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Case Report

Ostiomeatal complex inflammation with a rare ethmoid sinolith utilizing cone-beam computed tomography: A clinical and radiological approach to diagnosis ☆,☆☆

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ABSTRACT

A 28-year-old female patient was referred for panoramic radiography during a regular dental check-up. The dentist pointed out an additional suspicion of odontogenic maxillary sinusitis as she had complained of nasal obstruction, nasal discharge, postnasal drip, and frontal headache at the time. In this present case, cone-beam computed tomography (CBCT) imaging modality was utilized to evaluate the paranasal sinuses and detect any pathologic signs. This study aims to highlight the potential value of the modality for the identification of paranasal sinus diseases by presenting a rare finding of an ethmoid sinolith associated with a persistent ostiomeatal complex inflammation. The insufficient data currently available on the incidence of ethmoid sinoliths emphasize the significance of reports intended to inform practitioners about the imaging properties of these calcifications. To the author's

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knowledge, this is the first case report that demonstrated the primary utilization of dental CBCT in detecting ethmoid sinolith in a straightforward manner.

Introduction

Sinolith is a calcified mass that presents in paranasal sinuses, primarily in the maxillary sinus which is also known as an antrolith, rhinolith or antral stone. Literature findings of sinolith in other sinuses, such as in the frontal, ethmoid, and sphenoid sinuses, are still limited, thus making it an extremely and consequently uncommon entity [1–3].

Cone beam computed tomography (CBCT) is a diagnostic imaging modality that is the three-dimensional (3D) imaging of choice for the oral and maxillofacial region. CBCT has been increasingly used in clinical dentistry as it offers a simple, faster, and cheaper yet competitive way of examination compared to medical CT. Through lower doses of radiation and good visualization of hard tissues comparable to that provided by CT, CBCT can also be used to provide a whole sinus topography assessment [4]. In a 2017 study on imaging considerations for different sinonasal diseases, CBCT was recommended by the American College of Radiology as 1 practical diagnostic imaging tool [5]. However, the available sinus calcification cases that have been published, including sinoliths, were often identified using CT as it is considered to be the gold standard for the diagnosis and preoperative evaluation of any sinonasal diseases [6,7]. To our knowledge, only 1 scientific significant database-indexed study has documented the presence of ethmoid sinolith in CBCT imaging, in which the scan was indicated for other purposes and the entity was found as an incidental finding [1]. In this present case, we used this modality to primarily evaluate the paranasal sinus and detect any pathologic diseases within.

This study aims to highlight the potential value of the CBCT modality for the identification of any paranasal sinus diseases by presenting a rare example of an ethmoid sinolith associated with persistent ostiomeatal complex inflammation. The insufficient data currently available on the incidence of ethmoid sinoliths emphasize the significance of reports intended to inform practitioners about the imaging properties of these calcifications.

Case report

This case report was prepared and written in accordance with the CARE reporting guidelines for case reports [8] in order to support accuracy and transparency in the dissemination of case reports and the reporting of data from patient interactions.

A 28-year-old female patient was referred to the radiology department of Universitas Padjadjaran Dental Hospital after a dental check-up at the same hospital for a panoramic radiography examination. Besides the intention of checking the dentition and previous restorations, the dentist also pointed out an additional suspicion of odontogenic maxillary sinusitis as she had complained of nasal obstruction, nasal discharge, postnasal drip, and painful frontal headache at the time which was increased in head down position and when being exposed to cold temperature.

During the evaluation, the patient reported that she had been experiencing all symptoms for the last 3 months despite occasionally taking over-the-counter cold and flu medications without any previous expert consultations. Any medical history of past or recurrent sinusitis, allergic rhinitis, infectious diseases, sinus trauma, or sinus surgery were denied. No notable pathologic signs were observed extra-orally during clinical evaluation while the intraoral assessment recorded some missing teeth, an impacted #38 (left mandibular third molar), caries on #46 (right mandibular first molar), and dental restorations on #27 (left maxilla second molar) and #47 (right mandibular second molar).

