



Paranasal Sinus Cysts Encountered Over Twenty Years: Varieties, Management, Recommendations, and Outcome

Ramadan Hashem Sayed, Khaled Gamal Dahy* and Al Hussein Awad Ali

Department of Otolaryngology-Head and Neck Surgery, Faculty of Medicine Sohag University, Egypt

*Corresponding author: Khaled Gamal Dahy, Sohag Faculty of Medicine, Department of ENT, Sohag, Egypt, Tel: +20-93-4606594/ +20-1000796380; Email: khgamal272@gmail.com

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Abstract

Background: We recorded all cases presenting with paranasal sinus cysts to draw attention to those with unusual presentations and their management outcome in a retrospective original study.

Methods: All patients were subjected to history-taking, ENT examination, endoscopic examination, CT scanning, and managed with endoscopic sinus surgery/or a combined approach.

Results: We encountered 54 patients with paranasal sinus cysts. Mucocele/pyocele was the commonest (79.6%), followed by radicular cyst (9.3%), dentigerous cyst (5.55%), and fungocele (5.55%). The frontal sinus was the commonest sinus of cyst origin (55.6%). Maxillary sinus cysts occurred in 16.6% of cases. The sphenoid sinus was the "least" sinus of cyst origin in 7.4% of cases.

Conclusion: Ethmoid mucocele occurred mostly by extension from other sinuses, predominantly frontal. With exclusive frontal sinus cysts, it is essential to look for possible underlying pathology or associated complication. Fungocele, a new nomenclature, could complicate those with allergic fungal sinusitis.

Keywords: Paranasal Sinus Cysts; Mucocele; Pyocele; Odontogenic

Abbreviations: PNS: Paranasal Sinus, DC: Dentigerous Cyst, RCC: Rathke's Cleft Cysts, CT: Computed Tomography, AFS: Allergic Fungal Sinusitis.

Background

A wide variety of paranasal sinus (PNS) cysts had been reported in the literature including extrinsic, which originate in nearby structures like the dental tissues (radicular cyst, dentigerous cyst, and keratocystic odontogenic tumors), or intrinsic, which originate in the mucosa of the paranasal sinuses especially mucocles and pyocele. Radicular cysts (apical periodontal cysts, dental root end cysts) are the most common inflammatory odontogenic cysts in tooth-bearing areas of the jaws they originate from an epithelial

rest of Malassez in periodontal ligaments secondary to inflammation. They are most frequently found at the apices of the involved teeth with infected or necrotic pulps; however, they may also be found on the lateral aspects of the roots in relation to accessory root canals [1,2]. Most commonly, radicular cysts occur between the third and the sixth decades of life with male predominance and could occur in relation to the maxillary sinus. Dentigerous cyst (DC) or follicular cyst is the second most common type of odontogenic cyst [3]. It forms during tooth development and is associated with the pressure exerted by the crown of an unerupted (or partially erupted) tooth on the fluid within the follicular space. They almost exclusively occur in permanent dentition [4]. The most common location of DC is the mandibular third molar, more rarely occurring in the maxillary sinus. Enucleation is

the standard treatment [4]. Keratocystic odontogenic tumors are rare and benign lesions of odontogenic origin with a potential for aggressive and infiltrative behavior [5]. They are more common in the third to sixth decades with a male to female ratio is approximately 2:1 and the majority are found in the mandible and could occur in the maxillary sinus [6]. Mucocoeles of the PNSs are benign, chronic expanding lesions that develop because of obstruction of the sinus ostium. It is the commonest cyst affecting the PNSs, and if infected, it forms pyoceles. The frontal sinus is the most common sinus to be affected by a mucocoele, which usually results from trauma or inflammatory processes [7], followed by ethmoid, maxillary, and to a lesser extent the sphenoid sinus. Onodi cell mucocoele is extremely rare and might cause visual loss owing to its close relation to the optic nerve [8] and a higher rate of optic nerve protrusion and dehiscence [9]. Posttraumatic mucocoele was the least frequent form of PNS mucocoeles and was located predominantly in the frontal sinus, causing symptoms long after the forehead injury [10]. Although mucocoeles are benign; yet, their expansion may lead to bony erosion and extension into the orbit and cranium, leading to ocular and visual complications, and rarely frontal lobe syndrome i.e. mass effect on the frontal lobe [11], meningitis, meningoencephalitis, intracranial abscess, seizures, or cerebrospinal fluid fistula [12]. Very rarely, they will present as a subcutaneous forehead mass or swelling [7], and more rarely they can cause frontocutaneous fistula and a forehead skin ulcer [12]. The endoscopic marsupialization technique could easily be applied even to a very large mucocoele that has extended to the intracranial region, without any serious complications [13]. In-office drainage of sinus mucocoeles under topical/local anesthesia is well tolerated by patients, especially for mucocoeles that were accessible endoscopically in the office setting, in non-anxious patients, aiming to widely open and drain the mucocoeles with high success, and low complication rates, even in large mucocoeles with bone erosion. Orbital and skull base erosion are not contraindications [14]. External and radical procedures for the treatment of frontal sinus mucocoeles have major complications including high surgical morbidity, esthetic concerns of postoperative scar formation, and difficulty in radiologic diagnosis of recurrence after obliteration [13]. Terranova PA, et al. [15] found a complete self-reconstruction of bone that had previously been eroded by the mucocoele, in the postoperative CT scan in 66.6% of their patients [15]. There is no need to reconstruct the eroded bone, as sparing the mucoperiosteum is enough to enable the bone to regenerate. No enophthalmos, meningocele, or other facial deformities were noted, despite not having undergone surgical reconstruction of the bone [15]. Other types of cysts related to PNS include aneurysmal bone cysts, Rathke's cleft cysts (RCC), and epidermoid cysts. An aneurysmal bone cyst is a benign multicystic mass that is locally destructive and rapidly expandable and rare in the sinuses [16]. Rathke's

cleft cyst is a benign epithelium-lined cyst containing mucoid material, typically occurring entirely intrasellar, or intrasellar with suprasellar extension (intra-suprasellar). Extrasellar location is uncommon [17]. Kasliwal MK, et al. [17] reported a rare patient with an intrasphenoidal RCC [17]. Epidermoid cysts are the commonest of head and neck cutaneous cysts Golden BA, et al. [18] found in various regions [19]. In contrast to dermoid, epidermoid cysts do not include skin appendages within their wall [20]. Ren D, et al. [21] reported a huge epidermoid cyst in the maxillary sinus with sinus cavity expansion and sinus wall thinning [21].

