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Questionable Nasolabial Lump: A Case Report

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Authors' contributions

This work was carried out in collaboration among all authors. Author KR managed the preoperative and postoperative care and data collection. Author CC was the operative surgeon. Author HKV wrote the protocol and was the chief surgeon. Author BS managed the formulation of study. Author SA managed the literature searches. Author GH was the assisting surgeon. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

As an otorhinolaryngologist it is not an everyday occurrence to find a swelling in the nasolabial fold area. The challenge however lies in the diagnosis, proper and timed treatment of the existing pathology, especially in a non – infective case. There are various pathologies that can lead to a swelling in the nasolabial area. We here report a rare case of a female patient who presented to our outpatient department with a slowly progressive nasolabial swelling which radiologically pointed towards a Nasolabial cyst. However, the final diagnosis was an entity on the other end of spectrum, an Ameloblastoma, an odontogenic tumor.

Keywords: Ameloblastoma; mandible; odontogenic; nasolabial cyst.

1. INTRODUCTION

Nasolabial cysts are rare nonodontogenic soft-tissue lesions of nasal vestibule, fossa canina and sublabial region. The overall incidence of the cyst is 0.7%. These lesions are usually painless swellings involving sublabial fold, lips, face and leads to nasal obstruction. Pain can occur if it becomes infected [1].

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Ameloblastoma on the other hand are odontogenic lesions. According to the World Health Organization (WHO), 1991, it act as a benign tumour which is locally aggressive with a high tendency to recur [2]. Ameloblastoma, subject to late and rare presentation, can result in facial asymmetry, displacement of teeth, malocclusion and pathologic fractures attributed to its size progression.

2. CASE REPORT

A 50 year old female reported to our hospital with complaints of swelling over the region of right nasolabial fold [Fig. 1] with associated pain since one year. This slowly progressive growth did not cause any nasal obstruction or difficulty in breathing. There was no history of nasal trauma, bleed, discharge, nasal blockage, fever, hyposmia or anosmia. There was no history of swelling on other parts of the body.

The external appearance comprised of a well-defined, hard, nontender swelling in the right nasolabial region measuring (2*3) cm with normal overlying skin colour or any localised skin changes.

On anterior rhinoscopy deviated nasal septum towards left was noted. On examination of the oral cavity a hard swelling over the right side of the hard palate was observed measuring (1*2) cm. Computed tomography of the paranasal sinuses revealed a large lytic expansile cystic lesion involving the upper alveolus in the right paramedian plane with extreme thinning of the cortex along the anterior aspect and cortical erosions along the posterior and lateral aspects. A small lobulated relatively hyperdense soft tissue component was noted along the posterior aspect predominately in the region of cortical erosions and extension into the adjacent region of oral cavity. There was no evidence of unerupted tooth with erosions of the roots of the adjacent teeth, the lesion measuring approximately (3.8*3.2*2.3) cm. Small polypoidal soft tissue density in the left maxillary sinus representing polyp/retention cyst was suggested [Fig. 2].

With past and personal history being insignificant, all routine blood investigations were sent which were within normal limits and the patient was thus posted for surgery.

A right nasolabial cyst excision was performed in which a sublabial incision was taken in the upper gingivolabial sulcus and cyst was exposed and dissected off. The final sample was sent for histopathological examination (HPE.)

On HPE,[Fig. 3] a cystic mass showed cyst wall lined by odontogenic epithelium with basaloid columnar cells showing peripheral palisading. Subepithelium showed fibrocollagenous tissue along with dead bony trabeculae. Solid mass showed a tumor mass composed of well differentiated odontogenic epithelium showing peripheral palisading with cystic areas with plasmaceous material, congested blood vessels along with large areas of fibrin deposition compressing the odontogenic epithelium. There was also presence of dense mixed inflammatory cell infiltrate, foreign body giant cells and foamy macrophages.

Thus, features of benign odontogenic epithelial tumor favoring amelobastoma were seen. The patient had an incident-free hospital stay and followed up for six months. The patient underwent Computed tomography of the paranasal sinuses after six months in which no recurrence was observed.

