

## ORIGINAL ARTICLE

# Global Variation in Cleft Palate Repairs: An Analysis of 352,191 Primary Cleft Repairs in Low- to Higher-Middle-Income Countries

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**Objectives:** Resources for repair of cleft lip and palate may be lacking in low- and middle-income countries. The Smile Train is a registered charity that supports cleft repair in resource-poor settings. In the global health care challenge, it has been suggested that many babies born with cleft palates are not repaired. This study aims to determine whether any variation exists in the proportion of cleft lip and cleft palate repairs undertaken in low- and middle-income countries.

**Methods:** Data were obtained from the Smile Train database of 352,191 consecutive cleft operations performed between 2008 and 2011 in low- to higher-middle-income countries. The ratio of cleft lip to palate repair was analyzed as a function of geographic region and by country income (gross national income).

**Results:** A significant correlation exists between both the income of a country and its geographical region to the ratio of lip and palate repair procedures undertaken. Higher-income countries had a higher ratio of cleft palate repairs. Countries in sub-Saharan Africa have the lowest proportion of cleft palate repairs.

**Conclusion:** This study emphasizes that many babies born with cleft palates in resource-poor regions do not have their palates repaired. This finding may be explained by an increased neonatal mortality in cleft palate babies. Furthermore, fewer isolated palatal clefts may present to an appropriate health care facility or there may be a reluctance to treat cleft palate due to concerns regarding higher perioperative risks or the lack of available surgical and anesthetic expertise.

KEY WORDS: *cleft lip, cleft palate, developing world, neonatal mortality, resource-poor economy, Smile Train*

Cleft lip and cleft palate constitute one of the most commonly occurring congenital abnormalities and represent a global health care challenge (World Health Organization, Human Genetics Programme, 2002). There is significant variation in the global incidence of orofacial clefting, and the provision of treatment depends on numerous factors including gender, ethnicity, geographic region, and socioeconomic status (Rozendaal et al., 2012). The Smile Train charity has made a significant impact on the global management of patients with cleft lip and cleft

palate by supporting the provision of orofacial cleft care in resource-poor countries over the past decade (Hubli and Noordhoff, 2013). However, the management of cleft lip and palate remains an international challenge.

In a retrospective analysis of cleft practice in Uganda, it was noted that a significant discrepancy exists between the proportion of cleft palates being repaired (compared with the rate cited elsewhere in the literature) and the number of children born with cleft palate deformity (Dreise et al., 2011; Wilson and Hodges, 2012). This study investigates whether this observation is an isolated finding or whether it is seen elsewhere in other resource-poor countries.

## METHOD

All cleft operative procedures recorded in the Smile Train database were interrogated for the period between January 1, 2008, and December 31, 2011. The Smile Train database is composed of Smile Train reports that are completed by the Smile Train partner at the time of surgery. Only primary repairs were included in the analysis. Patient demographic data, type of cleft deformity, and the date and nature of the operative procedure were recorded. Each country was

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TABLE 1 Number of Cleft Repairs and Demographic Data per Geographic Region

<i>Geographic Region</i>	<i>Unilateral Lip</i>	<i>Bilateral Lip</i>	<i>Palate</i>	<i>Total Repairs by Region</i>	<i>Mean Age in Years Lip/Palate</i>	<i>Male:Female Ratio</i>
East Asia and Pacific	61,933	11,704	62,549	136,186	3.7/7.3	1.49:1
Europe and Central Asia	1410	319	2120	3849	1.2/3.0	1.40:1
South Asia	86,204	14,980	65,140	166,324	7.0/6.5	1.58:1
Latin America	6642	1915	6873	15,430	2.8/4.6	1.48:1
Middle East and North Africa	994	235	829	2058	5.1/5.2	1.57:1
Sub-Saharan Africa	21,315	3551	3478	28,344	10.6/6.7	1.64:1

categorized by geographical region and by income (gross national income [GNI]) according to data from the World Bank (2011). For each country, region, and income, the total number of cleft lip and cleft palates repairs was calculated, and subsequently, the percentage of palate repairs was determined. Formal institutional review board approval was not sought for this study; however, the principles of the Declaration of Helsinki were followed.

