

# Occult nasolacrimal duct ectasia in a child with acute sinusitis: clinicopathological considerations and literature review

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**Summary.** *Background:* Enlargement of the nasolacrimal duct (NLD) in the absence of neoplasm is rare. As there are few reports on this condition in the literature, its pathologic significance and proper management remain unclear. We report a case of asymptomatic NLD enlargement incidentally discovered on high resolution computed tomography scan performed for a complicated acute sinusitis. *Methodology:* Case report and literature review. *Results and conclusions:* NLD enlargement, defined as a truly isolated asymptomatic dilatation of the NLD, is rarely encountered. Etiology and clinical significance are unclear although the absence of nasolacrimal pathway or sinonasal disease and radiological findings of bone remodelling suggest that it may be congenital. In such cases, diagnosis may be incidental and based on imaging studies, and the term NLD *ectasia* seems to be more appropriate to describe this distinct entity. NLD enlargement may favour osteomeatal complex obstruction, thus leading to severe acute sinusitis. ([www.actabiomedica.it](http://www.actabiomedica.it))

**Key words:** nasolacrimal duct ectasia, dacryocystorhinostomy, sinusitis, diagnosis

## Introduction

Three-dimensional configuration of nasolacrimal duct (NLD) is highly variable. Differences in size have been reported for age, gender, and race (1). Enlargement of the NLD in the absence of neoplasm is rare. As there are few reports on this condition in the literature, its pathologic significance and proper management remain unclear (2).

We report the case of an 8-year-old boy with a massive enlargement of NLD incidentally discovered during a diagnostic work-up for a rhinogenic orbital cellulitis. The aim of this paper is to discuss the clinical

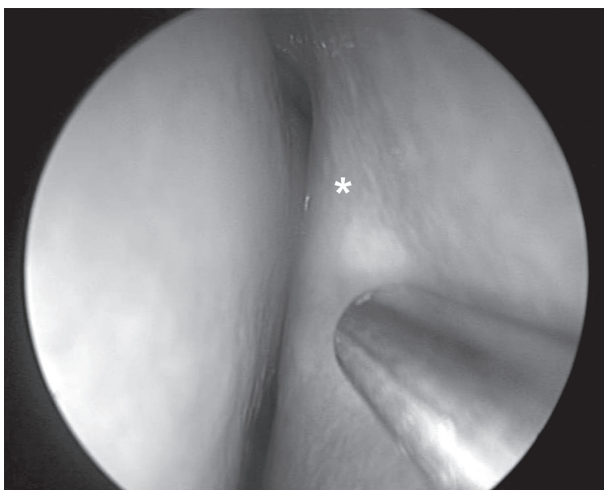
significance of this malformation, and its possible relationship with the onset of the acute sinusitis.

## Case Report

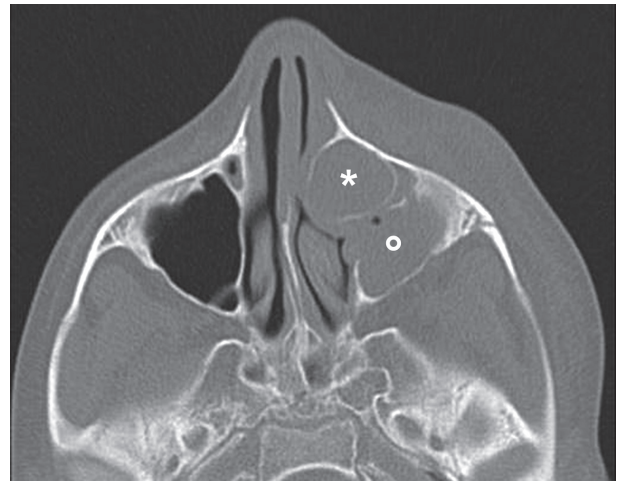
An 8-year-old boy came to our department complaining of left-sided painful zygomatic and orbital swelling, proptosis, and fever. Two weeks before, he had experienced left-sided facial pain during an acute rhinitis that resolved after a short course of oral azithromycin and acetaminophen. His parents denied any history of persistent or recurrent nasal obstruc-

This study was presented in part at the "96° Congresso Nazionale della Società Italiana di Otorinolaringoiatria" (Rimini, 13-16 May, 2009) and won the first prize in the poster session

tion. Diagnostic fiberoptic endoscopy of the left nasal cavity was not feasible because of a large-curved bulging of the lateral nasal wall corresponding to the lacrimal crest, masking the middle meatus (figure 1). A full-course of intravenous ampicillin/sulbactam was started, and a high resolution computed tomography (HRCT) scan was obtained. Imaging revealed a massive opacification of the left the maxillary and anterior ethmoid sinuses. Orbital fat showed marked inflammatory changes, with the ocular bulb displaced laterally and anteriorly without any signs of subperiosteal or intraorbital collection. Besides the inflammatory involvement of paranasal sinuses, HRCT scan showed a massive enlargement of the left-sided NLD, which was filled by hypodense material, showed no erosions of its walls (figure 2). The maximum anteroposterior diameter of the left NLD measured 18.3 mm and the maximum transverse diameter measured 18.6 mm. The anteroposterior diameter of the right NLD measured 7.0 mm and its transverse diameter measured 5.7 mm. Measurements were taken at the level of the infraorbital margin, as suggested by other anatomical studies (1,3). Given these findings, we investigated the anamnestic signs of nasolacrimal obstruction. No history of epiphora, dacryocystitis, or congenital malformation emerged, and a careful examination of the left medial canthal region failed to reveal any mass. The child underwent endoscopic endonasal left dacryocystorhinos-