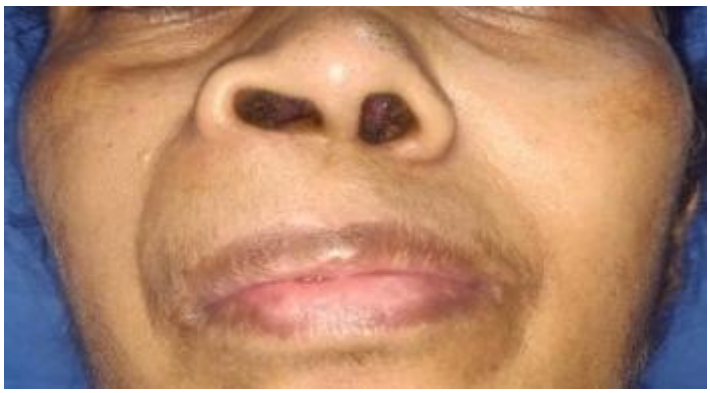

Fig. 1. Preoperative view showing swelling over right nasolabial fold

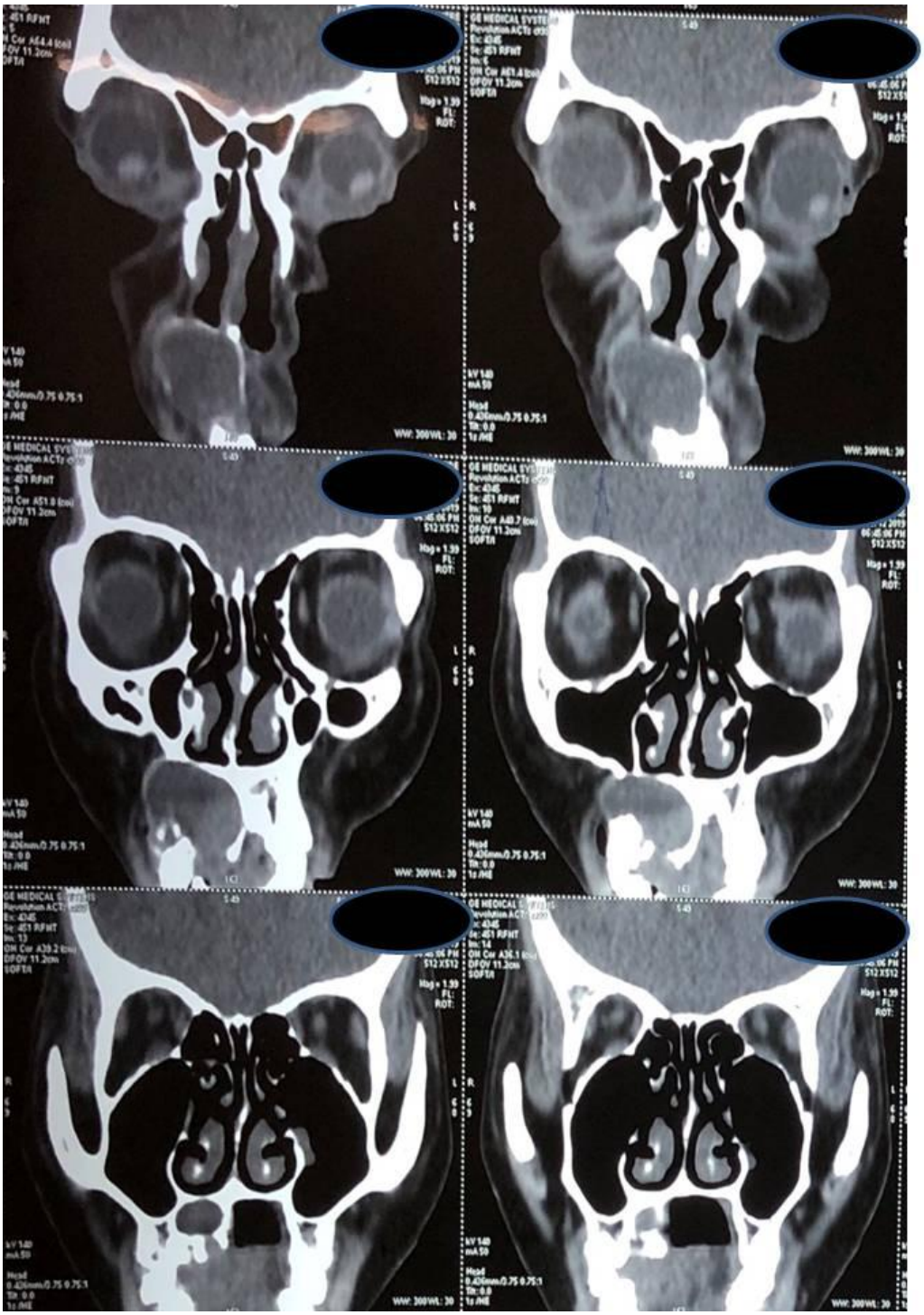

Fig. 2. Preoperative computed tomography of the paranasal sinuses

3. DISCUSSION

Various differentials of swelling can occur in anterior maxilla or in soft tissues of alar labial region and may include odontogenic and nonodontogenic lesions. Nasolabial swelling differentials include nasopalatine duct cyst, globulomaxillary cyst, soft tissue cysts such as

dermoid or epidermoid cyst, furunculosis of the base of the nose, benign tumours, soft tissue and salivary gland tumours. Malignant tumours may include mucoepidermoid carcinoma [3].

Nasolabial cysts are rare and also are said to be under-reported. They comprise of about 0.7% of all cases of maxillofacial and only 2.5% of the maxillofacial non-odontogenic cysts [4]. They are commoner in black women in the 4th to 5th decades of life and mostly unilateral (90%) of cases and 10% bilateral [5]. Also regularly called as nasoalveolar and Klestadt cysts, in 1951, Rao introduced the term 'Nasolabial cyst [6].

