloc*. Lymph node dissection, including the bilateral neck and right retropharyngeal nodes, was performed while a second otolaryngologist began harvesting the left radial forearm free flap.

Reconstruction began with evaluation of the defect and formation of a template, which was subsequently utilized to assist in the donor site harvest from the forearm overlying the radial artery. The radial forearm flap was harvested in standard fashion. The free flap was inset via circumferential sutures to the remaining tongue and floor of the mouth. Microvascular anastomosis was accomplished via 9-0 nylon suture for the radial artery—facial artery anastomosis and via a 2.5 coupler for the cephalic vein—facial vein anastomosis. Adequate perfusion of the flap was then confirmed using doppler. The forearm donor site was closed using a 7.5 cm × 7.5 cm split-thickness skin graft harvested from the left thigh and the patient was transferred to the PICU in stable condition.

The patient had a satisfactory post-operative recovery, and he received post-operative chemotherapy. Prior to discharge from the hospital, a port was placed for chemotherapy infusion and the patient subsequently developed a methicillin-susceptible *Staphylococcus aureus* (MSSA) bloodstream infection which was successfully treated with vancomycin and cefazolin. His tracheostomy was decannulated on post-operative day 16. His nasogastric tube

Highlight box

Key findings
- Microvascular free-tissue transfer is a versatile option for head and neck malignancies in children with high success rates.

What is known and what is new?
- Surgical resection with free flap reconstruction and radiotherapy are both utilized in the management of pediatric head and neck masses, but radiotherapy is known to have many adverse effects.
- We add two cases to a literature review of free flap reconstruction in children, displaying the decreased incidence of adverse outcomes when compared to radiotherapy.

What is the implication, and what should change now?
- Surgical resection with free flap reconstruction is a successful alternative to primary radiotherapy in the treatment of head and neck malignancies and should be strongly considered by physicians when evaluating pediatric patients.

Table 1 Literature review of pediatric free flap case reports/series

Source	Age	Flap utilized	Comorbid/predisposing condition	Complications
Ferri *et al.* (1)	13 years	Anterolateral thigh flap	Severe trismus	None
	15 years	Anterolateral thigh flap	Severe trismus	None
Crawley *et al.* (2)	9 years	Parascapular fasciocutaneous flap	Desmoid tumor resection leading to scarring and fibrosis	None
Bedogni *et al.* (3)	12 years	Fibular osteomuscular bone flap	Central giant cell granuloma, Evan syndrome	None
Weizman *et al.* (4)	13 years	Anterolateral thigh flap	Rhabdomyosarcoma	None
	14 years	Rectus abdominis flap	Ewing's sarcoma	Not reported
	17 years	Rectus abdominis myocutaneous flap	Osteosarcoma	Revision of venous anastomosis
	2 years	Gracilis myocutaneous flap	Rhabdomyosarcoma	Not reported
	4 years	Rectus abdominis flap	Rhabdomyosarcoma	Not reported
	14 years	Fibula osteocutaneous flap	Rhabdomyosarcoma	Not reported
	16 years	Gracilis myocutaneous flap	Rhabdomyosarcoma	Not reported
	18 years	Gracilis myocutaneous flap	Osteosarcoma	Not reported
	15 years	Anterolateral thigh flap	Large contracting scar	None
Faria *et al.* (5)	8 months	Fibular osteocutaneous flap	Melanotic neuroectodermal tumor	Slight valgus deformity of donor ankle
Piette *et al.* (6)	6 years	Latissimus dorsi	Rhabdomyosarcoma	None
Guo *et al.* (7)	7 months	Fibular flap	Germ cell tumor of the mandible	None
	15 years	Fibular flap	Hemifacial microsomia	None
	14 months	Fibular flap	Rhabdomyosarcoma	Bone formation over the reconstruction plate
	6 years	Fibular flap	Ewing's Sarcoma	Failed growth of fibular bone graft
Rashid *et al.* (8)	14 years	Fibular flap	Aneurysmal bone cyst	None
	13 years	Fibular flap	Giant cell granuloma	None
Gharb *et al.* (9)	3 years	Anterolateral thigh flap	Giant hairy nevus involving cheek, ear and neck	None
	6 years	Anterolateral thigh flap	Cheek noma (orofacial gangrene)	None
	8 years	Anterolateral thigh flap	Exposed hardware mandible	None
	11 years	Anterolateral thigh flap	Ossifying fibroma of maxilla	None
	15 years	Anterolateral thigh flap	Large contracting scar	None
Iida *et al.* (10)	14 months	Rectus abdominis myocutaneous flap	Malignant rhabdoid tumor	None
Nayak *et al.* (11)	7 years	Anterolateral thigh flap	Epithelioid sarcoma	None
Sadiq *et al.* (12)	21 months	Anterolateral thigh flap	Intraoral caustic burn	None
	3 years	Radial forearm flap	Intraoral caustic burn	None

was removed on post-operative day 17 and he subsequently resumed a regular diet before being discharged to home. At 12 years after surgery, the patient remains disease free.

