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Case Series

Combined Endoscopic Endonasal and Endoscopy-Assisted Transoral Approach for Resection of Maxillary Ameloblastoma: A Case Series

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Abstract

Introduction: Ameloblastoma is a benign tumor of odontogenic origin arising in the mandibular region, and less commonly, the maxilla. Maxillary tumors are complex, and left untreated, can cause destruction of surrounding structures. The aim of this case series is to introduce an endoscopically guided, transnasal and transoral method for the resection of maxillary ameloblastoma with appropriate tumor margins.

Cases Summary: Our study includes 3 cases of recurreny maxillary ameloblastoma. Revision resection was completed via a transnasal or combined transnasal and transoral approach. Our combined approach involved removal of the floor of the maxillary sinus through an incision on the upper jaw and endoscopic resection of tissue. Diseased areas of the maxillary bone were removed with appropriate margins (1cm) without bony resection and facial reconstruction. Post-op imaging at 15 months (case 1) and 12 months (cases 2 and 3) show no recurrence.

Conclusion: Endoscopic sinus surgery (ESS) offers a minimally invasive technique for sinonasal access. However, its application in ameloblastoma remains underexplored, with only 7 reported cases. ESS enables precise resection with enhanced visibility, and a transoral approach can achieve wide-margin removal while minimizing facial incisions. This hybrid method is a promising option for managing maxillary AM without radical surgery.

INTRODUCTION

Ameloblastoma is a rare, benign tumor of odontogenic origin that arises from the embryonic enamel organ, basal epithelial cells of oral mucosa, or the remnants of the dental lamina in the mandible, or less commonly, the maxilla [1-3]. While not malignant, AM is locally invasive and left untreated, can cause damage to bony structures with high rates of morbidity and facial disfiguration [3-5]. The most common AM type is the solid or multicystic tumor, which is more aggressive, with multiple cystic areas that spread through the bone marrow [2-4]. Others include the unicystic type, showing a single cyst-like lesion within the bone, and uncommonly, the peripheral type, where the tumor occurs outside the bone [2-7]. In rarer instances, AM manifests as the metastasizing type, which transforms into malignant cells and spreads to distant sites and organs [2-7].

Diagnosis of AM relies on histopathological analysis for tumor characterization [8], which further directs treatment [9]. Histologically, AM presents with multilayered odontogenic epithelium with palisaded and elongating peripheral basal cells, and depending on subtype, can present with single or multiple microcysts in the tissue [8]. Computed Tomography (CT) can further be used for assessment of disease presence, degree of tissue involvement and tumor size [6]. Radiographic findings

include lytic lesions and a "soap bubble" appearance of the bone in multicystic disease, characterizing the presence of an ameloblastoma [10].

Surgical excision of the tumor is the sole definitve treatment for AM [5]. The choice of surgical technique often depends on tumor classification and degree of invasion [5,6]. Typically, solid and multicystic subtypes are removed using radical surgery, regardless of tumor size [11]. Given more invasive disease, radical surgery is followed by primary bony reconstruction, but complications with speech, swallowing, and future access to the maxillary sinus may be a challenge [11-14]. Therefore, other surgical methods that maintain low recurrence while preventing major facial structural change should be explored. This paper presents three cases of ameloblastoma managed via a complete endoscopic endonasal surgery, with or without a transoral approach, and using wide margin resection.

Case 1

This case, previously reported by our research team [13], presents a 64-year-old Caucasian male with a history of asthma, who was initially diagnosed with recurring allergic fungal rhinosinusitis following symptoms of nasal blockage, congestion, and facial pain. During revision Endoscopic Sinus Surgery (ESS), an incidental mass originating from the floor of the left maxillary sinus was discovered and biopsied. Histopathologic analysis showed cyst-like structures lined by columnar epithelial cells and divided by stroma-appearing spindle cell material, leading to the diagnosis of maxillary AM (Figure 1A). A computed tomography scan of the head and neck showed a 3.8 x 2.4 x 1.0 cm tumor, with borders limited to the inferior aspect of the left maxillary sinus and tethered inferomedially to the dental root of tooth 2-7 (upper left third molar), with extention into the left cheek inferolaterally (Figures 2A and 2B). A subsequent endoscopic approach was carried out 3 months after initial biopsy with the aim of complete surgical resection.