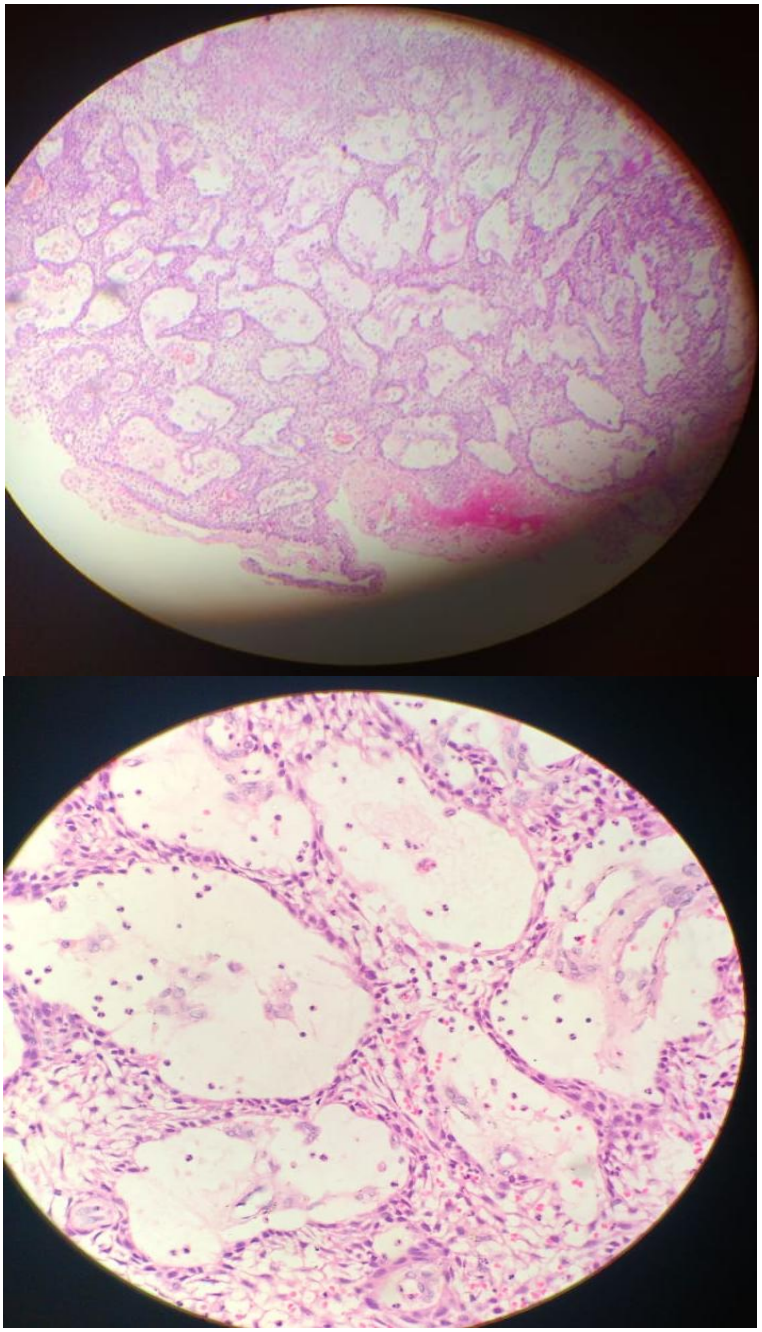

Fig. 3. Low-power and high power magnification showing histopathological sections of ameloblastoma with follicular and cystic variant

There are three theories for the formation of the cyst: Embryological formation by detention cells in the maxilla, medial and lateral nasal wall; by detention cells from the inferior nasolacrimal channel redundant cells- the most accepted theory given by Bruggemann; by detention cells from the inferior nasolacrimal channel endodermal cells [7].

History of trauma hastens the formation of the cyst .It is quite constant to find nasolabial cysts at the anterior nasal floor. This location is distinctive too. Bull et al. in 1967 have described it as pathognomonic [8]. From here the possibility of the cysts to grow can be in following three directions: to the nasolabial fold, the mouth vestibule and the nasal vestibule. Patients either can be asymptomatic or may come to you with at least one of these three symptoms: partial or complete nasal obstruction, well-circumscribed swelling and/or localized pain [8]

Ultrasonography acts as an office-based diagnostic modality. A computerized tomographic (CT) scan provides clues to proper diagnosis and is also the imaging modality of choice. CT findings include a well-demarcated, rounded, homogeneous, low-density soft tissue lesion at the nasolabial region with or without evidence of scalloping and bone remodelling. It is of utmost importance in pre-operative. MRI scans are important to shed light on the origin of the cysts and they avoid unwarranted needle aspiration or dental surgery [9]. However histological examination provides ultimate diagnosis. Resection of the cyst allows both, cure and final diagnosis. Various management modalities include endoscopic marsupialization, surgical excision, incision and drainage, injection of sclerotic agents, simple aspiration and cauterization.

