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Unilateral maxillary swelling: a varied spectrum

Deepthi Satish, C. B. Pratibha*, A. M. Balasubramanya, Sumy Philip

Department of ENT, St Johns Medical College Hospital, Bangalore, Karnataka, India

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***Correspondence:**
Dr. C. B. Pratibha,
E-mail: cb.pratibha@gmail.com

ABSTRACT

Background: Diverse disease entities may present with mass in the maxilla and include benign conditions and malignant tumours. A detailed evaluation including preoperative imaging is essential to plan approach to management of isolated maxillary sinus lesions optimally.
Methods: We did a retrospective chart review of 14 patients with isolated maxillary swelling and involvement of maxillary sinus who presented to the Department of ENT in a tertiary care hospital over a period of 4 years. A detailed history and examination were done and investigations including contrast enhanced CT scan of the nose and paranasal sinuses was done in all patients.
Results: A total of 14 patients were included in this study. 7 were male and 7 were female. The mean age of the patients was 52.6, the age group of patients varied from 6 to 77 years. In our series we had patients with benign lesions like dentigerous cyst, fibro-osseous lesions, mucocele, radicular cyst, aspergilloma, and mucormycosis. Malignant cases included Adenoid cystic carcinoma, Low grade myofibroblastic sarcoma, Diffuse B cell lymphoma and undifferentiated carcinoma. All patients were treated surgically, they underwent a combined endoscopic and Caldwell Luc approach.
Conclusions: In patients with unilateral maxillary swelling, a systematic approach to diagnosis with preoperative contrast enhanced CT scan is essential with a high index of suspicion. Trans nasal endoscopic approach with sinus surgery is the preferred approach for these lesions, however, in cases of jaw cysts and odontogenic cysts extending laterally it can be combined with a Caldwel-Luc approach.

Keywords: Maxillary sinus, Caldwell–Luc, Endoscopic sinus surgery

INTRODUCTION

Unilateral maxillary swelling are rare and diverse disease entities may present with mass in the maxilla.[1] Being a large cavity the maxillary sinus lesions are initially asymptomatic and cause symptoms only when they have filled the sinus completely, cause pressure symptoms or spread into the surrounding areas. Many pathologies in this area share common symptoms but have unique features.[2] Our aim is to review patients presenting with unilateral maxillary swelling with involvement of maxillary sinus and to study the clinical, radiological and pathological correlation and appropriate management with review of literature.

METHODS

Study design: This is a retrospective chart review.

Patient selection

Patients included in this study presented to the Department of ENT in a tertiary care hospital (St Johns Medical College Hospital Bangalore, India) over a period

of 4 years (September 2014- May 2018). Fourteen patients with isolated maxillary swelling and involvement of maxillary sinus were included in this study.

Data collection

A detailed history and examination were done and investigations including contrast enhanced CT scan of the nose and paranasal sinuses was done in all patients.

Treatment

Biopsy was done by either endoscopic approach alone or combined with Caldwell-Luc approach. In malignant lesions patients were referred to oncology for further management. In cases of benign pathology like mucocele and fungal sinusitis patients were treated with endoscopic sinus surgery.

Statistics

The data collected was entered in to Microsoft excel and analysed. Descriptive data was presented in the form of frequency and percentage using pie chart and bar diagrams.

Ethical approval

Ethical approval was obtained from the Institutional Ethics Committee with study no 380/2018.

RESULTS

A total of 14 patients were included in this study. 7 were male and 7 were female. The mean age of the patients was 52.6, the age group of patients varied from 6 yrs to 77 yrs. All our patients presented with unilateral cheek swelling. Majority of them 21.4% had associated facial pain and headache also. Patients with malignant lesions had additional symptoms of loose tooth 14.2% and epistaxis 7.1%, remaining 7.1% presented with nasal obstruction (Figure 3).

