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Differential diagnosis for an unusual calcification in the maxillary sinus: case report

Diagnóstico diferencial para calcificação incomum em seio maxilar: relato de caso

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ABSTRACT

The aim of this study was to report a rare case of an asymptomatic calcification of unusual size and shape, inside the maxillary sinus identified on a cone beam computed tomography exam and to discuss the importance of knowledge of the anatomy of maxillary sinus and its changes carefully evaluating the entire volume of the images, regardless of the region of interest. An 83-year-old female patient underwent a cone beam computed tomography exam for other diagnostic purposes in the maxillofacial region. When analyzing the entire volume, an image of unusual limits, hyperdense and calcified was found in the right maxillary sinus, close to the anterior and medial wall, with a rounded and homogeneous shape, occupying approximately one third of the maxillary sinus. There was no lytic or erosive lesions on the maxillary sinus wall. The main diagnostic hypotheses raised were giant anthrolith, mucous retention phenomenon and osteoma. Due to the anatomical complexity of the maxillary sinus, diagnosis in this region becomes a challenge. Considering the limitations of 2D exams, the cone beam computed tomography exam can be used to evaluate these structures. Knowledge of differential hypotheses is extremely important for the case to be conducted correctly, but it does not replace biopsy and histopathological examination.

Indexing terms: Calcinosis. Cone beam computed tomography. Diagnosis. Maxillary sinus.

RESUMO

O objetivo desse estudo foi relatar um caso raro de uma calcificação assintomática de tamanho e formato incomum, no interior do seio maxilar, diagnosticado em um exame de tomografia computadorizada de feixe cônico, além de discutir a importância do conhecimento da anatomia do seio maxilar e suas alterações, avaliando cuidadosamente todo o volume das imagens, independentemente da região de interesse. Paciente do sexo feminino, 83 anos de idade, realizou uma tomografia computadorizada de feixe cônico para outros fins de diagnóstico na região bucomaxilofacial. Ao analisar todo volume da tomografia encontrou-se uma imagem de limites incomuns, hiperdensa e calcificada, no seio maxilar direito, próximo a parede anterior e medial, com formato arredondado e homogêneo,

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ocupando aproximadamente um terço do seio maxilar, com as suas paredes intactas e sem nenhuma lesão lítica ou erosiva. As principais hipóteses diagnosticas levantadas foram antrólito gigante, fenômeno de retenção mucoso e osteoma. Devido a complexidade anatômica do seio maxilar, o diagnóstico nessa região se torna um desafio. Considerando as limitações dos exames 2D, a tomografia computadorizada de feixe cônico pode ser empregada para a avaliação dessas estruturas. O conhecimento do raciocínio diagnóstico e das hipóteses diferenciais são de extrema importância para o que o caso seja conduzido corretamente, porém não substituem a biópsia e o exame histopatológico.

Termos de indexação: *Calcinose. Tomografia computadorizada de feixe cônico. Diagnóstico. Seio maxilar.*

INTRODUCTION

The maxillary sinuses (MS) form a pair of bony chambers located in the maxillary bone that contain air inside [1,2]. Its proximity to important structures can lead to the development of sinus pathologies and inflammatory processes of odontogenic origin [3], requiring special attention in dental planning. These changes include thickening of the sinus mucosa, mucous retention phenomenon, polyps, antroliths, opacified images of the indefinite origin or related to the inflammatory reaction, and less commonly, the presence of periostitis related to osteolytic lesions [3,4]. The identification of these changes is essential to establish the correct diagnosis and treatment of the patient [5], when necessary.

Because it is a specific exam of the dentomaxillofacial region [6], cone beam computed tomography (CBCT) is often the exam of choice for analysis of the paranasal sinuses, as it promotes three-dimensional images with lower radiation dose and cost when compared helical computed tomography (HCT) [7]. CBCT clearly shows sinus changes4 and their relationship with adjacent teeth [8].

MS are part of the region of dental interest and their careful evaluation is essential when planning surgical and non-surgical procedures in the maxillary region. In addition, nonspecific symptoms such as pain and drainage may come from sinus pathologies. The aim of this manuscript is to report a case of a large calcification in the MS detected in a CBCT examination of an asymptomatic patient.

CASE REPORT

An 83-year-old female patient was referred to a radiology and dental imaging clinic to perform a CBCT exam for rehabilitation planning with implants in the posterior region of the maxilla.