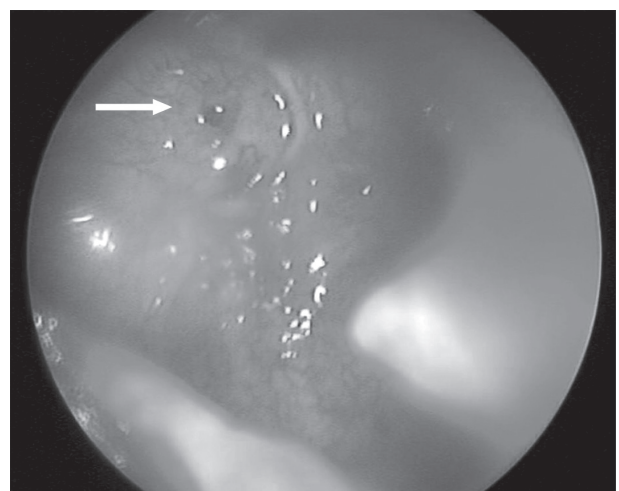


**Figure 1.** Diagnostic fiberoptic endoscopy showing a large-curved bulging (\*) of the left lateral nasal wall corresponding to the lacrimal crest, masking the middle meatus



**Figure 2.** Axial high resolution computed tomography scan showing a massive enlargement of the left-sided NLD filled by hypodense material (\*). Complete opacification of the maxillary sinus is also visible (°)

tomy (DCR). Endoscopic opening of the left NLD through its medial wall showed a dilated NLD covered by flogistic mucosa and filled with mucopurulent secretion; we found the distal orifice of the lacrimal sac at the top of its medial wall (figure 3). Irrigation by probing through both lacrimal puncta documented the patency of the lacrimal pathway. A specimen of exudate was sent for microbiologic studies, and mucosal biopsies were taken. Drainage of sinus infection was achieved by functional endoscopic sinus surgery with



**Figure 3.** Removal of the NLD medial wall revealed a large cavity covered by flogistic mucosa. The distal orifice of the nasolacrimal sac is clearly visible at the top of the medial wall of the cavity (arrow)

left middle meatal antrostomy and anterior ethmoidectomy. Accurate inspection of the inferior meatus failed to detect internal dacryocystocele. The microbiology report showed a growth of *Streptococcus constellatus* and the definitive pathology report on NLD mucosa showed acute inflammation.

Patient's signs and symptoms resolved completely after surgery and he was discharged on the 4th post-operative day. Flexible fiberoptic nasal endoscopy performed two months after surgery showed a complete healing of surgical site.

## Discussion

The case we discuss presents quite a peculiar clinical picture, given the nasolacrimal pathology and the possible relationship between NLD deformity and severe acute sinusitis.

Anatomic and radiological studies have defined the upper normal limit for the anteroposterior diame-

ter of the NLD as 8-10 mm (1,4). Whitnall, and more recently Groessl and Shigeta, conducted anatomic and radiologic studies on the three-dimensional configuration of the NLD (1,3,4). They found a high variability in size depending on age, gender, and race, but did not detect any significant difference in symmetry. According to these authors, the upper limit of normal anteroposterior diameter of the NLD should be considered 8-10 mm. When this limit is exceeded, some authors advocate using the term "enlargement" of the NLD (2,5). In our opinion, whether this condition should be considered an anatomical variant, or is a result of chronic lacrimal drainage obstruction, as suggested by some authors, must still be clarified. When a NLD enlargement is encountered, differential diagnosis includes congenital malformations, dacryocystoceles, and tumours (6). To our knowledge, four cases of NLD enlargement in the absence of tumour have been reported in the literature, the main features of which are summarized in Table 1. The present case shows relevant differences when compared with these