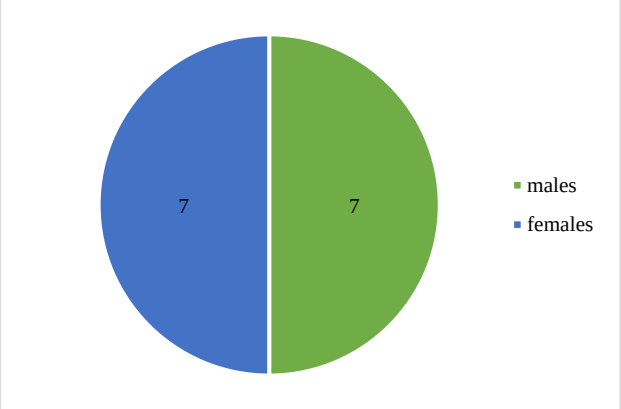

Figure 1: Pie chart depicting the sex distribution.

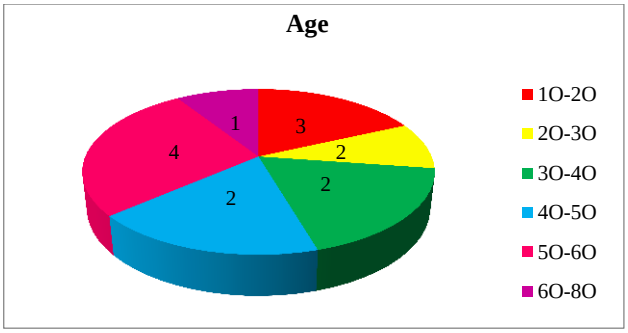

Figure 2: Pie chart depicting the age distribution of patients.

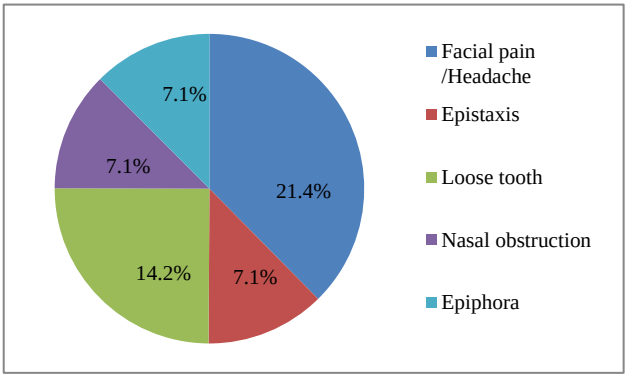

Figure 3: Pie chart showing associated symptoms with percentages.

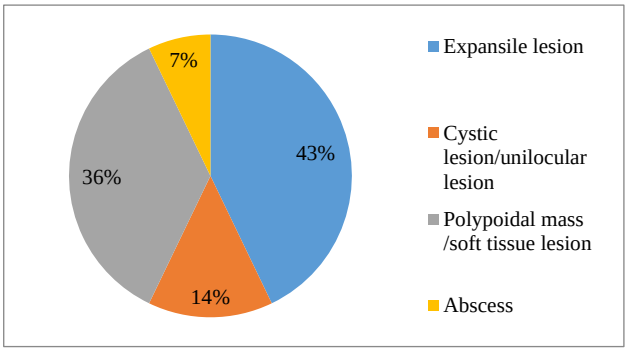

Figure 4: Pie chart showing various findings on CECT scan.

A contrast enhanced CT scan was done pre-operatively in all cases. The findings are given in (Figure 4). All patients were then taken up for biopsy under GA, endoscopic approach, Caldwell Luc approach or a combination of both. One patient presented with a left cheek swelling with CECT showing a Homogenous opacity involving the left maxilla with collection with Erosion of anterior wall of maxilla. In view of the abscess a sub labial incision was taken and abscess drained followed by endoscopic middle meatal antrostomy. Tissue sent for histopathology revealed an aspergilloma. Post-operatively patient was treated with antifungal medication and on follow up she improved symptomatically. In 3 patients, endoscopic approach was

done for excision and biopsy of the mass. In 8 patients, combined endoscopic and Caldwell-Luc approach was done and tissue sent for histopathology. Single procedure sufficed for patients with benign conditions namely cystic lesions, fungal sinusitis and mucocele. However, 2 patients with fibro-osseous lesions required multiple procedures to achieve diagnosis and complete excision.

Among the 14 patients, 3 patients were diagnosed with dentigerous cyst 2 patients with cemento-ossifying fibroma, 1 patient each with ossifying fibroma, mucocele, radicular cyst, aspergilloma, and mucormycosis involving the maxillary sinus on one side. The remaining 9 patients are on regular follow up and are asymptomatic.

