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Endoscopic-assisted approaches for enucleation of invaded sinonasal region by benign odontogenic cysts: a case series study

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Abstract

Background: During surgical planning for the excision of odontogenic cysts and tumors, surgeons should consider choosing the appropriate endoscopic approach to reach all parts of the maxillary sinus and cyst walls during its enucleation. Using the endoscopic trans-nasal approaches results in less than one-third of the maxillary sinus being reached regardless of antrostomy or angled instruments used. Also, the expansion process and involvement of maxillary sinuses in odontogenic cysts and tumors are variable, adding more difficulties during excision. This study aimed to assess the expansion process of large benign odontogenic cysts and tumors involving nasoantral regions and adaptability to different endoscopic approaches for enucleation. Six patients were included in this case series study. Preoperative panoramic views plain film radiography, and computed tomography scans were obtained. Lesion characteristics, relation to teeth origin, the nose, and maxillary sinus wall displacement, location of lesions about typical vertical and horizontal nasomaxillary constructions, and associated inflammation of the paranasal sinuses were evaluated. Three surgical approaches were tailored, allowing viewing and endoscopic enucleation.

Results: Four periapical (radicular) cysts, one ameloblastoma, and one calcifying epithelial odontogenic tumor, ranging in size from 3 to 5 cm, were enucleated endoscopically in patients aged 15–40 years. Intra-sinus expansion of the cyst and lesion level above the nasal floor allowed endoscopic enucleation using wide middle meatal antrostomy in 1 (17%). Lesions localized to the anterior wall and the nasal process of the maxilla permitted endonasal minimal rhinotomy approach in 1 (17%). Central lesions, cyst extension below the level of the nasal floor, and lateral wall expansion with or without anterior maxillary wall scalloping had adequate exposure using a minimal sublabial approach in four patients (67%). Together with its role in complete enucleation, endoscopy allowed associated rhinosinusitis surgical treatment in five patients (83%).

Conclusion: Tailoring surgical approaches to the expansion process allowed endoscopic enucleation of large odontogenic cysts/tumors without wall remnants. The integrity of the Schneiderian membrane was preserved, protecting the critical surrounding structures.

Keywords: Odontogenic cysts, Odontogenic tumors, Nasal endoscopy, Cyst enucleation, Endoscopic sinus surgery, Rhinosinusitis, Sinusitis, Periapical cyst, Ameloblastoma, Calcifying epithelial odontogenic tumor

Background

An extraordinarily diverse collection of lesions are cysts and tumors produced from odontogenic tissues. Depending on their histogenesis and etiology, jaw cysts can be

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divided into several groups [1]. Dental lamina, reduced enamel epithelium, and epithelial remains of tooth-forming tissues (Rests of Malessez) are predecessors to the teeth from which odontogenic cysts are generated. Non-odontogenic cysts originate in different epithelial structures [1]. Developmental or inflammatory odontogenic cysts are classified as periapical cysts, dentigerous cysts (16.6–21.3%), and odontogenic keratocytes (OKC) (5.4–17.4 %), with periapical cysts being the most prevalent (52.3–70.7%) [2–4]. Contrary to cysts, benign odontogenic tumors are often categorized based on histogenesis. Ameloblastoma, keratocyte odontogenic tumors, and calcifying epithelial odontogenic tumors are examples of odontogenic epithelium tumors without odontogenic ectomesenchyme. Odontoma is an example of an odontogenic epithelium tumor with odontogenic ectomesenchyme [5]. Such tumors seem to strike differently in various parts of the world [3, 4].

The maxillary sinus and nasal cavities are the paths of least resistance for establishing odontogenic cysts and benign tumors affecting the upper jaw. People may still have massive benign tumors or cysts without clinically evident jaw enlargement [6]. For the growth of odontogenic cysts and benign tumors affecting the upper jaw, the maxillary sinus and nasal passages represent the path of least resistance.

Cystic disorders of the maxilla can be treated surgically using either marsupialization or enucleation, with or without bone grafting. The plan of care depends on lesion size, jaw bone integrity next to the lesion, and closeness of the cyst to critical anatomical structures, e.g., the dental and maxillary antrum [7].