All steps of the subsequent surgical excision were completed via an image-guided endoscopic endonasal approach. Through an endoscopic endonasal medial maxillectomy, and with the aid of angled instrumentation, the entire floor of the maxillary sinus was accessed. The endoscopic view permitted precise visualization and navigation along the inferior aspect of the maxillary sinus floor. The borders of the ameloblastoma were clearly demarcated, and a 0.5-mm margin was marked circumferentially. Histopathologic analysis confirmed negative tumor margins. The delineated mucosa was elevated, leaving exposed and dehiscent bone. The lesion

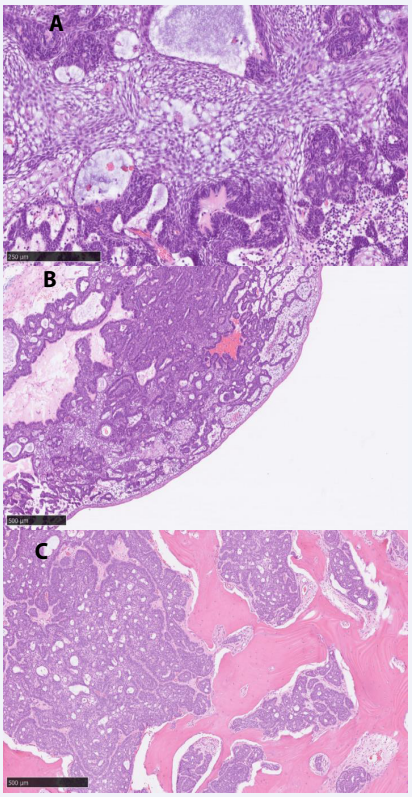

Figure 1 Histopathologic slides of ameloblastoma, multicystic type (A) High power view (200x) showing stellate reticulum and basaloid cells; (B) HE section showing low power view (100x) of ameloblastoma under the sinus epithelium arranged in follicular and plexiform architecture; (C) Low power view (100x) of tumour invading the bone.

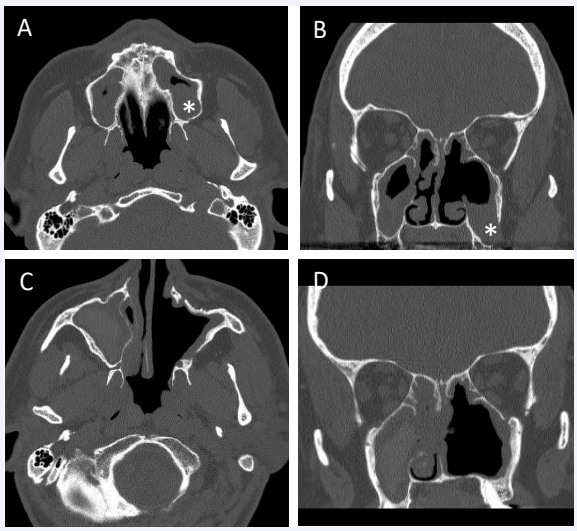

Figure 2 Case 1; (A) Axial view, (B) Coronal view; Pre-operative CT scan of left maxillary sinus AM measuring 3.8 x 2.4 x 1.0 cm present within inferior aspect of the left maxillary sinus with bony erosion at the posterolateral aspect of the maxillary sinus wall and into the left cheek; (*) denotes tumor soft tissue adjacent to eroded bone. (C) 15-month post-operative axial view of left maxillary sinus, (D) 15-month post-operative coronal scan left maxillary sinus shows no tumor recurrence in left maxillary sinus; opacification in the right maxillary sinus due to fungal rhinosinusitis.

