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Marlinda Adham

Oncology Division, Department of Otorhinolaryngology-Head, and Neck Surgery, Faculty of Medicine Universitas Indonesia, marlinda.adham@yahoo.com

Ferucha Moulanda

Oncology Division, Department of Otorhinolaryngology-Head, and Neck Surgery, Faculty of Medicine Universitas Indonesia, drferuchamoulanda@gmail.com

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Different Approaches of Suspected Unilateral and Bilateral Maxillary Sinus Myxoma, Two Rare Cases Report

Marlinda Adham,¹ Ferucha Moulanda

Oncology Division, Department of Otorhinolaryngology-Head, and Neck Surgery, Faculty of Medicine Universitas Indonesia.

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Marlinda Adham  <https://orcid.org/0000-0003-2461-9628>



Abstract

Maxillary sinus myxoma is a rare benign tumor with locally aggressive behavior and can infiltrate surrounding structures. Surgical intervention is considered the primary procedure with several approaches for wider tumor exposure, complete tumor removal, and better wound healing.

In this article, two cases of suspected unilateral and bilateral maxillary sinus myxoma were presented with different approaches applied for both cases. We managed two patients, a 37-year-old male with mass at right maxilla and a 60-year-old male with mass at both maxillae. Physical examination, radiology imaging, and pre-operative histopathology examination showed maxillary sinus myxoma. The total maxillectomy procedure with Weber Ferguson incision was performed for the 1st patient; meanwhile, the combined sub-labial degloving and left Weber Ferguson incision was performed for the 2nd patient.

Result: Clear and wide surgical fields were obtained in both cases with the approaches applied. Complete resection was performed, and follow-up showed no residue with an excellent clinical appearance.

Conclusion: Different approaches were planned based on the extent of the tumor, considering important structures nearby. Complete removal is the treatment of choice for proper healing, reconstruction procedures afterward, and prevention of recurrences.

Keywords: Maxillary sinonasal myxoma, sub-labial degloving, total maxillectomy, Weber-Ferguson incision.

Introduction

Incidence of sinonasal neoplasms is rare, with less than 10% of head and neck cancers, with an incidence of 0.5 to 1 per 100.000 people in the USA annually. Most sinonasal neoplasms rarely showed symptoms early. Because the paranasal sinuses are air-filled structures with significant potential space. A high index of sinonasal neoplasm suspicion should be maintained, especially in older patients with unilateral symptoms.¹

Sinonasal myxomas are histologically benign but locally aggressive tumors. Their origin is dental mesenchymal. Intraosseous myxoma has been found mainly in the jaws and occurred in the second or third decade of life.² The most frequent symptoms reported are swelling of the jaws, with only a few patients complaining of pain, paresthesia, and ulceration, 28% for pain and 56% for swelling. Clinical and radiological aspects of myxomas are mostly inconclusive. Histopathological examination of the lesion is mandatory to make an accurate diagnosis. Surgery is the mainstay treatment of myxoma. It has a high recurrence rate related to lack of encapsulated nature and leads to the infiltration process to the surrounding bony tissue.³ Incomplete removal can also be the reason for recurrence; thus, complete surgical treatment through bone resection is the most advocated modality. The patient must be followed-up closely for years.^{4,5}

This paper reports two cases of suspected myxoma that affected unilateral and bilateral maxilla and which surgical approach was used for management to ensure clear visualization and clean margin to prevent a recurrence.

Case illustration

A 37-year-old male presented with a mass at the right maxilla enlarging for eight months. On examination, a 4 x 5 cm² mass was noted in the right cheek and extended until the right hard palate. The mass was expanded to the right side of the nasal cavity causing nasal obstruction and leftward deviation of the nasal septum. There was no history of fevers, nosebleeds, or nasal discharge. No palpable lymph node. No history of eating and severe breathing impairment.

