Endoscopic dacryocystorhinostomy to treat congenital nasolacrimal canal dysplasia: A retrospective analysis in 40 children

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Research article

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Abstract

Purpose: To investigate the therapeutic effectiveness and safety of endoscopic dacryocystorhinostomy (EN-DCR) to treat congenital nasolacrimal canal dysplasia (CNCD). Methods: Forty children (50 eyes) with congenital nasolacrimal duct obstruction (CNLDO) and lacrimal bony dysplasia, including 8 with bony atresia (10 eyes) and 32 with bony stenosis (40 eyes), were recruited in this retrospective study. Standardized EN-DCR was performed in all cases. The post-operative observations included relief of symptoms, fluorescein dye disappearance test (FDDT), syringing of lacrimal passages and anastomotic patency under nasal endoscopy. Patients were followed up for 8-18 months. Results: The standardized EN-DCR surgery had a success (cure and improvement) rate of 100%, including a cure rate of 82% and an improvement rate of 18%. The cure rate of 40 bony nasolacrimal duct stenosis was 82.5% and 10 bony nasolacrimal duct atresia was 80%. Statistical analysis showed that neither the receipt of other treatments before surgery nor the type of bony nasolacrimal duct dysplasia affected the cure rate. No significant complications were observed during the post-operative follow-up, except for four cases (4 eyes) that suffered middle turbinate and nasal mucosal adhesion and two cases with sinusitis. Conclusions: CNCD is a type of CNLDO that does not respond to conservative and conventional treatment. EN-DCR provides a safe and effective treatment for children with CNCD. It has a high success rate with a low incidence of complications.

Introduction

Congenital nasolacrimal canal dysplasia (CNCD) is a sub-type of the common congenital nasolacrimal duct obstruction (CNLDO).\textsuperscript{1,2} This type of congenital lacrimal duct deformity is induced by a rudimentary or immature bony nasolacrimal duct. Several studies using computed tomography (CT) scans have characterized the abnormal features of CNCD.\textsuperscript{1-5}

Conservative and conventional therapy, such as spontaneous relief, massage, irrigation, lacrimal duct probing, and intubation surgery, are ineffective. In recent years, nasal endoscopic surgical techniques are more frequently used by ophthalmologists and ear-nose-throat physicians to treat lacrimal duct obstruction diseases and dacryocystitis. At present, endoscopic dacryocystorhinostomy (EN-DCR) is generally believed the only effective treatment for CNCD.\textsuperscript{6-8} However, few reports of using EN-DCR to treat CNCD include children.\textsuperscript{9-11} In the present study, we reported the clinical data of 40 cases (50 eyes) of CNCD treated with EN-DCR in our hospital from 2012 to 2016.

Methods

Patients

Forty children (50 eyes) with CNCD, who received EN-DCR in the Department of Ophthalmology, Beijing Children's Hospital, Capital Medical University, from February 2012 to March 2016, were recruited into this study.
The diagnosis of CNCD was based on the history provided by parents, the symptoms of postpartum tear discharge with eye discharge, tests of the patency of the nasolacrimal canal by irrigating the lacrimal sac, and CT examination of the lacrimal duct. All cases completed lacrimal CT angiography within one month before surgery. Following the report of Zhang et al.,¹,² lacrimal bony dysplasia was divided into bone nasolacrimal duct stenosis and bone atresia using CT angiography (Figure 1).

The demographic and clinical information of patients is summarized in Table 1. The inclusion criteria were CNLDO with lacrimal bony dysplasia and age 2-18 years. Exclusion criteria included anomalies of the initial segment of the lacrimal passage, such as absence of the inferior lacrimal puncta, absence or atresia of the inferior canaliculitis, traumatic lacrimal duct obstruction, lacrimal fractures induced by trauma, or lacrimal sac damage or displacement.¹²

The indications for EN-DCR include cases that had been clearly diagnosed as CNCD. This type of case was also suitable for external dacryocystorhinostomy (DCR). In all cases, the parents in this group chose EN-DCR.

**Ethics statement**

This study was a retrospective study, conducted in accordance with the principles of the Helsinki Declaration, and approved by the Institutional Review Committee of Beijing Children's Hospital. Written informed consent was obtained from the parents of all patients before surgical treatment.