Unexpectedly, the panoramic radiograph did not reveal any specific abnormalities relating to the patient's major complaint (Fig. 1). Although there was a slight difference in opacity between the internal structures of the left and right maxillary sinuses, uncertainty still arose as to the lack of evident pathologic indications, and both sinuses were normal in size and had intact cortical boundaries. Due to the unspecified findings and the overlap of anatomical structures in the panoramic image, the patient had been subsequently recommended to undergo a 3D CBCT radiography examination for further investigation, using the large field of view (FOV) to evaluate the maxillary and other paranasal sinuses. The patient consented to get the examination after being informed about the justification and the necessity of CBCT's multiplanar assessment.

CBCT scans were performed using the Orthopantomograph OP300 Maxio unit (Instrumentarium Dental, PaloDEx Group Oy, Tuusula, Finland) under the following protocol of acquisition: FOV 13 $\times$ 15 cm, 90 kVp, 4 mA, 9 s, and voxel size of 380 μm (standard resolution). The images were evaluated and reconstructed on OnDemand3D Dental v1.0 Build 1.0.10.7462 (Cybermed Inc, Daejeon, Republic of Korea). Mucosal thickening of the paranasal sinuses was seen in the coronal, sagittal, and axial slicing images. The CBCT radiographs revealed uniform opacities occupying the ostiomeatal complex (ipsilateral/left maxillary, anterior ethmoid, and frontal sinus) with the presence of maxillary sinus air-fluid level. The opacified lumen was also occurred posteriorly to the sphenoid sinus. Additionally, an ovoid radiopaque calcified stone-like entity with the size of 3.4 $\times$ 3.6 $\times$ 4.5 mm was identified in the left anterior ethmoid cell, within the suprabullar ethmoid air cell, and proximate to the fovea ethmoidalis of the cranial base (Fig. 2). The ITK-SNAP version 3.8.0 software (Cognitica, Philadelphia, PA) (http://www.itksnap.org) software was used to perform segmentation and calculate the total volume of the calcification (Fig. 2D), being 54,1587 mm [3]. Based on all these

Fig. 1 – The panoramic radiograph did not show any specific signs related to both patient's complaint and the dentist's suspicion of an odontogenic maxillary sinusitis. The overlap anatomical structures around the sinus complicates the assessment by the radiologist.

findings, a differential diagnosis of ethmoid sinolith associated with sinus inflammation was suggested.

The patient was referred to an otolaryngologist (ENT) for further treatment who had been subsequently prescribed a set of medications of oral antibiotics, nasal decongestants, and saline nasal sprays. A post-treatment CBCT after 1-month duration following the medication with the referral from the ENT was performed to evaluate the sinus. The CBCT images, however, revealed the existing ethmoid sinolith as well as an additional homogenous radiopaque dome-shaped lesion with dimensions of 14 × 19 × 10 mm attached to the superoanterior wall of the left maxillary sinus, suggesting a radiographic impression of a mucous retention cyst with a differential diagnosis of mucocele. The patient was advised to undergo a rhinoendoscopy and surgical intervention to remove the sinolith but was rejected due to its asymptomatic condition. She has been followed up regularly since then. The head-to-head imaging comparison of the first and second CBCT scans of the patient is shown in Fig. 3.

Discussion

Calcification in the paranasal sinuses, particularly an ethmoid sinolith, is a rare phenomenon [1,3]. In the present case, CBCT was primarily used to assess the paranasal sinuses rather than CT, which is the gold standard for evaluation of the paranasal sinuses, since it is the advanced imaging modality available in the dental hospital to evaluate the dentomaxillofacial region. In addition to its image quality, validity, reproducibility, and low radiation dose, the modality has sparked attention in the field of otolaryngology since it clearly demonstrates changes in all paranasal sinuses and their relationship with surrounding structures [9–11]. However, the available sinus calcification cases that have been published, including ethmoid sinoliths, were often identified using CT [6,7,12].

