Mandibular Defects and Reconstruction Post Oncological Resection in Paediatric Patients

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KEYWORDS

Paediatric Mandibular Neoplasms – Mandibular Reconstruction – Free Fibular Flap.

ABSTRACT

Introduction: Tumours of the paediatric mandible are rare clinical entities, which require a multidisciplinary team management. Surgery is frequently utilized as a modality for local control in the management of paediatric mandibular neoplasms. However, mandibular reconstruction in such age group is more challenging than in their adult counterparts, owing to the developing nature of the mandible and the craniofacial skeleton. Materials & Methods: The medical records of patients below the age of 18 years of age presented to the Paediatric Oncology and Head and Neck Surgical Oncology Departments at the National Cancer Institute of Cairo University from January 2014 to January 2024 with mandibular tumour were reviewed. The review includes; radiological site of the lesion, histo-pathological diagnosis, multidisciplinary team management. Patients that underwent segmental mandibular resection were evaluated for the type of the resultant mandibular defects according to Brown's classification of mandibular defects and the subsequent reconstruction procedures. Results: Forty-two patients were reported with mandibular neoplasm. Primary mandibular tumours were found in 35 cases (83.3%), of which non odontogenic tumours were diagnosed in 29 cases (69.0%). The anatomical subunits of the mandible; ramus, angle and body were involved in 14 (33.3%), 13 (31.0%) and 15 (35.7%) cases, respectively. Twenty-nine patients underwent segmental mandibular resection. According to Brown's classification; Class I, Class Ic, Class II, Class IIc, Class III, Class IV and Class IVc were the resultant defects in 0 (0%), 2 (6.9%), 10 (35.5%), 10 (35.5%), 3 (10.3%), 0 (0%), and 4 (13.8%) cases, respectively. Conclusion: The different pathologies of the mandibular neoplasms affecting the paediatric age group frequently involve the ramus of the mandible. With the surgery being the modality of choice in most of such pathologies, the resultant defects following segmental resection and the reconstruction approach in such age group vary greatly from the adult age group. Hence, the free fibular bone segments used for restoration of the ramus projection would be of a relatively larger length. Condylar resection, unfortunately, would hinder the restoration of occlusion relationship, essential for normal development of the maxillofacial skeleton.

1. Introduction

Tumours of the paediatric mandible are rare clinical entities, with most of these tumours being described in children over eight years of age. Most of the paediatric mandibular neoplasms are benign. Malignant neoplasms represent less than 10% of the cases. Mandibular tumours are classified according to their origin into odontogenic or non-odontogenic tumours. Odontogenic tumours arise from quiescent tooth-forming tissues of the mandible, yet their etiology remains unknown and their level of aggression vary widely. Non-odontogenic tumours arise from the mesenchymal or osseous tissue of the mandible, and encompass a broad range of pathologic conditions (1).

The incidence of non-odontogenic tumours is twice that of odontogenic tumours. Osteosarcoma and Ewing sarcoma are the most common non odontogenic tumours, and frequently presented with destructive bony lesion involving the mandibular ramus associated with extra osseous soft tissue component (2). Ameloblastoma, an odontogenic tumour, is the most common tumour diagnosed in paediatric patients and mainly found within the third molar region (3).

The extent of surgical resection depends on the pathology of the tumour, as it ranges from simple enucleation for benign tumours, segmental mandiblectomy only for locally malignant tumours, and segmental mandiblectomy with cervical lymph node dissection for malignant tumours as indicated. Overtreatment should be avoided, as implications of surgical management on the cosmetic outcome and maxillofacial skeleton development are profound in such age group (4).

In contrast to maxillectomy, continuity of the mandible after the resection surgery is required and an

appropriate reconstruction plan should be tailored according to presumed extent of resection. In all cases of segmental resection, a reconstruction plate to unite the native mandible to the bone graft, whether a non-vascularized bone graft for small sized defects or vascularized free fibular bone graft for larger defects, is needed (5).

The main difference between adult and paediatric patients in mandibular reconstruction is the growth potential of the utilized bone grafts segment with the subsequent affection of the growth of the native mandible. There are only few studies in the literature reporting on such growth potential, with some of those articles reported the hypertrophy of length and height of bone segments, while other studies described the arrest of bone segments' growth (6).

