Acquired lacrimal drainage apparatus obstruction in children

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BACKGROUND Pediatric acquired lacrimal drainage apparatus (LDA) obstruction is much rarer than congenital LDA obstruction. Its etiology and treatment outcomes have not been well defined. Our aim was to examine the etiology and management of acquired LDA obstruction in children and report the results of its management.

METHODS We retrospectively reviewed the medical records of patients ≤16 years of age who presented with acquired epiphora to investigate the causes and describe the management of this condition.

RESULTS A total of 31 patients (16 males [52%]) were included. Mean age of patients was 10.9 years (range, 3-16). The main causes of acquired LDA obstruction were keratoconjunctivitis, herpes simplex blepharokeratoconjunctivitis, and trauma. Silicone tube intubation, endonasal or external dacryocystorhinostomy, and the insertion of lacrimal bypass tubes were the mainstays of management.

CONCLUSIONS It is important to suspect acquired LDA obstruction in children with acquired, persistent epiphora. Surgical management is similar to that in adults. (J AAPOS 2019;23:217.e1-5)

Congenital lacrimal drainage apparatus (LDA) obstruction with symptomatic, defective drainage occurs in up to 20% of children during the first year of life.[1] Acquired LDA obstruction in children, however, is rare. A population-based study in Olmsted County, Minnesota, found only 11 cases of acquired LDA obstruction in a population of 409,635 children aged 5-14 years from 1976 to 2000.[2] The etiology of acquired LDA obstruction has not been well defined. We aimed to establish the relative frequency of the various causes of acquired LDA obstruction in children and to analyze its clinical characteristics. Where possible we speculate on mechanisms leading to the condition. We outline the management and outcome of acquired LDA obstruction in children.

Subjects and Methods

Ethical approval for this study was obtained from the research and ethics committee of the Royal Victorian Eye and Ear Hospital, East Melbourne. We retrospectively reviewed the medical records of patients ≤16 years of age who presented with acquired epiphora at the practices of JE, TH, and AM from January 1990 to May 2018. Patients with reflex lacrimation secondary to acute ocular surface conditions were excluded. Data collected included age at presentation, age at the onset of symptoms, and sex. Pertinent features of the history and the findings of the examinations of eyelids, conjunctivae, and corneas were noted. LDA obstructions were classified according to anatomical location, as determined by the clinical examination or examination under anesthesia.

Possible etiologies of acquired LDA obstruction were identified, including herpes simplex keratoconjunctivitis,[3] and previous keratoconjunctivitis, where the diagnosis depended on the findings of symblepharon, diffuse corneal scars, or several round subepithelial corneal opacities. In some patients, noncausative factors could be identified.

Surgery was undertaken when deemed appropriate in accordance with the location of the LDA abnormality. Outcomes were determined on the basis of subjective symptoms and/or objective findings at clinical examination, as outlined previously by Kay and colleagues[4] and described in Table 1. Outcomes were based on symptoms *and* examination findings where possible. The outcome was recorded as that least desirable in those cases wherein the symptoms recorded did not equate with the anatomical findings or vice versa, that is, the outcome was recorded as poor if the patient had persistent epiphora even if patency of the nasolacrimal system was demonstrated at irrigation or, conversely, if patency was not demonstrated despite resolution of epiphora.

Results

A total of 31 patients (16 males [52%]) with acquired LDA obstruction were identified. Mean patient age was 10.9 years (range, 3-16 years). Patients were symptomatic with epiphora for a mean duration of 26.1 months prior to presentation (range, 2-108 months). LDA obstruction

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Table 1. The determination of outcome in cases of pediatric acquired lacrimal drainage apparatus obstruction[a]

Outcome	Symptoms	Examination findings
Good	Resolution of epiphora	Normal tear meniscus height ± patent nasolacrimal system at syringing
Fair	Improvement in symptomatic epiphora; ongoing occasional tearing	Periocular discharge and/or partial obstruction of the nasolacrimal system at syringing
Poor	No improvement in symptomatic epiphora	Dacyrocystitis and/or nasolacrimal system obstruction at syringing

[a]Adapted from Kay and colleagues.[4]

was bilateral in 8 patients (26%) and unilateral in 23 (81%); in the latter it was right-sided in 13 patients (42%) and left-sided in 10 (32%).

