Open Journal of
Clinical & Medical
Case Reports

Volume 3 (2017)
Issue 4

ISSN 2379-1039

Ameloblastic fibrosarcoma in a 10-year-old girl: Literature review and a rare case report

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Abstract

Ameloblastic fibrosarcoma is a rare malignant odontogenic tumour consisting of a benign epithelial and malignant mesenchymal component. It is regarded as the malignant counterpart of the benign ameloblasticfibroma with the epithelial component remaining benign, but the mesenchymal component becoming malignant. The diagnosis is made by histopathological and immunohistochemical evaluation, since the epithelial component remains benign and the mesenchymal component becomes malignant. The treatment is still controversial, without a definition regarding chemotherapy and radiotherapy as coadjutant treatment. Here, we present a case of ameloblastic fibrosarcoma in a 10-year-old girl.

Keywords

ameloblastic fibrosarcoma; odontogenic; mandible

Introduction

Ameloblastic fibrosarcoma (AFS) is a rare odontogenic neoplasm exhibiting a benign ameloblastomatous epithelial component admixed within a sarcomatous mesenchyme[1]. AFS is composed of proliferating odontogenic epithelium resembling of ameloblastoma embedded in a cellular mesenchymal tissue resembling dental papilla but without the development of odontoblasts having features of sarcomalike of fibrosarcoma[2]. Due to rarity of this neoplasm little is known about its molecular pathogenesis. AFS is considered to arise either as a de-novo lesion in the jaw or malignant transformation of pre-existing ameloblastic fibroma (AF) [3,4].To our knowledge 114 well-documented cases have been published in the literature (Table I) [1-87]. We report a rare case of AFS arising de-novo in a 10-year-old girl.

Case Report

A 10-year-old girl was referred to the Department of Oral Pathology, with a rapidly increasing painful mandibular swelling of 5 months duration. Extra-oral examination revealed a bony hard painful swelling, extending from the right parasymphyseal to left parasymphyseal area, measuring about 4x 5 cm in size. The overlying skin was erythematous, taut and intact with no sign of sinus formation. There was no palpable cervical lymphadenopathy or paraesthesia of the lower lip. Intra-oral examination disclosed a soft erythematous ulcerated mass, with focal areas showing features of necrosis and had bad odour, extending from deciduous mandibular left first molar to deciduous mandibular right canine. There was

There was bicortical expansion with displacement of the mandibular incisors (Figure. 1A). Patient's past medical and family history was negative Orthopantomogram (OPG) revealed a large ill-defined radiolucency involving the mandibular symphyseal region with floating teeth appearances of 31, 32, 41, 42 (Figure.1B). Computerized tomographic scan of the mandible showed a 5.2 X 3.5 cm expansion of both buccal and lingual cortical plates of the mandible with areas of stippled calcification and perforation (Figure.1C).

An incisional biopsy was done from labial aspect of the lesion mesial to left canine under local anaesthesia. Microscopically haematoxylin and eosin stained section showed a consistent biphasic appearance in which malignant mesenchymal and benign epithelial odontogenic components blended together in varying amounts. The epithelial component was arranged in islands with a peripheral layer of columnar cells. These cells resembled preameloblastic cells or cells of the inner enamel epithelium, some of which were surrounded by a homogenous eosinophilic hyalinized zone, often described as representing an inductive effect which seen in peripheral odontogenic fibroma cases [88] (Figure.2A & B). There was presence of reversal of polarity and hyperchromatic nuclei; cytoplasm was clear and vacuolated. In the central part of epithelial islands, polyhedral cells resembling stellate cells of the enamel organ. The mesenchymal component showed fascicular arrangement along with pleomorphism (Figure. 2C). Numerous mitotic figures were also present. On immunohistochemical studies, tumour cells in the islands expressed cytokeratin (CK) focally (Figure.2D). It was immunonegative for EMA, SMA, desmin, synaptophysin, Mic-2, CD34, CD31, S 100 protein & HBM-45. The histologic features were consistent with those of AFS.