Moreover, the development of the mandible in paediatric patients depends on the primary growth centre which is located in the proximal zone of the conical subcondylar ridge. Thus, preservation of mandibular condyle and reconstruction of mandible in the rapid phase growth which occur between 8 and 12 years of age had great influence on growth of the rest of the mandible. Nevertheless, the development of the entire paediatric craniofacial skeleton is dynamic and continue via complex mechanisms attempting for proper facial symmetry, which requires an adequate occlusion relationship between the mandible and the maxilla to be established (7).

This study aimed to address the site of the tumours of different pathologies affecting the anatomical subunits of the mandible in the paediatric population, and whether the resultant mandibular defects following oncological resection and the subsequent reconstruction procedure would differ than those encountered in the adult age group.

2. Materials & Methods

The medical records of patients presented to the Paediatric Oncology and Head and Neck Surgical Oncology Departments at the National Cancer Institute of Cairo University from January 2014 to January 2024 with primary complaint of a jaw swelling or pain related to the mandible and diagnosed with mandibular tumour, were reviewed. The records of the patients below the age of 18 years of age at initial presentation were selected for detailed clinical data analysis, including; radiological site of the lesions, histo-pathological diagnosis, multidisciplinary team management. Those patients who underwent segmental mandibular resection had their defects classified according to Brown's classification of mandibular defects and the subsequent reconstruction procedures done were reported.

3. Results

Forty-two patients, below the age of 18 years, were diagnosed with mandibular malignancies of different histopathologies at the NCI in the period between January 2014 and January 2024, illustrated in Table (1). Primary tumours of the mandible represent the majority of the cases (35 cases), of which non-odontogenic & odontogenic tumours were found in 29 and 6 cases, respectively. The mandible was secondarily affected in the remaining 7 cases; through direct extension from nearby tumours in 6 cases and being a site of blood borne metastasis in one case.

Table (1): Pathology of Mandibular Tumors

Pathology	No. of Cases (%)
Primary Tumors	**35 (83.3%)**
Non-Odontogenic Tumors	**29 (69.0%)**
Ewing Sarcoma	11
Osteosarcoma	10
Fibrosarcoma	3
Burkitt Lymphoma	2
Langerhans Cell Histiocytosis	2
Cavernous Hemangioma	1
Odontogenic Tumors	**6 (14.3%)**
Ameloblastoma	4
Odontogenic Carcinoma	2
Secondary Tumors	**7 (16.7%)**
Via Direct Invasion	**6 (14.3%)**
Rhabdomyosarcoma	4
Mucoepidermoid Carcinoma	2
Via Metastasis	**1 (2.4%)**

Retinoblastoma	1

The age at the diagnosis range from 2 years to 18 years with the mean age of diagnosis being at 11.3 years of age. There was a slight female predilection of 1.47 ratio (25 females and 17 males). The panoramic radiographs were initially done to all patients, followed by CT scan of the head and neck. MRI evaluation was additionally requested in 29 patients for the assessment of the soft tissue extension of the tumour.

The site of the mandibular tumours with regards to the anatomical subunits of the mandible are illustrated in Table (2). All patients required an image guided needle core biopsy for definitive diagnosis, except for two patients; one being radiological diagnosed (Cavernous haemangioma) and the other was excised as an excisional biopsy (LCH). 16 cases required bone marrow aspiration & biopsy and PET/CT for proper staging of their disease.

Table (2): Site of Mandibular Tumours

Patholgy	Ramus	Angle	Body
Ewing Sarcoma	2	9	1
Osteosarcoma	8	1	1
Fibrosarcoma	1	1	1
Burkitt Lymphoma	0	0	2
Langerhans Cell Histiocytosis	0	0	2
Cavernous Hemangioma	0	0	1
Ameloblastoma	0	0	4
Odontogenic Carcinoma	1	0	1
Rhabdomyosarcoma	2	1	1
Mucoepidermoid Carcinoma	0	1	1
Retinoblastoma	1	0	0
No. of Cases (%)	**14 (33.3 %)**	**13 (31.0%)**	**15 (35.7 %)**

