CASE REPORT

Rhinology, 41, 41-44, 2003

Bilateral congenital dacryocystocele as a cause of respiratory distress in a newborn*

A. Teymoortash[1], L. Hesse[2], J.A. Werner[1], B.M. Lippert[1]

[1] Department of Otolaryngology, Head and Neck Surgery, Philipps University of Marburg, Germany
[2] Department of Ophtalmology, Philipps University of Marburg, Germany

SUMMARY

Newborns with respiratory distress and nasal obstruction must be examined for congenital dacryocystocele. This disease is caused by a stenosis in the proximal and distal area of the nasolacrimal duct and leads to a cystic dilatation of this duct. A case of a newborn with bilateral dacryocystocele and dyspnoea is presented. The otorhinolaryngologic as well as the paediatric examination could only reveal in the rhinoscopic examination a tumor of the left nasal cavity that partly obstructed the endonasal space. No other pathologic findings were detected. To clarify the origin and the localization of the tumor as well as to exclude an intracranial relation, a magnetic resonance imaging of the middle face and the frontal skull base was performed. After probe and rinsing of the lacrimal ducts the symptoms improved rapidly. In newborns with nasal obstruction a bilateral rhinoscopy of the lower nasal meatus is required to exclude the existence of a dacryocystocele.

Key words: dacryocystocele, congenital, lacrimal duct stenosis, rhinoscopy

INTRODUCTION

In newborns the canalisation of the lacrimal ducts is often not fully developed. Thus the connate obstruction of the lacrimal ducts occurs rather often. About three-quarters of all newborns have a persisting Hasner's membrane at the opening of the nasolacrimal duct into the lower nasal meatus. However, despite the frequency only 5% of the cases develop clinical symptoms. Those symptoms are mucopurulent discharge and epiphora that often regress spontaneously in the first months. Rarely an additional proximal stenosis at the valve of Rosenmüller at the junction of the lacrimal canaliculi and lacrimal sac may lead to a progressing accumulation of the amnion fluid or the lacrimal fluid in the lacrimal duct. This proximal narrowness works like a valve that impedes the reflux of the lacrimal fluid to proximal and allows the passage of the amnion or lacrimal fluid only in one direction (Cassady, 1952; Levy, 1979).

The obstruction of the drain and the secretory accumulation provoke a cystic enlargement of the lacrimal duct by intracanalicular increase of pressure and lead to the formation of a dacryocystocele, also called dacryocele or amniotocele (Goralowna and Tarantowicz, 1979; Harris and DiClementi, 1982). Those congenital mucoceles may protrude into the nasal lumen passing the still soft bony limitation of the lacrimal duct and may lead to a nasal obstruction. Due to the fact that infants are obligate nasal breathers within the first three weeks this may cause dyspnoea. In newborns who suffer from nasal obstruction, congenital dacryocystocele must be considered in the differential diagnosis. In the present study, the case of a newborn with bilateral dacryocystocele and respiratory distress is described.

CASE REPORT

A full-term male infant with uneventful birth and pregnancy anamnesis revealed tachydyspnoea with evidence of nasal obstruction immediately after birth. To exclude a congenital choanal atresia an endoscopic nasal inspection was performed. Anterior rhinoscopy revealed a spheroidal tumor of the left nasal cavity that partly obstructed the endonasal space. This tumor presented as elastic whitish greyish mass. The other paediatric and otorhinolaryngologic specific examinations did not reveal any further pathologic findings.

To clarify this finding a magnetic resonance imaging (MRI) of the middle face and the frontal skull base was performed in order to determine the origin and the localization of the tumor as well as to exclude choanal stenosis. The imaging showed an enlarged nasolacrimal duct, hyperintense on T2-weighted images, and hypointense with liquor signal intensity on T1-weighted images. In the left nasal cavity a cystic structure protruded below the inferior turbinate as extension of the naso-