Large cysts usually require enucleation to remove the cyst's epithelium lining and the cholesterol crystals that prevent or delay periapical wound healing [8]. The surgical management of large odontogenic cysts and tumors depends on several factors. The surgical technique chosen for each patient should have the least likelihood of iatrogenic damage. Failure to see around corners and attachment sites in traditional surgical techniques can result in cyst marsupialization instead of enucleation, with an increased incidence of recurrence [6]. Interventions for cysts expanding into the maxillary sinus may lead to oroantral fistula formation and chronic rhinosinusitis (CRS). Thus, it is necessary to maintain intact boundaries between the Schneiderian membrane and the oral cavity. It is also necessary to treat any inflammation of the nasal and paranasal sinuses associated with these cysts and tumors. So, endoscopy can be the key to successful surgical performance. There have been few reports of endoscopically assisted surgery for the enucleation of such large odontogenic cysts involving a small proportion of patients [9–12]. Removal of intra-sinus dentigerous cysts with intra-sinus expansion via trans-nasal endoscopic approach has been reported in individual patients [10, 11]. Another series reported patients with cysts removed by a trans-nasal endoscopic approach via the inferior meatus lateral wall; however, 5 of the 13 (38%) cyst walls were not wholly excised [12].

Access to lesions at the anterior wall and alveolar recess of the maxillary sinus is challenging. Possible endoscopic options are the application of a 70° endoscope following a type III sinusotomy, endoscopic standard medial maxillectomy, endoscopic pre-lacrimal window approach (PLWA), Endoscopic endonasal minimal rhinotomy approach, modified Denker's and by way of a canine fossa trepanation [13]. During surgical planning for cyst enucleation, surgeons should consider choosing the appropriate endoscopic approach. The desirable approach is the one that allows reaching all parts of the maxillary sinus and cyst walls during its enucleation with intact paranasal sinus membrane and barriers from teeth and oral cavity. Also, the expansion process and involvement of maxillary sinuses in odontogenic cysts and tumors are variable, which adds more difficulties during excision.

We, therefore, assessed the expansion and involvement of larger odontogenic cysts and tumors in the Sino-nasal passages and, subsequently, their adaptability for different endoscopically assisted surgical approaches in individual patients.

Methods

Patient demographic and clinical characteristics were evaluated. We prospectively enrolled a case series of patients who presented with large (> 2 cm diameter) benign odontogenic cysts and tumors extending into the nose and paranasal sinuses. Plain film radiography panoramic views, and computed tomography (CT) were obtained for all patients.

Lesion characteristics, identification of the displaced walls of the nose and maxillary sinuses, relation to vertical (nasal septum, medial, and lateral maxillary sinus walls), and horizontal constructions (nasal and orbital floors) associated inflammation of the paranasal sinuses were evaluated. Also, associated rhinosinusitis was scored according to Lund/Kennedy scoring system. Teamwork management, including dentists with maxillofacial experiences, evaluated all patients preoperatively and postoperatively and utilized nonsurgical dental root canal therapy (NDRCT) for necrotic teeth or intraoperative assistance. The ethics committee of our hospital approved the study, and each patient provided written informed consent.

Surgery was tailored according to the origin of each cyst, its direction of growth, and the wall affected in the scalloping process. All patients underwent cystic

enucleation through different corridors according to the manner of expansion of the lesion and were assisted with 0 and 30°, lengths (6, 14, 18 cm) and diameters of (2.7 and 4 mm) endoscopes and nasal and paranasal sinus instrumentation. We evaluated the ease of access of each exposure, accessibility of all cyst walls, wall remnants after excision, preservation of intact surrounding tissues (Schneiderian membrane, oral mucosa, teeth, and surrounding nerves), and complications. Under general anesthesia and adequate infiltration with 1/200,000 adrenaline, three surgical approaches were tailored, allowing viewing and single-handed endoscopic enucleation:

Endoscopic-assisted minimal sublabial approach

A trapezoidal flap was created through a minimal sublabial approach. The incision starts from the lateral portion of the lateral incisor to the lateral portion of the second premolar and first molar through the upper gingivobuccal sulcus. The flap of the upper mucogingival flap was elevated to total thickness, followed by exposure of part of the cyst. In cysts originating from the central incisors, a small bony window flap from the anterior wall of the maxilla was fashioned using bone drills. In cysts originating from lateral incisors and canines, where the anterior bony wall was thinned or perforated, the latter was removed peripherally with a rongeur until it reached the compact bone. The small bony windows can allow the insertion of the endoscope and the instruments. Complete enucleation was done using the elevators. The cysts were dissected from the lateral wall, the antral mucosa, the mucosa of the nose floor, and the oral mucosa in that order. We cauterize the remaining attached pedicle and suture the flap. A small inferior meatal antrostomy was done to drain and follow the cavity if the cyst's superior wall exceeds the inferior turbinate's height.

Endoscopic endonasal minimal rhinotomy approach

An incision was made in front of the anterior end of the inferior turbinate. The incised edges were retracted with hook retractors, and periosteal elevators were used under endoscopic guidance. A small part of the thinnest bony wall over the cyst wall was removed using bone drills or osteotomes and a hummer to create a nasal window to approach the cyst but not the maxillary sinus. With the expansion process, the related bone can be observed mainly at the junction of the nasal floor and the inferior part of the frontonasal process of the maxilla (site of the endonasal window).