Over twenty years we recorded all cases presenting with PNS cysts, their clinical presentations, radiological findings, a management protocol, and surgical outcome to draw attention to those patients with unusual and odd presentations and their management outcomes.

Methods

A retrospective study was done with all patients presenting with PNS cysts to the outpatient clinic were included in the study. Ethical clearance was obtained from the institutional ethical board with ethics committee No. Soh-Med-22-03-30. Informed consent was obtained from all patients who agreed to participate in this study. These patients underwent history-taking, ENT examination, endoscopic examination, computed tomography (CT) scanning of the nose and PNSs, and routine laboratory investigations. They were managed with endoscopic sinus surgery/or a combined approach under general anesthesia, and the postoperative outcome was recorded. Those with serious inflammatory complications were followed up for one year.

Results

Over twenty years, between January 2002 and December 2021, we encountered 54 patients with various PNS cysts. The age range was 7-70 years, with a mean age of 28.6 years, and the male/female ratio was 3:2. Table 1 demonstrated the distribution of these cases. Mucocoele/pyocoele was the commonest cyst encountered in the PNSs (43/54=79.6%), followed by radicular cyst (5/54=9.3%), dentigerous cyst (3/54=5.55%), and finally, what we termed fungocoele (3/54=5.55%). Most patients presented with an insidious, painless, slowly progressive, disfiguring swelling, and diplopia. Painful swelling was the presenting symptom in 4 patients with pyocoele; complicated in one of them with a left orbital pain and orbital cellulitis, and another with a left-sided orbital pain, orbital abscess, and cavernous sinus thrombophlebitis. Two patients with sphenoid fungocoele presented with visual deterioration (Figures 1-11).

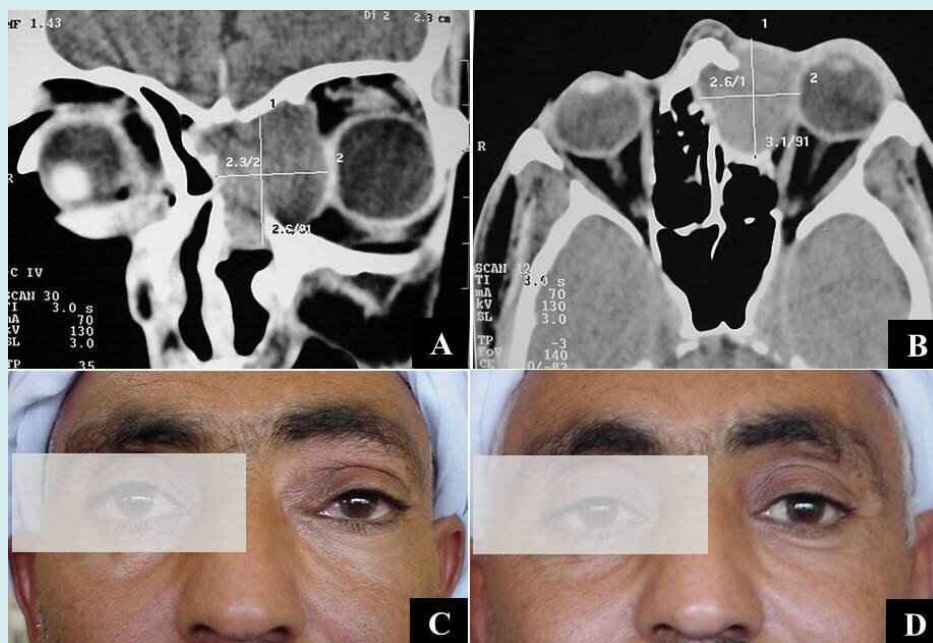


Figure 1: (A) CT scan with a coronal soft tissue cut showing a left fronto-ethmoid mucocoele displacing the inter-frontal septum to the right, eroding the sinus floor, and displacing the globe; (B) CT scan with axial soft tissue cut showing the mucocoele involving the anterior ethmoid air cells with eroded lamina papyracea and displaced globe; (C) Pre-operative photo showing swelling on the inner aspect of the left globe with down and out proptosis; (D) Postoperative photo 4 days following ESS.