*Received for publication: May 19, 2003; accepted: September 25, 2003

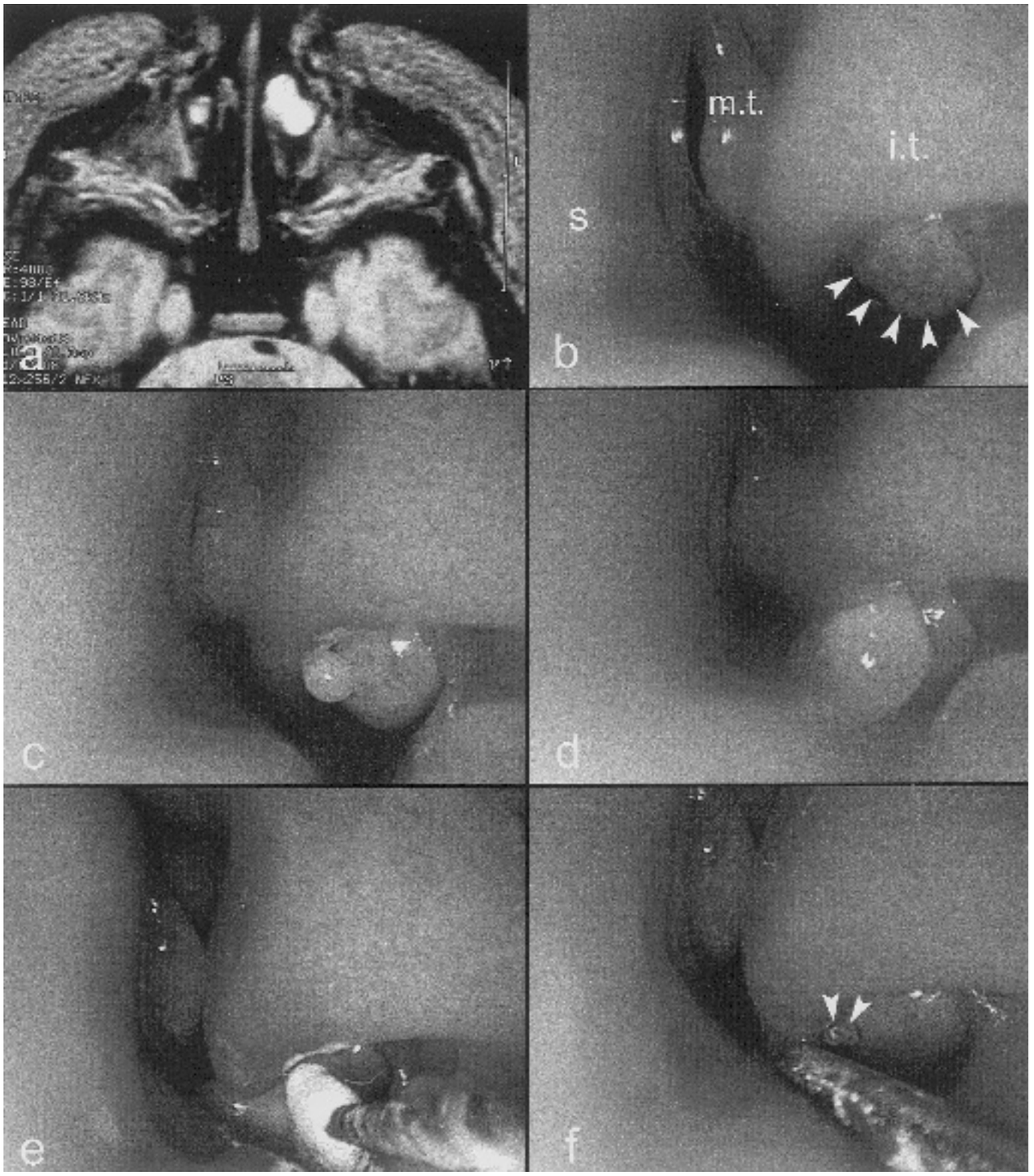

Figure 1. a) axial MRI (T2-weighted) of a bilateral dacroycystocele with intranasal extension with more significant finding on the left side. b) endonasal tumor in the region of the left nasal meatus (arrows). c,d) whitish concentrated secretion from the lacrimal duct after rinsing the lacrimal duct with normal saline. e) smear probe for bacteriologic diagnosis. f) aspiration of secretion and description of the opening of the lacrimal duct in the region of the lower nasal meatus after rinsing (arrows). s=nasal septum, m.t.=middle turbinate, i.t.=inferior turbinate.

lacrimal duct (Figure 1a). On the right side an enlargement of the lacrimal duct was visible. The frontobase and the rhinobase were inconspicuous. There was no sign of choanal atresia.

Rigid nasal endoscopy was then performed under general anaesthesia. After decongestion of the nasal mucosa with xylometazolin 0.025% solution the mucosa was examined with a 30° telescope. First the left inferior turbinate was examined. In the area of the lower nasal meatus a cystic tumor could be detected (Figure 1b). Contralaterally, also a cystic structure could be revealed in the area of the lower nasal meatus, however, significantly smaller, so that initially the imaging could not easily determine the tumor. Choanal atresia could be excluded. Afterwards a probing and rinsing of the lacrimal ducts with normal saline solution was performed. Nasal endoscopy was performed concomitantly with lacrimal duct probing. During rinsing of the left side, a small resistance had to be surmounted which leaded to the drainage of viscous, concentrated secretion from the endonasal tumor (Figure 1b-f). After rinsing the lacrimal duct and suction of the secretion from endonasal tumor, the tumor collapsed. The following irrigations could be performed without any problem and showed a good drainage into the endonasal region. The irrigation of the right side produced only a little amount of concentrated secretion. After the above mentioned intervention the symptoms regressed rapidly. Further treatments were not necessary. After a follow-up of more than three years, no recurrence could be observed.