Following a thorough search of the PubMed and Scopus databases, only 1 study was discovered that reported an ethmoid sinolith using CBCT [1]. A more general search within the keywords limited to "sinolith" and "ethmoid sinus" found 3 additional reoirts of ethmoid sinoliths, all of which were detected by CT as the primary diagnostic imaging modality [2,3,12]. We also explored the Google Scholar database to explore the nonindexed references which yielded 2 other reports of ethmoid sinoliths using CBCT from the same author and publication year [13,14]. Of these publications, the CBCT imaging was performed without the intention of assessing the paranasal sinus, hence the lesion was discovered as an incidental finding [1,13,14]. Thus, our study is the first report to utilize dental CBCT straightforwardly to detect the sinolith and assess its associated sinonasal diseases. The main comparison between the present case and earlier reported cases is shown in Table 1. Based on the present and previous documented reports, the ethmoid sinolith could occur across all age groups. There were 8 patients (4 males, 4 females) with a mean age of 45 (range 22-71 years). It should be noted that due to the scarcity of data, it was unable to determine any particular predilection. However, a previous report suggested that sinus calcifications appear to be found more commonly in women and young adults [15]. Clinical symptoms typically displayed may vary, while the patient described in the present case had nasal obstruction, nasal discharge, frontal headache, and postnasal drip that were identical to previous reports [2,3,12]. Other previously mentioned symptoms that were absent in this patient include epistaxis [3], anosmia [2,3], facial pain [3], and dry cough [2].

The present sinolith was located in the left anterior ethmoid cell within the suprabullar air cell. Earlier reports had different sites, ranging from within bulla ethmoidalis [2] and posterior ethmoid cell [13,14], to being attached to the lamina payracea within the anterior ethmoid cell [1,2,12]. Most cases also reported a single and unilateral lesion of sinolith, while only 1 study had a bilateral presence [13]. In terms of size, the sinolith in our case was 3.4 × 3.6 × 4.5 mm, which is close to

Table 1 – Published reports of ethmoid sinolith in electronic databases.

Indexing database	Study	Patient's data			Imaging modality	Sinolith morphology			Radiographic signs of inflammation			
		Age	Sex	Clinical presentation		Site	Size	Shape	Presence	Degrees	Description	
Scopus & PubMed	Kanzaki and Sakamoto [12]	61	M	Nasal obstruction, nasal polyp, 30 y previous bilateral nasal polypectomy	CT	Unilateral: Left anterior ethmoid sinus, adjacent to the left lamina papyracea	Ø 10 mm	Irregular	√	Moderate	Inflammatory signs in the left ethmoid sinus	
	Almaši et al. [2]	52	F	1-mo nasal obstruct ion, postnasal drip, dry cough. No history of sinusitis.	CT	Unilateral: Right middle nasal meatus, bulla ethmoidalis	N/A	Ovoid, smooth-margined	√	Moderate	Mucosal and bony thickening in the right maxillary sinus	
		71	M	Repeated polypectomies, gradual worsening of nasal breathing and loss of olfaction	CT	Unilateral: Anterior ethmoid adjacent to the left lamina papyracea	N/A	Ovoid	√	Severe	Mucosal thickening in ethmoid	
	Nayak et al. [3]	45	M	20-y recurrent nasal obstruction, episodic sneezing spells, headache, nasal discharge, watering eyes and itching in the nose	CT	Unilateral: Right anterior ethmoidal cell blocking the frontal recess	Dimension: 30 × 29 × 25 mm Volume: 21,75 cm^3	Irregular	√	Severe	Mucosal thickenings in all right (ipsilateral) paranasal sinuses with presence of polyps	
	da Costa et al. [1]	22	F	Nasal obstruction, constant coryza	CBCT	Unilateral: Right anterior ethmoid cell, attached to the medial aspect of the right lamina papyracea	Dimension: 11 × 9 × 4 mm Volume: 247,438 mm^3	Oval-shaped, well-defined	X	N/A	Nasal obstruction without any radiographic signs of inflammation	Incidental finding

