MALNUTRITION IN CHILDREN WITH UNREPAIRED OROFACIAL CLEFT: A SYSTEMATIC REVIEW

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ABSTRACT

Background: The anatomical defect of the oral cavity in children with orofacial clefts presents them with feeding challenges which increase their risk of becoming malnourished. The objective of this review was to investigate the extent of malnutrition among children under 5 years with unrepaired cleft of the lip and/or palate.

Materials and Methods: A systematic literature search of published articles that assessed malnutrition in children with unrepaired cleft lip and/or palate was conducted. Pubmed Central, Cochrane library, Pubmed (MEDLINE) and Google Scholar databases were searched.

Result: A total of 4,489 papers were found of which 8 were included in the review after meeting the inclusion criteria. Malnutrition was found to be higher in isolated cleft palate and cleft lip and palate infants than in isolated cleft lip infants who had nutritional status close to that of non-cleft infants. Syndromic cleft infants were highly malnourished compared to non-syndromic infants. Weight-for-age was the most assessed anthropometric indicator of nutritional status. Average prevalence of underweight (low weight-for-age) was 21.5%.

Conclusion: Malnutrition is high especially within the first year of life in infants with unrepaired cleft lip and/or palate.

Keywords: Cleft lip and palate, malnutrition, growth impairment, failure to thrive

Abbreviations: ICL- Isolated cleft lip, ICP- Isolated cleft palate, CLP- Cleft lip and palate, CP±L- Cleft palate with or without cleft lip, FTT- Failure to thrive, BMI- Body mass index

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1.0 Introduction

Orofacial clefts (cleft lip and cleft palate) are among the most common congenital anomalies globally affecting between 1 in every 1000 live births to 2.69 in every 1000 live births in different parts of the world (McLeod et al., 2004).

The most significant difficulty that babies born with orofacial clefts encounter is feeding and babies having cleft palate with or without cleft lip (CP±L) are more likely to suffer from malnutrition compared to those with cleft lip only (Cubitt et al., 2012). The anatomical defect of the palate and upper lip usually leads to poor latch and generation of insufficient negative pressure at the nipple required for proper intraoral suction as well as poor organization and retention of bolus before swallowing is initiated (Reid, 2004). Infants therefore, have prolonged feeding times and burn more calories in the feeding process than they take in. There also tends to be problems of nasal regurgitation of milk, aspiration and poor maternal-child bonding. These challenges eventually lead to impaired weight gain, malnutrition and a weakened immune system (Ize-Iyanu and Saheeb, 2011: Reid et al., 2006). Although there is lip and palate surgery for correcting this birth defect, some infants are unable to go through the surgical procedures due to low weight and low haemoglobin levels, typical of malnutrition, whereas others die potentially out of malnutrition and infections before their scheduled dates of surgery (Wilson and Hodges, 2011).

The purpose of this systematic review was to sum up recent evidence about the scope of malnutrition in children with unrepaired cleft lip and/or palate.

2.0 Materials and Methods

The online databases of Pubmed Central, Cochrane library, Pubmed (MEDLINE) and Google Scholar were searched for research articles published in English language from the year 2000 to 2017. ‘Cleft lip and palate’ as a free text term was combined with each of the following using the Boolean operator AND; malnutrition, growth impairment, underweight, nutrition, and weight gain. The outcomes of relevance were: underweight, wasting, stunting, failure to thrive and weight gain.

Studies that involved infants and children up to 5 years old with unrepaired cleft lip and/or palate were included in this review. Studies in which participants received lip or palate repair surgery during the duration of the research were included if nutritional status was recorded before primary surgery otherwise, they were excluded. Animal studies were rejected as well as studies in which cleft children used prosthetic palates.

The online searches in the aforementioned databases generated four thousand, four hundred and eighty-nine (4,489) publications out of which 4,470 were rejected based on their titles leaving 19 publications. Further, 4 papers were found to be duplicates and thus rejected as well, leaving a total of 15 publications with the potential of being included in the review. The abstracts and methods of the 15 papers were scrutinized with subsequent elimination of 7
papers that failed to meet the inclusion criteria stated earlier leading to retention of 8 papers. The selection process is displayed in a PRISMA flow diagram as captured in figure 1.