Using elevators, the cysts were dissected from the nasal floor, the antral mucosa, and the oral mucosa.

Approach using an endoscopic endonasal broad middle meatal antrostomy

A pediatric side-biting forceps was used to dissect the uncinate retrogradely quickly. From the inferior turbinate to the agger nasi, the free margin of the uncinate served as the starting point for this retrograde dissection. The tool was continuously utilized without taking it out of the nose between each mouthful. We ensured that this window was finished, maintaining the entire lacrimal system and cystic walls intact. Additionally, it is crucial to enlarge the middle meatal antrostomy beyond normal limits to widely fenestrate the maxillary sinus' medial wall. The superior floor of the orbit, the anterior lacrimal duct, and the inferior floor of the nose serve as the boundaries of this prolonged antrostomy (with the consequent removal of the inferior turbinate).

Associated pathologic changes in the nasal and paranasal sinuses were treated using trans-nasal endoscopic sinus surgery (ESS). The surgical specimens were sent for histopathological analysis. All the cases were followed for 2 years through ORL and dental evaluation of symptoms and examination.

Results

We assessed six patients, ranging in age from 15 to 40 years; of these, 4 (67%) had periapical (radicular) cysts, and 2 (33%) had calcifying odontogenic tumor and ameloblastoma; all were enucleated endoscopically. Patient demographic, clinical, and CT characteristics, as well as the histopathological types of the cysts/benign tumors and endoscopic surgical approaches, were shown in Table 1. An overall opacity and weakening of the sinus walls were seen on a CT scan of cysts. The densities of the lesions were constant and homogenous. Some cysts still seem convex even if the sinus walls have eroded and the neighboring structures were not infiltrated (Figs. 1 and 2). Fake teeth and the origin of pathology can affect the walls affected in the scalloping process (Fig. 1). Cysts related to central incisors displaced the floor of the nose and soft palate (Fig. 1A, B). Cysts related to the canine displaced the floor of the nose and maxillary sinuses together with the medial wall and anterior wall of the same sinuses (Fig. 1C, D). Cysts related to the molar affected the maxillary sinus floor and medial walls (Fig. 1E, F). Three lesions (50%) were contacting the nasal septum. Medial maxillary wall scalloping existed in four patients (67%). Expansion of the lateral wall existed in 3 (50%) patients. Extensions below the level of the nasal floor were in four lesions (67%). Four patients had associated pan rhinosinusitis (67%) (Figs. 1 and 2), maxillary rhinosinusitis alone [1 patient (17%)] and no rhinosinusitis [1 patient (17%)] (Fig. 2). The mean CT scan score was

Table 1 Demographic, clinical, CT characteristics, histopathological types of the cysts, and endoscopic surgical approaches

Variable	Characteristics	Number (%) of patients [Total 6]
Patient demography	-Age: 15–40	
	-Sex:	
	-Male	2 (33%)
	-Female	4 (67%)
Patient presentations	- Asymptomatic	1 (17%)
	- Facial swelling	2 (33%)
	- Nasal congestion and discharge	2 (33%)
	- Facial pain	1 (17%)
CT criteria of bone displacements	-Septal and nasal floor	1 (17%)
	-Nasal and maxillary floor	1 (17%)
	-Maxillary floor alone	2 (33%)
	-Anterior wall	2 (33%)
	-Medial wall of maxillary sinuses	3 (50%)
	-Lateral wall of the maxillary sinus	2 (33%)
Histopathological types	- Periapical (radicular cysts)	4 (67%)
	- Calcifying odontogenic tumor	1 (17%)
	- Ameloblastoma	1 (17%)
Endoscopic surgical approaches	-Endoscopic-assisted minimal sublabial approach	4 (67%)
	-Endoscopic endonasal minimal rhinotomy	1 (17%)
	-Endoscopic wide middle meatal antrostomy	1 (17%)

11 with a range (of 3–14). An endoscopic assisted minimal sublabial approach was used [4 patients (67%)] for central lesions (cysts originating from the medial incisors with expansion only into the nasal cavity floor) [1 patient (17%)], as well as for cysts originating from the canines (elevating the nasal and antral floors) [1 patient (17%)], extension below the level of the nasal floor and lateral expansions and extensions with or without scalloping of the anterior wall of the maxillary bones [2 patients (33%)]. Ease of access was evident in patients treated with an endoscopic assisted minimal sublabial approach. An inferior meatal antrostomy was done in such an approach to follow-up the cavity.

