# ORIGINAL ARTICLE

# WILEY

# Computed tomography for guidance in the diagnosis and surgical correction of recurrent pediatric acute dacryocystitis

Chengyue Zhang | Yanhui Cui | Qian Wu | Li Li

National Key Discipline of Pediatrics, Ministry of Education, Department of Ophthalmology, Beijing children's hospital, Capital Medical University, National Center for Children's Health, Beijing, China

#### Correspondence

Li Li, Department of Ophthalmology, Beijing children's hospital, Capital Medical University, National Centre for Children's Health, Beijing 100045, China Email: liliyk1@163.com

Received: 17 August, 2018 Accepted: 29 Janurary, 2019

#### ABSTRACT

**Importance:** This is the first retrospective study of the effect of computed tomography (CT) in diagnosis and surgical correction of recurrent pediatric acute dacryocystitis (PAD).

**Objective:** To explore the pathogenesis of recurrent PAD and the impact of CT in guidance of surgical planning.

**Methods:** Medical histories, clinical manifestations, and CT results of 10 patients with recurrent PAD were reviewed. Etiologies and treatment effectiveness were recorded for all patients.

**Results:** CT revealed that three patients had congenital dacryocystocele with lacrimal sac cyst, enlargement of the nasolacrimal canal, and intranasal cyst of affected sides. After regression of local inflammation, marsupialization was performed. CT showed that four patients had PAD secondary to congenital nasolacrimal canal dysplasia; these patients exhibited normal upper portions of the nasolacrimal canals, but had stenotic or atretic middle and terminal segments. After improvement of local inflammation, endonasal dacryocystorhinostomy was performed. Three patients had PAD secondary to congenital lacrimal sac diverticulum; after contrast injection, CT showed that the cysts at the lacrimal sac. After the topical infection was controlled, transcutaneous dacryocystorhinostomy was performed in combination with excision of the lacrimal sac diverticulum. No recurrence of PAD was detected at 6-month follow-up.

**Interpretation:** Causes of PAD include congenital dacryocystocele, congenital lacrimal sac diverticulum, or congenital nasolacrimal canal dysplasia. Marsupialization with endoscope, endonasal dacryocystorhinostomy, and transcutaneous dacryocystorhinostomy constitute distinct surgeries for PAD treatment. CT provides an important diagnostic function and facilitates selection of specific surgical approaches for recurrent PAD.

### **KEYWORDS**

Acute dacryocystitis, Nasolacrimal canal, Computed tomography

# **INTRODUCTION**

Acute dacryocystitis (AD) is a rare, but severe, infectious ocular disease, which is characterized by rapid onset of

erythema, swelling, and tenderness in the area of the lacrimal sac, with or without epiphora or mucopurulent discharge. In some patients with severe AD, systemic manifestations can include the onset of fever.

DOI: 10.1002/ped4.12115

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Pediatric AD (PAD) is a subtype of AD with features that are unique and distinct from those of adults. In a case series describing patients with congenital nasolacrimal duct obstruction, Pollard<sup>1</sup> reported that the incidence of PAD was 2.9%. Without timely treatment, PAD will rapidly develop into lacrimal abscess, orbital cellulitis, and meningitis, which can threaten vision and may lead to death.<sup>2,3</sup> Prompt and effective treatment modalities, based on etiology, are very important to prevent the recurrence and incidence of serious complications as listed above. Congenital nasolacrimal duct obstruction, traumatic obstruction of the nasolacrimal duct, and the presence of a lacrimal foreign body have been reported as major causes of PAD.<sup>2,4-6</sup> Application of systemic antibiotics and lacrimal probing are the primary treatments for PAD.<sup>2,3</sup>

Computed tomography (CT) can precisely delineate the shape, direction, and abnormal structure of the bony nasolacrimal duct; thus, it is the sole examination method that can verify the existence of congenital nasolacrimal canal dysplasia.<sup>7</sup> CT can also indicate the anatomical characteristics of congenital dacryocystocele and congenital lacrimal sac diverticulum.<sup>8,9</sup>

Herein, we retrospectively analyzed 10 patients with recurrent PAD and summarized their etiologies and prognosis after the completion of surgery. Furthermore, we assessed the effect of CT in guidance of diagnosis and management of recurrent PAD. PAD who visited the Department of Ophthalmology at Beijing Children's Hospital from June 2010 to June 2017. The study protocol was in accordance with the Declaration of Helsinki and was approved by the Ethics Committee of Beijing Children's Hospital. Each child's parents or guardian provided written informed consent after detailed counseling on the disease and the purpose of this study.

