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CASE REPORT

Diagnosis of ethmoid sinolith by cone-beam computed tomography: case report and literature review

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Abstract

Introduction The aim of this study was to report the first case of diagnosis of a rare ethmoid sinolith by cone-beam computed tomography (CBCT) and discuss the importance of carefully assessing the entire volume of the images regardless of the region of interest.

Case presentation A 22-year-old woman underwent CBCT examination to assess the upper permanent canine teeth. Analysis of the entire volume of CBCT revealed an oval-shaped hyperdense, homogeneous, expansive formation with defined boundaries located in the ethmoid bone. The differential diagnosis of a sinolith in the ethmoid sinus was confirmed after removal of the lesion by nasoendoscopy and histopathological examination.

Conclusion The diagnosis of lesions in the ethmoid sinus is challenging because of the complex anatomy of this region. Considering the limitations of two-dimensional X-ray examination, CBCT examination has become an important imaging modality for the diagnosis of these calcifications. In addition, the ability of the dental surgeon to assess the entire volume of the CBCT images and the knowledge of imaging features of these calcifications allows for recognition of incidental findings, facilitating early diagnosis and appropriate treatment.

Keywords Cone-beam computed tomography · Ethmoid sinus · Paranasal sinus

Introduction

The paranasal sinuses are air spaces delimited by the nasal cavity and are divided into the maxillary, frontal, ethmoidal, and sphenoidal sinuses. The paranasal sinuses are rarely affected by calcification, but when present, these calcifications assume different names depending on their location. Sinolith is the term used to describe the presence of calcification in the frontal, ethmoidal, and sphenoidal sinuses, while rhinolith and antrolith are terms used to describe the presence of calcifications in the nasal cavity and maxillary sinus, respectively. Rhinoliths and antroliths are quite common, while sinoliths are extremely rare, especially when located inside the ethmoid sinus [1, 2]. Sinoliths can be single or multiple, generally have a hardened and uneven consistency, and vary in size [3]. Although their etiopathogenesis is unclear, some possible predisposing factors are chronic infection, the presence of a foreign body, poor drainage, and sinus fungal infection [1–5].

Computed tomography (CT) is the imaging technique of choice to locate and assess such calcifications and their relationship with the surrounding tissues [1, 4, 5]. An alternative to CT for the oral and maxillofacial region is cone-beam CT (CBCT), the advantages of which include less exposure of patients to radiation and lower cost; in addition, CBCT provides good images of hard tissues equivalent to those provided by CT [6, 7].

To the best of our knowledge, the data available to date on the occurrence of sinoliths in the ethmoid sinus are quite scarce, and diagnosis has only been obtained by CT, highlighting the importance of reports aimed at educating

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clinicians regarding the imaging characteristics of these calcifications [1, 2, 4]. The aim of this study was to report the first case of diagnosis of a sinolith in the ethmoid sinus diagnosed by CBCT and discuss the importance of carefully assessing the entire volume of the images regardless of the region of interest.

Case report

A 22-year-old woman was referred to a dental radiology clinic for a CBCT examination to assess the upper canines (teeth 13 and 23), which were retained. Thus, the patient underwent CBCT examination for dental reasons, because this examination is specific for the dentomaxillofacial region, and CBCT is used much more frequently than CT in dental clinics.