DISCUSSION

In the sixth fetal month, the nasolacrimal duct develops by canalization of the embryonic epithelial cord resulting of invagination of the ectodermal tissue localized between the maxillary process and the lateral aspect of the nasal process. The development of the draining lacrimal ducts is completed with the opening of the lacrimal points at the lid margins and with the opening of the nasolacrimal duct into the nose (Sevel, 1981). Rarely an obstruction in the upper as well as lower part of the nasolacrimal drainage system may lead to a cystic extension of the lacrimal duct that can already be enlarged prenatally with amnion fluid (Harris and DiClementi, 1982). Already at birth or in the first weeks they can become symptomatic. In the most extended study of 54 patients those cysts occurred bilaterally in 10% of the cases with strong female preponderance. About three-quarters of all cases were female infants (Mansour et al., 1991). Only rarely a relation to syndromes was reported (Sharony et al., 1999).

As in the described case, a dacryocystocele can protrude according to the direction of the lacrimal duct endonasally below the inferior concha and cause a nasal obstruction with dyspnoea especially during feeding (Yee et al., 1994; Hepler et al., 1995). Furthermore a cystic extension of the lacrimal sac may lead to an external swelling below the medial canthal angle. This tumor often occurs bluish-grey and tight (Grin et al., 1991). These cysts cannot be compressed and massaging does not lead to secretion out of the lacrimal points because of the proximal obstruction. Due to unimpeded secretion of lacrimal fluid, epiphora occurs. Generally there is no indication for inflammation. In case of infection a secondary dacryocystitis can develop. Not always all mentioned symptoms can be observed. The external swelling below the medial canthal angle was described in most of the reports in the literature as the main clinical appearance that is often accompanied by an additional cystic part that is endonasally visible (Peloquin et al., 1995). In the present case, the dacryocystocele occurred clinically only as an endonasal tumor. Furthermore, there was a bilateral dacryocystocele that revealed different extents.

In differential diagnosis of infants, the possibility of an endonasal extension of a meningocele or encephaocele must be considered. Furthermore angiomas, dermoids and neoplasms of the lacrimal duct have to be excluded. In case of nasal obstruction without indication of a nasal tumor, a choanal atresia or rhinitis must be considered in newborns (Harris and DiClementi, 1982; Hepler et al., 1995). The mentioned diseases are separate entities with different prognoses and require different treatment procedures. So an exact diagnosis is of high importance.

The ultrasound examination allows a good topographic description of the lacrimal sac, of the nasolacrimal duct as well as neighbour structures (D'Addario et al., 2001). A hypoechoic cystic extension of the draining lacrimal ducts can be observed sonographically in dacryocystoceles. Further radiological examinations to clarify this disease are dacryocystography, MRI and computed tomographic (CT) scans that are to be recommended to determine the extent and to exclude intracranial relations. MRI is the method of choice for description of the soft parts as well as possible relations with the central nervous system before biopsy and planning of surgical interventions. Furthermore, MRI allows a better diffentiation between mucocele and a solid tumor (Rubin et al., 1994). In the CT scan, bony structures can be better revealed and a choanal atresia is easier to detect (Rand et al., 1989).

Spontaneous healing may be expected in some cases by treating the patient conservatively with warm compresses and decompression by massages as well as topic antibiotic ointments and decongestive nasal drops (O'Keefe et al., 1994; Schnall and Christian, 1996). In case of breathing distress and inflammation a surgical intervention becomes necessary. A distinct nasal obstruction in infants caused by dacryocystocele should be considered as an urgent case like choanal atresia since infants are obligate nasal breathers. In most of the cases, as also in the presented one, probing and rinsing of the lacrimal duct lead to a symptom free state. This requires open-

ing of the draining lacrimal ducts because of the persisting distal stenosis to the nose. In other cases an endonasal surgical marsupialisation should be performed (Roy et al., 2002).

