Nasolabial cyst: diagnosis mainly based on topography?

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SUMMARY

The nasolabial cyst is described in three patients and literature is shortly reviewed. The condition manifests itself by a smooth, fluctuating swelling in the nasolabial fold next to the ala of the nose. This clinical manifestation represents the most important criterion for diagnosis. Histopathology and cytologic findings are of minor importance for diagnostic purposes. Etiology is still uncertain. According to the most accepted theory the lesion has to be classified among the fissural cysts, but histopathologic findings may vary and do not give support to any of the theories of etiology. Computertomogaphy is better than conventional roentgenography to demonstrate the extension of the lesion into surrounding structures. Echography can give important preoperative information. Nomenclature related to this cystic lesion is not uniform. Nasolabial cyst is the most adequate term, as it is describing the lesion only topographically and is not referring to controversial etiologic theories or to variable histopathology. Therapy consists of surgical removal using an intraoral approach and prognosis is excellent.

INTRODUCTION

Cysts in the maxillary region occur rather frequently, but seldom the nose is involved. Symptoms and complaints of disturbed nasal function usually manifest in advanced cases only. It may be difficult to diagnose these lesions correctly because a uniform and generally accepted nomenclature is still not present. The next three cases may illustrate this.

Case histories

Patient 1. A 46-year-old woman had a swelling for some weeks at the right side of the nose, which had grown rather progressively. Examination revealed a nontender swelling in the right nasolabial fold elevating the ala of the nose (Figure 1). This lesion was bulging into the nasal vestibule and into the gingivolabial fold. The patient was edentulous. Routine radiographic findings (Tschebull's projection, orthopantomography) were negative. At needle aspiration non-transparent mucous fluid was obtained, mainly containing granulocytes and some histiocytes. Nasolabial cyst was the presumptive diagnosis and an operation was

Figure 1.

Frontal view of patient 1 showing facial asymmetry due to a nasolabial swelling at the right side.



performed under general anaesthesia. The approach consisted of an incision in the gingivolabial fold and a cyst of 2 cm diameter was removed by blunt dissection. A small perforation was made in the nasal mucosa and had to be sutured. Postoper-

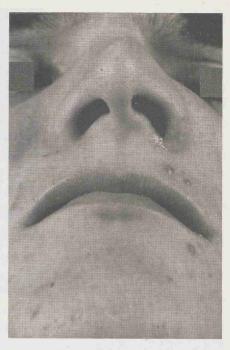
Microscopic examination revealed that the wall of the cavity consisted of connective tissue with a lining of pseudostratified columnar epithelium with numerous goblet cells (Figure 2) and some ciliated respiratory epithelium.

Figure 2. Photomicrograph of the wall of the cyst in patient 1 showing connective tissue with a lining of pseudostratified columnar epithelium with goblet cells.



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Figure 3. Patient 2 with a swelling in the left nasolabial fold.



Patient 2. A 20-year-old woman had a swelling for about six months at the left ala of the nose (Figure 3).

At rhinoscopy an enlargement of the left inferior turbinate was manifest. Dentition was normal. Fluctuation was present at palpation. Routine radiological findings were normal and there were no indications of dentigorous or periapical pathology. However, computertomography showed a density in the inferior turbinate, the piriform aperture and the ventral aspect of the maxillary sinus (Figure 4). A small discontinuity in the bony outline marked the extension of the lesion into the maxillary sinus.

Ophthalmologic and neurologic examinations revealed no abnormalities. At needle biopsy yellowish fluid was obtained containing macrophages, granulocytes and cholesterol crystals. An operation was performed and the lesion was explored via an incision according to Denker in the gingivolabial fold. A part of the canine fossa was removed enabling a clear view of the extension of the lesion into the maxillary sinus (Figure 5).

A small bony dehiscence of the lateral nasal wall was observed, furthermore the cyst extended into the inferior turbinate. The lesion was completely removed, together with a part of the nasal mucosa, leaving an antrostomy. Postoperative course was uneventful and healing was by first intention.

At microscopic examination of the specimen a cyst was seen with a wall of connective tissue partially covered with stratified squamous epithelium. Irregularity

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Figure 4.

Computertomographic picture of patient 2 demonstrating a well-confined density in the left vestibule of the nose. A small dehiscence in the piriform aperture is present, marking the extension of the lesion into the maxillary sinus.





Figure 5. Operative view of patient 2. The cyst is exposed through a sublabial incision and a part of the ventral wall of the maxillary sinus is removed. The arrow indicates the extension of the cyst into the maxillary sinus.

of the bony outline and osteoclastic activity were present wherever the cyst was in close proximity with the bony structures (Figure 6).

The cyst contained not only fluid but also some loose connective tissue with scattered leukocyte infiltration and cholesterol particles.