Case 2

The second patient, a 9-year-old male, presented with complaints of right-sided jaw pain, trismus, and localized swelling. A computed tomography (CT) scan showed a right mandibular mass concerning for malignancy and subsequent biopsy confirmed a diagnosis of Ewing sarcoma. Further workup was negative for metastatic disease and the patient completed 12 weeks of neoadjuvant chemotherapy with vincristine, doxorubicin, and cyclophosphamide. At that time, the decision was made to attempt local control with surgical resection followed by free flap reconstruction.

Resection was accomplished via right mandibulectomy, resection of the floor of the mouth, partial glossectomy, and modified radical dissection of the right neck. The operation began with cervical and pre-auricular incisions, and subplatysmal flaps were raised to allow for a radical neck dissection of levels 1–5. The parapharyngeal space and skull base were exposed, allowing for adequate visualization of the tumor. Composite resection of the mandible was then performed, which necessitated sacrifice of the right lingual nerve as it coursed directly through the tumor. Following resection of the mass, the remaining native mandible was fixated via maxillary and mandibular arch bars.

Reconstruction was accomplished via osteocutaneous right fibular free flap transfer with temporomandibular joint reconstruction. The right leg was chosen for free tissue transfer, and the fibular flap was harvested in standard fashion. The fibula was secured within the temporomandibular joint capsule using locking screws, with good bony contact at all edges. The pedicle was then brought down to the neck and the peroneal artery was anastomosed to the right superior thyroid artery using 9-0 nylon suture. End-to-end anastomosis was performed between the two accompanying peroneal veins and the common facial vein using vein couplers. Adequate perfusion to the flap was confirmed. Following closure of the donor site, the patient was transferred to the Pediatric Intensive Care Unit (PICU) in stable condition.

The patient's post-operative course was complicated by prolonged non-healing of the oral and neck wounds as well as a small orocutaneous fistula along the anterior aspect of the flap. The airway was initially managed via nasotracheal intubation and the patient was extubated on post-operative day 3. The presence of a fistula delayed resumption of oral intake, and the patient received nutrition via a nasogastric tube until post-operative day 17. He began a regular diet on post-operative day 19. A 6-week course of adjuvant chemotherapy was completed and the patient has remained disease free at 5 years after surgery.

Ethical considerations

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patients' parents or legal guardians for the publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Discussion

Considerable progress has been made in the field of surgical reconstruction with free tissue transfer, specifically in the management of pediatric malignancies of the head and neck. Historically, advanced head and neck malignancies in children were treated with primary radiotherapy for local control, since functionally and cosmetically safe surgical reconstruction options were limited. However, such reconstruction is now a viable option, as the utilization of free flap surgical techniques has allowed for larger and more complex resections. This, in certain cases, may obviate the need for radiation therapy and its associated adverse effects, including growth suppression, osteonecrosis of the irradiated site(s), and generation of second primary cancers. These effects are magnified in the pediatric population. These patients are more susceptible to the growth suppressive effects of radiation, and have a longer lifespan in which they may develop radiation-induced malignancies.

The facial skeleton is especially susceptible to radiation-induced growth suppression, as it has been shown that 93% of patients treated with mild to severe radiation will have resultant soft tissue or bone damage. In a study of 26 pediatric patients who underwent primary radiotherapy for rhabdomyosarcoma or nasopharyngeal carcinoma, the authors concluded that there were significant alterations in craniofacial skeletal measurements indicative of treatment-induced asymmetry and deformity (15). In a study of 17 patients who underwent primary radiotherapy for head and neck rhabdomyosarcoma, facial growth retardation

was reported in 11 (65%), neuroendocrine dysfunction in 9 (53%), dental abnormalities in 7 (41%), and hypothyroidism in 3 (17.7%) patients. All 17 patients displayed some late effect of the radiation, with one patient who was treated for rhabdomyosarcoma of the supraglottic larynx developing low-grade mucoepidermoid carcinoma of the parotid gland 20 years post-treatment (14).

Of particular relevance to this case series, is a study done on 266 patients with Ewing's sarcoma treated with primary radiotherapy. In this population, 16 (6%) had developed second malignancies at median follow-up of 9.5 years (16). Other possible adverse effects of head and neck radiotherapy include panhypopituitarism, behavioral and developmental abnormalities, and xerostomia, the latter of which may be permanent (17).

The two patients described in this series both presented with sarcomatous head and neck malignancies. For the patient in case 1 with biphasic synovial cell sarcoma, the acuity of the presentation and the progressive symptoms prompted surgical management. Furthermore, the preoperative imaging studies indicated that wide resection with overall preservation of function was likely. Thus, surgical excision with free flap reconstruction obviated the need for adjuvant radiotherapy, which is often considered for this disease process following inadequate pathologic margins (18). For the patient with Ewing sarcoma described in case 2, the decision to manage surgically was determined given the potential of achieving negative margins and sparing the need for radiation therapy. Ewing sarcoma of the head and neck is typically managed with neoadjuvant chemotherapy followed by local control with either surgical extirpation or radiation therapy, depending on response to treatment and extent of local disease (19). Radiation therapy is indicated for situations where surgery would be considered incomplete or would cause "intolerable morbidity". As demonstrated in case 2, the ability to achieve clear margins with impressive functional and cosmetic outcomes via free flap reconstruction may further decrease the number of head and neck tumors considered to be too large or complex for surgical resection.