(*continued on next page*)

Table 1 (*continued*)

Google Scholar	Sava and Rusu [13]	34	F	N/A	CBCT	Bilateral: 1. Left ethmoid: 1 mm inferior to the ethmoid roof within a suprabullar ethmoid air cell 2. Right ethmoid: 1.5 mm beneath the ethmoid roof, within the central posterior ethmoid air cell	1.6 mm^2 (sagittal) 7.6 mm^2 (sagittal)	Ovoid Ovoid	X	N/A	No clinical symptoms and radiographic signs of infections	Indicidental finding
	Sava CJ, et al. [14]	52	M	N/A	CBCT	Unilateral: Right posterior ethmoid, in front of the spheno idal concha	Dimension: 3.24 × 3 × 4.51 mm	Irregular	X	N/A	No symptoms and radiog raphic signs of infections	Incidental finding
Present Case		28	F	3-mo nasal obstruc tion, nasal discharge, postnasal drip, frontal headache	CBCT	Unilateral: Left ante rior ethmoid cell, within suprabullar ethmoid air cell	Dimension: 3.4 × 3.6 × 4.5 mm Volume: 54,1587 mm^3	Ovoid	√	Severe	Inflammation signs with opacities in the left maxillary, ethmoidal sphenoid and frontal sinus	

F, Female; M, Male; N/A, Not available.

