Malocclusion and treatment need in children and adolescents with sickle cell disease

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ABSTRACT
Objective: To assess the prevalence of malocclusion and treatment need in children and adolescents with sickle cell disease (SCD).

Materials and Methods: In this cross-sectional study, the sample size comprised 35 five-year-old children and 36 adolescents of both sexes, aged between 12 to 18 years, with SCD. Dental occlusion was assessed using two indexes: the Malocclusion Index (World Health Organization) and the Dental Aesthetic Index (DAI).

Results: The prevalence of malocclusion in the preschool children was 62.9%. The main malocclusions observed in this age group were Class II (37.1%), increased overjet (28.6%), reduced overbite (28.6%), and open bite (17.1%). In the 12- to 18-year-old subjects, the prevalence of malocclusion was 100%, and the most prevalent types of malocclusion were maxillary overjet (63.9%) and maxillary misalignment (58.3%). It is noteworthy that the majority of adolescents (80.6%) had very severe or disabling malocclusions.

Conclusion: The results revealed a high prevalence of malocclusion in children and adolescents with SCD. According to DAI score, the majority of the sample presented with very severe malocclusion and a compulsory treatment need. (Angle Orthod. 2014;84:467–472.)

KEY WORDS: Anemia; Sickle cell; Malocclusion; Maxillofacial abnormalities

INTRODUCTION
Malocclusion, considered a worldwide public health problem, is defined as a change in teeth position and skeletal growth1,2 that causes esthetic and functional problems. Bone development, heredity, environmental factors, and the general health state of the child influence the onset and/or worsening of this anomaly.3,4 Occlusal disorder is the third—most important dental public health problem, surpassed only by tooth decay and periodontal disease.5 This problem can negatively influence the quality of life6 in terms of the social interactions and psychological well-being of affected individuals.1,2,6 Epidemiological studies2,7–11 have shown a high prevalence of malocclusion in children and adolescents in Brazil.

Patients with sickle cell disease (SCD) also frequently present with malocclusion problems.12 As a result of the compensatory expansion of their bone marrow, these patients usually have an increased overjet,13–16 higher teeth angulation causing upper incisor separation,17,18 prognathism, and diastemas.18–23 Their maxillary protrusion varies from 21% to 88.5%,18,20–22,24 and their deep overbite frequency ranges from 30% to 80%.19 Based on those facts, this study aimed to determine the prevalence of malocclusion and treatment need in children and adolescents with SCD in the state of Pernambuco, Brazil.

MATERIALS AND METHODS
This study was approved by the Ethics Committee of the Center for Hematology of Pernambuco (under protocol number 047/2011). This was cross-sectional study, with clinical epidemiological characteristics, that studied specific populations. The sample comprised 71 patients: 35 five-year-old children and 36 adolescents.
aged between 12 and 18 years with SCD and with both sexes represented. The subjects received clinical and laboratory SCD diagnosis at the Center for Hematology of Pernambuco (Hemope). The Hemope is a reference unit in hemoglobinopathies located in the state of Pernambuco, Brazil. Data collection was conducted from January through June 2012. The exclusion criteria were other systemic diseases, psychiatric or neurological disorders, or other factors that precluded oral examinations and patients who had undergone previous orthodontic treatment.

As a result of the specificity of the study population and the difficulty associated with random selection of patients with SCD, we used a nonprobability (convenience) sample. As the World Health Organization (WHO) has suggested that the study sample for certain age groups and indices should be selected according to oral health surveys, in this study the ages chosen were 5 years and 12 to 18 years, according to the methodology proposed by the last epidemiological survey on oral health conducted by the Ministry of Health (SB Brazil 2010).

Dental occlusion was assessed using two indexes: the Malocclusion Index, with modifications included from Foster and Hamilton criteria, and the Dental Aesthetic Index (DAI). The measurements were carried out using a Community Periodontal Index (CPI) probe designed by the WHO.

The first index assesses malocclusion in the primary dentition considering the following occlusal problems: canine classification (Classes I, II, and III), overjet (normal, increased, edge to edge, and anterior crossbite), overbite (normal, reduced, open, and deep), and posterior crossbite. The overjet was considered normal when the primary upper central incisors did not exceed 2 mm. The changes proposed by Foster and Hamilton established a specific classification criteria for occlusal disharmony in primary dentition.