CBCT was performed on the i-CAT® CB500 system (Imaging Sciences, Hatfield, PA, USA), following the acquisition protocol: 120 kVp, 5 mA, voxel size of 0.125 mm, Field of View (FOV) of 8 × 8 cm. The images were evaluated using the e-Vol DX® software (CDT software, Bauru, SP, Brazil) in Full HD screen (resolution 1920X1080 pixels), with 1-mm spacing.

The upper posterior teeth and upper right canine were absent. In the right MS, a hyperdense image was observed, with well-defined and homogeneous edges, rounded shape, with density compatible with calcified material, showing a close relationship with the floor of the MS and close to the anterior and medial walls, which were intact, with no signs of lysis or erosion (figure 1). There was a thickening of the SM lining mucosa on the floor and anterior wall. The thickening also involved the lesion. The calcification was not connected to the SM walls and its interior was more hypodense in relation to the margins (figure 2). The total volume and the largest diameter of the calcification were calculated using the ITK-SNAP 3.0 software (Cognitica, Philadelphia, PA, USA) (https://www.itksnap.org), being 2253 mm^3 and 15.5 × 14.4 × 16.5 mm, respectively.

When the patient was questioned, she reported not knowing the alteration and showing no symptoms. She denied episodes of sinusitis, previous infectious diseases, allergic rhinitis, asthma crisis, or history of surgery in MS, as well as facial trauma or insertion of a foreign body in the nasal cavity. Based on the clinical and imaginogical findings, the initial

