

## Bilateral nasolabial cyst: a rare case report and literature review

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### Abstract

Nasolabial cysts (NLCs) are rare, non-odontogenic lesions that typically present as painless swellings in the nasolabial region. Bilateral cases are even more uncommon and can pose diagnostic challenges. A 65-year-old female presented at the Department of Otorhinolaryngology, Kütahya Health Sciences University Evliya Çelebi Training and Research Hospital, Turkey, in December 2021, with nasal congestion and intraoral swelling persisting for four months. Imaging revealed bilateral cystic lesions extending from the nasal base to the maxillary incisors. The cysts were completely enucleated via an intraoral sublabial approach under general anaesthesia. Histopathology confirmed the diagnosis of bilateral infected NLC. The patient recovered uneventfully, with no recurrence or complications observed over a two-year follow-up period. Early diagnosis and appropriate surgical management of NLCs ensure favourable outcomes with minimal morbidity.

**Keywords:** Bilateral nasolabial cyst, Maxillary cyst, Non-odontogenic cyst.

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### Introduction

The nasolabial cyst (NLC)—also known as the nasoalveolar cyst, Klestadt's cyst, fissural cyst, nasal vestibular cyst, and nasal wing cyst—is a relatively rare cystic lesion that is considered benign and non-odontogenic and is typically located in the anterior maxillary region.<sup>1</sup> It was first described by Zuckerkandl in 1882.<sup>2</sup> NLC is more commonly seen in women than men, and it often occurs in adults during the fourth and fifth decades of life.<sup>3</sup> Patients with NLC often present with a swelling near the nose, specifically in the nasolabial region. This swelling can lead to deformity and may

extend into the nasal or oral cavity. As a result, individuals may experience symptoms such as nasal obstruction, a feeling of pressure, and overall facial discomfort. In some cases, when the cyst becomes infected, patients can complain of pain in the affected area. The infection may lead to additional symptoms such as redness, warmth, and tenderness around the cyst.<sup>2</sup> The diagnosis of NLC involves a combination of clinical, radiological, and histopathological assessments. Radiological diagnosis includes radiography, ultrasonography, computed tomography (CT), and magnetic resonance imaging (MRI).<sup>4</sup> This article aims to discuss the clinical and radiological findings, and treatment methods of bilateral NLC, which is a rare case in the light of the literature.

### Case Report

A 65-year-old female presented to the Department of Otorhinolaryngology, Kütahya Health Sciences University Evliya Çelebi Training and Research Hospital, Turkey, in December 2021, with complaints of nasal congestion and swelling in the oral cavity that had been present for four months. Physical examination revealed two swellings on the floor of the bilateral nasal vestibules, with the right side being more prominent. Additionally, the bilateral nasolabial folds had disappeared, and fluctuation of the cysts was palpated over the incisive teeth in the oral cavity. Both CT and MRI were utilised, revealing two cystic lesions, 26mm in diameter on the right and 16mm in diameter on the left (Figure 1). These lesions extended from the nasal base to the roots of the incisive teeth. Both lesions were hyper-intense and non-contrast-enhancing in T1 and T2 series on MRI (Figure 1). Following the patient's informed consent, intraoral sublabial approach was planned under general anaesthesia. The procedure aimed for the complete enucleation of the cyst. A semilunar incision was carefully made parallel to the sulcus, just below the keratinised gingiva, extending from the right maxillary lateral incisor to the left lateral incisor. Blunt dissection was used to separate the soft tissues from the cyst epithelium, ensuring that the cyst lining remained intact throughout the procedure (Figure 2).

Histopathological evaluation of the excised tissue demonstrated inflamed polypoid mucosal structures, supporting the diagnosis of bilateral infected NLC. Microscopic examination identified a cyst lined with pseudostratified squamous epithelium, containing