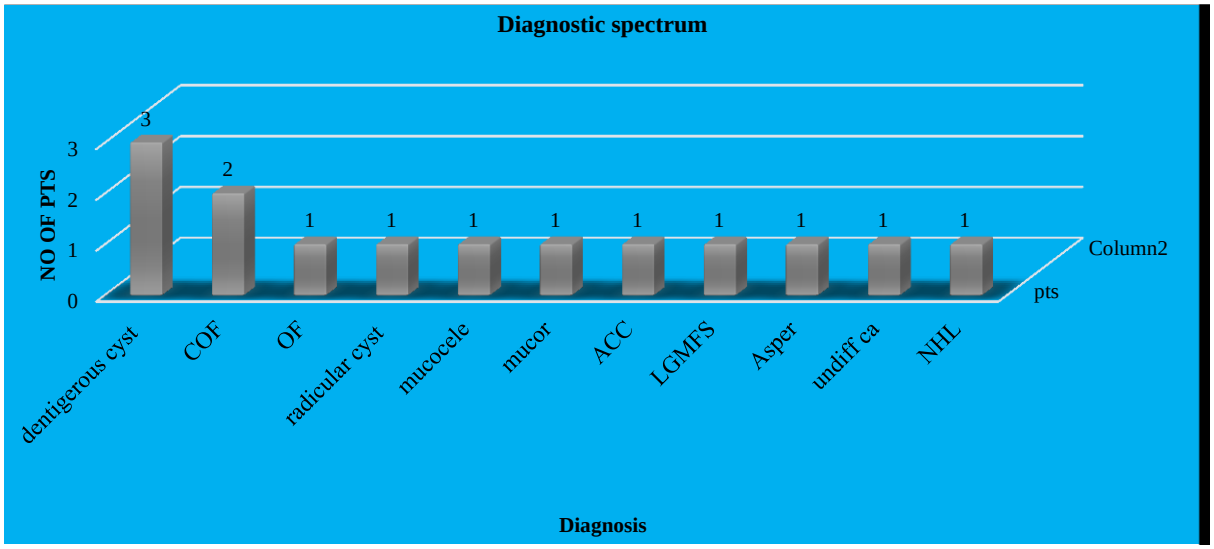

Figure 5: Graph showing the varied diagnostic spectrum.

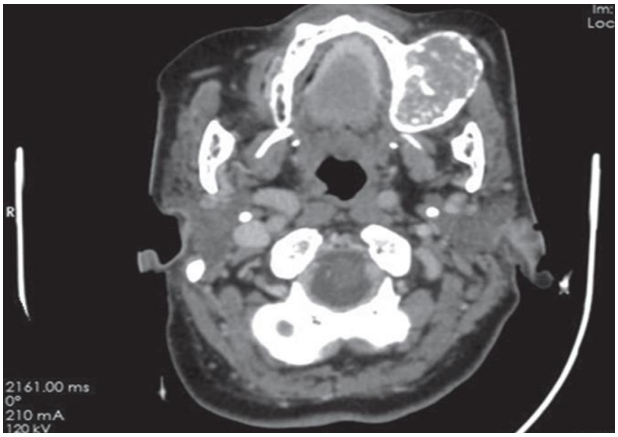

Figure 6: Axial CECT images of the nose and PNS of a patient with cemento-ossifying fibroma, showing a large expansile lesion arising from the left maxilla.

Four patients were diagnosed with malignant lesions which included Adenoid cystic carcinoma, Low grade myofibroblastic sarcoma, Diffuse B cell lymphoma and undifferentiated carcinoma (1 patient each). The patient with Low grade myofibroblastic sarcoma presented only with unilateral cheek swelling. The patient with adenoid cystic carcinoma presented with unilateral cheek swelling and epiphora. The patient with diffuse B cell lymphoma presented with unilateral cheek swelling and pain and patient with undifferentiated carcinoma presented with unilateral cheek swelling and loose tooth. CECT in these cases showed bone erosion of the maxilla and extension into the orbit in adenoid cystic carcinoma and extension into the infratemporal fossa and ptyregopalatine fossa in the patient with undifferentiated carcinoma. All patients underwent a combined endoscopic and Caldwell-Luc approach and tissue taken for biopsy. Histopathology revealed the diagnosis. Patients with adenoid cystic carcinoma and low grade myofibroblastic sarcoma underwent complete excision at an outside hospital and received post-operative radiotherapy. Patient with lymphoma was scheduled for chemotherapy and later lost to follow up. Patient with undifferentiated carcinoma underwent radiotherapy and is on regular follow up.