Patient 3. A 45-year-old woman had since three months a slowly growing swelling at the right side of the nose, possibly "the result of a trauma". No complaints of nasal obstruction were present. At clinical examination a smooth, fluctuating non-tender swelling of about 2 cm diameter could be palpated in the right nasolabial sulcus. The right nasal vestibule was slightly narrowed by this lesion. She was edentulous in both dental arches. Tomography of the paranasal sinuses and orthopantomography showed no radiolucencies or erosions of bony structures. At echography a hollow structure of 8×18 mm was present 5 mm under the skin surface (Figure 7).

Diagnosis was nasolabial cyst and at operation a cystic lesion was enucleated through an incision in the gingivolabial sulcus (Figure 8).

The cyst was located at the ventral aspect of the right inferior angle of the piriform aperture and no signs of bony erosion were found. A small perforation in the nasal mucosa resulted but healed spontaneously afterwards.

At histopathologic examination a cyst was seen filled with seromucous fluid and

Figure 6.

Photomicrograph of the cyst in patient 2. The wall is built up by connective tissue partially covered with stratified squamous epithelium. Note the irregularity of bony architecture with osteoclastic activity in the area of contact between the cyst and the bone.



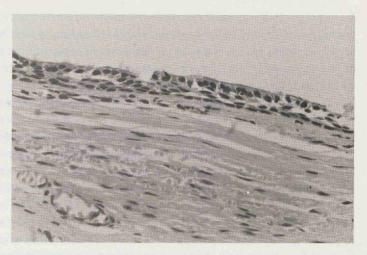


Figure 7. Echograph of the cyst in patient 3 demonstrating the exact dimensions of the lesion.



Figure 8. Operative view of patient 3. The nasolabial cyst is clearly exposed through an intraoral incision.

Figure 9. Photomicrograph of the cyst in patient 3. The wall of the cyst consists of a fibrous sheet covered here with stratified columnar epithelium.



a lining of a thin fibrous sheet covered with simple squamous, simple cuboidal and stratified columnar epithelium with some mucous glands (Figure 9).

These patient histories may give rise to some questions. In each of these cases nasolabial cyst was the diagnosis but it may be doubtful whether this was justified. Which criteria are needed to make a correct diagnosis? Has this diagnosis specific consequences? Before these questions can be answered some data described in the literature have to be discussed.

REVIEW OF LITERATURE

Most authors agree that Zuckerkandl was the first to describe the nasolabial cyst in 1882. Since that time this rather uncommon condition has been the subject of numerous publications. Comprehensive surveys were published by Roed-Petersen (1969) and Allard (1982). They demonstrated that until now 218 cases are described in the literature, which means that the nasolabial cyst represents about 0.3% of all cysts described in the maxillary region. Male-female ratio is about 1/3.5, localization is unilateral in 90% of the cases and the condition occurs probably more frequently in the negroid race. Onset of symptoms is usually between the fourth and fifth decade. Allard (1982) found 20 synonyms for the nasolabial cyst in the literature. Klestadt's cyst, naso-alveolar cyst and nasolabial cyst are the terms mostly used.

About *etiology* no consensus is present. In the beginning of this century several theories have been described. However, most of them have been rejected. Only two theories of etiology deserve attention because they are still widely recognized and are potentially acceptable.

Brüggeman (1920) suggested that the cyst might originate from a part of the nasolacrimal duct. The localization of the cyst in the upper lip next to the nasal ala is hard to explain by this hypothesis. However, during embryonic development posterior displacement of the nasolacrimal duct may occur due to outgrowth of the maxillary process (Stoelinga, 1971).

Klestadt (1913) classified the nasolabial cyst among the fissural cysts. According to this hypothesis the lesion originates from a proliferation of epithelial components entrapped in the developmental fissures between medial nasal, lateral nasal and maxillary processes. Therefore the condition is supposed to be an intermediate stage in the development of a hare lip. Later most authors supported this theory. However, the question has to be raised why this cyst manifests itself never at birth but only during later life. Another important question is why this condition is most often found in females. Until now no universal explanation has been provided for these questions and the exact etiology is still uncertain.

Clinical features include the presence of a swelling next to the ala of the nose, which is non-tender, well-confined, smooth and fluctuating. This swelling usually extends into the buccogingival sulcus and into the nasal vestibule. Some authors mention bony erosion (Walsh-Waring, 1967; Roed-Petersen, 1969; Brandao et al., 1974; Schmallenbach and Austermann, 1976), others state that the lesion is strictly confined to the soft tissues (Atterbury et al., 1961; Brons and Jongebreur, 1967; Bull et al., 1967; Bone, 1972; Karmody and Gallagher, 1972). The latter is supported by the observation that radiographic evidence of bony erosion is seldom present (Kwast and Roorda, 1963; Santora et al., 1970). Differential diagnostic possibilities include furunculosis of the nose (Roed-Petersen, 1967), several maxillary cysts like globulomaxillary cyst, nasopalatine duct cyst and all types of odontogenic cysts (Brons and Jongebreur, 1967). Furunculosis of the nose manifests itself by a painful swelling and this infectious condition has a different clinical course. Maxillary cysts usually can be distinguished from the nasolabial cyst by their localization and the presence of evident radiolucensies in the maxillary bone.