The tumour showed extreme acceleration in growth after the incisional biopsy, with the size of the lesion doubling within a period of 6 months incapacitating the patient to close her mouth at all (Figure.3). Although evidence indicates that reactive growth is caused by an increase in the proliferation of fibroblasts, histopathologic differences between initial and postbiopsy lesions remain unexplored. Patient was referred to a tertiary care centre for further management where she underwent six cycles of presurgical chemotherapy (constituting of injection. emeset, ifosfamide, mensa, cisplatin and etoposide) followed by segmental resection of the mandible. The patient passed away after 6months after the initial operation. An autopsy was not performed.

Discussion

AFS is an extremely rare malignant odontogenic neoplasm, first reported as “spindle cell sarcoma” of the mandible by Heath in 1887 [5,6].It is considered as the malignant counterpart of AF and depending on the occurrence of dentin or dentin admixed with enamel, it has been further sub-classified as ameloblastic dentinosarcoma (ADS) or ameloblastic odontosarcoma (AOS) respectively [7].Many authors suggested that ADS and AOS are the same as AFS, because of consistent biological behaviour of these neoplasms [5].Also, according to the WHO, the presence or absence of dental hard tissue in odontogenic sarcoma is of no prognostic importance [8].Less than 5% malignant odontogenic neoplasms and even fewer sarcomatous neoplasms have been reported in literature [1].After a thorough literature search, we found that only 114 cases of AFS have been reported so far. Taking into consideration the present case, the mean age of presentation of these115 cases of AFS was 26.76 years, with age ranging from 4 months to 89 years. Of these, 61 cases were reported in males (53.33%), 47 in females (40%) and 7

unknown (6.66%), giving a male to female ratio of 1.29:1.Sixty six (62.85%) tumors were reported in the mandible, while 29 (27.61%) in maxilla, 9 (8.57%) unknown and 1 (0.95%) in the thyroid gland.

The clinical presentation in the present case was suggestive of an aggressive neoplasm. The presence of a tumor mass in the anterior region of the mandible with swelling and ulceration and radiographic signs of an ill-defined, lytic lesion with internal calcification raised the possibility of wide range of clinical diagnosis. In our case, destructive behaviour of the lesion was obvious in both clinical and radiographic images differential diagnosis considered were osteosarcoma, round cell neoplasm and aggressive odontogenic neoplasia, especially odontogenic myxoma or AFS. AFS must be differentiated from AF, ameloblastoma, and AOS histologically. To distinguish the malignant potential of AFS the significant points to kept in mind are: (1) hypercellularity of mesenchymal component with patchy distribution of hypercellular and hypocellular areas; (2) pleomorphic cells with hyperchromatic nuclei, occasionally malignant multinucleated giant cells and (3) arrangement of the cells in the bundles stimulating the herring-bone pattern, cartwheel, or storiform pattern, and occasional extracellular pink material in a lace-like deposition [3].These characteristics helped us to make the differential diagnosis with ameloblastoma in which the malignant mesenchymal component is deficient, with AF in which the mesenchymal component is benign with AOS in which enamel and dentin in addition to the fibrosarcomatous component is seen.

Although approximately two-thirds of AFS cases seemed to have arisen de-novo [7], literature review yielded 31 (26.95%) AFS cases with history of previous AF at the same site. Many reports of AF transforming into AFS after aggressive surgical treatment have been reported. In the present case, the tumour may have arisen de-novo as AFS, because the patient had not been treated before undergoing incisional biopsy. The commonest site of occurrenceof AFS is the mandibular posterior region, mostly presenting as a painful intraosseous swelling and ulcerated mass occasionally. These tumors are locally aggressive having tendency to erode through bony structures and adjacent soft tissues, and intracranial invasion [8-10]. A high local recurrence rate of 36.53 % inthe gingiva, floor of the mouth and neck has been reported in the literature. Although metastasis is an unusual characteristic of this tumour, 17.30% patients have died within 3 months to 19 years, resulting from locally aggressive tumour growth [8].