The CBCT examination was conducted on an i-CAT® CB500 system (Imaging Sciences, Hatfield, PA) under the following acquisition protocol: 120 kVp, 5 mA, voxel size of 0.25 mm, and field of view of 13 × 8 cm. In the axial, sagittal, coronal, and three-dimensional CBCT reconstructions (Figs. 1, 2), we observed not only the retained canines, but also the presence of an oval-shaped hyperdense, homogeneous, expansive formation of calcium density. The formation had defined boundaries, measuring 11 × 9 × 4 mm at its greatest extent and occupying the ethmoid cells anterior to the right side, close to the medial aspect of the right lamina papyracea. The presence of the calcium formation resulted in light reduction in the frontonasal channel, but without significant obstruction of its light. We also verified veiled ethmoid cells anterior to the right side, adjacent to the lesion, and in close proximity to the medial wall of the orbit on the right side and to the nasal cavity. The volume of the calcium formation was 247.438 mm^3, calculated using the software Insight ITK-SNAP version 2.4.0 (http://www.itksnap.org) [8]. During assessment of the patient's clinical signs and symptoms, the patient reported nasal obstruction and constant coryza. She did not have a medical history of sinusitis, surgery in the sinuses, or trauma in the region. After assessment of the images and correlation of the imaging findings with the clinical signs and symptoms, our main differential diagnosis was a sinolith in the ethmoid sinus. The patient was transferred to an otolaryngologist for a more detailed assessment. After nasoendoscopy, the calcium formation was removed and the histopathological examination confirmed a sinolith, free of fungal contamination. At the time of this writing, the patient had been undergoing dental monitoring for 2 years to check for signs of recurrence.

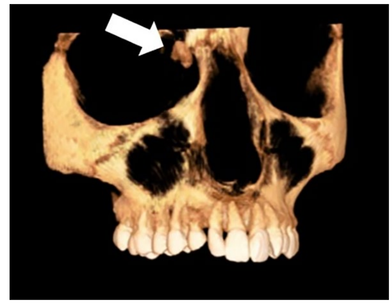

Fig. 2 Three-dimensional cone-beam computed tomography reconstructions showing the sinolith (white arrows)

Discussion

The occurrence of calcification in the paranasal sinuses is unusual. When present, it is most prevalent in the maxillary sinuses, while it is extremely rare in the ethmoid sinus [1, 2]. Calcifications in the paranasal sinuses may be classified according to their origin as either exogenous (formed from foreign bodies such as cotton and paper) or endogenous (resulting from complications after extractions, blood clots, purulent secretions, mucus, desquamated epithelium, bone fragments, and *Aspergillus* fungi) [1–5]. Some studies have affirmed that ethmoid sinoliths are predominantly of endogenous origin [2].

The development of these calcifications depends on two factors: one is the presence of chronic infection with accumulation of pus and deposition of salts, and the other is poor sinus drainage that leads to the accumulation of secretions,

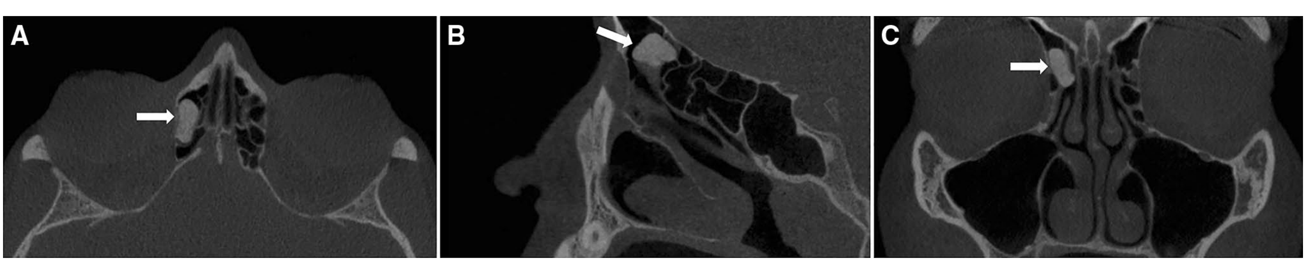

Fig. 1 Cone-beam computed tomography reconstructions showing the sinolith (white arrows). **a** Axial section. **b** Sagittal section. **c** Coronal section

which serve as a focus for sinolith formation [2, 5]. Other suggested that mechanisms include accumulation of calcium salts in foreign bodies, trauma, radiation therapy, or neoplastic lesions. In addition, ectopic bone formation stimulated by repeated polypectomies may result in the formation of calcification in the ethmoid sinus [1, 3].

