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Incidence of Cleft Lip and Palate Between Siblings in South Indian Population

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Abstract

Aim: This study set out to look at the incidence of cleft lip and palate between siblings in the South Indian population Materials and methods: Saveetha Dental College and Hospital's Oral & Maxillofacial Surgery Unit treated fifty cleft patients who also have siblings. The incidence of clefting in the family was researched using the history of consanguineous marriage. Results: Eighty percent of patients had a cleft history, most commonly in younger siblings, while also there is an Increased incidence of the cleft with parents who have consanguineous marriage.

Conclusion: There is a strong correlation between CP and being related by blood. This study's findings on the frequency of consanguinity and the resulting heaviness of congenital disabilities highlight the critical importance of establishing a national registry for congenital anomalies such as craniofacial defects.

1. Introduction:

The most common congenital anomaly in humans is a cleft lip, cleft palate, or orofacial cleft (OFC) cleft lip and palate (CL/P) [1]. The incidence and prevalence of OFC vary greatly among geographical areas. From a global viewpoint, there is a wealth of epidemiological data accessible; yet, many countries lack access to this information [2]. One in every 600 newborns is affected with CL/P, according to estimates from throughout the world. While there is little data on the prevalence of OFC in India, the most recent estimates place the rate at 1.09 per 1,000 live births. In contrast, Mossey and Little [4] estimated a national incidence of 28,600. Nonsyndromic OFC is a complex condition with several causes. Multiple variables, including genetics, environmental factors, and cultural context, contribute to the development of CL/P. In addition to being linked to 400 various syndromes, genetic factors play a crucial role in the genesis of CL/P.

Cultural customs make it normal for Indians to marry within their own families. Consanguineous marriages accounted for 7.40% of cleft lip cases and 14.29% of cleft palate cases in a research by Akshay et al. A thorough screening for further birth abnormalities should be performed on all infants with cleft lip and palate, but particularly those born prematurely and with a low birth weight. [18]It is well recognized that close genetic relatives have a greater risk of passing on genetic defects, especially autosomal recessive ones. Cultural practices of consanguinity may have a causal influence in epigenetic alterations that lead to birth defects. Consanguinity is a cultural component that interacts with genetic and environmental variables to contribute to the genesis of CL/P [5]. Many cultures practice consanguineous marriage, which involves two people who are very closely connected by blood, such as first cousins or even uncle and niece. It is estimated that 10.4% of all marriages in the world are second cousins or closer, and that this tight link is carried on through subsequent generations [6]. This study's

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authors wanted to evaluate cleft lip and palate prevalence in a South Indian community since it differs so much throughout the world.

2. Materials and Methods:

Fifty adults with clefts and 50 of their siblings were included in a retrospective research conducted by the Oral and Maxillofacial Surgery Department at Saveetha Dental College and Hospital. Fifteen of the 50 cases were older siblings, while 35 were younger.

The institute's ethics board gave its permission for the research to be conducted in hindsight, and all personal

information was handled in accordance with the standards outlined in the 1975 and 1983 updates to the Helsinki Declaration. The hospital's electronic medical records system was used to review the patient's histories. The patient's family history of consanguineous marriage detailed with cleft histories and siblings (elder or younger) with a cleft were also recorded.

IBM SPSS statistics 20.0 (Chicago, IL) was used to conduct the chi-square test for the purpose of calculating the cleft prevalence among patients, and differences at the p 0.05 level were regarded to be significant.

3. Results:

Table 1: Incidence of cleft lip and palate with parental history of consanguineous marriage			
	Parental H/O Consanguineous Marriage		
Patient	Yes	No	P value*
Elder	66.7%	33.3%	0.197
Younger	80%	20.0%	0.0004
P value*	0.0035	0.564	

*Chi-Square Test

Table 1 reveals a correlation between a family history of consanguineous marriage and cleft lip and/or palate. Thirty-eight of the fifty individuals with cleft lip and/or palate had a family history of consanguineous marriage. There was a significant difference between older and younger siblings (P Value 0.0004). In contrast, the data for the older sibling group were not statistically significant (P Value = 0.197). Consanguineous marriages between parents also correlate with an increased risk of cleft lip and palate in offspring. (P = 0.0035)



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4. Discussion:

Marriages between blood relatives have been common since the beginning of time. Primitive societies' limited numbers necessitated them, and up until the early 19th century, they were widely practiced all throughout the planet. The benefits and drawbacks of this approach have been debated. Although intermarriage is becoming less common worldwide, it is still common in certain parts of the Middle East, including Saudi Arabia, Iran, and Southern India [7]. Consanguineous marriages occur in a broad variety of places and ways. There are several legal, cultural, religious, and societal considerations [7,8,9] in this system of marriage amongst blood relatives. Consanguinity is practiced differently in different regions of the globe, and this is also true within the Indian subcontinent. This type of marriage is culturally, socially, spiritually, and legally recognized in India, especially in the southern regions. In the cities of Bengaluru and Mysore in the state of Karnataka in South India, 21% of Hindu marriages are between uncle and niece, as observed by Bittles et al. [10]. It is unclear whether the frequency of consanguineous marriage has increased or decreased since the 1990s when statistics last showed a nationwide fall of 1%. Consanguinity's influence on health and illness is a topic that has been explored and studied extensively. No previously published studies from India could be found. consanguinity and common craniofacial malformations, especially OFC, are being investigated. The present research found a 66.7% prevalence of CLP clefts in elder siblings and 80% of CLP clefts in younger siblings. These findings corroborate those of prior research involving Scandinavian peoples. Though cleft incidence is comparable throughout the Scandinavian nations, the prevalence of different forms of clefts varies. 76.0% percent of our patients reported the incidence of a cleft in their families to have a history of consanguineous marriage (38 patients out of 50). It was shown that the inheritance was statistically significant (p=0.0035). Patients diagnosed with CLP had a much higher rate of clefting due to genetic factors. Cleft lip and/or cleft palate tend to follow a similar pattern of inheritance in each family. The percentage of patients who have a history of clefts in their families varies among research [11].

In India, rural regions have greater illiteracy rates and poorer socioeconomic status [12]. Consanguinity also has social benefits, especially in rural areas, including the reduction of domestic violence, the compatibility of in-laws, the financial benefits of a smaller dowry, the reduction of divorce, and the preservation of any landholding. These benefits exceed the many potential drawbacks, like as genetic abnormalities that cause birth defects and infant death. Related research explains why factors such consanguinity, ascertainment, cleft history in the family, and intracluster correlation between OFC-affected siblings are important [14].

Arguments have been made for and against consanguineous marriages ever since Darwin's time. Our understanding of the impact of this practice on health and illness has been greatly enhanced by the development of genetics and its application to the medical sciences for the purpose of diagnosis and prevention. Many congenital deformities, including hearing loss in children, cataracts at birth, heart malformations, and cleft lips and palates, are linked to autosomal recessive genes that are expressed during intimate biological unions [7,11]. Numerous studies have shown links between consanguinity and OFC, although this does not imply that genetic relatedness is the cause of this disorder. Because of this biological plausibility, further study is required. Recently, the gene mutated in consanguineous Palestinian families causing CL/P was discovered [15]. Consanguinity has been hypothesized to have genetic repercussions. [9] Bittles predicted that cultures where consanguineous marriage was common would see an increase in the prevalence of recessive genetic diseases [13]. The consanguineous marriage system is of interest to the public at large due to the wide range of contextual factors at play, including geography, religion, and social class. Consanguinity may rise or fall depending on a variety of factors related to marriage, including the preferences of the bride and husband, the preference of one caste over another, and the prevalence of love marriage over arranged marriage. Consanguinity is regarded to be one of the several etiological risk factors for CL/P and other congenital abnormalities. Considering the various risk factor etiology for CL and palate abnormalities, the findings of this research are important and give useful information on the prevalent consanguineous marriage system practice. Identifying the frequency with which consanguinity occurs would allow for the development of effective preventative measures and public education campaigns. According to the World Health Organization, India should create



a national register for congenital abnormalities. In order to convince policymakers in India of the need of multidisciplinary collaboration in the study of database design, epidemiology, gene-environment interaction, and the prevention of congenital abnormalities including craniofacial deformities [3, 16, 17], this study lays the groundwork for doing so.

The purpose of the current research was confined to describing the association between consanguinity and CL/P using a cross-sectional design.