## RESULTS

Overall, there were 352,191 primary cleft repairs performed in 77 countries during the 4-year period of the analysis. There were 211,202 cleft lip and 140,989 cleft palate procedures. The mean age at surgery (for all procedures) was 6.3 years. For those children having a cleft lip repair, the mean age was 6.1 years, and for those having cleft palate repair, it was 6.7 years. The demographic data for each geographic region studied are summarized in Table 1.

Sixty-two percent of all patients were male; of those having a cleft lip repair, 64% were male, whereas for those having a cleft palate repair, 59% were male. This overall gender discrepancy is reflected throughout the different geographical regions and GNIs as demonstrated in Tables 1 and 2. The lower-income countries have a higher male-to-female ratio than the higher-income countries.

Figure 1 demonstrates the relationship between the percentage of cleft palate repairs (as a proportion of the total number of cleft procedures) and geographic region. Sub-Saharan Africa has the lowest percentage of cleft palate repairs at 12%. These data are also presented cartographically in Figure 2 for every country analyzed (with data for selected developed world countries included for reference and taken from data published elsewhere and not the Smile Train database).

As the wealth of the country increases, the percentage of cleft palate operations increases (Table 2; Fig. 3). Low-income countries (GNI less than \$567) have the lowest percentage of palatal repairs at 22%, compared with 38% in countries of low middle income (GNI \$568 to \$1760) and 52% in upper-middle-income countries (GNI \$1761 to \$6522). This latter figure is approaching that seen in developed countries, where the percentage of cleft repairs is in the order of 70% (Vallino-Napoli et al., 2004; Deacon, 2010).

Table 2 also demonstrates that as the wealth of the country increases, the mean age at which the child has his or her primary cleft lip surgery decreases from 9.5 years to 2.6 years; however, the mean age of primary cleft palate repair actually increases from 6.2 years to 7.0 years.

## DISCUSSION

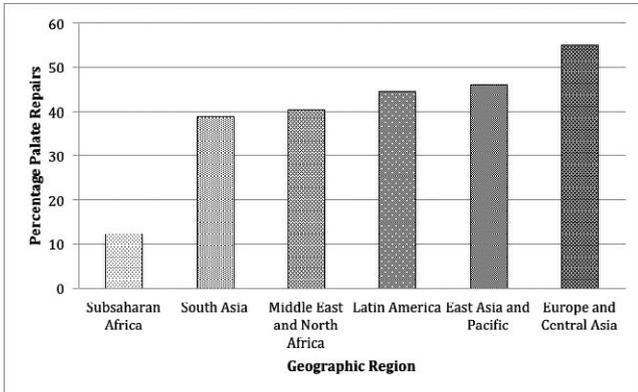
Children with cleft lip and/or palate deformities are an inevitable component of every culture and community throughout the world, and as the global population continues to increase, the global burden of cleft deformities escalates. Early surgical intervention is essential to improve the quality of life for these patients, and in many cases, the operation is life-saving (Cubitt et al., 2012). Despite the variations in overall clefting incidence between geographical and racial groups, the underlying correlation between cleft lip and cleft palate incidence should remain universally constant (Sayetta et al., 1989).

Our analysis has demonstrated that the highest proportion of cleft palates repaired is in the higher-income countries. Evidently, many babies with cleft palate born in low-resource regions are not repaired. It is likely that the increased mortality associated with a cleft palate is more pronounced in low-income countries, with malnutrition being one of the principal factors as these children are unable to adequately breastfeed (Lazarus et al., 1999).

TABLE 2 Percentage of Clefts Presenting for Surgical Repair and the Actual Percentage of Cleft Palates Repaired as a Function of the Gross National Income\*

<i>Gross National Income</i>	<i>Unilateral Lip</i>	<i>Bilateral Lip</i>	<i>Palate</i>	<i>Mean Age in Years Lip/Palate</i>	<i>Male:Female Ratio</i>	<i>Percentage Palate</i>
Low	29,021	4976	9698	9.5/6.2	1.59:1	22
Lower middle	104,392	19,533	74,504	6.6/6.5	1.56:1	38
Upper middle	45,085	8195	56,787	2.6/7.0	1.41:1	52

\* The mean age at which surgical repair of the palate was undertaken is also stated.