After an extensive review of the literature in the PubMed database using the keywords “sinolith”, “paranasal sinus”, and “ethmoid sinus”, we found only three studies involving four cases of the presence of a sinolith in the ethmoid sinus, all published in the English language [1, 2, 4]. The main comparisons between the present case and earlier reported cases are shown in Table 1. In the previous cases, the size of the sinoliths ranged from a diameter of 10 mm [4] to an area of $30 \times 25 \times 29$ cm^2.

In the present case, the size of the sinolith was $11 \times 9 \times 4$ mm, which is equivalent to the diameter of the sinolith reported by Kanzaki and Sakamoto [4]. Although the size of the calcification found in this case is close to that reported previously [4], we cannot conclude that this is the average size of sinoliths in the ethmoid sinuses, because the number of cases reported in the literature is extremely limited. With respect to location, previously reported sinoliths were found in the left anterior ethmoid sinus, adjacent to the left lamina papyracea ethmoid [4]; in the bulla ethmoidalis and anterior ethmoid cell, attached to the left lamina papyracea [1]; in the right anterior ethmoid cell [2]; and, in the present study, in the right anterior ethmoid cell, attached to the medial aspect of the right lamina papyracea. In all the previous reports, the diagnosis was obtained by CT [1, 2, 4]. Thus, the present case is the first to be diagnosed by CBCT.

Regarding the imaging examination, CT is considered the gold standard for evaluation of the paranasal sinuses [9, 10], because it allows for detailed visualization of the anatomy in this region and the presence of calcifications in the paranasal sinuses, in addition to their location, size, and relationship with adjacent structures [2, 5]. Such examination is not possible with conventional X-rays because of expansion and overlapping structures [6]. Thus, the overlapping of structures inherent to the radiographic examination and the complex anatomy of the ethmoidal sinus makes it more difficult to visualize calcifications present within the ethmoidal sinus and the precise delimitation of calcifications with adjacent structures using conventional X-rays. Moreover, because the sinolith was small and close to the walls of the interior aspect of the ethmoidal sinus in the present case, its diagnosis by radiographic examination would have been even more difficult.

As an alternative to CT, CBCT is another three-dimensional examination technique that has aroused interest in in the field of otolaryngology [11, 12]. CBCT is used to evaluate structures of the maxillofacial region using small voxel sizes [13], resulting in higher spatial resolution and image quality for bone tissues than CT [10, 13] as well as lower exposure of patients to radiation [10–12]. However, despite the advantages of CBCT, CT is still preferred for large lesions and those with soft tissue involvement because of the lower contrast resolution of CBCT for soft tissue [11, 12].

In the present case, CBCT was not the examination technique of choice for diagnosis of the sinolith, because the examination was requested for dental purposes; the sinolith was an incidental finding. However, because the area of calcification was small, CBCT had the advantage of producing high-quality images, allowing for identification of the sinolith and precise delimitation in relation with the walls of the ethmoidal sinus. This was advantageous in terms of surgical planning and prevention of complications involving adjacent structures (margin of the orbit and nasal cavity).

Notably, because CBCT images of the anatomy of the paranasal sinuses are best visualized in the coronal plane [14], we encourage professionals to use this reconstruction in their evaluations and use other planes (sagittal and axial) as complementary images. Thus, in the present case, it was possible to see the proximity of the calcification to the margin of the orbit and nasal cavity in the coronal images. We also performed three-dimensional reconstruction of the sinolith using the software ITK-SNAP version

