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Intraosseous Adenoid Cystic Carcinoma of Mandible: A Peculiar Case of Its Kind with Review of Literature

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Abstract: *Intraosseous adenoid cystic carcinoma is a rare variant of a malignant slow growing adenoid cystic carcinoma (ACC), which is most commonly found in major and minor salivary glands. Due to its subclinical nature to spread through perineural invasion and aggressive propensity for metastasis and recurrence, diagnosis of ACC is late and results in poor prognosis due extensive spread.. We have reviewed previously published reports regarding clinical behaviour and treatment outcome of intraosseous adenoid cystic carcinoma of mandible. In this report we present a case of a 65 year old male patient who was diagnosed with central ACC of mandible which was quite aggressive and had undergone radical treatment.*
Keywords: *intraosseous, adenoid cystic carcinoma, mandible, cylindroma, intraosseous salivary tumour*

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I. Introduction

Adenoid cystic carcinoma (ACC) is a malignant slow growing neoplasm, which shows tendency for perineural invasion beyond clinical palpable margins[1]. ACC is seen most commonly in 4-6th decade of life[2]. In head and neck region occurrence in major and minor salivary gland is more common, 6% of all tumors of salivary gland, accounts for 15-30% of submandibular gland ,2-15% of parotid and 30% of intra oral minor salivary gland tumors.[3] Intraorally there is a definite propensity for posteriolateral region of hard palate.[4] Other intraoral sites are cheek, floor of the mouth and tongue. Involvement of unusual sites includes external auditory canal, nasopharynx, lacrimal glands, breast, vulva, esophagus, cervix, and Cowper glands.[3]

An extremely rare clinical variant of ACC is central ACC with about on a few cases reported. Clinically central ACC presents with a bony swelling which is initially painless but later with nerve involvement may presents with pain. In late stages, lymph node involvement and distant systemic metastasis are common[2]. Also it has a propensity for perineural spread beyond clinically apparent borders.[5] Therefore, a wide excision of the lesion is treatment of choice. In cases of parotid gland involvement, facial nerve sacrifice is unavoidable.[2] In cases with positive surgical margins, radiotherapy has been an adjunct to surgical treatment with a necessary long term follow up[6]. Here we report a case of intraosseous adenoid cystic carcinoma of mandible without any distant metastasis, treated surgically and still under followup since 1 year.

II. Material And Methods

We did the literature search to analyse the clinical behavior and treatment outcome of intraosseous adenoid cystic carcinoma. For this different search data base like Cochrane, science direct, pubmed, ebesco, google scholar were gone through. Keywords used to potentiate our search were "intraosseous adenoid cystic", "cylindroma", "intraosseous salivary tumour". All the articles published in english were considered. Site specificity i.e primary involvement of mandible was kept in mind while searching for the data. Other facial sites involvement were excluded. Soft tissue lesions were also not included. Article search was confined to intraosseous adenoid cystic carcinoma. Other forms of salivary gland neoplasm were excluded. Due to retrospective nature of the study it was granted an exemption in writing by institutional review board.

III. Results

After considering all the decided exclusion and inclusion criteria 36 relevant articles were included for the review.(table 1) There were 32 case reports and 4 case series amounting to 50 cases of intraosseous adenoid cystic carcinoma of mandible. The age of the patients ranged from 23 to 83 years, mean age being 53. Maximum prevalence of this lesion was seen in fourth to fifth decade of life. There were 26 males and 24 females suggesting no significant gender predeliction. The ratio of male: female involvement noted was 1.08:1. Presence of lesion showed site variability. Body of mandible was the most common site of occurance (68%), in 8 cases lesion occurred on angle of mandible. Ascending ramus and anterior part of mandible was seen to be involved in 4 cases (8%) respectively. In 6 cases lesion involved both angle and body of mandible. In two cases lesion occurred in condyle and coronoid process of mandible. Clinical analysis of the disease revealed that pain and swelling was the most common chief complaint. 10 cases presented with paresthesia . other symptoms included trismus and tooth mobility. Otalgia was seen in 2 cases. Clinically, buccal and lingual cortical expansion was mentioned in 11 cases, cortical erosion was seen in 19 cases. Most of the lesions were initially diagnosed as odontogenic tumor or periapical lesion. Histopathological examination was a definitive diagnostic aid. Radiographically, lesion was seen as radiolucency of variable size in 47 cases(95%). Other reports did not describe radiologic appearance. Poorly defined radiolucency was seen in 29 cases (63%). In 3 cases lesion crossed the midline. Mainstay of the treatment was surgery and radiotherapy. 8 cases received both radiotherapy and chemotherapy. Lungs were most common site of metastasis, seen in 12 cases (24%) (table 1). Bone metastasis was seen in 2 cases. More than 5 years post treatment survival was seen in 7 cases. Metastasis to breast was seen in one case. Cervical node metastasis was seen in 5 cases(10%). Mean post treatment disease free duration noticed was 4 years. Maximum post treatment disease fee survival wasa seen for 14 years. Recurrence was reported in 5 cases (10%). Minimum time of relapse was 10 months and maximum was 12 years. Mean duration of recurrence noted is 5.6 years. All these inferences suggests the aggressive nature of intraosseous adenoid cystic carcinoma.

