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Cone-beam computed tomographic imaging of silent sinus syndrome: A case series and a literature review

Nisha G. Manila[1,*], Mehrnaz Tahmasbi Arashlow[1], Scott Ehlers[1], Hui Liang[1], Madhu K. Nair[1]

[1]*Department of Diagnostic Sciences, Texas A&M University College of Dentistry, Dallas, TX, USA*

ABSTRACT

While silent sinus syndrome (SSS) is familiar to otolaryngologists and ophthalmologists, it is a rare clinical entity in dentistry and is likely to be underdiagnosed due to dentists' lack of awareness of this condition. SSS presents a diagnostic challenge to dentists, as patients typically have no history of trauma or sinusitis. The characteristic feature of SSS is a gradual retreat of the maxillary sinus walls, resulting in enophthalmos and hypoglobus. Multidetector (multislice) computed tomography is the imaging modality of choice for SSS and other paranasal sinus diseases. Cone-beam computed tomography promises to be an alternative low-dose imaging modality. This report describes 3 cases of SSS in adults, who had no identified clinical symptoms except diminutive and opacified maxillary sinuses, as well as the inward bowing of the sinus walls as noted on cone-beam computed tomographic imaging. ***(Imaging Sci Dent 2020; 50: 365-71)***

KEY WORDS: Paranasal Sinus Diseases; Maxillary Sinus; Cone-Beam Computed Tomography; Multidetector Computed Tomography

Silent sinus syndrome (SSS) is a well-known entity in otolaryngology, originally named by Soparker et al. in 1994.[1] In that study, the authors presented a group of 14 patients with spontaneous enophthalmos and hypoglobus with maxillary sinus opacification and collapse, along with a lack of history of related trauma or surgery. Earlier, in 1964, comparable observations were published by Montgomery.[2] Also referred to as imploding antrum and chronic maxillary sinus atelectasis, SSS is characterized by painless enophthalmos and inward retraction of the maxillary sinus walls as detected on imaging studies.[1] Many such cases have since been identified and reported in the otolaryngology and ophthalmology literature. However, this condition is generally unfamiliar to dentists, and it is thus seldom reported in the dental literature despite being observable on advanced imaging studies acquired for other oral and maxillofacial diagnostic purposes.

SSS is a relatively rare clinical condition that is likely under-reported due to dentists' lack of awareness and its unique radiographic appearance.[1] This is particularly important when surgery is planned in the area of interest. SSS generally presents in the third through fifth decades of life.[3] The term SSS is often used interchangeably with chronic maxillary atelectasis; however, SSS is characterized in particular by the absence of chronic sinusitis.[4] Although SSS can be initially identified using clinical features, the final diagnosis must be confirmed radiographically.

The most common tool used for 3-dimensional imaging is multidetector computed tomography (MDCT).[5] Two fundamental problems with this imaging modality are the relatively high dose of radiation administered and the relatively high cost.[5] In contrast, cone-beam computed tomography (CBCT) produces images with high spatial resolution (approaching 76 μm) with a much lower radiation burden.[5] Additionally, artifacts from metallic dental restorations are less apparent with this technique. Veldhoen et al.[6] compared MDCT and CBCT and indicated that, at a lower radiation dose, CBCT provided better subjective image quality and resolution. Bacher et al.[5] showed that, independent of imaging technique, the salivary glands, brain, and thyroid received the highest equivalent dose during paranasal sinus

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*Correspondence to : Dr. Nisha G. Manila
Department of Diagnostic Sciences, Texas A&M University College of Dentistry, 3302 Gaston Ave, Dallas, TX 75246, USA
Tel) 1-214-828-8479, E-mail) nishomfr@tamu.edu

