

Characteristic of Ameloblastoma in Oral and Maxillofacial Surgery Hasan Sadikin Hospital: 1 Year Retrospective Study

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Abstract: Introduction: Ameloblastoma is the most common odontogenic tumour. Ameloblastoma is a borderline tumour because ameloblastoma is a benign but locally aggressive tumour with a high recurrence rate if the excision is not complete. The principle of treatment for ameloblastoma is excision all the tumour. Method: This study is a retrospective study with 76 patients diagnosed with ameloblastoma during 2019-2020. Data taken from each patient were gender, age, radiological features, the histopathological diagnosis of the location of ameloblastoma, management, defects, reconstruction. Result: A total of 35 patients were plexiform ameloblastoma, 22 patients with follicular ameloblastoma, 3 patients with mixed plexiform and follicular ameloblastoma and 16 patients with unspecified ameloblastoma. 30 patients were treated radically, while 16 patients were treated conservatively. Discussion: Radical treatment is more often used to reduce recurrence rates, whereas conservative measures are indicated in children and adolescents, as well as adult patients with unilocular ameloblastoma types. Conclusion: The most characteristic of ameloblastoma is plexiform ameloblastoma and located in the mandible. Ameloblastoma performed radically and reconstructed with an AO plate.

Keywords: Ameloblastoma, Characteristic, Epidemiology

1. Introduction

Ameloblastoma was the most common odontogenic tumor, based on Nalabolu et al. in 2016 showed that ameloblastoma occurred in 49% of odontogenic tumor cases. Based on its nature, ameloblastoma is an intermediate (borderline) tumor because ameloblastoma was benign but locally invasive with a high recurrence rate if the excision is incomplete.

A systemic review by McDonald-Jankowski et al. showed that the number of ameloblastoma in hospital was significantly higher in Asian or African populations than in European or American hospitals. Lu et al. studied a Chinese population and showed a mean age of 31.4 years with a 1.5:1 male:female ratio and 90.8% of the tumors were in the mandible. A study by Hatada et al. in the Japanese population showed a mean age of 34.7 years with a ratio of 1.6:1 male: female and 92.6% located in the mandible.

Research by Ruslin et al. in Eastern Indonesia, from 84 patients diagnosed with ameloblastoma, 14 patients were diagnosed with unicystic ameloblastoma (25%), thirty-two patients with follicular ameloblastoma (57%) and 10 patients with multicystic ameloblastoma (18%). The principle of therapy for ameloblastoma of the jaw was the removal of the tumor as a whole. Many clinicians prefer radical therapy because of its relapse nature. Treatment with radical methods can reduce the recurrence rate.

This tumor had a high recurrence rate, so close evaluation and observation after treatment is necessary. Hasan Sadikin Hospital is a referral center from the province of West Java and there had been no research on the characteristics of ameloblastoma that occurs in West Java, especially in Hasan

Sadikin Hospital. Based on the formulation of the problem stated, the purpose of the study is to conduct a retrospective study to analyze the characteristics of ameloblastoma in patients at the RSHS Oral Surgery KSM in the period January 2019 – December 2020.

2. Material and Method

The sample in this study were patients diagnosed radiologically and histopathologically as recurrent ameloblastoma at Hasan Sadikin Hospital in the period January 2019 – January 2020. This study was conducted using a total sample.

The age category in this study was the age when the patient was diagnosed with ameloblastoma, grouped into 5 age groups, namely 0-10 years; 11-20 years old; 21-30 years old; 41-50 years and above 50 years. The gender category is divided into 2, male and female. Based on the supporting radiological examination, it was taken by reading the radiological images, both panoramic images and head CT scans where ameloblastomas were grouped into 3 types, unilocular, multilocular (soap bubble appearance) and desmoplastic (scallop appearance).

Based on histopathological examination, ameloblastomas were grouped into 6 types, unilocular, plexiform, follicular, acantomatous, granular, basal cell and desmoplastic. The locations of ameloblastomas were divided into 6 locations, anterior maxilla, right maxilla, left maxilla, anterior mandible, right mandible and left mandible.

Management of ameloblastoma was generally divided into 2 types, conservative and radical. There are two types of

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conservative management, enucleation and dredging, while the radical treatment includes marginal resection, segmental resection, hemimandibulectomy/hemimaxillectomy and total mandibulectomy/maxillectomy.

