

The Impact of the COVID-19 Pandemic on Cleft Care

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Background: The COVID-19 pandemic had multiple effects on the provision of health care, including the suspension of elective and nonessential surgeries. The objective of this study was to determine the early effect of the COVID-19 pandemic on the surgical care of patients with cleft lip and/or palate at a high-volume cleft center.

Methods: A retrospective comparative cohort study of patients with cleft lip and/or palate undergoing lip adhesion, cleft lip and nose repair, and palatoplasty before and during the pandemic was conducted. There were 50 patients in the prepandemic cohort and 53 in the pandemic cohort.

Results: Mean age at lip adhesion was 3.1 ± 1.1 months prepandemic ($n = 8$) and 3.5 ± 2.5 months in the pandemic cohort ($n = 8$) ($P = 0.75$). One lip adhesion was delayed by 1.6 months. Mean age at cleft lip and nose repair was 6.6 ± 1.9 months prepandemic ($n = 23$) and 8.0 ± 2.1 months in the pandemic cohort ($n = 23$) ($P = 0.03$). Six pandemic cleft lip and nose repairs were delayed; the mean delay was 2.6 ± 1.8 months. The mean age at palatoplasty was 13.9 ± 2.2 months prepandemic ($n = 26$) and 14.1 ± 2.9 months in the pandemic cohort ($n = 26$) ($P = 0.79$). Seven pandemic palatoplasties were delayed; the mean delay was 3.3 ± 1.4 months.

Conclusions: The COVID-19 pandemic caused delays at each stage of repair for cleft lip and/or palate-related procedures; however, only cleft lip and nose repair were significantly affected. This study emphasizes the importance of remaining vigilant regarding the care of this vulnerable population during this challenging time. (*Plast Reconstr Surg Glob Open* 2021;9:e3587; doi: 10.1097/GOX.0000000000003587; Published online 28 April 2021.)

INTRODUCTION

The coronavirus disease 2019 (COVID-19) pandemic has had multiple effects on the provision of health care, including the suspension of elective and nonessential surgeries in the United States and abroad.¹ In response to the growing COVID-19 pandemic and resultant moratorium on elective and nonessential procedures, the American Cleft Palate-Craniofacial Association (ACPA) released a statement on April 1, 2020 regarding cleft lip and palate (CL/P) related surgeries.² ACPA guidance recommended delaying primary cleft lip repair and prioritizing patient and provider safety when considering whether to delay palate repair. It

is also important to consider that intraoral surgeries, such as cleft palate repair, are particularly high risk for COVID-19 transmission due to aerosolization of the virus,³ and in early April, national focus was on mitigation efforts to minimize spread and preserve health system resources including intensive care beds and personal protective equipment. Other publications have echoed the ACPA guidelines to delay cleft lip repair, but the risk of delaying palate repair must be weighed against the risk of exposure to COVID-19 for the patient and healthcare team.^{4,5}

The first year of life for children with cleft lip/palate requires careful adherence to institutional protocols to ensure the best surgical and speech outcomes. Earlier lip repair may support family integration of the infant in addition to improving nasal aesthetics, which may have important psychosocial implications later in life.⁶ Early palate repair impairs maxillary growth, but several studies describe worse speech outcomes with delayed cleft palate repair, as velopharyngeal insufficiency can affect phonological development and lead to maladaptive articulation patterns, decreased speech intelligibility, and social stigmatization.⁷⁻¹⁰ Although protocols and timing of repair

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can vary by institution, definitive lip repair is typically performed at the age of 3–6 months, followed by palate repair between the age of 6 and 18 months.^{11–13} A survey of pediatric craniofacial surgeons conducted in April 2020 found that only 29% would offer cleft lip repair currently or in situation-specific manner, while 38% would offer cleft palate repair currently or in situation-specific manner.¹⁴ As of now, the effect of surgical delay on CL/P repair as a consequence of COVID-19 remains unknown.

The goal of the present study was to evaluate the early effect the COVID-19 pandemic has had on cleft care at a high-volume cleft center. We hypothesize that the COVID-19 pandemic delayed care for CL/P patients. To test this hypothesis, we compare CL/P patients undergoing lip adhesion, cleft lip repair, and palatoplasty to a matched pre-pandemic cohort at a high-volume tertiary academic cleft center.

METHODS

This is a retrospective comparative cohort study approved by the Institutional Review Board. The study population included patients with CL/P undergoing lip adhesion, cleft lip/nose repair, and palatoplasty at a single tertiary care, stand-alone pediatric hospital with >300 beds. The pandemic cohort included patients who underwent surgery from March 2020 to August 2020. The pre-pandemic cohort included matched patients who underwent surgical repair during 2018 and 2019. Patients with sub-mucosal cleft palates and those carrying syndromic diagnoses were excluded from analysis.