The DAI accounts for a different criteria that expresses the occlusal status of the individual in the permanent dentition as well as treatment needs (Table 1). Altogether there are 10 measures obtained that contribute to the three major dimensions to be evaluated: the dentition (number of missing incisors, canines, and premolars), spacing (incisal segment crowding, incisal segment spacing, diastema, and maxillary and mandibular misalignment), and occlusion (maxillary overjet, mandibular overjet previous, vertical anterior open bite, and anterior-posterior molar relationship). Mixed dentition is not evaluated as a result of the appearance of some transitional occlusal characteristics during development in this phase of the dental arch.

A sample of 20 patients, different from the main study sample, was conducted to determine the agreement between the gold standard and the examiner (Kappa = 0.86). The intraexaminer calibration was based on the reexamination of one patient in every 10 children from the main study (71 patients), making a total of seven children (Kappa = 0.89).

Clinical examinations were performed at the dental office of Hemope and were conducted under artificial light, with the child sitting on a chair and facing the viewer. We used personal protective equipment (gloves, hats, masks), gauze, disposable wooden spatulas, mouth mirrors, and millimetric probes (CPI). The data collection instrument was a clinical record with information related to dental occlusion developed with the same criteria used in the SB Brazil Project 2010.

The parents or guardians were informed about the proposed study and signed an informed consent document, after which they were invited to answer a formulary to evaluate sociodemographic status. The data were analyzed using absolute frequencies and percentages to the categorical variables and the statistical measures (mean and standard deviation to the variable age). The software used for data entry and to obtain statistical calculations was the SPSS Statistical Package for the Social Sciences, version 13 (SPSS Inc, Chicago, III), and the margin of error was 5.0%.

### RESULTS

The 36 patients aged between 12 and 18 years presented a mean age of 14.42 years, with a standard deviation of 1.61 years.

The majority of patients were male (53.5%) and belonged to the 12–18-year-old group (50.7%), with parents and/or guardians with at least a primary level of education (43.7%). It was also observed that 74.6% of patients had family incomes that were one to three times the minimum wage, and 49.3% of patients lived in the metropolitan area (Table 2).

Table 3 shows that most (62.9%) 5-year-old children presented canine Class I; slightly more than half of the sample had a normal overjet (54.3%) and overbite...
The presence of posterior crossbite was recorded in 8.6% of the patients, and the occurrence of malocclusion at 5 years of age was 62.9%.

The data in Table 4 indicate that the majority (58.3%) of patients had tooth loss, maxillary misalignment (58.3%), and maxillary overjet (63.9%). According to DAI scores, all adolescents had some type of malocclusion. Most adolescents had very severe or disabling malocclusions (80.6%) (Table 5).

**DISCUSSION**

Craniofacial bone abnormalities are considered factors that may contribute to the development of dental malocclusion in any individual. In SCD patients, these abnormalities may occur as a result of hyperplasia and expansion of the bone marrow to compensate for the short life of red blood cells as a result of disease progression.13,14

There are few published epidemiological studies on SCD and occlusal disorders, especially in children. The results should be interpreted with caution as a result of the difference in terms of the methodology used, the variety of existing indices, and a lack of consensus regarding the optimal measuring instrument11 and sample size.

The prevalence of malocclusion in preschool children in Brazil ranges from 33.9% to 87%.7,30,31 In the last epidemiological survey on oral health conducted by the Ministry of Health (SB Brazil 2010),11 the prevalence of malocclusion was 69% at 5 years of age. Similar numbers were found in the present study (62.9%) and were slightly higher than those (49.7%) reported by Soares et al.12 in a study developed with 704 children, aged 6 months to 8 years, with SCD in the state of Bahia, Brazil. This difference must be related to the methodology applied.

The most prevalent malocclusions found in preschool children were Class II canines (37.1%), increased overjet (28.6%), reduced overbite (28.6%), and open bite (17.1%). With the exception of Class II canines, these results are similar to those found by Oredugba and Savage21 in Nigeria, with 229 patients aged between 1 and 18 years (177 patients with SCD and 122 healthy patients). The cited study assessed the anthropometric measurements and dental occlusion without age stratification of patients. The following malocclusions were found: Angle Class II (21%) and maxillary protrusion (21%).