Assessment of the magnitude of maxillary defects after ameloblastoma treatment based on the Classification System for Maxillectomy Defects (Figure 1) was divided into Limited maxillectomy defects involving one or two maxillary walls accompanied by buccal soft tissue covering the maxilla or critical structures such as lips, nose and eyelids. Subtotal Maxillectomy Defects covered the palate but the dentoalveolar and orbital walls remained intact. Subtotal maxillectomy defects are divided into 2 types, type IIA and type IIB. Type IIA defects involve less than 50% of the maxilla and do not cross the midline, whereas type IIB involve more than 50% of the maxilla and cross the midline. Total Maxillectomy Defects involve the orbital floor, dentoalveolar and may be accompanied by orbital resection. This defect was divided into 2 types, type IIIA and type IIIB. Type IIIA defect involves the entire maxillary wall

including the orbital floor but leaves structures within the orbit. Type IIIB involves the entire maxillary wall including structures within the orbit. Orbitomaxillectomy defect involving all maxillary structures, superior orbital wall and deep orbital structures.

Classification of mandibular defects by Roman numerals (I, II, and III) indicating the bone defect and letters (A, B, C, D) indicating soft tissue quality (Figure 2). Type I involves the anterior mandibular arch (mandibular symphysis and/or parasymphysis either unilateral or bilateral). Type II involves half of the mandible (corpus, angle, ramus, with or without condyles). Type III involves a lateral defect involving one or two or three parts of the mandibular structure (corpus, angulus, corpus).

Type A showed no soft-tissue defects. Type B involves intraoral soft tissues. Type C indicates a defect in the skin or extra-oral soft tissue. Type D defects occur in either intraoral or extra oral soft tissue.

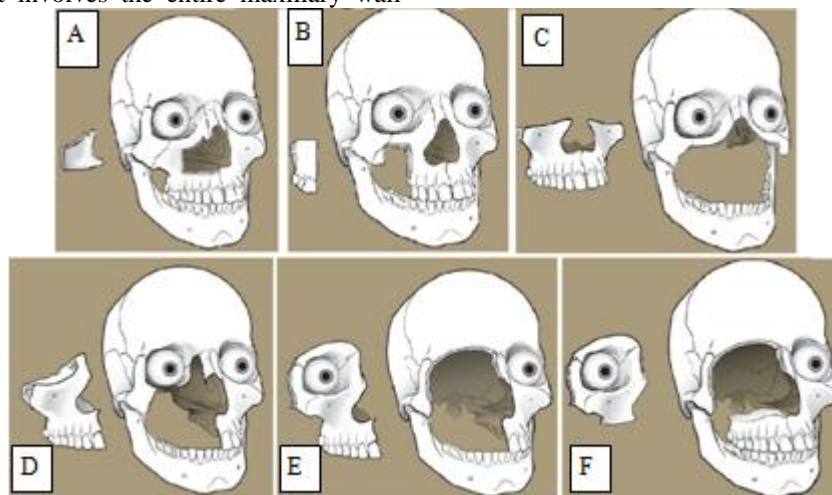


Figure 1: Classification of maxillary defects; 7a. Type I. Limited Maxillary Defects; 7b. Type IIA. Subtotal Maxillectomy Defects; 7c. Type IIB Subtotal Maxillectomy Defects; 7d. Type IIIA Total Maxillectomy Defects with no orbital involvement; 7e. Type IIIB Total Maxillectomy Defects involving orbit; 7f.

		Bony defect		
		Anterior	Hemimandible	Lateral
Soft tissue defect	None	IA	IIA	IIIA
	Intraoral only	IB	IIB1 ≤ 2 zones IIB2 ≥ 3 zones	IIIB
	Skin only	IC	IIC	IIIC
	Intraoral + skin	ID	IID	IIID

Figure 2: Defects Mandible Classifications

3. Results

The results of collecting data on prevalence and characteristics of recurrent ameloblastoma in patients at the RSHS Oral Surgery KSM in the period January 2019 - January 2020 found 1 patient who received treatment. The results of data collection characteristics of ameloblastoma in patients at KSM Oral Surgery RSHS were as followed.

