

Ameloblastic carcinoma: A clinicopathologic dilemma – Report of two cases with total review of literature from 1984 to 2012

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ABSTRACT

Ameloblastic carcinoma (AC) is a rare primary odontogenic tumor that has histological features of both ameloblastoma and carcinoma. A total number of 92 case reports speak about its rare incidence, affecting mostly the mandible as a locally destructive lesion. The maxilla is affected even more rarely as only 35 cases have been reported until 2012 in scientific literature. The clinical course of AC is generally aggressive, with extensive local bone destruction. The most common clinical features include swelling, pain, trismus, significant bone resorption with tooth mobility, dysphonia and intraoral fistula. We report two cases of AC with aggressive behavior.

Keywords: Ameloblastoma, ameloblastic carcinoma, odontogenic carcinoma

INTRODUCTION

Ameloblastic carcinoma (AC) is characterized by malignant cytological features in combination with the overall histological pattern of ameloblastoma. The term malignant ameloblastoma is confined to those ameloblastomas that metastasize despite an apparently typical benign histology in both the primary and the metastatic lesions.^[1] The incidence ratio of AC to malignant ameloblastoma is 2:1.^[2]

In this article, we report two cases of AC, one involving left maxilla (primary type), which would be an addition to existing 35 cases previously reported and the other one affecting the left side of mandible arising ex ameloblastoma.

CASE REPORTS

Case 1

This paper reports a case of a 21-year-old male patient who reported to our department with the complaint of a painless, rapidly growing swelling, over the left side of mid-face since

4 months [Figure 1]. He had difficulty in chewing on the left side and three episodes of epistaxis for the last 1 month. On extraoral examination, a firm diffuse, non-tender swelling with normal overlying skin over a left side of mid face measuring 5 cm × 6 cm × 4 cm was observed. The swelling was seen extending superiorly from left infraorbital region inferiorly up to 1 cm below the commissure, medially from the philtrum region obliterating the nasolabial fold and extending laterally up to 3 cm anterior to the left ear lobule. Intraorally, a firm swelling involving left maxilla extending from distal aspect left maxillary central incisor up to the second molar region with more buccal cortical expansion, obliterating left buccal vestibule and displacing lateral incisor, canine, premolars and maxillary first molar palatally, but without any mobility was observed. All associated teeth were vital. Swelling was negative on aspiration. On neck examination, no cervical lymphadenopathy was found. Orthopantomograph showed a unilocular radiolucent lesion affecting the left maxilla displacing lateral incisor, canine, both premolars and molars [Figure 2]. The patient's vital signs were as Follows: Heart rate – 76/min, blood pressure – 126/78 mm Hg and respiratory rate – 14/min. His medical condition was not

suggestive of any systemic diseases and the biochemical and hematological investigations were all within the normal limits.

On performing incisional biopsy, it was found that the outer cortical plate was lost, the growth being attached to the mucoperiosteum. The gross appearance of the specimen was slimy in character. The initial histopathology report was that of follicular variant of ameloblastoma with a suspicion of AC.

The patient was planned for wide resection of tumor with normal margins through the modified Weber Ferguson approach under general anesthesia [Figure 3]. Resection was done with adequate



Figure 1: Preoperative photograph of case 1 showing expansile swelling over the left anterior maxilla



Figure 3: Intraoperative photograph showing the tumor involving the left maxilla exposed via modified Weber-Ferguson incision