**Table 1.** Review of the Literature

Case	Age	History	Clinical evaluation	CT scan	Diagnosis	Treatment
1 (2)	10	Two episodes of dacryocystitis in the previous 9 months	Soft palpable mass in the medial canthal region with mucopurulent epiphora	Enlargement of the NLD and sac	Acquired dacryocystocele	Lacrimal probing with silicone stenting
2 (2)	37	Mass in the medial canthal region in the previous 2 years, which erupted and disappeared	Mass in the medial canthal region in the absence of lacrimal puncta	Enlargement of the NLD and sac	Acquired dacryocystocele	DCR with lacrimal sac and NLD biopsy
3 (2)	46	Recurrent sinusitis; occasional mild epiphora "all her life"; one previous occasion of a slightly elevated mass in the medial canthal region with mucus reflux lasting a few days	Normal; also the dye disappearance test and lacrimal irrigation were normal	Enlarged NLD; patent maxillary sinus ostium	Idiopathic acquired or congenitally enlarged NLD	Observation and follow-up with CT scan
4 (5)	65	Left-sided epiphora that resolved after an external DCR performed during childhood; chronic sinonasal symptoms such as cheek pressure, nasal congestion and drainage	Normal; patent rhinostomy site	Enlarged NLD; mucosal thickening and air-fluid level in the maxillary sinus	Acquired idiopathic NLD enlargement	Removal of the posterolateral wall of NLD and antrostomy by a combined endoscopic transnasal and endoscope-assisted Caldwell-Luc approach

NLD: nasolacrimal duct; DCR: dacryocystorhinostomy; CT: computed tomography

four cases. There was no history of lacrimal drainage obstruction or nasal sinus disease; inspection of the medial canthal region and inferior meatus failed to detect any signs of lesion suggesting a lacrimal pathway disease; flexible fiberoptic endoscopy of the nasal cavities revealed a large-curved bulging of the lateral nasal wall corresponding to the lacrimal crest in the absence of any other abnormal finding. These considerations suggest that the case we describe may represent an isolated form of NLD enlargement, probably congenital. Thus, in the presence of a truly isolated NLD enlargement, we suggest the term *NLD ectasia*, as it should be considered a distinct entity. As a consequence, cases 1 and 2 should be considered separately. As this type of clinical picture may be completely asymptomatic its diagnosis is generally incidental, suggesting that the incidence of NLD ectasia may be underestimated.

Etiology of NLD enlargement is unclear. Some authors have hypothesized that chronic NLD obstruction may produce enough intraluminal pressure to cause dilation by a bone remodelling process (7). Nevertheless, the case here presented did not show any feature of nasolacrimal drainage obstruction. Moreover, in case n. 3 the dye disappearance test and lacrimal irrigation were normal despite referred occasional mild epiphora. Other authors have suggested that NLD enlargement should be considered an idiopathic acquired condition or a congenital malformation in patients with patent lacrimal drainage (2). In Shloegel's opinion, these two hypotheses can be reconciled by considering that an acquired obstruction may resolve itself shortly after or even before birth, leaving an empty, dilated NLD (5).

As shown in figure 2, NLD enlargement involved the left osteomeatal complex region. This may have contributed to impeding the normal drainage of the maxillary and anterior ethmoidal sinuses. Such a scenario resulted in severe acute sinusitis. In the presence of a sac and/or NLD enlargement with clinical and radiological suspicion of a neoplastic process, biopsy is mandatory. Once malignancy has been ruled out, coexistent symptoms should be investigated. If a nasolacrimal obstruction is diagnosed, lacrimal probing with silicone intubation or DCR with biopsies of the lacrimal mucosa have been proposed.

When an isolated NLD enlargement is detected incidentally in an asymptomatic patient, biopsy or radiologic follow-up with CT imaging at 6 and 12 months have been suggested (2). We emphasize the need to consider the paranasal sinuses involvement as well. The paper presented by Schloegel deals with a NLD *ectasia* causing chronic maxillary sinusitis, whose treatment consisted in the removal of the posterolateral wall of the dilated NLD and wide antrostomy using a combined endoscopic transnasal and endoscope-assisted Caldwell-Luc approach. We hypothesized that our case of NLD enlargement, by obstructing the ostiomeatal complex of the left nasal cavity, favoured the acute sinusitis and orbital cellulitis. The patient underwent a left-sided endoscopic endonasal dacryocystorhinostomy with meatal antrostomy and anterior ethmoidectomy. The aims of this surgical procedure were to treat the infection, to restore the paranasal sinus drainage, and to ensure the patency of the nasolacrimal pathways. Finally, biopsies of the NLD mucosa were taken to rule out a neoplastic process.

## Conclusions

Nasolacrimal duct enlargement, defined as a truly isolated asymptomatic dilation of the NLD, is rarely encountered. Its etiology and clinical significance are unclear. Given the absence of nasolacrimal pathway, sinonasal disease, or radiological findings of bone remodelling suggesting that it may be congenital, we interpret the presence of mucopurulent secretion in the NLD in our patient as a spread of infection from a nearby focus to the dilated but functionally normal NLD. In such cases, diagnosis may be incidental and based on imaging studies, and the term *NLD ectasia* seems to be more appropriate to describe this distinct entity. NLD enlargement may favour osteomeatal complex obstruction, thus leading to severe acute sinusitis. We emphasize that the anatomical relationship between a *NLD ectasia* and crucial regions of the lateral nasal wall should be taken into account in order to provide proper treatment, which appears to be dependent on coexisting signs of nasolacrimal drainage obstruction and sinus disease.

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