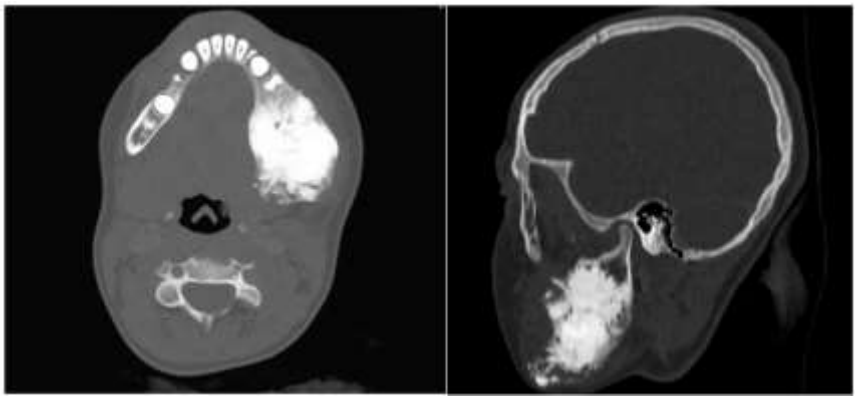

Figure (1): CT scan of 14 years female patient with Ameloblastoma involving the right posterior of the body of the mandible. A; Axial View, B; Sagittal View.

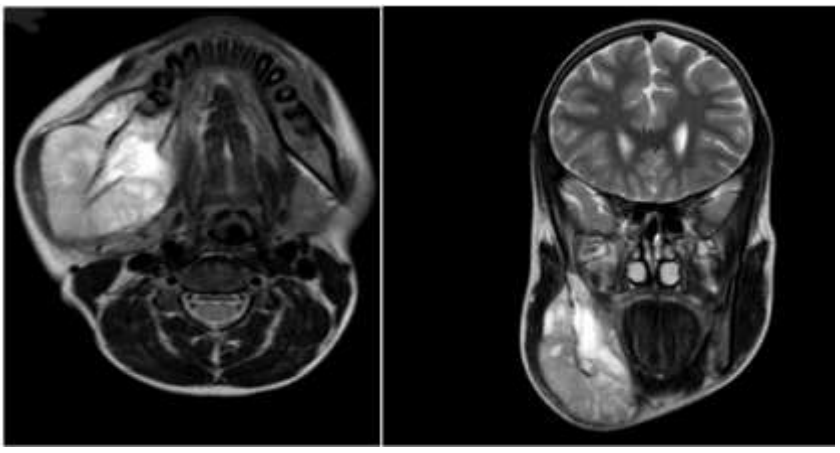

Figure (2): MRI of 16 years male patient with Ewing Sarcoma centered on the left mandibular angle. A; Axial View, B; Coronal View.

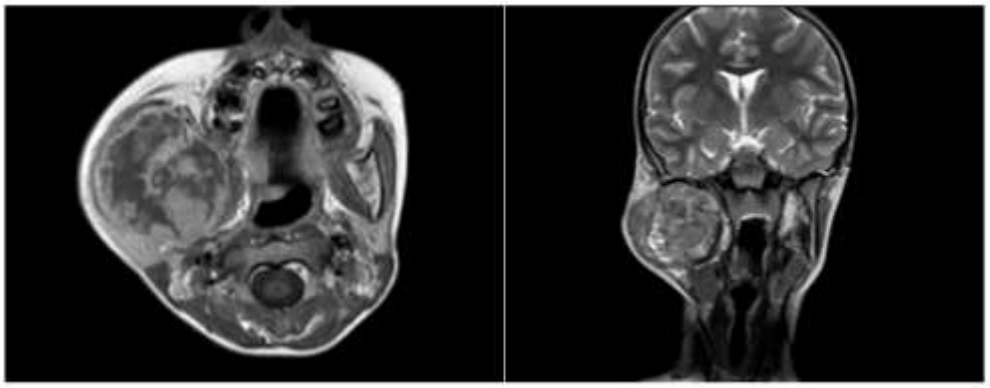

Figure (3): MRI of 8 years female patient with Osteosarcoma of the right mandibular ramus. A; Axial View, B; Coronal View.