Histopathology may vary considerably. In the lining of the cyst different types of epithelium can be found: simple squamous epithelium, stratified squamous epithelium, pseudostratified columnar epithelium, simple cuboidal epithelium and ciliated respiratory epithelium. The latter is the most frequently found and usually a great number of goblet cells is present (Klestadt, 1953; Santora et al., 1970; Karmody and Gallagher, 1972). This variety in histopathologic features may be due to the pluripotential character of embryonic epithelial rests (Bone, 1972). According to most investigators *therapy* consists of surgical removal. The lesion has to be enucleated using an intraoral approach. Recurrence is rare and prognosis is excellent.

DISCUSSION

The patients described here are women and the lesion is unilaterally located. This seems to confirm to the prevalence among females and the tendency towards unilateral manifestation as described in literature. Age distribution in these three patients is as usual too. Clinical features in these cases correspond with data in literature: a fluctuating non-tender swelling in the nasolabial fold extending into the nasal vestibule.

All routine radiographic findings were normal, in patient 2, however, computertomography showed a bony dehiscence in the piriform aperture. In the literature we could not find a description of computertomographic findings in patients with a nasolabial cyst. This method has higher resolutive properties than conventional roentgenography resulting in a more accurate visualization of bony structures around the lesion. The nasolabial cyst is probably able to cause bony erosion in an advanced stage more often than assumed until now. However, extension of the cyst into maxillary sinus as seen in patient 2 remains extraordinary.

In patient 3 echography was performed and proved to be of much value. It demonstrated not only the presence of a fluid-filled cavity, but also showed the exact dimensions of the lesion.

Histopathologic findings varied considerably in these patients: in the lining of the cyst stratified and pseudostratified columnar epithelium, partially ciliated with goblet cells were found, moreover simple squamous, simple cuboidal and stratified squamous epithelium was observed. These histopathologic findings correspond with data described in literature and do not give evidence whether the diagnosis is correct or not. Cytologic findings of the aspirated fluid were not uniform as well. In patient 2 cyst fluid contained cholesterol crystals, which is rather unusual in nasolabial cysts (Walsh-Waring, 1967). Moreover the epithelial lining consisted in this case of stratified squamous epithelium, furthermore dimensions were unusual. Therefore diagnosis is controversial here. The differential diagnosis is some type of odontogenic cyst. However, clinical features of this patient are characteristic for a nasolabial cyst. Histopathologic findings in these patients did not give support to any of the theories of etiology.

Therefore it can be stated that the disorder can be diagnosed almost by clinical symptomatology alone. Pathognomonic symptoms include the presence of a well-confined, smooth, fluctuating swelling in the nasolabial fold. For this reason a descriptive nomenclature is satisfactory. The term nasolabial cyst is to prefer, which was first mentioned by Rao (1955). This topographic diagnosis does not suggest any histopathologic or etiologic entity and allows distinction with other cysts in the maxillary region.

Once a nasolabial cyst is diagnosed it is recommended that therapy consists of surgical removal. The intraoral approach is preferably because the operation leaves no visible scars. The lesion can be removed mainly by blunt dissection, but

the firm attachment to the nasal mucosa, in the patients described, required sharp instrumentation. In advanced cases, like patient 2, with extension into the inferior meatus and the maxillary sinus the incision can be widened and removal of a part of the lateral nasal wall may be necessary. The intraoral approach always provides a sufficient exposure to allow complete removal of the cystic lesion and to bring about healing without any sequellae.

ZUSAMMENFASSUNG

Die Nasolabialzyste wird beschrieben an Hand der Krankengeschichten von 3 Patienten und Angaben in der Literatur. Das klinische Bild ist gekennzeichnet durch eine weiche, fluktuierende Schwellung unter dem Nasenflügel. Dieses Symptom ist weitgehend bestimmend für die Diagnose weil es sich gezeigt hat dass dazu zytologische und histologische Untersuchungsergebnisse nur von untergeordneter Wichtigkeit sind. Die Ätiologie dieser Zyste ist noch immer unsicher. Nach den heutigen Ansichten soll die Theorie der Gesichtspaltengenese, der sich stützt auf das Vorkommen von persistierenden Epithelresten im Bereich der ehemaligen Gesichtsspalten, als wahrscheinlich angenommen werden. Die variierende histologische Befunde sind jedoch nicht in der Lage irgendeine Theorie der Ätiologie zu unterstützen oder zu wiederlegen. Ausdehnung in den knochigen Strukturen des Oberkiefers ist leichter mittels Computertomographie als mit konventionellen röntgenologischen Untersuchungsmethoden fest zu stellen. Die Echographie hat sich herausgestellt als eine präoperative Untersuchungsmethode von grosser Bedeutung. Die Nomenklatur ist verwirrend aber wegen fehlender Uniformität der histologischen Befunde und unsicherer Genese soll der rein topographischer Name Nasolabialzyste vorgezogen werden. Die Therapie besteht aus Extirpation vom intraoral und die Prognose ist vorzüglich.

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