Journal of Oral Biology and Craniofacial Research 12 (2022) 238–247

Contents lists available at ScienceDirect

Journal of Oral Biology and Craniofacial Research

journal homepage: www.elsevier.com/locate/jobcr

Reconstruction options in pediatric population with hemi or total mandibulectomy defects: A systematic review

Adity Bansal [a,*], Shakil Ahmed Nagori [b], Ashi Chug [a], Ashutosh Dixit [a], Rebecca Chowdhry [a], Srinivas Gosla Reddy [c]

[a] Department of Cranio-Maxillofacial Surgery/Dentistry, All India Institute of Medical Sciences (AIIMS)Rishikesh, Uttarakhand, 249203, India
[b] 21 Corps Dental Unit, c/o 56, APO, India
[c] GSR Institute of cranio-Maxillofacial and Facial Plastic Surgery, Vinay Nagar Colony, I S Sadan, Hyderabad, Telangana, 500059, India

ARTICLE INFO

Keywords:
Mandibular reconstruction
Fibula graft
Costochondral graft
Mandibular pathology
Hemi-mandibulectomy
Total mandibulectomy

ABSTRACT

Purpose: Mandibular pathologies causing extensive defects in pediatrics is relatively rare, and sparse literature is available for the reconstructive options of the same. The main aim is to provide optimum esthetics and function.
Materials and methods: PubMed, Cochrane CENTRAL, Embase, MEDLINE and Scopus databases were searched for articles published only in English language up to May 2021, involving reconstruction of hemi or total mandibulectomy defects in patients with the age of 8 years or less, associated with benign or malignant pathology. Primary outcome variable was the success of reconstruction. Qualitative analysis was performed using a microsoft excel-sheet.
Results: Of the 2201 articles reviewed, only fourteen were selected for data extraction. 22 patients were included. Fifteen were benign, six were malignant pathologies, and one was not defined. Hemi-mandibulectomy was performed in twenty-one cases, while one underwent total mandibulectomy. Condyle was preserved in five cases, while was removed in nine. Single-stage reconstruction was done in nineteen cases, while second-stage reconstruction was done in the other three. Reconstruction was done with fibular graft in thirteen cases, while CCG was used in others with variable follow-up time. Though minor complications were observed, success was observed in all cases post reconstruction with either fibula or CCG graft, defined either by function, or growth.
Conclusion: Irrespective of the age-group, nature of pathology and size of the defect, reconstruction should be considered with either fibula or CCG in single-stage, as they are equally efficacious, with minor complications. Well-defined reconstructive paradigm should be developed for pediatric mandibular reconstruction.

1. Introduction

Reconstruction of the mandibular defects remains a challenge in the pediatric population. Varied techniques have developed over time. Earlier attempts were made at the use of non-vascularized autogenous cortico-cancellous grafts like iliac crest and the costochondral (CCG) grafts, where the structural support was provided by the cortical portion, and surrounding soft tissues were responsible for revascularization.[1] However, when used in large defects, results were inconsistent; especially when placed in poorly vascularized or irradiated bone defects.[2] Regional muscle flaps then came into use as an attempt to improve the vascularity of the above grafts, wherein they provided both soft tissue and osseous support, along with muscle acting as a vascular carrier. But their use was limited in mandibular reconstruction as they are bulky with tenuous blood supply between muscle and periosteum, and a restricted arc of rotation of the pedicle.[3]

This led to the advent of the vascularized bone transfer, offering several advantages as it maintains the viability of the cells, and thus resulted in bone healing analogous to the healing of the fracture, rather than creeping substitution. Also, they provide an intact blood supply with greater resistance to infection.[4] Literature has described various such osseous flaps like scapula, ribs, second meta-tarsal, iliac crest, radius, and fibula. The use of each of these flaps had their limitations like the quality of available bone and donor site morbidities. However, free fibular grafts have been used most extensively for the reconstruction of long bone defects, mainly in adults. Another postulated phenomenon is

* Corresponding author.
E-mail addresses: aditybansal@rediffmail.com (A. Bansal), drshakilnagori@gmail.com (S.A. Nagori), ashichug@gmail.com (A. Chug), dixitdrashutosh@gmail.com (A. Dixit), rebeccachowdhry1987@gmail.com (R. Chowdhry), goslareddy@gmail.com (S.G. Reddy).

https://doi.org/10.1016/j.jobcr.2022.03.001
Received 10 March 2021; Received in revised form 23 February 2022; Accepted 14 March 2022
Available online 16 March 2022

"Spontaneous regeneration of mandible (SBM)", where osteoprogenitor cells reservoir i.e periosteum provides vascularity for new bone formation.[5] Adequate defect stability and soft tissue cover are principal factors in SRM.

Though there are well-defined guidelines for mandibular reconstruction in adults, however, the reconstructive paradigm in pediatrics is lacking and is controversial. The goal is to provide optimal aesthetic and functional restoration, as the child is still in the growing phase. This systematic review aimed to investigate the current evidence to access the efficacy of various reconstruction options in Hemi or total mandibulectomy defects in patients with the age of 8 years or less.

2. Materials and methods

2.1. Protocol and registration

This systematic review protocol was registered in PROSPERO i.e the International Prospective Register of Systematic Reviews (CRD42020201610). The planning and reporting of this review were by the recommendations defined by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA)[6] and the Cochrane Handbook for Systematic Reviews.[7]

2.2. Eligibility criteria

This review included studies involving the reconstruction of hemi- or total mandibulectomy defects in pediatric patients with the age of 8 years or less, associated with benign or malignant pathology including odontogenic lesions, traumatic injuries, vascular malformations, osteomyelitis, osteoradionecrosis (ORN), or medication-related osteonecrosis (MRONJ), involving only the mandible. Patients with the age >8 years, syndromic patients, patients with only soft tissue defects (e.g burns) or congenital deformities or temporomandibular (TMJ) ankylosis, were excluded. Only English language literature published up to May 2021 was included. Case report, case series, retrospective reviews, prospective studies, randomized clinical trials (RCTs), and controlled clinical trials were included. However, editorials, book chapters, and cadaveric studies were excluded.