The leading causes of acquired LDA obstruction were trauma and viral infection, each with 7 cases (23%); the latter group included 3 cases of adenoviral conjunctivitis (43%), 3 of presumed herpes simplex virus infection (43%), and a single case of herpes zoster virus infection (14%). These cases are summarized in Tables 2 and 3. Other causes for LDA obstruction were Centurion syndrome (3 cases [10%]) and congenital duplicated inferior lacrimal puncta (3 cases [10%]). See Table 4. In the patients with congenital duplicate punctum, there had been a clear interval without epiphora between birth and the development of epiphora at 13, 15, and 16 years of age. One patient with acquired epiphora had signs of atopy, and one had had a previous episode of canaliculitis. See Table 5. In 9 cases (29%), the cause of LDA obstruction could not be identified. See Table 6.

Surgical management was undertaken in 25 cases (81%). Silicone tube intubation of the nasolacrimal system, external and/or endoscopic dacryocystorhinostomy with silicone tube intubation of the nasolacrimal system, and the insertion of Lester Jones tubes were the mainstays of management. A total of 34 major operative interventions were undertaken (excluding examinations under anesthesia, simple syringing and probing of the nasolacrimal system, the removal of silicone tubes under general anesthesia, and reinsertion or replacement of Lester Jones tubes). Each patient had, on average, 1.4 procedures. When ancillary procedures are included, 27 patients (87%) had an average of 1.7 procedures each.

Seventeen patients (54.8%) required only one major operative intervention; 7 (23%) required two surgeries. One patient (3%) required three procedures. Five patients had a Lester Jones tube inserted. These were placed at a mean age of 12.4 years (range, 6-16 years). One patient's tube had to be exchanged for that of a different size, which was then replaced on two occasions. The remainder of these patients all had their tube replaced on one occasion.

Outcomes for the management of 26 of the 39 (67%) nasolacrimal systems with acquired obstruction were available. The outcome was good in 17 cases (65%), fair in 6 cases (23%), and poor in 3 cases (12%).

Discussion

There are few reports of pediatric acquired LDA obstruction in the literature. One study reported on 213 cases of congenital LDA obstruction and 45 cases of acquired LDA obstruction (82.6% vs 17.4% of cases).[5] In another, silicone intubation was performed for LDA obstruction in 59 (93.7%) children with congenital LDA obstruction and 4 (6.3%) with acquired LDA obstruction.[6] Both studies focused on the management of LDA obstruction and did not elaborate on its etiologic factors or clinical characteristics.

In our study, many cases of acquired LDA obstruction were related to viral infection. Harley and colleagues[7] reported 4 cases of pediatric acquired LDA obstruction after ocular herpes simplex infection. Another study reported 20 pediatric patients who underwent lacrimal duct surgery for obstruction that developed after viral infection; herpes simplex in 8 cases and varicella zoster in 5 cases.[8]

Epidemic keratoconjunctivitis (EKC) caused by adenovirus is another cause of LDA obstruction in children. Most pediatric cases of acquired LDA obstruction in Japan are caused by EKC.[9] Our findings may reflect the relatively high prevalence of EKC in Australia, where it occurs both epidemically and sporadically throughout the year, regardless of season.[10] It is also possible that some cases of "idiopathic" LDA obstruction described herein were in fact due to viral infection, but this was not readily discernible on the basis of patient history and/or examination.

Herpes simplex virus, varicella zoster virus, adenovirus, and papilloma virus can all cause inflammation in the epithelial lining of the lacrimal drainage system, which may lead to LDA obstruction in children by a variety of mechanisms.[6,7,11,12] In viral conjunctivitis, inflammation extends beyond the mucosal epithelium into the deeper elastic layers of the LDA.[7,12,13] In nonviral conjunctivitis, inflammation does not extend into the elastic layer. Stenosis of the lacrimal system is not observed. Membranes in EKC are fibrinous.[14] Fibrin modifies the adhesive properties of connective tissue cells in conjunctival membranes, so it likely plays a role in conjunctival adhesion formation.[15] Narrow drainage structures like the canaliculi can be expected to be more vulnerable to obstruction if they become inflamed because of their mucosal wall proximity.[16] In the cases of pediatric acquired LDA obstruction that we identified as being caused by viral infection, obstruction beyond the common canaliculus was not identified. Typically, the site of obstruction in patients with LDA obstruction after HSV infection is at the mid-canaliculus.[3]

Table 2. Pediatric acquired lacrimal drainage apparatus obstruction caused by trauma

Patient	Sex	Age, years	Laterality	Duration of symptoms, months	Level of lacrimal obstruction	Management	Outcome
1	M	7	R	7	NLD	External DCR Redo external DCR	—
2	F	11	L	36	NLD	External DCR	Fair
3	F	10	R	—	UL canaliculus @ 5 mm; LL canaliculus @ 5 mm	External DCR + LJT	Good
4	F	16	L	—	NLD	External DCR	Good
5	F	3	R	—	NLD	External DCR + medial canthoplasty	Good
6	F	13	R	48	Lacrimal sac	Endonasal DCR External DCR + LJT	Good
7	F	15	R	2	LL canaliculus common canaliculus	External DCR + LJT	Good

DCR, dacryocystorhinostomy; *LJT*, Lester Jones tube; *LL*, lower eyelid; *NLD*, nasolacrimal duct; *UL*, upper eyelid.