An endoscopic endonasal minimal rhinotomy approach was used for cysts and tumors localized to the anterior wall and the nasal process of the maxilla in one patient (17%) (Figs. 2 and 3). Continuous traction of the incised lateral wall of the nose was encountered during the endoscopic endonasal minimal rhinotomy approach. A cyst with preserved maxillary bone boundaries (intra-sinus cysts) originating from the molars and induced intrusions expansion of the cyst with the maxillary floor at the level of the nasal floor was evident in one patient (17%) (Figs. 1 and 2). It was removed via endoscopic wide middle meatal antrostomy, followed by cystic enucleation. The cyst walls were completely removed in all patients. The teeth were preserved in all. No wall remnants were observed after excision, with preservation of the intact surrounding Schneiderian membranes, oral mucosa, teeth, and surrounding nerves. In postoperative follow-up, minimal edema was observed in the four patients treated via endoscopic assisted minimal sublabial approaches. The edema resolved after treatment with anti-inflammatory drugs for 5 days. One patient with a periapical cyst related to incisor teeth had a postoperative septal abscess drained trans nasally successfully. With follow-up for 2 years through ORL and dental evaluation of symptoms and examination, no recurrences were observed.

Discussion

Large odontogenic cysts and tumors are uncommon, presenting as ordinary CRS or swelling in the palate, gingivolabial sulcus, and cheek [14]. The present clinical series studied the expansion process of large odontogenic cysts and tumors into the sinonasal passages. To the best of our knowledge, this is the first study examining different endoscopic approaches used in enucleation according to the expansion process of large odontogenic lesions into the sinonasal passages. The affected teeth and origin of pathology can affect which sinonasal walls participate in the scalloping process. In addition, the most critical bony landmarks identified

