

Systematic Review

THE IMPACT OF SOCIAL STIGMA ON CHILD PATIENTS WITH CLEFT LIP AND PALATE

Yuni Ariani^{1*} , Diska Astarini²¹Department of Plastic Reconstructive and Aesthetic Surgery, dr. Ramelan Central Naval Hospital, Surabaya, Indonesia²Department of Obstetrics and Gynecology, Universitas Hasanuddin, Makassar, Indonesia

ABSTRACT

Social rejection still happens to a large number of children who have cleft lip and palate. Stigma has a detrimental impact on children with cleft in the aspects of education, employment, marriage, and community acceptance, which can be exacerbated by barriers to high-quality child care. A literature study related to this topic was conducted by searching for articles from 2018 to October 2021 through three databases (i.e., PubMed, Embase, and Medline). The term used for the literature search was "cleft", which generated results that were sorted alphabetically and by relevance. This yielded 15 articles that focused on social stigma's influence on cleft lip and palate patients. In conclusion, the articles generally reported that the awareness of social stigma impacts on children with cleft lip and palate are lacking in various areas. It is important to identify social stigma's influence on children with cleft lip. This can help surgeons prioritize resource allocation and provide further evidence in incorporating quality of life measures into the treatment outcome assessment.

Keywords: Social stigma; cleft lip; children; social determinant of health

***Correspondence:** Yuni Ariani, Department of Plastic Reconstructive and Aesthetic Surgery, dr. Ramelan Central Naval Hospital, Surabaya, Indonesia. Email: dokteryuniariani@gmail.com

Article history

•Submitted 30 Nov 2022 • Revised 10 Jan 2023 • Accepted 2 Feb 2023 • Published 10 Mar 2023

How to cite: Ariani Y & Diska A (2023). The impact of social stigma on child patients with cleft lip and palate. *Folia Medica Indonesiana*, 59 (1), 70-74. <https://doi.org/10.20473/fmi.v59i1.39641>



Copyright: © 2023 Folia Medica Indonesiana.

This is an open-access article distributed under the terms of the Creative Commons Attribution License as stated in <https://creativecommons.org/licenses/by-nc-sa/4.0/deed.id>.

pISSN: 2355-8393, eISSN: 2599-056x

Highlights:

1. Awareness of social stigma impact on children with cleft lip and palate is lacking in various areas.
2. Identifying social stigma's influence on children with cleft lip is important to help surgeons prioritize resource allocation and provide further evidence in incorporating quality of life.

INTRODUCTION

One of every 700 children is born with a cleft lip, making it the most common congenital craniofacial anomaly in the world (Tanaka et al. 2012, Fell et al. 2014, Allori et al. 2017). The cleft lip and palate prevalence rates in children vary widely across socioeconomic class, gender, and ethnicity. However, children with cleft lip and palate may encounter additional challenges in low- and middle-income countries (LMICs). Significant disadvantages, such as speech difficulties, delays in physical and cognitive development, persecution by bullies, and social exclusion, can arise from cleft lip later in life.

Children who have cleft lip and palates are more susceptible to psychosocial influences, such as those

from the community on their ability to learn, work, and even get married. If this persists into adulthood, higher-level work and education are more difficult to obtain for them (Camille et al. 2014, Mzezewa et al. 2014, Maine et al. 2017). There is currently no literature or scientific writing that analyzes the impact of social stigma on child patients in a variety of areas. Clarifying and identifying the effects of social stigma can help with resource allocation and gathering evidence to incorporate quality of life measures into outcome assessment in the care of children with cleft lip (Allori et al. 2017).

MATERIALS AND METHODS

This study followed the screening process guideline of systematic reviews and was registered in the

International Prospective Register of Systematic Reviews (PROSPERO). Three databases (i.e., PubMed, Embase, and Medline) were utilized in the literature search, limited to studies from 2018 to October 2021. The term "cleft" was used as a keyword in the literature search to produce maximum results, which were then sorted alphabetically and by relevance. Table 1 exhibits the literature search process using the Medical Subject Headings (MeSH) in order to yield article titles from the three databases. The inclusion criteria in this study were limited to literature discussing social stigma and cleft lip and palate in pediatric patients, who were no older than 18 according to the United Nations.