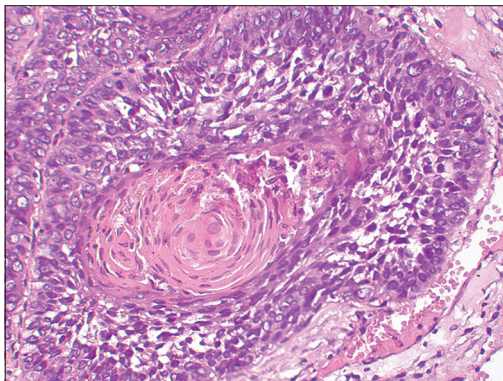


Figure 5: Photomicrograph showing odontogenic epithelial island with peripheral ameloblast-like cells (pre-ameloblasts) and centrally stellate-reticulum like cells. Also seen is acanthomatous change (squamous metaplasia with keratin pearl formation) within the follicle, which also shows comedo necrosis (H and E, ×40 objective)

margins, orbital floor was spared and proper hemostasis achieved. The surgical procedure was uneventful. The defect was closed with a surgical obturator. The resected specimen was sent for histopathologic examination [Figure 4]. The histopathological study revealed that the tumor was composed of follicles of odontogenic epithelial islands in a dense, mature connective stroma. Islands resembled dental organs with peripheral layer of ameloblast-like cells and centrally stellate reticulum-like cells. Furthermore seen was acanthomatous change (squamous metaplasia with keratin pearl formation) within the follicle, which also showed comedo necrosis [Figure 5]. These cells showed nuclear hyperchromatism, nuclear pleomorphism, increased nuclear-cytoplasmic ratio and mitotic figures or abnormal mitosis [Figure 6]. The histopathological examination also showed clear cells and fascicles of plump spindle cells merging with



Figure 2: Orthopantomograph showing a well-defined unilocular radiolucent lesion affecting left maxilla with multiple displaced teeth

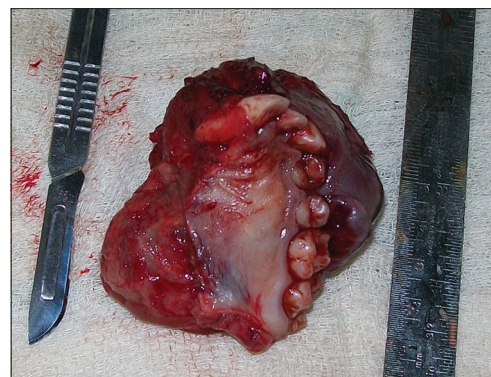


Figure 4: Resected tumor after sub-total maxillectomy

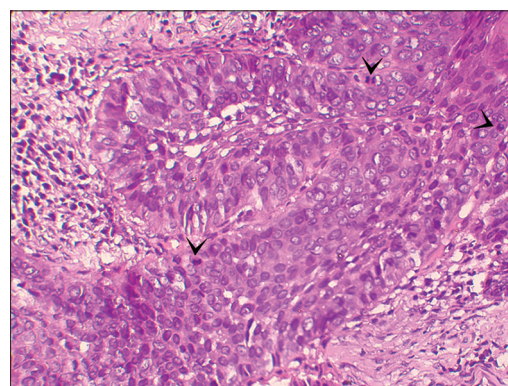


Figure 6: Photomicrograph showing nuclear hyperchromatism and pleomorphism. Arrowheads point out mitotic figures (H and E, ×40 objective)

other cells of the follicular ameloblastomatous epithelium. This confirmed the diagnosis as AC (spindle cell variant). The patient was then referred to radiotherapy department, where he received 25 fractions of 1.8 Gy each for a period of 5 weeks (45 Gy). Post-radiation mucositis was reported.

The patient was regularly followed-up for 1 month interval for a period of 6 months. To rule out distant metastasis, he was sent to the Department of Pulmonary Medicine and Gastroenterology of this institution, but they have cleared for any distant metastasis. Presently, after the 15 month postoperative period, there has been no recurrence at the primary site and no distant metastasis or regional lymphadenopathy has been observed. Patient is regular to his follow-up and maintaining his oral hygiene [Figure 7].

Case 2

This was a case of a 70-year-old male patient who reported to our department with the complaint of rapidly growing painless swelling of the left side of lower jaw with difficulty in chewing, multiple mobile teeth over left side of both jaws and progressive loss of teeth for last 6 months [Figure 8]. He had the past history of a similar lesion over the left lower jaw, which was treated by en-bloc resection 20 years back. The histopathology then was suggestive of ameloblastoma.

