# ORIGINAL RESEARCH

# Diversity in Lacrimal Pathway Morphology Among Patients with Congenital Nasolacrimal Duct Obstruction

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**Purpose:** This prospective observational study aimed to explore the diversity in lacrimal pathway morphology among patients with congenital nasolacrimal duct obstruction (CNLDO) by examining dacryocystography (DCG) images.

**Patients and Methods:** The study included 64 patients who underwent DCG before undergoing general anesthesia probing for unilateral CNLDO. Several parameters were measured from the lateral view of the DCG images: (1) the lacrimal sac (LS) and the nasolacrimal duct (NLD) angle, (2) the angle formed by the superior orbital rim (SOR), LS, and the NLD, (3) LS length, and (4) bony NLD length. Additionally, frontal views of the DCG images were utilized to measure (5) LS–NLD angle and (6) LS angle concerning the midline.

**Results:** The average age of the patients was 34.3 months. The mean  $\pm$  standard deviation of the measurements of the above parameters was  $(1) -1.2^{\circ} \pm 16.5^{\circ}$  (range:  $-44.6^{\circ} \pm 46.6^{\circ}$ ),  $(2) -5.0^{\circ} \pm 10.3^{\circ}$  (range:  $-24.0^{\circ} \pm 19.0^{\circ}$ ),  $(3) 10.2 \pm 2.4$  mm (range: 6.5 - 16.0 mm),  $(4) 8.0 \pm 2.5$  mm (range: 3.1 - 14.8 mm),  $(5) 15.6^{\circ} \pm 11.2^{\circ}$  (range:  $-16.8^{\circ} \pm 41.0^{\circ}$ ), and  $(6) 15.1 \pm 5.2^{\circ}$  (range:  $3.3^{\circ} - 29.8^{\circ}$ ). All parameters, except for parameter (3), conformed to a normal distribution.

**Conclusion:** This study provides valuable anthropometric data derived from DCG images, highlighting the substantial variability in lacrimal pathway morphology among patients with CNLDO. Furthermore, anatomical constraints made probing with a straight metal bougie anatomically infeasible in 25.0% of the patients included in this study.

**Plain Language Summary:** Understanding the morphology of the lacrimal pathway is crucial for the successful probing treatments in patients with congenital nasolacrimal duct obstruction (CNLDO). This study represents an initial effort to quantify anthropometric parameters of the lacrimal drainage system through dacryocystography images, specifically aiming to highlight the limitations of blind probing procedure. The results underscore significant variations in the morphology of the lacrimal drainage system among patients, which could impact diagnostic approaches and treatment strategies. Additionally, the findings suggest that patients with CNLDO who do not respond to blind probing may have underlying anatomical complexities. Therefore, rather than relying on repeated blind probing, employing dacryoendoscopy-guided probing under direct visualization could offer a more effective therapeutic alternative for complicated cases of CNLDO.

Keywords: anatomy, congenital nasolacrimal duct obstruction, dacryocystography, dacryoendoscopy, epiphora, lacrimal lavage, probing, radiology

#### Introduction

Congenital nasolacrimal duct obstruction (CNLDO) is primarily associated with the congenital obstruction of the distal portion of the nasolacrimal duct (NLD), frequently characterized by a membranous obstruction.<sup>1</sup> CNLDO has a notable

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spontaneous resolution rate, ranging from 89%–96%, with conservative management causing resolution within the first year.<sup>2–4</sup> Patients who do not achieve spontaneous resolution require surgical intervention. Globally, probing is the primary surgical approach. It involves metal bougie insertion into the lacrimal pathway from the punctum to the distal end of the NLD, where an obstruction is typically located, to create a perforation. Therefore, understanding the morphology of the lacrimal pathway is crucial for the success of probing. Previous investigations have revealed significant variations in the morphology of the lacrimal pathway in adulthood. However, fewer studies have investigated the detailed analysis of the lacrimal pathway in pediatric CNLDO.

Our present investigation aimed to conduct a morphological analysis of the lacrimal drainage system in patients with CNLDO using dacryocystography (DCG) images obtained under general anesthesia at our institution. Our study aims to develop a foundational understanding of lacrimal passage diversity in children, which holds great potential for enhancing future CNLDO diagnosis and treatment.