The primary outcome variable used to assess the efficacy of the reconstruction method was the success of the reconstruction option, which was defined by the maintenance of its form and function. The additional outcome variable was complications encountered.

2.3. Information sources, search and study selection

An initial electronic search of PubMed, Cochrane CENTRAL, Embase, MEDLINE, and Scopus databases was performed for articles published only in the English language up to May 2021. A broad search was implemented utilizing a combination of Medical Subject Headings (MeSH) and free-text terms, using the following search strategy: ((("pediatric" OR "paediatric") AND ("mandibular" OR "mandible" OR "lower jaw") AND ("reconstruction" OR "resection and reconstruction")); 'Pediatric mandibular reconstruction'; 'Pediatric mandibular resection and reconstruction'; 'Pediatric mandible grafting'; 'Pediatric mandible reconstruction'; 'Pediatric mandibular resection'.

An electronic search for relevant articles in the journals of oral and maxillofacial surgery was also performed, including 'British Journal of Oral and Maxillofacial surgery (BJOMS)', 'Journal of Maxillofacial and Oral Surgery (MAOS)', 'Journal of Oral and Maxillofacial surgery (JOMS)' and 'Journal of Cranio-Maxillofacial surgery (JCMFS)', 'International Journal of Oral and Maxillofacial Surgery (IJOMS)'. Other sources scanned for additional records were bibliographic databases, the reference list of potentially included articles, conference proceedings, trial registers, and gray literature.

2.4. Data collection and analysis

Two reviewers (AB, SN) independently analyzed all the relevant studies based on the inclusion criteria mentioned above. At first, the screening of title and abstract was performed by the reviewers, followed by analysis at the full-text level. Any disagreements encountered were resolved by discussion with a third reviewer (GSR), until a consensus was reached. The following data were extracted from the included studies: patient age, gender, author, year of publication, journal, study location and type, number of eligible patients, type and nature of pathology, pathology dimensions, reconstruction method, stage of reconstruction, time is taken for reconstruction post-resection in double-stage cases, surgical approach, type of resection, whether condyle removed or preserved, graft type and its size (if mentioned), success rate (if defined by study), resorption of the graft (yes or no), maintenance of form and function, complications, follow-up period, pre- or post-surgery chemotherapy or radiotherapy, any previous surgery, recurrence, and any additional surgery at a later stage.

2.5. Quality assessment of individual studies

Considering the scarcity of studies on this topic, the included studies were only case reports and retrospective studies. Joanna Briggs Institute – University of Adelaide tool was used to critically appraise the case reports included.[8] They were rated as either "yes", "no", "unclear" or "not applicable" for every parameter.

2.6. Data analysis

Qualitative analysis was performed using the data as reported by the included studies using a Microsoft Excel sheet.

3. Results

3.1. Study selection

The designed search strategy yielded a total of 2201 records. After manual removal of the duplicates, 1009 articles were screened based upon their titles and abstracts, as per the defined inclusion criteria. 16 articles were then finalized for analysis of the full-text, following which two articles were excluded[9,10] Therefore, 14 articles were included in this review[11,12,13,14,15,16,17,18,19,20,21,22,2,23] No additional records were identified in the gray literature search or in the manual search of other databases or reference lists. Fig. 1 shows a flowchart depicting the selection process.

3.2. Quality assessment

Quality assessment of individual case reports is presented in Table 1. A clear description of the demographics, intervention, adverse events and take-away message was provided by all the case reports.[11,12,13,14,15,16,17,18,19,20,21,22,2,23] Maximum articles reported the patient's history[14,15,16,17,19,20,21] and clinical presentation adequately.[14,15,16,17,19,20,21,22] Diagnostic methods were delineated by all the case reports except one.[18]

3.3. Characteristics of the included studies

The characteristic of the studies included was evaluated and tabulated in Table 2.

Total of 14 articles were included in this systematic review, in which case reports were 11 in number[14,15,16,17,18,19,20,21,22,2,23] with the remaining being retrospective studies[11,12,13] Cases were reported to form different areas of the world like Brazil,[14] Israel,[15] Texas,[11] United Kingdom[16,18], Germany[17,20], New York,[13] Chile,[21] Boston,[12] Italy,[22] Canada,[2] United States,[23] and Malaysia.[19] The total number of the