The patients with PAD included in this analysis showed signs of erythema, swelling, and tenderness at the area of the inner canthus. Some affected eyes exhibited mucopurulent secretions (Figure 1). None of the patients presented with fever or other symptoms; there was no history of trauma among the patients. Routine blood examinations showed leukocytosis in all patients. All patients had histories of AD and had undergone lacrimal probing at least once in other hospitals; they exhibited postoperative recurrence of AD (Table 1). Importantly, patients with a single manifestation of AD were excluded from this study.



# **METHODS**

We performed a retrospective cohort study of patients with

FIGURE 1 Appearance of a patient with pediatric acute dacryocystitis.

TABLE 1 Demographics and clinical characteristics of 10 patients with recurrent pediatric acute dacryocystitis

		<u> </u>			·		
Patient	Gender	Age	Number of episodes <sup>†</sup>	Number of probing	Primary disease <sup>‡</sup>	Pathogen	Medical history
1	Female	15 d	2	1	Dacryocystocele	HI	Bluish mass at inner canthus after birth
2	Female	21 d	2	1	Dacryocystocele	SP	Bluish mass at inner canthus after birth
3	Male	26 d	2	1	Dacryocystocele	SP	Bluish mass at inner canthus after birth
4	Male	12 mo	3	2	Nasolacrimal duct abnormalities	SP	Epiphora after birth
5	Female	16 mo	3	2	Nasolacrimal duct abnormalities	SA	Epiphora after birth
6	Male	22 mo	2	1	Nasolacrimal duct abnormalities	SA	Epiphora after birth
7	Female	24 mo	2	1	Nasolacrimal duct abnormalities	HI	Epiphora after birth
8	Female	8 mo	2	1	Lacrimal sac diverticulum	SA	Inner canthus mass arose& resolved automatically for several times before the first AD
9	Male	42 mo	2	1	Lacrimal sac diverticulum	SP	Inner canthus mass arose& resolved automatically for several times before the first AD
10	Female	20 mo	2	1	Lacrimal sac diverticulum	SP	Inner canthus mass arose& resolved automatically for several times before the first AD

<sup>+</sup> "episodes" refers to episodes of dacryocystitis; <sup>‡</sup>All patients exhibited congenital disease.

HI, Haemophilus influenzae, SA, Staphylococcus aureus; SP, Streptococcus pneumoniae; AD, acute dacryocystitis.

For each patient, we first performed intravenous infusion of broad-spectrum antibiotics to control local inflammation. Lacrimal irrigation, anterior segment examination by slit-lamp microscope, and orbital CT examination were also conducted. For some patients, 48% lipiodol contrast agent was injected into the lacrimal passage before CT examination. During lacrimal irrigation, we ensured adequate outward drainage to remove the purulent material from the lacrimal sac. We collected regurgitated purulent secretions for bacterial culture and drug sensitivity assessments, and further adjusted the antibiotics based on these results. We made the final accurate diagnosis after analysis of clinical characteristics, examination results, and imaging findings.

# RESULTS

Ninety-six children were diagnosed with PAD in the Department of Ophthalmology at Beijing Children's Hospital from June 2010 to June 2017. Ten patients with recurrent PAD (10 eyes; six girls and four boys; numbered for identification here as 1–10), were included in this study (Table 1). Three patients were diagnosed with PAD secondary to congenital dacryocystocele, four with PAD secondary to congenital nasolacrimal canal dysplasia, and three with PAD secondary to congenital lacrimal sac diverticulum. Bacterial pathogens detected by bacterial culture were *Streptococcus pneumoniae* (five patients), *Staphylococcus aureus* (three patients), and *Haemophilus influenzae* (two patients).

Orbital CT scans of patients 1-3 showed a cystic mass

in the affected area of the lacrimal sac, with an enlarged bony nasolacrimal duct on the affected side and a cystic mass in the inferior nasal meatus (Figure 2A–D). The anterior segment of the affected eye was normal. Lacrimal irrigation revealed ipsilateral lacrimal passage obstruction, accompanied by a large volume of purulent discharge. These three patients were diagnosed with PAD secondary to congenital dacryocystocele. The patients first received intravenous infusion of broad-spectrum antibiotics to control local inflammation; then, under general anesthesia, they underwent marsupialization with the aid of a nasal endoscope. To avoid recurrence, the cyst wall of the inferior meatus was thoroughly removed with a microdebrider during the operation.