was then excised in its entirety. A 4-mm, 70-degree angled diamond burr was subsequently employed to polish and smooth the bony floor of the left maxillary sinus extending to the root of the second to seventh molars. At the molar root, monopolar cauterization was applied to eliminate any potential tumor remnants. Finally, Surgiflo hemostatic matrix was placed over the resection site following complete tumor clearance. The patient remained disease-free for four months, but a minor recurrence was identified at the 6-month follow-up visit, spanning from the anterior maxillary wall to the posterior aspect of the inferior turbinate. The recurrent tumor was excised via an endoscopic transnasal approach with 1.0cm margins, and the resection margins were confirmed negative on reapeat frozen and permanent histopathology. At the final follow-up, 15 months post-surgery, no evidence of recurrence was observed (Figure 2C and 2D). The affected tooth was further extracted by the patient's dentist post-operatively. The patient's nasal cavity and sinuses were imaged via camera guidance before and after the procedure, and show satisfactory healing of the nasal cavity (Figure 3).

Case 2

We present a 63 year-old Iranian female with a diagnosis of AM confirmed on biopsy. On initial occurrence of the tumor one year prior, the patient had presented to a different center with facial pain, pressure, and congestion. She had undergone an endoscopic endonasal modified medial maxillectomy for the removal of the left maxillary ameloblastoma shortly after this presentation. Documentation from this initial surgery were unavailable.

In early 2025, the patient presented to our center with an asymptomatic recurrent AM in the left maxillary sinus, as displayed on a 1-year post-op MRI scan, showing the AM extending from the second molar, through the pterygoid plates and into the maxillary sinus, measuring 2.3 x 2.7 x 2.8cm (Figure 4). On presentation, the patient did not report any numbness or pain; however, she did experience nasal congestion. We planned a combined endoscopic endonasal,

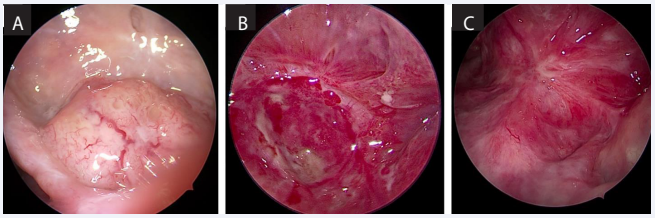

Figure 3 Case 1 (A) Nasal endoscopy of the left maxillary sinus shows AM tumor; (B) 3-months post-operative endoscopy of left maxillary sinus, no tumor recurrence and healing; (C) 15-months post-operative endoscopy of left maxillary sinus, continues to have no AM recurrence, healed mucosa.

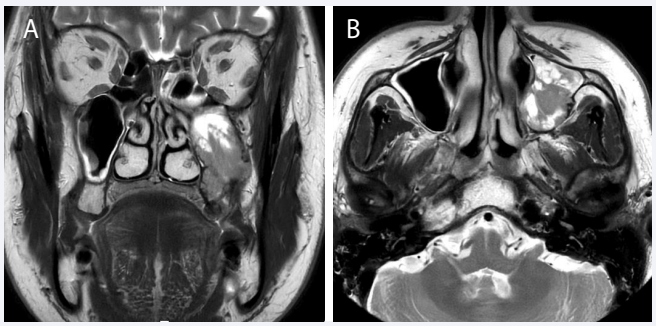

Figure 4 Case 2; (A) Coronal view, (B) Axial view; T1, Gadolinium-enhanced pre-operative MRI of left maxillary sinus AM measuring 2.3 x 2.7 x 2.8cm. Expansion of tumor from the left 2nd molar with involvement of the left pterygoid plates; hyper-densities visible within AM tumor.

computer-assisted resection of the bony tumor along with an endoscopic-assisted transoral approach for complete tumor excision, dental extraction, and reconstruction of the resulting oroantral defect using local flaps. To achieve adequate access to the maxillary floor, a left medial maxillectomy was performed. This was accompanied by a drill-out of the involved medial and lateral pterygoid plates up to the root of the pterygoid process. A frozen section of the posterior wall of the maxillary sinus was obtained and sent for histopathological evaluation, which showed a multicystic AM with arranged in follicular and plexiform architecture and visible invasion into the bone (Figure 1C). Once negative tumor margins were confirmed, we proceeded to drill an additional 1 cm margin toward the infratemporal fossa.

