CASE REPORT

Ameloblastic fibro-odontosarcoma of the mandible: imaging findings

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We report a histologically verified case of ameloblastic fibro-odontosarcoma (AFOS) affecting the right mandible. CT scanning showed an expansile intraosseous mass with ill-defined borders and slightly dense opacities.

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Odontogenic tumours are very infrequent lesions, accounting for less than 4% of all oral and maxillofacial specimens.^{1–4} Malignant neoplasms account for a minimum percentage of all odontogenic tumours, as they represent between 0% and 6.1% according to diverse series published worldwide.^{1–9}

The 2005 World Health Organization (WHO) classification of odontogenic sarcomas presented two entities: ameloblastic fibrosarcoma (AFS); and ameloblastic fibro-dentinosarcoma (AFDS) and/or fibro-odontosarcoma (AFOS).¹⁰

The AFS consists of malignant connective tissue admixed with epithelium similar to that seen in an ameloblastoma or ameloblastic fibroma (AF). If there is also dentine, this is known as an AFDS and if there is also enamel, it is called AFOS. Those extremely rare lesions that combine carcinomatous and sarcomatous elements, but are recognizable as odontogenic through their epithelial component resembling ameloblastic carcinomas, have been called odontogenic carcinosarcoma or odontogenic carcinoma with sarcomatous proliferation.^{11,12} To date only 15 cases of AFDS/AFOS have been reported in the English-language literature.^{10,13}

To our knowledge, CT findings of AFOS have not been described in the radiology literature. This report describes the CT and histopathological findings in a 45-year-old Chinese woman with AFOS of the right mandible.

Case report

A 45-year-old Chinese woman with painful swelling in the right mandible was referred to our hospital in January 2009 for maxillofacial consultation. She reported expansion of the mass for approximately 2 months. The patient was generally healthy and a clinical examination revealed no numbness. She had no history of radiotherapy and was taking no medications.

Panoramic radiograph revealed an irregular, poorly demarcated osteolysis. The expansile lesion extended from the right lower second molar to about the whole ramus and was approximately 6 × 4.5 cm in diameter (Figure 1). Contrast-enhanced CT scan (a LightSpeed-16, GE Medical Systems, Waukesha, WI) of the gnathic lesion showed the medullary portion of the right ramus was osteolytic and destroyed. The lesion appeared as expansile multilocular lesions and the cortexes were broken (Figure 2a). The soft-tissue component was fairly uniform and included slight radiopaque foci, which might have been calcium deposits. After infusion of the contrast material, the soft-tissue portion of the tumour showed an unhomogeneous enhancement (Figure 2b,c).

A partial mandibulectomy was performed while the patient was under general anaesthesia. The mandible contained a fleshy mass with a white to yellowish cut surface. The histopathological examination revealed that the epithelium was composed of budding and branching cords of small polygonal epithelial cells mixed with islands and knots. Larger islands had a border of columnar cells with hyperchromatic nuclei. Structures resembling enamel matrix and dentine were noted (Figure 3). Immunohistochemically, malignant spindle cells were positive for vimentin but negative

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Figure 1 Panoramic radiograph revealed an irregular, poorly demarcated osteolysis. The expansile lesion extends from the right lower second molar to about the whole ramus (black arrows)

for S-100 protein. Ameloblastic epithelium showed positive immunoreactivity for pancytokeratin. A diagnosis of AFOS was made.

No clinical sign of recurrence was observed 12 months after the partial mandibulectomy was performed.

Discussion

AFS is an odontogenic tumour with a benign epithelial and a malignant ectomesenchymal component.¹⁰ A tumour with histological features of AFS, together with dysplastic dentine, is known as an AFDS and with enamel/enameloid and dentine/dentinoid is known as an AFOS.¹⁰

When reviewing the literature on AFDS/AFOS, 15 cases^{10,13} have been reported in the English-language literature up to 2009. To the authors' knowledge, the case presented here is the sixteenth and the sixth in a female patient. The age range is 12–83 years with a peak in the third decade.¹⁰ Clinically, AFDS/AFOS presented as a painful swelling and intraosseous mass.

Ameloblastic fibro-odontomas are regarded as possible precursor lesions.^{14–16} The histological study of the present case revealed a highly cellular mesenchymal tissue with spindle-shaped cells with marked pleomorphism, arranged in a fascicular pattern. There were also small areas with deposition of enamel matrix and dentine material adjacent to ameloblastic epithelium. The WHO distinguishes odontogenic sarcoma devoid of dental hard tissue (AFS) from those displaying focal evidence of dentinoid (AFDS) or dentinoid plus enameloid (AFOS), but the WHO panel acknowledges

that presence or absence of dental hard tissue in an odontogenic sarcoma is of no prognostic significance.¹⁷

Radiographically, AFDS/AFOS can show a uni- or multilocular appearance with poorly circumscribed outlines being associated with tooth and one or more dense opacities. The present case showed a multilocular appearance associated with the right lower second molar and slight dense opacities. These radiographic findings and the location of the mass highly suggest the possibility of odontogenic cysts and tumour. The clinico-radiographic differential diagnosis should include ameloblastoma, ameloblastic carcinoma, keratocystic odontogenic tumour and dentigerous cysts. However, a case with an irregular border and expansion and perforation of the cortexes should be interpreted with caution and the possibility of malignant odontogenic tumour should be suspected.