CT scans of patients 4–7 showed that the upper portion of the nasolacrimal canal was relatively normal, but became considerably stenotic or atretic at the middle and terminal segments (Figure 3A–C). The anterior segment of the affected eye was normal. Lacrimal irrigation revealed impatency of the ipsilateral lacrimal passage, accompanied by a large volume of purulent secretion. The patients were diagnosed with PAD secondary to congenital nasolacrimal canal displasia. After local inflammation was controlled, these patients underwent dacryocystorhinostomy with the aid of a nasal endoscope, under general anesthesia.

Orbital CT scans of patients 8–10 revealed a cystic mass situated at the ipsilateral medial canthus of the affected eye. Injection of contrast agent showed a cystic mass filled with high-density contrast agent; this mass was located near the lacrimal sac, with a connection between

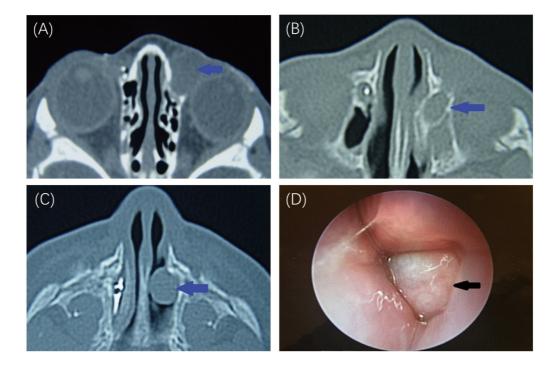


FIGURE 2 Congenital dacryocystocele. (A) Left cyst located at the lacrimal sac area on axial computed tomography (CT). (B) Left lacrimal sac fossa enlargement on axial CT. (C) Left intranasal cyst on axial CT. (D) Intranasal cyst under endoscope.

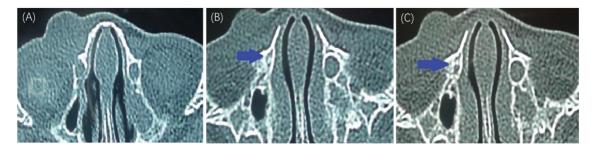


FIGURE 3 Anatomical characteristics of congenital nasolacrimal canal dysplasia. (A) Initial segment of nasolacrimal canal on axial computed tomography (CT). (B) Middle segment of nasolacrimal canal on axial CT. (C) Terminal segment of nasolacrimal canal on axial CT.

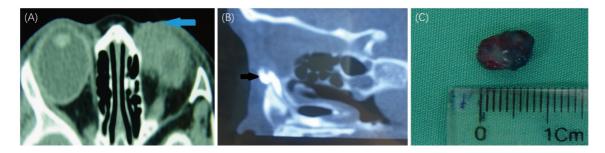


FIGURE 4 Congenital lacrimal sac diverticulum. (A) A mass located at the lacrimal sac area. (B) Mass filled with contrast, connected to the lacrimal sac. (C) Lacrimal sac diverticulum after surgery.

the cystic mass and lacrimal sac (Figure 4A–C). The anterior segment of the affected eye was normal. After pressurized irrigation, lacrimal irrigation revealed that the ipsilateral lacrimal passage was patent, with minimal purulent secretion. These patients were diagnosed with PAD secondary to congenital lacrimal sac diverticulum. After local inflammation was controlled, these patients underwent subcutaneous dacryocystorhinostomy and excision of the lacrimal sac diverticulum under general anesthesia.

Six-month postoperative follow-up assessments revealed that none of the patients in this study experienced any complications or recurrence of AD.

# DISCUSSION

In this study, we retrospectively reviewed the diagnosis and treatment modalities of 10 patients with recurrent PAD. We found three distinct causes of recurrent PAD in these patients: congenital dacryocystocele, congenital nasolacrimal canal dysplasia, and congenital lacrimal sac diverticula. The main pathogens were *S. pneumoniae*, *S. aureus*, and *H. influenzae*, Three patients with PAD secondary to congenital dacryocystocele underwent marsupialization with the aid of a nasal endoscope. Four patients with PAD secondary to congenital nasolacrimal canal dysplasia underwent dacryocystorhinostomy with a nasal endoscope. Three patients with PAD secondary to congenital lacrimal sac diverticulum underwent transcutaneous dacryocystorhinostomy and excision of the lacrimal sac diverticulum.