This was followed by a transoral excision of the second maxillary molar. The anterior and medial bony margins were further evaluated and drilled out. As a result, an oroantral defect measuring approximately 3 cm was created (Figure 5A). Reconstruction was performed using an inlay buccal fat graft and an onlay rotational palatal mucosal flap from the ipsilateral side. The donor site was covered with Biodesign, Surgicel, and Gelfoam, secured with a bolster, and sutured to the adjacent tooth. The maxillary sinus was packed with Surgiflo at the end of the procedure. The patient remained disease-free, as confirmed by postoperative CT imaging at 3 month follow-up (Figure 6A and B). At the 6 month follow-up, no healing problems or fistula were observed (Figure 5B). Additional follow-up MRI imaging and assessment at 12 months showed no recurrence (Figure 6C and D).

Case 3

Our third case involves a 68-year-old male with a history of Crohn's disease, in whom an incidental diagnosis of ameloblastoma was made on CT performed to rule

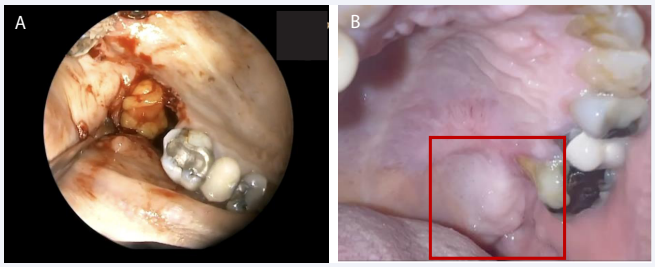

Figure 5 Case 2 (A) Intra-operative endoscopic view of the oral cavity with trans-oral removal of tumor; (B) 6-month post-operative visualization of the oral cavity shows healed mucosa (red box) with the 2nd molar removed, no fistulae visible.

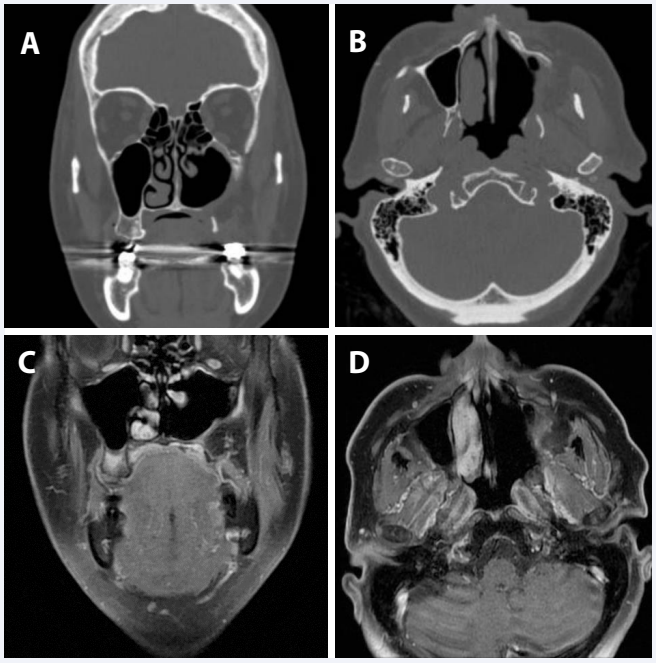

Figure 6 Case 2 (A) Coronal view, (B) Axial view; Post-operative CT scan of left maxillary sinus at 3 months with no tumor recurrence. (C) Coronal view, (D) Axial view; T1, Gadolinium-enhanced post-operative MRI left maxillary sinus at 12 months with no tumor recurrence, fat pad remains in place in axial view.

out stroke. He had previously undergone a right-sided endoscopic medial maxillectomy for AM resection four years prior at our center. At the time of this initial surgery, tumor tissue was removed from the floor of the maxillary sinus and pterygopalatine fossa. Healing was complete with no presence of oro-antral fistulae. CT imaging at 6 months and 1 year post-operatively showed no tumor recurrence. At a follow-up visit four years after surgery, soft tissue growth was identified and biopsy confirmed tumor recurrence. Imaging revealed a 2.3 x 2.4 x 1.8cm lesion extending from the infratemporal fossa, involving the right pterygoid plate and the floor of right maxillary sinus (Figure 7).