IV. Case Report

A 65 year old male reported with chief complain of swelling with lower jaw region since 4-5 weeks. Extra orally there was slight swelling seen with anterior part of the mandible which was non tender on palpation. Intraorally, there was expansion of buccal and lingual cortical plates bilaterally extending from third molar region right side to anterior border of ramus to the left side.(fig1) Swelling was bony hard, non compressible, non tender and no mobility with teeth was present. There was generalized attrition present and patient maintained poor oral hygiene with generalized gingival inflammation and calculus present. No lymphadenopathy was present and orthopantomograph was adviced to the patient.

A panoramic radiography revealed osteolytic changes in trabacular pattern, with irregular margins and a large radiolucent area, involving the body and ramus on right and left of the mandible. An incisional biopsy was performed which histomorphologically revealed to be ameloblastoma. A C.T. scan revealed an osteolytic space occupying lesion present in mandible extending bilaterally up to ramus region (fig 2). The patient was planned for surgery in our center under general anesthesia. Extended submandibular incision was taken. After exposing the mandible, demarcation of the lesion was noticed and total mandibulectomy was done leaving the condylar heads(fig 3). Suprahyoid muscles and submandibular gland was also excised along with the mandible primary reconstruction using a reconstruction titanium plate, which was fixed to condylar head with help to giving primary contour to the lower third of the face.(fig 5,6) Closure was done layers . Post operative recovery of the patient was uneventful.

The surgical specimen (fig 4) was sent for biopsy and histopathological report suggested malignant cells arranged in sheets, islands, ducts and tubular pattern with cyst like spaces noted and negative surgical margins confirmed the diagnosis as intraosseous adenoid cystic carcinoma. Post operative necessary clinical and radiological investigations were performed which were within normal limits. The patient is well, with no evidence of recurrence or distant metastases 9 month from the original diagnosis. Post operative chest X-ray and ultrasonography abdomen and CT was done to rule out metastasis. Patient has been referred for radiation therapy and still kept under observation and follow up.

V. Table Of Reported Cases In Literature

Cases Of Primary Adenoid Cystic Carcinoma Of The Mandible Published In The Literature

s.no.	Author	Year	Age (yr) /Gender	Location	Initial Diagnosis	Symptoms	Treatment	Observations
1	Bumsted[7]	1955	54/M	Body	Ameloblastoma	Pain, swelling	Surgery and radiotherapy	Lung metastases
2	Bradley[8]	1968	64/M	Angle	Muco-epidermoid carcinoma	Trismus, pain, swelling	Surgery	

