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An Alternative Surgical Approach to Cemento-Ossifying Fibroma of the Maxilla

ABSTRACT

Objective: To report an alternative combined gingivobuccal and endoscopic endonasal approach to treating ossifying fibroma of the left maxilla.

Methods:

Design: Case Report

Setting: Tertiary Private University Hospital

Patient: A 19-year-old girl with a progressively enlarging left cheek mass for 3 years.

Results: Our patient underwent left medial maxillectomy via a combined gingivobuccal and endoscopic endonasal approach with complete resection and significant improvement of symptoms, with good functional outcome.

Conclusion: Large ossifying fibromas of the maxilla can be completely and successfully excised via a combined gingivobuccal and endoscopic endonasal approach.

Keywords: *Cemento-ossifying Fibroma; SNOT-22; Nasal Obstruction Symptom Test; Visual Analog Scale; combined approach; conservative approach*

Ossifying fibroma is a rare benign fibro osseous neoplasm of the jaw, composed of varying amounts of bone or cementum-like tissue in a fibrous tissue stroma.¹ It commonly occurs in the 2nd to 4th decades of life.^{1,2} Small lesions may be asymptomatic and may be diagnosed incidentally, and are usually solitary, may displace teeth and, in cases involving the maxilla, extend into the maxillary antrum. Some lesions may grow to become massive causing considerable aesthetic and functional deformities.^{1,3} Complete resection is curative and the exact surgical approach depends on the size, location and extent of the lesion.⁴ Small, asymptomatic lesions may benefit from a “wait-and-see” strategy with regular monitoring or may undergo curettage.⁵ Large lesions of the maxilla especially with infiltration to surrounding structures such as the anterior skull base, orbit and nasal cavity, may present a dilemma with regard to achieving complete resection while minimizing postoperative morbidities of a radical approach. We report a case of ossifying fibroma of the left maxilla treated with an alternative approach, combined gingivobuccal and endoscopic endonasal surgery.

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CASE REPORT

A 19-year-old girl presented with a 4 x 4 x 4 cm firm, non-tender progressively enlarging left cheek mass for 3 years. (Figure 1) Associated symptoms included left unilateral nasal obstruction, clear nasal discharge, left cheek numbness (75% sensory deficit) and occasional facial heaviness over the frontal area. She did not experience epistaxis, changes in smell, or loose dentition. Other pertinent physical examination findings were a nontender mass in the left hard palate and widening of the left gingivobuccal sulcus. (Figure 2) There was no history of trauma or atopy.

Nasal endoscopy showed the lateral nasal cavity wall bulging towards the septum, obstructing the left nasal cavity with clear nasal discharge; the right nasal cavity was unremarkable. (Figure 3) The Sino-Nasal Outcome Test (SNOT-22) was 89. She also scored 16 on the Nasal Obstruction Symptom Evaluation (NOSE) test, and 9/10 on the Visual Analog Scale (VAS) based on severity of nasal obstruction and facial pain. Punch biopsy via Caldwell-Luc approach showed Fibrous Dysplasia.

A contrast-enhanced CT scan of the Paranasal Sinuses (Figure 4) showed a low density non-enhancing soft tissue lesion with ground glass thickening of the maxillary bony walls completely occupying the left maxillary antrum. The lesion extended posteriorly into the nasopharynx, laterally with obliteration of the adjacent ostiomeatal unit, medially with erosion of the middle and inferior nasal turbinates and deviation of the nasal septum to the right, and inferiorly with downward bulging of the left hard palate and maxillary alveolar ridge. The orbital contents were unaffected.

The patient underwent a left medial maxillectomy via combined gingivobuccal and endoscopic endonasal approach under general anesthesia. Through the gingivobuccal approach, curettage of the left maxillary mass was performed until the lateral nasal wall was reached. (Figure 5A-B) By this time, the nasal cavity was accessible endoscopically, hence a modified endoscopic medial maxillectomy was performed. (Figure 5C) Curettage of the maxillary mass was completed and hemostasis was achieved. (Figure 5D-E) The resulting maxillary sinus cavity was packed with gauze soaked with petroleum jelly. The patient tolerated the procedure well and was sent home on the third postoperative day after nasal packing was removed.