The patient underwent an image-guided combined endoscopic endonasal and endoscopic-assisted transoral

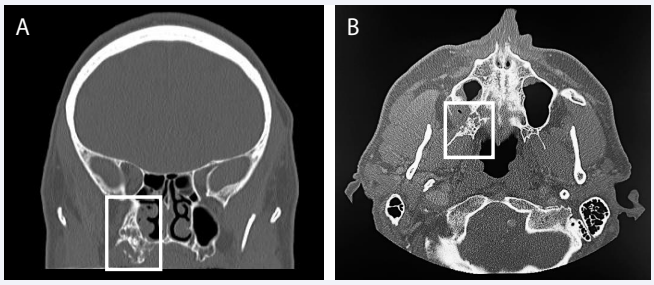

Figure 7 Case 3 Recurrent right maxillary AM pre-operatively; (A) Coronal view, 2.3 x 2.4 x 1.8cm lobulated soft tissue extending from the infratemporal fossa into the nasal cavity, soft tissue extension into roof of maxillary sinus with bony dehiscence and lytic changes; (B) Axial view recurrent AM. Involvement of the right pterygoid plate outlined by white squares.

approach for complete tumor excision, dental extraction, and reconstruction of the resultant oroantral defect with local flaps. A revision right medial maxillectomy was performed, and endoscopic evaluation demonstrated a large multiloculated ameloblastoma filling the right maxillary sinus. Tumor attachment to the pterygoid plate was completely drilled and removed. As in the previous case, margins from the posterior, anterior, and lateral walls, including both soft tissue and bone, were submitted for histopathologic evaluation, yielding similar results with bony tumor invasion (Figure 1). Intraoperative frozen section confirmed ameloblastoma. Resection was extended with a 1-cm margin off the pterygoid plate, which was confirmed negative intraoperatively.

The procedure continued with an endoscopic-assisted transoral approach, during which two molar teeth were extracted. The resultant 4-cm defect was reconstructed using an inlay buccal fat flap combined with an onlay local palatal flap (Figure 8A). The donor site was covered with Surgicel and Gelfoam, secured with a bolster, and sutured to an adjacent tooth, which was removed one week later. CT imaging at 3 months post-operatively revealed no tumor recurrence (Figure 9A and B). At the 6-month follow-up, healing was satisfactory with no evidence of fistulization,

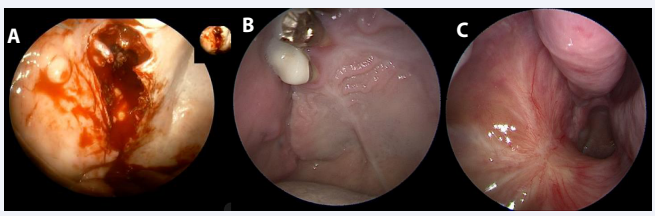

Figure 8 Case 3 (A) Intra-operative view of the oral cavity after complete tumor resection and tooth extraction; (B) 6-month post-operative view of the oral cavity shows appropriate healing, there is an area of soft tissue prominence due to scarring; (C) 6-month post-operative transnasal endoscopic view, scar healing after a medial matxillectomy.