Figure 1: Selection process for published articles

3.0 Result

The following data were extracted from the studies:
- Publication details (author, publication date)
- Country in which study was carried out
- Study design
- Characteristics and number of participants
- Aim or objectives of study
- Main findings
Six (75%) of the 8 articles included in this review were published from 2012 to 2017 which could be interpreted as increased recent research interest in the topic especially in Sub-Saharan Africa which had three (50%) of the six recent studies conducted. Brazil was the most represented country in this review with 3 studies (37.5%), followed by Uganda with two studies (25%). Three studies each (37.5%) were conducted in Africa and South America. There were four cross-sectional studies (50%), three retrospective (37.5%) and one case-control study (12%). Weight-for-age was the most frequently assessed anthropometric indice, being used alone or with other anthropometric indices in 75% of the studies reviewed (Babalola et al., 2016; Miranda et al., 2016; Tabari et al., 2015; Cubitt et al., 2012; Beaumont, 2008 and Montagnoli et al., 2005). One study assessed Failure to thrive (FTT). Three studies (37.5%) employed the use of Z-scores whereas the remaining five studies (62.5%) used percentiles cut-off for determining nutritional status. Four (50%) studies analysed secondary data for the determination of malnutrition among cleft lip and/or palate children. A total of one thousand, nine hundred and fifty-nine (1,959) cleft lip and/or palate children were involved in the studies. All the studies were hospital-based and infants from birth up to 12 months old were the most studied age group (3 studies i.e., 37.5%) followed by infants up to 24 months old (2 studies i.e., 25%). Table 1 provides a summary of the characteristics and key findings of the studies included in this review.

Isolated cleft lip infants (ICL) were found to experience less growth impairment or undernutrition compared to cleft lip and palate (CLP) or isolated cleft palate (ICP) infants with the former having similar growth to that of non-cleft infants (Miranda et al., 2016; Cubitt et al., 2012; Beaumont 2008; Montagnoli et al., 2005). Beaumont (2008) retrospectively assessed failure to thrive in cleft lip and/or palate infants by comparing their weight-for-age data with the UK 1996 Growth Reference Charts and reported that at the time of lip surgery, FTT was present in 7%, 14% and 28% of ICL, UCLP and BCLP infants respectively. At the time of primary palate surgery, prevalence of FTT was 20% in ICP infants but had increased from 14% to 23% in UCLP infants and slightly decreased from 28% to 22% in BCLP infants. FTT prevalence was higher in syndromic than non-syndromic cleft infants (33% and 5% in syndromic and non-syndromic ICP infants respectively at the time of lip surgery). Miranda et al. (2016) and Tabari et al. (2015) reported that 4.1% and 5% respectively of cleft palate with or without cleft lip infants had weight-for-age values above the 95th percentile, suggesting that some CP±L children could be overweight. Based on the classification of malnutrition as below 5th percentile using the WHO Growth Curves as reference, the average prevalence of underweight (low weight-for-age) for the two studies was 21.5%. Based on the WHO Z-Score classification, Babalola et al. (2016) recorded the lowest prevalence of wasting (low-weight-for-length) i.e., 18% whereas Tungotyo et al. (2017) recorded the highest prevalence of 68% in a group of CLP infants aged from birth to 10 months old in Uganda. Severe wasting, i.e. Z-score less than -3 standard deviations, was present in the 39% of the said CLP population.
Table 1: Data Extraction (Summary of studies on malnutrition in children with cleft lip and / or palate from 2000 to 2017)

