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ARTICLE

Congenital nasolacrimal duct obstruction update study (CUP study): paper I—role and outcomes of Crigler's lacrimal sac compression

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Abstract

Purpose To assess the exclusive role and outcomes of Crigler's lacrimal sac compression in the management of congenital nasolacrimal duct obstruction (CNLDO).

Methods Retrospective interventional case-series was performed on patients diagnosed with CNLDO and who were advised Crigler's lacrimal sac compression (CLSC) at a tertiary care Dacryology Institute from Jan 2016 to June 2019. CNLDO patients who were practicing incorrect techniques of lacrimal sac compression at presentation were separately assessed. All the patients were assigned to four groups (Gr 1: 0–3 months, Gr 2: >3 & <6 months, Gr 3: >6 & <9 months and Gr 4: >9 and <12 months) based on the age at which the CLSC was initiated and followed up quarterly or as needed till at least 1 year of age. The parameters studied include patient demographics, clinical presentation, age of initiation of CLSC, success rate with CLSC, and need for additional interventions. Success was defined as the subjective resolution of epiphora and discharge with objective measures of normal tear meniscus height and dye clearance on fluorescein dye disappearance test.

Results A total number of 1240 patients with CNLDO were assessed. Of these, 1037 patients were advised correct techniques of CLSC from the beginning, and the remaining 203 patients were referred but performing it incorrectly at presentation. Of the 1037 patients, 236 were lost to follow-up; hence, a total of 1004 patients (801 + 203) were included for final analysis. CLSC was found to be an effective conservative strategy in the management of CNLDO. The rate of resolution of CNLDO in Gr 1 to Gr 4 was 87.3%, 78.9%, 77.9%, and 76.8%, respectively. There were no statistically significant differences in the outcomes based on the age of CLSC initiation. The referred patients whose techniques were rectified following the initial incorrect techniques showed a resolution of 61.2% (79/129). The correct techniques of CLSC appeared to influence the outcomes. However, the age of its initiation did not substantially impact the outcomes. Significantly high resolution was noted even beyond nine months of age and encouraging results beyond 12 months of age.

Conclusions It is crucial to initiate the correct techniques of Crigler's lacrimal sac compression to achieve favourable outcomes. Age of initiation of CLSC in infancy does not appear to influence the outcomes. The resolution rate continued to be significantly high up to 1 year of age. There is a need to assess the role of CLSC beyond 12 months of age.

Introduction

Congenital nasolacrimal duct obstruction is the most common congenital lacrimal drainage disorder and a common cause of paediatric epiphora [1]. It can be of a simple variant when associated with only a membranous obstruction at the distal end of nasolacrimal duct (NLD) or complex when it is associated with syndromes or certain other embryonic anomalies involving the lacrimal drainage system [2, 3]. The incidence of epiphora varies from 1.2 to 20% based on methods and types of studies reporting them [4, 5]. The studies that assessed the natural history of the disease or spontaneous resolution or resolution with conservative measures like Crigler's lacrimal sac compression (CLSC) had several limitations [4, 6, 7]. The age of entry into the studies, unconfirmed diagnosis, the age of initiation of lacrimal sac compressions, variety of interventions, and

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ill-defined follow-up have limited the conclusions that could be confidently drawn into the practice. The spontaneous resolution rate of symptomatic epiphora was found to be 96% by the age of 1 year, and the success of probing was found to decrease beyond 13 months of age [4, 8]. This led to the widespread practice of probing at or beyond 1 year of age.

Lewis Crigler in 1923 proposed his technique of digital compression of the lacrimal sac to enhance the hydrostatic pressure within the lacrimal drainage system to overcome the membranous obstruction at the distal end of the NLD [9]. Several variations of this technique were proposed with variable success rates. However, the original description of Crigler's appear to be more logical and is being more advocated in recent times [6, 8, 10, 11]. However, few have studied the role of isolated lacrimal sac compression and rarely in big cohorts of CNLDO [5, 6, 8, 10, 12–18]. Systematic reviews have recommended lacrimal sac compressions as a conservative modality and recognized them as a non-invasive modality that increases the spontaneous resolution rate and has high compliance [19, 20]. The current study assessed the role and outcomes of CLSC alone in a group-wise classified large cohort of patients diagnosed with CNLDO.