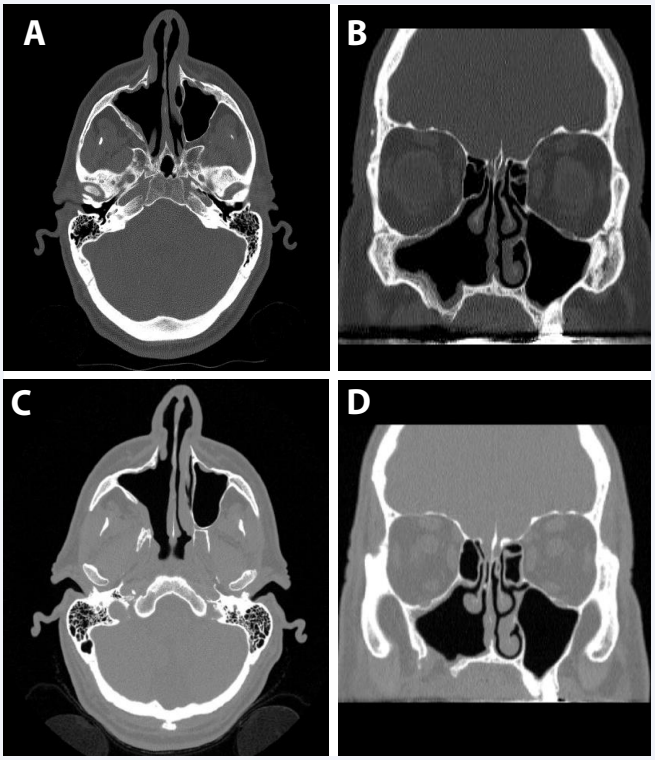

Figure 9 Case 3 (A) Axial view and (B) Coronal view; post-operative CT images at 3 months; no tumor residue or recurrence noted. (C) Axial view and (D) Coronal view; post-operative CT images at 12 months show no recurrence.

except for a residual prominence from scarring found on the hard palate (Figure 8B and C). CT imaging and clinical assessment at 12 months further showed no recurrence of tumor (Figure 9C and D).

DISCUSSION

Ameloblastomas are benign, locally-invasive tumors, with slow-growing spread to surrounding soft tissue and bony structures. As noted in the literature, A majority of cases (80%) occur in the mandible and among younger adults, while there fewer instances with maxillary involvement, which is present more commonly in those above 60 years of age [6]. Due to infiltration of the maxillary sinus, our patients presented with nasal congestion, facial pressure, and pain, aligning with other reported cases of maxillary AM in the literature [16-18]. As indicated in other case reports and reviews, this slow-growing tumor can culminate in diffuse bony spread and facial disfigurement [19-21]. In addition, it can severely impact the patients quality of life due to pain, cranial nerve involvement, and facial functional impairment. In some cases it can be life-threatening with invasion into the brain and skull base structure, warranting timely and adequate surgical management [22].

Surgical treatment of ameloblastomas can be accomplished through conservative or radical methods, with the latter traditionally preferred to reduce chances of tumor recurrence for multicystic tumors [23,24]. Radical approaches, while highly effective in preventing tumor recurrence and currently accepted as standard treatment, can cause substantial effects on the patient quality of life [25]. To mitigate these issues, reconstructive surgery is often a needed aspect of radical surgery, typically with the use of bony grafts [26,27].

While the mandible is more easily accessible, maxillary tumors are more difficult to reach with far more expansive bony removal and more challenging reconstruction [28]. Additionally, bone grafts can compromise access to the maxillary sinus [28]. If not used in anticipation of this issue, inadequate facial reconstruction causes difficulties in swallowing and speech [28]. With our suggested surgical methods, we present a technique that does not require facial reconstruction and eliminates the challenge of accessing the maxillary sinus, if needed for treatment of other pathologies or recurrences. Our method uses a video-assisted system with direct intransal or transoral visualization, combined with accepted margins of 1-1.5cm used in radical procedures [29]. Furthermore, radical approaches in maxillary tumor removal involve a large incision from the philtrum, vertically along the side of the nose, and across the lower lids horizontally, posing risks of scarring and difficult healing for patients [30,31]. This can impact the patient's self-image and appearance satisfaction [32]. In comparison, our patients did not have any visible scarring, and all incisions were done intranasally or intraorally, maintaining aesthetic satisfication.

Conservative methods of treatment include curettage, marsupilization, enucleation and cryotherapy. The primary challenge remains to be high recurrence rates, depending on the subtype of ameloblastoma [17]. For example, a study of 3 cases of unicystic AM of the mandible with decompression and enucleation reported no recurrence after 1 year [33]. However, studies show that other forms of AM, namely solid or multicystic tumors, may have recurrence rates of 29-91% using this approach [11-34]. Like our presented technique, conservative management has the benefit of preventing iatrogenic facial disfigurement and preserves critical functionality that may be compromised by radical surgery [20]. As described, this procedure differs from conservative approaches in maintaining appropriate margin removal when treating solid maxillary AM, demonstrating a technique that warrants further exploration in larger patient cohorts.

