

Odontogenic myxoma and its differential diagnoses based on the apparent diffusion coefficient: a case report and brief literature review

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ABSTRACT | *Objective:* This report discusses the main imaging findings observed in odontogenic myxoma (OM) with an emphasis on the importance of apparent diffusion coefficient (ADC) values for diagnosis. *Case report:* In the present report, a female patient sought care due to an edema in the maxilla region associated with nocturnal dyspnea. Cone beam computed tomography showed a hypodense image invading the right maxillary sinus. The mean ADC value was $2.07 \pm 0.30 \times 10^{-3} \text{ mm}^2/\text{s}$ contributed to the elucidation of the diagnostic hypotheses, differentiating the lesion from its main differential diagnoses. Histopathological examination confirmed the suspicion of an odontogenic myxoma. *Conclusion:* OM is a rare benign mesenchymal neoplasm of locally aggressive nature. DWI parameters and ADC values can be useful to differentiate OM from other odontogenic lesions.

DESCRIPTORS | Odontogenic Tumors; Myxoma; Diffusion Magnetic Resonance Imaging.

RESUMO | **Mixoma odontogênico e seus diagnósticos diferenciais com base no coeficiente de difusão aparente: relato de caso e breve revisão da literatura** • *Objetivo:* Este relato discute as principais descobertas advindas da análise de imagens radiográficas relacionadas ao mixoma odontogênico, enfatizando a importância dos valores do coeficiente de difusão aparente (ADC) no diagnóstico. *Relato de caso:* Uma paciente do sexo feminino buscou tratamento por conta de um edema na região da maxila, associado a dispnéia noturna. O uso de tomografia computadorizada de eixo cônico revelou uma imagem hipodensa invadindo o seio maxilar direito. O valor médio do ADC foi $2,07 \pm 0,30 \times 10^{-3} \text{ mm}^2/\text{s}$ e contribuiu para a definição da hipótese de diagnóstico, diferenciando a lesão de seu principal diagnóstico diferencial. Exame histopatológico confirmou a suspeita de mixoma odontogênico. *Conclusão:* Mixoma odontogênico é uma rara e benigna neoplasia mesenquimal, de caráter localmente agressivo. Parâmetros de sequência de difusão e valores ADC podem ser úteis para diferenciar o mixoma odontogênico de outras lesões odontogênicas.

DESCRITORES | Tumores Odontogênicos; Mixoma; Imagem de Difusão por Ressonância Magnética.

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INTRODUCTION

Odontogenic myxoma (OM) is a rare neoplastic lesion that originates from the ectomesenchyme of a developing tooth. Most cases affect patients between 20 to 40 years of age and women. The mandible is more affected than the maxilla, mainly in the posterior region.^{1,2} Despite being classified as a benign lesion, OM is highly invasive and aggressive, with high recurrence rates.³

OM is clinically described as a slow-growing odontogenic tumor, commonly diagnosed when it affects a large dimension of the patient's maxillary-mandibular complex.^{3,4}

The diagnosis is based on the histopathological results, Diffusion Weighted Imaging (DWI) – a form of Magnetic Resonance Imaging (MRI) – is used to distinguish OM from other odontogenic conditions by forming images from the mobility of water in the tissues, promoting a molecular and cyto-architectural visualization of the lesions.⁵⁻⁷ The Apparent Diffusion Coefficient (ADC) is useful to quantify the image of the lesion and its values are inversely proportional to the lesion's cellularity.^{5,6}

We described a case of odontogenic myxoma and evaluated the effectiveness of ADC values in

its diagnosis, comparing them to those of other odontogenic lesions.

CASE REPORT

A 28-year-old woman came to the office with progressive swelling on the right side of the maxilla associated with nocturnal dyspnea in the last three months. Clinical examination revealed facial asymmetry on the right side and expansion of the vestibular and palatal bone cortical areas.

Cone beam computed tomography (GXCB-500™, powered by i-CAT® obtained with a 16 x 6.0 cm field of view (FOV) and 0.2 mm voxel size) was used to obtain coronal, axial and sagittal images; subsequently, parasagittal/cross-sectional images were cut to 1.0 mm thickness and 1.0 mm spacing in the maxilla. Tomography showed a hypodense lesion and osteolysis with an inflating pattern in the right maxilla. There was a complete invasion of the right maxillary sinus of the alveolar bone crest from the region of maxillary right second premolar, maxillary right first molar, and maxillary right second molar, the floor of the orbit and the nasal cavity (Figure 1).