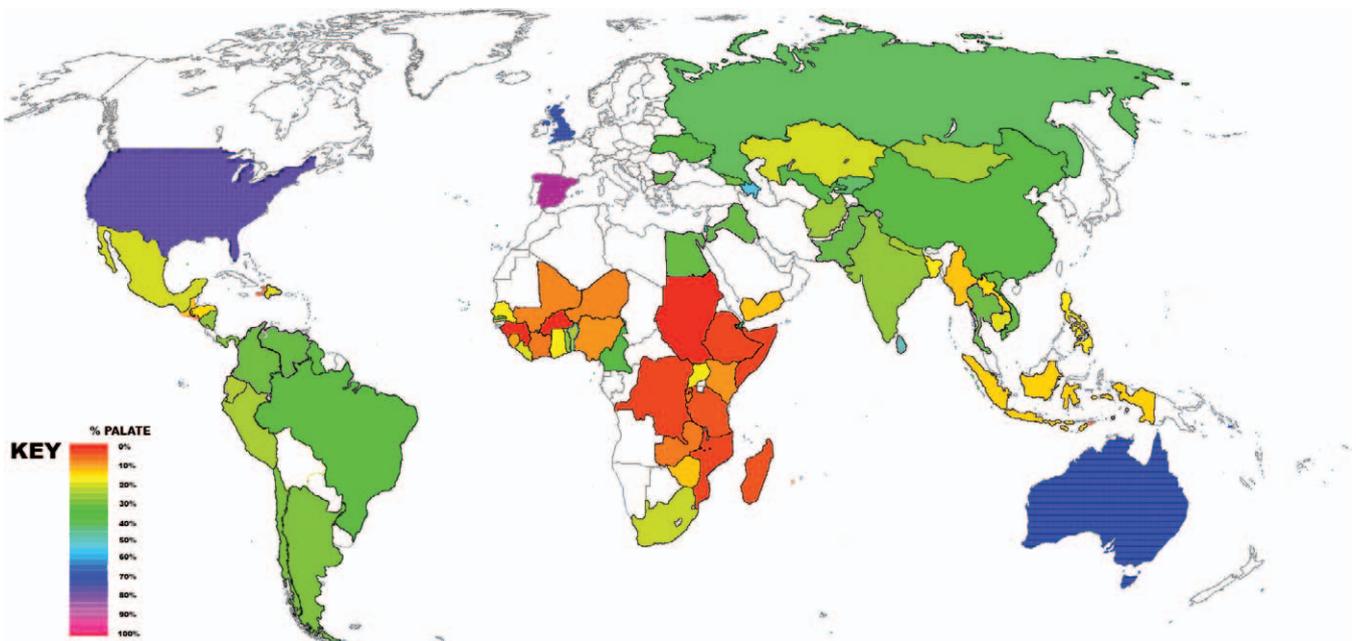
**FIGURE 1** Percentage of cleft palate repairs undertaken as a proportion of all primary cleft procedures as a function of geographic region.

Complex feeding devices are not necessary; simple maneuvers such as cup feeding can have a dramatic influence on neonatal nutritional status in cleft children, but such methods are poorly disseminated (Lang et al., 1994). It is also likely that infanticide may account for a proportion of deaths in children born with orofacial clefting (Strauss, 1985; Scheper-Hughes, 1990).

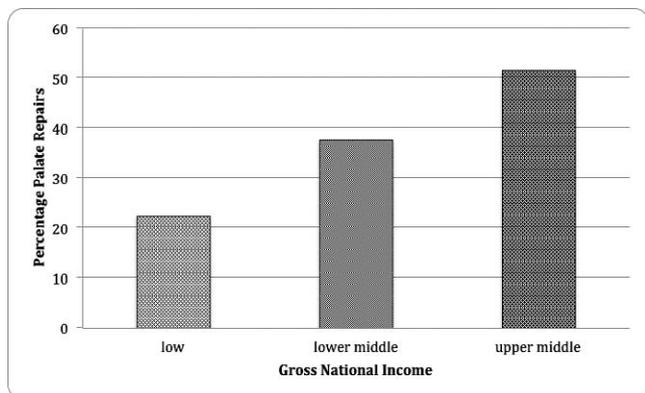
Babies born with an isolated cleft palate may not attend for treatment; an incidence survey recently conducted in Uganda demonstrated that the incidence of isolated cleft palate appears unexpectedly low (Dreise et al., 2011). It is feasible that the hidden isolated cleft palate anomaly may be missed at birth, a situation frequently observed in our practice in Kampala, Uganda. Moreover, our empirical observations indicate that parents frequently do not