After removal of the nasal pack, the patient reported significant decrease in symptoms with a VAS score of 5/10. During the first 2 weeks of follow-up, she had minimal nasal congestion with occasional mucoid discharge. A repeat SNOT-22 showed a score of 32, and a NOSE score of 5 on the second week after surgery, with reported overall improvement. She reported an improved sensory deficit to 30% over the left cheek. Nasal endoscopy at 4 months after surgery (Figure 6) showed smooth



Figure 1. 19-year-old girl with a 4 x 4 x 4 cm firm, non-tender progressively enlarging left cheek mass for 3 years



Figure 2. Oral cavity view showing nontender mass in the left hard palate with widening of the left gingivobuccal sulcus

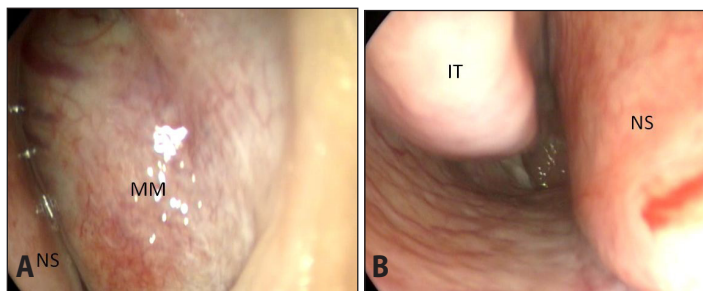


Figure 3. Preoperative nasal endoscopy: A. maxillary mass (MM) bulging from left lateral nasal wall obstructing left nasal cavity with clear secretions; B. unremarkable right nasal cavity. (IT, inferior turbinate; NS, nasal septum)

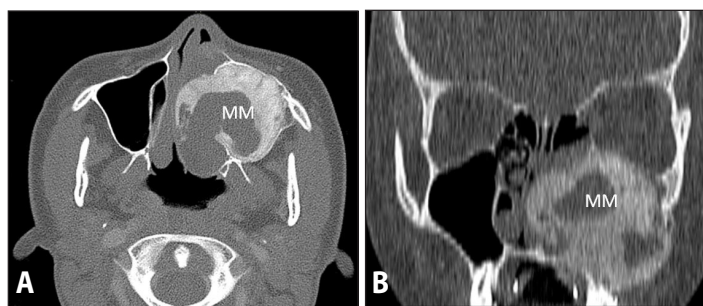


Figure 4. Contrast-enhanced CT scan of the paranasal sinuses, bone window: A. axial view with non-enhancing soft tissue maxillary mass (MM) with ground glass thickening occupying the left maxillary antrum and extending into the posterior nasopharynx, obstructing the left nasal cavity; B. coronal section showing maxillary mass (MM) bulging medially and deviating the septum to the right; also bulging inferiorly to the hard palate

nasal mucosa with no tumor recurrence seen. The maxillary antrum was well visualized. Histopathology of the excised specimen revealed cemento-ossifying fibroma.