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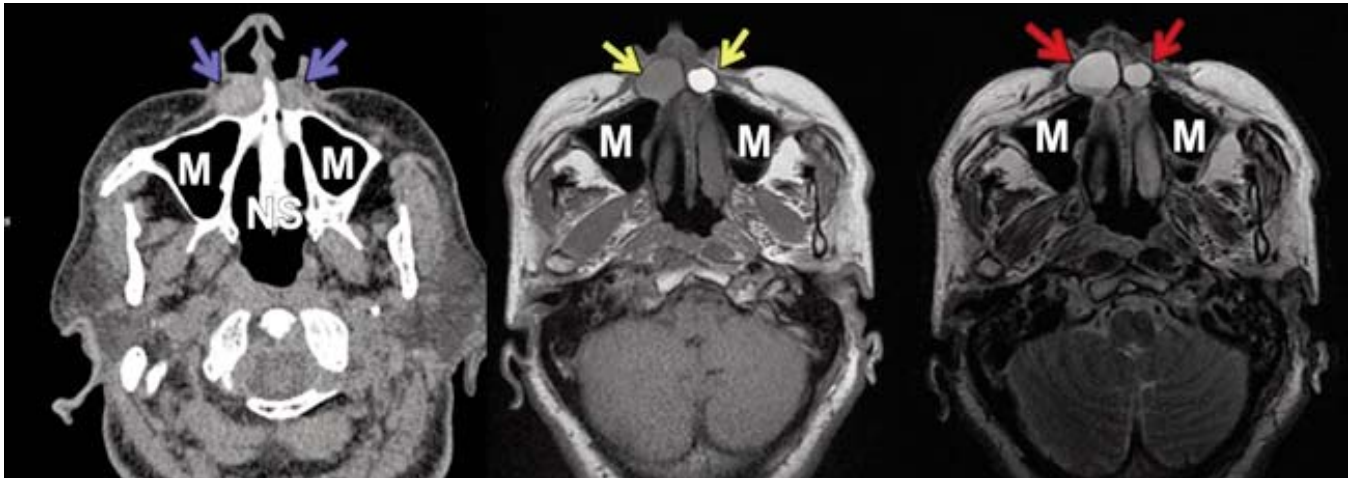
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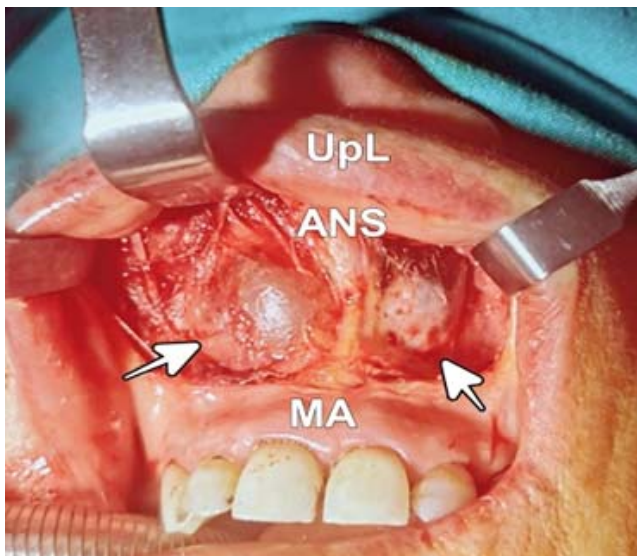
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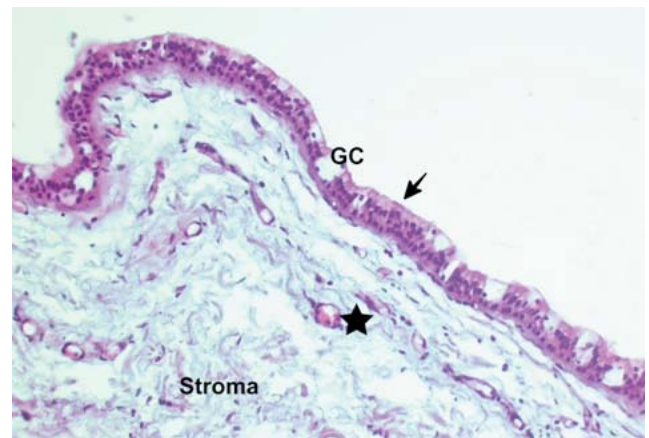
**Figure-1:** Axial section of CT image of cysts (blue arrow) and T1-T2 series MRI image of cysts (yellow and red arrow) (M: maxillary sinus, NS: nasal septum).