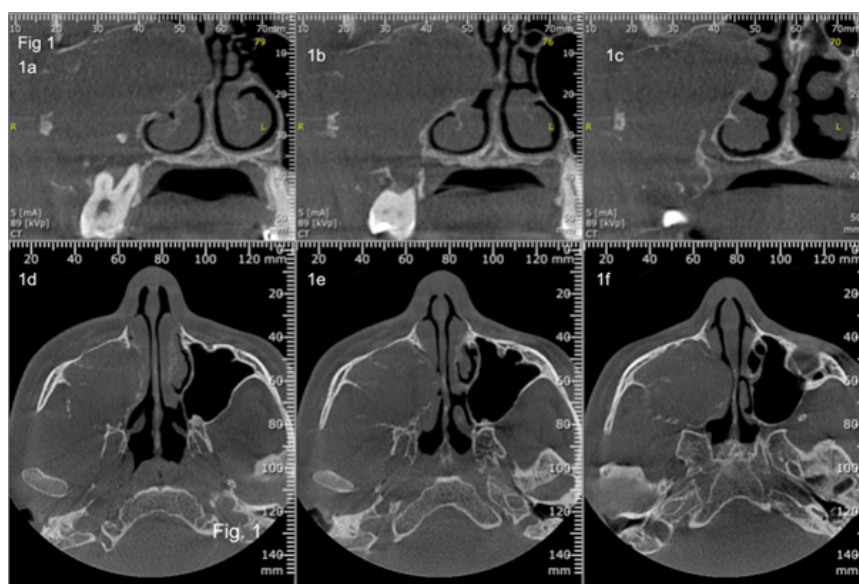


FIGURE 1 | Cone beam computed tomography in coronal section (1a, 1b, 1c) and axial section (1d, 1e, 1f) showing an osteolytic lesion with an inflating pattern in the right maxilla. Produced by the authors.

Tomography allowed the visualization of the expanded, thinned and ruptured areas of alveolar bone cortical areas, as well as bone cortical of the maxillary sinus walls, displacement and thinning of orbit floor and lateral wall of ipsilateral nasal fossa. An invasion and partial obstruction of the corresponding nasal fossa was also found, besides a delicate and significant trabecular meshwork in a multilocular pattern in the maxillary tuber and peripheral regions of the lesion.

MRI was performed using a 1.5 Tesla scanner (Sigma General Electric, Milwaukee, USA) with a skull coil for axial, coronal, and sagittal anatomical planes. T1 and T2 weighted images were obtained with an 8-channel head coil, phase matrix and T1 (TR = 478 ms, 0.72 mm isotropic voxel size, TE = 16 ms, 1.0 × 21 FOV, 0 cm, cut-off interval = 2.0 mm) and T2 sequences (TR = 6.5 ms, 0.72 mm voxel isotropic size, TE = 90.0 ms, FOV of 21.0 × 21.0 cm, slice interval = 2.0 mm). MRI showed the complete filling of the right maxillary sinus with material characterized

by an intermediate signal in T1 and heterogeneous hypersignal in T2, in a solid aspect, with tenuous foci of impregnation by paramagnetic contrast in the lower and lateral contour of the lesion, remodeling the maxillary sinus right walls, with bulging of the superficial soft part planes of the malar region, reduction of the amplitude of the middle meatus and nasal fossa on the same side, and the tapering of the alveolar ridge of the maxilla also on the right side. The lesion had expansive characteristics, without significant bone erosions or infiltrations (Figure 2).

From the analysis of the DWI (Figure 3), the ADC values showed a low to intermediate signal, indicating diffusion restriction, and the mean value was $2.07 \pm 0.3 \times 10^{-3} \text{ mm}^2/\text{s}$.

The anatomopathological report revealed odontogenic neoplasm of mesenchymal origin consisting of many spindle and star cells, loose and myxoid stroma. Shavings of mineralized tissue could be seen in focal areas. Based on histopathology, the final diagnosis was odontogenic myxoma (Figure 4).