On extraoral examination, a firm diffuse non-tender swelling over left side of face extended superiorly from the left infraorbital region to the lower border of mandible inferiorly. Medially, it extended up to one-third of both upper and lower lips with an elevated angle of mouth and laterally 1 inch anterior to the angle of mandible with more medio-lateral expansion measuring about 10 cm × 8 cm × 6 cm. Neither paresthesia was associated with the swelling nor was any regional lymph node palpable. Intraoral examination revealed a painless firm proliferative growth from midline up to the left anterior border of ramus with an obliterated left side buccal sulcus and extended lingually with expansion of the lingual cortex and a medially displaced tongue [Figure 9]. The swelling was negative on aspiration. No cervical lymphadenopathy was found. Orthopantomogram showed a multilocular radiolucent lesion involving left angle and ramus of mandible with multiple areas of resorption over anterior border of ramus and maxillary alveolus distal to premolar region [Figure 10].

His medical history was suggestive of Type 2 diabetes mellitus, under irregular medication. His vitals were as follows: Blood pressure – 148/96 mm Hg, heart rate – 76/min, respiratory rate – 14/min and afebrile. Fasting blood glucose (FBG) came out with 202 mg/dl. Endocrinology consultation was done and he was advised injection Human insulin 28 U in three divided doses 6/8/14 U subcutaneously. After 3 days, the FBG level reported to be 92 mg/dl. The patient was then planned for an incisional biopsy under the local anesthesia. It was suggestive of follicular variant of ameloblastoma. He was then taken up for surgical resection of the lesion with 2 cm of bony margins on the symphysis region. After hemi-mandibulectomy, the defect was reconstructed with the placement of 2.4 mm titanium reconstruction plate with condyle and surgical wound was closed primarily in layers [Figure 11]. Postoperative recovery was uneventful and the surgical wound healed well. The resected

specimen was sent for histopathologic examination [Figure 12]. To our surprise, the histopathology report came out with diagnosis of AC. He was sent to radiotherapy department for further intervention, but he refused to undergo it.

After about 8 months, the patient again turned up with a firm, tender swelling over the left side mid and lower face extending to the neck [Figure 13]. The computed tomography (CT) scan showed a hypodense image of approximately 12 cm × 16 cm size present over left side mandibular and submandibular region. He was planned for resection of tumor under general anesthesia. The submandibular incision was given and the tumor appeared firm in consistency, surrounding the reconstruction plate, it was in close approximation to the great vessels and while removing it, the left internal carotid artery got injured; the vessel was clamped and repaired with 6-0 prolene suture. The tumor was removed with normal margins along with the reconstruction plate. Hemostasis achieved and closure done in layers. The postoperative recovery was delayed, with unstable blood pressure. Patient was shifted to intensive care unit and placed on ventilator support. The next day right side hemiparesis of whole body was detected and the CT scan showed left parietal infarct. Serum urea and creatinine were also marked elevated. After 3 days, the patient died due to multiorgan failure followed by cardiorespiratory arrest.

DISCUSSION

Odontogenic tumors are rare tumors of tooth forming apparatus involving jaw bones. In North America, between 11% and 24% of odontogenic tumors are ameloblastomas,^[2] while among sub-Saharan Africans it accounts for 66-99%.^[3] The tumor is frequently quoted as comprising 1% of all oral tumors among Caucasians,^[4] approximately 80% are found in the mandible and 20% in the maxilla. Mostly it is asymptomatic and the symptoms appear with the expansion of the jaw. In 1955, Small and Waldron showed that 47% of maxillary ameloblastomas occur in the molar region, 33% occur in the antrum and floor of the nose, 9% occur in both the premolar and the canine regions and 2% occur in the palate.^[5] There are three variants of this tumor, including the solid or multicystic variant, the unicystic variant and the peripheral variant. A review of the international literature by Reichart *et al.* in 1995 found the solid or multicystic variant to be the most common, comprising 92% of the 3677 cases of ameloblastoma, while the unicystic and peripheral variants accounted for 6% and 2% of the cases, respectively.^[6]