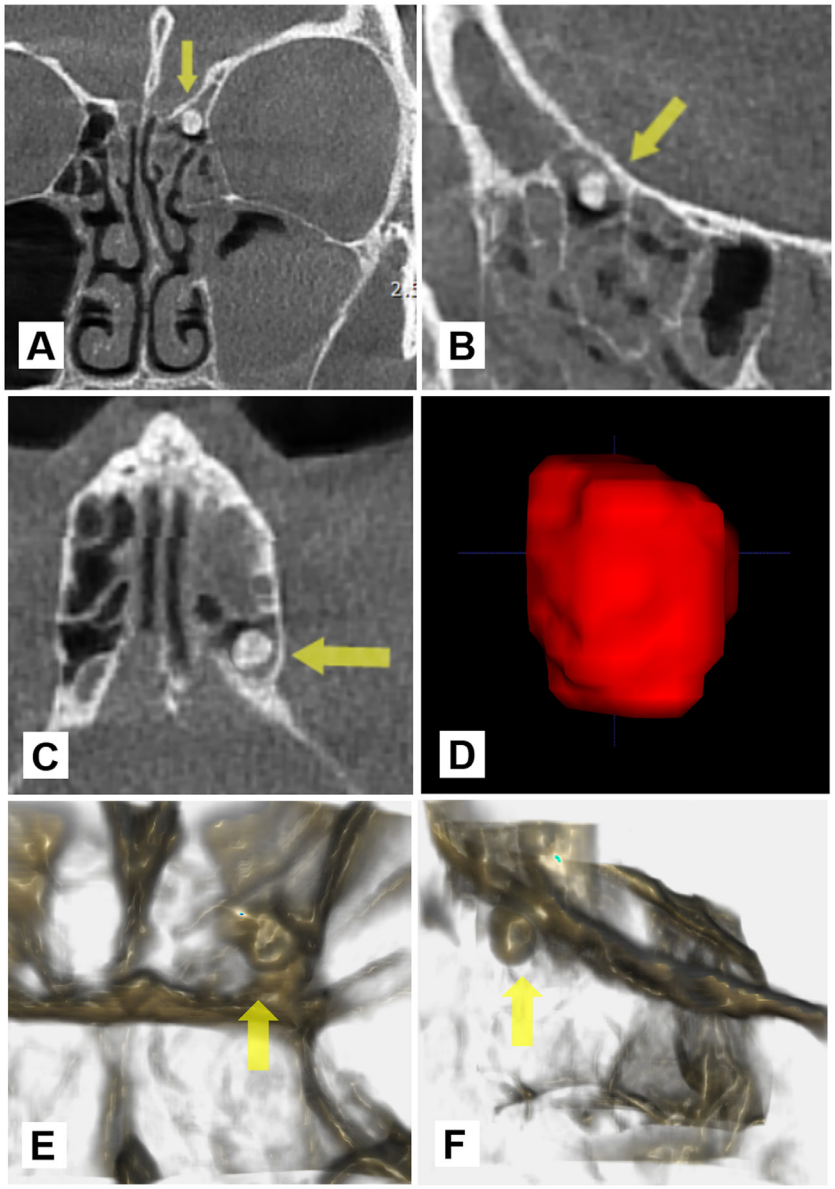

Fig. 2 – The calcified entity which located within the suprabullar cell in the left ethmoid sinus. The (A) coronal, (B) sagittal, and (C) axial view of the lesion on CBCT images also consistently showed opacities and mucosal thickening in the surrounding ethmoid cells. (D) The 3D segmentation of the lesion for volume measurement using ITK-SNAP and the clipped 3D reconstruction of the CBCT images around the calcification in (E) anterior and (F) lateral views.

the diameter of the entity reported by Sava et al. [14]. Despite being comparable, the calcification was discovered in a variety of sizes in other reports. The vast majority of sinoliths were small and appeared as oval to irregularly shaped objects, with the most significant dimension reported being 30 × 29 × 25 mm [3], which is more than 10-fold larger than the one in our case. These findings conform to the previous theory that sinoliths can be single or multiple, vary in size, and typically have a hardened and irregular consistency [16].

Long-standing infections, inadequate sinus aeration and drainage, and fungal infection are thought to be the main predisposing factors for sinolith formation, although the precise etiopathology is not well known [2,12,16]. Other suggested mechanisms include accumulation of calcium in foreign bodies, trauma, radiation therapy, history of repeated polypectomies, and neoplastic lesions [12]. In our situation, it is likely that the sinolith developed as a result of a persistent sinus infection, supporting a prior report that a sinolith might be discovered together with inflammatory alterations in the paranasal sinuses [2]. Although not entirely understood, suppuration can promote the development of sinolith by precipitating the deposition of calcium and other mineral salts in the afflicted area when it coexists with acute and chronic inflammation [12,15].

The intensity, length, and frequency of sinus inflammation were hypothesized to affect the calcification, affecting its shape and size [17]. It was also indicated that severely inflamed sinuses tend to form multiple calcifications [17].