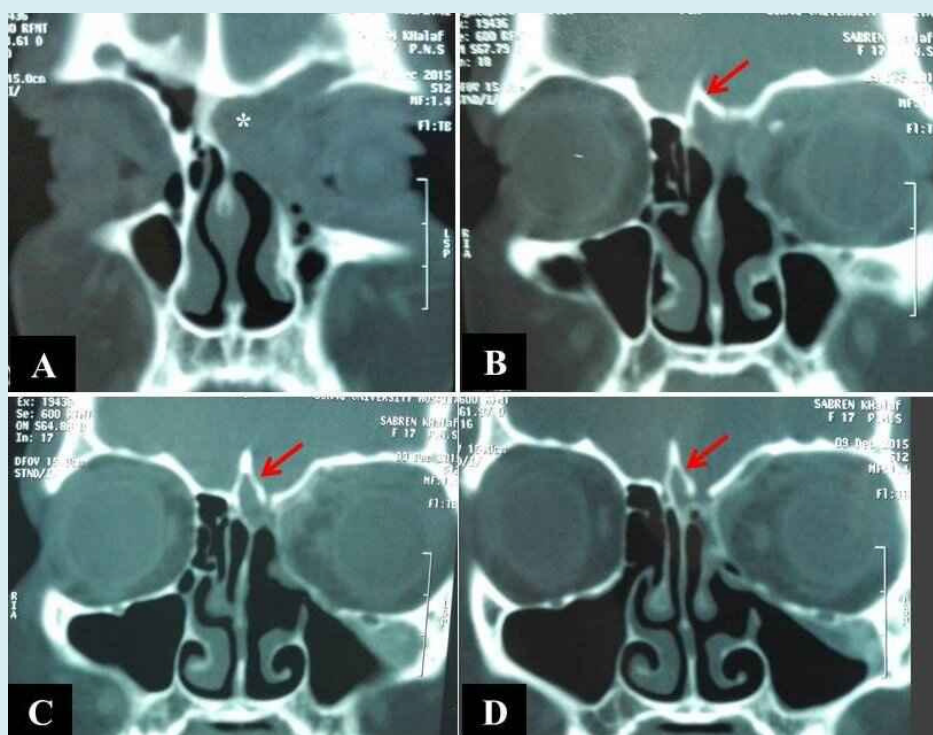


Figure 2: CT scan with coronal bone window cuts showing a left-sided FEM (white asterisk) eroding the floor of frontal sinus and lamina papyracea and displacing the globe (A), with distension of a pneumatized crista Galli by the mucocoele (red arrows in B, C, D).

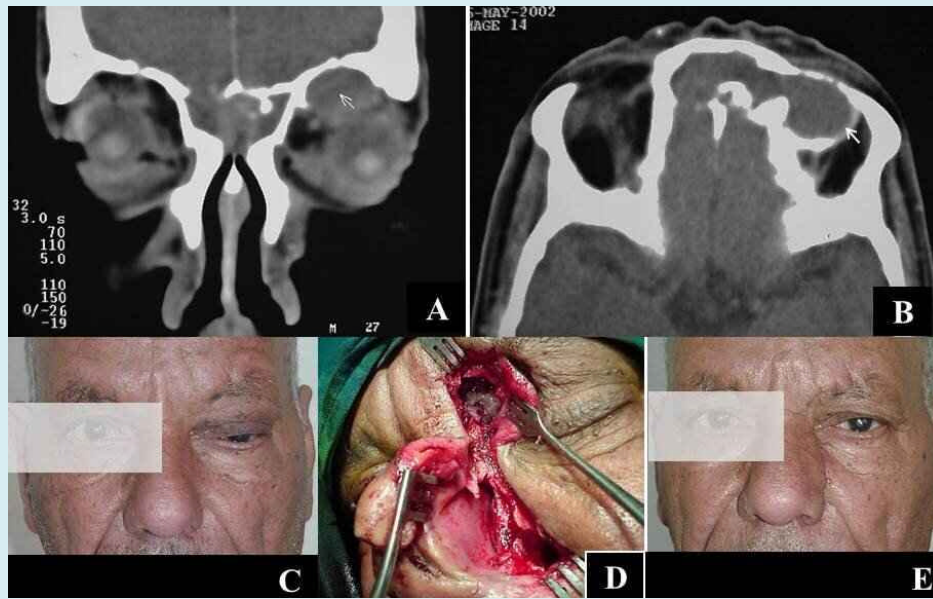


Figure 3: CT scan with coronal (A) and axial (B) soft tissue cuts showing a soft tissue shadow involving both frontoethmoid complexes with an absent inter-frontal septum and eroded posterior wall of right frontal sinus due to inverted papilloma, and a well-defined localized swelling in the medial orbital roof (white arrow) that is in continuity with the soft tissue shadow in the frontal complex (B) that proved to be pyocoele; (C) A preoperative photo showing a left-sided upper lid dusky swelling with a down displacement of the globe and orbital cellulitis; (D) Extended Moure's lateral rhinotomy with localized frontal osteotomy to facilitate removal of the inverted papilloma that could be seen through the osteotomy; (E) Postoperative photo of the patient two weeks after surgery.



Figure 4: CT scan with sagittal (A, B, C) and axial (D) soft tissue cuts showing a well-defined frontal subcutaneous isodense soft tissue shadow (white arrows), with scattered high-density shadows of AM, with absent anterior walls of frontal sinuses, involving both frontal sinuses (D) with a localized erosion of the posterior wall of the right frontal sinus (red arrows in B, D), that proved to be a fungocele. Note the pan-sinus opacity with scattered high-density shadows of AM (red asterisks) in the maxillary sinus (A), sphenoid sinus (B), and ethmoid air cells (C)