AC is an extremely rare malignant odontogenic epithelial neoplasm that may arise *de novo* or from a pre-existing odontogenic lesion.^[1,7] There are about 92 cases reported from 1984 to 2012 in scientific literature. The age of presentation ranges from 7 to 91 years. Males are more frequently affected with M:F ratio of 2.3:1. The most common site is mandible with about 56 reported cases, 35 cases in maxilla and in only one case arising from anterior skull base.^[8] Posterior portion of jaw is more commonly involved. Clinically, AC causes expansion of the jaw, grows rapidly, frequently causes pain and often results in perforation of the cortex. Involvement of the nasal cavity is usually related to local invasion of the maxillary ACs. Although regional and distant metastasis is the feature



Figure 7: Postoperative photograph after 6 months



Figure 8: Preoperative photograph of Case 2 showing swelling over left mid and lower face



Figure 9: Case 2: Intraoral view showing the swelling involving left mandibular alveolus, obliterating the vestibule with buccal cortical expansion



Figure 10: Orthopantomograph showing multiple ill-defined radiolucencies over the angle and ramus region of left side of mandible with displaced adjacent teeth

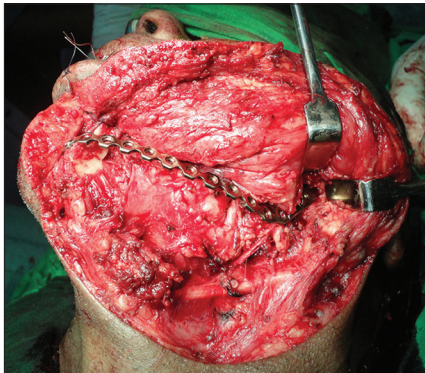


Figure 11: Operated site after the resection of tumour (hemimandibulectomy) done followed by placement of 2.4 mm titanium reconstruction plate

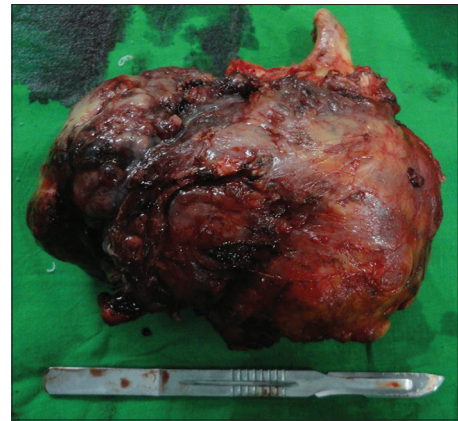


Figure 12: Resected hemi-mandible with 2 cm margin



Figure 13: Case 2: Recurrence with marked swelling over left side lower face extending to the neck after 8 months of surgery

of malignant ameloblastoma, but in few cases of AC, these lesions have been known to metastasize mostly to the lung or regional lymph nodes.^[9] Few cases reported with metastasis to the brain, bone marrow and liver.^[10-14] MacIntosh pointed out that the first site of reported ameloblastic metastasis was the lung and originally believed to be due to aspiration from the oral lesion rather than a true hematogenous or lymphatic spread. Indeed, enucleation and curettage surgeries might liberate neoplastic cells into the upper airway that could find their way into the lower airway.^[15] Zwahlen *et al.* reported a case with histologically proven myocardial metastasis of a maxillary malignant ameloblastoma.^[16]

The pathogenesis for malignant transformation of ameloblastoma may occur either spontaneously or because of induction following chemotherapy or post-surgical radiation.^[6,17] Aggressive behavior and metastatic potential associated with ameloblastoma has been noted in two of its variants – one is the granular cell type and the other clear cell type.^[18,19] Pulmonary metastasis was reported in two of the 20 granular cell ameloblastomas reviewed by Hartman.^[20] Furthermore, the cases of malignant ameloblastoma reported by Tsukada *et al.*,^[21] Hoke and Harrelson^[22] were of granular cell type.