Methods

The manuscript adhered to the Tenets of the Declaration of Helsinki. Institutional review board and Ethics committee approval was obtained. Retrospective chart reviews were performed for patients diagnosed with CNLDO and who were advised Crigler's lacrimal sac compression (CLSC) at a tertiary care Dacryology Institute from Jan 2016 to June 2019. Patients who were practicing incorrect techniques of lacrimal sac compression at presentation were separately assessed. The patients who had associated complexities like syndromes, craniofacial anomalies, dacryoceles or acute dacryocystitis or any previous surgical interventions were excluded. The patients were assigned to four groups (Gr 1: 0–3 months, Gr 2: >3 & <6 months, Gr 3: >6 & <9 months and Gr 4: >9 and <12 months) based on the age at which the CLSC was initiated and followed up quarterly or as required till at least 1 year of age. All patients were advised ten strokes of lacrimal sac compression, four times per day using the original Crigler's technique [1, 9, 11]. The parameters studied include patient demographics, clinical presentation, age of initiation of CLSC, success rate with CLSC, and need for additional interventions. Success was defined as the subjective resolution of epiphora and discharge with objective measures of normal tear meniscus height and dye clearance on fluorescein dye disappearance test.

Results

A total number of 1240 patients with CNLDO were assessed. Of these, 1037 patients were advised correct techniques of CLSC from the beginning. The remaining 203 patients were referred to the authors and were performing CLSC incorrectly at presentation. Table 1 presents the demographic details of the 1037 patients and their respective group-wise divisions. Of the 1307 patients, 236 were lost to follow-up; hence, a total of 1004 patients (801 + 203) were included for final analysis (Table 1). Of the 801 patients, 6 continued massage beyond 12 months and hence were excluded, leaving a total number of 795 with correct techniques for the final analysis. There was no gender predilection noted for the study patients. CLSC was found to be an effective conservative management strategy in the management of CNLDO. The rate of resolution of CNLDO in Gr 1 to Gr 4 was 87.3%, 78.9%, 77.9%, and 76.8%, respectively (Table 2). There were no statistically significant differences in the resolution rates between Gr 1 and Gr 2 ($P = 0.0759$), Gr 2 and Gr 3 ($P = 1$) and Gr 3 and Gr 4 ($P = 1$). The same was true when a comparison was performed between those who were initiated at <6 months versus >6 months of age ($P = 0.06$) (Table 2). Overall, the generalized resolution rate of CNLDO in infancy with CLSC was 81% (644/795). Of those who did not respond to CLSC [19% (151/795)], all underwent irrigation, and probing and subsequently, 7 patients refractory to probing (Complex CNLDO) underwent dacryocystorhinostomy.

Table 1 Demographics of the CNLDO-Sac compression study group.

Initiation age groups	Males	Females	Total	LFU	Final analysis Nos
Gr 1: 0–3 months	152	146	298	60	238
Gr 2: >3 & <6 months	183	165	348	82	266
Gr 3: >6 & <9 months	158	142	300	78	222
Gr 4: >9 & <12 months	41	44	85	16	69
Gr 5: > 12 & <18 months	1	5	6	0	6
Total	535	502	1037	236	801

CNLDO congenital nasolacrimal duct obstruction; initiation—age at which lac sac compression was initiated; *LFU* lost to follow-up, *Nos* numbers, *Gr* group.

Table 2 Outcomes of CNLDO with Crigler's Lacrimal sac compression.