Table 3. Pediatric acquired lacrimal drainage apparatus obstruction caused by viral infection

Patient	Sex	Age, years	Laterality	Duration of symptoms, months	Etiology	Level of lacrimal obstruction	Management	Outcome
8	M	—	R	24	HSV	LL canaliculus @ 8 mm	Observation	—
9	M	16	R	6	HSV	UL canaliculus @ 5 mm LL canaliculus @ 5 mm	Nil undertaken	—
10	F	12	R	3	HSV	UL canaliculus @ 5 mm LL canaliculus @ 5 mm	External DCR + LJT	Good
11	F	11	L	24	VZV	Common canaliculus	Endonasal DCR + repair canaliculus	Good
12	M	10	L	108	EKC	UL punctum LL punctum	Nil undertaken	—
13	M	4	L	42	EKC	Common canaliculus	External DCR	—
14	F	3	L	30	EKC	Patent	S&P	Good

DCR, dacryocystorhinostomy; *EKC*, epidemic keratoconjunctivitis; *LJT*, Lester Jones tube; *LL*, lower eyelid; *S&P*, syringe and probe; *UL*, upper eyelid; *VZV*, Varicella zoster virus.

Table 4. Pediatric acquired lacrimal drainage apparatus obstruction in association with Centurion syndrome (CS) and congenital duplicate lower eyelid puncta

Patient	Sex	Age, years	Laterality	Duration of symptoms, months	Etiology	Additional lacrimal system abnormality	Management	Outcome R/L
15	M	15	Bilat	24	CS	—	Bilat medial canthoplasty	Fair/good
16	M	14	Bilat	12	CS	—	Bilat medial canthoplasty + RPD Bilat DCR	Fair/fair
17	M	10	Bilat	—	CS	—	Bilat division MCT + RPD + punctoplasty	Good/poor
18	M	16	R	12	Congenital duplicate LL punctum	Lacrimal sac diverticulum	Observation	—
19	F	15	R	24	Congenital duplicate LL punctum	Absent upper eyelid punctum and canaliculus	External DCR + MC stent	Good
20	F	13	L	60	Congenital duplicate LL punctum	NLD patent	External DCR + joining LL canaliculi	Poor

Bilat, bilateral; *DCR*, dacryocystorhinostomy; *DCR*, dacryocystorhinostomy; *LL*, lower eyelid; *MC*, monocanalicular stent; *MCT*, medial canthal tendon; *NLD*, nasolacrimal duct; *RPD*, retropunctal diamond; *UL*, upper eyelid.

Table 5. Other causes of pediatric acquired lacrimal drainage apparatus obstruction

Patient	Sex	Age, years	Laterality	Duration of symptoms, months	Etiology	Level of lacrimal obstruction	Management	Outcome R/L
21	F	12	R	18	Canaliculitis	LL canaliculus	Incision and curettage canaliculus	Fair
22	M	14	Bilat	36	Atopy	Right common canaliculus; bilat NLD	Bilat DCR	Poor/poor

Bilat, bilateral; *DCR*, dacryocystorhinostomy; *LJT*, Lester Jones tube; *LL*, lower eyelid; *NLD*, nasolacrimal duct.

In our study, females who acquired LDA obstruction following trauma outnumbered males 6:1. LDA obstruction at the level of the nasolacrimal duct occurred in 4 cases (57%). In all of these cases, there was a history of significant trauma—a motor vehicle accident in 3 of the 4 cases and blunt trauma from a surfboard in the fourth—with facial and/or orbital fractures in all. In 2 cases (29%), LDA obstruction occurred at the level of the canaliculi and in the last of these cases (14%) at the level of the lacrimal sac.