Coming to our case, the word ameloblastoma derives itself from an English-Greek aggregation, Amel meaning enamel and Blastos meaning germ. First described in 1827 by Cusack, in 1885, Malassez introduced the name 'Adamantinoma' which according to Fisher in 1913, is now a rare form of bone cancer. It was described in detail first by Falkson in 1879. The term ameloblastoma was coined by Ivey and Churchill in 1930 and is considered as a true neoplasm. Ameloblastomas are extremely rare odontogenic tumours comprising only about 1% of all jaw tumours and being the second most common odontogenic tumours (9-10%). Fortunately, they are a benign tumour but with locally aggressive behaviour. The tumour is commoner in mandibular region than in maxillary. Amongst maxillary sites, 98% occur posteriorly and 2% anteriorly [10]. Ameloblastomas usually are asymptomatic until people notice intraoral or facial swelling. Patients often present with slow, progressive maxillary or mandibular expansion and facial asymmetry, mostly without pain and altered sensation. There can be complaints of change in bite and loose teeth. Smaller tumors are usually detected first on a regular ear, nose, throat or routine dental radiographic examinations. Ameloblastomas of maxillary region may prove fatal in some cases because of the lack of a thick cortical plate, plentiful cancellous bones and the proximity of the maxilla to the nasal cavity, nasopharynx, paranasal sinuses, orbits and skull base, there is commonly a delay in the recognition of the extension into these structures [11]. This progression of the lesion can lead to difficulty in mastication, deglutition, epistaxis, nasal obstruction with rhinorrhoea, however uncommon.

Maxillary ameloblastoma is an unusual epithelium-derived odontogenic tumour. Ameloblastomas are more common in 30-40 years of age [12].