REFERENCES

1. Cassady JV (1952) Developmental anatomy of the nasolacrimal duct. Arch Ophtalmol 47: 141-158.
2. D'Addario V, Pinto V, Anfossi A, Del Bianco A, Cantatore F (2001) Antenatal sonographic diagnosis of dacryocystocele. Acta Ophthalmol Scand 79: 330-331.
3. Goralowna M, Tarantowicz W (1979) Imperforation of the nas lacrimal duct as a cause of nasal obstruction in the newborn. Rhinology 17: 173-175.
4. Grin TR, Mertz JS, Stass-Isern M (1991) Congenital nasolacrimal duct cysts in dacryocystocele. Ophthalmology 98: 1238-1242.
5. Hepler KM, Woodson GE, Kearns DB (1995) Respiratory distress in the neonate. Sequela of a congenital dacryocystocele. Arch Otolaryngol Head Neck Surg 121: 1423-1425.
6. Harris GJ, DiClementi D (1982) Congenital dacryocystocele. Arch Ophthalmol 100: 1763-1765.
7. Levy NS (1979) Conservative management of congenital amniotocele of the nasolacriminal sac. J Pediatr Ophthalmol Strabismus 16: 254-256.
8. Mansour AM, Cheng KP, Mumma JV, Stager DR, Harris GJ, Patrinely JR, Lavery MA, Wang FM, Steinkuller PG (1991) Congenital dacryocele. A collaborative review. Ophthalmology 98: 1744-1751.
9. O'Keefe M, Shaikh A, Bowell R, Lanigan B (1994) Management of congenital dacryocele. Acta Ophthalmol 72: 122-123.
10. Peloquin L, Arcand P, Abela A (1995) Endonasal dacryocystocele of the newborn. J Otolaryngol 24: 84-86.
11. Rand PK, Ball WS Jr, Kulwin DR (1989) Congenital nasolacrimal mucoceles: CT evaluation. Radiology 173: 691-694.
12. Roy D, Guevara N, Santini J, Castillo L (2002) Endoscopic marsupialization of congenital nasolacrimal duct cyst with dacryocoele. Clin Otolaryngol 27: 167-170.
13. Rubin PA, Bilyk JR, Shore JW, Sutula FC, Cheng HM (1994) Magnetic resonance imaging of the lacrimal drainage system. Ophthalmology 101: 235-243.
14. Schnall BM, Christian CJ (1996) Conservative treatment of congenital dacryocele. J Pediatr Ophthalmol Strabismus 33: 219-222.
15. Sevel D (1981) Development and congenital abnormalities of the nasolacrimal apparatus. J Pediatr Ophthalmol Strabismus 18: 13-19.
16. Sharony R, Raz J, Aviram R, Cohen I, Beyth Y, Tepper R (1999) Prenatal diagnosis of dacryocystocele: a possible marker for syndromes. Ultrasound Obstet Gynecol 14: 71-73.
17. Yee SW, Seibert RW, Bower CM, Glasier CM (1994) Congenital nasolacrimal duct mucocele: a cause of respiratory distress. Int J Pediatr Otorhinolaryngol 29: 151-158.

Dr. Afshin Teymoortash
Department of Otolaryngology, Head and Neck Surgery
Philipps University
Deutschhausstr. 3
35037 Marburg
Germany

Tel: +49-6421-286 6478
Fax: +49-6421-286 6367

E-mail : teymoort@mailer.uni-marburg.de
Website: www.ent-marburg.de

SOCIETY NEWS

FELLOWSHIPS IN RHINOLOGY

Sponsored by

THE EUROPEAN RHINOLOGIC SOCIETY

Available to Trainee-Surgeons from Eastern Europe

One place available on each of the following Courses in 2004-2005

International Course on in Functional-Aesthetic Nasal Surgery
Ulm, Germany, March 8-11 2005
Course Director: Prof.Dr. G. Rettinger
Language: English

International Course on FESS (Basic and Advanced Techniques)
Graz, Austria, July 8-10 2004
Course Director: Prof.Dr. H. Stammberger
Language: English

5th International Course on Reconstructive and Aesthetic Surgery of the Nose and Face "Around the Nose"
Nijmegen, The Netherlands, May 12-14 2004
Language: English

International Course in Functional Corrective Nasal Surgery
Utrecht, The Netherlands, June 8-11 2004
Course Director: Dr. A.F. van Olphen
Language: English

An Endoscopic approach to Rhinosinusitis
London, United Kingdom, October 20-23 2004
Course Directors: Prof. V.J. Lund and I.S. Mackay
Language: English

Sponsorship includes both registration fees and costs of accommodation. Application forms are available from the Secretary of the ERS. Applications should be submitted at least three months before the course starts. Applications and/or queries should be directed to either the President or the Secretary of the European Rhinologic Society.

Prof. dr. G. Rettinger (President ERS), Department of Otorhinolaryngology, University of Ulm, Prittwitzstrasse 43 D89075 Ulm, Germany

Dr. K.J.A.O. Ingels (Secretary ERS), Department of Otorhinolaryngology, University Hospital "St. Radboud", Geert Grooteplein Zuid 18, NL-6525 GA Nijmegen, The Netherlands