Congenital dacryocystocele is characterized by the presence of a bluish cystic mass at the inner canthus in newborns; it is caused by obstruction at both ends of the outflow pathway, which results in expansion of the lacrimal sac, enlargement of the nasolacrimal canal, and formation of a nasal cystic mass. Congenital dacryocystocele tends to develop into dacryocystitis or orbital cellulitis, which can be life-threatening.<sup>10</sup> Paysse et al<sup>9</sup> found that, in children, nearly 60% of patients with congenital dacryocystocele eventually developed PAD.

CT is regarded as an important imaging method in the diagnosis of congenital dacryocystocele.<sup>11</sup> Typical manifestations of congenital dacryocystocele on CT include a cystic mass in the lacrimal sac, as well as ipsilateral nasolacrimal duct enlargement, with or without a mass in the inferior nasal meatus. In this study, three patients with AD secondary to congenital dacryocystocele were diagnosed on the basis of patients' clinical and imaging features. However, unlike patients in previous reports, the three patients in the present study exhibited inferior nasal masses, PAD, and previous failure of lacrimal surgery. We suspect that lacrimal surgery failed because the wall of the inferior nasal cystic mass was not completely removed. Moreover, in patients who underwent penetration of the cystic wall during lacrimal probing surgery, the "sticky" inflammatory substances in the cystic mass could not be quickly discharged. The remaining substance likely led to further adhesion and closure of

the probed hole.<sup>12</sup> Therefore, we ultimately performed marsupialization with the aid of a nasal endoscope under general anesthesia. During the operation, we removed the thickened cystic wall with a debrider, in order to prevent recurrence.

Congenital nasolacrimal canal dysplasia is a rare and distinct form of maldeveloped lacrimal duct, which is characterized by bony obstruction at the nasolacrimal canal.<sup>13</sup> Currently, the literature regarding nasolacrimal canal dysplasia is sparse.<sup>14</sup> Zhang et al<sup>7</sup> reviewed CT images of 25 children with congenital nasolacrimal canal dysplasia and summarized its common characteristics as a normal initial segment, with apparent stenosis or atresia of the middle or terminal segments of the nasolacrimal canal. Notably, we could not locate any studies regarding PAD secondary to congenital nasolacrimal duct dysplasia.

A lacrimal sac diverticulum is an outpouching of the lacrimal sac.<sup>15</sup> Patients with lacrimal sac diverticula are typically asymptomatic until the diverticulum becomes sufficiently large to cause tearing, discharge, and other severe symptoms.<sup>8</sup> The communication between a diverticulum and the lacrimal sac may be open, or may serve as a one-way valve. When the communication becomes narrow or occluded, a lacrimal cyst forms.<sup>15</sup> Recurrent AD and orbital cellulitis may also occur as a result of this mechanical blockage.<sup>16</sup> Lacrimal sac diverticula have been reported to lead to AD and orbital cellulitis in adults. However, congenital lacrimal sac diverticulum has not been reported as a cause of PAD.

In the present study, three patients were diagnosed with PAD secondary to lacrimal sac diverticula; these diagnoses were made on the basis of medical history, clinical characteristics, and CT dacryocystography, which clearly showed a cystic mass filled with contrast agent and connected with the normal lacrimal sac. In these three patients, because of the large size of the lacrimal sac diverticula, we performed transcutaneous dacryocystorhinostomy and excision of the lacrimal sac diverticulum, in order to ensure the patency of the lacrimal passage.

CT scanning aids in differentiation of conditions that cause cystic lesion, and can assist clinicians in modifying their surgical approaches.<sup>17</sup> An increasing number of studies include the use of CT to investigate the lacrimal duct anatomy of children and patients with congenital lacrimal duct diseases.<sup>14,18</sup> In the present study, we found that CT scanning and CT dacryocystography aided in the diagnosis of PAD secondary to congenital dacryocystocele, congenital lacrimal diverticulum, or congenital nasolacrimal canal malformation. Furthermore, CT can facilitate selection of the most suitable surgical approach. Considering the potential radiation risk associated with use of CT, we utilized a low-dose CT protocol to minimize adverse effects of it. So Therefore,

we can finally confidently state that the benefits of CT in guiding the diagnosis and surgical correction of recurrent dacryocystitis PAD in children outweigh its risks mentioned above.