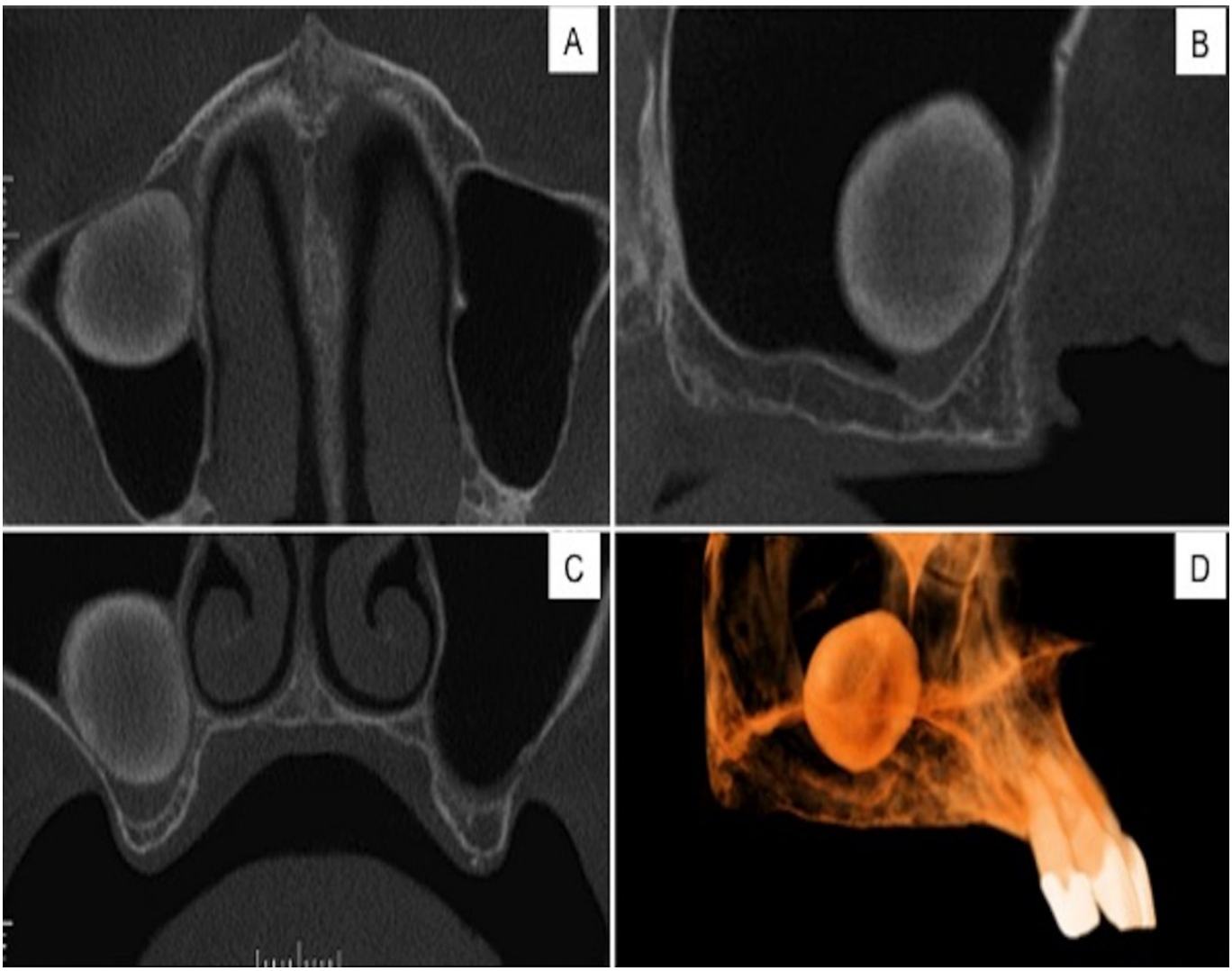

Figure 1. CBCT. Axial (A), sagittal (B), coronal (C) and tridimensional (D) reconstructions showing a calcification in the right MS in its largest diameter.

following hypothesis were suggested: a hard-tissue neoplasm (osteoma), mucous retention phenomenon and antrolith. As the region was edentulous, pathologies of odontogenic origin were discarded. Due the fact of the lesion is not in contact with any maxillary sinus wall, the osteoma was the least likely hypothesis. Likewise, the increased density and the corticalization of the lesion contest the hypothesis of a mucous retention phenomenon. Thus, our first presumptive diagnosis was an antrolith. The patient was referred to the otorhinolaryngologist but, due her advanced age and absence of symptoms, only periodic clinical-imaging monitoring was adopted.

DISCUSSION

The MS can be affected by several conditions, and its clinical access is difficult. 3D images are shown to be very helpful in the better clarification of those conditions [2]. The radiologist has to carefully evaluate the MS to rule out any significant pathologic changes [6]. The higher prevalence of pathologies in asymptomatic patients emphasizes the significance of interpretation of the whole volume scans as it might have an impact on its medical status and in planning

