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Diagnostic Algorithm and Management Protocol with Special Consideration to Chronic Granulomatous Lesions Involving the Maxillary Sinus

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ABSTRACT

Tumor growth of the maxilla and maxillary sinus, causing eye proptosis along with a diffuse swelling over the malar region of the face mostly point towards a diagnosis of maxillary sinus neoplasm and /or granulomatous lesion. This provisional diagnosis is further confirmed with the help of radiological and histopathological evaluation. Maxillary sinus is a unique part of the maxillofacial region as it is in close proximity to the odontogenic apparatus, the nasal cavity which are a very potent source of pathologies. Owing to the hollow nature of the sinus any neoplastic and non neoplastic growth can easily grow in to the sinus from the oral and/ or nasal cavity. Also the maxillary sinus is lined by the pseudo stratified ciliated squamous epithelium which is very much pleuripotent in nature, can easily give rise to pathologies arising from the maxillary sinus itself. This uniqueness of maxillary sinus makes it important to focus on following a particular algorithm for diagnosis and management. This article focuses on the development of algorithm to be followed for maxillary sinus pathologies, with a special consideration on chronic granulomatous lesions producing tumor like growth in the maxillary sinus.

Keywords: Maxillary Sinus, Chronic Granulomatous Lesions, Eye Proptosis, Nasal Obstruction, Growth on Maxilla

BACKGROUND

The maxillary sinus (MS) among other sinuses is unusual among the other sinuses because of its well established vascular relationship with the odontogenic apparatus and nasal cavity.[1] Other unique feature of MS is its epithelial lining which is a ciliated columnar epithelium helps in production and movement of mucous often almost against gravity, from where the mucous travels to the nasal cavity through the ostia and further to the nasopharynx.[2] In case a disease is present in the maxillary sinus, the movement of mucous is obliterated due to inflammation, which brings about the major symptoms.[3] The anatomical positioning of the ostia of anterior ethmoid, frontal and maxillary sinus are in close approximation, therefore, pathologies involving any of the sinus may affect another.[4]

The ostium of MS is high on the medial wall and is about 2.4 cm in diameter. The development of MS is first among other sinuses. There are two growth spurts of MS development in which first one is at 0-3 years and another one is at 7-12 years. These growth spurts are in correspondence with the developing permanent dentition.[5] A lot of disease process can involve the MS

arising either from the sinus lining, the adjacent para nasal sinuses, nasal cavity and odontogenic origin, thus making the diagnosis quite difficult. These pathologies may sometimes show protrusion into any of the surrounding cavities.[6] MS pathologies present as masses casing proptosis of eye ball and such lesions are usually Neoplastic or Chronic granulomatous disease.[7] Therefore, it necessitate the need to develop certain algorithms for the diagnosis of MS pathologies.

Chronic granulomatous lesions are a group of diseases which show the presence of granuloma formation.[8] These granulomas are formed in an immune response to indigestible agent. These granulomatous changes can be associated with an underlying infection, inflammation or neoplasm. When these affect the head and neck region, mostly they present with a tumor like growth. Therefore, making it difficult to diagnose clinically as well as radio logically, since they sometimes mimic a malignant lesion in conventional radio graph.[9] Hence, it requires a set of clinical, radiological, surgical and pathological evaluations to reach a diagnosis and manage accordingly. Moreover, a few of symptoms in a chronic granulomatous disease overlap with each other making the diagnosis and management further complicated.[10]

These facts indicate towards development of a unique algorithm with each and every single case of growth involving the MS suspected to be a Chronic granulomatous disease. In the present short communication we have emphasized the need of development of algorithms while diagnosis using one such case as an example.

Development of Algorithm for a Case of Growth Involving the MS

A 35 year old female patient reported to Department of Dentistry with a chief complaint of swelling and dull pain on left side of face present since 3 - 4 months. On inspection, the she had diffuse swelling involving the left side of face, producing asymmetry. On further evaluation there was drooping of the eye of the affected side. On intraoral examination there was no apparent lesion present, but there was slight mobility in 26,27 and 28. A provisional diagnosis of growth left maxillary sinus was given. A para nasal sinus view (Water's position) was ordered initially, just to rule out if the swelling was cystic or there was any underlying solid growth. On evaluation of the radio graph, there was an ill defined radio opacity involving the left maxillary sinus. The margins of the radio opacity were irregular suggestive of a malignant neoplasm and/or chronic granulomatous lesion. Also this growth was causing an undisplaced fracture of the left zygomaticofrontal suture. Later, a 3D CT scan of the face and maxillary sinus was advised to rule out the extension of the lesion in axial, coronal and sagittal sections. The CT scan revealed a radio dense growth involving the left maxillary sinus, the nasal cavity, the orbital cavity where it was pushing the eye ball upwards hence causing the eyelids to droop. The coronal section, revealed large non-enhancing soft tissue mass within the left nasal cavity which extended into the left Maxillary antrum as well as ethmoidal air cells. The mass extended posteriorly through the posterior choana into the nasopharynx. The adjacent bony boundaries appeared sclerotic with areas of bone thinning, suggestive of pressure atrophy. Looking at this clear picture of the lesion the provisional diagnosis was narrowed down to chronic granulomatous lesion. To further establish a confirmatory diagnosis and to rule out any underlying malignancy, aspiration cytology was performed. The smears were stained with Hematoxylin and Eosin, Giemsa and Papinacoloau stain. These smears had presence of foamy macrophages in a background having lymphocytes and plasma cells. These cells had central to eccentric nucleus with an indistinct nucleoli. Few areas also showed presence of eosinophilic refractile bodies suggestive of Russell's bodies. There were no atypical cells. At this