<table>
<thead>
<tr>
<th>Author/ Year/ country</th>
<th>Study design</th>
<th>Number of participants</th>
<th>Characteristics of participants</th>
<th>Study objective</th>
<th>Main findings/ conclusions</th>
</tr>
</thead>
<tbody>
<tr>
<td>Tungotyo et al. (2017)</td>
<td>Cross-sectional</td>
<td>44</td>
<td>Syndromic and non-syndromic Cleft lip and/or palate infants less than 12 months old</td>
<td>To determine prevalence and factors associated with malnutrition among infants with cleft palate and/or lip at Comprehensive Rehabilitation for Uganda Hospital in Uganda</td>
<td>Malnutrition among cleft lip and/or palate infants born in Uganda was high Malnutrition prevention strategies with special emphasis on feeding/ nutrition counseling for cleft lip and palate infants are needed</td>
</tr>
<tr>
<td>Babalola et al. (2016)</td>
<td>Case-control</td>
<td>100 (50 each for case and control groups)</td>
<td>Children up to 60 months old with non-syndromic cleft lip and/or palate</td>
<td>To assess the influence of non-syndromic cleft lip and/or palate on nutritional status of children</td>
<td>Differences in underweight, wasting and stunting between cleft infants and control were not statistically significant (p value = 0.334, 0.585 and 0.538 respectively)</td>
</tr>
<tr>
<td>Miranda et al. (2016)</td>
<td>Cross-sectional</td>
<td>381</td>
<td>Non-syndromic cleft lip and/or palate infants under 24 months</td>
<td>To study growth of length-for-age, weight-for-age and body mass index of children with cleft lip and palate receiving normal diet To establish specific growth curves for children with cleft palate with or without cleft lip who had not undergone palatoplasty and for children with isolated cleft lip</td>
<td>Compared with non-cleft children, children with isolated cleft palate or cleft lip and palate had impaired weight-for-age and body mass index growth but experienced catch-up growth/recovery from about 5 months old Isolated cleft lip children had similar growth to that of non-cleft children</td>
</tr>
<tr>
<td>Tabari et al. (2015)</td>
<td>Cross-sectional</td>
<td>61</td>
<td>Cleft lip and palate</td>
<td>To evaluate nutritional and</td>
<td>Pre-operative weight-for-age was lower</td>
</tr>
<tr>
<td>Country</td>
<td>Study Type</td>
<td>Study Population</td>
<td>Study Design</td>
<td>Study Objectives</td>
<td>Findings</td>
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<tr>
<td>Iran</td>
<td></td>
<td></td>
<td></td>
<td>Growth status of children with cleft lip and palate before and after lip and palate reconstructive surgery</td>
<td>Nutritional status of children with cleft lip and palate improved after surgery</td>
</tr>
<tr>
<td>Cubitt et al. (2012) Uganda</td>
<td>Retrospective</td>
<td>321 (243 cleft infants, 78 non-cleft infants)</td>
<td></td>
<td>To compare nutritional status of cleft lip and/or palate infants with non-cleft infants</td>
<td>Cleft lip and palate infants had weight-for-age Z-scores that were significantly lower than either isolated cleft lip or non-cleft infants.</td>
</tr>
<tr>
<td>Da Silva Freitas et al. (2012) Brazil</td>
<td>Retrospective</td>
<td>112</td>
<td></td>
<td>To evaluate first year growth of infants who have cleft palate with or without cleft lip who do not use palatal plates</td>
<td>Within the first few months of life, infants having cleft palate with or without cleft lip weighed less than normal, unaffected infants but weight gain was similar between the two groups at the end of the first year</td>
</tr>
<tr>
<td>Beaumont (2008) UK</td>
<td>Retrospective</td>
<td>187</td>
<td></td>
<td>To assess level of failure to thrive in cleft lip and/or palate infants</td>
<td>Elevated levels of failure to thrive was present in infants with cleft of the palate. Provision of feeding support by clinical nurse specialists could improve weight gain in cleft lip and palate children</td>
</tr>
<tr>
<td>Montagnoli et al. (2005) Brazil</td>
<td>Cross-sectional</td>
<td>881</td>
<td></td>
<td>To analyze growth impairment differences according to gender in the first 2 years of life in children with cleft lip and/or palate</td>
<td>Isolated cleft palate and cleft lip and palate children experienced severe weight and length impairment compared to isolated cleft lip children</td>
</tr>
</tbody>
</table>
Discussion

The high prevalence of FTT especially in the cleft palate with or without cleft lip (CP±L) group is perturbing as it has been suggested that FTT in infants could lead to long term cognitive development impairments or behavioural deficits (Corbett and Drewett, 2004). It is worth noting that the FTT prevalence of 7% in ICL infants was close to the prevalence of 5% in the reference UK population suggesting that ICL infants had similar weight-for-age and BMI-for–age growth to that of non- cleft children (Miranda et al., 2016; Cubitt et al., 2012). The severity of undernutrition in cleft infants is linked to the cleft type and the occurrence of other anomalies or syndromes with CLP and ICP infants being more undernourished compared to ICL infants (Miranda et al., 2016; Cubitt et al., 2012; Beaumont et al., 2008; Montagnoli et al., 2005). This is so considering that cleft infants with associated syndromes like Pierre Robin sequence spend a great deal of energy on the basic function of breathing thus having little for growth and development and that feeding difficulties are far fewer in ICL infants (Pandya and Boorman, 2001; Jones, 1988).