Histologically it shows features of ameloblastoma with cytologic atypia, high mitotic index, reverse polarization, peripheral palisading and necrosis, neural and vascular invasion. There are various tumors which may mimic AC histologically or clinically which includes primary intra-alveolar epidermoid carcinoma,^[23] squamous cell carcinoma arising in the lining of an odontogenic cyst,^[24] acanthomatous ameloblastoma and kerato-ameloblastoma,^[25] squamous odontogenic tumor,^[26] calcifying epithelial odontogenic tumor, salivary gland neoplasms such as pseudoadamantine adenocarcinoma, ductal carcinoma, high-grade mucoepidermoid carcinoma and metastatic carcinoma to the jaws from lung, breast and gastrointestinal tract. Yoon *et al.* compared the immunohistochemical markers and found that the significant expression of cytokeratin 18, parenchymal matrix metalloproteinases-2 (MMP-2), stromal MMP-9 and Ki-67 differentiated AC from ameloblastoma.^[27]

The management of the AC is similar to that for ameloblastoma along with radiotherapy as reported by various authors. Literature also suggests only few case reports in which neck was addressed. The neck dissection was not performed in both the cases because no neck nodes were detected on clinical examination. Addressing no neck dissection in cases of squamous cell carcinoma is still in controversy. As our diagnosis was AC with no palpable neck nodes, we have not addressed the neck. Prior to the mid-1980s,

the literature was lacking with well-documented evidence regarding the relative radioresponsiveness of the ameloblastoma. Until this point, it was believed that ameloblastoma was radioresistant. In 1984, Atkinson *et al.* published their review of 10 patients undergoing the delivery of megavoltage irradiation. Nine of the patients responded, three of whom underwent surgical salvage. Of the 10 patients, seven showed no evidence of disease after surgery and/or radiation therapy, with follow-up ranging from 1 to 10 years.^[28] Additional evidence suggesting the value of radiation therapy in treating the ameloblastoma has been reported by Gardner.^[29]

Radiotherapy and chemotherapy seem to have limited value in AC and although primary radiotherapy was expected to be useful in the cases with perineural or massive soft-tissue invasion and in positive surgical margins.^[13,30] Dhir *et al.* reviewed 18 patients with maxillary AC. In 11 of 18, radiotherapy was used either as primary or secondary treatment in the case of metastasis and/or recurrence.^[30] Jensen *et al.* in 2011 introduced carbon ion therapy for multiple recurring AC.^[31] Horváth *et al.* reported a case of mandibular AC with pleura-pulmonary and bone marrow metastasis in a 8-year-old girl. They prescribed five cycles of chemotherapy with vincristine, endoxane, adriamycin, carboplatin and etoposide. It resulted in tumor shrinkage but no significant effect upon pulmonary site and the patient died after 8 months.^[32] Hence, the role of chemotherapy is still unpredictable.

Survival rate depends upon local recurrence, regional or distant metastasis. Infante-Cossio *et al.* reported a maximum 5 years of follow-up in two patients with no evidence of local recurrence, regional or distant metastasis. Third patient showed local recurrence and he died after few months of it. Hence, after surgery and radiotherapy, 5 years of survival rate can be accepted as a standard if no recurrence or metastasis occurs.^[11] Case reports of AC affecting both the jaws, which have been published at the time of writing this article, have been listed in Table 1.