**Figure-2:** Anatomical landmarks and surgical steps of the intraoral sublabial approach for NLC excision. (I) Pre-operative illustration showing the anatomical location of the NLC in relation to key structures, including the anterior nasal spina (ANS), maxillary alveolus (MA) and upper lip (UpL). (II) Initial mucosal incision: A semilunar incision is made parallel to the gingivolabial sulcus, extending from the lateral incisor of one side to the contralateral lateral incisor. (III) Blunt and sharp dissection: the cyst is carefully separated from the surrounding soft tissues using blunt dissection, ensuring the preservation of the cyst wall to prevent rupture (white arrow). (IV) Complete enucleation: the cyst is removed in its entirety while minimising trauma to adjacent structures. (V) Closure: the incision is sutured primarily using absorbable sutures, ensuring proper healing and minimal postoperative discomfort.

abundant cilia and mucin-secreting goblet cells. The cyst wall was composed of fibrous tissue with mild infiltration of polymorphic chronic inflammatory cells and congested blood vessels. No granulomatous formations or malignant features were detected (Figure 3).

During the two-year follow-up, no recurrence was



**Figure-3:** Microscopic view of the excised specimen. The cyst is lined with pseudostratified squamous epithelium (black arrow) containing mucin-secreting goblet cells (GC). The capsule consists of fibrous tissue with mild infiltration of polymorphic chronic inflammatory cells and congested blood vessels (black star).

observed. The patient experienced minimal post-operative discomfort and pain levels did not exceed a visual analog scale (VAS) score of 2 after the first week. Facial swelling gradually decreased and resolved completely within two weeks. No impairment of speech, chewing or lip mobility was reported. Sensory examination revealed no paraesthesia, numbness or other signs of infraorbital nerve involvement. Furthermore, there were no post-operative complications of infection, wound dehiscence or prolonged healing. The patient followed the post-operative care instructions, including prescribed analgesics and oral hygiene recommendations. At follow-up visits, she expressed satisfaction with the cosmetic and functional results and reported no visible deformity, scarring, or asymmetry in the perioral region.

**Table-1:** Differential Diagnosis of Nasolabial Cyst: Key Clinical and Radiographic Features.

| Differential Diagnosis                      | Localisation                              | Radiographic Findings                                    | Clinical Features                                     | Distinguishing Factors                                 |
|---|---|--|---|--|
| Odontogenic Cysts (Periapical, Dentigerous) | Intraosseous, related to teeth            | Well-defined radiolucency, may have cortical perforation | Often associated with non-vital teeth, swelling, pain | Vitality testing: Adjacent teeth are usually non-vital |
| Nasopalatine Duct Cyst                      | Midline of anterior maxilla               | Heart-shaped radiolucency between central incisors       | Painless swelling in the anterior palate              | Located in midline, intraosseous                       |
| Globulomaxillary Cyst                       | Between lateral incisor and canine        | Well-defined radiolucency                                | Asymptomatic, may cause divergence of roots           | Intraosseous, located outside the midline              |
| Dermoid/Epidermoid Cyst                     | Subcutaneous, floor of the mouth, maxilla | No osseous involvement                                   | Soft, slow-growing mass, yellowish colour             | More common in childhood, mucosa appears yellowish     |
| Sebaceous Cyst                              | Skin, subcutaneous tissues                | No osseous involvement                                   | Mobile, fluctuant nodule, may have central punctum    | Located in dermis, often with sebaceous material       |
| Benign/Malignant Salivary Tumours           | Palate, upper lip, buccal mucosa          | Variable, may show bone invasion                         | Slow-growing, firm mass, possible ulceration          | May be associated with salivary gland dysfunction      |
| Nasal Furunculosis                          | Nasal vestibule                           | No specific radiographic changes                         | Painful, erythematous swelling, possible abscess      | Associated with infection, often presents with fever   |
| Acute Maxillary Sinusitis                   | Maxillary sinus                           | Mucosal thickening, sinus opacification                  | Facial pain, pressure, nasal congestion               | Sinus-related symptoms, may have purulent discharge    |