Figure 7: Contrast enhanced CT scan of a patient with radicular cyst showing a well-defined cystic lesion with a cortical rim arising from the left upper molar.

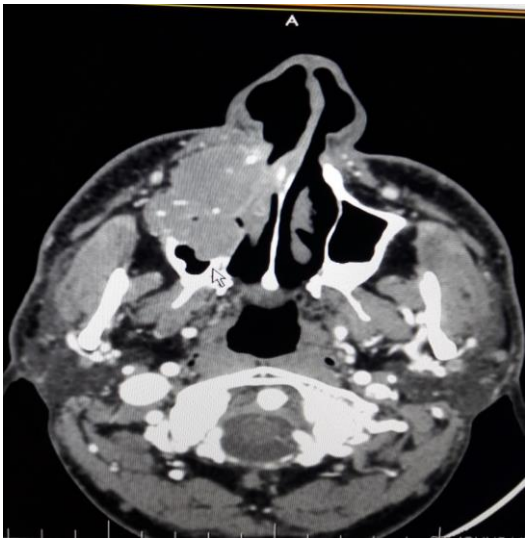

Figure 8: Contrast enhanced CT scan of a patient with fungal sinusitis showing a well-defined soft tissue lesion involving the right max sinus.

DISCUSSION

The maxillary sinus is typically pyramidal shaped with a volume of 5-20 ml, it consists of a medial wall, roof, and floor, anterior and posterior wall. A thorough understanding of the maxillary sinus anatomy is essential to plan approach to management of isolated maxillary sinus lesions optimally.[3]

Many of the maxillary sinus lesions are asymptomatic till they reach a larger size. Common symptoms at presentation are nasal obstruction, unilateral cheek swelling in benign lesions. Malignant lesions may present with epistaxis, proptosis and loose tooth.

Diverse disease entities may present with mass in the maxilla and include benign conditions and malignant tumours like squamous cell carcinoma, adenoid cystic carcinoma, and lymphoma. Benign lesions represent inflammatory pathologies. Neoplastic lesions can arise from the tissues that form the normal anatomy of the maxillary sinus like the mucosa, bone, salivary gland, muscle, mesenchyme and vessels most of them being rare tumours.[2]

Fibro-osseous lesions

In our case series of 14 patients, 3 (25%) presented with fibro-osseous lesions, 2 patients with cemento-ossifying fibroma (COF) and 1 patient with ossifying fibroma. Fibro-osseous lesions (FOL) are a poorly defined group of lesions affecting the jaws and craniofacial bones, they replace the bone by cellular fibrous tissue containing foci of mineralization. World Health Organization has classified fibro-osseous lesions and sub-divided these into ossifying fibroma and cementifying fibroma (CF) or cemento-ossifying fibroma (COF). Due to the presence of cementum-like material OF has been called as cemento-ossifying fibroma.[4] When this tumour arises in children, it has been named the juvenile cemento-ossifying fibroma which presents at an earlier age and is more aggressive.[5]

COF is a bony tumour of the maxilla arising from the periodontal ligament it is composed of varying amount of cementum, bone, and fibrous tissue. This neoplasm occurs in patients of wide age range, with greatest number of cases between third and fourth decade of life. There is a female predilection. Maxillary lesions are large at the time of presentation. The differential diagnosis for COF includes chondrosarcoma or osteosarcoma, fibrous dysplasia, odontogenic cysts, squamous cell carcinomas, calcifying odontogenic cysts (Gorlin cysts), and calcifying epithelial odontogenic tumours (Pindborg tumours). COF can be differentiated from sarcomas and carcinomas by its well-defined border. Fibrous dysplasia has a characteristic “ground glass” appearance not seen in cemento-ossifying fibroma. Radiologically COF may appear as unilocular or multilocular radiolucent lesion. The margin of the lesion is relatively well defined and shows the presence of sclerotic rim. The recommended treatment for COF is complete excision of the tumour. COF of the mandible are common, in the maxilla it is rare and only few cases have been reported in the literature.[3] in our series the 77-year-old female had COF and the 55-year-old female had ossifying fibroma. Both were treated by combined endoscopic and Caldwell-Luc approach and are on follow up with no recurrence. However, the Juvenile ossifying fibroma is aggressive with a high tendency to recur. The third case an 18-year-old female underwent endoscopic excision, histopathology revealed COF, she presented with a recurrence 6 months later and was planned for an endoscopic medial maxillectomy but was lost to follow up.