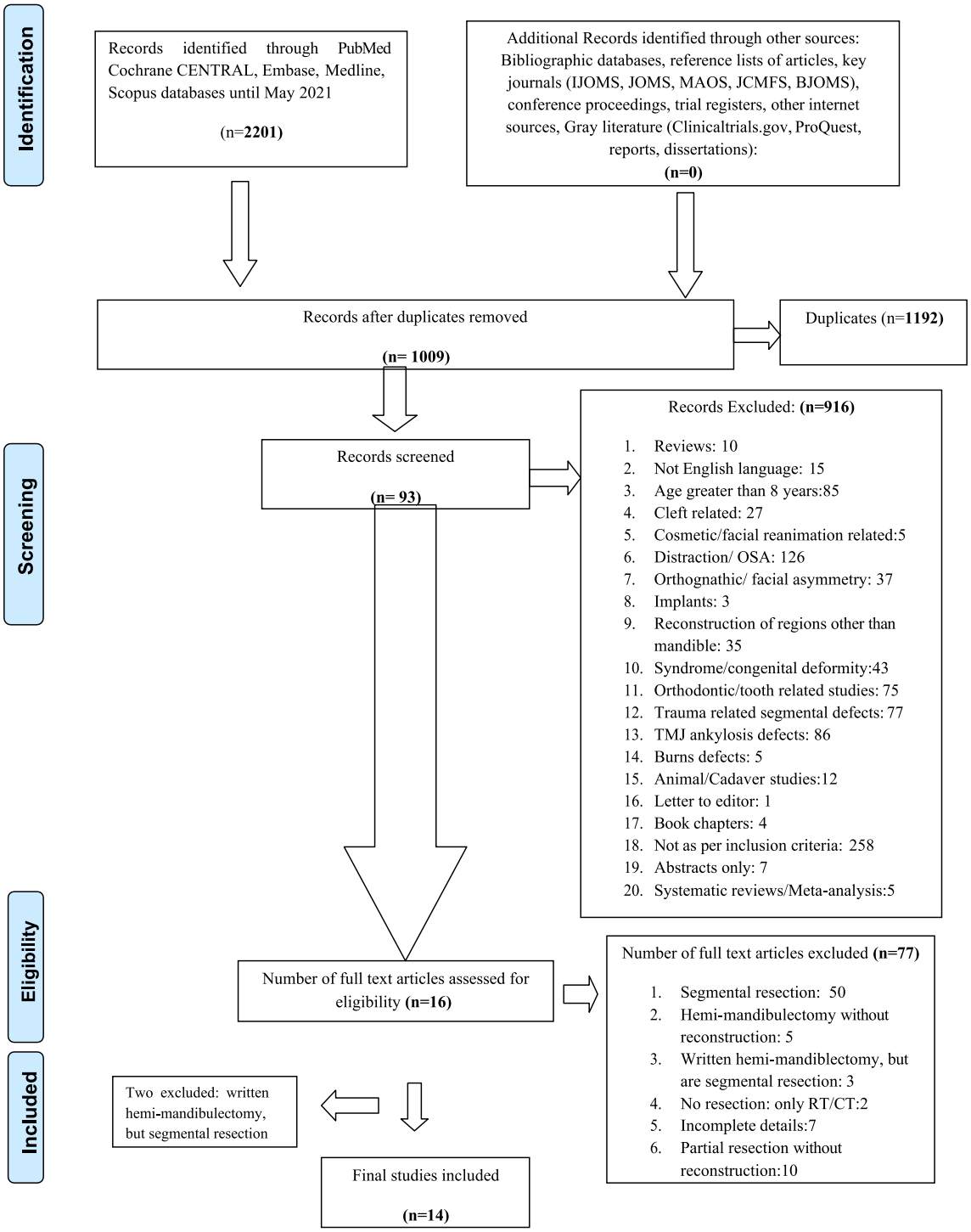

Fig. 1. Shows the PRISMA flowchart depicting the selection process.

patients included were 22, out of which 12 were females[11,14,15,17,12,22,19,20,23] and 5 were males[16,12,13,21].[2] The mean age of affected males and females was 48.8 months and 37.67 months respectively. The youngest children to undergo reconstruction in our review were found to be of 6 months (two cases).[17,20]

Of the total reported pathologies, 15 were benign[13,14,16,17,18,19,20,21,22,2,23] in nature and 6 were malignant,[15,11,12] and one was not defined.[8] The most common benign pathology reported were desmoplastic fibroma[21,22,23] with 5 cases; and four cases of melanotic neuroectodermal tumor (MNTI).[14,16,17,20] Ewing's sarcoma (2)[11,12] and rhabdomyosarcoma (2)[12] were the most common pathologies in the malignant category. Dimensions of the pathology were described in two of the malignant cases only. One case[15] described the three-dimensional length of a lesion as 7.8 cm × 5.5 cm x 5 cm; while the other case[11] only mentioned the length as 7 cm. Benign pathologies delineating the dimensions were 5 in number,[16,20,21,22] with the greatest being 5 cm × 4.5 cm x 3.5 cm in measurement.[20]

Hemi-mandibulectomy was performed in 21 cases; where condyle was preserved in 5 cases.[13,17,19,22,23] and removed in 9 cases[14,15,16,17,20,21,22,2] Total mandibulectomy was performed in only one case, along with bilateral removal of the condyles.[12] Reconstruction was done at the same time of mandibulectomy i.e single stage in 19

Table 1
Critical appraisal of the case reports included in the systematic review with Joanna Briggs Institute-University of Adelaide tool.

Author (year of publication)	Were patient's demographic characteristics clearly described?	Was the patient's history clearly described and presented as a timeline?	Was the current clinical condition of the patient on presentation clearly described?	Were diagnostic tests or assessment methods and the results clearly described?	Was the intervention(s) or treatment procedure(s) clearly described?	Was the post-intervention clinical condition clearly described?	Were adverse events (harms) or unanticipated events identified and described?	Does the case report provide takeaway lessons?
Volk et al. (2020)	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Kazakydasan et al. (2019)	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Skinner et al. (2017)	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Potter et al. (2016)	Yes	Unclear	Unclear	Unclear	Yes	Yes	Yes	Yes
Faria et al. (2013)	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Reiser et al. (2013)	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Ferri et al. (2012)	Yes	Unclear	Yes	Yes	Yes	Yes	Yes	Yes
Eckardt et al. (2010)	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Ahmed et al. (2007)	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Eckardt et al. (2001)	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Posnick et al. (1993)	Yes	Unclear	Unclear	Yes	Yes	Yes	Yes	Yes

cases.[11,12,13,14,15,16,17,18,19,20,21,22,2,23] Second stage reconstruction was done in the other three cases.[12,22]

Reconstruction was performed with grafts in all the single-stage reconstruction cases. Fibular grafts were used in eleven of the above cases[11,12,13,14,15,21,2,23], with eight cases reconstructed with costochondral grafts.[16,17,18,19,20,22] Fibular grafts were used for second stage reconstruction in two cases,[12] while CCG graft was used in the third case.[22] The time gap between mandibulectomy and reconstruction in double stage cases was found to be 6 years and 10 months,[12] 5 years,[12] and 1 year[22] respectively. The sub-mandibular approach was the most common surgical approach used.[16,17,19,20]

3.4. Data analysis and outcomes of the included studies

A descriptive synthesis of the study characteristics was evaluated and have been summarized in Table 3.