In general, the primary modality of treatment is aggressive surgical excision with clear margins. Adjuvant radiotherapy and chemotherapy have also been used in some cases. These treatment modalities help in prevention of recurrence of the tumour. However, specific chemotherapy protocols have yet to be established because of the rarity of this neoplasm.

Figures

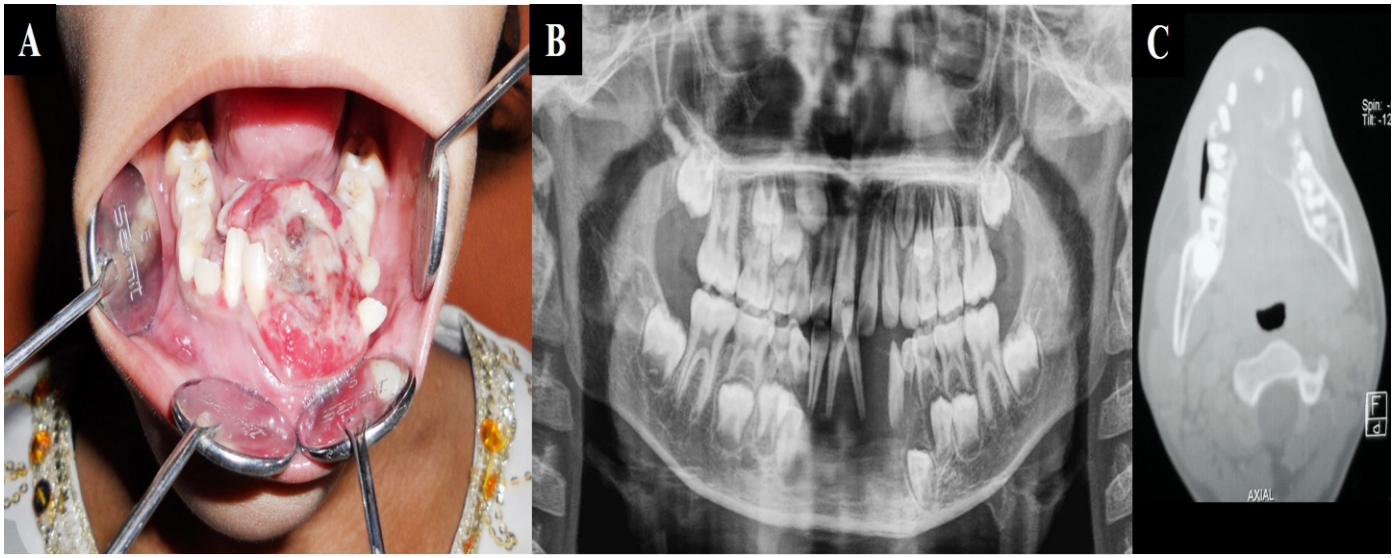

Figure 1: Clinico-radiographical features of primary tumour (**A**) Intraoral view showing ulceroproliferative of the mandibular anteriorregion before incisional biopsy; (**B**) Orthopantomogram showing large radiolucent area in the symphyseal region, displacing the incisors; (**C**) Axial CT scan showing heterogeously enhancing lesion in the mandibular anterior region with labial and lingual cortical perforation.