#### Methods

#### Patient Selection

We reviewed the medical records of 204 patients who underwent probing under general anesthesia as part of a prospective observational study on patients with CNLDO from November 2011 to April 2023 at Kanagawa Children's Medical Center in Japan. Specifically, we analyzed 64 patients with unilateral CNLDO who had undergone DCG before probing. CNLDO diagnosis was established based on clinical symptoms of epiphora and mucous discharge and further confirmed through the fluorescein dye disappearance test. We excluded patients who presented with bilateral involvement, incomplete obstruction, punctum or canaliculus obstruction, congenital malformation syndromes, congenital NLD agenesis, facial anomalies, dysmorphisms, congenital dacryocystocele, a history of epidemic keratoconjunctivitis infection, facial trauma, or eyelid injury (Figure 1). Bilateral CNLDO cases were also excluded due to the overlap of contrast in the lateral view of the DCG images. Observation of the flow of contrast medium into the pharynx on DCG helped to distinguish between the cases in which it was difficult to determine whether an obstruction was complete or incomplete after the lacrimal irrigation test.

#### **DCG** Procedure

Patients with a documented history of allergy to contrast media or iodine were excluded from the examination. In our protocol, DCG was employed as a preoperative investigation in the operating room immediately prior to probing. Probing under general anesthesia was specifically reserved for patients with CNLDO who did not respond to conservative



Figure I Flow diagram of the included cases in this study.

treatment and blind probing conducted in the office. To minimize radiation doses, the exposure was meticulously targeted to the specific area where the lacrimal duct is located. Images in frontal and lateral views were acquired after injecting a nonionic, water-soluble contrast medium (iopamidol of 300 mg/mL) through the dilated upper and lower lacrimal punctum. This was achieved using an inverter-type mobile X-ray system with a minimal radiation dose (Sirius Star Mobile; Hitachi, Tokyo, Japan, with a mean dosage of 70 peak kilovoltage [kVp] and 3.2 milliampere-seconds [mAs]). Dedicated computer software (RapideyeCore v1.5; Canon Medical System, Tochigi, Japan) was used to measure the length and angles of the images.

## Investigated Parameters on DCG Images

Our analysis of DCG images assessed six specific parameters. Four of these measurements were acquired from lateral view images (Figures 2 and 3). The first parameter was the angle between the lacrimal sac (LS) and the NLD (lateral LS–



Figure 2 Methods for measuring the lateral LS–NLD and SOR–LS–NLD angles. The measurements indicated angles with positive and negative values representing anterior and posterior inclination, respectively. The left figure demonstrates the original image, the center exhibits the lateral LS–NLD angle measurement, and the right shows the SOR–LS–NLD angle measurement. Retention of contrast media is evident in dilated mNLD. The images were converted to monochrome to facilitate the observation of contrast media.

Abbreviations: LS, lacrimal sac; bNLD, bony nasolacrimal duct; mNLD, membranous nasolacrimal duct.



Figure 3 Methods for measuring the lengths of LS and bNLD. The long axes of both LS and bNLD were measured from the lateral view of the DCG. The images were converted to monochrome to facilitate the observation of contrast media. Abbreviations: LS, lacrimal sac; bNLD, bony nasolacrimal duct.

NLD angle). The second parameter was the angle formed by the superior orbital rim (SOR), the center of the LS, and the NLD (SOR–LS–NLD angle). The described technique was used to evaluate the SOR–LS–NLD angle. A straight line was initially drawn from the center of the LS toward the SOR, and the point where this line intersected the SOR was determined as the tangent point. Subsequently, we measured the angle formed between the line connecting the tangent point on the SOR to the center of the LS and the line relating the center of the LS to the distal end of the interoseous NLD. Further, we measured the lengths of the bony NLD (bNLD length) and the LS (LS length) within the lateral view image (Figure 3). Furthermore, we assessed whether the SOR in our analysis of the lateral view impeded the bNLD trajectory. In particular, we extended the bNLD line anteriorly to ascertain any obstruction by the SOR. Blockage of this path is expected to pose a challenge for the passage of a straight metal probe through the bNLD caused by this interference due to the SOR (Supplementary Figure 1).

We evaluated two more parameters in terms of frontal view image (Figure 4). The first was the angle between the LS and the NLD (coronal LS–NLD angle), and the second involved measuring the angle formed by the LS in connection to the midline, which extends through the nasal septum (LS–midline angle). Angles were designated by positive and negative values for lateral view image measurements to represent anterior and posterior inclination, respectively. In contrast, angles were designated by positive and negative values for frontal view image measurements to indicate medial and lateral inclination, respectively.

#### Statistical Analysis

Statistical analyses were conducted using the online statistics calculator DATA*tab* (Graz, Austria). The Shapiro–Wilk test was used to assess the normal distribution of measurement data, with a *p*-value of >0.05 indicating normality in the distribution.