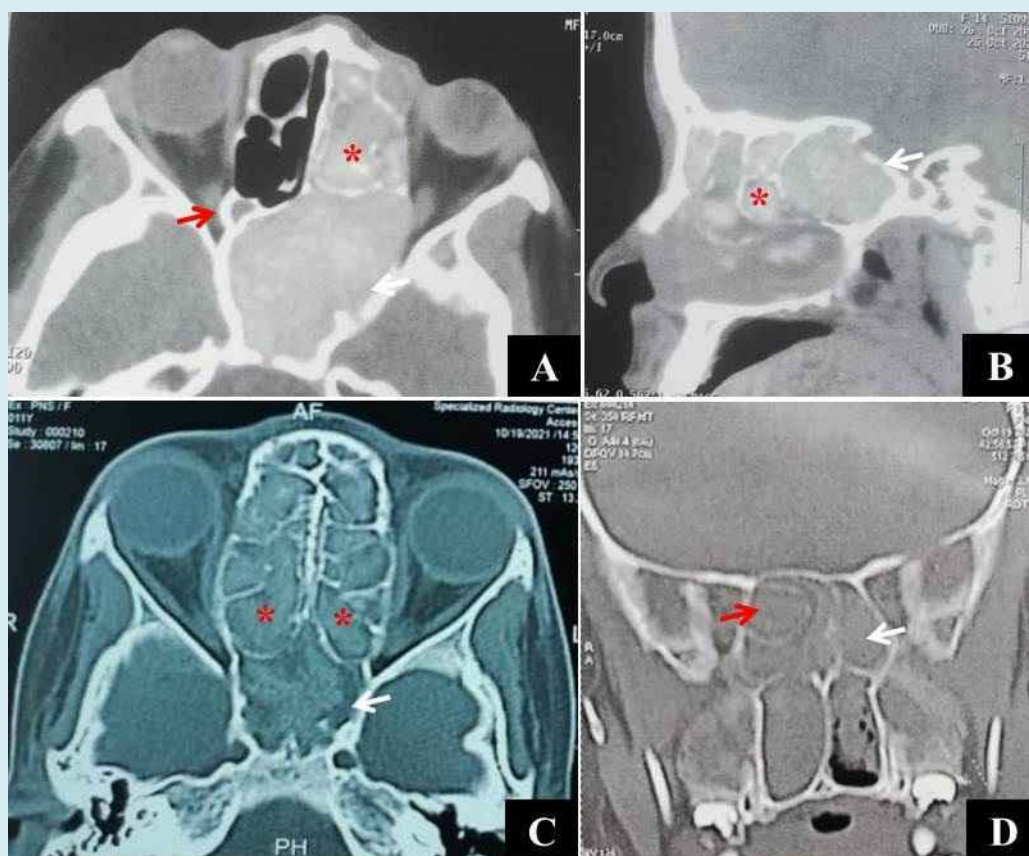


Figure 5: CT scan with axial (A) and sagittal (B) soft tissue cuts showing a left-sided sphenoid fungocele (white arrows), very small right sphenoid sinus (red arrow), and scattered high-density shadows of AM in ethmoid air cells (red asterisks); CT scan with axial (C) and coronal (D) bone window cuts showing a bilateral sphenoid fungocele with an absent inter-sphenoid septum and merged sinuses (white arrows), bilateral AFS involving ethmoid air cells (red asterisks) and left Onodi cell invading the right sphenoid sinus (red arrow in D)

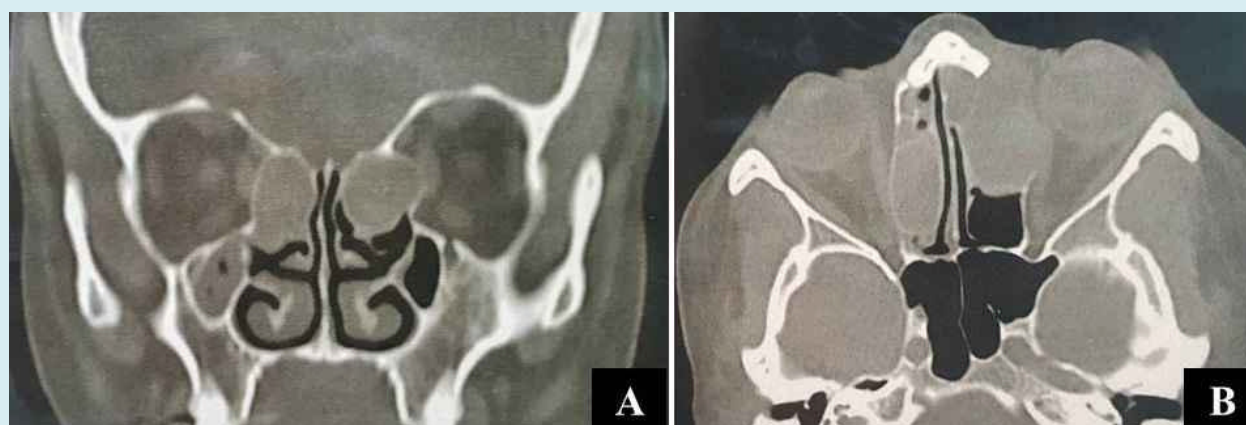


Figure 6: CT scan with coronal (A) and axial (B) bone window cuts showing soft tissue shadows involving anterior ethmoid air cells on both sides, proved to be pyoceles, with an erosion of the left lamina papyracea, orbital extension, and displacement of the left globe.