Low grade myofibroblastic sarcoma

Myofibroblasts are believed to originate from proximal resident mesenchymal cells, in particular fibroblasts. In adults, myofibroblasts are known to exist in the periodontal ligaments. These cells are mesenchymal spindle- shaped cells that share ultrastructural features with both fibroblasts and smooth muscle cells.[6]

Myofibrosarcoma is a malignant tumour of the myofibroblasts. Myofibroblastic sarcomas can be classified into low, intermediate and high-grade entities. Low-grade myofibroblastic sarcoma (LGMS) has a predilection for the head and neck, especially in the tongue and oral cavity, with a high tendency to local recurrences and metastases. LGMS arising in the maxillary sinus are extremely uncommon.[7] Patients usually complain of a slow growing painless swelling. Our patient presented with a painless swelling over the right cheek. His contrast enhanced CT scan showed a Soft tissue lesion involving left maxilla with extension into left Infratemporal fossa and pterygomaxillary fissure,

he underwent surgery at an outside hospital and is now is lost to follow up with us.

The treatment for LGMS is a wide local resection with tumour-free margins. LGMS is known to present with recurrences and metastasis even after a prolonged period of time so post-op chemotherapy and radiotherapy is advised.[6]

Non-Hodgkin's lymphoma of maxillary sinus

Non-Hodgkin's Lymphoma (NHL) are a group of neoplasms that originate from the cells of the lymphoreticular system. 40% of Non-Hodgkin's lymphoma arises from extra nodal sites like the stomach, liver, soft tissue, dura, bone, intestine and bone marrow. The nasal cavities and paranasal sinuses are rarely affected by primary NHL.[8] It more commonly affects the middle aged and the elderly with slight male predilection, in our case it was a 29-year-old healthy male. Clinical features of NHL of the maxilla may be nonspecific, or may present as local bony swelling, loose tooth, ulcerations of the mucosa, and rarely facial or dental pain. There is an increased risk of NHL in HIV positive patients so it is important to stress on early diagnosis to ensure appropriate treatment, to improve prognosis, and quality of life. Differential diagnosis includes fungal infections, dento-alveolar abscess, Wegener's granulomas, midline lethal granulomas, squamous cell carcinomas and metastatic tumour.[9]

Fungal infections of the maxillary sinus

Fungal infections of the nose and paranasal sinuses can be categorized into invasive and non-invasive forms. The clinical presentation and course of the disease is primarily determined by the immune status of the host and can range from harmless to life threatening complications.[10] Aspergillosis is the commonest fungal infection of the nose and paranasal sinuses. Of these, *Aspergillus fumigatus (*90%) is the most common fungal pathogen followed by *Aspergillus niger* and *Aspergillus flavus*. Mucormycosis, Candidiasis may also occur. Maxillary sinuses are commonly involved in fungal infections. Endoscopic sinus surgery followed by antifungal therapy is the mainstay of treatment of fungal sinusitis[11].

In our series of cases 2 patients had fungal infections. One patient presented with a pre-maxillary tense swelling, showing an abscess on CECT scan with erosion of anterior wall of maxillary sinus. Hence, a sub labial approach was done to drain the abscess. The biopsy was reported as *Aspergillus*. The patient was started on itraconazole for 3 weeks and on follow up did not show any recurrence. The second patient was taken up for endoscopic biopsy of maxillary lesion. Intraoperatively whitish necrotic material was seen in the maxillary sinus, middle meatal antrostomy and debridement was done. Histopathology revealed mucormycosis and hence post operatively treated with IV amphotericin (target dose 2 grams was given).

How do we approach unilateral maxillary swelling?