The literature search generated 477 entries, 32 of which included full-text articles. After reviewing the literature, 15 articles were identified to match the inclusion criteria of this study. Evidence-based theories were utilized to provide descriptive analyses, which were employed in the article review process. The selected literature reported several factors that would arise if cleft lip patients did not receive special care.

RESULTS

Only fifteen studies found in the literature search results that addressed the impact of social stigma on pediatric patients with cleft lip and palate (Table 2). These studies indicated a 47% involvement of international cooperation in the care of child patients with cleft lip and palate. The majority of the studies were conducted in India (n=4) and Nigeria (n=4). Several factors were investigated in the studies, i.e., public trust (60%), social influence (46%), marriage (46%), education (40%), employment (33%), and psychological distress (20%).

There is a belief in the society that cleft lip and palate are caused by the parents' actions or faults (Adeyemo et al. 2016), such as punishing their children with abuse to their faces (Maine et al. 2017). Children with cleft lip were frequently perceived as though they were not human (Conway et al. 2015). Most people realize that cleft lip affects only the lip and face. However, they do not realize its etiology and how to prevent it (Wong Riff et al. 2017).

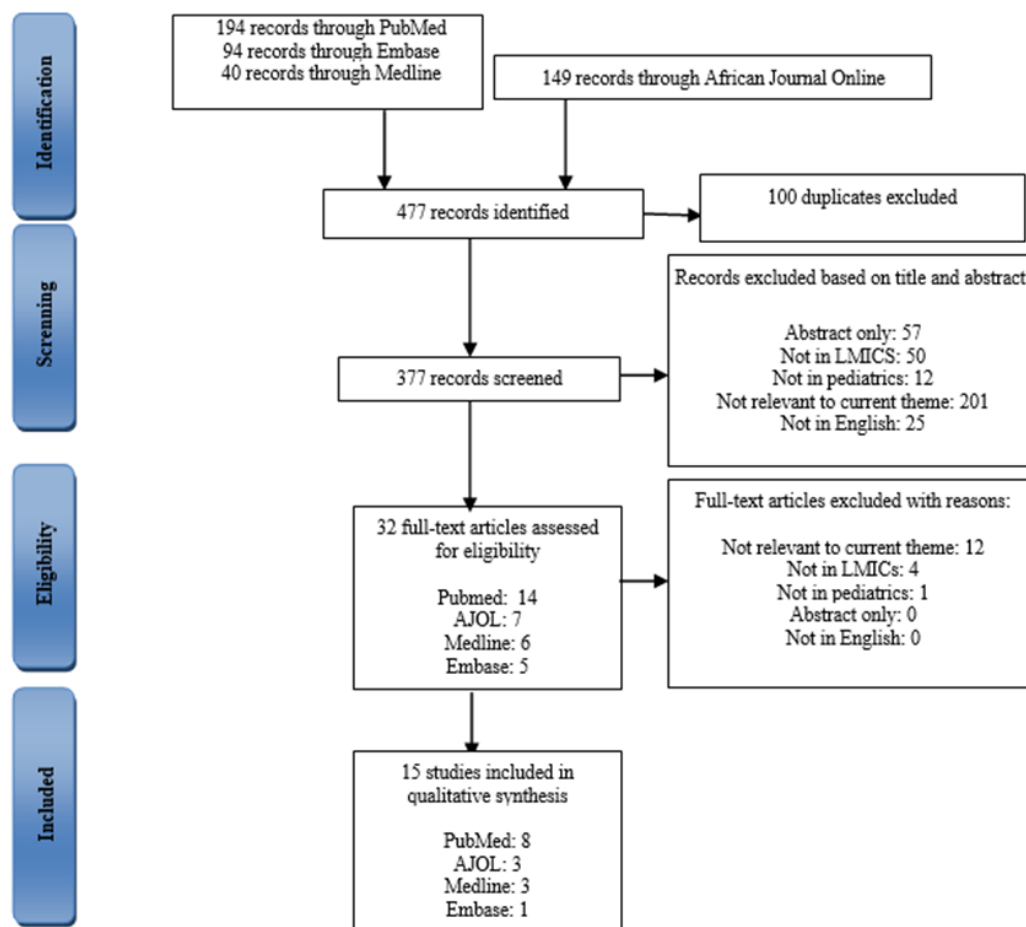


Figure 1. Screening process diagram of the literature search results.