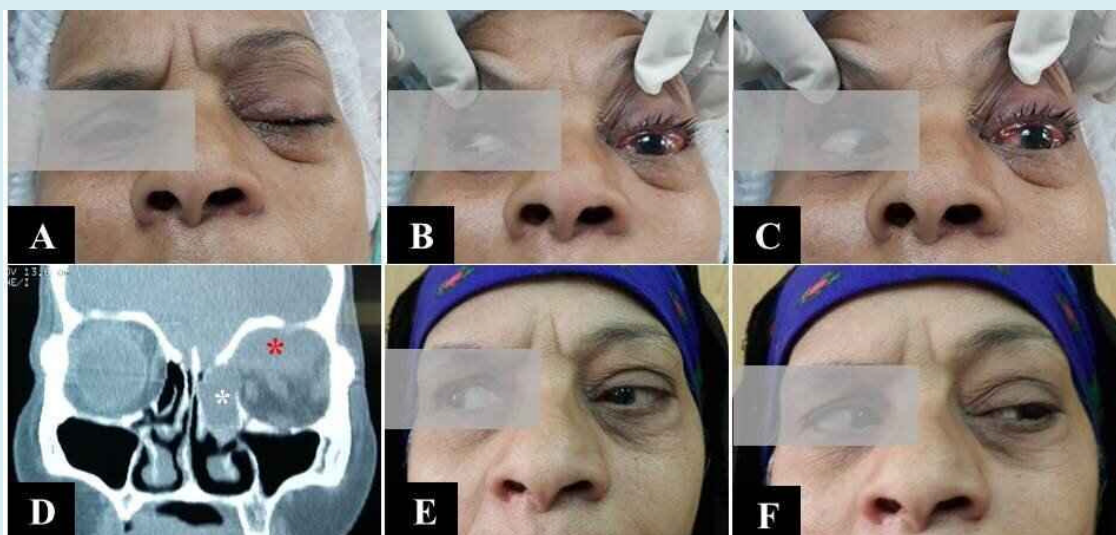


Figure 7: A 50-year-old female having left-sided total ophthalmoplegia due to cavernous sinus thrombophlebitis presented with ptosis and lid edema (A); a left-sided proptosis, orbital cellulitis, and ophthalmoplegia on looking to the right (B), and on looking to the left (C); (D) CT scan with coronal soft tissue cut showing a left-sided ethmoid pyocoele involving a concha bullosa and anterior ethmoid cells (white asterisk) with an erosion of the lamina papyracea, and a well-defined soft tissue shadow on the upper medial wall and roof of the orbit (red asterisk) due to sub-periosteal abscess; (E, F) Postoperative photos of the patient showing resolution of orbital cellulitis and regain of ocular motility on looking to the right and left.

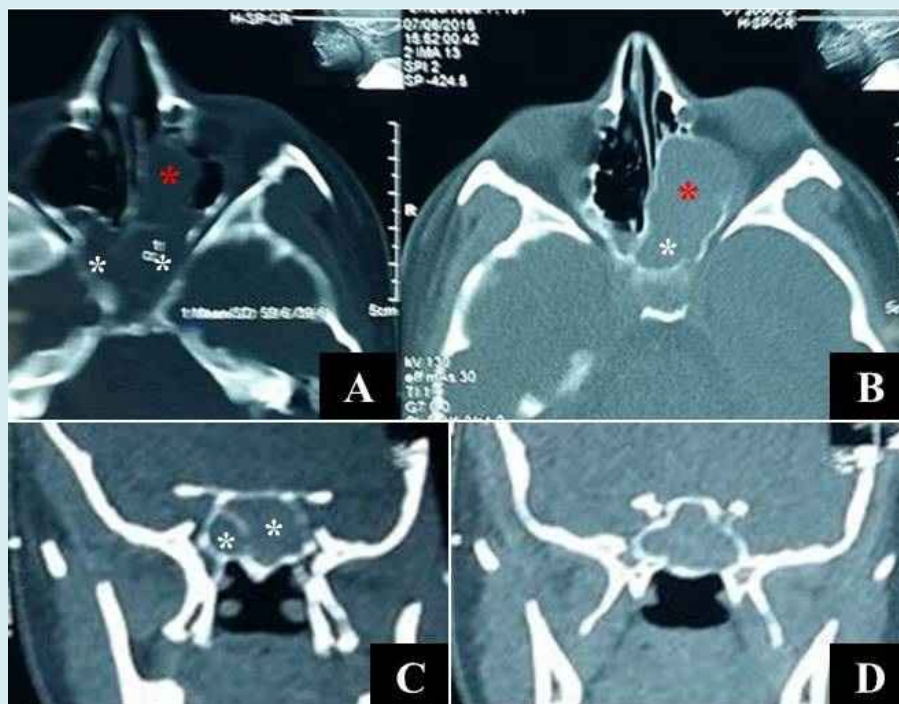


Figure 8: CT scan with axial bone window cut (A) and axial soft tissue cut (B) showing a mucocoele involving both sphenoid sinuses with a smaller right and a larger left sphenoid sinus (white asterisks), and involving the left posterior ethmoid air cells (red asterisks). Note the partially eroded inter-sphenoid septum and anterior face of the left sphenoid sinus; CT scan with coronal soft tissue cuts showing mucocoele involving both sphenoid sinuses (white asterisks) with a partially eroded inter-sphenoid septum and a smaller right sphenoid (C), and merged both sphenoid sinuses with an absent inter-sinus septum and distended both lateral walls (D).