3	Hámori and Krasznai[9]	1969	43/M	Body	Osteomyelitis	Otalgia	Surgery	Lung metastases
4	Shin et al[10]	1969	59/F	Angle	Fibroosseous lesion	Trismus, pain		
5	Dhawan et al[11]	1970	40/F	Ascending ramus	Malignant tumor	Swelling	Inoperable	Lung metastases
6	Slavin and Mitchell[12]	1971	35/M	Angle	Ameloblastoma	Swelling	Surgery	Lost to follow-up
7	Burkes[13]	1975	50/F	Body	Odontogenic infection	Pain	Surgery	Lung metastases
8	Yoshimura et al[14]	1978	47/F	Body	Cyst lesion	Dental loss, trigeminal paresthesia	Surgery and radiotherapy	Metastases in Gasser ganglion; 2 years and 7 months of disease free duration
9	Mushimoto et al[15]	1978	24/M	Body	Ameloblastoma	Swelling	Surgery	
10	Kaneda et al[16]	1982	47/M	Body	Spontaneous mandibular fracture, cyst lesion	Dental loss, swelling	Surgery	5 years disease free
11	Gingell and Siegel[17]	1983	72/F	Body	Dental irritation	Pain	Surgery	7 years disease free
12	Hirota and Osaki[18]	1989	82/M	Body	Malignant tumor	Swelling	Radiotherapy and chemotherapy	Died of pneumonia after end of treatment
13	Johnson et al[19]	1989	68/F	Ascending ramus	Ameloblastoma	Swelling	Surgery and radiotherapy	Relapse 4 years after end of treatment
14	Brookstone et al[20]	1990	33/F	Angle, body	Malignant tumor	Pain, trismus	Surgery and radiotherapy	One year disease free
15	Brookstone and Huvos[21]	1992	67/M	Body	No mention by author	No mention by author	Surgery	Neck metastases
16	Tarayre et al[22]	1996	83/M	Body	Spontaneous mandibular fracture, cyst lesion	Pain	Surgery	4 months disease free
17	Blanchard et al[23]	1998	49/M	Symphysis	Cyst lesion	Swelling, pain, dental mobility	Surgery, radiotherapy, and chemotherapy	Lung metastases
18	Favia et al[24]	2000	46/F	Body	Periapical lesion	Swelling, pain	Surgery	14 years disease free duration after treatment
19	Clark et al[25]	2000	54/M	Body	Periapical lesion	Swelling	Surgery and radiotherapy	4 years and 6 months disease free
20	Madrigal et al[26]	2000	51/M	Angle	No mention by author	No mention by author	Surgery and radiotherapy	Died after 4 yrs of disease
21	Madrigal et al[26]	2000	48/M	body	No mention by author	No mention by author	Surgery and radiotherapy	Died at 3 years
22	Madrigal et al	2000	24/F	angle	No mention by author	No mention by author	Surgery and radiotherapy	Live with disease for 7 years with pulmonary metastasis
23	Madrigal et al	2000	35/F	angle	No mention by author	No mention by author	Surgery and radiotherapy	Lived for 5 years
20	Chen et al[27]	2004	56/M	Body	Periapical lesion	Pain	Surgery	No mention by author
21	Capodife et al[28]	2005	36/f	angle	No mention by author	Pain swelling paresthesia	Radiotherapy and chemotherapy	3 year with disease and pulmonary metastasis
22	Gumgum et al[29]	2005	61/F	Angle	tumour	swelling	Surgery and radiotherapy	No mention by author
21	Al-Sukhun et al[2]	2006	80/F	Body	Periapical lesion	Pain	Surgery	3 years disease free
22	Shamim et al[30]	2008	45/M	Body and angle	Odontogenic tumor	Swelling and pain	Could not survive till treatment	Pulmonary metastasis
22	García de Marcos[31]	2008	57/F	Body	Periapical lesion	Dental nerve paresthesia	Surgery and radiotherapy	3 years disease free
23	Carlos bregni et al[32]	2009	46/M	Angle and body of mandible	Odontogenic tumor	Pain and paresthesia	surgery	Pulmonary metastasis