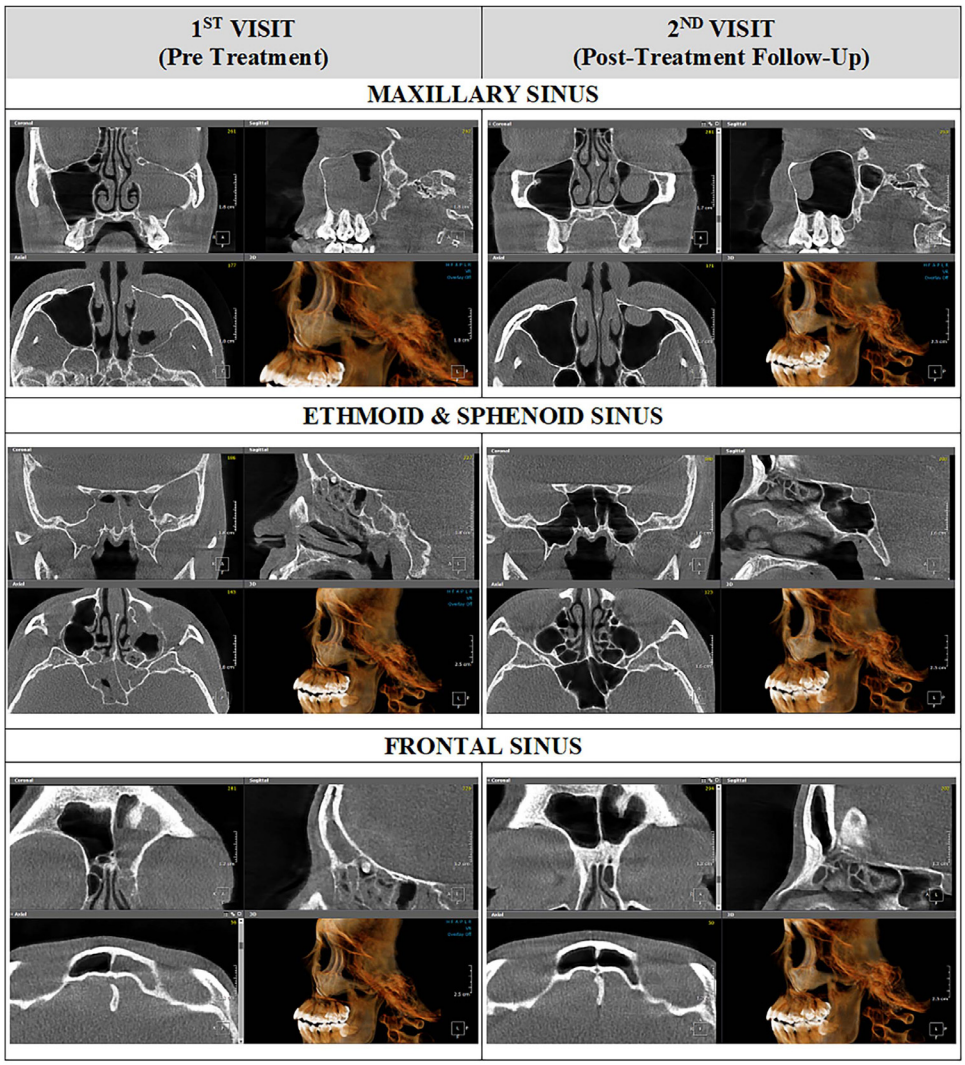

Fig. 3 – Comparison of CBCT examination of all paranasal sinuses of the patient at the first visit (pretreatment) and second visit (post-treatment follow-up). Note the uniform opacification and mucosal lining thickening of all sinuses at the first visit. The clouding had mostly vanished 1 month after receiving the medication, but a secondary dome-shaped radiopaque lesion was discovered on the anterosuperior wall of the existing maxillary sinus.

Based on the theory, we attempted to determine the radiographic signs of inflammation within the relevant sinus in this study and categorize its degree (Table 1) into following grading groups by Cho et al. [17]: (1) mild (less than one-third opacification of the sinus); (2) moderate (from one-third to two-third opacification of the sinus); or (3) severe (greater than two-thirds opacification of the sinus). The largest ethmoid sinolith was found in the report by Nayak et al. [3] with a 20-year background of severely inflamed sinus, thus supporting the theory. As in our patient who similarly had severely inflamed sinus, it had a much smaller size of calcification that we may assume to correlate with the shorter duration of 3 months symptoms. However, the only multiple and bilateral ethmoid sinolith case [13] from all studies showed a contradiction with the absence and no history of sinus inflammation. Limited by the availability of reported cases, more investigation into the interrelationships of other potential variables is required.

Air-fluid levels, mucosal thickening, and opacification of the sinus lumen are the distinctive features of sinus inflammatory disease [7]. Secretions accumulating in a persistently inflamed sinus may remain fluid, become condensed, hyperdense, become over-infected with fungi, or gradually increase in volume [18]. In this study, the homogenous opacified paranasal sinuses within the ostiomeatal unit was noted, which included the middle meatus of the nasal cavity, the adjacent left anterior and middle ethmoid cells, as well as the maxillary and frontal sinuses. This finding significantly pointed to the obstruction and inflammation condition of unilateral chronic sinusitis within all 3 sinuses. Furthermore, clouding was also observed within the sphenoid sinus, indicating that the inflammatory area extended further.