1. Success of reconstruction

The success of reconstruction was defined as qualitatively and distinctively by all the studies. The function was considered as one of the most common success criteria, described as no masticatory difficulty in five studies.[14,18,19,20,22] Three studies described success radiographically, mentioning it to be a stable bone reconstruction with a well-integrated graft.[15,11,13] Others described success as perfect bone healing and growth in coordination with craniofacial development.[16,17,21,12,2,23]

Nine cases received either pre or post-operative chemo or radiotherapy, with a variable number of cycles.[11,14,16,12,18,12] The cases observed varied follow-ups, with a maximum reported follow-up of 17 years.[22] Recurrence was not observed in the thirteen cases,[11,14,15,16,17,19,20,23] while it was not reported in the other nine cases.[12,13,18,21,22,2]

2. Complications

3.5. Recipient site complications

Recipient site complications were observed in the form of slight contour asymmetry or deficit of the mandible,[14,17] malocclusion, and deviation to the reconstructed site,[11] overgrowth in vertical direction,[20] extrusion of the miniplate,[16] transient paralysis of the facial nerve,[21,19] enlargement of rectus abdominis flap,[12] failure of growth of neo-mandible in stage-I surgery,[12] loss of vestibular depth intraorally,[2] and partial resorption of the proximal side of the graft.[22]

3.6. Donor site complications

Deviation of the ankle valgus,[14] great toe flexion contractures,[11] discomfort, and stiffness in the ankle[13] and sural nerve anaesthesia[21] were amongst the observed donor site complications.

4. Discussion

Reconstructive surgery following mandibular resection of benign or malignant pathology remains a controversial topic in the pediatric population. One can expect varied aesthetic deformities and irreversible functional deficits owing to the wide spectrum of pathologies involving the mandible.[24] It might result in wide continuity defects like hemi or total mandibulectomy, depending upon a multitude of factors like size and site of the lesion, nature of the lesion (benign or malignant), age of the patient, and concomitant radiotherapy or chemotherapy.[25] The age-group for this study has been chosen to be 8 years or below, as reconstruction options in such age-group has sparse literature, and continues to a topic of debate.

Reconstruction of such defects is a challenge in pediatrics as the aim remains to match the growth potential of the patient, which is impossible to predict.[26] Also, the graft donor sites fundamentally used in adults, like fibula, iliac crest, and CCG might lead to significant deformity locally in the younger population, due to the disruption of their growth center. Autogenous grafts, vascularized free flaps, or prosthesis have been utilized for such reconstruction, however, prosthetic material is not considered suitable in growing children as their dimensions are fixed, and there is a risk of condylar-head wear or fracture of the

Table 2
Depicts the characteristics of the included studies.

Ser no	Author	Year	Study location	Study type	Sample size	Age/ Gender	Pathology	Nature of pathology (Benign/ Malignant)	Pathology dimensions	Type of reconstruction	Stage of reconstruction	Time for reconstruction in double stage cases	Surgical approaches
1.	Volk et al.	2020	United States	Case report	1	2/F	Desmoid fibromatosis	Benign	NR	Graft and plating	Single	NA	NR
2.	Kazakydasan et al.	2019	Malaysia	Case Report	1	8 y/F	Basal cell ameloblastoma	Benign	NR	Graft and reconstruction plate	Single	NA	Sub-mandibular approach
3.	Skinner et al.	2017	Chile	Case report	1	3 y 7 m/M	Desmoplastic fibroma	Benign	5 cm × 4.5 cm x 3.5 cm	Graft and resorbable polylactate plate, and angle fixed with titanium plate	Single	NA	Pre-auricular approach extending to neck
4.	Potter et al.	2016	United kingdom	Case report	1	3 y/NR	Neuroblastoma	Benign	NR	Graft and mini fragment plates (IMF for two weeks)	Single	NA	Curvilinear incision
5.	Faria et al.	2013	Brazil	Case report	1	8 m/F	Melanotic neuroectodermal tumor	Benign	NR	Graft	Single	NA	NR
6.	Reiser et al.	2013	Israel	Case report	1	6 y/F	Pediatric ameloblastic fibro-odontosarcoma	Malignant	7.8 cm × 5.5 cm x 5.1 cm	Graft plus 2 mm reconstruction plate	Single	NA	Midline lip split extra oral approach (as per image)
7.	Ferri et al.	2012	Italy	Case report	3	3 y/F	Desmoplastic fibroma	Benign	45 x 50 × 43mm	Graft and miniplates	Single	NA	Combine cervical and intra-oral approach
						2 y/F	Desmoplastic fibroma	Benign	40 mm × 35 mm x43mm	Graft and miniplates	Single	NA	Cervical approach
						2 y/F	Desmoplastic fibroma	Benign	NR	Stage I: Graft and miniplates Stage II: Removal and second graft	Double	1 y	Cervical approach
8.	Eckardt et al.	2010	Germany	Case report	2	6 m/F	Melanotic Neuroectodermal tumor	Benign	NR	Graft	Single	NA	Submandibular approach
						2 y/F	Ameloblastoma	Benign	NR	Graft plus 1.5 mm titanium miniplates	Single	NA	Combined submandibular and transoral approach
9.	Crosby et al.	2008	Texas	Retrospective study	1	5 y/F	Ewing's sarcoma	Malignant	7 cm	Graft plus 2.4 mm reconstruction plate	Single	NA	NR
10.	Guo et al.	2008	Boston	Retrospective study	6	10 m/ NR	Germ cell tumor	NR	NR	Graft	Single	NA	NR
						14 m/ M	Rhabdomyosarcoma	Malignant	NR	Stage 1: titanium plate with acrylic spacer, which was covered with myocutaneous rectus abdominis free flap Stage II: Graft	Double	6 y 10 m	NR
						6 y/F	Ewing's Sarcoma	Malignant	NR	Stage I: Graft plus reconstruction plate Stage II: free fibula plus osseointegrated implants	Double	5 y	NR
						8 y/NR	Fibrosarcoma	Malignant	NR	Graft	Single	NA	NR

(*continued on next page*)

Table 2 (*continued*)