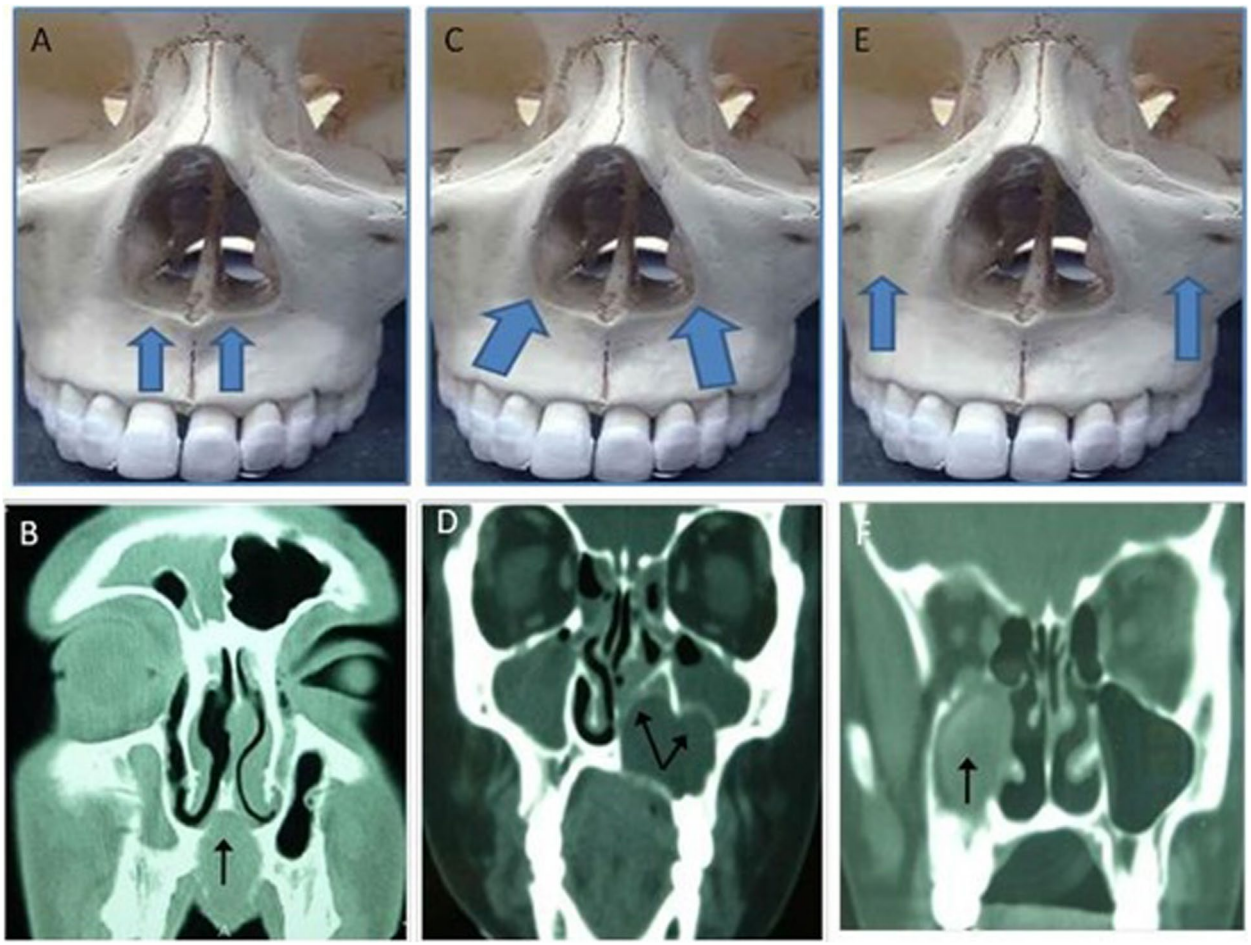

Fig. 1 Relationship between the origin of odontogenic cysts and direction of expansion of the maxillary walls. **A** Cysts originating from the central incisors with the direction of expansions (arrow). **B** Coronal CT image of a periapical cyst, showing an expansive process displacing the nasal cavity floor (black arrow) with intact and sclerotic borders. **C** Cysts originating from the lateral incisors and canines with directions of expansion (arrows). **D** Coronal CT image of a periapical cyst of the maxilla originating from non-vital left canine, showing an expansive process displacing part of the nasal cavity floor, inferior turbinate, and maxillary sinuses, with intact and sclerotic borders and occupying the majority of the left maxillary sinus (arrows). Associated pan rhinosinusitis was also observed. **E** Cysts originating from premolars and molars with the direction of displacements of the maxillary sinus floor (arrow). **F** Coronal CT image of a radicular cyst originating from the right second molar, expanding, and occupying the majority of the left maxillary sinus (arrow) with a very thin sclerotic border (arrow)

in choosing the appropriate approach are the level of the lesion to the nasal septum, the floor of the nose together with the lateral maxillary wall expansions.

In the present study, odontogenic cysts originating from the central incisors can give rise to septal and nasal floor elevations. Maxillary sinuses can be spared as they are the farthest from the roots of the incisors. This expansion results in ultra-thin floors of the nose bone barriers, making the endoscopically assisted sublabial approach appropriate for enucleation with safe barrier preservation. Following minimal removal of the bones at the anterior wall of the cysts, endoscopy can safely identify nearby roots.

The front sinus wall is formed by the lateral wall of the maxilla, which ranges in thickness from 2 to 5 mm [15]. Above the infraorbital foramen, the labial levator and the orbicularis oculi muscle join this wall and send growth to the maxillary sinuses. We found that limited wall thickness allowed scalloping, thinning, and loss of a portion of the lateral wall of the maxilla. The cysts always originate from nearby lateral incisors and canines, allowing elevation and expansion of the nose floor and antral mucosa. Subsequently, the endoscopic transnasal

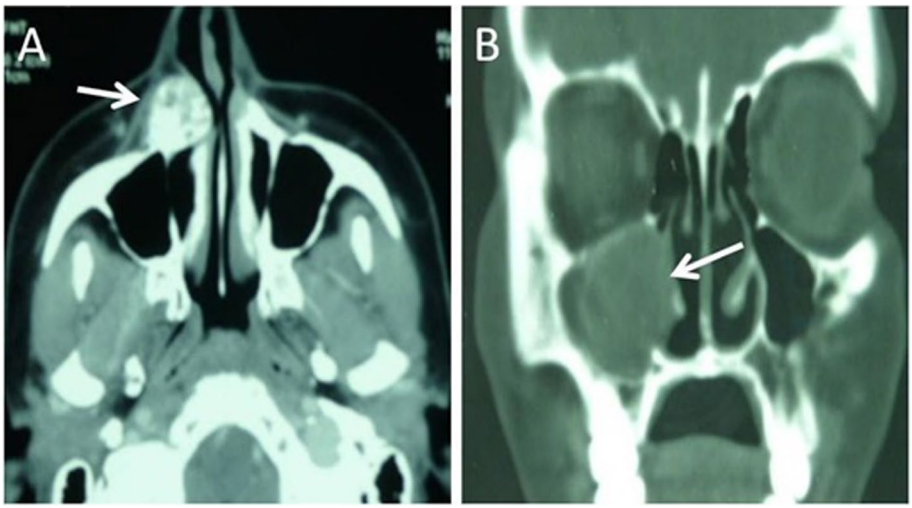

Fig. 2 CT scan of lesions enucleated endoscopically via trans-nasal approaches. **A** Calcifying odontogenic tumor (white arrow) excised via endoscopic endonasal minimal rhinotomy. **B** Right periapical maxillary intra-sinus cyst with scalloping of the maxillary sinus walls (white arrow) enucleated via endoscopic wide middle meatal antrostomy