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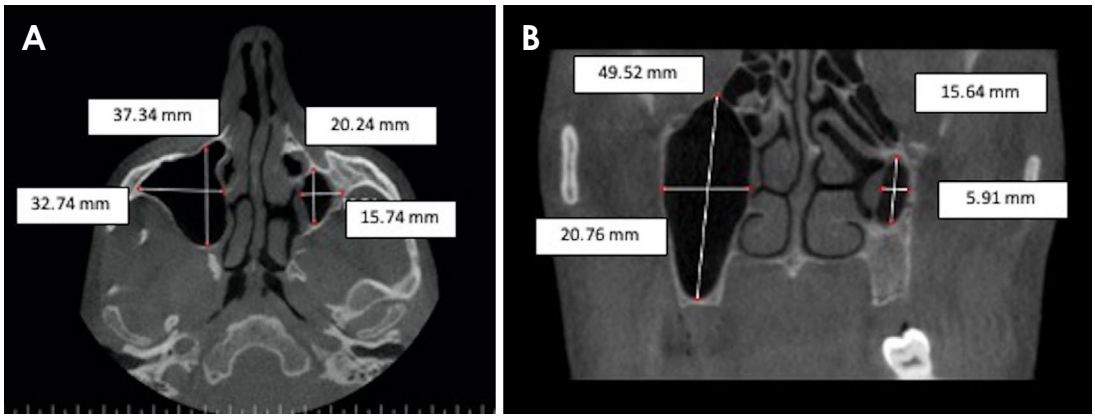

Fig. 1. Case 1. Axial (A) and coronal (B) images demonstrate left maxillary sinus volume loss relative to the right side.

imaging; the effective dose with CBCT was 30 μSv, whereas low-dose and standard MDCT protocols resulted in effective doses of 200 μSv and 1200 μSv, respectively. The comparison between MDCT and CBCT remains controversial in many settings due to the latter's lack of capacity to adequately image soft-tissue entities. However, justification for using a higher-dose examination in the absence of associated soft tissue pathoses is difficult. The judicious use of radiation is advocated by the American Dental Association and the U.S. Food and Drug Administration, as well as all dental specialty organizations through their support of the Image Wisely campaign. It is important to keep doses low while maintaining image quality.[7] The purpose of the present report was to describe relatively less well-known imaging features of SSS and to advocate for the use of CBCT for diagnostic imaging purposes.

Case Reports

A retrospective review of clinical and imaging data from between March 2019 and October 2019 was performed using the patient database at our institution. Patients' demographic data, clinical presentation, and radiographic findings were reviewed. The studies were analyzed with regard to the overall appearance of the maxillary sinuses, including the overall volume, the configuration of cortical contours, and the appearance of the sinus infundibulum and the uncinate process. Adjacent structures including the orbits, middle meatus, and middle turbinates were also evaluated. No history of trauma, sinus surgery, or space-occupying masses of any kind was reported. CBCT studies were performed using a dedicated maxillofacial CBCT unit (iCAT FLX; Imaging Sciences International, Hatfield, PA, USA), at 120 kVp, 5 mA, and a 20-second exposure time. The size of the field of view was 16 cm × 13 cm for case 1 and 23 cm × 17 cm for cases 2 and 3. All images were acquired and viewed as Digital Imaging and Communications in Medicine files by board-certified oral and maxillofacial radiologists with several years of experience reading such studies. These images were saved in JPG format with a resolution of 300 dots per inch.

For all cases, measurements were made on the images in which the smallest dimensions were observed in all planes of interest for the purpose of comparison with the normal side. These measurements illustrate the diminutive size of the affected sinus in views orthogonal to the x-, y- and z-axes.

Case 1

A 32-year-old man was referred for evaluation of an unerupted left maxillary tooth. Physical examinations of the head and neck region revealed no abnormal findings. All of the patient's vitals were within normal limits, and no other abnormal extraoral findings were detected. A CBCT study revealed a relatively small left sinus with its superior third positioned lower than its anatomic counterpart along the x-axis. The left orbital floor appeared to be retreating into the maxillary sinus, with its cortical integrity maintained. Concha bullosa was observed on the right, with mild deviation of the cartilaginous portion of the nasal septum. The ostiomeatal complex appeared to be well-aerated and patent with no evidence of occlusion (Fig. 1).