## Discussion

NLC is classified as a non-odontogenic jaw cyst and represents a relatively small percentage of all jaw cysts. Specifically, it covers approximately 0.7% of all jaw cysts.<sup>1</sup> These cysts are typically seen in the soft tissues around the nasal alar region and are considered benign and non-aggressive.<sup>4</sup> The prevalence of NLC is reported to be higher in Asian populations.<sup>3</sup> Additionally, these cysts are typically unilateral. However, there are rare cases where nasolabial cysts can be bilateral, occurring on both sides of the nasal alar region.<sup>3-5</sup> It is estimated that approximately 10% of NLC cases may present bilaterally.<sup>5,6</sup>

There are two main theories for the pathogenesis of NLC. According to one theory, cysts are thought to arise from embryonic nasal respiratory epithelium remaining in a facial cleft formed by the fusion of the maxillary, medial, and lateral nasal processes. Another theory assumes that the cysts may arise from the misplaced epithelium of the nasolacrimal duct, as their location and histological findings are the same.<sup>1</sup> In histopathology, stratified or pseudostratified epithelial tissue consisting of goblet cells and cuboidal and columnar epithelial cells is frequently observed. There is no sign of atypia or malignancy.<sup>6</sup>

NLC typically manifests as a painless, round mass located in the soft tissue adjacent to the nasal alar, specifically around the uppermost part of the nasolabial fold.<sup>7</sup> The typical presentation involves a well-localised, fluctuating swelling with a cystic consistency in the nasolabial sulcus. This specific location and the palpable nature of the cyst

contribute to the characteristic clinical findings associated with NLC. The overlying skin is normal, with no drainage. The cysts may potentially expand into various facial structures, affecting areas such as the nasolabial fold, nostril, and labioalveolar sulcus. The consequences of this expansion may include the disappearance of the nasolabial fold, elevation of the nasal area, swelling in the floor of the nasal cavity, and protrusion from the buccal sulcus' mucosa in the lateral upper incisor or canine tooth. If the cyst is asymptomatic, and there is no nasal obstruction, infection, or deformity, i.e. in the absence of certain conditions or complications, its presence may not be noticed.<sup>5-7</sup> In approximately 30% of the patients, the primary reason for seeking medical attention is an infection; according to a case series by Kuriloff, half of the patients in that series developed an infection. The infected cyst can become painful and rupture and drain into the oral cavity or nose. The size of these cysts can reach up to 5cm, and when they reach a large size, they can rarely cause resorption of the underlying bone.<sup>8</sup>

The differential diagnosis and distinguishing features of nasolabial cysts are summarised in Table 1. Odontogenic infection and odontogenic cysts (e.g. periapical, dentigerous cysts) with cortical perforation are among the differential diagnoses for NLC in the nasolabial fold region. However, they are typically tooth-related and intraosseous lesions. Particularly in the context of the diagnosis of NLC, vitality testing can be used to distinguish lesions of odontogenic origin. The vitality testing on adjacent maxillary teeth is expected to be positive. In addition, nasopalatine duct cysts and



globulomaxillary cysts, other differential diagnoses, are intraosseous and have typical localisations. A nasopalatine duct cyst is a lesion in the maxillary anterior midline and the labial or oral region.<sup>8</sup> In the maxillary bone, globulomaxillary cysts are situated outside the midline between the lateral incisors and canines.<sup>9</sup> The NLC should also be differentiated from dermoid and epidermoid cysts of the maxilla. While the colour of mucosa in those cysts is yellow, it is a natural pink or bluish tone in NLC. In addition, epidermoid and dermoid cysts are typically seen in childhood, while NLC is more common in adults.<sup>8</sup> Other differential diagnoses include sebaceous cysts as well as malignant or benign salivary gland tumours.<sup>5</sup> It is less likely that infection spreading from the cysts may cause acute maxillary sinusitis, periodontal abscess, nasal furunculosis, or facial cellulitis.<sup>8</sup>