Ser no	Author	Year	Study location	Study type	Sample size	Age/ Gender	Pathology	Nature of pathology (Benign/ Malignant)	Pathology dimensions	Type of reconstruction	Stage of reconstruction	Time for reconstruction in double stage cases	Surgical approaches
						5 y/NR	Neural ectodermic tumor	Benign	NR	Graft	Single	NA	NR
						3 y/NR	Rhabdomyosarcoma	Malignant	NR	Graft	Single	NA	NR
11.	Ahmed et al.	2007	United Kingdom	Case report	1	7 m/M	Melanotic neuroectodermal Tumor	Benign	4.2 cm x 3 cmx 3 cm	Graft and 6 hole miniplate	Single	NA	Submandibular incision
12.	Eckardt et al.	2001	Germany	Case Report	1	6 m/F	Melanotic Neuro-ectodermal Tumor	Benign	3 cm × 4 cm	Graft	Single	NA	Submandibular approach
13.	Genden et al.	2000	New York	Retrospective Study	1	8 y/M	Aggressive Juvenile Fibromatosis	Benign	NR	Graft along with reconstruction plates and osseointegrated implants	Single	NA	NR
14.	Posnick et al.	1993	Canada	Case Report	1	7 y/M	Aggressive fibromatosis	Benign	NR	Graft and miniplates	Single	NA	NR

y, years; m, Months; NR, not reported; NA, not applicable; IMF, Inter-maxillary fixation.

prosthetic plate,[27] lead to inappropriate results due to rapidly evolving dynamics of the face. Also, no author has discussed the iliac crest bone graft reliability in such cases.[28]

Earlier, there was a consensus against the use of micro-vascularized bone graft in infants due to their incomplete ossification.[29] Posnick et al. first reported the use of vascularized fibula flap for reconstruction of the pediatric mandible.[2] During the wide literature search for the use of such grafts in hemi or total mandibulectomy defects, in children less than or equal to 8 years, yielded only case reports or retrospective studies, including 11 patients in this review.[12] All the cases showed varied age-groups and the nature of pathologies, with the two youngest age groups being 8 months[14] and 14 months.[12] Our review did not show any effect of the nature of lesion on the growth potential post-operatively, in contrast to a review which observed greater continued growth in benign lesions as compared to malignant lesions.[30] Though radiotherapy is known to cause impaired bone remodeling, however, none of the included studies reported growth restriction due to the same. Literature also could not identify any study focusing on the effect of chemo- or radiotherapy on graft success.

The potential for growth of the reconstructed mandible with vascularized fibular graft has been controversial. Three techniques have been defined for the fibular reconstruction of the larger defects.[30] In the first technique, distal fibular end substitutes the condyle, and the soft tissue covers its head, known as the "fibula substitute condyle technique".[27] In the "condyle graft technique", the resected condyle is transplanted to the fibular end.[31] In the third "condylar preservation" technique, the condylar head is preserved while resection.[32] This last technique is often preferred over others as it preserves the native condyle with the growth potential. Zhang et al. showed greater growth when the condyle was preserved (81.5%),[30] which is in contrast to our review where both the groups (condyle preserved or removed) retained their growth ability.

Literature reveals that even after condyle preservation, the growth is not consistent with the native mandible due to the interruption of continuity.[30] An animal study showed regeneration of the condyle is possible after condylectomy.[33] It is also observed that the surrounding tissues also facilitate the growth of the mandible.[34] Spontaneous regeneration of bone has been reported in literature if there is intact adjacent periosteum,[5] however, it does not apply to the wide continuity defects. Hence the growth of the un-resected mandibular segment along with graft remains uncertain. If condyle is resected, whether it should be reconstructed or not is also a matter of debate. Some studies recommend against its reconstruction.[11] Also, fibular graft carries the risk of ankle joint deformity,[14] therefore, it is suggested to preserve the distal end of the graft to stabilize the ankle.[12]

The use of CCG grafts in such wide hemi-mandibulectomy defects is also sparse. Only six case reports could be ascertained in literature, including nine patients, with 6 months being the youngest child to undergo the same.[17,20] All were benign pathologies, with no observed effect of chemo- or radiotherapy on the growth potential. Ahmed et al. advocated CCG graft for reconstruction of extensive defects of the mandible in the infants,[16] which is in accordance to the findings of Eckardt et al.,[17] where he concluded it to be ideal as they cause minimal donor site morbidity, and no resorption even after 15 years of follow-up. Potter et al. described superior epigastric artery based vascularized CCG graft, especially in cases where radiotherapy is indicated.[18] Usually CCG reconstruction is done with two consecutive ribs, in a "double-barrel" manner, to increase the height of the mandible, with its cartilaginous stump acting as the condylar head. Though it is considered anatomically similar to the mandibular condyle, it carries the risk of unpredictable overgrowth, governed by the amount of the graft cartilage.[31] CCG can also cause ankylosis due to the conversion of cartilage to bone.[35]

The success of the grafts, whether fibular or CCG, was evaluated via growth potential or radiological methods like orthopantomogram or computed tomography in the included studies, which have suggested that both the grafted fibular bone and the residual mandible facilitates the mandibular growth.[11] It has been observed that the width of the

Table 3
Depicts a summary of descriptive synthesis of the study characteristics.

