Unilateral Maxillary Swelling: A Retrospective Study

Hetal Patel · Abhineet Lall

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Abstract Unilateral maxillary swelling is a rare condition, however we being in tertiary care centre did see a large number of these cases. All patients presenting with unilateral maxillary seen during the last 3 years were included in this retrospective study and evaluated concerning histories, diagnostic and therapeutic approaches and achieved outcomes. A total of 31 patients were seen during this period. 27 of these were benign lesions and were subdivided as follows: odontogenic cystic lesions (11/27), fibro-osseous lesions (8/27), secondary to extensive allergic fungal sinusitis (5/27) and mucocoele (2/27). The remaining five were malignant lesions, two each were squamous cell cancer and adenocarcinoma of the maxillary sinus, and one was rhabdomyosarcoma. All patients received treatment with due merit of the underlying etiology. Understanding and recognizing key diagnostic features helps in appropriate management and reducing morbidity.

Keywords Maxilla · Maxillary sinus

Introduction

Unilateral maxillary swelling are not only rare but a variety of conditions cause these lesions. Our aim in the study has been to establish a clinical, radiological and pathological correlation, review literature and lay down guidelines on appropriate management of these lesions as a whole.

The aim of this publication is to deliver up to date information on modern diagnostic and therapeutic options for the management of these lesions on the basis of a retrospective study from a large, tertiary otorhinolaryngological referral centre.

Materials and Methods

The design of the study is a retrospective observation with the observation period from December 2005 to December 2008. In a review of medical and surgical documentation of our tertiary otorhinolaryngological referral centre, all patients with unilateral maxillary swelling were included into the observation. Exclusion criteria were soft tissue swelling, primarily arising in the buccal space appearing as a maxillary swelling. In addition total of patients who were treated for any condition involving the maxillary sinus were evaluated.

The inpatient notes of the patient identified in search of the primary measure were collected and each patient's data set thoroughly evaluated for history of presenting complaints, previous surgery, and diagnostic workup including imaging studies and result of the histopathological examination, therapeutic measures (surgical approach and procedures) as well as postoperative course. Exemplary images from different imaging studies and intraoperative sites were gathered as well.

Results

A total of 31 patients were included in the retrospective observation. The patient had a mean age of 47.2 years (range 11–74 years), with two patients under 15 years and

H. Patel · A. Lall (✉)
Department of ENT and Head Neck Surgery, Seth G S Medical
College and KEM Hospital, Parel, Mumbai, India
e-mail: abhineetlall@hotmail.com

two patients above the age of 60 years. The observed group of patients consisted of 19 males and 12 females. Four of the 27 benign lesions had history of previous surgery. Two of them were allergic fungal sinusitis. The other two were fibro osseous lesions who had been operative at a younger age (8 years and 10 years of age). Both these fibro osseous lesions were operated externally and significantly enough at a time when their only complaint was cosmetic deformity.

The diagnosis of the 31 patients included in the observation were odontogenic cyst in 11 cases, eight cases of fibro osseous lesions (including six of fibrous dysplasia and two cases of cement ossifying fibroma), five cases secondary to extensive polyposis in allergic fungal sinusitis, two cases each of maxillary mucocele, squamous cell carcinoma and adenocarcinoma, one case of rhabdomyosarcoma. All patients had complaints of nasal obstruction. Seven patients of all had some amount of diplopia with one patient having decreased vision. Three patients had a history of epistaxis of which two were malignancy and one was an infected polypoid. The radiological diagnostic workup of the patient included both computed tomography (CT) and magnetic resonance imaging (MRI) in nine of the 31 patients. Additionally MR angiogram was done in three cases of fibro osseous lesions to have a better idea about the vascularity. A diagnostic biopsy was done for all the malignancy. Apart from these biopsy was also done for two cases of fibro osseous lesion and one case of odontogenic cyst for ambiguous presentation on imaging. Care was taken that the biopsy was a representative tissue for which an incision with maxillary sinus ostium widening done before taking a biopsy from the maxillary sinus.

All patients received treatment by endoscopic approach. Malignancy had a different protocol and will be elaborated later. In all 28 cases were managed endoscopically. The use of powered instruments cannot be understated, with the shaver and the endodril being used extensively. The surgical steps were same as classically described in endoscopic sinus surgery. A satisfactory clearance was achieved in all the 26 benign conditions with the exception of two fibro osseous lesions. Both the two cases have been following up regularly and have not shown any signs of recurrence. Intraoperative blood loss were kept at the minimum with only two patients requiring intraoperative blood transfusion. Merocel pack was used in all the patients and removed on the third postoperative day.