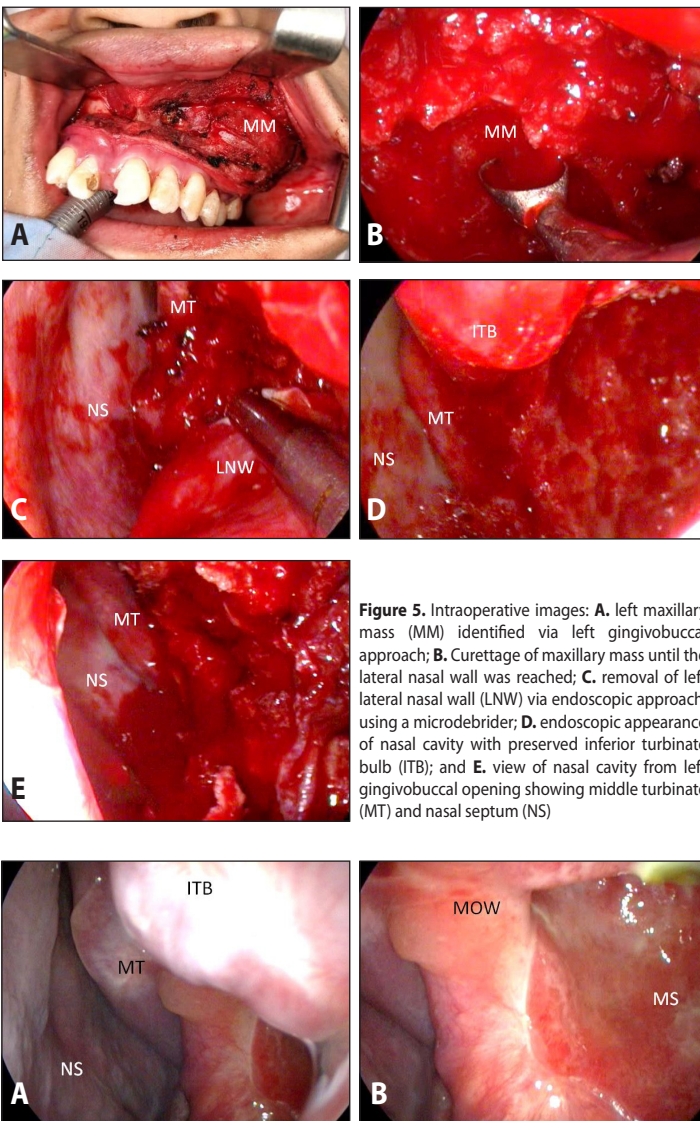


Figure 5. Intraoperative images: **A.** left maxillary mass (MM) identified via left gingivobuccal approach; **B.** Curettage of maxillary mass until the lateral nasal wall was reached; **C.** removal of left lateral nasal wall (LNW) via endoscopic approach, using a microdebrider; **D.** endoscopic appearance of nasal cavity with preserved inferior turbinate bulb (ITB); and **E.** view of nasal cavity from left gingivobuccal opening showing middle turbinate (MT) and nasal septum (NS)

Figure 6. Postoperative nasal endoscopy after 4 months: **A.** post-operative left nasal cavity using zero-degree scope showing healed mucosa; **B.** maxillary antrum with smooth mucosa using a 30-degree scope. (NS, nasal septum; MT, middle turbinate; ITB, inferior turbinate bulb; MOW, medial orbital wall; MS, maxillary sinus)

DISCUSSION

Ossifying fibroma is a rare, benign neoplasm of the maxilla and mandible, occurring mostly in patients in the 2nd to 4th decade of life, with a higher incidence in females compared to males. In a study by Chang *et al.*, 71% of cases occurred in the 2nd to 4th decade, with a mean age of 34 years, and a female predominance of 79%.³ In a case series by Liu *et al.*, the mean age was 37 years, occurring in females in 69% of cases.⁵ The lesion can affect both the maxilla and the mandible, with majority of cases occurring in the mandible, especially in the molar and premolar regions.^{1,3,6,7} The origin of ossifying fibromas is not clearly understood. However, it is considered that these lesions arise from

the periodontal membrane^{1,5,6} which contains pluripotent stem cells capable of producing cementum, bone and fibrous tissue.^{8,9}

The most common clinical presentation for ossifying fibromas is an asymptomatic slow-growing painless mass on the affected area.^{3,5,6,7} Other presentations include pain, surface ulceration, cortical plate perforation and periodontal ligament widening.^{3,6} Patients with maxillary lesions may present with nasal obstruction and/or epistaxis.^{5,7}

The Sino-Nasal Outcome Test (SNOT-22) is a symptom-based outcome measure that can measure changes in symptoms and also be used to predict postoperative outcome and improvement.¹⁰ The Nasal Obstruction Symptom Evaluation (NOSE) scale is another instrument that is reliable, valid, and responsive to changes in a patient’s clinical status before and after treatment, and may be used to or to compare different treatments such as medical versus surgical therapy, and outcome difference between surgical techniques.¹¹ For severity of symptoms, the visual analog scale (VAS) is a simple method of assessment recorded by the patient by giving a score on a scale of 1 to 10.¹² In this case, the therapeutic intervention resulted in a significant decrease in the SNOT-22, NOSE and VAS scores.

A computed tomography (CT) scan is the preferred imaging modality for these types of lesions.⁴ Predominant radiographic features of ossifying fibromas are a round, well-defined, expansile mass with a corticated border and varying degrees of radiopacity.^{1,5,6} It is diagnostically challenging to distinguish between fibrous dysplasia and ossifying fibroma.¹³ Both may exhibit similar clinical, radiographic and histologic features.² Radiographically, the most helpful distinguishing feature is the well-circumscribed appearance of ossifying fibroma.¹⁴

The definitive management for ossifying fibromas is surgical resection. The circumscribed nature of these lesions generally permits complete resection with relative ease.² The decision to pursue an endoscopic versus an open approach depends on the discretion and experience of the surgeon. Recent studies have reported excellent outcomes using an endoscopic approach, with improved endoscopes and instruments, increased surgical experience and routine use of navigation systems.¹⁵ Endoscopic advantages include direct visualization, enhanced magnification, absence of external deformities, and decreased morbidity versus open surgery.¹⁶ However, limitations and contraindications include tumor extension to the anterior wall of the frontal sinus, extension beyond the optic nerve, or extension to the anterior wall of the maxillary sinus.¹⁵ Larger lesions may require a more extensive approach due to tumor aggressiveness and high recurrence rates after incomplete removal.⁶ Characteristics of aggressive lesions include rapid growth, cortical bone thinning or perforation, orbital involvement and tooth displacement.¹⁷ These tumors can exhibit early recurrence, therefore a more radical approach is warranted.¹⁷⁻¹⁹