Between the time of the first outpatient appointment to the day of surgery, an average of just 16 days, Cubitt et al. (2012) observed that there was a decrease in mean Z-score of the CLP group from -2.90 to -3.21 in spite of efforts to increase weight. The average Z-score of -3.21 for the CLP population at surgery is indicative of the presence of severe underweight in most, if not all the CLP infants in that population. The mean Z-score when used as a marker for severity of nutrition problems raises awareness that, if a condition is severe, an intervention is needed not just for those who have been categorized as ‘malnourished’ but for the whole population (WHO, 1995).

Babalola et al. (2016) observed that though the prevalences of underweight (26%) and wasting (18%) in the CLP population were higher than that of the control group (18% and 14% respectively), the difference was not statistically significant (p value = 0.334 and 0.585 respectively). This could be explained by the fact that the mean age of the CLP population in that study was 17 months whereas the most marked effects of undernutrition in CLP children occurred within the first 12 months of life mainly due to impaired suction which affects attachment to breast or artificial nipple for feeding (Miranda et al., 2016; Da Silva Freitas et al., 2012; Reid, 2004; Avedian and Ruberg, 1980).

Although CP±L children have more impaired BMI and weight-for-age growth compared to their ICL counterparts, the former experience a ‘catch-up’ growth, even without palatoplasty, beginning from the 9th month to the 13th month by which time the growth of the two groups are similar and close to the growth of normal children (Miranda et al., 2016; Da Silva Freitas et al., 2012) and this presents the possibility of affected children achieving adequate nutritional status when properly stimulated and supported. Miranda et al. (2016) surmised that it could be a case of CP±L infants getting adapted to their anomaly with time.
Two studies showed a link between undernutrition in cleft lip and/or palate children and post-delivery nutritional knowledge or support. According to Tungotyo et al. (2017) undernutrition was 3.8 times more likely to occur in cleft infants if their caretakers lacked post-natal feeding and nutritional information. Weight gain can be improved with the provision of feeding support (monitoring feeding, weight gain and giving advice on feeding aids) by clinical nurse specialists in hospitals and during domiciliary visits within the first few weeks after birth or referral of a cleft infant (Beaumont, 2008).

A good number of studies have documented the growth and nutritional status of children with cleft lip and/or palate, bringing the feeding difficulties and consequent malnutrition and growth impairment experienced by these children to the fore. Increased nutritional assessment of affected infants is needed especially in Africa where studies have been fewer, to increase awareness and strengthen the willpower of stakeholders (parents and caretakers, policy makers, governments, charity organizations etc.) to deal with malnutrition in this vulnerable population.

**Conclusion and recommendation**

Evidence from the studies show that children born with cleft lip and/or palate are a high risk population for undernutrition especially within the first year of life for whom prompt post-natal interventions are needed before surgical repair of the malformations. All the studies in this review were hospital based but community-based studies as well as studies during free-sponsored cleft care by charity organizations should be conducted to assess malnutrition in cleft lip and/or palate children who otherwise would not have presented for treatment in Hospitals due mainly to poverty. In developing countries, poverty (lack of money for surgery, transportation to treatment facilities etc.) and ignorance of availability of cleft repair surgery have been documented as causes of under-reporting, late presentation and non-presentation of cleft patients for treatment (Aimiede et al., 2013; Adeyemo et al., 2009; Schwarz and Bhai Khadka 2004).

**Declaration**

The Authors declare that there is no conflict of interest.
Authors Contribution

Author 1: literature search and manuscript write-up
Author 2: editing and review of manuscript
Author 3: editing and review of manuscript

References