Original article

Narrative Review on Prosthetic Reconstruction of Neurofibromatosis Cases Undergoing Hemi-Mandibu lectom

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ABSTRACT

Neurofibromatosis (NF) is an autosomal-dominant inherited syndrome with teeth retention and complex tooth deficiencies which can affect swallowing, speech, esthetic, and airway patency. That's where the rehabilitation of structures, functions, esthetics deformities, and life quality with different prosthetic choices should be considered. The present literature review inspected the prosthetic treatment plans to rehabilitate NF type 1 after hemi-mandibulectomy. By an extensive literatures search, some articles were chosen. The prosthetic treatment options for dental reconstruction of the hemi-mandibulectomy can be conventional removable prostheses, implant-supported fixed protheses, and implant-supported removable protheses. Ultimately, choosing among prosthetic treatment plans is made based on clinician opinion, patient preference, and ridge condition in each case.

Keywords:Bone *deformity; Mandibular reconstructions; Neurofibromatosis*

INTRODUCTION

A neurocutaneous disease called neurofibromatosis (NF) is an autosomal-dominant inherited syndrome with three different types.[1,(2] NF1 occurs almost 20 times more than NF2.[3,4] NF1discovered by Von Recklinghausen[3,5] shows symptoms such as café-au-lait macules, Lisch nodules of the iris, and interriginous freckling.[6)-8] Different malformations and tumors manifest with this syndrome however, neurofibroma, the benign nerve sheath tumor is more common among them.[9,10]

NF1 can act as a bone disease and cause jaw deformity if the plexiform of neurofibroma cells (PNF) are involved.[11] This jaw deformity can cause significant problems in the dental procedure and surgical approach.[12] Also, in the facial region, tumors can disfigure the soft tissue extensively, usually unilateral.[(13)(14)] Teeth retention and complex tooth deficiencies are the syndrome's oral manifestation.[13),15]

NF2, in contrast to NF1, is characterized by bilateral schwannoma involvement of the superior vestibular branch of the eighth cranial nerve and it doesn't have oral features.[16] That's why NF2 wasn't included in this review.

To treat the neurofibroma tumor, still, no significant approach is introduced. Surgical

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removal is the gold standard 00 treatment plan.[1,12,] However, it can affect swallowing, speech, esthetic, and airway patency.[11,17)] This is the point where the rehabilitation of structures, functions, esthetics deformities, and life quality with different prosthetic choices is brought out.[18] The present literature review inspected the prosthetic treatment plans to rehabilitate five cases of NF type 1 which went through hemi-mandibulectomy.

MATERIAL AND METHODS

An extensive literatures search had been carried out by combing through the Embase, Medline, and PubMed databases.

Inclusion criteria were met if (1) abstract topic related to NF1 and prosthetic rehabilitation; (2) article was in English language; and (3) full-text article was available.

Records were then excluded if one of the following criteria pertained: (1) inadequate patients' information was provided (age, sex, type of NF, jaw lesion location, clinical and radiographic findings); (2) no oral involvement was mentioned. The collected data organized and analyzed. Table 1 presenting the data of the investigated 5 cases.

FINDING

The study was based on 5 patients, 3 females and 2 males. The youngest case was 21 years old and the oldest was 65 years old. Based on the location of the jaw lesions, all cases were in the mandible along with maxillary lesions in 4 cases. (Table1)

The Clinical, radiographic, and pathological characteristics were evaluated and listed on table 2.

The radiographic appearance was radiolucent in all cases. When the effect of tumors on the mandibular canal was assessed, the enlargement of the canal was seen in Abrah an et al[22], Schneider et al[20], and Kokovic et al[23] case reports and the involvement of canal was

Table 1: Characteristics of the studied cases.

Case number	age	sex	Lesion location	Frequency	Age at diagnosis of NF 1	Familial history	References
1	58	Female	• Maxilla: In right maxillary sinus • Mandible: All over the arch	Multiple	15	None	Tosios et al 2018[19]
2	21	Female	• Maxilla: In the left side • Mandible: In the left side	Multiple	21	Not mentioned	Schneider et al 2017[20]
3	45	male	• Left posterior part of the mandible crossing the midline	Single	Not mentioned	Not mentioned	Narang et al 2017[21]
4	65	Female	• Maxilla: All over the arch • Mandible: All over the arch	Multiple	4	None	Abrahan et al 2016 [22]
5	57	Male	• Maxilla: In the right and left sides (not anterior portion) • Mandible: All over the arch	Multiple	Not mentioned	Not mentioned	Kokovic et al 2012[23]

stated in Narang et al study.(21) However, in Tosios et al study the mandibular canal and mental foramen were not identifiable.(19) Teeth displacement was observed on Narang et al case report. One of the patient suffered from left hemi-facial paralysis due to the tumor.(20)

The surgical procedure and prosthesis treatment plan in these articles were listed in table 3. As can be seen, different cases have

Table 2: Clinical, radiographic, and pathological characteristics of the studied cases.