The treatment of mandibular tumours necessitate the utilization of different modalities, which included; surgery, chemotherapy and/or radiation therapy, illustrated in Table (3). Eight patients were not subjected to any sort of surgical intervention. Four patients were treated with non-surgical modalities. The other four patients were planned initially for surgical resection of their mandibular tumours, however, the progression of their disease precluded them from curative surgical resection and were offered only palliative treatment.

Table (3): Different Management Modalities of Mandibular Tumours

Management Modality	No. of Cases (%)
Surgery	10 (23.8%)
Surgery & Chemotherapy	11 (26.2%)
Tri-modality	11 (26.2%)
Chemotherapy	7 (16.7%)
Chemotherapy & Radiation Therapy	3 (7.1%)

Thirty-two patients underwent mandibular resection surgery of different types with subsequent reconstructive procedures. Marginal mandibular resection had been done for three patients with no necessitation for bony reconstruction; two patient underwent horizontal marginal maniblectomy and one patient underwent vertical marginal mandiblectomy.

Twenty-nine patients underwent segmental resection of the mandible, illustrated in Table (4). Based on Brown classification, Class II defect, either with or without condylar resection, was the predominant defect type. (II; 10 patients and IIc; 10 patients). Class III and IVc defects were found in 3 and 4 cases, respectively. Two cases were classified as a Class Ic defect.

The masticatory muscles were resected as a safety margin in 16 cases, and were successfully preserved in the other 13 cases. The mucosa of the oral cavity was closed primarily in five cases with no need for mucosal reconstruction. The facial and cervical skin were preserved in all cases except for one advanced case.

Table (4): Brown's Classification of Mandibular Defects

Brown's Class	No. of Cases (%)
Class I	0 (0%)
Class Ic	2 (6.9%)
Class II	10 (35.5%)
Class IIc	10 (35.5%)
Class III	3 (10.3%)
Class IV	0 (0%)
Class IVc	4 (13.8%)

Two patients underwent segmental resection of the mandible and reconstruction with plate only with the mucosa being closed primarily and definitive bony reconstruction was delayed. One patient underwent resection of the mandibular ramus with neither mucosal nor bony reconstruction.

Twenty two patients had immediate reconstruction simultaneously with the resection surgery and four patients had the reconstructive procedure as a delayed intervention. Two of the four patients who underwent delayed reconstruction had their mucosal defect being reconstructed by pectoralis major myocutaneous flap at the time

of the surgical resection, while the other two patients had their mucosa closed primarily at the resection surgery.

A total of 26 patients underwent reconstruction of the mandible with a free fibular flap. An osteo cutaneous free fibular flap from the contralateral side was harvested in patients who underwent immediate reconstruction, where the skin paddle was utilized for mucosal reconstruction. In patients who underwent delayed reconstruction, an osseous free fibular flap from the ipsilateral side was harvested. In all of the patients, the pedicle was posteriorly directed and the fibular bone was osteomized for reconstruction of the horizontal contour of the mandibular body and the vertical projection of the mandibular ramus. In those patients who underwent condylar resection, the fibular bone segment were placed in contact with TMJ and held by a non-absorbable suture with the articular disc.

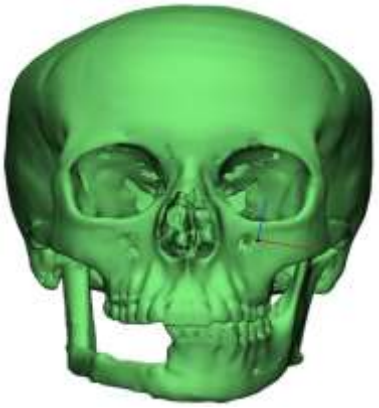

Figure (4): Frontal 3D reconstructed image of 16 years female patient with Class IIc defect reconstructed with free fibular flap.

Figure (5): Frontal 3D reconstructed image of 17 years male patient with Class II defect reconstructed with free fibular flap.

Figure (6): Frontal 3D reconstructed image of 5 years female patient with Class III defect reconstructed with free fibular flap.