## Results

This study included 64 patients with unilateral CNLDO, including 41 (64.1%) males and 23 (35.9%) females. Among them, 37 (57.8%) were on the right side and 27 (42.2%) were on the left. The mean  $\pm$  standard deviation of the age of patients during surgery, which also represents the time of data measurement, was  $34.3 \pm 19.2$  months (range: 14–84 months). Concerning physical measurements, the average height of the patients was  $90.1 \pm 11.4$  cm (range: 75.1–119.2 cm), and the average weight was  $13.4 \pm 3.2$  kg (range: 8.6–21.2 kg). All patients in this study exhibited membranous obstruction at the NLD terminus. No obstructions were observed at other anatomical sites, including the common canaliculus or the LS–NLD transition. In addition, preoperative examinations ruled out the presence of any bony obstructions. All measurement data were acquired from the obstructed side and are presented in Table 1, with further details provided in the <u>Supplementary Table 1</u>. Figure 5 depicts normal distribution diagrams for the assessed parameters.



Figure 4 Methods for measuring the frontal LS–NLD angle and LS–midline angle. The measurements denoted angles by positive values to signify medial inclination and negative values to represent lateral inclination. From left to right: the sequence displays the original image, the measurement of the frontal LS–NLD angle, and the measurement of the LS–midline angle. The images were converted to monochrome to enhance the contrast media visibility. **Abbreviations:** LS, lacrimal sac; bNLD, bony nasolacrimal duct; mNLD, membranous nasolacrimal duct.

Parameters	Lateral LS–NLD angle	SOR-LS-NLD angle	LS length	bNLD length	Frontal LS–NLD angle	Frontal Medial– LS angle
Mean value	-1.2° ± 16.5°	-5.0°± 10.3°	10.2 ± 2.4 mm	8.0 ± 2.5 mm	+15.6° ± 11.2°	15.1° ± 5.2°
Maximum value	+46.6°	+19.0°	16.0 mm	14.8 mm	+41.0°	29.8°
Minimum value	44.6°	-24.0°	6.5 mm	3.1 mm	-16.8°	3.3°
Normal distribution (p-value)*	Yes (p = 0.69)	Yes (p = 0.25)	No (p = 0.036)	Yes (p = 0.62)	Yes (p = 0.74)	Yes (p = 0.27)

Table I Lacrimal Duct Parameters Measured in This Study

**Notes**: \*The *p*-value in the normal distribution column was calculated by the Shapiro–Wilk test; p > 0.05 indicated that the distribution was normal.

Abbreviations: bNLD: bony nasolacrimal duct, CNLDO: congenital nasolacrimal duct obstruction, DCG: dacryocystography, LS: lacrimal sac, NLD: nasolacrimal duct, SOR: superior orbital rim.

#### Lateral LS-NLD Angle

The "lateral LS–NLD angle" is the angle formed by the long axis of the LS and NLD when observed in the lateral view of DCG images. We designated the anterior inclination as a positive value. On average, we observed a posterior inclination of  $-1.2^{\circ} \pm 16.5^{\circ}$ . The range for this angle differed from a maximum posterior inclination of  $-44.6^{\circ}$  to a maximum anterior inclination of  $+46.6^{\circ}$ . Our findings indicate that anterior inclination, characterized by LS–NLD angles of >0°, was observed in 24 (37.5%) cases. Conversely, posterior inclination, defined by LS–NLD angles of <0°, was identified in 31 (48.4%) cases. Meanwhile, 9 (14.1%) cases exhibited an LS–NLD angle of 0°. Our measurements indicated a pattern consistent with a normal distribution, as affirmed by the Shapiro–Wilk test, with a *p*-value of 0.69.

## SOR-LS-NLD Angle

The "SOR-LS-NLD angle" denotes an angle formed by two lines within the lateral view: one extending from the SOR to the center of the LS and the other connecting the center of the LS to the NLD. An anterior inclination was designated as a positive value. This value represents the angle formed by the probe and the NLD when the probe is inserted into the LS and in contact with the forehead (Supplementary Figure 2). On average, we recorded a posterior inclination of  $-5.0^{\circ} \pm 10.3^{\circ}$ . The range for this angle differed from a maximum posterior inclination of  $-24.0^{\circ}$  to a maximum anterior inclination of  $+19.0^{\circ}$ . We observed anterior inclination in 16 (25.0%) cases and posterior inclination in 44 (68.8%) cases. Additionally, 4 (6.2%) cases exhibited a SOR-LS-NLD angle of 0°. Our measurements conformed to a normal distribution (p = 0.25).