**Surgical procedures**

All procedures were performed under general anesthesia. Patients were required to remain supine with the head tilted back 15°. The nasal cavity on the surgery side was shrunk by using 1:10000 adrenaline brain cotton pieces. The endoscopic surgical system (Karl Storz SE and Co. KG, Tuttlingen, German) and 4.0 mm, 0° rigid nasal endoscopy (XiON GmbH, Berlin, German) were utilized. Other surgical equipment included the ENT power system (Medtronic PLC, Dublin, Ireland), 4.0 mm ear burr, ophthalmic devices, including rongeurs of 3.0 mm and 2.0 mm, and the Blasky children mucosal biting clamp.

The EN-DCR procedure was divided into three steps according to the guideline of rigid endoscopy¹³ (Figure 2). In step one, a 2.0 cm × 1.5 cm mucoperiosteal flap is made in the lateral nasal wall using a sickle knife, starting from the front of the uncinate process and located mainly above the middle turbinate axillary and reaching the periosteum. In step two, a bone window is made. As shown in Figure 1a-c, the frontal process of the maxilla is engaged using a rongeur from the suturae lacrimomaxillaris; the front lacrimal bone is separated and clamped, then a bone window is formed with a diameter of about 1 cm × 1.5 cm. If the upper frontal process of the maxilla bone is thick, it can be ground with an electric drill. Then, the medial lacrimal sac wall is exposed, with a light pale blue color. After the lacrimal sac has been exposed, the Bowman probe is placed through the canaliculus into the sac. A vertical incision is then
made with a sickle knife and the medial wall of the sac is removed or the mucosal flap of the lacrimal sac is placed posteriorly, as shown in Figure 1d-f. In step three, the lacrimal sac is filled with vampire gauze or a gelatin sponge, which presses against the lacrimal sac mucosal flap to reduce movement and promote epithelialization of the anastomosis, as well as to attenuate early post-operative bleeding.

**Post-operative treatment and follow up**

Systemic antibiotics and hemostatic were administered for three days after surgery. Saline nasal sprays are recommended for use in the month after surgery. A nasal wash was performed beginning on the fourth day using a nose washing apparatus. The first post-operative follow up was arranged on the seventh day after surgery, when patients were subjected to a lacrimal passage flush. Tobramycin and dexamethasone (diluted 1:10) was used to flush the lacrimal passage from the upper and the lower punctum. In general, the first flush met with significant resistance, and was accompanied by pus discharge and regurgitation. Sustained pressure washing is recommended until flushing is smooth, swallowing confirmed, and regurgitation disappears, or until discharge reflux becomes clear fluid. In the first month after surgery, the lacrimal passage flush was performed weekly, then every two weeks for another two months. Anastomotic exploration was routinely performed in all cases after one month under local anesthesia according to age, anastomotic epithelialization, and timely discovery and cleaning of proliferated granulation tissue around the pore. At 8-18 months after surgery, anastomotic exploration was again performed. Outpatient follow-up studies included taking history, the fluorescein dye disappearance test (FDDT), lacrimal passage flush, and anastomotic exploration under nasal endoscopy in children >6 years. The evaluation of duct drainage function by FDDT was considered a major endpoint (FDDT 0–1: none or a thin fluorescing marginal tear strip persists in the conjunctival sac and the lacrimal drainage function is normal; FDDT 2–3: fluorescein persists in the conjunctival sac and the lacrimal drainage system is obstructed).

**Clinical criteria for outcome of surgery**

Surgical effects were divided into cure, improvement, and invalid, according to clinical criteria. Cure was defined as lacrimal sac pore formation in the lateral nasal wall in front of the middle concha, epithelialization under endoscopic observation, no tearing or pus, FDDT=0, and smooth flush. Improvement was defined as lacrimal sac pore formation in the lateral nasal wall in front of the middle concha, epithelialization under endoscopic observation, symptom relief, FDDT=1, and flush unobstructed or pressure flush unobstructed. Invalid was defined as no relief of symptoms, FDDT=2-3, flush unsmooth or pressurized, and pore atresia. Cure and improvement were considered successful surgery, and surgery efficiency was calculated as the sum of the cure rate and the improvement rate.