Following surgical procedure all 28 patients treated endoscopically were treated according to the following regimen: Pack removed on third postoperative day, antibiotics were continued till the 10th postoperative day. The patient was simultaneously started on alkaline nasal douch, saline nasal sprays and liquid paraffin nasal drops. The patient was called on the 10th postoperative day and nasal crusts were removed as an OPD procedure. An endoscopic

clearance of the crusts was not done on regular basis. The patient was then asked to follow up for the first 4–8 weeks. Follow up imaging was not done routinely.

We lost five patients in the follow up and both the patients of squamous cell carcinoma died one due to pulmonary metastasis and the other due to unrelated cause. Of these 24 patients, three required further minor surgery (division of endonasal synechia under local anesthesia, another two were taking long term topical steroids and one further patient was suffering from ongoing nasal symptoms (hyposmia, nasal obstruction).

Odontogenic Cyst

We saw 11 patients with odontogenic cyst, with by far the largest number were of dentigerous cyst. All the patients were young adults in their 20s and 30s. They all presented with smooth, gradual onset, painless maxillary swelling. None of the patient had any significant dental history. Oral examination revealed hard palatal bulge. All our patients had absent molar tooth when specially looked for. A CT scan gave significant clue towards its diagnosis. Eight patients had the CT scan showing large unilocular cystic lesion involving the maxilla with the presence a tooth within the cyst pointing towards the diagnosis. We did have one case where radiologically it appeared to be odontogenic keratocyst, where it was multilocular; however presence of a tooth within the cyst on the scan and biopsy confirmed the diagnosis. All cases were managed endoscopically where the aim was Marsupialization of the cyst.

Fibro Osseous Lesions

Benign fibro osseous lesions are interesting group of disease all of which are easily diagnosed radiologically. Six of our patients were fibrous dysplasia and the two were cement ossifying fibroma. All patients had a history similar to that of odontogenic cyst with gradual onset slow progressing painless swelling. Two patients were recurrence and had been operated at a younger age group (8 years and 10 years). In four of the patients the eyeball was pushed upward and outward causing severe diplopia. Diplopia was more in the inferolateral quadrant. One of the patient who presented to us with recurrence had severe restriction of movement of eyeball and had complaints of decreased vision. Epiphora although present in four, the patients were not forthcoming with the complaint. Imaging for fibrous dysplasia is very classical. The CT shows a ground glass appearance but essentially depends on amount of the calcification and ossification. All our patients were above the age of 20 years except one who was an 11 year old child with cosmesis as the only problem. We decided to manage the child conservatively and will be operated upon at a later

age. Two patients had the disease limited to the maxilla, the others had minor extension of the disease into the ethmoid and the recurrences had extensive disease. The extensive disease was that of a 23 year old female who had been operated upon in the past and disease extended to all the sinuses. This was the case in which we did an MR angiogram to have a better idea about the vascularity of the disease. All patients were managed endoscopically using powered instruments (endodrill). We achieved a satisfactory level of clearance in all the patients save two in which lesion extended on to the optic nerve. We lost three patients in the following including the one where we had left some diseased tissue. Among those who have followed, none have shown any recurrence.

Maxillary Mucocele

We had two patients with maxillary mucocele, an extremely rare site for mucocele formation. The patients had the smooth painless maxillary swelling with complain of nasal obstruction. CT scan showed a smooth expansile low attenuation lesion arising in the maxilla. Both the patients were managed endoscopically with the aim of complete marsupialization of the cyst. Both patients were discharged on the second post operative day and have remained asymptomatic till date.

Allergic Fungal Sinusitis

Allergic fungal sinusitis (AFS) is common where the fungal elements act as inciting factors for polyposis. We had five patients with extensive polyposis secondary to AFS. Apart from maxillary swelling three patients had proptosis causing diplopia. All five patients had complains of recurrent rhinitis with sneezing bouts. CT showed a pan-polyposis picture in all the patients. The patients were initially started on a course of oral steroids in tapering dose over a period of 20 days along with steroid nasal sprays. All the patients showed dramatic symptomatic improvement with the use of steroids. It also improved our operative field thereby improving the results of the surgery. Hence we can say that use of oral steroids preop has improves long term results in polyposis. Patients were continued on nasal sprays for till about 6 months postoperative and an endonasal examination was done as and when required. Two patients developed syncheia and they were released under local anesthesia.

Adenocarcinoma

Both patients of adenocarcinoma were females in their 40s and presented with maxillary swelling, occasional epistaxis and epiphora. Here the patient had a diffuse pain limited

over the maxillary region. CT showed mass present in the maxilla. We did an MRI to see for the soft tissue spread and to our surprise both of them had a much limited extend than what was being appreciated on the CT scan. Having a high degree of suspicion of malignancy we took a biopsy confirmed the histopathology and then went in for an endoscopic clearance. Both the patients are doing well without any evidence of recurrence.