S. No.	Author's name	Resection type (HM, TM)	Condyle preserved or removed?	Type of graft used/Size	Success rate	Resorption of grafts	Maintenance of form and function	Complications	Follow up period	RT or CT/ prior or post surgery	Any previous surgery	Recurrence	Plate/graft removal at later stage OR any additional surgery
1.	Volk et al.	Left HM	Preserved	Vascularized free fibula flap	Growth observed for neo-mandible	No	Facial symmetry, good occlusion and MIO	No	3y	NR	NR	No	Hardware removal after 8 m
2.	Kazakydasan et al.	Right HM	Preserved	CCG	No trismus, acceptable occlusion	NR	Right facial asymmetry	FN paraesthesia	6 m	NR	Same site surgical tumor removal, 9 m earlier	No	NR
3.	Skinner et al.	Right HM	Removed	Free fibular graft/ 14 cm-long osseous flap, 6 × 4 cm cutaneous island	Good flap growth	No	B/L condylar movement palpable No speech impairment	Immediate transient FN paralysis 2 m: sural nerve anaesthesia	6 y	NR	NR	No	NR
4.	Potter et al.	Right HM	NR	Vascularized C/L anterior rib segment	Good mandibular function	No	Good mandibular function	None	After every 3 m (limit NR)	Post operative RT (number of cycles NR)	No	NR	ND
5.	Faria et al.	Right HM	Removed	Right fibular osteocutaneous free flap/fibular segment: 8.9 cm; skin paddle: 4.5 × 2.1 cm^2	Good bone flap growth	No	Adequate mandibular contour and function	Ankle valgus deviation on the donor leg Slight mandibular contour asymmetry	12 y	3 courses of CT prior	No	No	NR
6.	Reiser et al.	Left HM	Removed	Free vascularized osteocutaneous fibular flap	OPG and 3D CT showed a stable bone reconstruction	No	Normal functional movements and aesthetics	None	1 y	NR	No	No	Planned within 2 y of surgery
7.	Ferri et al.	**CASE 1** Right mandibulectomy (till canine)	**CASE 1** Removed	**CASE 1** CCG (two ribs)	**CASE 1** Good aesthetic and functional outcome	**CASE 1** NR	**CASE 1** Good aesthetic and functional outcome	**CASE 1** None	**CASE 1** 3 y	**CASE 1** NR	**CASE 1** NR	**CASE 1** No	**CASE 1** NR
		CASE 2 Left HM	**CASE 2** Removed	**CASE 2** CCG (two ribs)	**CASE 2** Good aesthetic and functional outcome	**CASE 2** NR	**CASE 2** Good aesthetic and functional outcome	**CASE 2** None	**CASE 2** 2 y and 2 m	**CASE 2** NR	**CASE 2** NR	**CASE 2** No	**CASE 2** NR
		CASE 3 Right HM	**CASE 3** Preserved	**CASE 3** Stage I: CCG (one) with miniplates Stage II: second rib graft	**CASE 3** Reasonable aesthetic results Good restoration of facial symmetry	**CASE 3** After 1 y of stage I: partial resorption of the proximal graft	**CASE 3** After 8 y of Stage II: mandibular deviation	**CASE 3** After 1 y of stage I: partial resorption of the proximal graft; with subsequent non union	**CASE 3** Total of 17 y	**CASE 3** NR	**CASE 3** NR	**CASE 3** No	**CASE 3** Stage II: plate removal and second rib) 8 y after second surgery: sagittal osteo-distraction
8.	Eckardt et al.	**FIRST CASE** Left HM	**FIRST CASE** Removed	**FIRST CASE** CCG (two)	**FIRST CASE** Perfect bone healing	**FIRST CASE** 15 y: No resorption	**FIRST CASE** At 2 and 6 y: Slight vertical	**FIRST CASE** 15 y: facial contour deficit	**FIRST CASE** 15 y	**FIRST CASE** NR	**FIRST CASE** No	**FIRST CASE** No	**FIRST CASE** At 15 y: lateral mandibular

(*continued on next page*)

Table 3 (*continued*)

S. No.	Author's name	Resection type (HM, TM)	Condyle preserved or removed?	Type of graft used/Size	Success rate	Resorption of grafts	Maintenance of form and function	Complications	Follow up period	RT or CT/ prior or post surgery	Any previous surgery	Recurrence	Plate/graft removal at later stage OR any additional surgery
							overgrowth and transverse growth inhibition of reconstructed mandible						bone augmentation with autogenous iliac crest graft
		SECOND CASE Right HM	**SECOND CASE** Preserved	**SECOND CASE** CCG (two)	**SECOND CASE** Stable results of reconstructed mandible	**SECOND CASE** No resorption till 5 y	**SECOND CASE** Stable till 5 y	**SECOND CASE** None	**SECOND CASE** 5 y	**SECOND CASE** NR	**SECOND CASE** No	**SECOND CASE** No	**SECOND CASE** NR
9.	Crosby et al.	Right HM	NR	Osteo-cutaneous vascularized fibula flap	Regular diet and normal MIO OPG showed good bone union and growth	No	Malocclusion and deviation to the reconstructed site	Recipient site: wound infection, contour deformity, hypertrophic scar Donor sites: Great toe flexion contractures	4 y 9 and m	Pre operative CT (cycles NR)	Yes (NR)	No	Yes (time period NR)
10.	Guo et al.	**CASE 1** Right HM	**CASE 1** NR	**CASE 1** Osteocutaneous free fibular flap	**CASE 1** Successful	**CASE 1** NR	**CASE 1** NR	**CASE 1** None	**CASE 1** 2 y	**CASE 1** CT (both pre and post surgery; cycles NR)	**CASE 1** NR	**CASE 1** NR	**CASE 1** NR
		CASE 2 Right HM	**CASE 2** NR	**CASE 2** Stage 1: myocutaneous rectus abdominis free flap Stage 2: Free fibula flap	**CASE 2** Successful	**CASE 2** NR	**CASE 2** NR	**CASE 2** Subcutaneous portion of rectus abdominis flap enlarged after 7 y	**CASE 2** 7 y after Stage I After fibula: 2 y	**CASE 2** Not given	**CASE 2** NR	**CASE 2** NR	**CASE 2** C/L vertical ramus osteotomy at age of 8 y
		CASE 3 TM	**CASE 3** Removed bilaterally	**CASE 3** Coast to coast fibular reconstruction: Stage I: left fibula Stage II: right fibula	**CASE 3** Successful	**CASE 3** NR	**CASE 3** NR	**CASE 3** No growth of fibula neo-mandible	**CASE 3** 5y in stage I Stage II: 2 y	**CASE 3** CT (Time and number of cycles NR)	**CASE 3** NR	**CASE 3** NR	**CASE 3** Latisimus dorsi flap in second stage
		CASE 4 Left HM	**CASE 4** NR	**CASE 4** Left osteocutaneous fibular graft	**CASE 4** Successful	**CASE 4** NR	**CASE 4** NR	**CASE 4** None	**CASE 4** 2 y	**CASE 4** RT (Time and number of cycles NR)	**CASE 4** NR	**CASE 4** NR	**CASE 4** NR
		CASE 5 Right HM	**CASE 5** NR	**CASE 5** Right osteocutaneous fibular graft	**CASE 5** Successful	**CASE 5** NR	**CASE 5** NR	**CASE 5** None	**CASE 5** 2 y	**CASE 5** RT (Time and	**CASE 5** NR	**CASE 5** NR	**CASE 5** NR

(*continued on next page*)

Table 3 (*continued*)