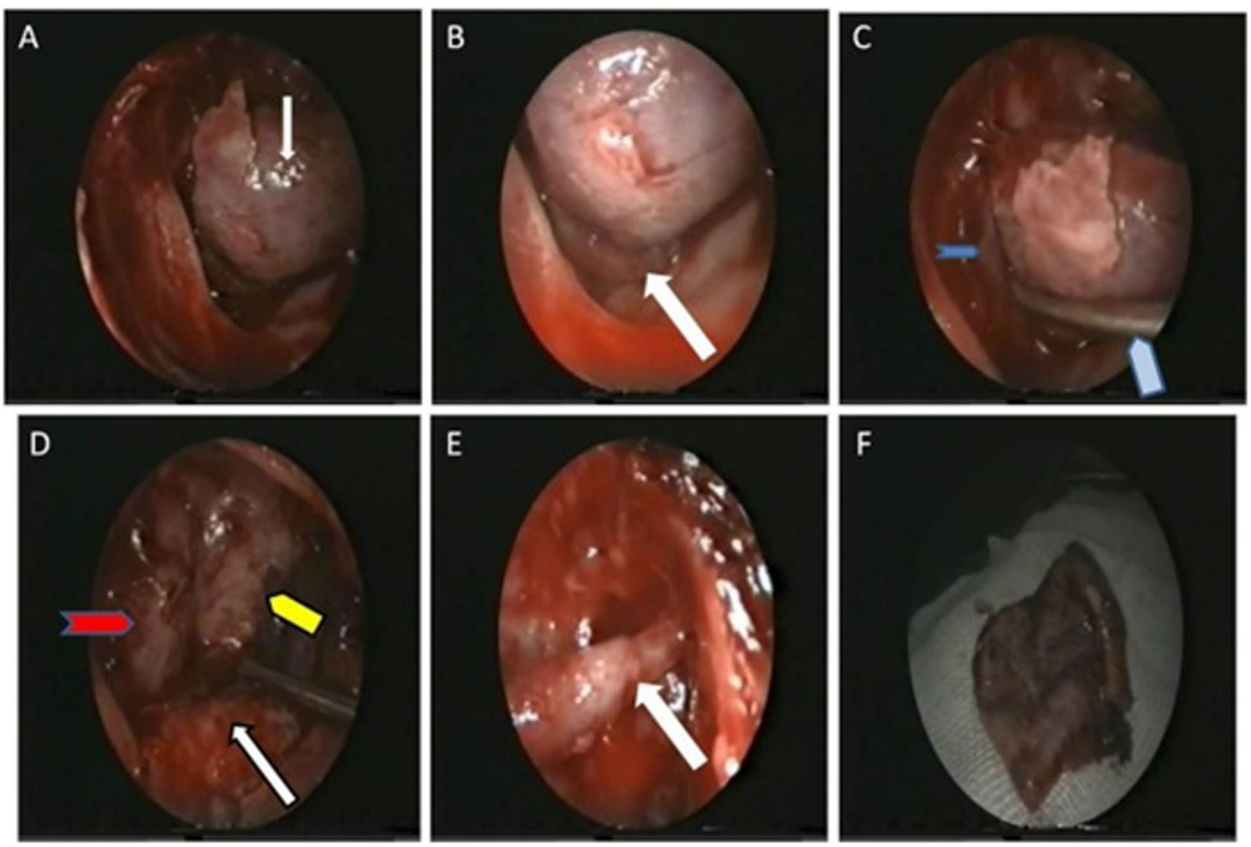

Fig. 3 Endoscopic view for endoscopic endonasal minimal rhinotomy approach: for enucleation of a left periapical cyst originating from the upper left canine. **A** The cyst wall (arrow) within its expanding cavity. **B** Dissection of the cyst wall (arrow) from part of the maxillary wall. **C** Dissection using a dissector (pentagon), elevating the cyst wall from the antral mucosa (chevron). **D** Complete dissection of the cyst (arrow) from the antral (chevron) and nasal (pentagon) mucosae. **E** Dissection of the cyst from the floor with a remaining attached pedicle that was cauterized. **F** The final cyst after excision

approach will not allow adequate exposure for excision without remnants [12]. The endoscopic assisted sublabial approach can expose the nasal floor and all maxillary sinus boundaries and identify the remaining cyst walls. Using the scalloped or perforated anterior maxillary wall as a window can result in adequate exposure with minor trauma to the existing bone and better cosmetic results. The odontogenic cysts elevate the inferior floor sinus wall above the level of the inferior meatus. We aim to preserve the mucoperiosteal wall of the maxillary sinus (that separates the sinus from the cyst wall) and to approach the cyst but not the maxillary sinus; the inferior meatal window allows dissection of the cyst, drainage of the cavity, and follow-up.