In the present study, the prevalence of malocclusion was 100% in the 12–18-year-old group. These results differ from the results found in SB Brazil 2010,11 in which the proportion was 38.8% at 12 years of age and 34.9% for 15–19-year-olds.11 The most prevalent types of malocclusion among adolescents were overjet (63.9%) and maxillary misalignments in the anterior segment (58.3%). Regarding overjet, the values were higher compared to those reported by Taylor et al.18

**Table 2.** Distribution of Children and Adolescents According to Age, Gender, Parent/Guardian Education, Family Income, and Origin (Recife-PE, Brazil, 2013)

<table>
<thead>
<tr>
<th>Variable</th>
<th>N</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (in yr)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>5</td>
<td>35</td>
<td>49.3</td>
</tr>
<tr>
<td>12 to 18</td>
<td>36</td>
<td>50.7</td>
</tr>
<tr>
<td>Gender</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>38</td>
<td>53.5</td>
</tr>
<tr>
<td>Female</td>
<td>33</td>
<td>46.5</td>
</tr>
<tr>
<td>Parent/guardian education</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Illiterate</td>
<td>2</td>
<td>2.8</td>
</tr>
<tr>
<td>Literate</td>
<td>3</td>
<td>4.2</td>
</tr>
<tr>
<td>Elementary</td>
<td>31</td>
<td>43.7</td>
</tr>
<tr>
<td>High school</td>
<td>26</td>
<td>36.6</td>
</tr>
<tr>
<td>College degree</td>
<td>9</td>
<td>12.7</td>
</tr>
<tr>
<td>Family income (Brazilian minimum wage)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt;1</td>
<td>15</td>
<td>21.1</td>
</tr>
<tr>
<td>1 to 3 times minimum wage</td>
<td>53</td>
<td>74.6</td>
</tr>
<tr>
<td>3 to 5 times minimum wage</td>
<td>2</td>
<td>2.8</td>
</tr>
<tr>
<td>5 to 7 times minimum wage</td>
<td>1</td>
<td>1.4</td>
</tr>
<tr>
<td>Origin</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Recife</td>
<td>15</td>
<td>21.1</td>
</tr>
<tr>
<td>Metropolitan region</td>
<td>35</td>
<td>49.3</td>
</tr>
<tr>
<td>Countryside</td>
<td>21</td>
<td>29.6</td>
</tr>
<tr>
<td>Total</td>
<td>71</td>
<td>100.0</td>
</tr>
</tbody>
</table>

**Table 3.** Distribution of Children (5-Year-Old) According to the Index Data of Malocclusion (Recife-PE, Brazil, 2013)

<table>
<thead>
<tr>
<th>Variable</th>
<th>N</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Malocclusion</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes</td>
<td>22</td>
<td>62.9</td>
</tr>
<tr>
<td>No</td>
<td>13</td>
<td>37.1</td>
</tr>
<tr>
<td>Canine classification</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Class I</td>
<td>22</td>
<td>62.9</td>
</tr>
<tr>
<td>Class II</td>
<td>13</td>
<td>37.1</td>
</tr>
<tr>
<td>Overjet</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Normal</td>
<td>19</td>
<td>54.3</td>
</tr>
<tr>
<td>Increased</td>
<td>10</td>
<td>28.6</td>
</tr>
<tr>
<td>No rating *</td>
<td>6</td>
<td>17.1</td>
</tr>
<tr>
<td>Overbite</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Normal</td>
<td>18</td>
<td>51.4</td>
</tr>
<tr>
<td>Reduced</td>
<td>10</td>
<td>28.6</td>
</tr>
<tr>
<td>Open and deep</td>
<td>6</td>
<td>17.1</td>
</tr>
<tr>
<td>No rating &amp;</td>
<td>1</td>
<td>2.9</td>
</tr>
<tr>
<td>Posterior crossbite</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Present</td>
<td>3</td>
<td>8.6</td>
</tr>
<tr>
<td>Absent</td>
<td>32</td>
<td>91.4</td>
</tr>
<tr>
<td>Total</td>
<td>35</td>
<td>100.0</td>
</tr>
</tbody>
</table>

* Presence of open bite.