24	Ti kamitsu et al[33]	2010	68/M	Body and chin	No mention by author	Tooth mobility paresthesia pain and swelling	Surgery radiotherapy and chemotherapy	Pulmonary and bone metastasis
25	Santos TS et al[34]	2011	48/M	Body	No mention by author	Pain and swelling	Surgery radiotherapy and chemotherapy	No mention by author
26	Hofert S et al[35]	2012	45/F	Bilateral body and chin	Periapical lesion	Pain and paresthesia	Surgery radiotherapy and chemotherapy	Pulmonary breast metastasis
27	Vinuth DP et al[36]	2013	64/m	Anterior mandible	Malignant tumor	Pain and swelling	surgery	15 months no recurrence
28	Ren ZH et al[37]	2014	23/F	Body	Odontogenic tumor	Pain and swelling	Surgery and radiotherapy	10 months no recurrence
30	Deng et al[38]	2014	81/F	Body	No mention by author	Tooth ache	Partial mandibulec tomy	Recurrence after 2.5 yrs
			63/F	Anterior mandible (chin)	No mention by author	swelling	Surgery and radiotherapy	Recurrence after 12 years
			42/F	Anterior mandible (chin)	No mention by author	toothache	Surgery and radiotherapy	No disease after six months
			46/F	Body	No mention by author	swelling	Surgery and radiotherapy	6 yrs disease free
			74/F	Body	No mention by author	toothache	Surgery and radiotherapy	Death after 2 yrs
			66/M	Ramus	No mention by author	swelling	Surgery with radiotherapy	No evidence of disease 21 months
			65/M	Body	No mention by author	swelling	Surgery and radiotherapy	No evidence of disease 74 months
			28/F	body	No mention by author	paresthesia	Surgery and radiotherapy	No evidence of disease 42 months
31	Han et al[39]	2016	57/F	Ramus and body	osteosarcoma	Swelling following tooth extraction and paresthesia	Surgery chemotherapy and radio therapy	Lived with disease at 3 trs and pulmonary metastasis
			41/M	Body, Angle, ramus, coronoid process and condylar neck(R)	Malignant jaw tumor	Toothache swelling paresthesia	Refused for treatment	Bone and lymph node metastasis after one year
			58/M	body	Jaw centricity carcinoma	Pain paresthesia	Surgery and radiotherapy	No evidence of disease 5 months
			54/M	body	ameloblastoma	swelling	Surgery and radiotherapy	No evidence of disease 3 months
32	Hong ying et al [40]	2017	38/F	Body and ramus	Multiple neoplasm	pain	Surgery and radiotherapy	No evidence of disease 2 years
			52/M	Condyle glenoid fossa and external auditory canal	Malignant tumor of TMJ with pulmonary metastasis	Mass in left preauricular region, otalgia	chemotherapy	2 yrs with disease

VI. Discussion

In 1859, Theodor Bilroth first described ACC histologically and named it "cylindroma" based on long cylindrical compartments formed by epithelial and connective tissue elements.[4] Later, in 1908, it was named as "Basalioma" by Krompecher.[4] Till 1940s, adenoid cystic carcinoma was well thought to be a benign tumor , a variant of mixed salivary gland but, Dockerty and Mayo, in 1943 stressed on malignant nature of ACC.[2] In 1954, Ewing (Foote and Frazell) coined the term " Adenoid Cystic Carcinoma".[4] .

In 1963, bhaskar reported two cases and analyzed the critera for central salivary gland tumors about their origin histology and pathogenesis.[1]

The origin of central malignant salivary gland tumors is still controversial theories includes

1) Ectopic salivary glands tissue that was developmentally entrapped in the jaws as reported by Richard and ziskind. Bhaskar (1963) said that the source if origin in mandible is probably the mucus glandular inclusion in retromolar area4
2) Neoplastic transformation of the sinus epithelium
3) Neoplastic transformation of the epithelial lining of an odontogenic cysts1 (42%)2.

Central ACC is a rare variant of tumor with most common complain of pain and swelling. Most common site of involvement is posterior body angle region of mandible.[2]

It is difficult to discriminate whether the tumors in maxilla perforated the cortical plate or surrounding tissues invaded the osseous structure. This might be the reason of lesser incidences of intraosseous salivary gland tumors reported in maxilla than mandible.1 central ACC are slightly more frequent in maxilla where it is more difficult to be sure of their intrabony origin.[19] Imaging holds a very important part in surgical planning of tumors which spread subclinically. CT scans and MRI helps to detect the subclinical spread.[2]

In 1992, brookstone and Huvos established the staging system based on the destruction of the jaws :

1) Stage I - Lesions with intact cortical plate with no evidence of bone expansion
2) Stage II – tumors with intact cortical plate but intrabony expansion
3) Stage III – lesions associated with cortical perforation or nodal disease

A strict diagnostic criteria has been stated for intraosseous salivary gland tumors (Batsakis,1979) which includes:

a) presence of osteolytic areas on the radiographs
b) the cortical plates of the involved region should be intact.
c) intact mucous membrane overlying the lesion
d) Histomorphologic confirmation of adenoid cystic carcinoma
e) elimination of an odontogenic or another primary salivary tumor[19]

Again histologically ACC presents in three main variant namely[4]

1) cribriform variant – sheets, bands or nests of basal/ myoepithelial cells and round or oval intercellular spaces termed as pseudocyts giving a Swiss cheese appearance
2) tubular variant- presence of duct like structures formed by cuboidal and columnar cells.
3) Solid variant – shows a solid group of cuboidal cells, with little tendency towards ducts or cyst formation.