stage, a malignant process was completely ruled out. Now the provisional diagnosis spectrum was narrowed down to chronic granulomatous lesions. A microbiological culture swab was performed from the nasal mucosa. This swab was inoculated on agar plate which came out inconclusive. Looking at the cytological smear, a differential diagnosis of Leprosy, Tuberculosis, Sarcoidosis, Rhinoscleroma and Mycotic infection was given. Leprosy was ruled out due to the absence of Acid fast bacilli using the Fite-Faraco stain. For ruling out tuberculosis, Ziehl Nelson stain was performed which was again ruled out due to the absence of any acid fast bacilli. None of the fields of the smears showed presence of any non-caseating granuloma, which ruled sarcoidosis. Also there were no fungal hyphae present which was enough to give up on the diagnosis of any mycotic infection. Therefore, looking at the presence of foamy macrophages, plasma cells and Russell's bodies, the provisional diagnosis was narrowed down to Rhinoscleroma. To get a confirmed diagnosis, an excisional biopsy was planned. The procedure which was first thought to be used for the biopsy was Caldwell-Luc operation but this procedure is rather more extensive and has higher chances of post-operative complications. Therefore, an advanced and more conservative approach was used such as that of Functional Endoscopic Sinus Surgery, hence performing an excisional biopsy. The biopsied specimen was sent for histopathology. During histopathology, it was found that the lesion was in the granulomatous phase in which there were pseudoepitheliomatous hyperplastic changes in the overlying mucosa. There was a dense inflammatory infiltrate which contained numerous lymphocytes, plasma cells with round to ovoid eosinophilic structures suggestive of Russell's bodies. The other significant group of cells which were large macrophages with clear vacuolated cytoplasm suggestive of Mikulicz cells. As per the literature, the cytoplasm of these macrophages is supposed to contain the causative bacteria Klebsiella rhinoscleromatis, which can be demonstrated using some special stains like Warthin-Starry silver stain, Giemsa stain and Gram stain. Knowing this fact, we used Giemsa stain for tissue sections, where we observed small blue rod shaped bacilli within the Mikulicz cells. Therefore with all these observations a final diagnosis of Rhinoscleroma was given. Post operatively the viral load was further reduced by presecribing Tablet Ciprofloxacin 500 mg twice daily and Tablet Amoxycillin cloxacillin 500mg thirce daily for seven days. She was kept on follow up for next two months where the swelling was observed to be kept reducing.

Fig-1: Extra-oral examination revealed a diffuse swelling causing disfigurement of face on the left side.

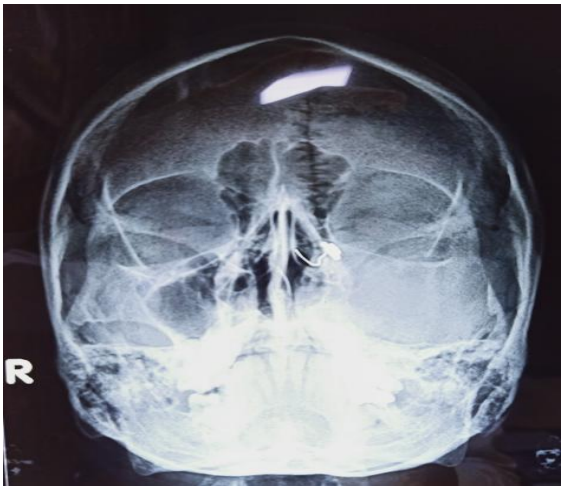

Fig 2: A paranasal sinus view (Water's Position) showed a aill defined radio opacity involving the left maxillary sinus. Also this growth was causing an undisplaced fracture of the left zygomaticofrontal suture.

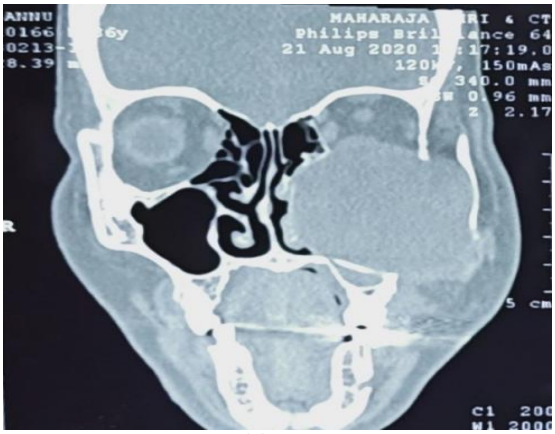

Fig-3: Transverse section of face and maxillary sinus shows a growth that involves left maxillary sinus, nasal cavity and orbital cavity, pushing the eyeball upwards.