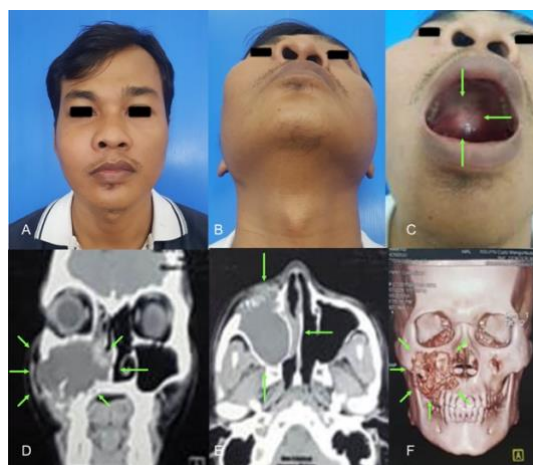


Figure 1. Patient with suspected right sinonasal myxoma extended to the right nose and palate. A. Anterior view, B. Inferior view, and C. Intraoral view. CT Scan showed a mass at the right maxilla extended to the right nasal cavity (figure D and E) and the anterolateral maxillary wall (figure F).

The mass's fine needle aspiration biopsy (FNAB) resulted in sinonasal myxoma. Computerized Tomography (CT) scan showed a mass at right maxilla extended to the right nasal cavity and ethmoid, destructed anterolateral right maxillary wall, maxillary bone, alveolar process until right hard palate.

Lateral rhinotomy with Weber Ferguson approach was performed. Anterior, lateral, medial, and superior walls of the destructed maxilla were resected. The interior wall of the maxilla was resected with part of the involved right hard palate, premolar, and molar teeth and removed en-bloc with a 5x4x4 cm³ mass. Intraoperative frozen section examination didn't perform since the mass margin was identified. Additional 0.5 cm margin excision was added, ensuring the free margin area. The posterior wall was thin with an eroded appearance and was excised. Reconstruction was performed with obturator and Mesh. Pathologic findings showed sinonasal myxoma with a clear margin without perineural and lymphovascular involvement. The patient recovered uneventfully postoperatively. No evidence of recurrence within six months of follow-up.



Figure 2. Weber Ferguson Incision approach to expose entire maxillary area (figure A), mass was identified and removed thoroughly with part of teeth and hard palate (figure B, C, D, and E). CT Scan after six months of surgery showed no recurrence (figure F) with good recovery (figure G). Obturator was placed on the palatal area (figure H).

A-60-year-old male with a mass at both maxillae that enlarged for three years. The symptoms are accompanied by nasal block in both nasal cavities. Mass was found with size 4x5 cm² and 5x6 cm² at right and left cheek respectively extended to the bilateral hard palate.

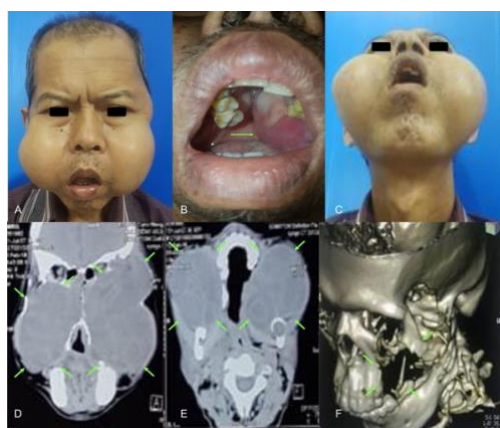


Figure 3. Patient with suspected bilateral sinonasal myxoma, extended to both nasal cavities, left hard palate, and left temporal area. A. Anterior view, B. Intraoral showing a mass enlarging, and C Inferior view. CT Scan showed a mass at both maxillary areas extended to paranasal sinuses, hard palate area, intracranial, left orbital, and left temporal (figure D and E), with the destruction of the left anterior maxillary wall (figure F).