**Statistical analysis**
Statistical Package for the Social Sciences (SPSS) software (SPSS Inc. Released 2008. SPSS Statistics for Windows, Version 17.0. Chicago: SPSS Inc.) was used for statistical analysis. The efficacy of preoperative treatment and the effect of type of bony nasolacrimal duct dysplasia on the cure rate were analyzed using the chi-square test, with P<0.05 representing statistical significance.

### Results

Fifty endoscopic DCRs were performed on 40 patients (50 eyes) with CNCD. There were no intraoperative complications. The mean age at surgery was 5.5 years (range 2–14 years). The male to female ratio was 1.9:1 (26:14). Previous interventions included probing in 32% of patients (16/50), massage in 56% (28/50) and intubation in 12% (6/50). The follow-up period was 8-18 months (average 16.5 months). Preoperative CT angiography of the lacrimal passage revealed 8 bony atresia cases (10 eyes) and 32 bony stenosis cases (40 eyes). The demographic information and clinical outcomes of cases are summarized in Table 1.

According to the criteria, the cure rate was 82% and the improvement rate was 18%. Patients in both the nasolacrimal canal stenosis and atresia groups achieved a high effective rate (Table 2). Chi-square analysis showed that the type of nasolacrimal duct dysplasia had no impact on the efficacy of surgical treatment based on the similar cure and improvement rates in both groups ($\chi^2=0.08, P=0.78$). According to Table 3, the chi-square test showed that the cure rate was not affected by treatments performed before EN-DCR ($\chi^2=1.17, P=0.28$).

We explored the stoma one month after surgery in patients who received general or local anesthesia. Among the 40 patients, 6 cases (8 eyes) had no follow up on time, and the other 34 cases (42 eyes) were followed up on time. These had round or oval nasal anastomosis with healed mucosa with a maximum diameter of 6-7 mm (Figure 3). However, visible mucosal thickening and edema remained, with bleeding upon tactile investigation. Among these 42 eyes, 32 had granulation in varying degrees. The early postoperative granulated tissue around the stoma was vulnerable and bled easier, making it accessible for removal using nasal mucosa pliers. After clearing the granulation tissue, bleeding was stopped with a gelatin sponge. Six cases (7 eyes) were followed up later than one month (2-3 months), by which time the mucosal edema was not obvious and the stoma had basically epithelialized. Four eyes had visible granulation tissue, which was surgically removed. Twenty-eight cases (36 eyes) who accepted anastomotic probing under general or local anesthesia were followed up after 8-18 months. The probing results showed that the anastomotic hole was round or oval, with no atresia. The diameter of the anastomotic hole was about 2-5 mm, with a maximum pore diameter of 5 mm (as shown in Figure 3). The mucosa around the anastomosis was well epithelialized and smooth without granulation tissue hyperplasia. Adhesion of the middle turbinate to the lateral wall nasal mucosa found in four cases (4 eyes) was separated through exploratory surgery.

No patients had significant complications during the post-operative follow-up. Four patients complained of occasional headache and were diagnosed with nasosinusitis. Further observations were needed,
because no nasosinusitis was found in these four patients prior to surgery.

**Discussion**

CNCD is a particular type of CNLDO. The vast majority of these cases are initially diagnosed as common CNLDO and are treated conservatively or conventionally, such as with massage, lacrimal duct probing, and intubation surgery. When these treatments prove ineffective, CT examination of the lacrimal duct can reveal CNCD. Prior to the use of lacrimal CT angiography, many children suffered repeated lacrimal probing, which can cause such complications as false passage and punctal tear. CT angiography can help diagnose congenital nasolacrimal duct abnormalities, thereby avoiding this lacrimal tissue damage. Meanwhile, CT angiography of the lacrimal passage can not only accurately determine the location and degree of obstruction, but also help to determine the developmental level of the agger nasi and its anatomic relationship with the lacrimal sac. In addition, it can help elucidate abnormalities in the paranasal sinuses and lacrimal sac, especially valuable information in patients with surgical histories.

