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CASE REPORT

CONTRAST ENHANCED EVALUATION OF NASOLABIAL CYST-A QUEST FOR THE ELUSIVE

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ABSTRACT

Nasolabial cyst is an uncommon, soft tissue cyst of non-odontogenic origin found in the maxillofacial region. It presents as a slowly enlarging, asymptomatic swelling in the nasolabial region. Being a soft tissue cyst, it cannot be appreciated radiographically. We present a case of nasolabial cyst in a 60 year old female patient who reported to us with a swelling in the left nasolabial region. Iodine contrast agent was injected into the cyst followed by a series of images to evaluate the entire extent of the lesion. This paper highlights the importance of use of radiographic contrast agent in the diagnosis of this rare lesion. A brief review of literature with emphasis on the conventional radiographic and computed tomography findings is presented.

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INTRODUCTION

Cysts affecting the head and neck region could either involve the bone or the soft tissues. They can be either developmental or inflammatory in origin (Patil *et al*., 2007). The nasolabial cyst is a rare developmental, soft tissue cyst that occurs as a swelling in the nasolabial fold at the base of the alae of the nose. It accounts for about 0.7% of cysts of the jaws and 2.5% of the non-odontogenic cysts (Amaral *et al*., 2005).

Case report

A 60 year old female patient, reported to our department with a chief complaint of swelling in the left nasolabial region since 20 days. The swelling was insidious in onset, associated with mild pain. Other associated symptoms like paresthesia or any discharge from the swelling was not reported. The patient had visited an independent dental practitioner 1 month back for pain in the left upper front tooth which was removed. She was otherwise in good health and her medical history was unremarkable. On examination, a diffuse extra-oral swelling (Figure 1) measuring about 3.5 X 2.5 cms was noted in the left nasolabial region causing mild elevation of the ala of the nose and obliteration of the nasolabial sulcus on the affected side.

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Intraorally, a solitary pale pink colored swelling was noted in the left maxillary labial vestibule, causing obliteration of the same in relation to missing 21, 22,23 region (Figure 2). The swelling was mildly tender on palpation and soft to firm in consistency. Co-relating with the history of extraction of the painful tooth and associated swelling, the lesion was provisionally diagnosed as residual cyst. The clinical differential diagnosis of odontogenic keratocyst, nasolabial cyst, epidermoid cyst, mucocele, benign salivary gland tumor was given. Intraoral periapical radiograph, maxillary anterior occlusal view and orthopantamograph revealed no evidence of any abnormality. Following negative results on radiographic examination, a diagnosis of nasolabial cyst was given. Aspiration via intra-oral route was performed which yielded around 4-5 ml of straw colored fluid. Following aspiration, an equal amount of iodine containing radiopaque contrast agent (Urografin 76%) was injected into the cyst, following which a series of radiographs were taken. Maxillary anterior occlusal view showed a homogeneously radio-opaque well defined, elliptical lesion measuring about 3.5 X 2.5 cm in the left maxilla (Figure 3).

Posterio-anterior view of skull (Figure 4a) showed the contrast media defining the cyst in the soft tissues of the left naso-maxillary region and lateral view of skull (Figure 4b) confirmed the extra-osseous nature of the cyst. A submentovertex view was taken to demarcate the axial extent

of the cyst and showing that it is a separate entity not arising from bone (Figure 4c). Axial, coronal (Figure 5) and sagittal computed tomography sections revealed a well defined, hyperdense lesion (3000 HU) in the floor of the nasal cavity on the left side. The cyst was enucleated through an intra-oral approach (Figure 6) under local anaesthesia and the specimen was sent for histopathological analysis.

Microscopic section revealed cyst lined by ulcerated stratified squamous epithelium. Walls showed fibrous tissue with dense acute and chronic inflammation and skeletal muscle fibres (Figure 7). Based on the clinical, imaging and histopathologic findings, a final diagnosis of nasolabial cyst was made. The post operative period was uneventful.

Figure 1. Extra-oral photograph showing fullness below the left nostril

Figure 2. Intra-oral photograph showing obliteration of the left maxillary vestibule

Figure 3. Maxillary anterior occlusal view show well-defined, elliptical, radio-opaque lesion following injection of contrast medium

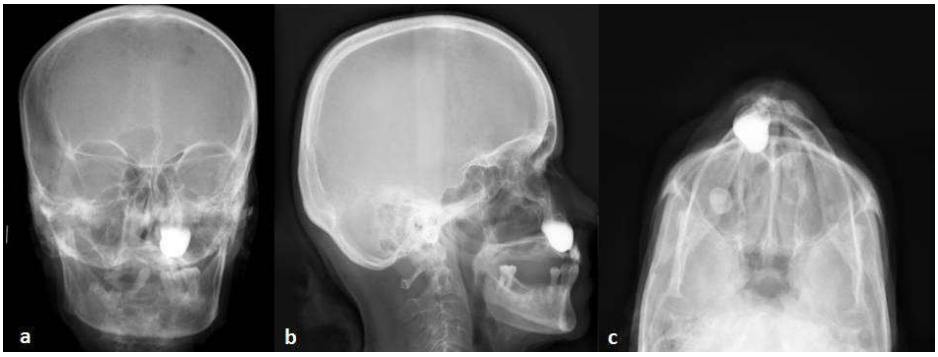

Figure 4. Postero-anterior, lateral skull and submentovertex views showing the extent of the lesion in three dimensions