understand the benefit of cleft palate repair and therefore fail to seek the advice of a health care professional. This unmet need for surgical care is in contradistinction to cleft lip deformity when patients (or their families) are usually more highly motivated to seek medical advice. Not infrequently, we have noted that children with cleft lip and palate who have had an initial lip repair may fail to return for a definitive cleft palate repair (Hodges et al., 2009). Indeed, this observation has recently encouraged the senior author (A.M.H.) to undertake a synchronous repair of the lip and palate to limit those children who might otherwise forgo their palatal closure (Hodges, 2010). Furthermore, in the context of sub-Saharan Africa, traditional healers might not always refer a cleft child to a practitioner of Western medicine (Dagher and Ross, 2004).

Numerous factors might make the prospect of undertaking palatal surgery less appealing for caregivers, thereby limiting the number of repairs performed. Frequently, babies with a cleft palate are significantly malnourished and require perioperative nutritional support. Malnourished babies have a higher incidence of intercurrent infections, particularly malaria and lower respiratory tract infections. Therefore, more resources are required to prepare a cleft palate patient for theatre. Cleft palates are generally more technically challenging than isolated lip repairs and require greater surgical and anesthetic expertise. Postoperative complications such as bleeding or airway compromise are more likely with palatal rather than with lip surgery. Thus, some centers may opt not to operate on more complex palatal clefts, and thus, in the absence of accessible specialized health care, the child is treated expectantly.



**FIGURE 2** Percentage of cleft repair as a proportion of all primary cleft procedures in the 77 countries studied depicted on a global map. Supplementary data taken from published data are presented for selected developed countries (United Kingdom, Spain, United States, Australia) for comparison (Vallino-Napoli et al., 2004; Genisca et al., 2009; Deacon, 2010; Yanez-Vico et al., 2012).



**FIGURE 3** Percentage of cleft palate repairs as a function of gross national income.

The increased numbers of cleft lip operations and the greater age of cleft lip repair in lower-income countries reflects the burden of cleft lip patients in these countries. Cleft lip deformities do not significantly affect survival; therefore, these patients may live with their cleft lip into old age, or at least until surgical intervention is available. The backlog of these patients in lower-income countries results in the mean age at repair being much greater than the equivalent patient in a lower-middle or upper-middle-income country.

The overall gender discrepancy among patients with cleft lip and palate is reflected throughout the geographic regions and the GNIs. The incidence of cleft deformities is known to vary with gender, with cleft lip and/or palate being more frequent in males (Tolarova and Cervenka, 1998). The variation in the male-to-female ratio is of interest, with lower-income countries having a higher proportion of males to females than the upper-middle-income countries (1.59 versus 1.42). Globally, there is a gender imbalance at birth, which is postulated to be related to natural factors and possibly gender control and deliberate gendercide (Courtwright, 2008). Countries with a high male-to-female ratio at birth (e.g., China at 1.67:1.0) do not have a similar increase in the male-to-female ratio of cleft lip and palate patients (1.4:1.0), whereas countries with a modest gender imbalance at birth (e.g., Ethiopia at 1.03:1.0) have a much greater discrepancy in clefting between the sexes (1.82:1.0). This gender imbalance may support environmental explanations for the etiology of the cleft deformity.

### CONCLUSION

Cleft palate repair varies both by geographic region and by country income. We conclude that in low-resource settings, fewer palate repairs are being performed compared with lip repairs. Possible reasons for this are considered, and we hope that by highlighting this situation, it may assist health care policy makers to develop strategies to reverse this trend.

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