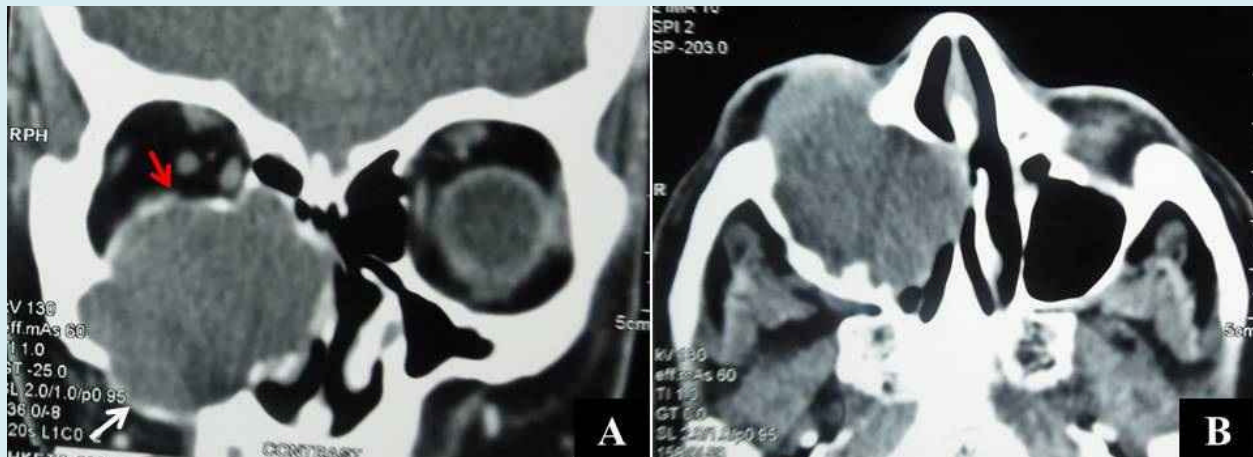


Figure 9: CT scan with coronal (A) and axial (B) soft tissue cuts showing a right-sided maxillary sinus mucocoele distending the sinus with a displaced and partially eroded roof (red arrow), anterolateral wall (white arrow), and markedly displaced and eroded medial maxillary wall, hugging the nasal septum.

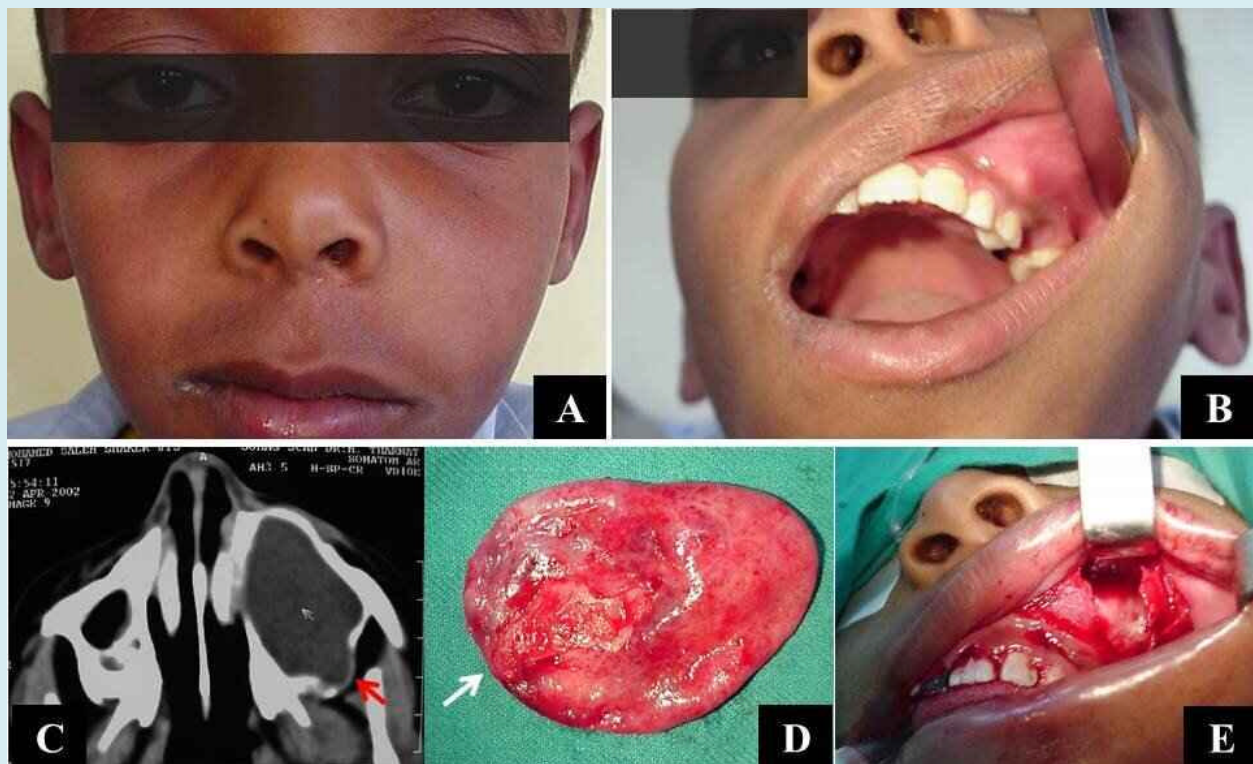


Figure 10: A nine-year-old boy with a left-sided cheek swelling caused by a radicular cyst (A), with obliteration of the left buccoalveolar sulcus, expansion of the anterior maxillary wall, and a carious upper left 2nd premolar tooth seen through the open mouth (B); (C) CT scan with axial soft tissue cut showing distension of the left maxillary sinus by the cyst with thinning out and erosion of the posterolateral maxillary wall (red arrow); (D) A photo of the removed cyst by enucleation; note the site of tooth reaction in the cyst wall (white arrow); (E) Operative photo showing a distended anterolateral maxillary wall with a window created in it through a sub-labial approach, with the cyst removed, and the sinus cavity seen.