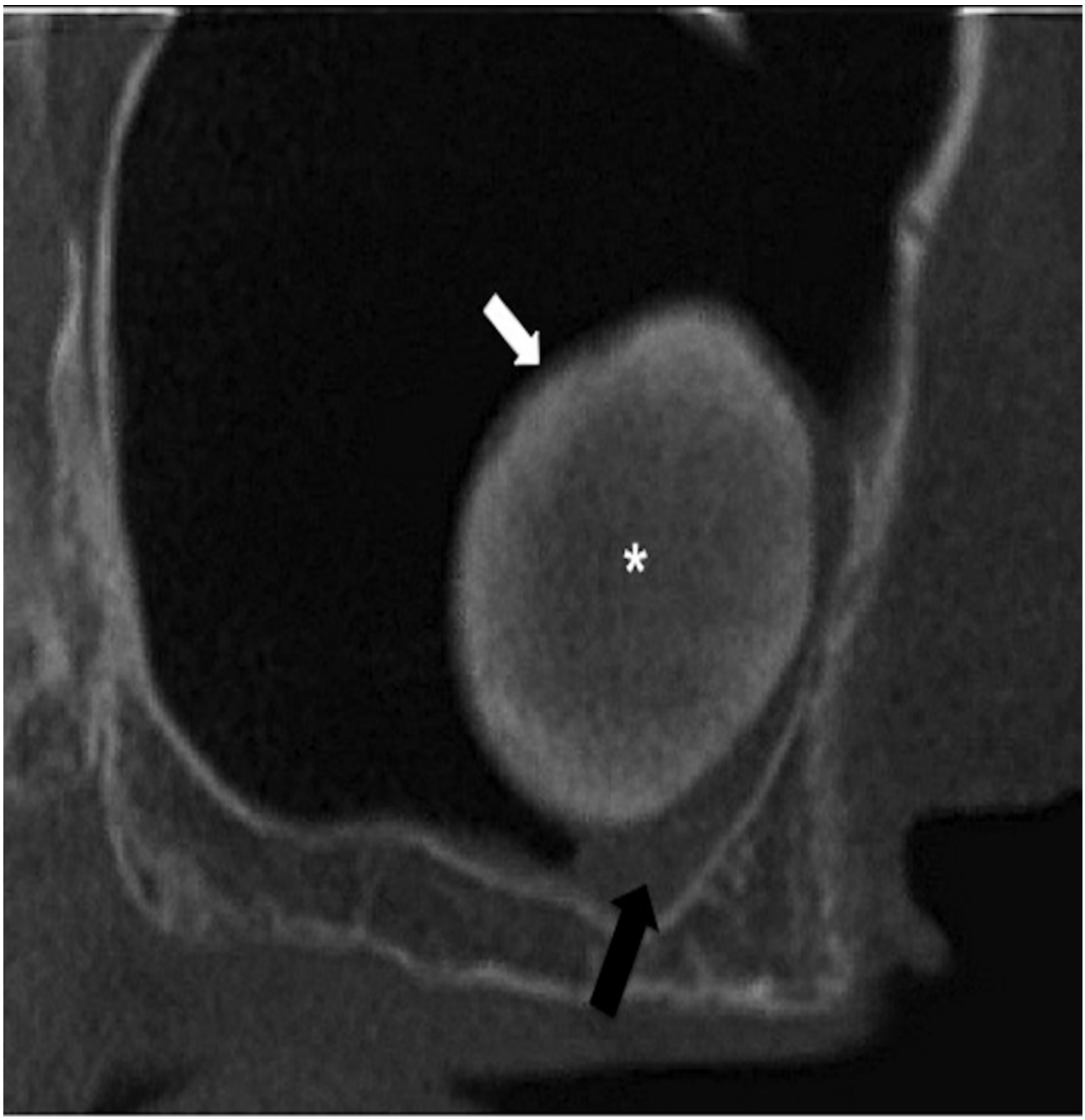

Figure 2. Sagittal reconstruction showing the lesion surrounded by the MS mucosa (white arrow), the mucosal thickening involving the floor and anterior wall of the MS (black arrow) and the hypodense interior compared to the margins (*).

Table 1. Reports of unusual antroliths (>1.0cm) in the literature.

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Study	Patient's data	Image modality	Size	Shape	Volume	Occurrence	Symptoms	Management
Cohen et al. [12]	32 years-old female	Panoramic radiography, Waters' view and HCT	3.0×3.0 cm	Irregular	Not reported	On the floor of right MS	Absent	Removal using the Caldwell-Luc approach
Nass Duce et al. [11]	47 years-old female	HCT	1.2 cm	Irregular	Not reported	In the middle of the left MS, non-related with the walls	Headache, postnasal discharge, and nasal stuffiness	Removal using the Caldwell-Luc approach

Table 1. Reports of unusual antroliths (>1.0cm) in the literature.

Study	Patient's data	Image modality	Size	Shape	Volume	Occurrence	Symptoms	Management
Nair et al. [18]	35 years-old female	HCT	1.0×0.3 cm	Irregular	Not reported	Right MS, frontal region, and anterior wall of ethmoid sinus	Nasal discharge, facial pain, and headache	Endoscopic sinus surgery
Shenoy et al. [19]	47 years-old female	HCT	2.0×1.0 cm	Irregular	Not reported	Right MS; no contact with the walls	Heaviness on the left side of the face since two months; foul smelling purulent nasal discharge	Endoscopic sinus surgery
Present study	83 years-old female	CBCT	1.5×1.6 cm	Rounded shape	2253 mm^3	Right MS, close to the floor, anterior and medial walls	Absent	Clinical and radiographic follow-up

treatment [9]. Incidental findings in scans primarily taken for other indications show a high prevalence of calcified findings [6], as noticed in the present case.

Antroliths, first described by Bowerman in 1969, are bodies located in the MS that received deposits of calcium phosphate minerals [10,11]. They form a rigid surface, which can be of endogenous (originating from dental or bone fragments, necrotic and suppurative tissues of the body, accumulation of erythrocytes and mucus) [11,12] or exogenous origin (originating from presence of foreign bodies in MS, such as vegetable matter, paper and cotton, dental implants, and material from dental canal overfilling [13]. Their etiology is undefined, but there are reports that associate Aspergillus sinusitis [14,15], long fungal infections due to immunosuppression, poor sinus drainage with an accumulation of mucus, presence of foreign bodies [10,13], which can be single or multiple [11]. Their prevalence varies between 0.15% and 2.40% [3,13,16] and they are less common than rhinoliths [17].

In imaging exams, the antrolith is described as an asymptomatic single [11] hyperdense image [18] in MS, usually small, with irregular shape, associated with mucosal thickening [3,13] revealed in imaging exams for other purposes [13]. Nass Duce et al. [13] reported a case of antrolith with similar characteristics, with hyperdensity of the edges and a more hypodense interior. The histopathological examination revealed necrotic material inside the lesion [11]. There are some reports of antroliths >1.0 cm in the literature (table I). However, all had an irregular shape, contrary to the present case, in which calcification was very well defined and rounded. As in our case, in only one study [12] the patient was completely asymptomatic. The other authors [11,19,20] reported symptoms such as heaviness on one side of the face; nasal discharge, facial pain and headache. All patients were females aging from 32-47 years.