Initiation age groups	Resol at 1 year age (%)	Not resolved (%)	Group comparisons	
			Groups	*p* value
Gr 1: 0–3 months	208 (87.3%)	30 (12.7%)	Gr1 vs Gr2	0.0759
Gr 2: >3 & <6 months	210 (78.9%)	56 (21.1%)	Gr1 vs Gr3	0.0468
Gr 3: >6 & <9 months	173 (77.9%)	49 (22.1%)	Gr1 vs Gr4	0.1958
Gr 4: >9 & <12 months	53 (76.8%)	16 (23.1%)	Gr2 vs Gr3	1
Overall <12 months	644/795 (81%)	151/795 (19%)	Gr2 vs Gr4	1
			Gr3 vs Gr4	1

Resol resolution at 1 year of age, *Gr* group.

Table 3 Outcomes with incorrect Crigler's techniques.

Total patients—203
Spontaneous resolution—1
Advised Crigler's (<12 months) and subsequently followed correct technique—129
Age beyond 12 months and advised probing—73
Of those 129 who rectified their techniques
Resolved with Crigler's lacrimal sac compressions—79 (61.2%)
Underwent additional interventions—44 (34.1%) (40 successful after probing, 1 failed probing—lost to follow up & 3 successful DCRs)
Lost to follow up—6 (4.7%).

Table 4 Broad probability of success with Crigler's compression (for teaching & parent counselling).

Initiation age groups	Likely resolution rate
0–3 months	90%
>3 & <6 months	80%
> 6 & <9 months	80%
> 9 months & < 12 months	75%

The numbers have been rounded to nearest 0 or 5 digit for easy memory.

The complex CNLDO includes refractory atonic sacs ($n = 4$) and one each of the buried probe, bony NLD obstruction, and refractory diffuse nasolacrimal duct stenosis. Interestingly, 6 patients' parents continued CLSC beyond 12 months on their own, of which 5 resolved before 18 months of age and 1 underwent successful probing. Of the 203 patients with incorrect techniques and subsequently rectified, 129 adhered to the modified techniques (Table 3). The resolution rate amongst these 129 patients was 61.2% (79/129) (Table 3). The correct technique of CLSC appeared to influence the outcomes and can be corroborated with the fact that incorrect techniques cannot anatomically achieve the goal of increasing the hydrostatic pressure within the lacrimal sac. However, the age of its initiation did not appear to substantially impact the outcomes till 1 year of age. Significantly high resolution was noted even beyond nine months of age. Table 4 demonstrates the resolution rate in this large cohort, rounded to the nearest "0" or "5" digit to help with parent counselling and teaching.

Discussion

The current study describes the influence of correct techniques and the age of initiation of Crigler's lacrimal sac compression on the outcomes of CNLDO. It may represent a paradigm shift in our understanding of the concept of spontaneous resolution. The resolution rate continued to be significantly high up to 1 year of age and encouraging results beyond 1-year of age. There is a need to reassess the role of CLSC beyond 12 months of age.

MacEwen and Young observed 4792 infants to assess the incidence of epiphora (20%, $n = 964$) and its spontaneous resolution [4]. They studied the natural resolution of the disease in a cohort of 834 symptomatic infants with epiphora and found an overall rate of spontaneous resolution to be 96% by the end of 1 year of age. This led to the widespread practice of probing at or beyond the age of 1 year for uncomplicated cases of CNLDO. The current study with comparable numbers, however, did not notice such a high resolution at the end of 1 year of age, even with the addition of CLSC [81% (644/795)]. While this can be ascribed to the hospital setting compared to a community setting in the other study, it is crucial to consider that hospital statistics provide a better representation of a physician's practice when it comes to managing children with CNLDO. The current study also demonstrated a continuous high-resolution rate even beyond nine months. This, combined with an incidental finding of resolution of CLSC in a small group beyond 12 months of age, should allow us further to contemplate the possible role of CLSC in older children. This notion also gains support from the work of Katowitz and Welsh, who demonstrated spontaneous resolution with CLSC and topical antibiotics in patients up to 24 months of age [13–18 months—10.4% (8/77; 19–24 months—11.9% (5/42)] [8]. Based on a population-based study, MacEwen and Young also believed that CNLDO may not be a pathology at all due to their high incidence (20%) and high

spontaneous resolution (96%) [4]. The authors of the current study do not agree with this concept since a vast majority of those children (80%) did not represent lacrimal drainage dysfunction. The lacrimal sac and the nasolacrimal duct function to transport the tears and are expected to be normal when the tear secretions begin. Any alterations in this arrangement cannot be considered normal.