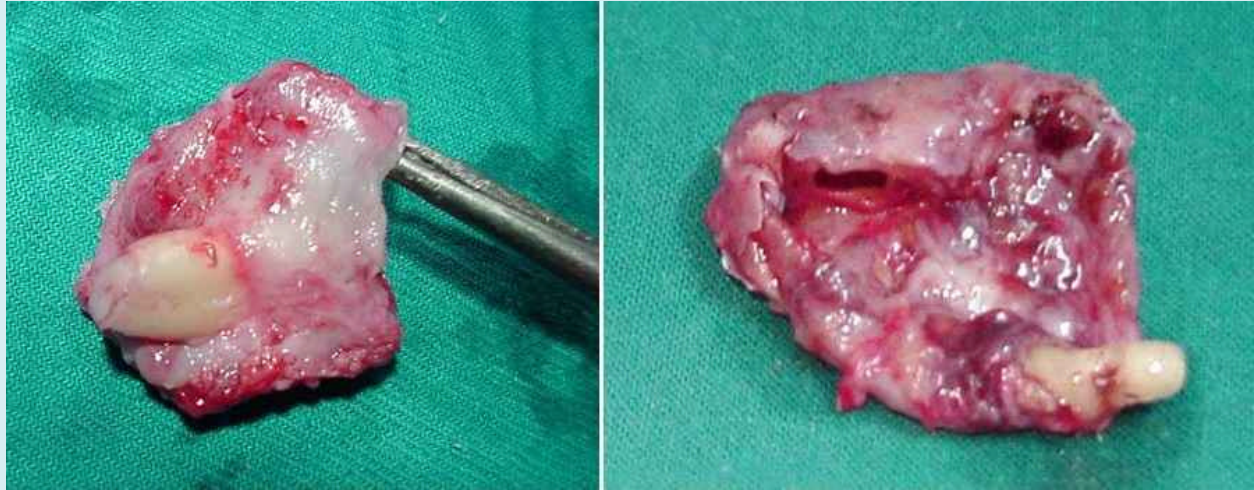


Figure 11: A photograph showing 2 freshly removed dentigerous cysts from 2 different patients with the offending missed tooth embedded in the cyst walls.

Cyst type	Site of origin	Number	Males	Females	Approach
Mucocele	Frontal	3	2	1	ESS
	Frontoethmoid	24	16	8	
	Ethmoid	9	5	4	
	Maxillary	1	0	1	
	Sphenoid (bilateral in one) and left posterior ethmoid	2	0	2	
Pyocoele	Frontal	2	2	0	1 ESS
	Ethmoid (bilateral in one)	2	0	2	1 CA
Fungocoele	Bilateral frontal sinuses	1	1	0	ESS
	Bilateral sphenoid sinuses	1	0	1	
	Left sphenoid sinus	1	0	1	
Radicular cyst	Maxillary	5	4	1	CA
Dentigerous cyst	Maxillary	3	2	1	
Total		54	32	22	

ESS: endoscopic sinus surgery; CA: combined approach.

Table 1: Distribution of patients with PNS cysts over twenty years.

The frontal sinus was the commonest sinus of PNS cyst origin in 30/54 cases (55.6%), being exclusively frontal in 6/30 cases (20%), and extending to involve the anterior ethmoid air cells forming frontoethmoid mucocele (FEM) in 24/30 (80%) cases (Figure 1). One of our patients with FEMs presented with a right-sided FEM at the age of 7 years that was managed with ESS, and eleven years later developed a left-sided FEM. A pneumatized crista Galli was involved as a part of FEM in one patient (Figure 2). Among the six patients with exclusive frontal sinus cysts, three had mucocele;

two had pyocoele, and one of them developed secondary to inverted papilloma involving both frontoethmoid complexes with a left frontal pyocoele and orbital cellulitis (Figure 3). This patient was managed with a combined extended Moure's lateral rhinotomy and endoscopic sinus surgery (ESS) to facilitate the removal of the neoplasm and drainage of the pyocoele. The patient achieved an excellent postoperative outcome with the resolution of the orbital cellulitis. The last case with exclusive frontal cyst presented by January 2014 with bilateral extensive allergic fungal sinusitis (AFS)

with pan-sinus involvement, and a large, non-tender, frontal subcutaneous doughy cystic swelling originating from both frontal sinuses, with absent anterior frontal walls, and an erosion in the right posterior frontal wall (Figure 4). He was managed with ESS, and the swelling was found to contain a viscid gelled mucoid discharge admixed with allergic mucin (AM); hence we termed it a fungocele, a nomenclature that was recorded for the first time in the literature. By 2001, we faced another 2 cases with sphenoid fungocele. The first was an 11-year female child who presented with bilateral deterioration of vision down to 6/24, bilateral AFS involving all PNSs, and a fungocele involving both sphenoid sinuses that merged. The second was a 14-year female child who presented with a left-sided AFS involving all the PNSs, a left sphenoid fungocele, a left proptosis, and left deterioration of vision down to 1/60 (Figure 5).

The ethmoid air cells were the sinus of cyst origin in 11/54 cases (20.4%). In all of them, the anterior ethmoid cells were involved, with 9 mucocèles and two pyocèles. One of the latter cases presented with bilateral ethmoid pyocèles and left-sided proptosis (Figure 6). The second presented with a left-sided pyocele complicated with orbital abscess and cavernous sinus thrombophlebitis (Figure 7). This patient was managed with ESS with a dramatic postoperative improvement, resolution of orbital abscess, and regain of ocular motility. We observed that 26/37 (70.3%) of ethmoid involvement occurred by extension from other PNSs, mostly the frontal sinus (24/26=92.3%), and only two from the sphenoid sinus. Among the ethmoid air cells, the anterior ethmoid cells were involved in 35/37 cases (94.6%), while the posterior ethmoid cells were involved in only 2/37 cases (5.4%) by extension from the sphenoid sinuses (Figure 8).

The maxillary sinus was involved with cystic swelling in 9/54 (16.6%) cases, one of them was a mucocèle expanding the sinus, and presenting with a right cheek swelling and proptosis (Figure 9). This patient was managed with ESS. The remaining 8 cases were cysts of dental origin; 5 were radicular cysts related to a carious upper tooth and expanding the maxillary sinus with a disfiguring cheek swelling (Figure 10) and 3 were dentigerous cysts expanding the sinus with a missed tooth included in the cyst wall (Figure 11). All odontogenic cysts were managed with a Caldwell-luc's approach combined with ESS to facilitate cyst enucleation, extraction of the offending carious tooth in case of radicular cysts, and performing a middle meatal antrostomy.