Oral cavity examination, nasal endoscopy, CT, and MRI are diagnostic methods for NLC.<sup>8</sup> Unless there is bone erosion, NLC typically do not show abnormalities on panoramic and intraoral radiographs. Its location is outside the alveolar bone, and when it reaches large sizes, "cupping" may occur on the underlying cortical plate.<sup>5,6,10</sup> This phenomenon was also observed in the case report by Parwani et al.<sup>10</sup> where a 69-year-old female patient presented with bilateral nasolabial cysts. In this case, occlusal radiography revealed a large scooped-out radiolucent area with 'cupping' surrounding the upper left central incisor in the middle and coronal thirds of its root, but not in the periapical area. This radiographic feature suggests that while NLCs are primarily extraosseous lesions, large cysts may exert pressure on the underlying bone, leading to characteristic radiological findings. Imaging methods (CT and MRI) are used to obtain more detailed information about the cystic structure of these lesions, their relationship with the nasal area and maxillary bone, and bone involvement.<sup>6</sup>

Two main treatment methods have recently been used for NLC: intraoral sublabial excision and transnasal endoscopic marsupialisation. The primary treatment method is intraoral sublabial excision, which is widely preferred due to its effectiveness and minimal risk of recurrence.<sup>5</sup> In the intraoral sublabial approach, the procedure is performed under local or general anaesthesia. A sublabial incision is made in the vestibulum of the maxillary anterior region, ensuring adequate exposure of the cyst. The mucoperiosteal flap is elevated carefully to prevent damage to adjacent structures, including the nasal floor and infraorbital nerve. The cyst is then meticulously dissected from the surrounding tissues and completely excised to minimise

the risk of recurrence. Haemostasis is achieved, and the incision is closed with absorbable sutures. The advantages of this approach include the absence of external scarring, a low recurrence rate, and a relatively straightforward dissection due to the common location of the cyst in the superficial soft tissues. Additionally, post-operative facial swelling and discomfort are generally mild compared to external approaches. However, certain challenges must be considered. Intraoperative bleeding from the rich vascular supply of the area may obscure the surgical field, necessitating careful haemostasis. The proximity to the infraorbital nerve requires delicate handling to avoid transient or permanent sensory disturbances. In large or adherent cysts, meticulous dissection is essential to prevent rupture, which could lead to incomplete excision and recurrence. In a case present in the literature, it was reported that the cysts were successfully removed by intraoral approach in a 30-year-old patient.<sup>5</sup> Similarly, in the present case, bilateral excision was performed using the intraoral sublabial approach, but this patient had larger lesions (26mm and 16mm) and shorter symptom duration. However, in both the cases, there were no post-operative complications and the patients were followed-up satisfactorily.

A study comparing the sublabial approach and the endoscopic transnasal marsupialisation technique concluded that endoscopic marsupialisation resulted in shorter operation time, reduced facial swelling, and lower pain scores.<sup>10</sup> However, another study reported no significant difference in recurrence rates between intraoral sublabial excision and transnasal endoscopic marsupialisation.<sup>3</sup> Studies are arguing that transnasal endoscopic marsupialisation is a simple, effective, less invasive, and easier treatment option, especially for large lesions.<sup>5,7</sup> As an alternative to these techniques, transnasal endoscopic marsupialisation using an Nd:YAG laser under local anaesthesia has been suggested, particularly for patients who prefer an outpatient procedure.<sup>1</sup> Additionally, alternative methods such as cryotherapy, injection of sclerotic agents, cauterisation, aspiration, and incision drainage have been described, but all have been associated with higher recurrence rates, except for complete surgical excision and transnasal endoscopic marsupialisation.<sup>5</sup>

In the current case, bilateral NLCs were successfully excised using the intraoral sublabial approach, with no recurrence observed during the two-year follow-up period. This highlights the efficacy of intraoral excision as a reliable and minimally invasive treatment option for NLCs.

## Conclusion

This case report presents a rare case of bilateral NLC, highlighting its clinical, radiographic, and histopathological characteristics. While NLCs are uncommon, their diagnosis should be considered in the differential diagnosis of anterior maxillary swellings. Advanced imaging techniques, such as MRI or CT, can aid in accurately identifying the lesion and differentiating it from other odontogenic and non-odontogenic pathologies. Early recognition of NLC is crucial to prevent misdiagnosis and unnecessary treatments. Therefore, all dental professionals should be familiar with its features to ensure timely and appropriate management, ultimately improving patient outcomes.

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## AUTHOR'S CONTRIBUTION:

**TCI & EI:** Concept, design, data acquisition, analysis, interpretation, drafting, revision, final approval and agreement to be accountable for all aspects of the work.