& Absence of the mandibular central incisor.
activity of the bone marrow, which results in changes in the trabecular bone associated with bone expansion and an increased overjet between the arches due to the maxillary protrusion.\textsuperscript{14,15,18,19,21,23,32–34}

The WHO recommends the use of the DAI index to verify the orthodontic treatment need and severity of malocclusion. However, this index includes tooth loss, and in the present study, once the tooth loss among adolescents was high (58.3%) this inclusion may have affected our results regarding the prevalence of severity of malocclusion. In addition, these results contrast with those obtained by Borges et al.,\textsuperscript{35} who reported that the prevalence of tooth loss in the health population was smaller (1.7%).

It is noteworthy that the majority of adolescents (80.6%) had very severe or disabling malocclusion. This finding corroborates the work of Onyeaso and da Costa\textsuperscript{23} in Nigeria with patients aged between 10 and 35 years, in which 50% of the subjects had severe malocclusion. The results of this study differ from those found in SB 2010 Brazil,\textsuperscript{11} which reports the prevalence of severe and very severe malocclusion in 15-year-olds and 19-year-olds to be 6.6% and 10.3%, respectively. At 12 years of age, the percentages of severe and very severe malocclusions were 11.9% and 7.1%, respectively. The findings of the present study indicate a high prevalence of malocclusion in children and adolescents with SCD because most adolescents had a mandatory orthodontic treatment need. These results reinforce the need to implement public health policies for inclusion of orthodontic treatment for this group of patients through early intervention programs and the adoption of measures to promote oral health to mitigate the occlusal problems.

It is recommended that oral health education aimed at people with SCD should be part of comprehensive health programs for children and adolescents. A preventive approach to malocclusion in children should be practiced by promoting particular measures, such

\begin{table}
\centering
\caption{Distribution of Adolescents (12–18 Years of Age) According to the Criteria of the Dental Aesthetic Index (DAI) (Recife-PE, Brazil, 2013)}
\begin{tabular}{lll}
\hline
Variable & N & \%
\hline
Total & 36 & 100.0
Malocclusion & 36 & 100.0
Number missing incisors, canines, and premolars
Any tooth & 15 & 41.7
One tooth & 9 & 25.0
Two to six teeth & 12 & 33.3
Incisal segment crowding
Present & 8 & 22.2
Absent & 28 & 77.8
Incisal segment spacing
Present & 6 & 16.7
Absent & 30 & 83.3
Diastema
Present & 5 & 13.9
Absent & 31 & 83.3
Maxillary misalignment
Present & 21 & 58.3
Absent & 15 & 41.7
Mandibular misalignment
Present & 14 & 38.9
Absent & 22 & 61.1
Maxillary overjet
Present & 23 & 63.9
Absent & 13 & 36.1
Mandibular overjet
Present & 36 & 100.0
Absent & 0 & --
Anterior open bite
Present & 1 & 2.8
Absent & 35 & 97.2
Antero-posterior molar condition
Normal & 12 & 33.3
Half cusp & 9 & 25
Entire cusp & 15 & 31.7
\hline
\end{tabular}
\end{table}

\begin{table}
\centering
\caption{Dental Aesthetic Index (DAI) Results for Adolescents (12–18 Years of Age) (Recife-PE, Brazil, 2013)}
\begin{tabular}{lll}
\hline
DAI Classification & N & \%
\hline
No abnormality or mild malocclusion (DAI ≤ 25) & 2 & 5.6
Malocclusion (26 to 30) & 2 & 5.6
Severe malocclusion (31 to 35) & 3 & 8.3
Malocclusion very severe or disabling (> 36) & 29 & 80.6
Total & 36 & 100.0
\hline
\end{tabular}
\end{table}
as encouraging breastfeeding to allow for proper facial growth, avoiding the development of oral habits, and providing guidance to those responsible for dental monitoring, to regulate orofacial growth and alleviate malocclusion problems.19

The main limitation of this study relates to the fact that it involves a specific population and a small number of cases, as detailed in the Materials and Methods section. Convenience sampling can generate potential selection bias because it is possible that the sample is not representative of the population, which prevents measurement of sampling error and a conclusive statement on the obtained results.36 However, given the difficulty in locating subjects for our study population, we adopted this sampling model.

CONCLUSIONS

- The obtained results revealed a high prevalence of malocclusion in children and adolescents with SCD.
- According to the DAI score, the majority of the sample presented with very severe malocclusion and a compulsory treatment need.

REFERENCES