Figure 10 showed that there were 76 patients diagnosed with ameloblastoma in the period January 2019 - January 2020 at RSHS, divided into 24 males (32%) and 52 females (68%). In Figure 11, the data showed that the age of ameloblastoma patients is 41-50 years old, <41 years old and >50 years old, which are 10 patients (13.4%), 36 patients (47%) and 25 patients (33. %). In the 0-10 year age range, the minimum age that can be affected by ameloblastoma is 5 patients (6.6%). The incidence of ameloblastoma in Indonesia occurs at an average age of 37-50 years.

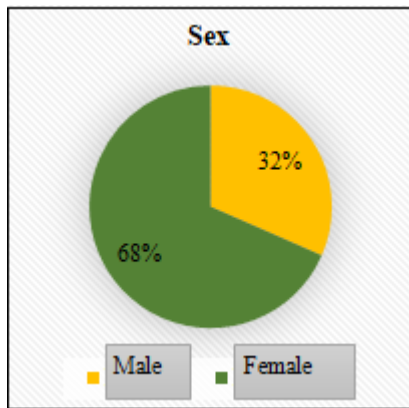


Figure 10: Sex Distribution of Ameloblastoma

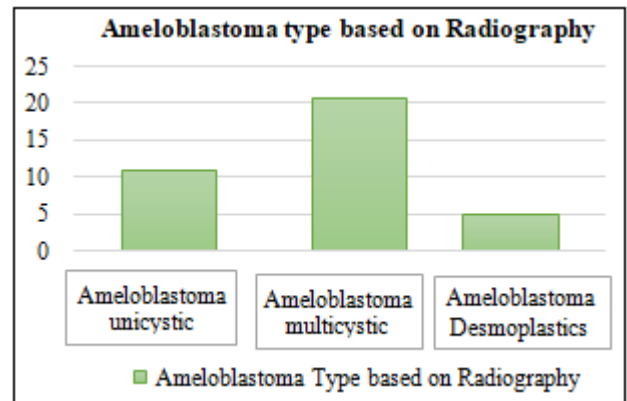


Figure 13: Ameloblastoma Type based on Pathology Anatomy

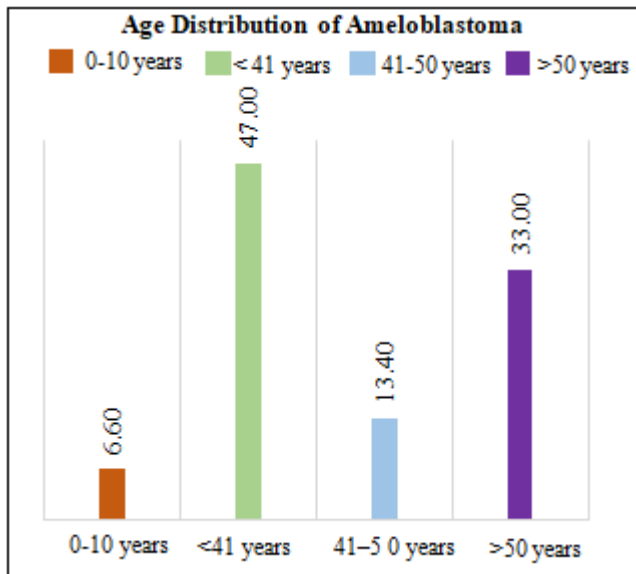


Figure 11: Age Distribution of Ameloblastoma

Based on radiographic examination and divided into 3, unicystic ameloblastoma, multicystic, and desmoplastic. Figure 12 showed that 11 patients (29.73%). Patients diagnosed from radiographic examination as multicystic ameloblastoma were 21 patients (56.76%). Patients diagnosed from radiographic examination as desmoplastic ameloblastoma were 5 (13.51%). Anatomical Pathology Examination (Figure 13) can be performed to support the diagnostic characteristics of ameloblastoma as in the data collected, 22 patients (59.46%), follicular ameloblastoma were found in 12 patients (32.43%), mixed ameloblastoma between plexiform and follicular ameloblastoma. as many as 3 patients (8.11%).

Ameloblastoma plexiform	Follicular	Mixed	Unspecified
33 Patient	22 patient	5	16

Figure 12: Ameloblastoma Type based on Radiography

Based on tumor location, found that 8 patients occurred in maxilla, and 66 patients occurred in mandible (Fig. 14). In the maxilla anterior, right, and left, there were 1 patient, 5 patients, and 2 patients, respectively. In the mandible, the majority of tumors occurred on the left as many as 34 patients, on the right as many as 18 patients, in the anterior as many as 5 patients.