Squamous Cell Carcinoma

We had two patients with squamous cell carcinoma causing irregular bulge of the maxillary sinus. Both patients were males and more than 70 years of age. They had history of epistaxis a, epiphora and diplopia. CT showed presence of bony destruction and an MR showed soft tissue extend with orbital extension. A multimodality treatment was planned along with radiation and medical oncologist. Patients were subjected to a radical surgery (a maxillectomy with orbital exenteration) and put on chemo radiation. Sadly we lost the patients, one due to pulmonary metastasis and the other due to unrelated cause. (Figs. 1, 2, 3).

Discussion

Odontogenic cyst has been the most common pathology in our series. The cyst lesions of the jaw have been classified into epithelial and non epithelial. Non epithelial are aneursymal bone cyst and solitary bone cyst. The epithelial variant has been divided into inflammatory and developmental. The last has been divided into odontogenic cyst

Fig. 1 Maxillary mucocele

Fig. 2 Clinical image

Fig. 3 Fibro osseous lesion

and nonodontogenic variants. Dentigerous cyst are a variety of odontogenic cyst and are most common in the mandible in the second and third decade [1]. Apart from dentigerous cyst other odontogenic cyst are gingival cyst, botryoid cyst, eruption cyst and odontogenic keratocyst. The odontogenic tumor is ameloblastoma. The nonodontogenic cyst are nasopalatine duct cyst, nasolabial cyst, midpalatal cyst of infant and fissural cyst. Whereas the nonodontogenic cyst may present soon after birth, the odontogenic variant may present at a much later in childhood.

There have been a number of terminology being used for fibro osseous lesions. They have been named as localized osteitis, osteofibroma, ossifying fibroma, fibrous osteoma or osteiod osteoma [2]. The confusion is compounded upon by the by the fact that within a histological specimen of say ossifying fibroma there may be areas of tissue which have

features of fibrous dysplasia [3]. Nomenclature will continue to confuse the issue until a tissue cell marker is found which can accurately differentiate the conditions on a histopathological basis. CT showing ground glass appearance are largely diagnostic. MR has variable presentation. Jee et al [4] in their study showed that fibrous dysplasia gives homogeneous hypo intensity on T1 weighted images throughout the lesion. On T2 weighted images MR images showed varying signal intensity, 62% in their study showed homogeneous hyper intensity and 38% showed relative homogeneous hyper intensity. These small areas of hyper intensity are believed to be areas of hemorrhage and cystic degeneration [5]. It is largely believed that older the lesion becomes, greater the tendency to increase in density due to formation of small calcified areas. Only when there is considerable calcification and ossification, does it give its trade mark characteristic of ground glass appearance. Ossifying fibroma are similar to FD, except that their radiological feature may vary from radiolucent to radiopaque [5], the CT exhibits larger non-ossified amount than FD and behave more aggressively [5]. Partially excised and curettaged lesions recur in 25% of cases [3]. We had two recurrence cases.

Mucocele of maxillary sinus are fairly rare. The reported incidence in adults vary from 3 to 10% [6]. A higher prevalence have been reported in Japanese population and have ascribed to the fact that more Caldwell Luc procedure are performed there [7, 8]. Our patient was a 14 year old male with no previous history of surgery. Mucocele have been described in association with neoplasia, trauma, surgery, inflammatory process (e.g. cystic fibrosis) and congenital abnormalities [9, 10]. Hence our case qualifies to be an idiopathic maxillary mucocele.

Allergic fungal sinusitis is by far the most common cause for allergic fungal sinusitis (AFS) [11]. Maxillary swelling in AFS is due to bony remodeling as a result of long standing disease. They respond very well to steroids. A CT scan with contrast study are diagnostic for AFS which shows increased attenuation and mucosal opacification on unenhanced images [11]. Aspergillosis are the main species as was seen in our series too. The swelling resolves over a period of time.

Malignant neoplasm of the paranasal sinuses and nose are rare, only 3% of all head neck malignancy [12]. These patients have classical clinical signs like epiphora, paresthesia and swelling, all of which was seen in our patients. Swelling is due to bony destruction. Additional symptoms which were seen in our series were epistaxis, nasal obstruction or discharge and diplopia. A CT scan is mandatory in these cases as bony destruction may not be evident on panoramic radiography. All such suspected malignancy should be confirmed with histopathology as their treatment changes on the histology.

Conclusion

Although there are variety of causes for unilateral maxillary swelling, a good history with clinical examination narrows down the differential diagnosis, and an equally good imaging helps to bring the possibilities further down. However, occasionally one may need to take a biopsy to (a) diagnose the lesions, (b) in malignancy to prove it histopathologically.

Although our series includes most of the conditions seen commonly, however it is not an exhaustive list and one needs to be vary of conditions like giant cell granuloma [13] and solitary plasmacytoma [14] which have been reported.

Key message With better preoperative investigation modalities, one needs to make a judicious discussion on management of such lesions.

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