#### SOR Interference

Passing a straight probe through the NLD is anticipated to be challenging when the anteriorly extended path of the bony NLD interferes with the SOR in a lateral view (Supplementary Figure 1). Among the 64 cases in our study, 16 (25%) encountered this specific interference.

## LS Length

The "LS length" represents the measurement of the longitudinal axis of the LS as observed in the lateral view. The mean LS length was determined as  $10.2 \pm 2.4$  mm. LS length values ranged from 6.5 mm to 16.0 mm. Notably, our measurements did not adhere to a normal distribution, as verified by the Shapiro–Wilk test verified (p = 0.036).

#### bNLD Length

The "bNLD length" is the measurement of the bony NLD's length as observed in the lateral view of DCG images. The average NLD length was recorded at  $8.0 \pm 2.5$  mm. The spectrum of bNLD lengths ranged from 3.1 mm to 14.8 mm. Our measurements matched to a normal distribution (p = 0.62).



Figure 5 Distribution of measurements. All parameters, except LS length, demonstrated a p-value of >0.05 in the Shapiro-Wilk test, indicating a normal distribution.

## Frontal LS-NLD Angle

The "frontal LS–NLD angle" denotes the angle of inclination from the LS to the NLD in a frontal view of DCG images, with medial inclination designated as a positive value. On average, we observed a medial inclination of  $+15.6^{\circ} \pm 11.2^{\circ}$ . The range for this angle differed from a maximum medial inclination of  $+41.0^{\circ}$  to a maximum lateral inclination of  $-16.8^{\circ}$ . Our measurements demonstrated conformity to a normal distribution pattern (p = 0.74).

## Frontal Medial-LS Angle

The "frontal medial–LS angle" is the angle created by the long axis of the LS and the median line running through the nasal septum in the frontal view. The average measured angle was  $15.1^{\circ} \pm 5.2^{\circ}$  (range:  $3.3^{\circ}-29.8^{\circ}$ ). Our measurements demonstrated a normal distribution pattern (p = 0.27).

#### Discussion

Probing is the primary choice for surgical treatment in patients with CNLDO, and a comprehensive understanding of the anatomy of the lacrimal pathway is crucial for achieving success in the procedure. Knowledge of the radiologic details of the lacrimal drainage system in children is limited.<sup>5–8</sup> To the best of our knowledge, this study is the first attempt to measure the anthropometric parameters of the lacrimal drainage system in patients with CNLDO, specifically highlighting the limitations of the blind lacrimal duct probing procedure. The length and morphology of the lacrimal drainage system demonstrated considerable differences among individuals and ethnic groups in adulthood.<sup>9-14</sup> Our group previously analyzed the morphology of the normal lacrimal pathway in adults within a Japanese population. This analysis was based on DCG images obtained using cone-beam computed tomography.<sup>15</sup> These results indicate a  $-6.3^{\circ}$  $\pm$  14.1° average sagittal LS–NLD angle in adults. Consequently, the value of  $-1.2^{\circ} \pm 16.5^{\circ}$  observed in patients with CNLDO in this study represented less posterior inclination in comparison with the measurements in adults. While this angle is anticipated to evolve with skull development, cadaveric studies in adults have also demonstrated significant variability in the lacrimal pathway among individuals, even within the same ethnic group. For example, Narioka et al conducted a measurement based on 46 lacrimal pathways from 23 Japanese adult cadavers and revealed that the sagittal LS-NLD angle exhibited anterior inclination in approximately 80% of cases and posterior inclination in the remaining 20%.<sup>16</sup> In contrast, Park et al involved measurements of 42 lacrimal pathways from 21 Japanese adult cadavers and revealed that approximately 90% of cases demonstrated posterior inclination with an average angle of  $-10.3^{\circ}$ .<sup>17</sup> It is presumed that an increased degree of flexure in the LS-NLD angle would make it more difficult for surgeons to probe through the LS-NLD transition.

Our current study also explored additional anatomical constraints related to the probing procedure. A SOR–LS–NLD angle with a positive value poses challenges for a straight metal probe passing through the bNLD due to interference from the SOR (<u>Supplementary Figure 2</u>). This interference was observed in 16 (25.0%) patients with CNLDO in this study, indicating the potential benefit of using a probe with a curved or bent tip for procedures in such cases. Additionally, 48 (75.0%) cases exhibited a SOR–LS–NLD angle of less than 0°, indicating the feasibility of passing the bNLD with a straight probe without the SOR interference.