Case 2

A 19-year-old man was referred for evaluation of the temporomandibular joints as part of orthodontic diagnosis and treatment planning. Physical examination of the head and neck region revealed no abnormal findings. All of the

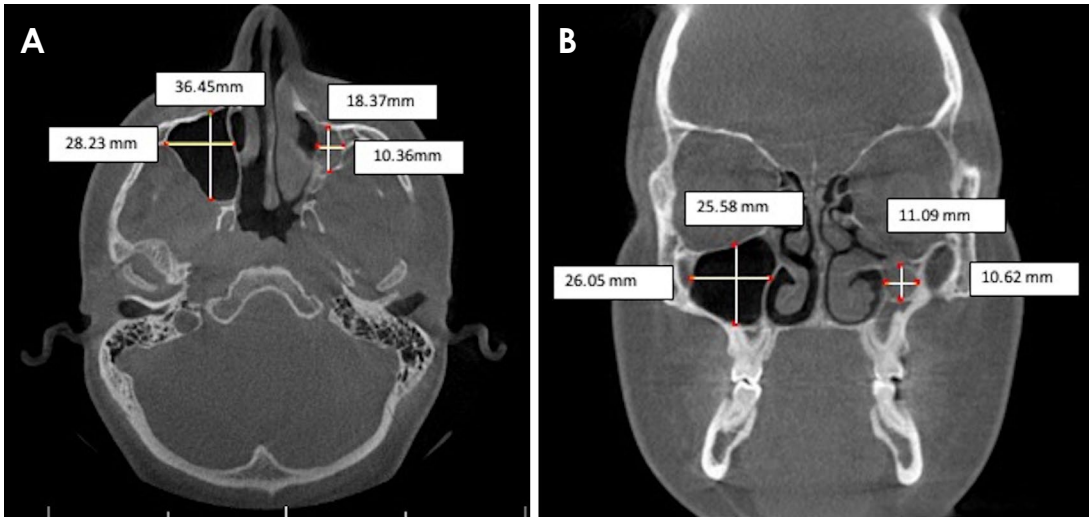

Fig. 2. Case 2. Axial (A) and coronal (B) images demonstrate a smaller and completely opacified left maxillary sinus.

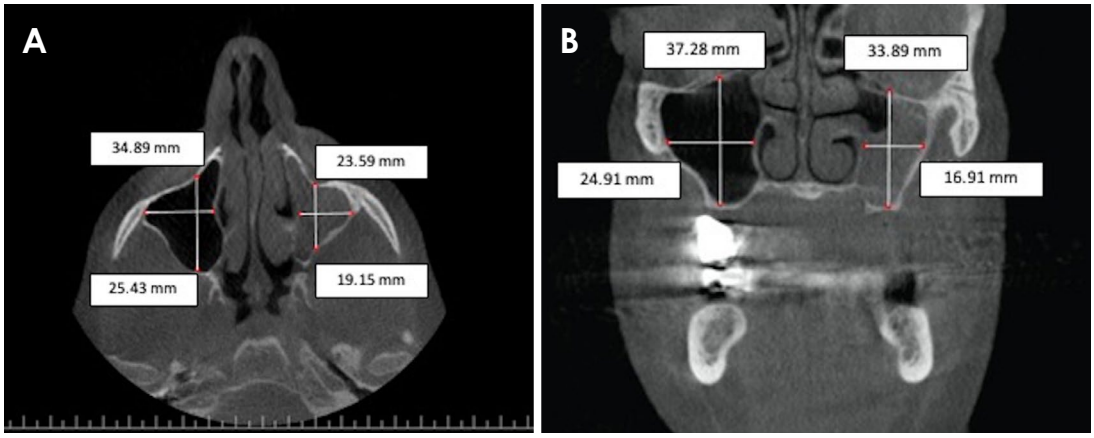

Fig. 3. Case 3. Axial (A) and coronal (B) images demonstrate opacification and inward retraction of the walls of the left maxillary sinus.

patient's vitals were within normal limits, and no other abnormal extraoral findings were detected. A CBCT study showed a hypoplastic left maxillary sinus, with a soft-tissue density filling the sinus. The anterolateral wall of the left sinus appeared to be retracted posteriorly. In addition, the floor of the left orbit (and the roof of the left maxillary sinus) was displaced inferiorly. On the affected side, the patency of the ostiomeatal complex could not be established (Fig. 2).