Table 1. The Medical Subject Headings (MeSH) for the literature search through PubMed, Embase, and Medline.

	Where	Who	What	
PubMed	Poverty	Child under 18 years old	Cleft lip palate	
	Poverty area		Cleft lip	
	Developing country		Cleft face	
	Social welfare		Cleft lip	
	Rural health		Unilateral cleft lip	
	Rural population		Cleft lip	
	Hospitals, rural		face	
	Rural health service		palate	
	International agency		Cleft lip palate	
	Socioeconomic factor			
	Embase	Socio International cooperation	Infant <to one year> Child	Cleft lip
		Social welfare	<unspecified age>	Cleft lip palate
		Developing country	1 to 6 years old child	Cleft face
Poverty		7 to 12 years old child	Cleft lip nose	
Rural health care		13 to 17 years old child	Unilateral cleft lip	
Rural population			Cleft lip face	
Rural area			palate	
Urban rural difference			Cleft palate	
Medline		Poverty	Child aged 23 months	Cleft lip
		Poverty	Child aged 0 to 18 years	Cleft palate
	Develop country	Child aged 1 month		
	Social welfare	Child aged 1 to 23 months		
	Rural health	Child aged 1 to 5 years		
	Rural population	Child aged 6 to 12 years		
	Hospital	Child aged 13 to 18 years		
	Rural health service			
	International agency			
	Socioeconomic factor			

Children with cleft lip and palate typically faced social repercussions, such as social isolation or being isolated from their surroundings (Yao et al. 2016, Wong Riff et al. 2017, Crerand et al. 2017, American Academy of Pediatric Dentistry 2019). Children with cleft lip and palate might be neglected by one or both parents since their physical appearance was seen as "terrifying" by their parents (Crerand et al. 2017, Hlongwa & Rispel 2018). However, several studies reported that there were parents who had a favorable attitude and interacted closely with their children who had cleft lip and

palate (Worth et al. 2017, Soeselo et al. 2019).

Table 2. The selected literature on children with cleft lip and the influence of society.

Research	Type of study
Tanaka	Retrospective
Allori	Cross-sectional
Camille	Retrospective
Abid	Qualitative
El-Shazly	Qualitative
Fadeyibi	Qualitative
Fell	Qualitative
Klassen	Case study
Mzezewa	Cohort
Naram	Qualitative
Butali	Qualitative
Owotade	Cross-sectional
Parmar	Narrative
Bluher	Qualitative
Mednick	Qualitative

Many studies have examined how children with cleft lip and palate struggled to attend school. Children with cleft lip were not allowed to attend school due to their "terrifying" appearance, which was often not tolerated in school. They were either denied admission to school because their appearance would frighten other children or they refused to attend school due to bullying. Surgery might have benefits for children with cleft lip and palate to avoid these situations (Kumar et al. 2014, Ness et al. 2015, American Cleft Palate-Craniofacial Association 2017). Despite the original size and completeness of the cleft, surgery might help to establish the symmetry of nasal width, philtral height, horizontal lip length, and vertical lip height in cleft lip and palate patients (Datusanantyo et al. 2021).

In the studies reviewed, children with cleft lip and palate might endure psychological distress because they were perceived as a burden on their families. Furthermore, they regarded their parents as having negative feelings toward them, resulting in a higher proportion of children and adolescents with cleft lip and palate feeling depressed. Social dysfunction, which restricted their opportunity to interact with peers, also contributed to their psychological distress. The children were often not allowed to interact with other people at all, indicating a higher incidence of social dysfunction (Worth et al. 2017, World Health Organization 2018).

DISCUSSION

The social repercussions on children with cleft lip and palate stemmed from misleading cultural belief that those children were born with supernatural curse and would bring negative influence to people (Wong

Riff et al. 2017). This would result in bullying and, in some cases, family seclusion for those children. This also occurred in school causing children to drop out and encounter barriers to higher education. In adulthood, they would be seen as a burden as they struggled to be hired for paid employment.