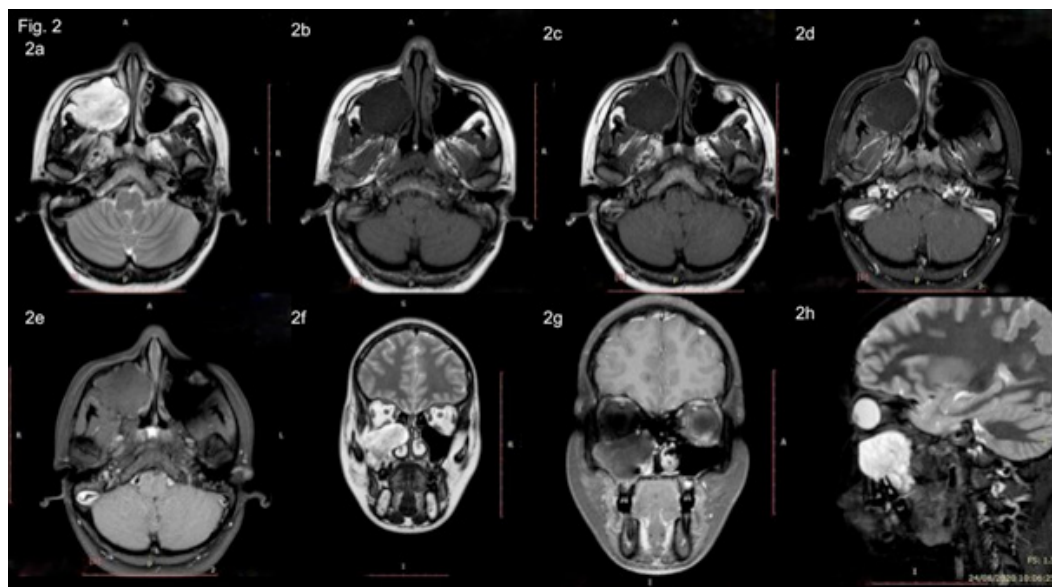


FIGURE 2 | Face magnetic resonance imaging in cross sections (2a: T2 - weighted images, 2b: T1-weighted images, 2c: T1 Spectral Presaturation with Inversion Recovery, 2d: T1 without contrast, 2e: T1 Spectral Presaturation with Inversion Recovery), coronal (2f: T2, 2g: T1 Spectral Presaturation with Inversion Recovery) and sagittal (2h: T2) showing a solid-looking mass in the right maxillary sinus with invasion to the ipsilateral nasal fossa. Produced by the authors.

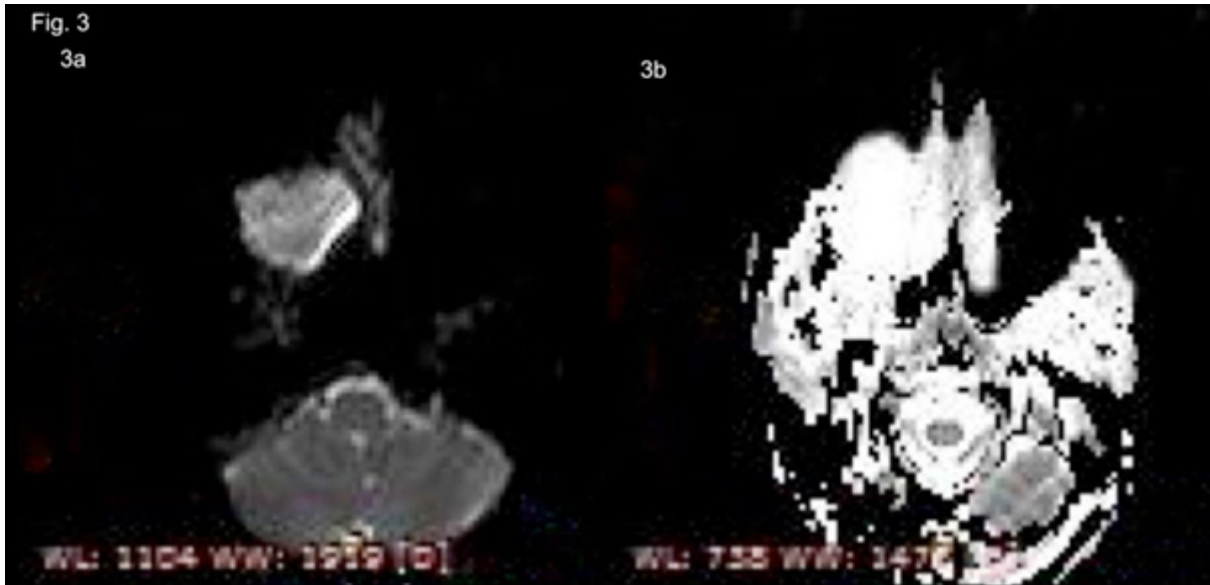


FIGURE 3 | DWI (3a) and ADC map (3b) in axial sections showing maxillary lesion with low to intermediate signal in diffusion, indicating partial diffusion restriction. Produced by the authors.