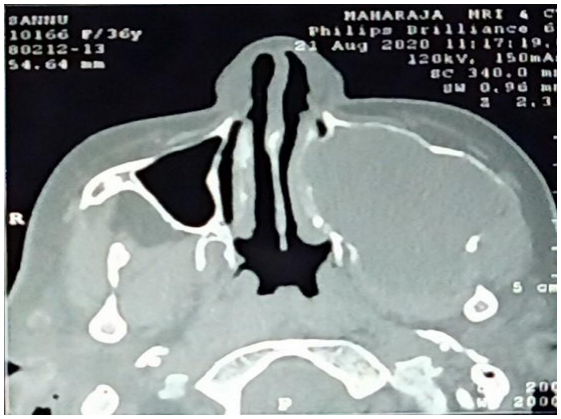

Fig-4: The CT scan on coronal section showed the mass was extending posteriorly till the ethmoidal air cells

Justification for the Role of Algorithm Development

The literary meaning of an algorithm is use of a step by step rule to reach a result. Chronic granulomatous lesions of the MS are sometimes difficult to diagnose sins these lesions are good mimickers of malignancy. Also with these lesions there is high chance of recurrence and if not given timely diagnosis and treatment they do cause a lot of local destruction. So in such cases, an algorithm comes into picture which will help in doing a step by step procedure to reach diagnosis. In the above mentioned case, we developed an algorithm to get to the final diagnosis and treat the patient accordingly. Rhinoscleroma is a chronic slowly progressing, non contagious granulomatous disease which is caused by gram negative Klebsiella rhinoscleromatis. This disease most commonly affects the nasal passages, pharynx and paranasal sinuses. Commonly occurs in low socioeconomic status population. Clinicopathologically this disease has three parallel stages which rhinitic / catarrhal followed by florid or granulomatous and lastly, fibrotic. In our case the patient presented during the granulomatous stage. During the diagnosis and management journey of this case we followed the following steps.

1. **Clinical Examination of lesion**
2. **Radiological Evaluation**
3. **Aspiration Cytology**
4. **Microbiological Evaluation**
5. **Surgical Management**
6. **Histopathological Evaluation**

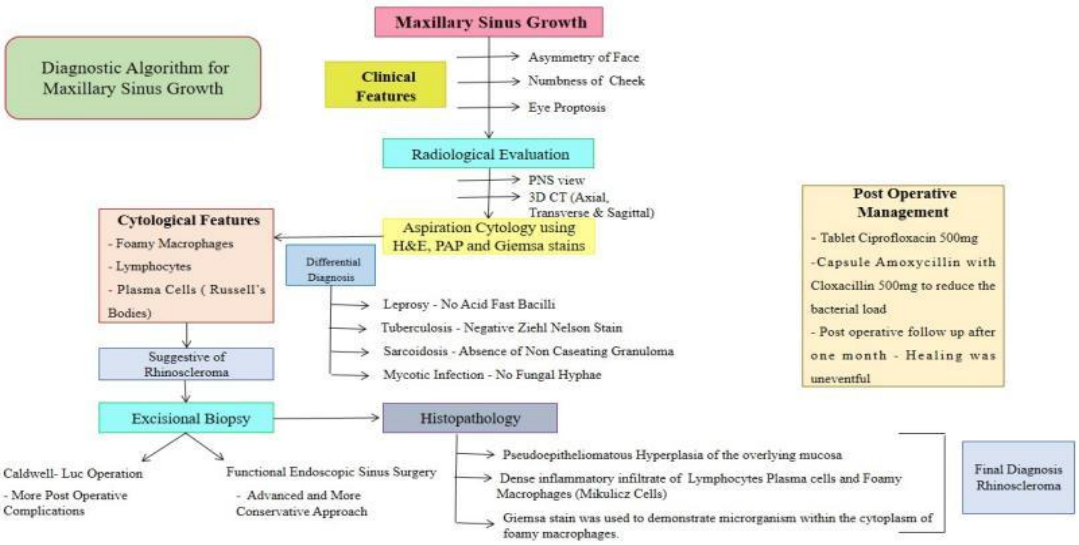

Fig-5: The pictorial representation of the diagnostic algorithm that was used during the diagnosis as well as management of the case. These algorithms should be developed with such lesions having a huge list of differential diagnosis.

CONCLUSION

To sum up, the chronic granulomatous lesions specially affecting the head and neck region, need a timely diagnosis and management in order to prevent the extensive destruction. Moreover, individuals with such granulomatous lesions can have overlapping features and without following a proper step by step algorithm an appropriate diagnosis is difficult.

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