The sphenoid sinus was the 'least' sinus of cyst origin in 4/54 (7.4%) of our cases; 2 were mucocèles, and in one of them, the cyst involved both sphenoid sinuses mainly the left (Figure 8). In both cases, the sphenoid mucocèle extended to involve the left posterior ethmoid cells forming sphe-no-ethmoid mucocèle. The other 2 sphenoid cysts were

fungocèles, in one of them involved both sphenoid sinuses that merged (Figure 5).

Discussion

There are several types of PNS cysts that have been recorded in the literature; mucocèle/pyocèle is by far the most prevalent, followed by cysts with a dental origin [22-24]. In our series, mucocèle/pyocèle formed 79.6% of PNS cysts, followed by odontogenic cysts in 14.8% of cases, with the radicular cysts being commoner than PNS dentigerous cysts (5:3) this coincides with previous studies about the prevalence of the cysts of the jaw [25-27]; lastly, fungocele in only 5.6%. The frontal sinus was the commonest sinus of cyst origin (30/54=55.6%), owing to its distinct ostium anatomy that is vulnerable to obstruction by various pathologies including inflammatory, traumatic, and neoplastic lesions, with the inflammatory one being the commonest cause of ostium obstruction and cyst formation as stated previously by Wilkerson [28]. In 88.9% of our cases with frontal sinus mucocèle (24/27 cases), it extended to involve the anterior ethmoid air cells forming FEM. This involvement could be explained by the close vicinity of the anterior ethmoid cells to the frontal sinus ostium this is in accordance with other published articles reporting the anatomic distribution of mucocèles [29-31]. According to several reports, isolated sphenoid mucocèles may occur in 1% - 3% of cases, this resembles our series in which, there were 2 sphenoid mucocèles (3.7%) [32,33]. Fungocele is a nomenclature that is recorded for the first time in the literature. It also involved the sphenoid sinus in 2 of our cases. Sphenoid fungocele was more serious and presented in a young age group with visual deterioration.

Paranasal sinus cysts of unusual sites of origin were observed in 6 of our cases. Interestingly enough, all were females. Four of these were mucocèles, one involving a pneumatized crista Galli as a part of FEM, one involving the maxillary sinus, and two involving the sphenoid sinuses and left posterior ethmoid. Few previous reports of mucocèle involving a pneumatized crista Galli were found in the literature [34,35]. The remaining two were fungocele involving sphenoid sinus. We could find no sound explanation for this gender predilection. Despite the rarity of bilateral involvement, Only a few reports have been recorded in the literature [36-39], in the present study bilateral PNS cysts were encountered in five patients, one had bilateral frontal fungocele, the second had bilateral ethmoid pyocèles, the third had bilateral sphenoid mucocèle, the fourth had bilateral sphenoid fungocele and the fifth had a right-sided FEM, followed eleven years later by a left-sided one.

Among our series, we met no case with post-traumatic mucocèle contrary to Kojima, et al. [10] who reported

posttraumatic mucocele in 5/68 (7%) of their patients [40].

Paranasal sinus cysts have a benign course, presenting with painless disfiguring swelling in most cases, frequently with diplopia; yet, serious complications could occur if get infected and with those with sphenoid fungocele. We faced four serious cases, one with an ethmoid pyocele complicated with orbital abscess, cavernous sinus thrombophlebitis, and total ophthalmoplegia. The second patient had an underlying unusual, high-up, inverted papilloma involving both fronto-ethmoid complexes with a left-sided frontal pyocele that extended into the orbit with orbital cellulitis. Both cases could be controlled, with rapid postoperative recovery and resolution of all deficits. To our knowledge, this is the first reported case in the literature of inverted papilloma involving bilateral frontoethmoid complex, and complicated with frontal pyocele and orbital cellulitis. The remaining two serious cases had sphenoid fungocele with visual deterioration that reached 1/60 in one of them.

Odontogenic cysts presented with painless slowly progressive maxillary sinus expansion with a disfiguring cheek swelling, related to a carious tooth in those with radicular cysts (5 cases) or a missed tooth in those with dentigerous cysts (3 cases). These odontogenic cysts could easily be enucleated through Caldwell-Luc's approach combined with ESS with a good outcome.

In contrast to obliterative techniques, endoscopic marsupialization is now the surgical method of choice for treating most paranasal sinus cysts [40-42]. This study, which shows no complication recorded and low recurrence rates (1/54=1.9%, with FEM that could be controlled with revision marsupialization with ESS), are similar to those of the previously published series, supporting the idea that the endoscopic approach is still a successful and safe mode of treatment and ought to be used for the majority of these patients. The endoscopic method reduces morbidity while also restoring mucociliary clearance, which permits ongoing endoscopic and radiographic evaluation of the operated sinus until the sinus returns to normal function.

Conclusion

The frontal sinus was the commonest sinus of PNS cyst origin. With exclusive frontal sinus involvement, it is essential to look for possible underlying pathology or associated complication. Fungocele, a new nomenclature, could complicate those with AFS. Sphenoid fungocele could lead to serious deterioration of vision. Ethmoid air cells are involved with mucocele mostly by extension from other sinuses, predominantly the frontal sinus. The anterior ethmoid cells were the commonest to be involved with mucocele/pyocele in 94.6% of cases, while the posterior ethmoid cells were

involved in 5.6% of cases by extension from the sphenoid sinus. Patients with mucocele should be followed up for possible development of another cyst on the contralateral side. All cases of PNS cysts could be controlled successfully with ESS except those with an underlying neoplastic lesion (one case) or odontogenic cysts; both required a combination of the external approach with ESS.

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Declarations

Ethics Approval and Consent to Participate: Study design and methodology were reviewed and affirmed by the local Institutional Review Boards No. Soh-Med-22-03-30, Faculty of Medicine.

Consent for Publication: All presentations of case reports must have consent for publication.

Competing Interests: The authors declare no competing financial or non-financial interests

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