Titinchi *et al.*²⁰ recommended the following treatment modalities: 1) Enucleation may be done in cases where the lesion is small/medium in size, well-defined, encapsulated, and easily accessible; 2) Curettage is warranted when no clear borders are seen around the lesion, in cases when the lesion is composed of soft bone fused with surrounding normal bone; and 3) Resection is performed in lesions extending into the maxillary antrum and/or nasal cavities with ill-defined borders. An open or combined surgical approach, such as in the patient's case, is recommended for complete removal of large tumors that extend to the sinuses, especially in this age group, although there is little information in current literature. A conservative approach may preserve orbital function, masticatory function, and normal bone growth especially in adolescents, as well as aesthetic preservation, with complete resection of the tumor.¹⁹ In this case, the young age and the relatively limited extent of tumor provided a unique opportunity for conservative surgery via a combined gingivobuccal and endoscopic endonasal approach, maintaining nasal function and minimizing aesthetic deformity while achieving complete tumor resection.

Prognosis is very good in these cases. However, recurrence is higher in the case of maxillary ossifying fibromas compared to mandibular ones due to greater difficulty in surgical resection and large size at the time of presentation.⁶ Recurrence rates are estimated to range between 0% and 28% of cases, and occur after a period ranging from 6 months to 19 years.²¹ Regardless of the type and approach of resection, the importance of close follow-up should be emphasized, with an average follow-up period of 10 years after surgery.²¹

In summary, we presented an alternative approach to the management of ossifying fibroma of the maxilla in a 19-year-old girl. A combined gingivobuccal and endoscopic endonasal approach is an acceptable alternative approach in large lesions, resulting in complete excision of the lesion and restoration of normal function with good aesthetic results, which greatly impact the overall quality of life.