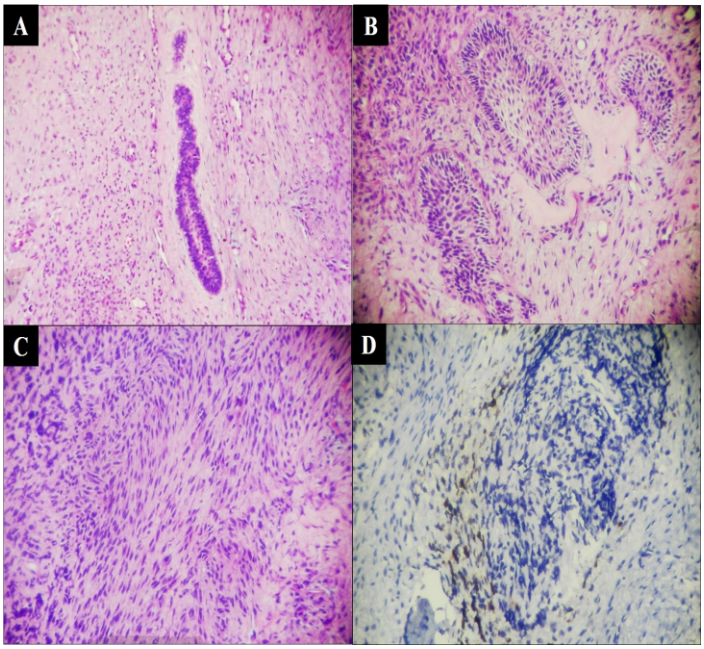

Figure 2: Histopathological features of recurrent tumour (**A**)Ameloblastic island in a sarcomatous background (Hematoxvlin-eosin stain X100); (**B**) Ameloblastic island with an adjacent area of induction (Hematoxvlin-eosin stain X100); (**C**) Malignant ectomesenchymal tissue of variable cellularity and polymorphism arranged in herring bone pattern (Hematoxvlin-eosin stain X100); (**D**)Focal positivity of ameloblastic epithelium for cytokeratin.

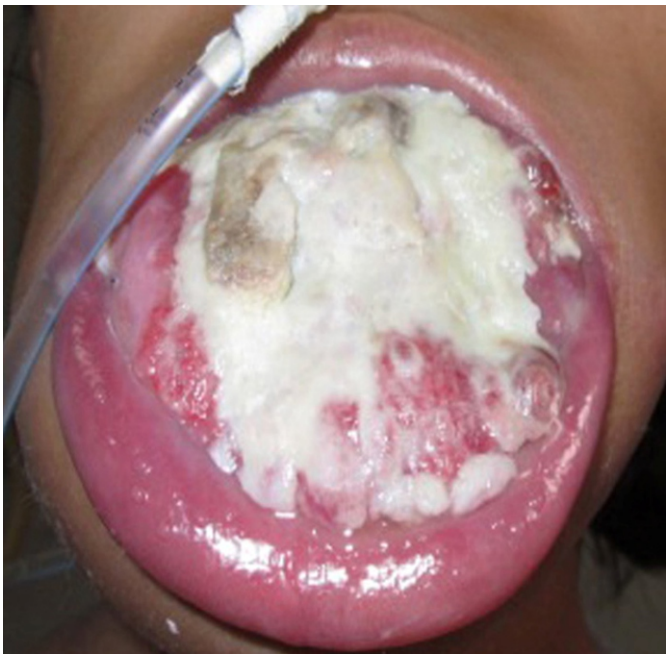

Figure 3: Clinical image showing rapid increase in the size of the tumour following incisional biopsy.

Table

Table 1: Summary of previous AFS cases reported in literature.