Table 1: Summary of ameloblastoma cases removed via ESS alone or combined approach.

Authors/year	Age	Sex	Surgery method	Tumor origin	Invasion	Tumor type	Tumor size	Recurrence
Kikuchi et al., 2024[31]	46	Female	Caldwell-Luc (via endoscopy) *and* Weber-Furguson minus subciliary incision	Left maxillary sinus	Bone invasion frontal sinus, pterygoid process, facial subcutaneous tissue	-	6cm x 5cm	None (6 month f/u)
Kosmidou et al., 2020[16]	74	Female	ESS	Right maxillary sinus	Nasal cavity, maxillary sinus periosteum	Multicystic	2.6cm x 3.1cm x 3cm	None (3 month f/u)
Leong et al., 2010 (abstract)[32]	-	-	ESS	Nasal ameloblastoma	-	-	-	None (12 month f/u)
Fiedler and Wunsch, 2021[15]	38	Male	ESS + Caldwell-Luc	Right maxillary sinus	-	-	-	None (4 month f/u)
Jain et al., 2013[33]	43	Female	ESS + transoral surgery with partial maxillectomy	Left maxillary sinus	Nasal cavity, bones above maxillary molars	-	-	None (12 month f/u)
Woodroffe et al., 2013[20]	60	Male	ESS + neurosurgery (trans-nasal)	Left maxillary sinus	Skull base and infraorbital fossa	-	-	None (18 month f/u) Adjuvant radiation therapy for 2 months
Karp et al., 2020 (Case 1 of this series)[13]	64	Male	FESS	Left maxillary sinus	Inferolateral walls of maxillary sinus	Multicystic	3.8 x 2.4 x 1.0 cm	Recurrent tumour at presentation; None at (15 month f/u)
Case 2 of this series	63	Female	Combined approach: ESS + endoscopic-assisted transoral surgery	Left maxillary sinus (recurrent)	Bony invasion into the left pterygoid plates from 2nd molar	Multicystic	2.3 x 2.7 x 2.8cm	Recurrent tumour at presentation; None at 12 month f/u)
Case 3 of this series	68	Male	ESS Combined approach: ESS + endocopy-assisted transoral surgery (recurrent tumor)	Right maxillary sinus (recurrent)	Bony invasion into right pterygoid plate and the floor of right maxillary sinus	Multicystic	2.3 x 2.4 x 1.8cm	Recurrent tumour at presentation; None at (12 month f/u)

Endoscopic Sinus Surgery (ESS) provides a minimally-invasive approach to treatment of sinus-related pathologies [35]. Within the literature, there are currently a total of 7 cases utilizing transnasal endoscopic approaches in the treatment of AM, of which 4 used an exclusively endoscopic approach and 1 used an endoscopic-assisted transoral approach, similar to what is presented in this seriess (Table 1). Notably, one of these reports has been published by our research team, and the case has been re-introduced in this case series [15]. Given the limited literature on this approach in AM removal, our group is providing additional evidence for potential successful treatment of AM using an image-guided endoscopic endonasal and endoscopic-assisted trans-oral approach. However, our study is limited in tumor recurrence timeline, as maxillary AM recurrence can occur anywhere between 1-10 years post-operatively, with some reports up to 30 years after primary surgery [9]. While our results are promising, our patient follow-up duration is between 12-15 months after initial surgery at this time. It would be of utmost importance to continue monitoring patient recovery longitudinally for more accurate assessment of successful treatment.

CONCLUSION

Maxillary ameloblastomas, though rare, present unique diagnostic and therapeutic challenges due to their proximity to critical anatomical structures and their potential for significant morbidity. As demonstrated in our case series and supported by limited literature, combined endoscopic endonasal and transoral approach allows for complete resection with adequate margins. Continued reporting of outcomes and long-term follow-up are essential to further define the role of ESS in the management of maxillary ameloblastomas and to establish evidence-based guidelines for surgical decision-making in this rare but impactful disease.

ETHICAL CONSIDERATIONS

Ethics approval (REB#H26-00858) has been obtained through our institution review board.

CONSENT TO PARTICIPATE

All participants in our case series provided written consent for the use of their information in this study.

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