Also, calcified mucous retention phenomenon was considered. The mucous retention phenomenon is usually asymptomatic, and radiographically dome-shaped, radiopaque, and characterized by the extension of the mucosa originating in the sinus wall [21]. CT scans reveal a non-corticalized hyperdense image with defined limits. The lining of the adjacent membrane is generally not visible. As it is asymptomatic, it is frequently detected in imaging exams for other purposes. Although its etiology is uncertain, some theories are supported in the literature, such as recurrent inflammatory allergic processes, trauma, odontogenic infections, and air humidity [22]. Its prevalence ranges from 5.8% to 36.7% [6].

The presence of a calcified mucous retention phenomenon was considered as a hypothesis in the present case, although the high density of the lesion makes us somewhat discredit this possibility. Vele et al. [22] reported a mucosa-covered globular mass of 4 cm inside the nasal cavity detected in a Waters radiography that was attached to the lateral nasal wall. It was removed by the lateral rhinotomy approach and macroscopic examination of the specimen showed a cyst lined with a paper-thin bony covering all round the inner lining mucosa. Microscopic examination revealed a cystic lesion with its wall showing calcification and osseous metaplasia [22]. Unfortunately, the patient was not submitted to tridimensional exam so that the image could be studied in detail and compared with our case. The calcification of the

mucous retention phenomenon is rare and was reported in only 5% of the cases. In some cases, it can be dense enough to simulate osteomas [23], which may explain the scarcity of reports in the literature. In a CBCT study, Yeung et al. [24] assessed these alterations and did not report calcified images.

Osteoma was initially considered in the present case. This benign, slow-growing neoplasm is more common in the paranasal sinuses than gnathic lesions. The frontal sinus is the most frequently involved, followed by the ethmoidal and maxillary sinuses. Most cases are asymptomatic [25], explaining why they are generally found as incidental findings in imaging studies [26,27]. Symptoms are reported only in cases where the lesion presses and occupies spaces of adjacent structures [26]. However, it hardly exceeds bone limits [27]. Traumatic, inflammatory, and embryological etiologies have been suggested [26,27]. The prevalence of the osteoma is about 2.6% [3]. In the tomographic exam, the osteoma can present several features. A hyperdense, heterogenous or homogeneous image, most likely well circumscribed lesion can be observed [28] and due to its tumoral growth, it is necessarily attached to the origin bone wall, by a broad to narrow osseous pedicle [25]. These conditions led us to classify this as the least likely hypothesis.

Due to the overlapping of radiographic image [29], sinus calcifications can be masked or underdiagnosed in these exams. CT is considered the gold standard exam for evaluation of paranasal sinuses, due to the details in the visualization of the anatomy [30]. In this context, CBCT stands out, as it is the three-dimensional exam of choice for the dentomaxillofacial region. Due to its isotropic volume, it generates less distortion, increasing sharpness, with lower doses of radiation when compared to HCT [13,29]. The absence of overlap, the reconstructed volume that often exceeds the area of interest and the increased demand for exams have increased the number of incidental findings in these exams. The prevalence of incidental findings in MS in CBCT exams varies from 18.8% to 76.1% [3,4,6,29].

In the present case, a hyperdense, rounded shape image was incidentally found in the MS when a CBCT scan was acquired for other purposes. The atypical presentations of sinus calcifications represent a challenge for the radiologist and the clinician. This is made even more difficult when the radiologist does not have the patient's clinical information. It is known that only a microscopic examination can confirm the diagnosis. However, in some cases, the absence of clinical symptoms or signs and the patient's condition postpone or contraindicate surgical removal, leaving only the presumptive diagnosis based on the imaging exam. Within the spectrum of differential diagnoses, our radiographic diagnosis for the case is an antrolith, although the possibility of a calcified mucous retention phenomenon or osteoma cannot be completely ruled out. For these diagnostic hypotheses, no treatment is necessary in the absence of symptoms. We emphasize the need for biopsy and histopathological examination to define the diagnosis, but the absence of complaints and the patient's age encourage us to adopt clinical-radiographic monitoring for the case. Regardless of the final diagnosis, the finding is certainly rare and deserves disclosure.

Collaborators

M HERREIRA-FERREIRA, image acquisition and conception of the case. GN SOUZA-PINTO and ES ESTOLENTINO, analysis and interpretation of the image. M CHICARELLI, drafting of article and critical revision. LCV IWAKI, drafting of article, critical revision and final approval.

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