The information available concerning the treatment, course and prognosis of AFDS/AFOS is limited because of the paucity of cases reported. According to previous reports, the treatment of choice is wide surgical excision with long-term follow-up.^{14,18} Ameloblastic sarcomas seem to have a better prognosis than other jaw sarcomas. The biological behaviour is that of a highly locally aggressive neoplasm with extremely low potential for distant metastasis. Only 1 out of 16 cases had metastasis to mediastinal node and liver.^{16,19–22}

In conclusion, we reported the radiographic and histological aspects of a new case of AFOS. Although uncommon, AFOS should be considered in the differential diagnosis of intraoral radiolucencies with poorly circumscribed outlines that contain radiopaque material.

Figure 2 Axial CT scan (soft-tissue algorithm; (a) showed that the medullary portion of the right ramus was osteolytic destroyed and the cortices were broken. Note the slight radiopaque foci, which might be calcium deposits (black arrows). Contrast-enhanced axial and multiplanar reformatted coronal CT scan (soft-tissue algorithm; (b,c) showed that the soft-tissue portion of the tumour had an unhomogeneous enhancement (black asterisk)

Figure 3 Photomicrograph (haematoxylin and eosin, original magnification $\times 100$) reveals a cord and island of odontogenic epithelium (short black arrows) in a loose primitive appearing connective tissue (black asterisks) with enamel (long black arrows) and dentine matrix formation (arrow heads)

References

- Regezi JA, Kerr DA, Courtney RM. Odontogenic tumors: analysis of 706 cases. *J Oral Surg* 1978; **36**: 771–778.
- Daley TD, Wysocki GP, Pringle GA. Relative incidence of odontogenic tumors and oral and jaw cysts in a Canadian population. *Oral Surg Oral Med Oral Pathol* 1994; **77**: 276–280.
- Mosqueda-Taylor A, Ledesma-Montes C, Caballero-Sandoval S, Portilla-Robertson J, Ruíz Godoy-Rivera LM, Meneses-García A. Odontogenic tumors in México. A collaborative retrospective study of 349 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1997; **84**: 672–675.
- Bhaskar SN. Oral pathology in the dental office: survey of 20,575 biopsy specimens. *J Am Dent Assoc* 1968; **76**: 761–766.
- Günhan O, Erseven G, Ruacan S, Celasun B, Aydıntug Y, Ergun E, et al. Odontogenic tumours. A series of 409 cases. *Aust Dent J* 1990; **35**: 518–522.
- Mothes P, Kreusch T, Harms D, Donath K, Schmelzle R. Die Häufigkeit odontogener Tumoren in Wachstumsalter. *Deutsche Zahnärztliche Zeitschrift* 1991; **46**: 18–19.
- Lu Y, Xuan M, Takata T, Wang C, He Z, Zhou Z, et al. Odontogenic tumors. A demographic study of 759 cases in a Chinese population. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1998; **86**: 707–714.
- Odukoya O. Odontogenic tumors: analysis of 289 Nigerian cases. *J Oral Pathol Med* 1995; **24**: 454–457.
- Arotiba JT, Ogunbiyi JO, Obiechina AE. Odontogenic tumours: a 15-year review from Ibadan, Nigeria. *Br J Oral Maxillofac Surg* 1997; **35**: 363–367.
- Barnes L, Eveson JW, Reichart P, Sidransky D (eds.) *World Health Organization classification of tumours. Pathology and genetics of head and neck tumours*. Lyon, France: IARC Press 2005, pp 294–295.
- Barnes L, Eveson JW, Reichart P, Sidransky D (eds.) *World Health Organization classification of tumours. Pathology and genetics of head and neck tumours*. Lyon, France: IARC Press 2005, pp 283–327.
- Slootweg PJ. Malignant odontogenic tumors; an overview. *Mund Kiefer Gesichtschir* 2002; **6**: 295–302.
- Mainenti P, Oliveira GS, Valerio JB, Daroda LS, Daroda RF, Brandao G, et al. Ameloblastic fibro-odontosarcoma: a case report. *Int J Oral Maxillofac Surg* 2009; **38**: 289–292.
- Altini M, Thompson SH, Lownie JF, Berezowski BB. Ameloblastic sarcoma of the mandible. *J Oral Maxillofac Surg* 1985; **43**: 789–794.
- Howell RM, Burkes EJ Jr. Malignant transformation of ameloblastic fibro-odontoma to ameloblastic fibrosarcoma. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1977; **43**: 391–401.
- Muller S, Parker DC, Kapadia SB, Budnick SD, Barnes EL. Ameloblastic fibrosarcoma of the jaws: A clinicopathologic and DNA analysis of five cases and review of the literature with discussion of its relationship to ameloblastic fibroma. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1955; **79**: 469–477.
- Lee JS. Odontogenic sarcoma and carcinosarcoma. *Semin Diagn Pathol* 1999; **16**: 325–332.
- Slater LJ. Odontogenic sarcoma and carcinosarcoma. *Semin Diagn Pathol* 1999; **16**: 325–332.
- Bregni RC, Taylor AM, Garcia AM. Ameloblastic fibrosarcoma of the mandible: report of two cases and review of the literature. *J Oral Pathol Med* 2001; **30**: 316–320.
- Chomette G, Auriol M, Guibert F, Delcourt A. Ameloblastic fibrosarcoma of the jaws—report of three cases. Clinicopathologic, histochemical and ultrastructural study. *Pathol Res Pract* 1983; **178**: 40–47.
- Dallera P, Bertoni F, Marchetti C, Bacchini P, Camorosi A. Ameloblastic fibrosarcoma of the jaws: report of five cases. *J Craniomaxillofac Surg* 1994; **22**: 349–354.
- Gustafsson H, Carlsoo B, Henriksson R. Ultrastructural morphology and secretory behavior of acinic cell carcinoma. *Cancer* 1985; **55**: 1706–1710.