S. No.	Author's name	Resection type (HM, TM)	Condyle preserved or removed?	Type of graft used/Size	Success rate	Resorption of grafts	Maintenance of form and function	Complications	Follow up period	RT or CT/ prior or post surgery	Any previous surgery	Recurrence	Plate/graft removal at later stage OR any additional surgery
										number of cycles NR)			
		CASE 6	**CASE 6**	**CASE 6**	**CASE 6**	**CASE 6**	**CASE 6**	**CASE 6**	**CASE 6**	**CASE 6**	**CASE 6**	**CASE 6**	**CASE 6**
		Right HM	NR	Right fibula	Successful	NR	NR	None	2 y	RT and CT (Time and number of cycles NR)	NR	NR	Rectus myocutaneous and right para-scapular flaps
11.	Ahmed et al.	Right HM	Removed	CCG	Stable graft	No	NR	Plate extrusion	2 m	9 courses of CT prior	No	No	Planned plate removal after 2 m
12.	Eckardt et al.	Left HM	Removed	CCG	Good graft healing Good functional	No	Vertical overgrowth and; growth retardation in transverse dimension	Vertical overgrowth and; growth retardation in transverse dimension	7 y	NR	None	No	NR
13.	Genden et al.	Left HM	Preserved	C/L free fibular graft	OPG observed symmetrical growth of face	No	No pain at rest or on physical therapy	At 2 y: "stiffness and discomfort" in ankle, but at y 2 m: no limitations	4 y 2 m	NR	NR	NR	Removal of reconstruction plates 18 m post operatively
14.	Posnick et al.	Left HM	Removed	Free fibular transfer/10 cm	Loss of vestibular depth	NR	Good MIO, and facial symmetry	loss of vestibular depth	2 y	No	Yes: Excision of aggressive fibromatosis of the left mandible	NR	NR

HM: hemi-mandibulectomy; TM: total mandibulectomy; y:years; m: Months; NR: not reported; CCG: Costochondral graft; B/L: Bilateral; ND: Not defined; FN: Facial nerve; OPG: Orthopantomogram X –ray; 3D CT: Three-dimensional computed tomography; C/L: Contra-lateral; MIO: inter-incisal mouth opening; RT: Radiotherapy; CT: Chemotherapy.

mandible shows a rapid increase either before 4 years or between 8 and 12 years, with a similar pattern for depth and height[36]; which is similar to the findings of a review where impaired growth was observed in more than half of children below the age of 8 years.[30] Fusion of the growth plates in case of the fibula is observed around 15–17 years of age.[13]

This systematic review aims to highlight that irrespective of the age-group, nature of pathology, and the defect size, reconstruction should be considered, preferably in single-stage, with either fibula or CCG, to prevent soft tissue relapse and also as it provides a bony bed for future prosthetic rehabilitation. Though the growth observed post-reconstruction at the age of 8 years or less might not be in total accordance with the native mandible, and hence might result in facial asymmetry with growth impairment at a later stage; however, several methods exist for the correction of the same. Early orthodontic treatment and augmentation osteoplasties would be useful for early control of growing deficits. Once skeletal maturity is attained, orthognathic surgery can be considered. Distraction osteogenesis has also been tried in mild contour defects, with un-clear results.[37] Also, some have suggested the use of vascularized scapular or iliac flaps at a younger age.[4] Consideration should be given for the removal of the implanted hardware after a while, as they might impede the mandibular growth.

Certain limitations were observed related to this review like limited sample size, variable age-group, and follow-up periods, differing methods of success evaluation, and lack of randomized controlled trials (RCT). Also, no set protocol was observed for the type of graft to be used in a particular age-group or the size of the defect. Therefore, we recommend RCT's with a larger sample size, so that a well-defined algorithm can be developed for the reconstruction of the pediatric mandible, especially in larger defects like hemi or total mandibulectomy.

This systematic review concluded that growth was observed in all the cases post reconstruction with either fibula or CCG graft. Also, no difference was observed in the growth potential if the condyle was resected or preserved. Neither the characteristic of the pathology, benign or malignant; or the pre or postoperative chemotherapy or radiotherapy hindered the growth. CCG grafts are usually preferred due to the ease of technique and low donor site morbidity, but microvascular grafts like fibula grafts are suggested in cases of malignancy. However, regular follow-up remains the mainstay of the treatment for growth monitoring. Also, it highlights the need for a well-defined reconstructive paradigm for pediatric mandibular reconstruction.

Funding source

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Declaration of competing interest

There is no conflict of interest to declare from any of the authors.

Acknowledgements

None.