We previously performed CT angiography in children with failed lacrimal probing, and found that these children had bony nasolacrimal canal stenosis or atresia. We recommend these children stop receiving lacrimal probing and undergo EN-DCR.

EN-DCR is now a well-accepted technique for managing nasolacrimal duct obstructions, with success rates comparable to those of external DCR. EN-DCR represents a mature technology to treat complex nasal lacrimal duct obstructions in children. In particular, the combination of EN-DCR with lacrimal CT angiography provides advantages over traditional lacrimal surgery. Previous studies have shown success rates for pediatric EN-DCR of 58-100%. Leibovitch et al. reported that the anatomical patency rate was 100% (endoscopic anastomotic opening), but the success rate (complete disappearance of symptoms) was only 92%. In our experience, EN-DCR is not recommended in children younger than two years. For cases with bone dysplasia shown by CT angiography, age is not a key consideration for EN-DCR. These children also have serious dacryocystitis symptoms and the intubation success rate is very low. Also, repeated intubation or probing can easily cause false passage and infection. If EN-DCR must be performed in very young children, surgical skills and equipment must be optimal.

For children with lacrimal duct obstruction, the initial EN-DCR can not only avoid skin scarring, but also protect the exhaust pump function of the orbicularis muscle and the medial canthal ligament. In addition, surgery through the nose significantly reduces the incidence of post-operative complications and increases patient compliance. We believe that EN-DCR has significant advantages over external DCR. Although most studies have shown comparable long-term efficacy for transnasal and external surgeries, EN-DCR does not affect appearance. Moreover, the improved EN-DCR is more concise, except for complicated cases such as those with traumatic dacryocystitis. In this study, implantation of a lacrimal duct diverging tube was not necessary, avoiding a second surgery to remove the tube. In adults and children over 6 years of age, the lacrimal sac can be filled to an appropriate size using a conical expansive sponge introduced through the stoma; however, the sponge must be removed one week after
implantation. For children, this procedure must be performed under general anesthesia, which some parents may not find acceptable. With its use of sophisticated endoscopic equipment and intranasal devices, EN-DCR may obtain a clearer operating field, with fewer injuries, little bleeding and shorter operation time than external DCR. Such advantages are important for children who need general anesthesia. Post-operatively, external operations require nasal packing, daily flush washing early in the post-operative stages, wound care, and stitches, with an increased chance of infection. Transnasal surgery requires simpler post-operative treatment. However, it is necessary to perform intranasal anastomotic exploration in young children at an early post-operative stage (one month), which is usually performed under general anesthesia. We explored the stoma under general anesthesia using sevoflurane and treated tissues with granulation in young children.

The narrow anatomical space in the nasal airway of children represents a challenge in performing EN-DCR, demanding a high level of surgical skill. High-definition endoscopic equipment, sophisticated intranasal devices, and accurate lacrimal positioning technology together enhance the success of this operation. In terms of equipment, the 0° and 30° diameter of 4.0 mm and 2.7 mm sinus endoscope is needed. A 4.0 mm endoscope is recommended, as it provides better lighting and a wider field of view. Because of the small anatomical space of young children, a 2.7 mm endoscope as well as nasal trumpet are recommended. However, some scholars hold that the 4.0 mm endoscope can be used in children younger than one year, because it can lead to better lighting and a wider viewing angle; however, results depend on the endoscopic surgeon's skill level, personal preferences, and device conditions. In this study, the youngest child was 2 years old and a 4.0 mm endoscope was successfully used, which had better lighting and a wider visual field than a 2.7 mm endoscope. The medial wall of the lacrimal sac may become thick due to inflammation, so we recommend using a corneal puncture with a 15° blade; however, some surgeons may be more accustomed to using a sharp sickle knife, myringotome, high-frequency electric knife, or laser. Experts agree that successful outcomes in pediatric DCRs require adequately sized and positioned osteotomy, full-length sac marsupialization, and a 360° mucosa-to-mucosa approximation to facilitate healing.