Cleft lip and palate surgery and reconstruction could provide these children hope for education, employment opportunities, social acceptance, and improved marriage prospects. It is advised to seek surgical treatment as early as possible. However, a study found that a patient's age at the time of cleft palate surgery was strongly related to the parents' income and access to information (Ningrum et al. 2021). Adults with severely neglected cleft lip and palate could still be treated, but communication difficulties accompanying these conditions might persist (Arista & Hutagalung 2021). Providing education, support, and outreach to people with cleft lip and palate could help them to integrate into the societal system and change the social stigma (Conway et al. 2015, Crerand et al. 2017). Primary care, including treatments for children with cleft lip and palate, should include physical, psychological, and social components (Roosihermatie et al. 2018).

Strength and limitations

The literature selected for this study had some limitations. Many of the articles did not employ the Patient-Reported Outcome Measures (PROMs), which might include questions about education, social impact, and psychological functioning for cleft lip patients. It is recommended to use the PROMs that can be a valid tool to assess and understand the impact of cleft lip cases and treatments. However, treatments for children with cleft lip and palate would only succeed if there is a solid cooperation between governments and non-governmental organizations to reduce social stigma and improve public education.

This study also had limitations because many cases of cleft lip and palate were unreported in some places. This review study covered only literature in English, so research published in other languages was excluded. To uncover further evidence of the social impact on children with cleft lip and palate, open discussion with colleagues from various regions is required.

CONCLUSION

Children with cleft lip and palate encounter social stigma, which has a major impact on their psychology and development. However, awareness of this phenomenon is still very lacking in many regions. The Patient-Reported Outcome Measures

(PROMs) are recommended for assessing surgical treatment or reconstruction's effectiveness in reducing the psychosocial burden on children with cleft lip and palate, particularly in low- and middle-income countries (LMICs). Collaboration between governments and non-governmental organizations is also suggested to decrease social stigma and enhance public education as part of these children's healthcare.

Acknowledgment

None.

Conflict of interest

None.

Funding disclosure

None.

Author contribution

YA designed the study, gathered the data, and approved the final manuscript. DA designed the study and prepared the manuscript.

REFERENCES

- Adeyemo W, James O, Butali A (2016). Cleft lip and palate: Parental experiences of stigma, discrimination, and social/structural inequalities. *Annals of Maxillofacial Surgery* 6, 195. doi: 10.4103/2231-0746.200336.
- Allori AC, Kelley T, Meara JG, et al (2017). A standard set of outcome measures for the comprehensive appraisal of cleft care. *The Cleft Palate-Craniofacial Journal* 54, 540–54. doi: 10.1597/15-292.
- American Academy of Pediatric Dentistry (2019). Policy on the management of patients with cleft lip/palate and other craniofacial anomalies. American Academy of Pediatric Dentistry. [Web Page]
- American Cleft Palate-Craniofacial Association (2017). Neonatal cleft lip and palate: Instructions for newborn nurseries. ACPA. [Web Page]
- Arista TH, Hutagalung MR (2021). Successful pharyngoplasty after Le Fort I advancement in a severely neglected cleft lip and palate patient. *Jurnal Rekonstruksi dan Estetik* 4, 24. doi: 10.20473/jre.v4i1.24351.
- Camille A, Evelyne A-K, Martial AE, et al (2014). Advantages of early management of facial clefts in Africa. *International Journal of Pediatric Otorhinolaryngology* 78, 504–6. doi: 10.1016/j.ijporl.2013.12.031.
- Conway JC, Taub PJ, Kling R, et al (2015). Ten-year