We included 3 patients in this study with congenital duplicated lower eyelid puncta. All 3 patients had associated congenital anomalies of the nasolacrimal system that could account for tearing—a lacrimal sac diverticulum, nasolacrimal duct obstruction, and absence of the upper canaliculus. They did not, however, present with epiphora until later in childhood.[17]

Kakizaki and colleagues[18] proposed a two-compartment model of lacrimal drainage. It is thought that with contraction of Horner's muscle, that is the lacrimal portion of orbicularis oculi, the canaliculi are compressed while the common canaliculus is pulled posteriorly and expanded. With relaxation of Horner's muscle, the common canaliculus is compressed against the anterior limb of the medial canthal tendon while the canaliculi expand and move anterolaterally to absorb fluid from the puncta. The site of origin of the accessory canaliculus in those with two puncta and its relationship to Horner's muscle may determine the direction of tear flow within the accessory canaliculus and, in turn, the effect of its presence on canalicular function and symptoms. An accessory canaliculus that originates from the common canaliculus may, during relaxation of Horner's muscle, transport tears back from the common canaliculus to the tear lake. This may compromise the function of the lower canaliculus to cause epiphora or see the earlier onset of symptoms in the setting of acquired nasolacrimal duct stenosis. Nasolacrimal duct pathology accounting for the onset of epiphora in the setting of compromised canalicular function has been described in the context of agenesis of a single punctum.[19] In the children we describe, it may be that, with time, the combination of an accessory lower eyelid punctum and another congenital nasolacrimal system abnormality saw the development of epiphora.

Centurion syndrome is a unique clinical entity that may manifest, most commonly in males, as epiphora with onset or exacerbation during puberty. The syndrome is

Table 6. Idiopathic paediatric acquired lacrimal drainage apparatus obstruction

Patient	Sex	Age, years	Laterality	Duration of symptoms, months	Level of lacrimal obstruction	Management	Outcome R/L
23	F	5	L	24	—	External DCR + LJT;	Good
24	M	14	B	4	LL punctum NLD	LL punctoplasty DCR + stent	Good/good
25	M	16	B	60	NLD	Endoscopic DCR External DCR	—
26	M	10	L	18	UL canaliculus @10 LL canaliculus @10	External DCR + excision of common canalicular obstruction	Good
27	M	3	R	12	NLD	Tube intubation	Poor
28	M	16	Bilat	40	Distal common canaliculus	External DCR Tube intubation	fair/fair
29	M	10	Bilat	6	Puncta	Tube intubation; LUL and LLL punctoplasty; RLL punctoplasty + MC stent	—/good
30	F	6	R	2	Common canaliculus	External DCR + excision of common canaliculus obstruction	Good
31	F	6	L	24	NLD	—	—

Bilat, bilateral; *DCR*, dacryocystorhinostomy; *LJT*, Lester Jones tube; *LL*, lower eyelid; *LLL*, left lower eyelid; *MC*, monocanalicular; *NLD*, nasolacrimal duct; *UL*, upper eyelid; *RLL*, right lower eyelid.

characterized by the presence of a prominent nasal bridge, an abnormally anterior insertion of the medial canthal tendon with consequent malposition of the lower eyelid away from the globe and displacement of the punctum out of the tear lake.[20] We encountered 3 cases of Centurion syndrome over the period studied, all occurring in males, with onset of epiphora at puberty in 2 of the 3 cases. Outcomes in this group were mixed. This may reflect ongoing debate as to the appropriate surgical management of this condition.[21] Some authors advocate that the condition can be managed with medial canthal tendon release alone[20,21]; others assert that this should be combined with conjunctivoplasty or lower eyelid retractor plication for success, explaining that while medial canthal tendon release restores the lid–globe apposition, it does not see retrodisplacement of the puncta, which requires the other procedure/s for correction.[22,23]

In cases of congenital LDA obstruction, many clinicians prefer to provide surgical management early, in the belief that increasing age decreases cure rate and increases the number and complexity of future procedures required.[24,25] There is thought to be a "complex" type of congenital nasolacrimal duct obstruction, more likely in older children and consisting of a firm stenosis that extends along the length of the nasolacrimal duct. It is thought to be due to prolonged inflammation and fibrosis.[26,27] In our study, in those in whom the cause of LDA obstruction could not be reliably identified, obstructions were variably sited along the length of the nasolacrimal systems. This is consistent with acquired LDA obstruction being different to congenital LDA obstruction. However, as with congenital LDA obstruction, early detection and management of an acquired LDA obstruction may lead to a higher cure rate.[4]

This study has several limitations. First, there may have been a bias in the selection of patients because the study was completed at tertiary referral hospitals and most patients were referred from other clinics. Second, the number of cases in our study was relatively small. Third, the diagnosis of EKC was based solely on "typical" clinical findings and not on serotyping of the viruses that caused the condition. Finally, outcome was reported by the surgeon undertaking the procedure and not by masked observers, as in other reports on the management of pediatric LDA obstruction.[28]

Literature Search

PubMed was searched, without date or language restriction, on June 30, 2018, using the following terms: *acquired*, *lacrimal*, *obstruction* AND *children* AND *pediatric*.

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