Imaging would be the first step in evaluation of unilateral maxillary swelling. Contrast enhanced CT scan is the gold standard imaging method. Benign tumours cause remodelling and thickening of adjacent bone, while malignant tumours cause bone erosion. Key areas to carefully evaluate on CT scan are the bony orbital walls, posterior wall of the maxillary sinus, pterygopalatine fossa, anterolateral wall, medial wall and dentition. MR imaging is complementary to CT in order to characterize the soft tissue components of the tumour and to evaluate the extent of tumour invasion beyond the bony sinus walls particularly orbital involvement.[12]

Histopathology is critical in patients with suspected neoplasms, atypical infections, or inflammatory disorders. Indications for biopsy would include atypical symptoms, abnormal findings on endoscopy (idiopathic nasal septal perforation, unexplained crusting, tissue loss), or on imaging(bone erosion or destructive lesions) new onset sinonasal symptoms at an advanced age, unilateral sinonasal process, and clinical findings suggestive of orbital, cranial nerve, intracranial, facial soft tissue, or maxilla involvement.[13] It is preferable to take a biopsy under general anaesthesia to obtain an adequate sample for diagnosis and to obtain haemostasis as a number of these tumours can bleed torrentially. An endoscopic biopsy will be the preferred approach. In the current case series an endoscopic biopsy was done in 3 out of 4 lesions with suspected malignancy. A Caldwell-Luc approach should be avoided in a suspected sinonasal malignancy as this could possibly seed the cheek flap with tumour and complicate a subsequent resection.[14]

Operating room consultations provide rapid gross and microscopic diagnosis for intra or perioperative patient management. It helps to confirm the presence of representative tissue for histopathology. It helps to ascertain malignancy when present and also to optimally process tissue for special studies to be used for further diagnosis.[15]

In our series intraoperative tissue sample were sent for frozen section in 2 patients. Of this one patient presented with a cheek abscess with a suspected mass arising from the middle meatus. The frozen section showed *Aspergillus* infection. Debridement of the lesion was then done in the same sitting. The other patient presented with a maxillary mass with erosion of the posterior lateral wall of the maxilla and extension into the infratemporal fossa and the frozen section revealed a malignant lesion. Further procedure was hence deferred. Final histopathology revealed myofibroblastic sarcoma. Complete excision followed by post-operative radiotherapy was done at an outside centre.

Endoscopic sinus surgery has become widely accepted as the standard of treatment for lesions of the paranasal sinuses. With increased skill with endoscopic surgical technique, advanced technologies such as intraoperative imaging systems, and a better understanding of the complex anatomy of the paranasal sinuses and surrounding vital structures, many otolaryngologists have increasingly applied their expertise in endoscopic sinus surgery to the resection of nasal and sinus neoplasms.[16] with the advent of angled endoscopes 30, 45 and 70-degree complete removal of the maxillary sinus lesions is possible. The endoscopic approach can be combined with other approaches like the Caldwell-Luc approach for extensive, recurrent lesions involving the floor and anterior wall of maxilla.

Caldwell-Luc surgery (CWS) is almost 120 years old now and is still done for certain indications. This approach intends to replace the diseased and scarred mucosa from maxillary sinus. In literature the indications for CWS at present would include dental conditions like dental and dentigerous cyst and for drainage of abscesses.[17] this approach is also used to repair blow out fracture of floor of orbit also as roof of maxillary antrum forms the floor of orbit in our study, we found CWS useful for odontogenic cysts and fibro-osseous lesions. Thus, CWS is a versatile and safe surgery and it still can be used for limited cases.

For extensive lesions involving the maxillary sinus, the external approaches such as midfacial degloving, external or intranasal Denker approach and medial maxillectomy are still applied in clinical treatment. However, with external approaches there is increased incidence of postoperative complications such as dry nose, epiphora, dacrocystitis, mucocele, facial neuralgia and external scarring.[18]

Thus, endoscopic sinus surgery/FESS has almost completely replaced the radical Caldwell-Luc approach and external approaches. With the help of angled endoscopes especially the 70-degree endoscope it is possible to see the complete maxillary sinus.

CONCLUSION

In our case series we had a varied spectrum of maxillary sinus lesions ranging from inflammatory, fungal to benign and malignant pathologies. A systematic approach to diagnosis with a high index of suspicion and an awareness of uncommon lesions is essential. Contrast enhanced CT scan is essential to look for tumour extent and bony involvement and also to plan the preferred surgical approach. Intra-operative cytology and frozen section examination of lesions of nose and paranasal sinuses are useful, quick, and reliable diagnostic technique for rapid and early diagnosis in the operation theatre and can be used as an adjunct to histopathology for better management of patients. Endoscopic sinus surgery is the preferred approach for these lesions. A combined endoscopic and Caldwell-Luc approach may be done in cases of jaw cysts and odontogenic cysts.