Table 4.1: Predilection for Ameloblastoma

No.	Predilection	Number
1.	Palatum	2
2.	Anterior Maxilla	1
3.	Maxilla Dextra	5
4.	Mandibula anterior	5
5.	Mandibula dextra	18
6.	Unspecified Mandibula	9
7.	Mandibula sinistra	34
8.	Maxila sinistra	2

Management in cases of ameloblastoma divided into conservative or radical management. Figure 15 showed the conservative management of ameloblastoma with enucleation of 6 patients and the dredging method of 10 patients. Radical treatment in these data was performed by marginal resection in 1 patient (1%), segmental resection in 2 patients (3%), hemimaxillectomy in 2 patients (3%), mandibulectomy in 2 patients (3%), and hemimandibulectomy in 11 patients (14%).

Table 4.2: Ameloblastoma Management

No	Management	Number	Percentage (%)
1.	Dredging	10	13
2.	Segemental resection	2	3
3.	Marginal resection	1	1
4.	Hemimandibulectomi	11	14
5.	Hemimaxillektomi	2	3
6.	Unspecified resection	12	16
7.	Enukleated	6	8
8.	Mandibulectomi	2	3
9.	Biopsy	18	24
10.	Others	12	16

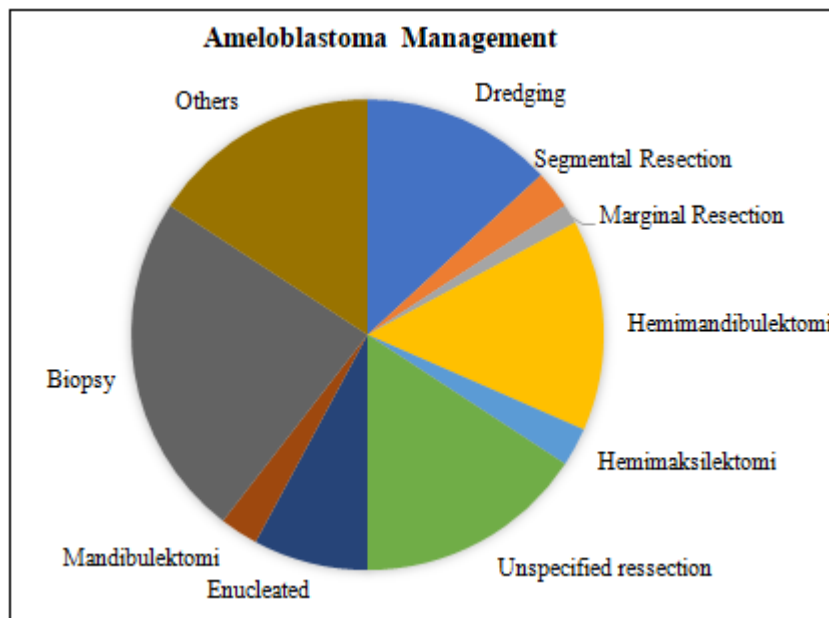


Figure 15: Ameloblastoma Management

Defects in the maxilla and mandible (figures 16 and 17) in maxillary ameloblastoma cases involved one or both maxillary walls and involved <50% of the maxilla (2 patients), did not cross the midline (1 patient) and did not leave the defect (1 patient), whereas in The mandible showed the most defects involving half of the mandible and there were extraoral and intraoral defects (13 patients).

No	Management	Numbers	Percentage (%)
1.	Plat AO Reconstruction	27	35
2.	Graft	4	5,3
3.	Protease	16	21