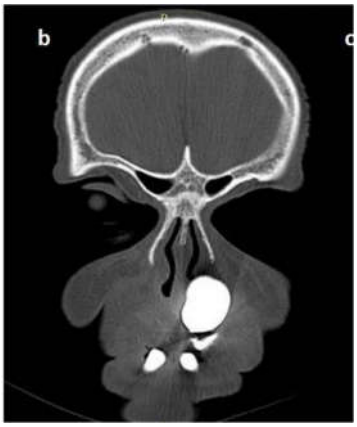

Figure 5. CT scan showing the coronal section of the lesion

Figure 6. Intra-operative view showing the excision was done via an intra-oral approach

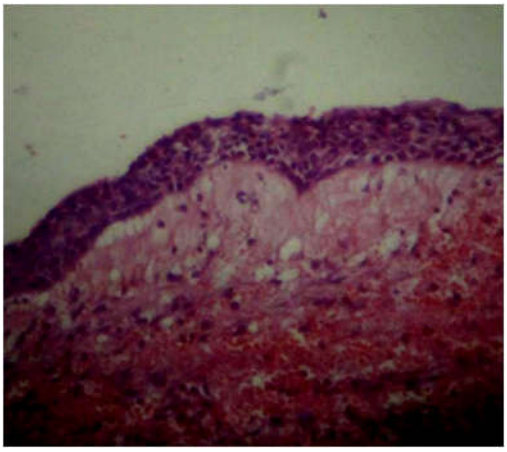

Figure 7. Hematoxylin & eosin stain showing the cyst wall lined by stratified squamous epithelium

DISCUSSION

The nasolabial cyst is an uncommon, non-odontogenic, benign, extraosseous lesion located in the paramedian region of the anterior maxilla and below the nasal ala (Friedrich *et al*., 2012). Nasolabial cysts were first described by Austro-Hungarian anatomist Emil Zukerkandl in 1892 (Toribio and Roehrl, 2011; Sahin, 2009). The first case was reported by McBride in 1892. In 1989, Brown-Kelly described this condition in greater detail (Aquilino *et al*., 2008). Thoma suggested the term nasoalveolar cyst to describe this lesion, but Rao in 1951 first used the term nasolabial cyst and defined nasolabial cysts as lesions located entirely within soft tissue, different from nasoalveolar cysts, which caused maxillary bone erosion (Aquilino *et al*., 2008; Parelkar *et al*., 2015). In 1953, Klestadt investigated nasolabial cysts in detail, after which the lesion became named Klestadt's Cyst in his honor (Vijay *et al*., 2014). It has been given many names such as Klestadt's cyst, nasoalveolar cyst, nasal vestibular cyst, mucoid cyst of the nose, and nasal wing cyst (Arul *et al*., 2012). The pathogenesis of this cyst is controversial. Previously proposed theories suggested their origin from inflamed mucous glands and considered them as retention cyst (Cure *et al*., 1996). The developing embryologic mid-face includes bilateral medial and lateral nasal and maxillary swellings. The medial nasal swellings form the middle portion of the nose, and, along with the maxillary swellings, contribute to the central upper lip. The nasal alae form from the lateral nasal swellings (Cure *et al*., 1996). Klestadt in 1913 suggested that the nasolabial cyst originates from entrapped epithelial tissue, which remains after the fusion of maxillary and nasal (median and lateral) process. According to this theory, the nasolabial cyst is a fissural or inclusion cyst (Matiakis *et al*., 2013; Pandav *et al*., 2012). Bruggemann in 1920 was the first to postulate that nasolabial cysts form from the anlage of the nasolacrimal duct (Cure *et al*., 1996; Matiakis *et al*., 2013). The nasolacrimal duct arises from the nasolacrimal groove, a deep furrow that separates the lateral nasal swellings from the maxillary swellings. Epithelial cords arise from ectoderm in the base of the nasolacrimal groove and are initially attached to the overlying ectoderm. After detaching from the overlying ectoderm, these cords canalize to form the nasolacrimal duct, and the maxillary and lateral nasal swellings subsequently fuse. Epithelial remnants of the nasolacrimal groove might therefore persist just deep to the junction of the nasal ala with the lip. A cyst may thus arise from these remnants and would develop more anteriorly than the opening of the nasolacrimal duct into the inferior meatus (Cure *et al*., 1996). This theory is supported by the fact that nasolacrimal duct is lined by pseudostratified columnar epithelium, the epithelium found in the cystic cavity of nasolabial cyst (Patil *et al*., 2007). This lesion has a strong predilection for women, in a ratio of approximately 3:1, with a mean age between the fourth and fifth decades of life, and higher incidence in individuals of the black race (Moitinho *et al*., 2013). Tiago *et al* studied 8 cases of nasolabial cysts and found that the mean age at which cysts were detected was 45.5 years. Due to their slow growth, nasolabial cysts tend to be detected in older patients (Aquilino *et al*., 2008). Contrary to what is described in literature, our patient presented with only a 20 day history of swelling. It might be possible that the cyst would have been there but was not noticed by the patient until it got infected and became painful. The clinical presentation is typical with a spherical swelling beneath the nasal ala causing its elevation and obliterating the nasolabial fold. Lesion distends the mucolabial sulcus intra□orally and can cause discomfort in denture users (Arul *et al*., 2012). Some patients may complain about difficulty in nasal breathing. The nasolabial cyst is asymptomatic but may be painful if infected (Amaral *et al*., 2005; Matiakis *et al*., 2013). Our patient also complained of pain associated with the swelling. In 90% cases cysts are unilateral and can rarely present as bilateral cyst (Goyal *et al*., 2014). It does not cause any displacement of the teeth (Patil *et al*., 2007). The cyst is soft, fluctuant and is best palpated bimanually with one finger on the floor of the nose and one finger in the labial sulcus. The teeth in the area of the lesion are vital. Sometimes the cyst may rupture spontaneously and drain into oral cavity or nose (Patil *et al*., 2007). In case of dentulous patients, nasolabial cyst must be differentiated from other pathologies that may present at this site namely, periapical abscess, granuloma or cyst that may perforate the bone and manifest in a similar fashion. Pulp vitality test may thus help in differentiating them from nasolabial cyst. Our patient was edentulous and gave history of extraction, thus possibility of residual cyst was considered. Other possible differential diagnoses could be epidermoid cyst, mucocele, lipoma, salivary gland tumors, odontogenic keratocyst (Arul *et al*., 2012).