No history of fevers, nosebleeds, or nasal discharge. He was still able to eat a soft diet. No visual impairment was reported. On examination, no palpable lymph node was found. CT scan showed an irregular solid mass at bilateral maxillary and extended to the sphenoid, posterior ethmoid sinuses, nasopharyngeal, para pharyngeal space, obliterated temporal, masticator, long capitis, levator veli palatine muscles, and hard palates with intracranial infiltration, left orbital and prominent to anterolateral reaching subcutaneous left temporal area. FNAB findings denoting maxillary sinus myxoma.



Figure 4. Sub labial incision was made for both bilateral areas (figure A and B). The Weber Ferguson incision was added for the left area to provide a wider surgical field until it reaches the orbital and temporal area (figure C). The mass could remove completely (figure D). Postoperative (figure E)

Discussion

Myxoma was known for penetrating the surrounding bone and vital structures. These could lead to radiologically unclear margins and increase the possibility of incomplete removal and recurrences.⁶ The treatment of choice for myxoma is mostly surgery through bone resection with challenging procedures to prevent recurrences and the chance of malignant transformation.^{3,4,7-9} Clinical and radiological aspects of myxomas are mostly inconclusive. For diagnostic purposes, it requires histopathological confirmation to exclude the differential diagnosis. Wide excision with margins around 5 to 10 mm is recommended in this condition.³

Previous studies proposed various approaches from conservative resections, segmental resection to aggressive surgeries, including wide local resection or en-bloc resection with enough margin and radical resection. Conservative resections such as enucleation or curettage offered minimum morbidity. However, it has a higher recurrence rate due to poorly defined tumor boundaries. Chrcanovic et al¹⁰ reported 31.3 % recurrences rate with curettage, 13.1% with enucleation and 1.3%-3.1% with resection. Dotta et al.¹¹ reported a 13.04% recurrence rate with resection approach and 25% with enucleation procedures. Zanetti et al.⁶ advocated the conservative treatment with a peripheral osteotomy to preserve the vital structures and maintain the oral function. Several surgeons proposed conservative surgery if the lesions were smaller than 3 cm, close to the vital structures, and young age.^{4,7,8,12} Extensive resection showed low rates of recurrence with a high morbidity rate. Recurrences commonly occur within two years after the resection ranging from 3 months until 15 years period.^{4,7,8,12} Due to the anatomical condition, recurrences after surgical resection is found slightly higher in the maxillary area than in the mandibular area. Recurrences can take place before the obvious appearance of clinical symptoms.¹³

As reported in these cases, more extensive tumors require a facial incision to provide adequate exposure, continued with aggressive surgeries. Anatomically, the maxillary area can be divided into suprastructure and infrastructure. This information provides input for

surgical approach consideration. Lateral Rhinotomy with the Weber Ferguson approach can be applied in both the suprastructure and infrastructure location of the tumor. The incision covers the medial canthus, lateral aspect of the nose, philtrum, upper lip, and gingivobuccal area. A surgeon can identify all maxillary walls and the rim of orbits and visualize the palate and oral cavity. Precise incision and trained surgeon result in superior aesthetic and functional outcomes.¹ For larger tumors with significant superior or lateral extension, modification with Lynch or sub ciliary extensions will be added. In the 1st and 2nd cases, lateral extension of the tumor became a point of consideration in performing the Weber Ferguson approach with subciliary extension. The incision takes a 90-degree turn laterally onto the infraorbital skin and follows the most prominent skin crease of the lower eyelid toward the zygomatic process. This extended version allowed the surgeon to perform en-bloc resection with an additional clear margin. Infrastructures dominant tumor can be achieved with sublabbial incision. As reported in the 2nd case, the right tumor mostly extends infero-laterally, thus treating with sub-labbial approach and continued with aggressive resection. This approach also has superiority in maintaining the aesthetic and function postoperatively.¹