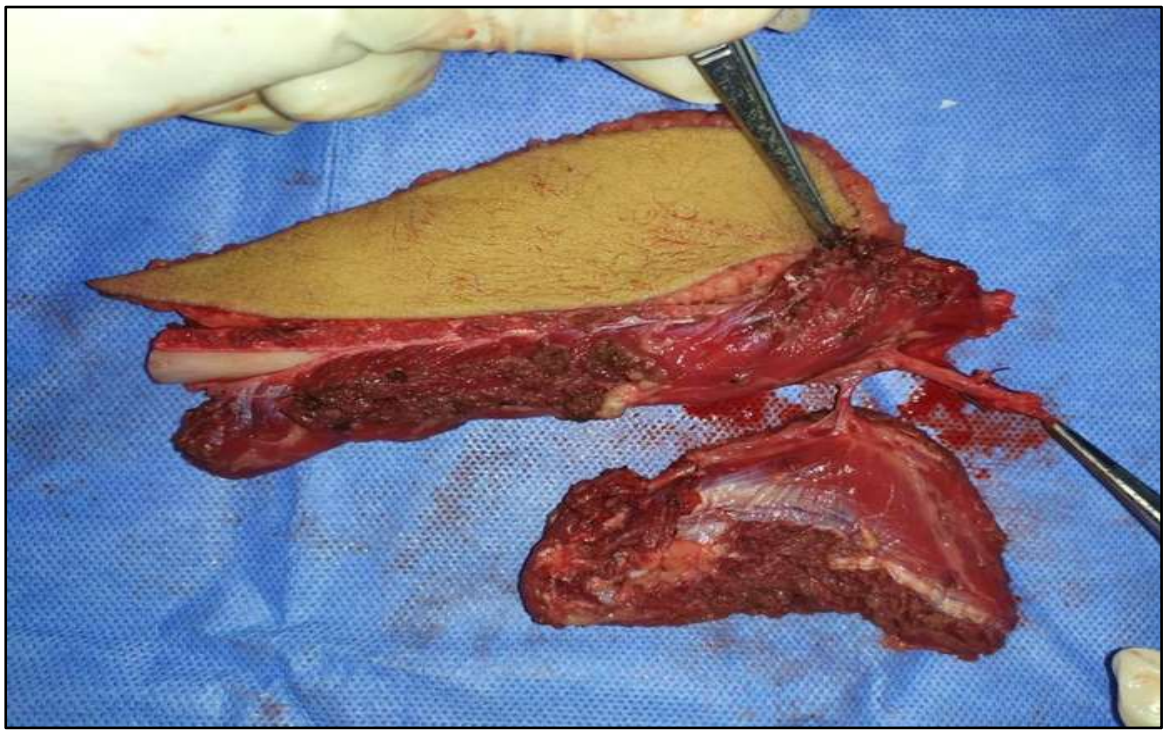

Figure (7): Chimeric Myo-Osteo-Cutaneous free fibular flap for reconstruction of Class IVc defect in 8 years female patient

Vascular comprise occurred in two cases. One case developed ischemia of the flap and resulted in complete loss of the flap and utilization of the pectoralis major myocutaneous flap for reconstructing the mucosal defect. The other case developed venous congestion of the flap and was successfully salvaged, however, this resulted in partial loss of the flap which necessitated debridement of the sloughed portion. Otherwise, all the remaining flaps showed complete survival. Other surgical complications included; Oral fistula (5 cases), hematoma (2 cases), surgical site infection (10 cases), necrosis of the cervical flap (4 cases), and hypertrophic scar formation (3 cases).

4. Discussion

Being a tertiary centre, neither the ratio between the malignant and benign lesions of the mandible, nor the ratio between odontogenic and non-odontogenic tumours are representative to the reported ratio in literature. However, the frequency of different pathologies of the malignant mandibular lesions in the paediatric age group are similar to that reported in literature, with osteosarcoma and Ewing sarcoma being the most common malignant pathologies of the mandible followed by direct malignant invasion of the mandible by rhabdomyosarcoma of the oral cavity (1).