Another endoscopic approach that can be used in managing odontogenic cysts is the endoscopic endonasal minimal rhinotomy approach. In the present study, this exposure can be utilized in lesions localized to the anterior wall and the nasal process of the maxilla, such as odontogenic cysts arising from premolar roots [15, 16]. These cysts, therefore, expand upwards and posteriorly, elevating the maxillary sinus floor. So, the same approach can allow adequate exposure and enucleation of the cyst wall. A case report used an endoscopic endonasal approach for marsupialization of odontogenic keratocytes in 6 years old boy [17]. Another series involved thirteen patients with cysts removed by a trans-nasal endoscopic approach via the inferior meatus lateral wall after resection of the anterior end of the inferior turbinate. Because the same approach was used without tailoring in all the patients, (38%) of the patients had cyst wall residues [12].

Above the infraorbital foramen, this wall serves as the location of the labial levator and orbicularis oculi muscles, which guide expansion to the maxillary sinuses [15, 16]. This proximity explains why cysts arising from the premolars and molars can invade the maxillary sinus. The cysts may take on the structure of the maxillary sinus throughout this growing process as well-defined intra-sinus cysts. Wide middle meatal antrostomy permitted the enucleation of these cysts. Also, the same endoscopic approach could be used in children due to the high floor of the nose and the presence of unerupted teeth. Other reports used a similar approach successfully in dentigerous cysts localized to the maxillary sinuses [10, 11]. Other possible endoscopic options are applying wide-angled endoscopic in prelacrimal window approach (PLWA), endoscopic standard medial maxillectomy, and modified Denker's by way of a canine fossa trepanation. Such approaches had some limitations in odontogenic cysts as we need to dissect and approach the cyst circumferentially but not the maxillary sinus. A crucial factor for the performance of a PLWA is the length of the bony window anterior to the nasolacrimal duct, with the prevalence of Simmen's complex feasibility types 1 in most of our cases. Also, the need for more downward and circumferential dissection of the whole cyst walls with preservation of maxillary mucosa, bones, and essential structures makes these approaches challenging to approach the goal.

Endoscopy in all patients with large benign odontogenic cysts extending into nasoantral regions allowed the accurate exploration of the operative field, including views surrounding the cyst (e.g., the corners and areas difficult to access) and improving the complete removal of the cystic lesion. Endoscopy also allowed us to monitor closely the separation of the cyst lining from the antrum, nasal mucosa, and essential structures such as the teeth and the inferior alveolar nerve. This advance also limits the extension of surgical approaches. The present algorithm (Fig. 4) allowed endoscopic enucleation of large odontogenic cysts without wall remnants. Only one of six patients experienced postoperative complications, a septal abscess.

In most cases, odontogenic CRS was associated with the present series (83%). A combination of medical and surgical approaches was required to treat such association. Concomitant endoscopic management of associated CRS was performed to ensure complete infection resolution and prevent recurrence and complications [18, 19]. As pan CRS was present in 67%, working through the natural ostia in ESS; allowed control and drainage of inflammation with the preservation of Schneiderian membrane barriers. Similar trans-nasal ESS has been advocated for the treatment of odontogenic sinusitis (due to different pathologies) in 64% [20], 70.4% [21], and 18% [22].

As one of the cases is ameloblastoma, it is well comprehended that it has unique and specific criteria and behavior compared to the other presented lesions; for instance: it is characterized by a high recurrence rate and has more aggressive behavior. Therefore, it needs a more specific way of treatment compared to other odontogenic cysts, which may include a wider excision with a safe margin and very intensive postoperative follow-up care compared to the others. Although this appears difficult with the closed endoscopic approaches mainly for multicystic lesion, no recurrence was observed in our unicystic amyloplastoma with endoscopic wider margins of resection, complete enucleation, and close follow-up.

As an alternative to trans alveolar or lateral window methods, the endoscopic procedure is linked with minimal morbidity and low recurrence rates. Additionally, endoscopic surveillance offers early recurrence identification that can be effectively treated with minimal