Recurrences of maxillary myxoma have high possibilities since it can hide behind bony trabeculations, with results challenging to access. Wide en-bloc resection with clear margins is primarily suggested for extensive or recurrence of myxoma. However, this approach has become more accepted for smaller and primary tumors since inadequate surgical margins and poorly defined tumor boundaries are associated with recurrences.¹² Murphy et al.¹⁴ suggested a 1 cm bony margin to prevent recurrences. Meanwhile, Takahashi¹⁵ reported that 5.4 mm (3.4–7 mm) was adequate for a clear margin. The intraoperative frozen section didn't perform in both cases since clear borders were identified. The five-millimeter margin was applied in both cases, and the histopathology result came as a free margin. Surgical defect at the maxillary area was reconstructed using Mesh, and an obturator was placed at the resected palatal area. Reconstruction using a bone graft or buccal fat pad can be performed if the size of defects is <5 cm. Prosthetic reconstruction will be needed if the size of the defect is >5 cm.¹⁶ Reconstruction could be performed directly after surgical resection or delayed until a disease-free period is achieved. Reconstruction with autogenous rib graft or vascularized fibular free flap may be used.¹⁴ Adjuvant treatment for local control is not suggested, and myxoma is considered as radioresistant.²

The final histopathology result in the 2nd case showed schwannoma and neurofibroma. Since the pre-operative FNAB was reported as myxoma, a surgical approach was planned based on myxoma's aggressive nature and extension. Unspecific clinical symptoms and imaging of schwannoma and neurofibroma could be misleading with other sinonasal masses, including myxoma. The schwannoma is usually capsulated, but lesion found in the sinonasal frequently is not, as seen in myxoma. Its slow-growing pattern can cause surrounding bone remodeling. Less than 100 cases of nasal schwannoma (neurilemoma) have been reported since 1943, and its association with neurofibromatosis type 2 (NF2) has been reported in isolated cases. Neurofibroma is mainly found with lacks capsule macroscopically.¹⁷⁻²¹ Complete surgical resection is the mainstay treatment of choice for both functional and aesthetic outcomes. Neurilemmomas are known to be generally resistant to radiotherapy. A low recurrence rate was found in schwannoma, but recurrence and re-growth are frequently seen in plexiform neurofibromas. The potential of malignant transformation has been reported in schwannoma and around 2-3% of neurofibroma. Therefore, as applied in the maxillary sinus myxoma, long term follow-up should be performed both clinical (if possible with nasal endoscopy) and radiologically.¹⁷⁻²¹

A contraindication to surgical intervention for advanced tumors includes the presence of trismus, which is caused by invasion of the pterygoid

muscles and soft tissues in the masticator space around the temporomandibular joint pterygomaxillary fossa. Invasion of the skull base with bone destruction of the posterosuperior wall and lateral walls of the sphenoid sinus is also considered a contraindication to a satisfactory surgical resection. Different approaches were planned based on the location, tumor extension, histopathology, and important structures nearby. Complete removal is a mandatory procedure for proper healing and prevention of recurrences, followed by reconstruction procedures maintaining the function and aesthetic result.¹

Conclusions

Tumor extension, histopathology, surrounding area, and potential functional impairment should be necessary for choosing the best surgical approach. A clear visual field of surgery is mandatory to provide adequate resection with a clear margin to prevent recurrences; however, preservation of important structures nearby should also be considered for a better quality of life. Thus, the surgery approach needs to be planned carefully in maxillary sinus myxoma.