Case 3

A 69-year-old patient (sex not stated) was referred for the evaluation of suspected pathosis involving a cloudy left maxillary sinus noted on a panoramic study. Physical examination of the head and neck region revealed no abnormal findings. All of the patient's vitals were within normal limits, and no other abnormal extraoral findings were detected. CBCT was performed to evaluate a potential underlying lesion and showed a relatively small left maxillary sinus with inward bowing of the anterolateral wall. Retraction of the medial wall into the sinus cavity was also noted. The lateral border of the sinus appeared sclerotic. The anterior floor of the left bony orbit appeared to be positioned inferior to its anatomic counterpart on the right. The left middle meatus was wider than the right, and the uncinate process was thin and appeared to be retracted into the orbital wall, obstructing the left maxillary sinus infundibulum. Mild mucosal thickening was noted within the right sinus. The patient had no symptoms that could be attributed to clinical sinusitis. No fluid accumulation was noted (Fig. 3).

Table 1. Literature review of imaging of paranasal sinus pathoses

Author	Objective	Imaging modality	Findings
Bacher et al. 2009[5]	To compare the effective dose associated with CBCT and MDCT	CBCT and MDCT	Paranasal sinus imaging with CBCT and digital tomosynthesis showed a large reduction in dose compared to low-dose and standard MDCT.
Veldhoen et al. 2017[6]	To compare MDCT and CBCT with regard to radiation dose, resolution, image noise, and image quality	CBCT and MDCT	Using the low-dose protocol, the assessed CBCT provided better objective and subjective image quality and equal resolution. Similar image quality, but better resolution was observed for CBCT than MDCT at higher exposure settings.
Casselman et al. 2013[7]	To assess the importance of non-dental CBCT indications	CBCT	Non-dental CBCT studies gradually replaced conventional X-rays and CT/ MDCT studies because they allowed imaging with higher resolution, lower radiation dose, and fewer metal artifacts.
Almashraqi et al. 2017[15]	To assess the radiation dose and image quality associated with different low-dose MDCT and CBCT imaging protocols in comparison with the standard MDCT protocol for maxillary sinus imaging	CBCT and MDCT	The low-dose MDCT and CBCT protocols are viable methods for maxillary sinus examination that yield a good diagnostic image quality using effective doses approximately 7 and 11 times lower than that of standard MDCT, respectively.
Xu et al. 2012[16]	Technical assessment of a new CBCT scanner (CS 9300; Carestream Health, Rochester, NY, USA) for applications in otolaryngology-head and neck surgery	CBCT	The CBCT scanner provided spatial and contrast resolution suitable for the visualization of high-contrast morphology in sinus, maxillofacial, and otologic imaging applications. The scanner appeared well suited to high-contrast sinus and temporal bone imaging at doses comparable to or less than that reported for conventional diagnostic CT of the head.
Ahmad et al. 2010[19]	To discuss benefits and limitations of CBCT images compared to other imaging methods; to compare CBCT with panoramic radiography and MDCT	Comparing CBCT with panoramic radiography and MDCT	In most maxillofacial diagnostic and surgical planning or follow-up circumstances, CBCT scans can replace multidetector CT scans.
Güldner et al. 2013[20]	To evaluate the specific dosages and identify potential optimizations of the performance of CBCT of the paranasal sinuses	CBCT	A crucial reduction of the applied dosage for CBCT visualization of the paranasal sinuses in cases of chronic rhinosinusitis is possible from a clinical and radiological point of view.
Fakhran et al. 2014[21]	To determine how often clinically important findings would be missed if CBCT was used routinely for sinus imaging	MDCT and CBCT	With appropriate selection of patients, CBCT can offer a substantial reduction in radiation dose and may provide a viable alternative to standard MDCT sinus imaging protocols.
Zojaji et al. 2015[22]	To assess the agreement between CBCT and sinus endoscopy findings and attempt to determine the diagnostic accuracy of CBCT in patients with chronic rhinosinusitis	CBCT and sinus endoscopy	Considering its high accuracy and lower costs and radiation doses, CBCT may be a proper alternative method for diagnostic sinus endoscopy in the assessment of chronic rhinosinusitis in patients with a contraindication for sinus endoscopy.
De Cock et al. 2015[23]	To evaluate image quality and radiation dose of CBCT and MSCT in patients with sinonasal polyposis	CBCT and MSCT	Given the lower radiation dose, CBCT can be considered for the evaluation of the sinonasal structures in patients with sinonasal polyposis.
Dierckx et al. 2015[24]	To assess organ dose and effective dose for 2 examination tasks of the head: the sinus, and the middle ear	CBCT, MSCT and projection radiography	In terms of dose delivered to the patient, it can be justified to replace some MSCT examinations with CBCT examinations.
Demeslay et al. 2016[25]	To assess the morphological concordance between CBCT and MDCT in the context of the sinonasal anatomy	CBCT and MDCT	This preliminary study indicated that CBCT represents a valid, reproducible, and safe technique for the identification of relevant sinonasal anatomical structures.
Al Abduwani et al. 2016[26]	To compare the absorbed doses in CBCT and conventional CT and to compare the clarity and image quality with regard to important structures in the sinus anatomy	CBCT and MDCT	CBCT is a quick and efficient alternative to conventional CT with a substantial reduction in radiation dose compared to conventional and low-dose MDCT techniques.
Yamauchi et al. 2017[27]	To investigate the accuracy of CBCT in the diagnosis of non-invasive chronic fungal rhinosinusitis	CBCT and MDCT	CBCT is inferior to MDCT in the detection of intrasinus calcification in patients with non-invasive chronic fungal maxillary sinusitis.