The current study measured the mean LS length in children at  $10.2 \pm 2.4$  mm, which is notably longer than the value observed in our previous research that included unaffected adults, which was  $8.9 \pm 2.3$  mm.<sup>15</sup> This indicates that LS may undergo dilation due to prolonged obstruction in patients with CNLDO.

DCG is instrumental in identifying the obstruction site and assists in planning the appropriate surgical approach for CNLDO. However, the DCG procedure involves increased radiation exposure, which is particularly concerning in infants due to their heightened radiosensitivity and the associated risk of malignancy.<sup>18,19</sup> Radiologic procedures carry inherent risks that should be balanced against their potential for significant clinical benefit. In our protocol, DCG was used as a preoperative investigation in the operating room immediately before the surgical procedure, which was reserved only for patients with CNLDO refractory to conservative treatment and blind probing. Confirming the obstruction site and understanding the morphology of the lacrimal pathway were helpful in planning an effective approach to the NLD. It is also imperative to minimize radiation doses through careful selection of kVp and mAs.<sup>18</sup> The radiation exposure dose of our DCG procedure was about 0.02 mSv. This dose was limited to the site of lacrimal pathway, which resulted in a radiation exposure that was significantly lower than that of a chest X-ray (about 0.1 mSv) and computed tomography scan (about 2.0 mSv).<sup>20</sup>

Recently, the effectiveness of dacryoendoscopy-guided probing in patients with CNLDO has been documented in several instances.<sup>21–25</sup> This procedure does not always necessitate general anesthesia, with notable success rates under local anesthesia in an office setting.<sup>26</sup> The findings of this study suggest that patients who do not respond to blind probing may present with anatomical complexities. Consequently, instead of relying on repeated blind probing, dacryoendoscopy-guided probing under direct visualization could offer a viable therapeutic alternative.

This study has three potential limitations. First, the study was conducted at a tertiary academic children's hospital, and the included patients with CNLDO were those who had not responded to conservative therapy or local anesthesia probing at the institution before referral. This introduces a selection bias regarding the extent to which the measurements in this

study align with the general anthropometric characteristics of children with CNLDO. Second, this study acquired the DCG images using a mobile X-ray system, which may not guarantee perfectly aligned frontal and lateral planes, thereby possibly affecting the measurement accuracy. Third, the data obtained from this measurement is expected to change with the growth of children. Previous studies indicated that the dimensional increases of the NLD are nonlinear, with most growth occurring within the first 6 months of life.<sup>6</sup> Similarly, the data collected in this study are anticipated to evolve alongside skull development.

## Conclusion

This study provides anthropometric data derived from DCG images to elucidate the diverse morphology of the lacrimal pathway in patients with CNLDO. These results indicate the considerable variation in the morphology of the lacrimal drainage system among individuals. From the surgical perspective, the lacrimal LS–NLD transition exhibited an average posterior inclination of 1.2° in the front–back direction, with observed variations ranging from a maximum posterior inclination of 44.6° to a maximum anterior inclination of 46.6°. In addition, the average medial inclination was 15.6° in the transverse direction, which demonstrates a wide range that extended up to a maximum medial inclination of 41.0° and a maximum lateral inclination of 16.8°. Furthermore, it was evident that probing with a straight metal bougie was anatomically infeasible in certain patients with CNLDO due to the anatomical constraints of the SOR. This observation has important implications for diagnostic approaches and treatment strategies.

## **Abbreviations**

bNLD, bony nasolacrimal duct; CNLDO, congenital nasolacrimal duct obstruction; DCG, dacryocystography; kVp, peak kilovoltage; LS, lacrimal sac; mAs, milliampere-seconds; NLD, nasolacrimal duct; SOR, superior orbital rim; SD, standard deviation.

## **Data Sharing Statement**

All data analyzed in this study are included in this article and elaborated upon in the <u>Supplementary Information</u>. For additional inquiries can be directed to the corresponding author (JN) on reasonable requests.

## Ethical Approval and Consent to Participate

This study and the data collection protocol were authorized by the institutional review board and the ethics committee of Kanagawa Children's Medical Center. Parents or guardians of all participating patients signed written informed consent. Additionally, the legal guardians of all patients provided consent to the publication of identifying information and images. All procedures used in this study were conducted following the principles of the Declaration of Helsinki.

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## Disclosure

The authors report no conflicts of interest in this work.

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