Disclosure

Authors declare no conflict of interest

References

1. Shah J. Nasal cavity and paranasal sinuses. In: Shah J, Patel S, Singh B, editors. Jatin Shah's Head and Neck Surgery and Oncology. 4th edition. Philadelphia: Elsevier; 2012:104-39.
2. Vasudev S, Kyvalya T, Balavikram K, Singh S. Odontogenic myxoma of the maxilla – A rare clinical sighting. *J Adv Clin Res Insights* 2020;7(2):30-3.
3. Hamama J, Khalif L, Fiqhi K, EL Kharim EK. Odontogenic Myxoma of Maxilla: Conservative or Radical Surgery? *Res Rep Oral Maxillofac Surg*.2018;2:1-5.
4. Prasannan L, Warren L, Herzog C, Lopez-Camarillo L, Frankel L, Goepfert H. Sinonasal Myxoma, a pediatric case. *J Pediatr Hematol Oncol* 2015; 27:90-2.
5. Kourda-Boutjemaa J, Farah-Klibi F, Rammeh S, Adouani A, Zermani R, Jlani-Baltagi S. Odontogenic myxoma: about four cases and revue of literature. *Ann de pathol*.2010;30:168-175.
6. Osman S, Hamouda GM, Eltohami YI. Clinical spectrum and treatment of odontogenic myxoma: analysis of 37 cases. *J Maxillofac Oral Surg*. doi: <https://doi.org/10.1007/s12663-020-01497-7>
7. Arul A, Verma S. Infiltrative odontogenic myxoma of the posterior maxilla: report of a case. *J natur sci biol med*. 2013;4: 484-487.
8. Koseki T, Kobayashi K, Hashimoto K, Ariji Y, Tsuchimochi M, Toyama M, et al. Computed tomography of odontogenic myxoma. *Dentomaxillofac Radiol* 2003; 32:160-5.
9. Kiresur M, Hemavathy S. An aggressive odontogenic myxoma of the maxilla. *Int J dentist*. 2014; 5:214-217.
10. Chrcanovic BR, Gomez RS. Odontogenic myxoma: an update analysis of 1692 cases reported in the literature. *Oral Dis*. 2019;25(3):676-83. doi: 10.1111/odi.12875.
11. Dotta JH, Miotto LN, Spin-Neto R, Ferrisse TM. Odontogenic myxoma: Systematic review and Bias Analysis. *Eur J Clin Invest*.2020;50:1-14.
12. Clarke J, Mandpe A, Weber P, Vogel H, Leng L. The unusual presentation of a myxoma within a sphenoid sinus: case report and literature review. *World Neurosurg*.2017;103:1-5.
13. Saalim M, Sansare K, Karjodkar FR, Farman AG, Goyal SN, Sharma SR. Recurrence rate of odontogenic myxoma after different treatments: a systematic review. *Br J oral maxillofac surg*.2019; DOI <https://doi.org/10.1016/j.bjoms.2019.09.005>

14. Murphy C, Hayes R, McDermott M, Kearns GJ. Odontogenic myxoma of the maxilla: surgical management and case report. *Ir J Med Sci.*2017;186:243-6.
15. Takahashi Y, Tanaka K, Hirai H, Harada H. Appropriate surgical margin for odontogenic myxoma: a review of 12 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol.* 2018;126(5):404-8. doi: 10.1016/j.oooo.2018.06.002
16. Leite-Lima F, Cunha JF, Lehman LF, Campos FE, Castro WH. Surgical management x recurrence of Odontogenic myxoma: Literature review and case reports. *J Oral Maxillofac Surg Med Pathol.* 2019;31:153-8.
17. Singh A, Haq M, Gautam P, Gautam D, Handa AC, Handa KK. The varied presentations of sinonasal regions schwannomas: Apropos of four cases. *Indian J Otolaryngol Head Neck Surg.* 2020. doi: 10.1007/s12070-020-01813-0.
18. Kumar S, Sayoo C. Sinonasal schwannoma: A rare sinonasal neoplasm. *Indian J Otolaryngol Head Neck Surg.* 2017;69:425-7.
19. Balaji SM. Surgical management of recurrent neurofibroma of infratemporal region: A case report with 20-year follow-up. *Indian J Dent Res.*2017;28:695-8.
20. Danesteh H, Sadeghzade A, Jafarpour D. Solitary central neurofibroma of the maxilla: A case report. *Braz Dent Sci* 2020;23(4). doi:10.14295/bds.2020.v23i4.2015.
21. Souza TSC, Amadeu NT, Fontana LC, Hamerschmidt R, Przysiezny PE. Nasal schwannoma: a challenging diagnosis of a rare neurofibromatosis type 2 manifestation. *J Head Neck Surg.* 2020;2:70-3.