Data were extracted from the electronic medical records of CL/P patients, including demographic information, clinical diagnoses, and surgical details. Patient information was compared between the 2 groups. Surgical delay, defined as deviation from our institutional protocol of lip adhesion by 3 months, primary lip repair by 6 months of age and palate repair by 12 months of age, was calculated. Patients in the pandemic cohort whose surgeries were delayed for reasons unrelated to COVID-19 (eg, patient illness other than COVID-19) were not included in the calculation for surgical delay. Demographic data were analyzed with Fisher’s exact test and age at surgery was analyzed with two-tailed Student’s *t*-test; a *P* ≤ 0.05 was considered statistically significant for all analyses. All analyses were performed using IBM SPSS, version 23.0 (IBM Corp., Armonk, N.Y.).

RESULTS

In total, 103 patients were included in the study (47 female, 56 male). Fifty-three patients underwent cleft surgical repair in 2020: 8 cleft lip adhesions, 23 cleft lip & nose repairs, and 26 palatoplasties. This cohort was matched 1 to 1 with 50 patients who underwent surgery before the pandemic: 8 cleft lip adhesions, 23 lip & nose repairs, and 26 palatoplasties. There was no statistically significant difference between the 2 groups (Table 1).

Mean age at lip adhesion was 3.1 ± 1.1 months pre-pandemic (n = 8) and 3.5 ± 2.5 months in the pandemic cohort (n = 8) (*P* = 0.75). One pandemic lip adhesion

was delayed by 1.6 months. Mean age at cleft lip and nose repair was 6.6 ± 1.9 months pre-pandemic (n = 23) and 8.0 ± 2.1 months in the pandemic cohort (n = 23) (*P* = 0.03). Six pandemic cleft lip and nose repairs were rescheduled due to the ban on elective surgeries; the mean delay was 2.6 ± 1.8 months. The mean age at palatoplasty was 13.9 ± 2.2 months pre-pandemic (n = 26) and 14.1 ± 2.9 months in the pandemic cohort (n = 26) (*P* = 0.79). Seven pandemic palatoplasties were delayed; the mean delay was 3.3 ± 1.4 months (Table 2).

DISCUSSION

To our knowledge, this is the first report describing the effects of the COVID-19 pandemic on the surgical care of patients with CL/P. The results of this study confirm that COVID-19 caused delays at each stage of repair, though only primary cleft lip and nose repair were significantly affected. These results are concordant with the recommendation from the ACPA to delay cleft lip repair during the moratorium on nonessential and elective surgeries during the height of the COVID-19 pandemic. The clinical significance of such delays in care may be small; however, the uncertainty of the duration of the pandemic and cumulative effect on multiple procedures over the course of a cleft patient’s care remains unknown.

The psychosocial impact of delayed cleft repair is considerable. Delay in cleft lip repair may lead to impaired cognitive functioning at 18 months as a consequence of disrupted early maternal-infant interactions before repair.¹⁵ One study found that, when compared with the general population, parents of children with CL/P show higher levels of depression in the first months of treatment, and maternal stress may be ameliorated by early surgical intervention.¹⁶ Similar studies have found that quality of life of families with cleft children improves in social and financial domains after surgical repair.¹⁷ While definitive lip repair at our institution occurs later (typically at 6 months of age), repair in 2020 was significantly delayed compared with 2019 secondary to COVID-19. The rate of NAM treatment was lower during the pandemic; there were 6 patients in the pre-pandemic cohort and only 1 in the pandemic cohort. Providers saw this patient with additional precautions and there were no interruptions or delays in their care. The one patient who experienced a delay in lip adhesion was performing DynaCleft taping

Table 1. Patient Demographics

	Prepandemic (n = 53)	Pandemic (n = 53)	Fisher’s Exact Test
Gender			<i>P</i> = 1.0
Male	28 (53%)	29 (55%)	
Female	25 (47%)	24 (45%)	
Diagnosis			<i>P</i> = 0.5
Cleft lip & palate	22 (42%)	19 (36%)	
Unilateral	18 (36%)	15 (28%)	
Bilateral	6 (12%)	4 (8%)	
Isolated cleft lip	13 (25%)	14 (26%)	
Unilateral	10 (20%)	12 (23%)	
Bilateral	1 (2%)	2 (4%)	
Isolated cleft palate	18 (34%)	20 (38%)	

Table 2. Comparison of Age at Surgical Intervention

	Mean Prepandemic Age, Mo \pm SD (N)	Mean Pandemic Age, Mo \pm SD (N)	P
Lip adhesion	3.1 \pm 1.1 (8)	3.5 \pm 2.5 (8)	0.75
Lip & nose repair	6.6 \pm 1.9 (23)	8.0 \pm 2.1 (23)	0.03
Palatoplasty	13.8 \pm 2.2 (26)	14.1 \pm 2.9 (26)	0.79

before surgery. For the children whose lip/nose repairs were delayed, none underwent lip adhesion or NAM before definitive lip repair. This delay in lip repair without presurgical intervention could complicate family dynamics and psychosocial functioning, especially without additional maternal/parental support.