Grading of ACC has a prognostic importance. Histopathologically tumor is graded into 3 stages[4]

Grade1 – if there is presence of cribriform and tubular patterns

Grade2 - a mixture of all three patterns i.e. cribriform, tubular and solid, with solid variety less than 30% of tumor

Grade3 – predominance of solid variety is more than 30% of the tumor.

Surgery is the best treatment for ACC. However it is difficult to obtain complete resection because of vascular invasion and perineural infiltration.3 the surgical ablation must be individualized because radically multilating surgery does not appear to improve the prognosis in highly aggressive tumors.[7] Neck dissection shoud be preformed in patients with positive clinical or radiographic cervical lymphadenopathy. Prophylactic neck dissection is generally not recommenced.[7]

Radiotherapy and chemotherapy

ACC is a radiosensitive tumor but not radiocurable. Though postoperative radiation enhances the local and regional control in ACC. Wang recommended that for inoperable tumors, a dose of 65- 70 Gy to the primary lesion was indicated and for microscopic disease a dose of 55 -60 Gy was considered adequate. It seems to have a limited value in intraosseous salivary gland neoplasms, but in cases of perineural invasion radiotherapy was initiated.[1]

Ueta et.al. used adoptive immunotherapy combined with chemotherapy and observed disappearance of tumor cells and remodelling of sinus walls with calcifications in sinus cavity.[1]

The occurrence of distant metastasis is double where there has been inadequate surgery compared to radiotherapy alone according to smith, lane and rankowin 1965.[6]

Prognosis is better with low grade tumor without peripheral invasion and with tumor free margin.[1]

In nonresectable tumors radiotherapy may remain the only treatment modality.[6] Patients with grade 1 tumors show better survival than grade3, from this other factors like perineural spread, lymph node metastasis further influence the prognosis.[4]

Excision of tumor requires a widest margin possible because it extends beyond the clinical palpable margins and it not only invasive the perineural tissue but also spreads perineurally.[1] Distant metastasis and loco regional recurrences is indicative of undiagnosed long standing lesion.[4] Distant metastasis is common in solid variant.[4] Local recurrence is seen in cases of positive microscopic margins.[6] Primary site of tumor influences the prognosis , with tumors of the minor salivary gland showing a more aggressive behaviour. Occurrence of four or more symptoms at presentation, perineural invasion and lymph node metastasis are also the predictors of poor prognosis. About 40-60% of patients develop distant Metastasis to lungs, bone and soft tissue and occurrence of bone metastasis corresponds to rapid tumor dessimination and death whereas lung metastasis demonstrated less aggressive clinical course and are susceptible to remission after surgical resection[2].

As ACC shows extensive subclinical invasion and early metastasis, hence it reinforces on a fact that tumor growth rate and metastatic capabilities are independent of tumor properties. The capability of tumor is to cause perineural invasion but it is because of perineural spread of the tumor that causes extension of tumor well beyond the clinical or radiographic margins. Tumor size greater than 4cm indicates even greate subclinical spread and therefore is associated with a worse prognosis.[5]

Expression of proliferating cell nuclear antigen and Ki-67 is correlated with aggressive clinical behaviour and poor prognosis in ACC.3 Minor salivary gland tumos show worse prognosis than major salivary gland tumors as minor salivary gland can be more readily infilterate extraglandular soft tissues and bone allowing increased dissemination of the tumors.

Recent studies have shown expression of c-kit (CD-117), which is a receptor for tyrosine kinase. This receptor is involved in growth and development of normal tissues. Also chemotherapy has not been of very effective in ACC as the slow growing nature of the tumor.[6]

Unlike to other salivary gland tumors, which spread via lymphatic, ACC spread by heamtogenous route. metastasis to lymph nodes and lungs is characteristically common than to kidney which is quite rare.[5] Long term survival rates are low 10yrs- 20% and 15yrs -10%. Postoperative radiotherapy has shown to improve the long term survival rates.[4]

VII. Conclusion

In conclusion, in our patient all the above mentioned diagnostic criteria have been fulfilled and histopathologically presence of malignant cells arranged in sheets, islands, ducts and tubular pattern with cyst like spaces noted and a negative surgical margin. No metastasis was seen in chest x- ray and CT abdomen, indicative of a better prognosis based on clinicohistopathologic grading and the patient has been kept on long term follow up mandatory so as to rule out and late events of regional and distant metastasis.