Numerous case reports and case series have been published within the last 10–15 years that detail successful treatment of pediatric pathologies utilizing surgical resection with free flap reconstruction (*Table 1*). The vast majority of these reports detail uncomplicated postoperative courses with good functional and cosmetic recovery. Lech *et al.* [2024] report a success rate of 90% in 136 pediatric patients treated with free clap reconstruction for head and neck disorders, which approximated the 94% and higher success rate previously described in the literature (20). In a case series, Weizman *et al.* [2011] describe 8 pediatric patients with varying pathology who had defects reconstructed with free flaps. One specific patient was linked with a venous thrombosis event requiring revision of the anastomosis. Other complications mentioned included singular cases of transient flap congestion requiring decompression, minor wound breakdown, and wound infection, although these were not ascribed to a specific patient(s) (4). A case report by Faria *et al.* [2014] details the development of a valgus deformity at the ankle of the donor leg for a fibular osteocutaneous flap approximately 6 years postoperatively. The patient underwent a corrective procedure of the deformity with an otherwise uneventful course (5). Piette *et al.* [2023] describe a 6-year-old patient who underwent surgical resection of a temporalis muscle rhabdomyosarcoma, followed by microvascular reconstruction utilizing a latissimus dorsi flap. The surgins achieved clear margins around the area of tumor, and the patient had healed well 15 days post-operation (6). These reports show the promising utility of free flap reconstruction in pediatric head and neck disease, with minimal complications and good outcomes.

Although the literature is rich with reports of successful management of pediatric head and neck tumors with surgical resection with free flap reconstruction, complications associated in the post-operative period have also been reported. A retrospective chart review of 16 patients who underwent free fibula grafts demonstrated two minor complications. A 15-year-old patient who underwent the procedure for hemifacial microsomia experienced a hematoma at the donor leg site and an 8-year-old patient who had suffered a gunshot wound to the neck experienced intraoral dehiscence (7). A retrospective study of 18 patients with benign mandibular tumors managed with vascularized fibular flaps demonstrated 100% flap survival. Complications included re-exploration for hematoma formation at the operative site in two cases and transient facial nerve neuropraxia in one case (8). In a case series evaluating the management of pediatric mandibular tumors, 56 patients were identified, 12 of whom were reconstructed using free tissue transfer. Complications from the surgery were not detailed (21). In a report on pediatric microvascular reconstruction from the Microvascular Committee, the authors reported on a consensus study involving 49 free tissue transfers. Four of 21 fibular flaps experienced minor dehiscence These were managed

without surgical intervention. One patient experienced breakdown at the radial forearm donor site due to infection, though this was adequately managed with appropriate wound care measures (22). Of note, a retrospective cohort study performed by Wolf *et al.* [2020] found that prior chemotherapy and radiotherapy were associated with a greater rate of early complications (23). While the reported complications have been relatively uncommon, complications can still occur with free flap reconstruction in the pediatric population and should be evaluated and managed appropriately. When evaluating a candidate for surgery, the risks should be thoroughly considered to ensure adequate understanding of these potential complications.

While the literature provides reports of promising evidence of the benefits of microvascular reconstruction with free tissue transfer, it is important to note that many of these have been case reports or small case series. As treatment algorithms evolve for local cancer control, further studies in the forms of cohort studies or clinical trials will be needed to further characterize the disease-related, cosmetic, and functional outcomes for children managed with radiation therapy *vs.* surgical resection with free flap reconstruction.

Conclusions

Free tissue transfer in the pediatric population is becoming increasingly utilized amongst surgeons for indications ranging from trismus release to resection of benign and malignant neoplasms. The literature is now rich with reports of pediatric patients treated with free flap reconstruction with successful outcomes and few postoperative complications or flap failures. Surgical resection with free flap reconstruction holds the potential of achieving clear pathologic margins with good functional and cosmetic outcomes, and should be considered as a therapy option in pediatric patients with head and neck tumors. Potential candidates for surgery should be thoroughly assessed and managed with the utmost care to minimize the risk of postoperative complications.

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Footnote

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Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://tp.amegroups.com/article/view/10.21037/tp-24-4/coif). C.A.E. has 3 patents [tracheostomy collar, otologic software, and mouthguard (Zotarix)]—2 for medical devices not relevant to this paper and one for a software diagnostic program; this author holds stock in 2 companies—one for a sinus relief device (TIVIC) and another for an otitis media software platform (Otologic); holds royalties in Grace Medical and Marpac; and provided expert testimony in a legal case for Elite Medical Experts which is not relevant to this paper. The other authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patients' parents or legal guardians for the publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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