Similar concerns surround timing of palate repair. Speech development, and outcomes are optimized by performing palatoplasty before the onset of speech production and a delay in care can affect psychosocial and functional outcomes.¹⁸ An intact palate not only prevents nasal regurgitation during feeding, but also allows for velopharyngeal competence and thus proper speech development and production.^{11,12,19} Previous studies suggest that the presence of an unrepaired cleft palate during the early speech development can lead to compensatory, maladaptive articulation patterns, hypernasality and nasal air emissions.^{7,8,20,21} Superior articulation and resonance outcomes have been observed in patients who underwent palatoplasty before significant lexical advancement.²² Decreased speech intelligibility can also be socially stigmatizing with negative psychological effects.⁸ Fortunately, palate repair was not significantly delayed at our institution. However, other centers which had higher rates of endemic disease may have experienced more significant delays in care.

While the moratorium on elective surgeries has been lifted, delays continued at our center due to COVID-19-related concerns. Reasons for ongoing delays included an initial case backlog immediately after the resumption of elective surgeries, family preference, social and economic difficulties, and diagnoses and/or exposure to COVID-19 in the immediate preoperative period. Understanding the effects of these delays is critical to approaching subsequent patient encounters with sensitivity. Surgery is often seen as the “cure,” but also is a great source of anxiety for families, especially as the date of surgery approaches.^{23,24} COVID-19 and the uncertainty surrounding surgery scheduling likely magnified this stress for families. The avoidance of in-person clinic visits during the start of the pandemic may have contributed to a perceived lack of support for affected families. Although the dramatic growth of telemedicine during the pandemic facilitated care for many, not all families have access to devices with audio/video capabilities and may have experienced difficulty accessing care. Social distancing during the COVID-19 pandemic may also have negative implications for families of children with CL/P, as their perceived support from family and friends may have been lower during these isolating times.²⁵ Further investigation is needed to understand the downstream effects these delays are having on the remainder of these patients’ care.

This study is limited to a retrospective review of short-term data; as such we are limited in the information available, and we are not able to definitively say what the effects of these delays in surgical care will be over time. Additionally, the study is limited because it is an observational study and potential confounders, such as patient’s social situation or family preference, cannot be controlled for. Given that it is possible these ongoing delays may affect patients’ psychosocial and speech development, it is important for providers caring for CL/P patients to remain alert to delays that may be occurring in the care of their patients. While follow-up studies are needed to determine effects of the extent of the delays observed due to the COVID-19 pandemic specifically, it is clear that significant delays in care are ideally avoided.

CONCLUSIONS

The COVID-19 pandemic caused delays at each stage of repair for CL/P-related procedures; however, only primary cleft lip and nose repair were significantly affected. These results are in accordance with the guidelines set forth by the ACPA. While the impact of isolated delays in care may be negligible, the downstream effect of such delays on cleft outcomes remains uncertain as the psychosocial aspects are not inconsequential and the cumulative effect on multiple procedures over the course of a cleft patient’s care remains unknown. Follow-up studies are needed to characterize the long-term effects and speech outcomes of these delays. As the pandemic continues, further delays may be expected while providers adapt to this rapidly evolving situation, especially in those practicing in resource-constrained health systems. This study emphasizes the importance of remaining vigilant regarding the care of this vulnerable population during this challenging time.

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REFERENCES

1. COVISOrg Collaborative. Elective surgery cancellations due to the COVID-19 pandemic: global predictive modelling to inform surgical recovery plans. *Br J Surg*. 2020;107:1440–1449.
2. ACPA. ACPA statement on cleft lip/palate related surgeries and team care during the COVID-19 pandemic. Published April 1, 2020. Available at: https://acpa-cpf.org/wp-content/uploads/2020/04/ACPA-Position-Statement-During-COVID-19_4.1.2020.pdf. Accessed August 10, 2020.
3. Wong S, Payton JI, Lombana NF, et al. A protocol for safe head and neck reconstructive surgery in the COVID-19 pandemic. *Plast Reconstr Surg Glob Open*. 2020;8:e3258.