A key factor for a successful operation is maintenance of the patency of the bone hole. The size of the bone hole is critical to prevent bone hole closure. On the one hand, a small bone window limits the size of the lacrimal sac flap which, if too small, prevents the effective draining of tears. A large bone window is recommended when exposing the inner lacrimal wall, which will improve the opening rate of the pore. On the other hand, a small bone window is susceptible to blockage by a post-operative blood scab, exudate, or granulation tissue, causing a failed operation. If the bone window is too large, the damage to surrounding tissue is severe. In addition, lack of support for the floating sac can lead to lacrimal sac flap retraction and an unsatisfactory surgical result.

The use of lacrimal stents in pediatric cases is controversial. Some recommend placing a lacrimal silicone tube in all EN-DCR patients for four weeks to expand the tear duct and sac opening instead of allowing the lacrimal sac to remain open. Zhou et al. reported that, instead of an expansion tube, an
appropriately-sized conical expansion sponge can be placed in the lacrimal sac through the stoma to expand the stoma. For reoperation cases, as well as those with small lacrimal sac and lacrimal stenosis, an indwelling silicone tube is necessary. We concluded that, in bony nasolacrimal duct stenosis or atresia cases, the lacrimal sac and the surrounding bones develop normally or even expand slightly, so that the lacrimal bone hole fits easily with the nasal mucosal flap and nasal mucosa. If there is no canalicular lesion, it is not necessary to place any anastomotic support. Ensuring a good nasal mucosal flap and lacrimal sac are important for the success of the operation. At the same time, we can also use vampire gauze or gelatin sponge as a short-term support in the anastomosis of the lacrimal sac, to press the nasal mucosa and lacrimal mucosa together and stop bleeding. When the vampire gauze or gelatin sponge is absorbed, anastomotic mucosal healing has been largely completed. Therefore, the decision to place a lacrimal duct support depends on the child's condition. An important prerequisite to ensuring the success of surgery is superb endoscopic sinus surgery techniques and familiarity with the sinonasal anatomy and the structural relationships around the eyes. In this study, none of the cases underwent lacrimal duct dilatation or catheter tube placement.

**Limitations**

The limitations of the current study include its retrospective and single center design. The limited sample size prevented group comparison.

**Conclusions**

In summary, CNCD is a type of CNLDO that does not respond to conservative and conventional treatment. EN-DCR is a safe and effective treatment for children with CNCD. It has a high success rate and a low incidence of complications. Multi-center studies should be performed to confirm these conclusions.

**Abbreviations**

CT\textsuperscript{\textregistered}computed tomography\textsuperscript{\textregistered}

EN-DCR\textsuperscript{\textregistered}endoscopic dacryocystorhinostomy\textsuperscript{\textregistered}

CNCD congenital nasolacrimal canal dysplasia\textsuperscript{\textregistered}

CNLDO congenital nasolacrimal duct obstruction\textsuperscript{\textregistered}

FDDT\textsuperscript{\textregistered}fluorescein dye disappearance test\textsuperscript{\textregistered}

SPSS\textsuperscript{\textregistered}Statistical Package for the Social Sciences\textsuperscript{\textregistered}

DCR dacryocystorhinostomy
Declarations

Ethics approval and consent to participate

This retrospective case series was approved by the ethics committee at Beijing Children's Hospital and conducted in accordance with the principles of the Helsinki Declaration, and informed consent was obtained from all parents of the patients before surgical treatment.

Consent for publication

Not applicable

Availability of data and material

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Competing interests

The authors declare that there is no conflict of interest.

Contributions of Authors

Dr. Cui carried out the endoscopic DCR surgery and drafted this manuscript. Dr. Zhang participated in the design of the study and performed the data analysis. Dr. Liu collected important background information. Dr. Wu drafted and revised the manuscript. Dr. Yu and Dr. Li participated in experimental design and coordination and helped to draft the manuscript. Dr. Wei designed this study and guided the surgery and data collection. All authors read and approved the final manuscript for submission.