Legends

FIG 1 :- clinical image showing buccal and lingual cortical expansion.
Fig 2 :- pre operative NCCT image showing osteolytic lesion showing buccal and cortical expansion.
Fig 3:- mandibulectomy done leaving behind bilateral condylar head.
Fig 4:- surgical specimen sent for histopathological examination.
Fig 5:- titanium reconstruction plate fixed to the surgical defect.
Fig 6:- post operative radiograph showing contoured reconstruction plate

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FIG 1 :- clinical image showing buccal and lingual cortical expansion

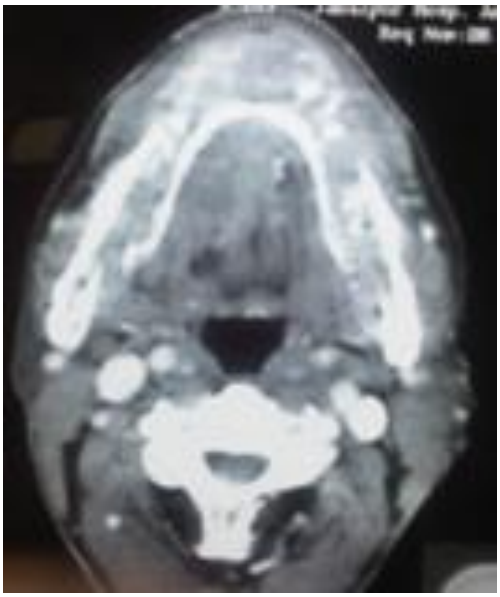

Fig 2 :- pre operative NCCT image showing osteolytic lesion showing buccal and cortical expansion

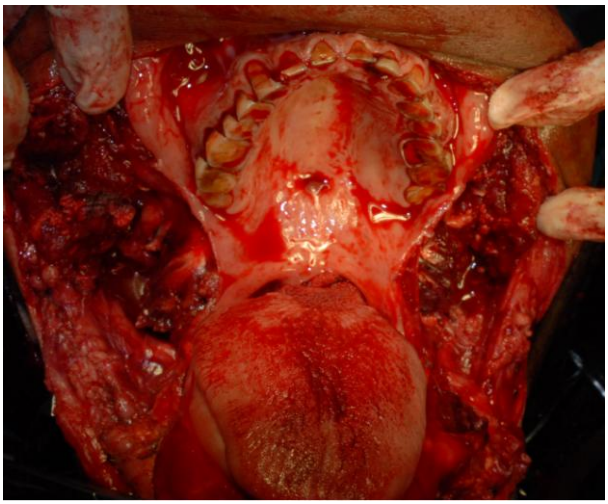

Fig 3:- mandibulectomy done leaving behind bilateral condylar head.

Fig 4:- surgical specimen sent for histopathological examination

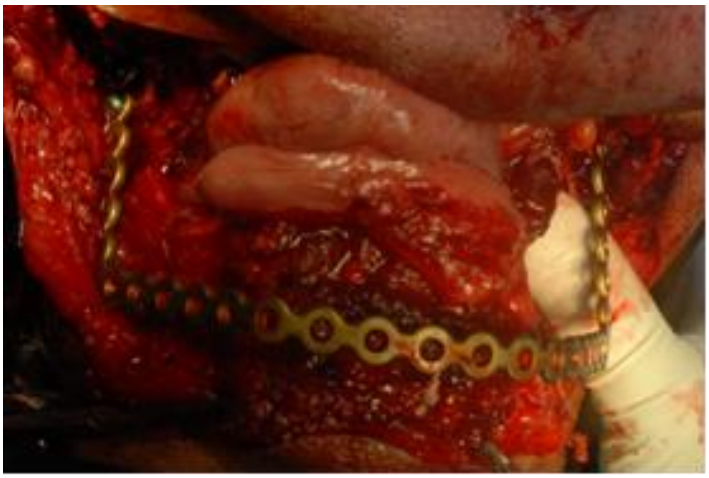

Fig 5:- titanium reconstruction plate fixed to the surgical defect.

Fig 6:- post operative radiograph showing contoured reconstruction plate

Dr. Ankita Saxena MDS. "Intraosseous Adenoid Cystic Carcinoma of Mandible: A Peculiar Case of Its Kind with Review of Literature." IOSR Journal of Dental and Medical Sciences (IOSR-JDMS), vol. 18, no. 8, 2019, pp 75-83.