On gross appearance it can be cystic, solid or cystic-solid with almost all ameloblastomas have cystic degeneration [13]. Small and Waldron believe that cystic degeneration is related to age. In conclusion, majority (85.7%) were solid, 4.8% cystic and 9.5% solid-cystic and become functional only after odontoblasts form the primary layer of dentin (the layer beneath enamel).

As per WHO and the International Agency for Research on Cancer, 2003, ameloblastoma is a benign tumour with odontogenic epithelium, mature fibrous stroma and without odontogenic ectomesenchyme [14]. Ameloblastoma is further classified into solid/ multicystic, extraosseous/peripheral, desmo-plastic ameloblastoma, unicystic.

Unicystic ameloblastoma (UA), first used by Robinson and Martinez in 1977, also called by the WHO as 'Çystogenic ameloblastoma', presents as a swelling with clinical and radiologic characteristics of an odontogenic cyst. HPE shows a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumour proliferation. UA comprise 5-15% amongst all types. With an unerupted tooth UA occurs with a mean age of

16 years as opposed to 35 years with no unerupted tooth. The mean age is considerably lower than that for solid/multicystic ameloblastoma with some stating no gender predilection while some opine of slight male preponderance. The stated recurrence rate is 6.7-35.7% with average interval for recurrence ~7 years. Thus, this tumour is prognostically distinctive.

The pathogenic mechanisms proposed in the formation of UA are reduced enamel epithelium from dentigerous cyst and due to cystic degeneration of solid ameloblastoma. Some studies also suggest role of deposits of these cells in the structures in and around the tooth, termed cell rests of Malessez and cell rests of Serres. Genetic mutations that activate a specific signalling pathway (MAPK) play a pivotal role in the pathogenesis [15].

UA has 6 radiographic patterns, ranging from well-defined unilocular to multilocular ones. There is an apparent predominance of a unilocular configuration in all studies of UA and they might mimic other odontogenic cysts. Radiographically ameloblastoma appears as expansile, radiolucent, often with multilocular or “soap-bubble” appearance, cystic lesions. There is thinning and expansion of the cortical plate with erosion through cortex. There might be unerupted tooth displaced and resorption of the roots of the adjacent teeth.

Radiodiagnosis can help to rule out abscesses and periapical inflammatory conditions. But the final diagnosis can be achieved by surgical biopsy with typical features [16].

Ameloblastomas are treated with complete surgical excision with normal bone margins of minimum 5-15 mm, though 10 mm margins are most common [15,17]. Upto 70% recurrence rates have been reported most commonly due to incomplete resection. Enucleation and curettage, cryotherapy and marsupialization have all been used to treat ameloblastomas, however, they are not standard of care now.

Differentials like malignant ameloblastoma and its two types which are metastasizing ameloblastoma and amelobastic carcinoma. Treatment of ameloblastic carcinoma is via surgical resection with margins of 2 to 3 cm. Concurrent chemoradiotherapy can been kept as an option post resection in case there are positive margins or perineural invasion. In metastasizing ameloblastoma, resection with 1 to 2 cm margins is preferred choice of treatment and no chemo-therapy or radiotherapy is generally required [15]. A Phase 2 study at Stanford University studying Dabrafenib and Trametinib for patients with BRAF mutated ameloblastoma is most likely to undergo completion by August 2020. Close patient follow-up for a minimum of five years is necessary to monitor for recurrence.

Mostly followed are current recommendations from the American Joint Committee on Cancer and the National Comprehensive Cancer Network for more detailed treatment modalities, indications for neck dissections, chemo, and radiotherapy.

4. CONCLUSION

To conclude, nasolabial swellings can be easily subjected to misdiagnosis on examination. As stated by the literature, ameloblastomas are a late diagnosis because of their poor symptoms and low prevalence. Therefore, a proper work up is of absolute necessity. The importance of radiological examination in diagnosing and preoperative evaluation cannot be understated. It is of extreme importance that the malignant variant of this tumour, however rare, must be taken into consideration while planning treatment options. Histopathological examination is the ultimate diagnostic tool. Excision surgery offers satisfactory result with a close watch during follow-up period.

CONSENT

As per international standard informed and written participant consent has been collected and preserved by the authors.

ETHICAL APPROVAL

As per international standard written ethical permission has been collected and preserved by the authors.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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