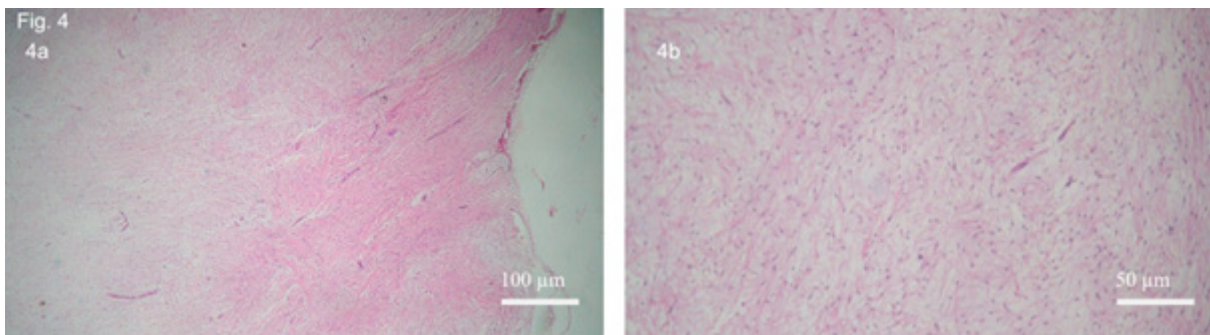


FIGURE 4 | Histological sections in hematoxylin and eosin confirming the diagnosis of odontogenic myxoma (4a: 4x) (4b: 40x). Produced by the authors.

DISCUSSION

Odontogenic myxoma is an infiltrative lesion with a loose and grayish-white appearance characterized by the pathological accumulation of mucoïd or myxoid stroma. OM consistency varies in proportion to the amount of collagen present in the lesion.⁴ Radiological and imaging screenings are important to establish the OM diagnosis; however, the characteristics of this lesion vary depending on its stage of development.⁶

Several differential diagnoses exist for OM such as dentigerous cyst, ameloblastoma, keratocyst and ameloblastic fibroma.^{5,6} In most cases, it is difficult

to distinguish OM from ameloblastomas based on radiographic imaging alone;⁸ the analysis of MRI resources can be useful to diagnose odontogenic myxoma and differentiate it from other lesions.

CT and conventional MRI can identify the extent and shape of odontogenic lesions and their effects on the surrounding structures.⁷ However, in cases where imaging exams cannot distinguish odontogenic lesions due to similar properties,⁵ ADC can be used because each lesion has a different value within a specific numerical range (Table 1).⁶ The DWI results and ADC values found in our case report were similar to the ones found in previous studies on OM.^{8,9}

TABLE 1 | Main differential diagnoses of odontogenic myxoma according to apparent diffusion coefficient values.

Odontogenic lesion	Average ADC value
Unicystic ameloblastoma (9)	$2.192 \pm 0.33 \times 10^{-3} \text{ mm}^2/\text{s}$
Unicystic ameloblastoma (10)	$2.518 \times 10^{-3} \text{ mm}^2/\text{s}$
Dentigerous cyst (10)	$2.150 \times 10^{-3} \text{ mm}^2/\text{s}$
Dentigerous cyst (18)	$1.67 \pm 1.06 \times 10^{-3} \text{ mm}^2/\text{s}$
Dentigerous cyst (9)	$1.23 \pm 0.09 \times 10^{-3} \text{ mm}^2/\text{s}$
Odontogenic myxoma (9)	$2.091 \pm 0.19 \times 10^{-3} \text{ mm}^2/\text{s}$
Keratocyst (9)	$1.019 \pm 0.07 \times 10^{-3} \text{ mm}^2/\text{s}$
Keratocyst (10)	$2.217 \pm 0.954 \times 10^{-3} \text{ mm}^2/\text{s}$
Keratocyst (18)	$1.03 \pm 0.31 \times 10^{-3} \text{ mm}^2/\text{s}$

Odontogenic myxoma appears in conventional MRI as a hypointense signal in T1-weighted images and hyperintense signal in T2-weighted images, the same as cystic ameloblastoma and dentigerous cyst, in other words, they are indistinguishable.⁷ However, odontogenic lesions express different ADC values according to diffusion, whether facilitated or restricted (Table 1). The ADC for OM is $2.122 \times 10^{-3} \text{ mm}^2/\text{s}$, a value similar to those found in ameloblastoma and dentigerous cyst ($2.337 \times 10^{-3} \text{ mm}^2/\text{s}$ and $2.150 \times 10^{-3} \text{ mm}^2/\text{s}$, respectively) since all these lesions facilitate water diffusion.^{5,6}

Keratocyst is a lesion characterized by anteroposterior growth within the medullary bone. Although less common, it can present an aggressive behavior, even sharing image aspects similar to those found in ameloblastoma and odontogenic myxoma, whereas the odontogenic myxoma has facilitated diffusion (ADC value $2.122 \times 10^{-3} \text{ mm}^2/\text{s}$), keratocyst presents lower ADC ($1.019 \times 10^{-3} \text{ mm}^2/\text{s}$) and diffusion restriction.^{5,6,8,10} Therefore, the analysis of DWI images and the quantification of ADC are fundamental role in the differential diagnosis between ameloblastomas, keratocysts and odontogenic myxomas.⁹

CONCLUSION

OM is a rare benign tumor with a multilocular and expansive tomographic aspect. DWI parameters

and ADC values can be useful to differentiate it from other odontogenic lesions.

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