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Acknowledgements
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Tables

Table 1. The general clinical and demographic information of subjects.
| Item                               | Data of all patients (n=40) |
|-----------------------------------|-----------------------------|
| Gender                            |                             |
| Male                              | 26/40 (65%)                 |
| Female                            | 14/40 (35%)                 |
| Age (years)                       |                             |
| Average                           | 5.5                         |
| Median                            | 6                           |
| Range                             | 2-14                        |
| Side (cases)                      |                             |
| Bilateral                         | 12/40 (30%)                 |
| Right                             | 12/40 (30%)                 |
| Left                              | 16/40 (40%)                 |
| Preoperative treatment (eyes)      |                             |
| Probing                           | 16/50 (32%)                 |
| Intubation                        | 6/50 (12%)                  |
| Massaging                         | 28/50 (56%)                 |
| Type of bony dysplasia (eyes)     |                             |
| Stenosis                          | 40/50 (80%)                 |
| Atresia                           | 10/50 (20%)                 |
| Effects                           |                             |
| Success rate                      | 50/50 (100%)                |
| Cure rate                         | 41/50 (82%)                 |
| Improvement                       | 9/50 (18%)                  |
| Complications (cases)             |                             |
| Middle turbinate and nasal mucosal adhesion | 4 |
| Nasosinusitis                     | 2                           |
| Other                             | 0                           |

**Table 2.** The effect of type of bony nasolacrimal duct dysplasia on cure rate.
| Type | Cure (%) | Improvement (%) | Invalid (%) | Total |
|------|----------|-----------------|-------------|-------|
| St.  | 33 (82.5)| 7 (17.5)        | 0           | 40    |
| At.  | 8 (80)   | 2 (20)          | 0           | 10    |
| Total| 41 (82)  | 9 (18)          | 0           | 50    |

St. = Stenosis of the nasolacrimal duct; At. = Atresia of the nasolacrimal duct.

The chi-square test showed that the cure rate was not affected by type of bony nasolacrimal duct dysplasia ($\chi^2=0.08, P=0.78$, no statistically significant difference).

**Table 3.** The effect of preoperative treatment on cure rate.

| Group | Cure (%) | Improvement (%) | Invalid (%) | Total |
|-------|----------|-----------------|-------------|-------|
| T     | 20 (91)  | 2 (9)           | 0           | 22    |
| N     | 21 (75)  | 7 (25)          | 0           | 28    |
| Total | 41       | 9               | 0           | 50    |

T=treated before surgery, such as receiving intubation and probing

N=not treated before surgery

The chi-square test showed that the cure rate was not affected by other treatments before endoscopic dacryocystorhinostomy ($\chi^2=1.17, P=0.28$, no statistically significant difference).

**Figures**
Computed tomography angiography of the lacrimal duct allows congenital nasolacrimal canal dysplasia to be clearly distinguished. (A, B) The sagittal position of bony nasolacrimal duct stenosis (black arrow). (C, D) The horizontal position of bony nasolacrimal duct stenosis (black arrow) and the contralateral contrast (white arrow). (E, F) Both the horizontal and sagittal position show bilateral hypomere bony nasolacrimal duct atresia (black arrow).
Figure 2

Illustration of the steps of the operation. (A) Remove the frontal process of the maxilla, moving the sutura lacrimomaxillaris forward with the rongeur (black arrow) (B) Use an electric drill to grind the thicker parts of the maxilla frontal process upper bone, if needed (black arrow); (C) After producing the bone window, expose the inner wall of the lacrimal sac (black arrow). (D) From the lacrimal point, insert the lacrimal probe into the lacrimal sac (black arrow), after endoscopy has verified accurate exposure of the lacrimal sac (black arrow). (E) A longitudinal incision is made along the lacrimal sac wall, and a crosscut incision is made at the top and bottom of the incision, forming a base at the edge of the lacrimal sac wall to form the mucosal flap. Flip the mucosal flap backward, to expose the front mucous membrane of the uncinate process, leaving the lacrimal sac cavity completely open (white outline). (F) The blood is absorbed blood with gauze stuffed into the lacrimal sac (white arrow) and the lacrimal sac mucosal flap is pinned, to reduce movement and bleeding.
Figure 3

Post-operative anastomotic opening. (A) The anastomotic opening is healing but still has mild edema at 1 month post-operation (black arrow). (B) The anastomotic opening at 12 months post operation, with epithelization (white arrow).