- experience of more than 35,000 orofacial clefts in Africa. *BMC Pediatrics* 15, 8. doi: [10.1186/s12887-015-0328-5](https://doi.org/10.1186/s12887-015-0328-5).
- Crerand CE, Conrad AL, Albert M, et al (2017). The Americleft psychosocial outcomes project: A multicenter approach to advancing psychosocial outcomes for youth with cleft lip and palate. *Clinical Practice in Pediatric Psychology* 5, 65–76. doi: [10.1037/cpp0000172](https://doi.org/10.1037/cpp0000172).
- Datusanantyo RA, Hutagalung MR, Rizaliyana S, et al (2021). Anthropometric outcome of primary unilateral cleft lip repair in Indonesia. *The Cleft Palate-Craniofacial Journal* 58, 1236–41. doi: [10.1177/1055665620982757](https://doi.org/10.1177/1055665620982757).
- Fell MJ, Hoyle T, Abebe ME, et al (2014). The impact of a single surgical intervention for patients with a cleft lip living in rural Ethiopia. *Journal of Plastic, Reconstructive & Aesthetic Surgery* 67, 1194–200. doi: [10.1016/j.bjps.2014.05.019](https://doi.org/10.1016/j.bjps.2014.05.019).
- Hlongwa P, Rispel LC (2018). “People look and ask lots of questions”: Caregivers’ perceptions of healthcare provision and support for children born with cleft lip and palate. *BMC Public Health* 18, 506. doi: [10.1186/s12889-018-5421-x](https://doi.org/10.1186/s12889-018-5421-x).
- Kumar S, Kroon J, Laloo R (2014). A systematic review of the impact of parental socio-economic status and home environment characteristics on children’s oral health related quality of life. *Health and Quality of Life Outcomes* 12, 41. doi: [10.1186/1477-7525-12-41](https://doi.org/10.1186/1477-7525-12-41).
- Maine RG, Linden AF, Riviello R, et al (2017). Prevalence of untreated surgical conditions in rural Rwanda. *JAMA Surgery* 152, e174013. doi: [10.1001/jamasurg.2017.4013](https://doi.org/10.1001/jamasurg.2017.4013).
- Mzewewa S, Hamese K, Mashego TAB (2014). Neonatal cleft lip repair in babies with breastfeeding difficulties at Polokwane Mankweng Hospital Complex. *South African Journal of Child Health* 8, 157. doi: [10.7196/sajch.693](https://doi.org/10.7196/sajch.693).
- Ness AR, Wills AK, Waylen A, et al (2015). Centralization of cleft care in the UK. Part 6: A tale of two studies. *Orthodontics & Craniofacial Research* 18, 56–62. doi: [10.1111/ocr.12111](https://doi.org/10.1111/ocr.12111).
- Ningrum LP, Saputro ID, Zarasade L (2021). Corelation of parents’ profiles of children with late cleft repair in Surabaya cleft lip and palate centre (January 2015–December 2017). *Jurnal Rekonstruksi dan Estetik* 5, 18. doi: [10.20473/jre.v5i1.24319](https://doi.org/10.20473/jre.v5i1.24319).
- Roosihermatie B, Isfandari S, Fauzia Y (2018). Case Report: Diagnosis and treatment of psychosomatic gastritis at a Primary Health Clinic in West Surabaya, Indonesia. *Folia Medica Indonesiana* 54, 155. doi: [10.20473/fmi.v54i2.8867](https://doi.org/10.20473/fmi.v54i2.8867).
- Soeselo DA, Suparman AS, Budi AS (2019). Parents’ knowledge, attitude and behaviour toward cleft lips and cleft palate in Kencana Hospital, Serang, Banten. *Journal of Craniofacial Surgery* 30, 1105–8. doi: [10.1097/SCS.00000000000005352](https://doi.org/10.1097/SCS.00000000000005352).
- Tanaka SA, Mahabir RC, Jupiter DC, et al (2012). Updating the epidemiology of cleft lip with or without cleft palate. *Plastic and Reconstructive Surgery* 129, 511e-518e. doi: [10.1097/PRS.0b013e3182402dd1](https://doi.org/10.1097/PRS.0b013e3182402dd1).
- Wong Riff KWY, Tsangaris E, Goodacre T, et al (2017). International multiphase mixed methods study protocol to develop a cross-cultural patient-reported outcome instrument for children and young adults with cleft lip and/ or palate (CLEFT-Q). *BMJ Open* 7, e015467. doi: [10.1136/bmjopen-2016-015467](https://doi.org/10.1136/bmjopen-2016-015467).
- World Health Organization (2018). Child causes of death, 2000–2016. WHO. [Web Page].
- Worth V, Perry R, Ireland T, et al (2017). Are people with an orofacial cleft at a higher risk of dental caries? A systematic review and meta-analysis. *British Dental Journal* 223, 37–47. doi: [10.1038/sj.bdj.2017.581](https://doi.org/10.1038/sj.bdj.2017.581).
- Yao CA, Swanson J, Chanson D, et al (2016). Barriers to reconstructive surgery in low- and middle-income countries. *Plastic and Reconstructive Surgery* 138, 887e-895e. doi: [10.1097/PRS.0000000000002656](https://doi.org/10.1097/PRS.0000000000002656).