Author	Year	Age (yr)	Gender	Location	Recurrence	Metastases	Death	De Novo AFS	Previous AF	Follow-Up
Krompecher[11]	1918	13	M	Mandible	No	No	Yes*	NA	NA	NA
Papadimitrou[12]	1928	28	F	Mandible	No	No	No	NA	NA	6 mo
Kegel[13]	1932	45	M	Maxilla	NA	NA	NA	NA	NA	NA
Hauenstein[14]	1937	NA	M	Mandible	NA	NA	NA	NA	NA	NA
Emminger[15]	1946	52	M	Mandible	No	No	No	NA	NA	3 mo
Hertz[16]	1952	38	F	Maxilla	Yes	No	No	NA	NA	12 yr
Pindborg[17]	1960	17	M	Left maxilla	Yes	No	Yes	Yes	No	26 mo
Cina et al[18]	1962	39	M	Right mandible	Yes	No	No	No	Yes	15 yr
	1962	32	F	Right mandible	Yes	No	Yes	No	Yes	30 mo
Muroya[19]	1962	43	M	Mandible	Yes	No	Yes	NA	NA	33 mo
Cataldo et al[20]	1963	78	F	Left mandible	NA	NA	NA	Yes	No	NFU
Hogeman[21]	1966	30	M	Right posterior mandible	Yes	No	No	No	Yes	3 yr
Peychl[22]	1971	17	F	Right maxilla	Yes	No	Yes	Yes	No	4.5 yr
Leider et al[6]	1972	26	F	Right mandible	Yes	No	No	No	Yes	16 yr
	1972	43	M	Left mandible	Yes	No	No	No	Yes	38 mo
	1972	9	M	Left mandible	No	No	No	Yes	No	62 mo
	1972	22	F	Left maxilla	No	No	No	Yes	No	42 mo
	1972	23	F	Left mandible	Yes	No	No	No	Yes	34 mo
	1972	12	F	Left maxilla	Yes	No	NA	No	Yes	NFU
Mori et al[23]	1972	3.5	F	Right posterior mandible	Yes	No	Yes	Yes	No	5 yr
	1972	40	F	Left mandible	Yes	No	Yes	No	Yes	19 yr
Hatzifotiadis[24]	1973	15	M	Left posterior maxilla	Yes	No	Yes	Yes	No	2 yr
Motegi et al[25]	1975	29	M	Maxilla	No	No	No	NA	NA	3 yr
Remagen et al[26]	1975	24	M	Mandible	NA	NA	NA	NA	NA	NFU
Matsumura et al[27]	1976	25	F	Maxilla	No	NA	NA	NA	NA	NA
Goldstein et al[28]	1976	37	M	Left mandible	Yes	No	Yes†	No	Yes	4 yr

Eda et al[29]	1976	13	F	Mandible	No	No	No	Yes	No	3 yr
Adekeye et al[30]	1978	26	M	Right maxilla	No	No	No	Yes	No	9 mo
Reichart[31]	1978	16	M	Left mandible	Yes	No	No	No	Yes	23 mo
Daramola et al[32]	1979	19	M	Right posterior mandible	No	No	Yes‡	Yes	No	NFU
Prein et al[33]	1979	24	M	Anterior mandible	No	No	No	NA	NA	4 yr
Iwasa et al[34]	1981	24	F	Mandible	No	NA	NA	NA	NA	NA
Chomette et al[35]	1983	9	NA	Left maxilla	Yes	No	No	No	Yes	10 mo
	1983	38	M	Right mandible	Yes	No	No	No	Yes	3.5 yr
	1983	27	M	Paramedian mandible	Yes	Yes	Yes	No	Yes	10 yr
Nasu et al[2]	1984	24	F	Right mandible	No	No	No	Yes	No	4 yr
	1984	29	M	Left maxilla	No	No	No	Yes	No	9 yr
Takeda et al[36]	1984	19	M	Left maxilla	Yes	No	Yes	No	Yes	9.5 yr
Yamamoto et al[37]	1987	7	F	Right mandible	No	No	No	No	Yes	53 mo
Edali et al[38]	1987	28	M	Right mandible	No	No	No	Yes	No	2 yr
Wood et al[39]	1988	19	M	Right mandible	No	No	No	Yes	No	4 mo
Joon et al[40]	1992	26	M	Right mandible	NA	NA	NA	Yes	No	NA
Sözeri et al[41]	1993	5	M	Right mandible	Yes	No	No	No	Yes	10 mo
Choi M et al[42]	1993	26	M	NA	NA	NA	NA	NA	NA	NA
	1993	48	F	NA	NA	NA	NA	NA	NA	NA
Dallera et al[3]	1994	34	M	Right mandible	Yes	No	No	Yes	No	15.5 yr
	1994	18	F	Right mandible	Yes	No	Yes	Yes	No	1 yr
	1994	25	M	Right mandible	Yes	No	No	No	Yes	9 yr
	1994	18	F	Right mandible	No	No	No	No	Yes	2 yr
	1994	44	M	Left mandible	No	No	No	No	Yes	55 mo
Muller et al[43]	1995	53	F	Left maxilla	Yes	No	No	No	Yes	12 mo
	1995	17	F	Right mandible	No	No	No	Yes	No	1 yr
	1995	21	M	Left mandible	No	No	No	No	Yes	8 yr
	1995	57	F	Left mandible	Yes	No	No	No	Yes	6 yr
Park et al[44]	1995	17	M	Right mandible	Yes	No	Yes	Yes	No	3 mo