In summary, this retrospective review of 10 patients with PAD cases revealed that recurrent PAD could be secondary to congenital dacryocystocele, congenital nasolacrimal canal dysplasia, and congenital lacrimal sac diverticulum. And All the patients in this study achieved a satisfactory resolution of PAD after appropriate lacrimal surgery, guided by use of CT. Therefore, we conclude that CT is an important ancillary clinical examination in diagnosis and selection of specific surgical approaches for patients with PAD.

# **CONFLICT OF INTEREST**

The authors declare that they have no conflicts of interest.

# REFERENCES

- Pollard ZF. Treatment of acute dacrycystitis in neonates. J Pediatr Ophthalmol Strabismus. 1991;28:341-343.
- Campallataro BN, Leuder GT, Tychsen L. Spectrum of pediatric dacryocystitis: medical and surgical management of 54 cases. J Pediatr Ophthalmol Strabismus. 1997;34:143-153.
- Baskin DE, Reddy AK, Chu YI, Coats DK. The timing of antibiotic administration in the management of infant dacryocystitis. J AAPOS. 2008;12:456-459.
- Marr JE, Drake-Lee A, Willshaw HE. Management of childhood epiphora. Br J Ophthalmol. 2005;89:1123-1126.
- Zuberbuhler B, Carifi G, Leatherbarrow B. Acute dacryocystitis in a 2-year old child caused by pantoea. Orbit. 2012;31:13-14.
- 6. Blankstein SS. Dacryocystorhinostomy in infants and children. AMA Arch Ophthalmol. 1952;48:322-327.
- Zhang C, Yu G, Cui Y, Wu Q, Wei W. Anatomical characterization of the nasolacrimal canal based on computed tomography in children with complex congenital nasolacrimal duct obstruction. J Pediatr Ophthalmol Strabismus. 2017;54:238-243.
- Zhang C, Wu Q, Yu J, Cui Y, Cao W, Fan Y, et al. Lacrimal sac diverticulum presenting as a lower eyelid mass with a secreting fistula. Chin Med J (Engl). 2014;127:3359-3360.
- Paysse EA, Coats DK, Bernstein JM, Go C, de Jong AL. Management and complications of congenital dacryocele with concurrent intranasal mucocele. J AAPOS. 2000;4:46-53.
- 10. Becker BB. The treatment of congenital dacryocystocele. Am J Ophthalmol. 2006;142:835-838.
- Meyer JR, Quint DJ, Holmes JM, Wiatrak BJ. Infected congenital mucocele of the nasolacrimal duct. AJNR Am J Neuroradiol. 1993;14:1008-1010.
- Kakizaki H, Takahashi Y, Sa HS, Ichinose A, Iwaki M. Congenital dacryocystocele: comparative findings of dacryoendoscopy and histopathology in a patient. Ophthalmic Plast Reconstr Surg. 2012;28:e85-86.
- Zhang C, Yu G, Wu Q, Cui Y, Zhang Y, Li C, et al. Radiographic observation of nasolacrimal canal in

congenital nasolacrimal canal agenesis. Chin J Ophthalmol. 2014;50:911-914. (In Chinese)

- 14. Coban DT, Beden U, Sonmez B, Elmali M, Erkan D. Unsuccessful probing and nasolacrimal canal agenesis in congenital epiphora. Orbit. 2010;29:363-366.
- Ormrod JN. Diverticulum of the lacrimal sac. Br J Ophthalmol. 1958;42:526-528.
- Akcay EK, Cagil N, Yulek F, Yuksel D, Simsek S. Congenital lacrimal sac diverticulum as a cause of recurrent orbital cellulitis. Can J Ophthalmol. 2009;44:e29-30.
- Epley KD, Karesh JW. Lacrimal sac diverticula associated with a patent lacrimal system. Ophthalmic Plast Reconstr Surg. 1999;15:111-115.
- Zhang C, Wu Q, Cui Y, Yu G. Anatomy of nasolacrimal canal in congenital nasolacrimal duct obstruction -18 cases retrospective study. Acta Ophthalmol. 2015;93:e404-405.

**How to cite this article:** Zhang C, Cui Y, Wu Q, Li L. Computed tomography for guidance in the diagnosis and surgical correction of recurrent pediatric acute dacryocystitis. Pediar Invest. 2019;3:39-44. https://doi.org/10.1002/ped4.12115