References

1 Manchester WM. Immediate reconstruction of the mandible and temporomandibular joint. *Br J Plast Surg*. 1965;18(C):291–303.
2 Zuker RM, Wells MD, Zuker RM. Use of the free fibular flap in the immediate reconstruction of pediatric mandibular tumors: report of cases. *J Oral Maxillofac Surg*. 1993;51(2):189–196.
3 Green MF, Gibson JR, Bryson JR, Thomson E. A one-stage correction of mandibular defects using a split sternum pectoralis major osteo-musculocutaneous transfer. *Br J Plast Surg*. 1981;34(1):11–16.
4 Phillips JH, Rechner B, Tompson BD. Mandibular growth following reconstruction using a free fibula graft in the pediatric facial skeleton. *Plast Reconstr Surg*. 2005;116(2):419–424.
5 Rai S, Rattan V, Jolly SS, Sharma VK, Mubashir MM. Spontaneous regeneration of bone in segmental mandibular defect. *J Maxillofac Oral Surg*. 2019;18(2):224–228.
6 Selcuk AA. A guide for systematic reviews: PRISMA. *Turkish Arch Otorhinolaryngol*. 2019;57(1):57–58.
7 Yoshii A, Plaut DA, McGraw KA, Anderson MJ, Wellik KE. Analysis of the reporting of search strategies in Cochrane systematic reviews. *J Med Libr Assoc*. 2009;97(1):21–29.
8 Munn Z, Tufanaru C, Aromataris E. Data extraction and synthesis. *Am J Nurs*. 2014;114(7):49–54.
9 Sharma P, Williams R, Monaghan A. Spontaneous mandibular regeneration: another option for mandibular reconstruction in children? *Br J Oral Maxillofac Surg*. 2013;51(5).
10 Thariat J, Schouman T, Brouchet A, et al. Osteosarcomas of the mandible: multidisciplinary management of a rare tumor of the young adult a cooperative study of the GSF-GETO, Rare Cancer Network, GETTEC/REFCOR and SFCE. *Ann Oncol*. 2013;24(3):824–831.
11 Crosby MA, Martin JW, Robb GL, Chang DW. Pediatric mandibular reconstruction using a vascularized fibula flap. *Head Neck*. 2008;30(3):311–319.
12 Guo L, Ferraro NF, Padwa BL, Kaban LB, Upton J. Vascularized fibular graft for pediatric mandibular reconstruction. *Plast Reconstr Surg*. 2008;121(6):2095–2105.
13 Genden EM, Buchbinder D, Chaplin JM, Lueg E, Funk GF, Urken ML. Reconstruction of the pediatric maxilla and mandible. *Arch Otolaryngol Head Neck Surg*. 2000;126(3):293–300.
14 Faria JCM, Batista BN, Sennes LU, Longo MVL, Danila AH, Ferreira MC. Mandibular reconstruction with a fibular osteocutaneous free flap in an 8-month-old girl with a 12-year follow-up. *Microsurgery*. 2014;34(1):51–53.
15 Reiser V, Alterman M, Shuster A, Kaplan I. Pediatric ameloblastic fibro-odontosarcoma of the mandible: a challenge of diagnosis and treatment. *J Oral Maxillofac Surg*. 2013;71(1):e45–57.
16 Ahmed J, Chuckwulobelu R, Sebire NJ, Hartley BEJ, Dunaway DJ. Hemimandibulectomy and autologous CCG rib graft reconstruction for a case of melanotic neuroectodermal tumour of infancy arising within the mandible. *Int J Pediatr Otorhinolaryngol Extra*. 2007;2(3):189–193.
17 Eckardt AM, Barth E-L, Berten J, Gellrich N-C. Pediatric mandibular resection and reconstruction: long-term results with autogenous rib grafts. *Craniomaxillofacial Trauma Reconstr*. 2010;3(1):25–32.
18 Potter MJ, Watt-Smith S, Giele H. Innovative free costo-chondral rib flap based on the superior epigastric artery for reconstruction of the paediatric hemi-mandible. *J Plast Reconstr Aesthetic Surg*. 2016;69(9):1315–1317.
19 Kazakydasan S, Zamhari AK, Achol LT. Basal cell ameloblastoma in a paediatric patient: a case report and review of oral basal cell tumours. *Oral Surg*. 2019;12(3):248–254.
20 Eckardt A, Swennen G, Teltzrow T. Melanotic neuroectodermal tumor of infancy involving the mandible: 7-year follow-up after hemimandibulectomy and CCG graft reconstruction. *J Craniofac Surg*. 2001;12(4):349–354.
21 Skinner HR, Vargas A, Solar A, Foncea C, Astorga P. Desmoplastic fibroma of the mandible in a pediatric patient: a case report of resection and reconstruction with A six-year follow-up. *J Oral Maxillofac Surg*. 2017;75(7):1568. e1-1568.e10.
22 Ferri A, Leporati M, Corradi D, Ferri T, Sesenna E. Huge desmoplastic fibroma of the paediatric mandible: surgical considerations and follow-up in three cases. *J Cranio-Maxillofacial Surg*. 2013;41(5):367–370.
23 Volk AS, Riad SSH, Kania KE, et al. Quantifying free fibula flap growth after pediatric mandibular reconstruction. *J Craniofac Surg*. 2020;31(7):e710–e714.
24 Benoit MMK, Vargas SO, Bhattacharyya N, et al. The presentation and management of mandibular tumors in the pediatric population. *Laryngoscope*. 2013;123(8):2035–2042.
25 Chen SJ, Zheng XW, Lin X, Liu H. Ameloblastic fibro-odontosarcoma of the mandible in a pediatric patient. *Eur Ann Otorhinolaryngol Head Neck Dis*. 2016;133(6):419–421.
26 Valentini V, Califano L, Cassoni A, et al. Maxillo-Mandibular reconstruction in pediatric patients: how to do it? *J Craniofac Surg*. 2018;29(3):761–766.
27 Wax MK, Winslow CP, Hansen J, et al. A retrospective analysis of temporomandibular joint reconstruction with free fibula microvascular flap. *Laryngoscope*. 2000;110(6):977–981.
28 Fang QG, Shi S, Sun CF. Odontogenic lesions in pediatric patients. *J Craniofac Surg*. 2014;25(3):e248–e251.
29 Kumar BP, Venkatesh V, Kumar KAJ, Yadav BY, Mohan SR. Mandibular reconstruction: overview. *J Maxillofac Oral Surg*. 2016;15(4):425–441.
30 Zhang WB, Liang T, Peng X. Mandibular growth after paediatric mandibular reconstruction with the vascularized free fibula flap: a systematic review. *Int J Oral Maxillofac Surg*. 2016;45(4):440–447.
31 Moore EJ, Hamilton SS. Mandibular condyle reconstruction with fibula free-tissue transfer. *Ear Nose Throat J*. 2012;91(9):E18–24.
32 Nahabedian MY, Tufaro A, Manson PN. Improved mandible function after hemimandibulectomy, condylar head preservation, and vascularized fibular reconstruction. *Ann Plast Surg*. 2001;46(5):506–510.
33 Hans MG, Enlow DH, Noachtar R. Age-related differences in mandibular ramus growth: a histologic study. *Angle Orthod*. 1995;65(5):335–340.
34 Gomes AS, De Lima EM. Mandibular growth during adolescence. *Angle Orthod*. 2006;76(5):786–790.
35 Guyuron B, Lasa CI. Unpredictable growth pattern of CCG graft. *Plast Reconstr Surg*. 1992;90(5):880–886.
36 Farkas LG, Posnick JC, Hreczko TM. Growth patterns of the face: a morphometric study. *Cleft Palate-Craniofacial J*. 1992;29(4):308–314.
37 Carter LC, Aguirre A, Boyd B, Delacure MD. Primary leiomyosarcoma of the mandible in a 7-year-old girl: report of a case and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 1999;87(4):477–484.