NogueiraTde et al[45]	1997	39	F	Right maxilla	No	No	No	No	Yes	2 yr
Tajima et al[10]	1997	14	M	Right maxilla	Yes	No	Yes	Yes	No	6 mo
DeNittis et al[46]	1998	32	M	Right maxilla	Yes	No	Yes	No	No	1 yr
Lu et al[47]	1998	NA	NA	NA	NA	NA	NA	NA	NA	NA
	1998	NA	NA	NA	NA	NA	NA	NA	NA	NA
	1998	NA	NA	NA	NA	NA	NA	NA	NA	NA
Sano et al[48]	1998	38	F	Right Posterior Maxilla	No	No	No	NA	NA	7 yr
Hayashi et al[5]	1999	22	M	Left mandible	No	No	No	Yes	No	3 yr
Bregni et al[7]	2001	19	M	Left mandible	No	No	No	Yes	No	7 mo
Huguet et al[49]	2001	31	M	Mandible	No	No	No	Yes	No	5 yr
Dufau et al[50]	2002	89	M	NA	Yes	NA	NA	No	Yes	1 mo, then LFU
Mosqueda Taylor et al[51]	2003	32	M	Left mandible	No	No	No	Yes	No	NFU
Batista de Paula et al[52]	2003	25	M	Left mandible	No	No	No	Yes	No	16 mo
Yamaguchi et al[53]	2004	31	M	Mandible	Yes	No	Yes	NA	NA	NA
Lee et al[54]	2005	62	F	Right maxilla	NA	NA	NA	NA	NA	NA
Kobayashi et al[55]	2005	26	M	Left mandible	Yes	No	No	No	Yes	5 yr
Iwamoto et al[56]	2005	15	M	Mandible	No	No	No	Yes	No	4 yr
Olgacet al[57]	2006	20-39	NA	Posterior maxilla	NA	NA	NA	NA	NA	NA
	2006	20-39	NA	Anterior maxilla	NA	NA	NA	NA	NA	NA
	2006	20-39	NA	Posterior mandible	NA	NA	NA	NA	NA	NA
Fujita et al[58]	2006	26.5	M	NA	NA	NA	NA	NA	NA	NA
	2006	26.5	F	NA	NA	NA	NA	NA	NA	NA
Williams et al[59]	2007	48	M	Left maxilla	Yes	No	No	No	Yes	6 mo
Okada et al[60]	2007	20-29	M	Posterior maxilla	NA	NA	NA	NA	NA	NA
Chidzongaet al[61]	2007	NA	F	Maxilla	NA	NA	NA	NA	NA	NA
Jing et al[62]	2007	4th D	F	Posterior maxilla	NA	NA	NA	NA	NA	NA
	2007	3rd D	F	Posterior mandible	NA	NA	NA	NA	NA	NA
Yoon et al[63]	2007	25	M	Mandible	No	No	No	NA	NA	18 mo