1	Intra oral clinical assessment	• Maxilla: Well-defined brown tumors in labial, Palatal expansion • Mandible: An indurated, yellow-colored mass extended from the alveolar ridge to the floor of the mouth	Tosios et al 2018[19]
	Radiographic assessment (Panoramic view and CBCT)	• Maxilla: A radiolucency with indiscrete borders • Mandible: Poorly defined, multilocular radiolucencies all over the arch	
	Pathological assessment	• Not mentioned	
2	Intra oral clinical assessment	• Maxilla: Narrow and deformed ridge, hyper plastic soft tissue • Mandible: Narrow and deformed ridge, hyper plastic soft tissue	Schneider et al 2017 (20)
	Radiographic assessment (Panoramic view)	• Maxilla: A radiolucency. • Mandible: A radiolucency, shortening of the ramus, narrowing of the mandible body, and rarefaction of the coronoid and articular process.	
	Pathological assessment	• Not mentioned	
3	• Intra oral clinical assessment	• Irregular shaped swelling with firm consistency from the left third molar region to the central incisor of the opposite side crossing the midline.	Narang et al 2017[21]
	• Radiographic assessment • (Panoramic view)	• Irregular radiolucency, teeth displacement, and teeth missing	
	• Pathological assessment	• Hyper cellular connective tissue stroma consisting of spindle shaped cells, with wavy nuclei arranged in fascicular and storiform patterns	
4	• Clinical assessment	• Multiple soft nodular masses on the tongue, maxillary and mandible edentulous arches	Abrahan et al 2016[22]
	• Radiographic assessment • (Panoramic view)	• Enlargement of the mandibular canal	
5	• Clinical assessment	• Maxilla: Narrow residual ridge on left and right side (without involvement of anterior portion), buccal soft tissue of the right cheek • Mandible: Narrow residual ridge	Kokovic et al 2012[23]
	• Radiographic assessment • (Panoramic view and CT)	• Maxilla: Increase in bone density • Mandible: Lateral bowing of the mandibular ramus, increase in dimensions of the coronoid notch, and a decrease in the mandibular angle	
	• Pathological assessment	• Not mentioned	

Table 3: Surgical procedure and prosthetic treatment plan of the studied cases.			
	Surgical procedure	Prosthetic treatment plan	Follow up
1	Was not performed due to patient desire.	• Maxilla: Tissue-supported removable complete denture • Mandible: Tissue-supported removable complete denture	Patient refused.
2	Hemi-mandibulectomy to remove the lesion and impacted teeth.	• Maxilla: Screw-retained implant-supported fixed partial prothesis • Mandible: Screw-retained implant-supported fixed partial prothesis	Several years
3	Left hemi-mandibulectomy along with the dissection of supra-omohyoid lymph nodes	• Mandible: Implant-supported fixed partial prothesis	Not mentioned
4	Hemi-mandibulectomy was performed before referring.	• Maxilla: Tissue-supported removable complete denture • Mandible: Tissue-supported removable complete denture	Not mentioned
5	Hemi-mandibulectomy was performed before referring.	• Maxilla: Implant-supported removablepartial prothesis • Mandible: Implant-supported removable complete denture	2 years

different treatment plans. In none of the studied cases, recurrence of the lesion was reported.

DISCUSSION

The prosthetic treatment options for dental reconstruction of the hemi-mandibulectomy can be conventional removable prostheses, implant-supported fixed protheses, and implant-supported removable protheses.

Conventional prostheses may not be appropriate [24][25] because they are incompatible with excessive soft tissue contour and defective bone morphology.[24] this is where implant-supported protheses are more welcomed.[26,27] However, in two studied case reports, the conventional removable prostheses were delivered.[19,22]

Despite the long-term success of the implant-supported fixed protheses, the placement of sufficient numbers of implants can be restricted by severe bone resorption and financial limitation.[28][29] In these conditions, Implant-supported removable prostheses become bold.[30]

Implant-supported removable prostheses are of great advantages in terms of improvement of mastication, speaking ability, and quality of life.[30][31] Also, implant-supported overdentures will create pink interdental papilla better than implant-supported fixed prostheses [32][33][34]and as well, they have flanges to rehabilitate the support-less soft tissue.[30][34] In deviated mandible cases where the forces on implants are angled, and in cases with the restricted mouth opening, removable prostheses perform better.[30]

To avoid peri-implantitis, in patients with poor oral hygiene, implant-supported removable prostheses have been mostly indicated because it's easy to keep them clean.[26,30]

It is worth noting that, implant-supported removable prostheses offer very significant improvement for patients with systemic-diseases because of the fewer implants required compared to implant-supported fixed prostheses.[30.35] Only in one of the studied cases, implant-supported removable prostheses were delivered.[23]

Along with the dental reconstruction, the reconstruction of the resected bone is important.[36] The jaw reconstruction is a challenging approach which can be more complicated if the fibrosis and scarring of soft and hard tissue happen due to delayed reconstruction.[38][37] A sound and healthy bone graft with a titanium reconstruction plate is an ideal choice to restore the segmented mandible.[39][40] In none of the cases reviewed in this manuscript, the bone reconstruction were conducted.

CONCLUSION

Ultimately, choosing among prosthetic treatment plans is made based on clinician opinion, patient preference, and ridge condition in each case. This statement is of great importance in hemi-mandibulectomy cases where the rehabilitation approaches are more challenging.

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