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Ethical approval: The study was approved by the Institutional Ethics Committee

REFERENCES

1. Patel H, Lall A. Unilateral Maxillary Swelling: A Retrospective Study. Indian J Otolaryngol Head Neck Surg. 2010;62(4):403–7.
2. Saleh H, Lund VJ. Benign maxillary Sinus Masses. The Maxillary Sinus. Thieme Publication. 2011;1:87-103.
3. Becker AM, Hwang PH. Surgical anatomy and embryology of the maxillary sinus and surrounding structures. The Maxillary sinus. Thieme Publication. 2011;1:1-7.
4. Prabhu S, Sharanya S, Naik PM, Reddy A, Patil V, Pandey S, et al. Fibro-osseous lesions of the oral and maxillo-facial region: Retrospective analysis for 20 years:J Oral Maxillofac Pathol. 2013;17(1):36–40.
5. Verma P, Rathore PK, Mrig S. Cemento-Ossifying Fibroma of the Maxilla:A Case Report. Indian J Otolaryngol Head Neck Surg. 2011;63(1):38–40.
6. Qiu JY, Liu P, Shi C, Han B. Low-grade myofibroblastic sarcomas of the maxilla. Oncol Lett. 2015;9(2):619–25.
7. Gómez-Oliveira G, Arribas-García I, Alvarez-Buylla AS, Burgos R, Martinez-Perez F, Alvarez-Flores M. Low-grade myofibroblastic sarcoma. Two rare tumors in two rare locations. Revesociroralmaxilofac .2015;37(2):108-12.
8. Adwani DG, Arora RS, Bhattacharya A, Bhagat B. Non-Hodgkin's lymphoma of maxillary sinus: An unusual presentation. Ann Maxillofac Surg. 2013;3(1):95–7.
9. Agrawal MG, Agrawal SM, Kambalimath DH. Non-Hodgkins lymphoma of maxilla:A rare entity. Natl J Maxillofac Surg. 2011;2(2):210–3.
10. Gavito-Higuera J, Mullins CB, Ramos-Duran L, Sandoval H, Akle N, Figueroa R. Sinonasal Fungal Infections and Complications: A Pictorial Review. J Clin Imaging Sci. 2016;6:23.
11. Karthikeyan P, Coumare VN. Incidence and Presentation of Fungal Sinusitis in Patient Diagnosed with Chronic Rhinosinusitis. Indian J Otolaryngol Head Neck Surg. 2010;62(4):381–5.
12. Eggesbo HB. Imaging of sinonasal tumours. Cancer Imaging. 2012:136–52.
13. Tabaee A, Hsu AK, Kacker A. Indications, technique, safety, and accuracy of office-based nasal endoscopy with biopsy for sinonasal neoplasm. Int Forum Allergy Rhinol. 2011;1:225–8.
14. McMonagle BA, Gleeson M. Nasal cavity and paranasal sinus malignancy. In: Scott-Brown's Otorhinolaryngology, Head and Neck Surgery. 7th

ed. Great Britian, UK: Edward Arnold; 2008: 2417-2435.
15. Nigam JS, Misra V, Dhingra V, S Jain, K Varma, and A Singh. Comparative study of intra-operative cytology, frozen sections, and histology of tumor and tumor-like lesions of nose and paranasal sinuses. J Cytol. 2013;30(1):13–7.
16. Banhiran W, Casiano RR. Endoscopic sinus surgery for benign and malignant nasal and sinus neoplasm. Curr Opin Otolaryngol Head Neck Surg. 2005;13(1):50-4.
17. Datta RK, Viswanatha B, Harsha MS. Caldwel luc surgery;Revisited. Indian J Otolaryngol Head Neck Surg. 2016;68(1):90–3.
18. Bing Z, Han DM, Cui SJ, Huang Q, Wang CS. Intranasal endoscopic prelacrimal recess approach to maxillary Sinus. Chin Med J. 2013;126(7):1276-80.

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