Table 1: Total review of literature of AC from 1984 to 2012

Author/year	Site of involvement	Age/sex	Treatment
Slootweg and Muller ^[1] (Oral Surg Oral Med Oral Pathol, 1984)	Mandible	23/F	Surgery + RT
	Mandible	75/M	Surgery + CT
Andersen and Bang ^[33] (J Max Fac Surg, 1986)	Maxilla	77/M	Surgery
Nadimi <i>et al.</i> ^[34] (J Oral Med, 1986)	Maxilla	15/F	Surgery
Corio <i>et al.</i> ^[35] (Triple O-1987)	8 cases (7 in mandible, 1 in maxilla)	15-84	Surgery
		M: F-1:1	
Donner <i>et al.</i> ^[36] (BJOMS, 1988)	Mandible with pulmonary metastasis	81/M	Surgery
McClatchey <i>et al.</i> ^[37] (J Otolaryngol 1989)	Maxilla, peripheral type	77/F	Surgery
Lee <i>et al.</i> ^[38] (J Cran Max Fac Surg 1990)	AC of maxilla, metastatic to the mandible	56/M	Surgery + RT
Bruce and Jackson ^[39] (J Cran Max Fac Surg 1991)	Mandible	57/M	Surgery + RT
Nagai <i>et al.</i> ^[40] (J Oral Pathol Med, 1991)	Mandible	50/M	Surgery
Gandy <i>et al.</i> ^[41] (JOMS, 1992)	Mandible	20/M	Surgery
		32/F	Surgery + CT
Lolachi <i>et al.</i> ^[42] (JLO, 1995)	Maxilla	82/F	Surgery
Ingram <i>et al.</i> ^[43] (Diagn Cytopathol 1996)	Maxilla	83/M	Surgery + RT
Simko <i>et al.</i> ^[10] (Head Neck, 1998)	Mandible	64/F	Surgery + RT
Infante-Cossio <i>et al.</i> ^[11] (J Cran Max Fac Surg 1998)	Maxilla	69/F	Surgery + RT
	Maxilla	77/M	Surgery + RT
	Maxilla	64/M	Surgery + RT
	Mandible	73/M	Surgery
Lau <i>et al.</i> ^[44] (O000E, 1998)	Mandible	23/M	Surgery
		59/M	Surgery
Khoo and Ong ^[45] (Annals Dent Univ Malaya, 1998)	Mandible with cervical node and pulmonary metastasis	25/M	Surgery
Cox <i>et al.</i> ^[46] (O000E, 2000)	AC ex ameloblastoma of mandible		Surgery

Contd...