Table 1 Case reports of sinoliths in the ethmoid sinus

Study (references)	Case number	Image modality	Sinolith size	Sinolith volume	Location
Kanzaki and Sakamoto [4]	1	CT	10 mm in diameter	Not reported	Left anterior ethmoid sinus, adjacent to the left lamina papyracea
Almasi et al. [1]	2	CT	Not reported	Not reported	Bulla ethmoidalis
	3	CT	Not reported	Not reported	Anterior ethmoid cell, attached to the left lamina papyracea
Nayak et al. [2]	4	CT	$30 \times 29 \times 25$ mm	Not reported	Right anterior ethmoid cell
Present case	5	CBCT	$11 \times 9 \times 4$ mm	247.438 mm^3	Right anterior ethmoid cell, attached to the medial aspect of the right lamina papyracea

CT computed tomography, *CBCT* cone-beam computed tomography

2.4.0 [8], obtaining the volume of the sinolith; this was not performed in any of the other cases reported in the literature.

Because CBCT allows visualization of structures without overlap and may include a larger area, the potential to identify incidental findings is greater. Incidental lesions may be found in imaging tests outside the region of interest and can be unrelated to the indication for performing the examination, but still have clinical relevance; they are also often associated with recurring signs and symptoms, as in the present case [6, 7, 15, 16].

In previously reported cases of sinoliths in the ethmoid [1] and sphenoid [3] sinuses, the presence of calcification was an incidental finding, as in the present report. According to Edwards et al. [7], the occurrence of incidental findings in the paranasal sinuses is quite frequent, indicating the importance of assessing the entire volume of data to achieve a complete diagnosis, regardless of the region of interest.

The ability to identify these findings is associated with the skill level and experience of the radiologist and the performance of a thorough systematic observation of every image, including structures adjacent to the maxillomandibular complex. Thus, to correctly interpret CBCT images, the radiologist must master the radiographic aspect of the maxillofacial anatomical structures in this modality to understand the spatial relation of volume and to become aware of possible pathological changes, anatomical variations, and anomalies that allow for a correct diagnosis [15]. This illustrates the importance of reports of rare cases, such as the present case, to alert surgeons to the radiographic aspects of incidental findings with clinical relevance to the patient.

In this case, the clinical finding that helped to confirm our differential diagnosis was the nasal obstruction and coryza reported by the patient, as in the previous cases of sinoliths in the ethmoid and sphenoid sinuses [1–4]. Other previously reported symptoms that were not found in the present case include headache, nosebleed, and facial pain [2, 4].

The treatment of sinoliths usually involves surgical removal through endoscopy. This method is preferred and was used in the present case, because it allows for easy access to and good visualization of the lesion is associated with low morbidity, and has a good prognosis without the need for additional treatment [1–5].

Importantly, in addition to the diagnostic function of CBCT for lesions and diseases present in the paranasal sinuses, CBCT examination is crucial to guide the surgeon in preoperative planning, because the anatomical relationships are precisely identified. This helps to avoid complications and increases the safety to the patient [15].

The differential diagnoses of ethmoid sinus calcifications on CBCT images include osteoblastoma, osteoma, ossifying fibroma, fibrous dysplasia, and calcification in the mucous membrane [2, 3, 5]. In the present case, the histopathological examination confirmed our suspicion of a sinolith, eliminating the risk of misdiagnosis.

The diagnosis of lesions in the ethmoid sinus is challenging, because of the complex anatomy of this region. The limitations of two-dimensional X-rays caused by overlapping of structures make CBCT the imaging modality of choice for the diagnosis of these calcifications. CBCT allows detailed, highly accurate visualization of the interior aspect of the paranasal sinuses. In addition, the ability of the dental surgeon to assess the entire volume of the CBCT images and the knowledge of the specific imaging characteristics of these calcifications allows for recognition of incidental findings, facilitating early diagnosis and appropriate treatment.

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Compliance with ethical standards

Conflict of interest Eliana Dantas da Costa, Francielle Silvestre Verner, Priscila Dias Peyneau, Deborah Queiroz de Freitas, and Solange Maria Almeida declare that they have no conflict of interest.

Human rights statements All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1964 and later versions.

Informed consent Informed consent was obtained from the patient for being included in the study.

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