Kushner demonstrated that of the 58 eyes with CNLDO in their series, whom they observed with just topical antibiotic drops, only 4 (6.8%) showed spontaneous resolution [6]. Of the 58 eyes who underwent gentle compressions, only to evacuate the lacrimal sac discharge, only 5 (8.6%) demonstrated spontaneous resolution as compared to 18 (30.5%) of the 59 eyes, which resolved with the hydrostatic massage [6]. However, the mean age in each group was approximately seven months. Although this would appear quite late, the findings should be interpreted in the light of new data from the present study showing high-resolution rates even beyond nine months of age. Also, Kushner's technique of hydrostatic massage, as demonstrated in their figure, does not appear to be exactly what was described originally.

The technique of Crigler's lacrimal sac compression has varied over the last century between different series. The basic principle is to compress the lacrimal sac in a way that the contents do not regurgitate from the puncta. The increased pressure within the sac would then get transmitted to the nasolacrimal duct overcoming the membranous obstruction [1, 11]. It is important to direct the contralateral index finger just within the anterior lacrimal crest to compress the sac well.[1] Any circular motion or downward sweep along the side of the nose will not generate the pressure within the lacrimal sac and hence would be ineffective [11]. The results of the current study and subgroup analysis of the improper technique group reflects the importance of performing it correctly.

Stolovitch and Michaeli found hydrostatic pressure to be an effective way of treating CNLDO in infants and reducing its morbidity [10]. The compressions were performed by the surgeon in the office for up to 3 such sittings with weekly intervals. The overall success rate was noted in 45% of the infants. The success rate was high in the first attempt and reduced as the number of attempts increased. Interestingly the younger the age, the better was the resolution [10]. The current study with similar numbers had an overall success rate of 81% in the infants, which was nearly twice that of the Stolovitch and Michaeli study. The authors of the present study believe that only 2–3 sittings are not enough. Hence, they educate and train the parents with the CLSC techniques, which are simple to learn. The protocol of ten strokes per sitting and four such sittings in a day has shown high CNLDO resolution rates. The current study has also found that resolution rates with correct techniques of CLSC continue to be high even beyond 9 months and the age of its initiation does not significantly impact the outcomes.

The limitations of the current study are the confounding bias of natural history, which cannot be excluded. Although the data from the wrong technique subgroup can provide some insights about this confounding bias, it cannot be completely eliminated. One way to study natural resolution would be to have a control group which can prospectively observe all children of CNLDO for up to 1 year without any sac compression or topical medications or any other intervention. However, given the demonstrable benefits of CLSC, it is ethically not feasible to prospectively deny CLS to study natural history. The strengths of this study include a large cohort and group-wise focused assessment of the role of CLSC in patients diagnosed with CNLDO.

In conclusion, the current study had demonstrated the benefits of CLSC in a large group of diagnosed CNLDO patients. The resolution rate with CLSC continued to be high throughout the infancy. The technique of lacrimal sac compression but not the age at its initiation in infancy that appears to influence the outcomes of CNLDO. The resolution of CNLDO with CLSC may continue beyond 12 months and focused prospective study would be useful for initiating a change in the current preferred practice patterns.

Summary

What was known before

- CNLDO has a high rate of spontaneous resolution.
- The role of Crigler's lacrimal sac compression is unclear.

What this study adds

- The techniques of Crigler's lacrimal sac compression appears to influence the outcomes.
- Resolution with Crigler's lacrimal sac compression may continue even beyond 12 months.

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Compliance with ethical standards

Conflict of interest MJA receives royalties from Springer for his treatise 'Principles and Practice of Lacrimal Surgery' (2nd ed) and 'Atlas of Lacrimal Drainage Disorders'. The authors declare that they have no conflict of interest.

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