The predilection of primary mandibular tumours to the anatomical subunits of the mandible was attributed to their pathology in our study. The osteosarcoma was commonly situated beneath the epiphyseal growth plate of the condyle. Meanwhile, Ewing sarcoma exhibits a more inferior position at the angle of the mandible. Such distribution is similar to their location in the long bones of the extremities. On the other hand, the odontogenic tumours tend to be located in the posterior portion of the mandible, the site of the permanent premolar and molar teeth. Lastly, the haematological malignancies are distributed all over the mandibular body, where there is abundance of the bone marrow.

The management of the paediatric mandibular tumours entails the integration of different modalities, which includes; chemotherapy, surgery and radiation therapy. In our study, surgery, in the form of segmental resection of the mandible, was the indicated as the modality of local control in more than 75% of the patients. Subsequently, the mandibular reconstruction procedure would be a challenging in such growing individuals, and would vary according to the age of the patient and the type of the resultant defect.

The timing of the definitive reconstructive procedure following oncological resection of mandibular tumours, whether immediate or delayed, is of a controversial debate, especially with malignant lesions. The possibility of adjuvant radiation therapy may adversely affect the functional and aesthetic outcome of the reconstructive procedure. Moreover, the occurrence of postoperative complication following such a complex procedure may

cause a delay in resuming the postoperative adjuvant therapy. This was observed with five cases in our study that experienced postoperative wound complications and were delayed for their adjuvant therapy (8).

On the other hand, delaying the reconstructive procedure had several disadvantages, which had been encountered in the four cases in this study that had their reconstructive surgery at a latter session after their resection surgery. First, the operative bed would be unfavourable medium for the flap and highly susceptible to infection. Second, the retrieval of vessels of good quality for proper micro vascular anastomosis would be more challenging. Third, releasing the contracture of the cervical skin may result in an outer defect, requiring additional external reconstruction. Lastly, the position of the mandible may be altered by postoperative fibrosis, with subsequent difficulty in re-establishing normal occlusion (9).

In our study, the types of the resultant defects following mandibular resection surgery in paediatric population according to Brown's classification were different than those encountered in adult population. This was noticed with the high incidence of condylar resection in more than half of the patients, with subsequent resection of the masticatory muscles. Moreover, the incidence of Class III defect was only 10%, denoting the high frequent involvement of the mandibular ramus within the resection surgery (10).

The reconstruction procedure following resection surgery with utilization of free fibular flap in paediatrics had also shown different technical aspects when compared to adult age group, apart from the growth concern regarding either the fibular bone segments or the native mandible. With condylar resection, the length of the fibular segment required for ramus reconstruction would be larger than usual to re-establish a neo TMJ. In adult population, as no concern regarding growth, reconstruction of the condyle and the TMJ would be easily accomplished via TMJ prosthesis. This will also not necessitate a longer fibular segment for ramus reconstruction (11).

Moreover, the importance of the condyle for mandibular reconstruction in paediatric population is pivotal, not only as a growth centre, but also as a counter regulator to the contralateral TMJ function. This was encountered in our study in those patients with condylar resection. Being unopposed, the masticatory muscles of the contralateral side resulted in difficulty in restabilising a normal occlusion relationship between the maxilla and the mandible. Such relationship is mandatory for normal development of the craniofacial skeleton.

5. Conclusion

The different pathologies of the mandibular neoplasms affecting the paediatric age group frequently involve the ramus of the mandible. With the surgery being the modality of choice in most of such pathologies, the resultant defects would alter the reconstruction procedure encountered in the adult age group. With the utilization of the free fibular flap for mandibular reconstruction, the fibular bone segments used for restoration of the ramus projection would be of relatively larger length. Moreover, the frequent resection of the condyle would hinder the restoration of occlusion relationship, which is essential for normal development of the maxillofacial skeleton.

6. Recommendations

The restoration of the vertical height of the mandibular ramus may entails some technical modifications of the utilized free fibular flap, with regards to the pedicle orientation and the laterality of the flap to the operative side. Also, condylar resection and the subsequent loss of the masticatory muscles would necessitate regular follow ups to adjust a normal occlusion relationship between the reconstructed mandible and the native maxilla.

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