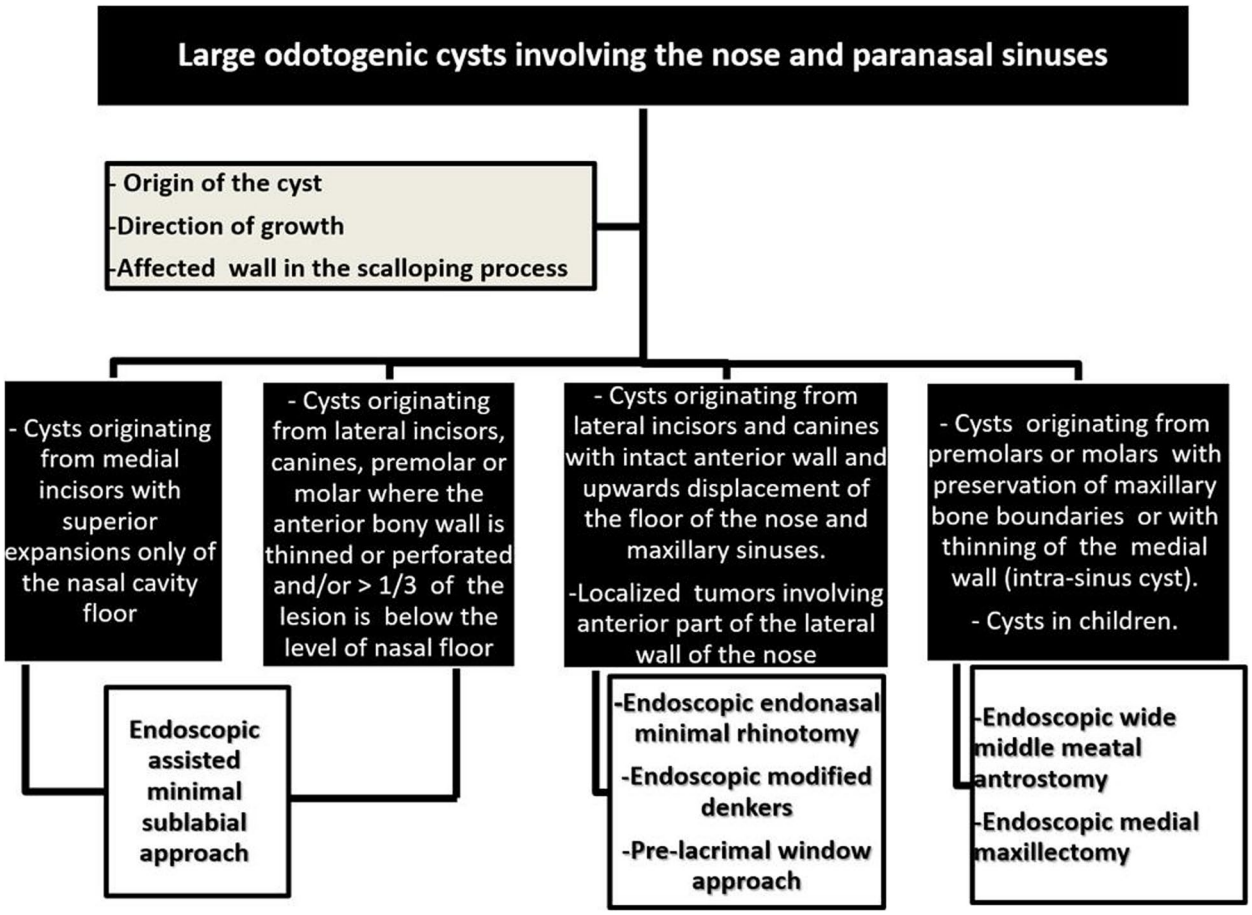

Fig. 4 Algorithm for endoscopic enucleation of large odontogenic cysts and tumors

associated morbidity [23]. Additionally, a combined technique provides comprehensive coverage of any pathological condition inside the paranasal sinuses and improves the surgeon's ability to manage it [24]. In the present study, we advise the present algorithm (Fig. 4) for endoscopic enucleation of large odontogenic cysts and tumors. This research has certain limitations, including the limited number of patients enrolled due to little referral and presentations to otorhinolaryngology specialty. So, we advise a multicenter study with a high number of cases to assess the different approaches according to our algorithm.

Conclusion

Tailoring surgical approaches according to the origin of cysts, the direction of growth, and the wall affected in the scalloping process can allow assisted endoscopic enucleation of large odontogenic cysts without wall remnants while preserving the integrity of the surroundings. More case-control studies are needed.

Abbreviations

CRS: Chronic rhinosinusitis; CT: Computed tomography; ESS: Endoscopic sinus surgery; NDRCT: Non-surgical dental root canal therapy; OKC: Odontogenic keratocyst.

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Authors' contributions

AR provided the concept, and design with the definition of the intellectual content, and conducted clinical studies. HA conducted clinical studies, data collection and analysis, and manuscript editing. All authors read and approved the manuscript.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

Approval was obtained from the Institutional Review Boards (IRB) of the Menoufia Faculty of Medicine and following the Declaration of Helsinki. After an explanation of all aspects of the study and being given the right to

withdraw at any time, informed written consent to participate in the study was provided by all participants (or their parent or legal guardian in the case of children under 16).

Consent for publication
Not applicable.

Competing interests
The authors declare that they have no competing interests.

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