CBCT: cone-beam computed tomography, MDCT: multidetector computed tomography, MSCT: multislice computed tomography, DSA: digital subtraction angiography, CT: computed tomography

Discussion

SSS has several unique imaging characteristics. Typical radiographic features include a marked unilateral decrease in the overall volume of the sinus due to inward retraction of the sinus walls, which in turn augments the ipsilateral orbital volume and results in the concomitant expansion of the middle meatus on the affected side.[3] The affected maxillary sinus also demonstrates opacification, while occlusion of the ostiomeatal complex and flattening of the malar eminence may be seen.[8] As demonstrated in Figures 1-3, maxillary sinus structures were measured.[9] Analysis of the axial and coronal CBCT sections revealed characteristics including unilateral volume loss in the maxillary sinus, retracted sinus walls, partial to complete soft-tissue opacification of the maxillary sinus, and inferior bowing of the orbital floor. A distinct lack of history of trauma or sinusitis was noted for these cases. The radiological differential diagnosis of SSS includes congenital maxillary sinus hypoplasia, chronic sinusitis, and mucocele, as well as changes seen after maxillary sinus and orbital trauma. Some of the distinguishing features of congenital maxillary sinus hypoplasia include the arresting of sinus development due to infection or trauma, as well as congenital first arch syndrome and developmental anomalies.[10] Congenital anomalies such as Treacher Collins syndrome are associated with unilateral maxillary sinus hypoplasia.[11] All features of maxillary hypoplasia were absent in the cases in the present study.

Based on a study of patients at the Massachusetts Eye and Ear Infirmary, chronic maxillary atelectasis has been classified into 3 stages. This classification is based on anatomical changes. Stage 1 (membranous deformity) is marked by lateralization of the maxillary fontanelle; stage 2 (bone deformity) involves the inward bowing of 1 or more walls of the maxillary sinus, and in stage 3 (clinical deformity), enophthalmos, hypoglobus, and/or deformity of the midface are noted.[12,13] Based on this classification, the cases in this study fell within stage 2.