4. Schoenbrunner A, Sarac B, Gosman A, et al. Considerations for pediatric craniofacial surgeons during the COVID-19 outbreak. *J Craniofac Surg.* 2020;31:e618–e620.
5. Schoenbrunner AR, Sarac BA, Janis JE. A summary of recommendations for plastic surgeons during the coronavirus disease 2019 outbreak. *Plast Reconstr Surg Glob Open.* 2020;8:e3039.
6. Hammoudeh JA, Imahiyerobo TA, Liang F, et al. Early cleft lip repair revisited: a safe and effective approach utilizing a multidisciplinary protocol. *Plast Reconstr Surg Glob Open.* 2017;5:e1340.
7. Follmar KE, Yuan N, Pendleton CS, et al. Velopharyngeal insufficiency rates after delayed cleft palate repair: lessons learned from internationally adopted patients. *Ann Plast Surg.* 2015;75:302–305.
8. Pearson GD, Kirschner RE. Surgery for cleft palate and velopharyngeal dysfunction. *Semin Speech Lang.* 2011;32:179–190.
9. Calteux N, Schmid N, Hellers J, et al. [Neonatal cleft lip repair: perioperative safety and surgical outcomes]. *Ann Chir Plast Esthet.* 2013;58:638–643.
10. GRABER TM. Changing philosophies in cleft palate management. *J Pediatr.* 1950;37:400–415.
11. Worley ML, Patel KG, Kilpatrick LA. Cleft lip and palate. *Clin Perinatol.* 2018;45:661–678.
12. Smith DM, Losee JE. Cleft palate repair. *Clin Plast Surg.* 2014;41:189–210.
13. Rohrich RJ, Gosman AA. An update on the timing of hard palate closure: a critical long-term analysis. *Plast Reconstr Surg.* 2004;113:350–352.
14. Sarac BA, Schoenbrunner AR, Wilson SC, et al. The impact of COVID-19-based suspension of surgeries on plastic surgery practices: a survey of ACAPS members. *Plast Reconstr Surg Glob Open.* 2020;8:e3119.
15. Murray L, Hentges F, Hill J, et al; Cleft Lip and Palate Study Team. The effect of cleft lip and palate, and the timing of lip repair on mother-infant interactions and infant development. *J Child Psychol Psychiatry.* 2008;49:115–123.
16. Grollemund B, Dissaux C, Gavelle P, et al; CLIP team (Cleft Lip & palate Infant Parent). The impact of having a baby with cleft lip and palate on parents and on parent-baby relationship: the first French prospective multicentre study. *BMC Pediatr.* 2020;20:230.
17. Emeka CI, Adeyemo WL, Ladeinde AL, et al. A comparative study of quality of life of families with children born with cleft lip and/or palate before and after surgical treatment. *J Korean Assoc Oral Maxillofac Surg.* 2017;43:247–255.
18. Dorf DS, Curtin JW. Early cleft palate repair and speech outcome. *Plast Reconstr Surg.* 1982;70:74–81.
19. Naran S, Ford M, Losee JE. What's new in cleft palate and velopharyngeal dysfunction management? *Plast Reconstr Surg.* 2017;139:1343e–1355e.
20. Hsieh ST, Zhu WY, Liou T, et al. Speech and audiology outcomes following single-stage versus early 2-stage cleft palate repair. *Plast Reconstr Surg Glob Open.* 2019;7(8 suppl):56–57. . eCollection 0000582019 Aug.
21. Willadsen E, Boers M, Schöps A, et al. Influence of timing of delayed hard palate closure on articulation skills in 3-year-old Danish children with unilateral cleft lip and palate. *Int J Lang Commun Disord.* 2018;53:130–143.
22. Chapman KL, Hardin-Jones MA, Goldstein JA, et al. Timing of palatal surgery and speech outcome. *Cleft Palate Craniofac J.* 2008;45:297–308.
23. Fletcher AJ, Hunt J, Channon S, et al. Psychological impact of repair surgery in cleft lip and palate. *Int J Clin Pediatr.* 2012;1:93-96.
24. Hasanzadeh N, Khoda MO, Jahanbin A, et al. Coping strategies and psychological distress among mothers of patients with non-syndromic cleft lip and palate and the family impact of this disorder. *J Craniofac Surg.* 2014;25:441–445.
25. Baker SR, Owens J, Stern M, et al. Coping strategies and social support in the family impact of cleft lip and palate and parents' adjustment and psychological distress. *Cleft Palate Craniofac J.* 2009;46:229–236.