Table 1: Continued

Author/year	Site of involvement	Age/sex	Treatment
Camelio <i>et al.</i> ^[47] (Ind J Dent Res, 2001)	Mandible	65/M	Surgery
Sastre <i>et al.</i> ^[48] (JOMS,2002)	Maxilla	40/M	Surgery
Avon <i>et al.</i> ^[49] (J Can Dent Res, 2003)	Maxilla	68/M	Surgery
Datta <i>et al.</i> ^[12] (Am J Otolaryngol, 2003)	Mandible, with multiple bony metastasis	22/M	Surgery+CT+RT
Oginni <i>et al.</i> ^[50] (Odontostomatol Trop. 2003)	Maxilla	61/F	Surgery
	Mandible	23/M	Surgery
	Mandible	65/M	Surgery+RT
Dhir <i>et al.</i> ^[30] (Oral Oncol, 2003)	Maxilla	72/M	Surgery+RT
Cizmecy <i>et al.</i> ^[7] (Otolaryngol Head Neck Surg, 2004)	AC ex ameloblastoma of mandible	44/F	Surgery+RT
Carinci <i>et al.</i> ^[51] (J Craniofac Surg 2004)	Mandible	81/M	Surgery
Goldenberg <i>et al.</i> ^[13] (Laryngoscope 2004)	Mandible with brain metastasis	60/F	Surgery+RT
	Maxilla	72/M	Surgery+RT
Uzum <i>et al.</i> ^[52] (J Oral Pathol Med 2005)	Mandible	66/M	Surgery
Ozlugedik <i>et al.</i> ^[8] (skull base 2005)	Anterior skull base	23/F	Surgery+RT
Arotiba <i>et al.</i> ^[53] (Nigerian J Surg Res 2005)	Mandible	52/M	Surgery
Suomalainen <i>et al.</i> ^[54] (O000E 2006)	Mandible	21/F	Surgery
Miyake <i>et al.</i> ^[55] (Pathol Int 2006)	Mandible	91/F	Surgery
Akrish <i>et al.</i> ^[56] (JOMS, 2007)	Mandible	80/M	Surgery
Ward <i>et al.</i> ^[57] (JOMS, 2007)	Maxilla	62/M	Surgery
Hall <i>et al.</i> ^[14] (O000E, 2007)	Reported 14 new cases, 8 in mandible and 6 in maxilla	7-75 years/ M: F-2.5:1	Surgery+RT CT (single case)
Adil <i>et al.</i> ^[58] (O000E, 2007)	Maxilla	90/M	Surgery
Naik and Kale ^[59] (Quintessence int 2007)	Maxilla	70/M	Surgery
Angiero <i>et al.</i> ^[60] (Anticancer Res, 2008)	Maxillary sinus	68/M	Surgery
Yazici <i>et al.</i> ^[61] (Pediatri Blood Cancer 2008)	Maxilla	10/M	Surgery+RT
Yoon <i>et al.</i> ^[62] (O000E, 2009)	Reported 6 new cases, 2 in mandible and 4 in maxilla	46-73 years/ M: F-5:1	Surgery+RT
Lucca <i>et al.</i> ^[63] (J Oral Maxillofac Surg 2010)	Maxilla (post)	73/M	No treatment
	Maxilla (post)	69/M	Surgery
Ram <i>et al.</i> ^[64] (J Maxfac Oral Surg, 2010)	Mandible	21/M	Surgery
Karakida <i>et al.</i> ^[65] (O000E, 2010)	AC ex ameloblastoma of mandible	43/M	Surgery
Jindal <i>et al.</i> ^[66] (Curr Oncol 2010)	Mandible (spindle cell variant)	60/M	Surgery
Roy Chowdhury <i>et al.</i> ^[67] (J Maxfac Oral Surg, 2010)	Mandible	67/M	Surgery
Jeremic <i>et al.</i> ^[68] (J Craniomaxillofac Surg, 2010)	Mandible	58/M	Surgery
Nicolotti <i>et al.</i> ^[69] (J Craniofac Surg, 2011)	Maxilla	77/M	Surgery
Maya <i>et al.</i> ^[70] (OMPJ 2011)	Reported 5 new cases-4 in mandible and 1 in maxilla	25-40 years/ M: F-2:3	Surgery
Pundir <i>et al.</i> ^[71] (J Oral Maxfac Pathol, 2011)	AC ex ameloblastoma of mandible	40/F	Surgery
Maheshwari <i>et al.</i> ^[72] (BMJ Case Rep 2011)	Mandible	35/M	Surgery+RT
Nai and Grosso ^[73] (Braz Dent J, 2011)	Mandible	74/F	Chemotherapy
Matsuzaki <i>et al.</i> ^[74] (O000E, 2011)	Maxilla (post)	73/F	Surgery
Horváth <i>et al.</i> ^[32] (Rom J Morphol Embryol., 2012)	Mandible, metastasis to lungs and bone marrow	8/F	Chemotherapy
Pirklbauer <i>et al.</i> ^[75] (J Craniomaxillofac Surg, 2012)	Mandible	86/M	Palliative RT
Casaroto <i>et al.</i> ^[76] (Anticancer Res 2012)	Mandible	68/M	Surgery
Routray and Majumdar ^[77] (J Oral Maxfac Pathol, 2012)	Mandible	60/M	Surgery
Present study	Maxilla	21/M	Surgery+RT
	Mandible ex ameloblastoma	70/M	Surgery

AC = Ameloblastic carcinoma, RT = Radiotherapy, CT = Chemotherapy, F = Female, M = Male

CONCLUSION

Based on the above study although, ameloblastoma is a slowly growing, locally aggressive tumor. Any change in its growth pattern should cause suspicion of malignancy. AC is likely to metastasize. Therefore, complete history and general systemic examination are required to rule out any distant metastasis. Radiotherapy has definite role in cure of AC as in our cases; recurrence was seen in the patient who refused to undergo it.

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