As the use of CBCT in diagnostic maxillofacial imaging has become more frequent, its range of applications has also expanded. Paranasal sinus imaging is one such CBCT application outside of dentistry.[7] The present report also provides a summary of relevant studies that have investigated the routine use of complex imaging in medicine for sinus pathoses, in an effort to provide some perspective regarding the potential applications of computed tomography for which low-dose CBCT techniques could be used. A narrative literature search of the PubMed database was performed with the following Boolean queries: 1) paranasal sinus AND imaging, 2) paranasal sinus imaging AND MDCT OR CBCT, 3) comparison of radiation dose CBCT AND MDCT, and 4) the above searches combined as follows: 2 AND (3 OR 1).

The search results were limited to papers in English published in the past 10 years. This time frame was chosen for 2 reasons. First, even though the widespread implementation of CBCT in dentistry started in the mid-2000s, relatively few studies are available on its use in paranasal sinus imaging. Second, the technology itself has rapidly developed over time, with improved spatial resolution, better signal-to-noise ratios, and significantly lower radiation doses. Therefore, findings from older studies may not be as relevant as more recent research. The 10-year cut-off reflects our need to include as many papers as possible while attempting to stay relevant in a changing technology landscape. The titles and abstracts of the retrieved papers were manually screened. Papers meeting the following inclusion criteria were selected for the final analysis: empirical investigations (quantitative and qualitative), comparison of the radiation dose associated with CBCT and MDCT, and paranasal sinus imaging. Snowballing from the results of this literature search, retrieved citations were used as the basis for new searches using the citation's bibliography, authors' names, and "related articles" as search options, as appropriate. A total of 14 papers met the inclusion criteria and repeatedly recommended CBCT as the preferred imaging modality over MDCT for the imaging of SSS and/or paranasal sinus disease or anomalies, due to the lower radiation dose associated with CBCT (Table 1).

In several studies, a significantly lower overall radiation dose has been demonstrated to be associated with CBCT compared to MDCT/multislice computed tomography, suggesting that CBCT should likely be the imaging modality of choice when appropriate demographic and clinical features are noted with an absence of history of trauma, surgery, or developmental anomalies.[14] A study by Almashraqi et al. compared a low-dose MDCT protocol with CBCT, and CBCT was found to be associated with approximately a 40% reduction in radiation dose and comparable image quality relative to MDCT.[15] Furthermore, a technical valuation report on imaging performance and radiation dose for a commercially available CBCT scanner illustrated some merits of CBCT imaging, including significantly better spatial and contrast resolution and reduced radiation dose.[16]

The exact pathophysiology of SSS is still a subject of debate, and 3 hypotheses have been proposed.[8] According to the first hypothesis, a prolonged subatmospheric (negative)

pressure is created in the sinus due to complete obstruction of the maxillary ostium, hypoventilation, and secretion/mucus retention. The cause of obstruction of the maxillary sinus may be inspissated secretions, hypermobility of the infundibular wall, mucocele or nasal polyp, and/or narrowing of the ostium by inflamed mucosa or adjacent Haller cells.[17] The negative pressure creates a vacuum that may induce osteopenia, bone remodeling, and sinus wall retraction. As a result, the orbital floor becomes thin, and support for the orbital contents is lost, leading to enophthalmos.[8] The second hypothesis suggests that inflammatory processes lead to osteopenia with subsequent erosion of the bony orbital floor. Stashenko et al. and Amano et al. have shown that inflammatory cells can produce cytokines that have the potential to inhibit osteoblast and collagen synthesis, leading to osteopenia.[8] The third hypothesis states that SSS develops in individuals with congenitally hypoplastic maxillary sinuses that become infected. However, studies have shown that SSS occurs in normal and well-developed maxillary sinuses.[8] Functional endoscopic sinus surgery is the preferred treatment for SSS, with expected results of improved drainage of the maxillary sinus and restoration of its anatomy.[18]

In summary, this report was written to highlight the incidental finding of SSS, a relatively rarely reported and often underdiagnosed sinus pathosis, on CBCT images. Furthermore, it underlines features that were observed using a relatively high spatial resolution modality with a significant reduction in radiation dose. Dental professionals should recognize the existence of SSS in otherwise asymptomatic patients and institute management where needed.

Conflicts of interest: None

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