5. Conclusion:

The current research concludes that there is a statistically significant link between CLP and being related through blood. This study's results provide more evidence that the high rates of consanguinity and the resulting handicap rates necessitate the creation of a national registry for congenital anomalies such craniofacial deformities. Finally, initiatives should be established to avoid consanguinity and educate the public about its consequences.

References:

- Centers for Disease Control and Prevention (CDC). Economic costs of birth defects and cerebral palsy – United States, 1992. MMWR Morb Mortal Wkly Rep 1995;44:6949.
- [2] Shaw W. Global strategies to reduce the health care burden of craniofacial anomalies: Report of WHO Meetings on International Collaborative Research on Craniofacial Anomalies. Cleft Palate Craniofac J 2004;41:23843.
- [3] Reddy SG, Reddy RR, Bronkhorst EM, Prasad R, Ettema AM, Sailer HF, et al. Incidence of cleft Lip and palate in the state of Andhra Pradesh, South India. Indian J PlastSurg2010;43:1849.
- [4] Mossey P, Little J. Addressing the challenges of cleft lip and palate research in India. Indian J PlastSurg 2009;42 Suppl1:S918.
- [5] Aljohar A, Ravichandran K, Subhani S. Pattern of cleft lip and palate in hospitalbased population in Saudi Arabia: Retrospective study. Cleft Palate Craniofac J 2008;45:5926.

- [6] Bittles AH, Black ML. Evolution in health and medicine Sackler colloquium: Consanguinity, human evolution, and complex diseases. Proc Natl Acad Sci U S A 2010;107 Suppl 1:177986.
- [7] Bittles AH, Black ML. Global Prevalence of Consanguinity; 2015. Available from: <u>http://www.consang.net/index.php/Global_</u> prevalence. [Last accessed on 2015 Mar 20].
- [8] Mehndiratta MM, Paul B, Mehndiratta P. Arranged marriage, consanguinity, and epilepsy. Neurol Asia 2007;12:157.
- [9] Bittles AH, Black ML. The impact of consanguinity on neonatal and infant health. Early Hum Dev 2010;86:73741.
- [10] Bittles AH, Shami SA Appaji Rao N. Consanguineous marriage in Southern Asia: Incidence, causes, and effects. In: Bittles AH, Roberts DF, editors. Minority Populations: Genetics, Demography, and Health. London: Macmillan; 1992. p. 1028
- [11] Hagberg C, Larson O, Milerad J. Incidence of cleft lip and palate and risks of additional malformations. Cleft Palate Craniofac J 1998;35:40–5.
- [12] Census India. Literacy and Level of Education in India; 2011. Available from: <u>http://www.censusindia.gov.in/2011census/</u> population_enumeration.html. [Last accessed on 2015 Mar 22].
- [13] Jose BA, Subramani SA, Mokhasi V, Jayan M. Consanguinity and clefts in the craniofacial region: A retrospective casecontrol study. J Cleft Lip Palate Craniofacial Anomalies 2015;2:1137.
- [14] Ravichandran K, Shoukri M, Aljohar A, Shazia NS, AlTwaijri Y, Al Jarba I. Consanguinity and occurrence of cleft lip/palate: A hospitalbased registry study in Riyadh. Am J Med Genet A 2012;158A: 5416.
- [15] Shahin H, Sharaha U, Lee MK, Watts A, King MC, van Aalst J, et al. Targeted Capture and Sequencing Identify Causative Alleles in Simplex and Multiplex Consanguineous Palestinian Families with Orofacial Clefts. 63rd Annual



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Meeting of the American Society of Human Genetics, Boston, USA; 2226 October, 2013.

- [16] World Health Organisation. Global registry and database on craniofacial anomalies. Human genetics programme: International Collaborative Research on Craniofacial Anomalies. Geneva, Switzerland: WHO Publications; 2003.
- [17] World Health Organisation. Addressing the global challenges of craniofacial anomalies. Human genetics programme: Report of a WHO

Meeting on International Collaborative Research on Craniofacial Anomalies. Geneva, Switzerland: WHO Publications; 2006.

[18] Akshay Mohan, Harish Babu, Nivethigaa B. Assessment Of Association Between Age, Gender, Consanguinity And Cleft Deformity - A Retrospective Analysis. Int J Dentistry Oral Sci. 2020;S1:02:0010:48-51. doi: dx.doi.org/10.19070/2377-8075-SI02-