Figure 18: Reconstruction and Rehabilitation

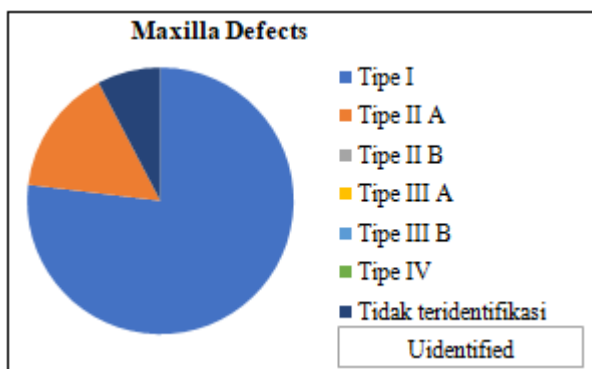


Figure 16: Maxilla Defects

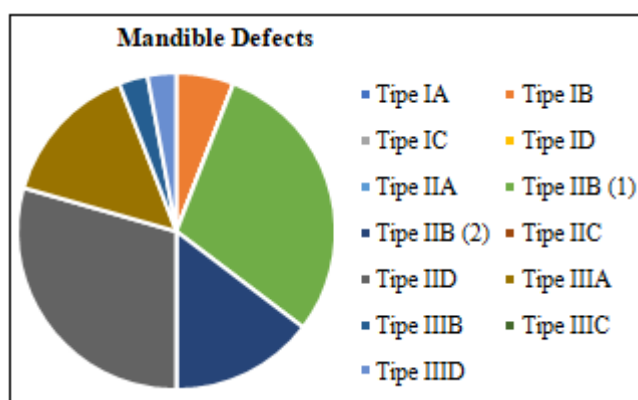


Figure 17: Mandible Defect

In the reconstruction and rehabilitation procedure, 27 patients (64.86%) had an AO plate inserted, in addition, 4 patients (10.81%) had grafts taken from the os costae, and 16 patients had prostheses made.

4. Discussion

Ameloblastoma was a benign tumor that grows slowly but locally invasive which is generally painless and can destruct bone. Characteristics of ameloblastoma based on radiographic examination and anatomical pathology are very varied, so analysis is needed to determine the characteristics of patients at KSM Oral Surgery RSHS. Based on the results of data collection, the incidence of ameloblastoma that occurred and was treated at the RSHS in the period January 2019 to January 2020 was 76 patients. Further investigation of 76 cases of ameloblastoma in the period January 2019 – January 2020 found 1 case of recurrent ameloblastoma. In the case of recurrence of ameloblastoma, marginal resection was carried out in the previous operation in 2020 and segmental resection was carried out in the next treatment in 2022. In the period January 2019 - January 2020, it was found that the average age of patients with ameloblastoma was 39 years. Based on research conducted by Ruslin on the average age distribution of ameloblastoma patients in the community in Makassar, Indonesia of 39.7 years, this strengthens the theory that the incidence of ameloblastoma was highest in the third to fourth decades.4 Ameloblastoma in a population is more common in women. compared to men. Based on primary data that has been obtained in patients diagnosed with ameloblastoma from a total of 76 patients, divided into 24 men (%) and 52 women (%).

In the case of recurrence ameloblastoma, a cystic type of ameloblastoma was found. The distribution of radiological examination of the results showed that the most diagnosed ameloblastoma was multicystic ameloblastoma by 56.76%, followed by unicystic ameloblastoma 29.73%, and desmoplastic ameloblastoma 13.51%. Research conducted by Gandhi et al which found that the most common in the

population was multicystic ameloblastoma as much as 77% and the remaining 23% were unicystic ameloblastoma, in addition, research from Saghravanian et al found 73% of cases of multicystic ameloblastoma that occurred in an Iranian population. Ruslin et al. also stated that in Eastern Indonesia the most cases of multicystic ameloblastoma were found by 57%, this indicates that the largest distribution of cases in the whole is multicystic ameloblastoma.

Other examinations carried out in establishing the diagnosis of ameloblastoma besides radiology was pathology anatomy examinations. The examination resulted that the cases that often occurred in patients treated at RSHS during the period January 2019 - January 2020 were the most cases of plexiform ameloblastoma, namely 22 cases, and followed by follicular ameloblastoma as many as 12 cases. Another case that occurred was mixed type ameloblastoma which is a combination of plexiform and follicular ameloblastoma, which occurred in 3 cases. Research by Rusdiana et al stated that the distribution of ameloblastoma in RSCM was 11 cases of follicular ameloblastoma, followed by 6 cases of ameloblastoma.