REFERENCES

- Jih MK, Kim JS. Three types of ossifying fibroma: A report of 4 cases with an analysis of CBCT features. *Imag Sci Dent*. 2020 Mar;50(1):65-71. DOI: 10.5624/isd.2020.50.1.65; PubMed PMID: 32206622; PubMed Central PMCID: PMC7078407.
- Khan SA, Sharma NK, Raj V, Sethi T. Ossifying fibroma of maxilla in a male child: Report of a case and review of the literature. *Natl J Maxillofac Surg*. 2011 Jan;2(1):73-9. DOI: 10.4103/0975-5950.85859; PubMed PMID: 22442615; PubMed Central PMCID: PMC3304240.
- Chang CC, Hung HY, Chang JYF, Yu CH, Wang YP, Liu BY, et al. Central ossifying fibroma: a clinicopathologic study of 28 cases. *J Formos Med Assoc*. 2008 Apr;107(4):288-94. DOI: 10.1016/S0929-6646(08)60089-3; PubMed PMID: 18445542.
- Jurlina M, Skitarelić N, Passal D, Passali FM, Mladina, R. Endonasal endoscopic resection of ossifying fibroma involving the ethmoid sinus, orbit and anterior skull base: case report and literature review. *Acta Otorhinolaryngol Ital*. 2016 Apr;36(2):144-8. DOI: 10.14639/0392-100X-674; PubMed PMID: 27196080; PubMed Central PMCID: PMC4907162.
- Liu JJ, Thompson LD, Janisiewicz AM, Shibuya TY, Keschnor DB, Garg R, et al. Ossifying fibroma of the maxilla and sinonasal tract: case series. *Allergy Rhinol (Providence)*. 2017 Mar;28(1):32-36. DOI: 10.2500/ar.2017.8.0190; PubMed PMID: 28381325; PubMed Central PMCID: PMC5380450.
- Kaur T, Dhawan A, Bhullar RS, Gupta S. Cemento-Ossifying Fibroma in Maxillofacial Region: A Series of 16 Cases. *J Maxillofac Oral Surg*. 2021 Jun;20(2):240-245. DOI: 10.1007/s12663-019-01304-y; PubMed PMID: 33927492; PubMed Central PMCID: PMC8041960.
- Han J, Hu L, Zhang C, Yang X, Tian Z, Wang Y, et al. Juvenile ossifying fibroma of the jaw: a retrospective study of 15 cases. *Int J Oral Maxillofac Surg*. 2016 Mar;45(3):368-376. DOI: 10.1016/j.ijom.2015.12.004; PubMed PMID: 26740351.
- Mintz S, Velez I. Central ossifying fibroma: an analysis of 20 cases and review of the literature. *Quintessence int*. 2007 Mar;38(3):221-7. PubMed PMID: 17333999.
- Marx RE, Stern D. Oral and maxillofacial pathology: a rationale for diagnosis and treatment. 2nd ed. Vol 1. Hanover Park: Quintessence Publishing Co., Inc.; 2012.
- Kennedy JL, Hubbard MA, Huyett P, Patrie JT, Borish L, Payne SC. Sino-nasal outcome test (SNOT-22): a predictor of postsurgical improvement in patients with chronic sinusitis. *Ann Allergy Asthma Immunol*. 2013 Oct;111(4):246-251. DOI: 10.1016/j.anai.2013.06.033; PubMed PMID: 24054358; PubMed Central PMCID: PMC3977600.
- Stewart MG, Witsell DL, Smith TL, Weaver EM, Yueh B, Hannley MT. Development and validation of the Nasal Obstruction Symptom Evaluation (NOSE) scale. *Otolaryngol Head Neck Surg*. 2004 Feb;130(2):157-163. DOI: 10.1016/j.otohns.2003.09.016; PubMed PMID: 14990910.
- Fokkens WJ, Lund VJ, Hopkins C, Hellings PW, Kern R, Reitsma S, et al. Executive summary of EPOS 2020 including integrated care pathways. *Rhinology*. 2020 Apr;58(2):82-111 DOI: 10.4193/Rhin20.601; PubMed PMID: 32226949.
- White SC, Pharoah MJ. Oral radiology: principles and interpretation. 6th ed. St. Louis: Mosby/Elsevier; 2009.
- Olifa BG, Nadia R, Safa N, Madiha M, Wafa A, Ines H, et al. Juvenile ossifying fibroma of the maxilla. *Egyptian Journal of Ear, Nose, Throat and Allied Sciences*. 2017 Jul;18(2):145-149. DOI: <https://doi.org/10.1016/j.ejenta.2016.09.002>.
- Appiani MC, Verillaud B, Bresson D, Sauvaget E, Blancal JP, Guichard JP, et al. Ossifying fibromas of the paranasal sinuses: diagnosis and management. *Acta Otorhinolaryngol Ital*. 2015 Oct;35(5):355. DOI: 10.14639/0392-100X-533; PubMed PMID: 26824919; PubMed Central PMCID: PMC4720932.
- Post G, Kountakis SE. Endoscopic resection of large sinonasal ossifying fibroma. *Am J Otolaryngol*. 2005 Jan-Feb;26(1):54-56. DOI: <https://doi.org/10.1016/j.amjoto.2004.06.009>.
- Kamal MS, Bahar MAA, Latif MA, Khan MMU. Juvenile ossifying fibroma of the maxilla: a case report. *Anwer Khan Modern Medical College Journal*. 2013;4(1):42-44. DOI: <https://doi.org/10.3329/akmmcj.v4i1.13685>.
- Hakeem AH, Hakeem IH. Juvenile Ossifying fibroma of paranasal sinuses— do we need to be radical in surgery?. *J Craniofac Surg*. 2013;24(3):e257-e258. DOI: 10.1097/SCS.0b013e318287d194; PubMed PMID: 23714983.
- Figueiredo LMG, de Oliveira TFL, Paraguassú GM, de Hollanda Valente RO, da Costa WRM, Sarmiento VA. Psammomatoid juvenile ossifying fibroma: case study and a review. *Oral Maxillofac Surg*. 2014 Mar;18(1):87-93. DOI: 10.1007/s10006-013-0400-y; PubMed PMID: 23435579.
- Titinchi F, Morkel J. Ossifying fibroma: analysis of treatment methods and recurrence patterns. *J Oral Maxillofac Surg*. 2016 Dec;74(12):2409-2419. DOI: 10.1016/j.joms.2016.05.018; PubMed PMID: 27311848.
- Aburas S, Bandura P, Al-Ibraheem A, Berger S, Meier M, Turhani D. A large maxillary cemento-ossifying fibroma superimposed with solitary bone cyst documented over 18 years: A case report. *Int J Surg Case Rep*. 2020;68:257-262. DOI: 10.1016/j.ijscr.2020.03.011; PubMed PMID: 32199251; PubMed Central PMCID: PMC7082601.