The CBCT imaging obtained at the second visit showed a different entity within left maxillary sinus, which was suggestive of a mucous retention cyst with a differential diagnosis

of mucocele and polyps. Based on this phenomenon, the cyst might have formed as a result of a preceded inflammatory sinus disease. This remark is in line with a previous study by Harar et al. [19] who found that maxillary sinus mucosal cysts appear more prevalent in patients with symptoms of chronic sinusitis, suggesting an etiological relationship.

Identifying any pathologic condition in the ethmoid sinus could be difficult due to its complicated architecture. In this report, we demonstrate the significant diagnostic value of CBCT for sinonasal evaluation. Conventional two-dimensional imaging, such as panoramic radiography, has several limitations in detecting maxillary sinus diseases and related areas due to overlapping anatomical structures and the complex anatomy of paranasal sinuses. Conversely, the multiplanar and 3D reconstruction images in CBCT radiographs was highly useful in locating any pathologic changes precisely within the paranasal sinuses.

Due to its isotropic voxels, CBCT is also known to generate less distortion and high-fidelity image reconstruction, increasing sharpness, with lower doses of radiation when compared to CT [1,20]. As inflammatory sinus disease is frequently recurrent and the fact that other circumstances may necessitate the need for repetitive imaging requests, makes CBCT appropriate in such cases and provides superior advantages for iterative evaluation, postoperative follow-up, and pediatric exploration [4]. On this basis of radiation optimization, we strongly encourage professionals and ENT specialists to consider adopting CBCT in their work. A previous survey conducted by Lata et al. [11] showed a significant gap in knowledge among ENT specialists regarding CBCT and its various applications, owing to the fact that CBCT is a relatively advanced and recent imaging technique that was not covered in the curriculum during their study, thus an in-depth introduction and understanding to the modality's usefulness in the field of ENT is still required. However, because of its limitations in terms of soft tissue contrast and density resolution, it should be noted that CT and MRI examinations are best suited for any lesions involving soft tissues [1].

Within the range of possible diagnoses, our final radiological diagnosis in this case is a sinolith. Several other possible differential diagnoses, including osteoma or bony exostosis, might be partially ruled out as the calcified mass was not attached to any of the walls of the ethmoid sinus or any bones in this particular case, based on the analysis of CBCT multiplanar images. The potential of a sinus calcification-mimicking osteoma, in which the osteoma might have been the initial disease and was dislodged due to certain sinus cavity conditions such as history of trauma, as reported in previous report [21], was also denied by the patient within our case. As the density of the mass in this case was similar to bony structure, it is possible that the sinolith originated with the nidus being bone rather than mucous, as is commonly observed. In some cases, including our case, the absence of clinical symptoms, along with the patient's better health condition, might delay or rule out surgical intervention, leaving only the presumed diagnosis based on the imaging test. We already highlighted the importance of a biopsy and histological analysis to establish a diagnosis, but because there were no more clinical symptoms, the patient did not feel the urge and rejected the plan of surgical removal. The patient was subsequently instructed to have periodic imaging to evaluate the lesion. Regardless of the eventual diagnosis, the discovery of this entity using CBCT is obviously rare and should be publicized.

Conclusion

CBCT 3D could serve as a first-choice advanced diagnostic imaging tool, an alternative to CT but with less radiation, for a complex case that cannot be adequately visualized with a conventional 2D radiograph, such as assessing paranasal sinuses. In the present case, the inflammation of the ostiomeatal complex, the ethmoid sinolith, and the pseudocyst, were all clearly apparent and accurately interpretable from the CBCT's multiplanar and 3D-reconstructed images, hence proving its promising diagnostic efficacy in otorhinolaryngology for identifying any sinonasal diseases.

Disclosure

The full copy of medical record and informed consent have not been made available due to hospital rules and patient's privacy. Any other data used to support the findings of this study are available from the corresponding author upon request.

Patient consent

Informed consent was obtained from the patient to be included in this study.

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