Management for ameloblastoma divided into conservative treatment and radical treatment. Recurrence after the first surgical treatment may occur from infiltrative growth of ameloblastoma through adjacent bone, which is responsible for local cancellous bone invasion beyond the radiographically visible margin. Mostly, recurrences are the result of inadequately performing the initial procedure. Hong et al (20), in a retrospective analysis of 239 patients with ameloblastoma, reported a recurrence rate of 4.5% after treatment with segmental resection or maxillectomy, 11.6% after marginal resection and 29.3% after conservative treatment (enucleation, curettage). and marsupialization), obtained a statistically significant correlation between the method of action and recurrence. In the case of ameloblastoma recurrence in the period January 2019 – January 2020, the initial surgical procedure performed was marginal resection. Radical and aggressive surgery were the preferred options for recurrent ameloblastoma management (4,7,21). This method supports that mandibular resection should be at least 1–2 cm beyond the radiological boundary to ensure that all microlesions are removed (7).

In another study, recurrent ameloblastoma was found in 12 patients, 6 of whom were male and 5 were female. The age range was 29 to 72 years, with a median age of 44.8 years. One tumor was identified in the anterior mandible (8%), 1 in the angle region, 1 in the ramus region, 3 in the mandibular body region (25%), and 6 in the angle and ramus region (67%). However, according to Ackermann et al. (24), aggressive surgery should be performed in some cases of unicystic ameloblastoma if the tumor is present in the connective tissue wall rather than the epithelium of the lesion, as this indicates infiltration. A radical approach is indicated for recurrent cases and larger ameloblastomas involving the inferior alveolar canal. The radical approach consists of segmental or marginal resection involving healthy bone 1.5–2 cm beyond the radiological margin (22, 23). The recurrence rate after conservative treatment is higher than after aggressive treatment (9, 10, 20). According to ... marginal resection was a treatment approach that

minimizes mandibular defects however, it can only be used in certain cases. Management of ameloblastoma with marginal resection has been reported to have a 15% recurrence rate.28-30

Based on the data that has been obtained at RSHS, the action distribution taken was marginal resection in 1 case, segmental resection in 2 cases, hemimandibulectomy in 11 cases, hemimaxillectomy in 2 cases, mandibulectomy in 2 cases. Conservative measures were taken at RSHS such as dredging in 10 cases and enucleation in 6 cases. Radical surgery was often used in RSHS because it aims to reduce the recurrence rate of ameloblastoma cases that occur, while conservative measures are indicated in pediatric and adolescent patients, as well as adult patients with unilocular ameloblastoma types.

Defects that often occur in the cases found in RSHS was occur in the mandible which provides defects in extraoral and intraoral, namely 27.03% of all ameloblastoma cases that occur in RSHS. According to research by Rameesh et al, it was found that in the Indian population, defects were found in the majority of the mandible more than 50%, and research by Tatapudi et al. stated that cases of ameloblastoma were more common and caused defects in the mandible by 80%. Based on these data, it can be associated that ameloblastoma defects are more common in the mandible

Reconstruction carried out in this case included AO plate placement, graft placement, and prosthesis fabrication. In the cases that occurred in RSHS reconstruction, the most cases were the installation of AO plates in 24 cases, making prostheses in 9 cases, and grafting in 4 cases. The choice of reconstruction is carried out based on the need and the extent of the defect that has been caused after surgery.

5. Summary

Research on the characteristics distribution and prevalence of recurrence rates in oral and maxillofacial surgery ameloblastoma patients at Hasan Sadikin Hospital for the period January 2019 – January 2020, it was found that ameloblastoma in female patients was more frequent than in men, with the percentage of incidence in women being 2% (52 patients) and the percentage of male incidence was % (24 patients). On further investigation of 76 cases of ameloblastoma in the period January 2019 – January 2020, 1 case of recurrent ameloblastoma was found. In the case of recurrence of ameloblastoma, marginal resection was performed in the previous surgery in 2020 and segmental resection was performed in the next treatment in 2022. Recurrence after the first surgical treatment may occur from infiltrative growth of ameloblastoma through adjacent bone, which is responsible for local cancellous bone invasion outside radiographically visible borders. For the most part, recurrences are the result of inadequate initial procedures

Marginal resection was treatment approach that minimizes mandibular defects however, it can only used in selected cases. Management of ameloblastoma with marginal resection has been reported to have a 15% recurrence rate.

The majority defects that occur in the mandible involve extraoral and intraoral tissues. A good reconstruction of the defect that occurs is by placing an AO plate, making prostheses, and installing grafts that function to restore the masticatory and aesthetic functions of the patient.

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