Nona Zabolinejad[8]	2008	4 mo	M	Left maxillary	No	No	No	Yes	No	4.5 yr
Delgado et al[64]	2008	19	F	Right mandible	No	No	No	Yes	No	5.3 yr
Kim et al[65]	2008	63	M	Left maxillary sinus	Yes	No	No	No	Yes	36 mo
Kousar et al[66]	2009	20	F	Left mandible	Yes	Yes	Yes	No	Yes	15 mo
Guthikonda et al[9]	2009	19	M	Right mandible	Yes	No	No	No	Yes	10 yr
Luo et al[67]	2009	32	M	Posterior mandible	NA	NA	NA	NA	NA	NA
Pontes et al[68]	2010	9	F	Right mandible	No	No	No	Yes	No	NFU
Faraj et al[69]	2010	17	M	Right mandible	No	No	No	No	Yes	NA
Nalinigupta et al[70]	2011	23	F	Left maxilla	NA	NA	NA	NA	NA	NA
Basavaraj[71]	2012	26	F	NA	No	No	No	Yes	No	4 mo
Jonathan Lai et al[1]	2012	17	F	Left Mandible	No	No	No	Yes	No	6mo
Noordhoek et al[72]	2012	36	F	Left Mandible	No	No	No	Yes	No	9 yr
Goldschmidt CD et al[73]	2012	12	M	Left Mandible	Yes	No	No	Yes	No	19 mo
	2012	13	M	Left mandible	Yes	No	No	No	Yes	18 mo
Costa DOP et al[74]	2012	1-19	F	Mandible	NA	NA	NA	NA	NA	NA
	2012	20-29	F	Mandible	NA	NA	NA	NA	NA	NA
Galvao et al[75]	2012	24	F	Posterior mandible	NA	NA	NA	NA	NA	NA
	2012	25	M	Posterior mandible	NA	NA	NA	NA	NA	NA
	2012	25	M	Posterior mandible	NA	NA	NA	NA	NA	NA
Cetin B et al[76]	2012	44	F	Thyroid gland	NA	NA	NA	NA	NA	NA
Lawal AO et al[77]	2013	28	F	Maxilla	NA	NA	NA	NA	NA	NA
Akinyamoju AO et al[78]	2013	28	F	Maxilla	No	No	No	NA	No	29 mo
Kapila R et al[79]	2014	26	F	Mandible	No	No	No	Yes	No	4.5 yr
Gilani SM et al[80]	2014	16	F	Mandible	NA	NA	NA	NA	NA	NA
Hu YY et al[81]	2014	22	M	Posterior Mandible	No	No	No	NA	NA	6 yrs
Balakrishna J et al[82]	2014	27	M	Mandible	NA	NA	NA	NA	NA	NA
Loya-Solis A et al[83]	2015	22	F	Left Mandible	No	No	No	Yes	No	1 yr

Nagori SA et al[84]	2015	23	M	Posterior Mandible	No	No	No	NA	NA	1 yr
Al Shetawi H et al[85]	2015	27	M	Mandible	No	No	No	Yes	No	6 mo
Pourdanesh F et al[86]	2015	34	F	Mandible	No	Yes	No	Yes	No	NA
Pillay RR et al[87]	2016	35	M	Left Maxilla	No	No	No	Yes	No	NA

Abbreviations: F: female; LFU: lost to follow-up; M: male; NA: not available; NFU: no follow-up; mo: month. D: decade

*From tuberculosis after operation.

†From melanoma.

‡From bleeding 9 days postoperatively.

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Manuscript Information: Received: September 27, 2016; Accepted: February 16, 2017; Published: February 22, 2017

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Citation: Singh JS, Desai RS, Bansal SP, Nehete AP, Mehta RV. Ameloblastic fibrosarcoma in a 10-year-old girl: Literature review and a rare case report. Open J Clin Med Case Rep. 2017; 1223

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