As this is a soft tissue lesion, the nasolabial cyst is only detected radiographically if deformation of the lateral and anterior limit of the nasal fossa, with convexity in the posterior direction is detected on the occlusal radiograph or if it causes bone erosion in the maxillary bone (Moitinho *et al*., 2013). Our case showed no changes on conventional radiographs. Thus contrast medium was injected and radiographs were taken. The visualization of cyst by injecting radiographic contrast medium is a safe, low cost, simple and rapid method. This procedure is precise for visualizing the definite extensions of the lesion, mainly in those cases when plain radiographs may not show any detectable changes and where advanced radiographic techniques (CT, MRI etc) cannot be accessed and patient cannot afford expensive investigations. Another disadvantage is exposure to higher radiation dose with computed tomography which can be avoided using this technique. Following injection of contrast medium, preferably two different views perpendicular to each other must be taken to provide information on height, width and depth of the cyst in three dimensions. The procedure is safe if the patient is not allergic to the iodine contrast agent. Whilst the injection of radiographic contrast medium could rupture the borders confining the cyst and then cause dissemination of bacteria into other facial planes, it can only happen when a different volume of radiographic contrast medium from the fluid aspirated is injected (Vijay *et al*., 2014). Gomes *et al*. have suggested that CT provides a superior view of surrounding bone around the cyst (Gomes *et al*., 1992). CT scans usually reveal a homogeneous, cystic lesion, anterior to the piriform aperture (Giongo *et al*., 2014). Contents of uncomplicated lesions may be hyperdense on unenhanced CT scans, slightly hyperintense relative to CSF on T1-weighted MR images, and isointense with CSF on T2-weighted MR images (Cure *et al*., 1996). Treatment with injection of sclerotic substances, marsupialization or surgical removal, have been attempted to

resolve these lesions. However, unlike the large intraosseous cysts, the soft-tissue lesions do not respond to marsupialization and surgical excision is the treatment of choice. As these cysts are closely related to the floor of the nose, the perforation of the nasal mucosa may occur during their removal and it needs repair before closure. The treatment of choice of this cyst is enucleation and transoral method is preferred approach.

Conclusion

Nasolabial cyst is a rare entity and requires careful diagnostic protocol as it is located in the danger area of the face and if secondarily infected, it may spread to cavernous sinus which may lead to serious consequences. The cyst is not readily visible on radiographs as it is a soft tissue entity. This warrants the use of contrast enhanced radiography for necessary diagnostic and therapeutic considerations; avoiding the use of techniques which require high radiation dose. Every case, howsoever insignificant or inconspicuous, is important in that it may lead to significant morbidity to the patient. In the head and neck region, nasolabial cyst appears elusive on conventional radiographs. Thus, best use of current imaging technology using contrast enhancement was made in this case to obtain precise diagnosis and thereby providing the best treatment possible.

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