Evaluation of Craniomandibular Dysfunction in Children 6 to 10 Years of Age With Unilateral Cleft Lip or Cleft Lip and Palate: A Clinical Diagnostic Adjunct

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It is becoming increasingly apparent that a clinical evaluation for craniomandibular dysfunction in children is important for predicting future problems in adults. Because of the dysmorphology inherent in children with clefts, there is potential for craniomandibular dysfunction in many cases. The prevalence of craniomandibular dysfunction in white children with unilateral cleft lip or cleft lip and palate from 6 to 10 years of age was investigated. Thirty children (22 males and eight females) were examined. Craniomandibular dysfunction was detected clinically by the following criteria: mandibular movements; deflection of the mandible on opening; temporomandibular joint (TMJ) sounds; and muscle and temporomandibular joint tenderness to palpation. Information related to subjective symptoms (headaches, difficulties in opening wide, pain in the temple region, pain in opening wide, pain in chewing, and reported clicking) was collected by interview. The results showed that the prevalence of objective and subjective symptoms was 76.6 and 53.3 percent, respectively. The most frequent symptom was muscle tenderness (60 percent), followed by temporomandibular joint tenderness (26.6 percent), temporomandibular joint sounds (20 percent), and headaches (16.6 percent). Statistically significant differences by cleft type were not found in the prevalence of any objective or subjective symptom. Significant correlation was found only between temporomandibular joint tenderness and muscle tenderness. Because the overall prevalence of symptoms is shown to be high in the sample studied, routine dental examinations of patients with clefts should include an evaluation of the masticatory system.

KEY WORDS: cleft lip and palate, temporomandibular joint, craniomandibular dysfunction

The prevalence of craniomandibular dysfunction in children and adolescents without clefts had been reported to be high. Objective and subjective symptoms have ranged from 27 to 74 percent and 19 to 85 percent, respectively (Nilner and Lassing, 1981; Egermark-Eriksson, 1982; Brandt, 1985; Dibbets et al., 1985; Ogura et al., 1985). A review of the literature pertaining to the prevalence of objective and subjective symptoms of craniomandibular dysfunction of children without clefts was made by Vanderas (1987a). However, there is no study on the prevalence of craniomandibular dysfunction in children with clefts, although some factors such as the impairment of the masticatory system, an increased frequency of malocclusion secondary to clefting, and variations of psychosocial factors related to clefts may contribute to the development dysfunction.

The purpose of the current study was to determine the prevalence of signs and symptoms of craniomandibular dysfunction in children, who range in age from 6 to 10 years, with unilateral cleft lip or cleft lip and palate and to describe a clinical routine for the diagnosis of these signs and symptoms.

Method

Subjects

The sample consisted of 30 children with either unilateral cleft lip or unilateral cleft lip and palate. There were 22 males and eight females, 6 to 10 years of age, selected from the Cleft Palate Center of the University of Pittsburgh. Subjects with associated malformations and syndromes were excluded from the sample (Rollnick and Pruzansky, 1981; Shprintzen et al., 1985; Vanderas, 1987b). The group with cleft lip only did not have alveolar clefts. Children who were present in the clinic for a dental check-up, for routine dental treatment, or for team evaluation and who met the requirements of the study were examined. None of the examined subjects had had an intraoral injection prior to the examination. The instructions given to the children in prep-
eration for the examination were at the level of comprehension and understanding appropriate to the age group.

Clinical Definition

Craniomandibular dysfunction was defined by the presence of a set of symptoms. These symptoms were divided into subjective symptoms (those reported by the patient) and objective symptoms (those detected clinically). Children with one or more of these symptoms met the criteria of craniomandibular dysfunction. This operational definition was based on the assumption that any of the symptoms might be the initiation of a more serious situation later in life.

Clinical Examination: Objective Symptoms

The objective symptoms of craniomandibular dysfunction used in this study are described below. The following items were included:

Mandibular Movements

1. Maximal opening was determined by measuring the distance from the tip of the interdental papilla of the upper central incisors to the tip of the interdental papilla of the lower central incisors (Landtwing, 1978) with a Boley gauge. The subjects were asked to open their jaws as widely as possible and then “a little bit more” (Nilner and Lassing, 1981; Brandt, 1985). An opening of 44 mm or less was considered restricted (Landtwing, 1978).

2. Maximal lateral movement was measured with a Boley gauge to the right and left with the aid of pencil markings on the labial surfaces of the upper and lower incisors. Any lateral movement less than 5 mm was considered restricted (Agerberg, 1974).

3. Maximal protrusion was determined by measuring the distance between labial surfaces of upper and lower central incisors, plus the overjet. Reduced protrusion was recorded if the distance was less than 5 mm (Agerberg, 1974).

All measurements were performed twice, and the highest value was recorded. All values were rounded to the nearest millimeter or half millimeter. In other words, decimal values less than or equal to 0.25 mm or less than or equal to 0.75 mm were rounded to 0 and 0.50 mm, respectively; values less than or equal to 0.25 mm or less than or equal to 0.75 mm were rounded to 0.50 mm or 1.00 mm, respectively.

Temporomandibular Joint Function

1. Deflection of the mandible on maximal opening was determined to the left or right by measuring the distance of the midline between the lower central incisors in relation to the upper midline. A pencil marker, a ruler, and a Boley gauge were used. In cases in which midline deviation was present in centric occlusion because of tooth movement, the appropriate position of the midline was marked with a pencil marker. Any deviation of 2 mm or more was recorded as a sign of craniomandibular dysfunction (Egermark-Eriksson, 1982).

2. Temporomandibular joint sounds, like clicking and crepitation, were determined by using a stethoscope. The subjects were asked to open the mouth wide and then to slowly close it.

Temporomandibular Joint and Muscle Palpation

1. Temporomandibular joint tenderness was determined by palpating from the side (laterally) and from behind (via the auditory meatus). A positive finding was recorded if the subject felt a difference between the right and left sides or described the palpation as painful or if the pain caused guarding or a palpebral reflex (Helkimo, 1974; Brandt, 1985).

2. Muscle tenderness was determined by palpation and recorded in the same way as was temporomandibular joint tenderness. The following muscle sites were palpated: the anterior and posterior portions of the temporal muscle; the superficial portion of the masseter muscle; and the lateral and medial pterygoid muscles. The palpation was carried out bilaterally, except for the lateral and medial pterygoid muscles, which were palpated individually.

Unilateral palpation of the lateral and medial pterygoid muscles was accomplished in the following manner (Williamson, 1977): The lateral pterygoid muscles were palpated by placing the examiner’s index finger posterior to the maxillary tuberosity in a manner similar to that used when a posterosuperior alveolar injection is given. The patient was asked to move the jaw to the side being palpated to allow more space for the fingertip. Firm pressure was then applied toward the infratemporal space. Although the muscle cannot be palpated directly by this means, indirect pressure can be exerted on the myofacial structures attached to the lateral pterygoid plate (Solberg et al, 1979). The medial pterygoid muscles were palpated by placing the examiner’s fingertip in the floor of the mouth near the third molar area. The gonial angle was supported extraorally by the other hand, and the examiner’s finger was able to apply pressure sublingually on the medial pterygoid insertion to the inner surface of the mandible.

The Interview: Subjective Symptoms

All subjects were interviewed by the examiner after the clinical examination. The questions were designed to gain information about headaches occurring once a week or more, pain in the temple region or when the mouth was opened wide or during chewing, difficulties in opening wide, and the occurrence of temporomandibular joint sounds and locking and luxation of the mandible on movements. Only headaches of unknown etiology were recorded. Locking was defined as a temporary fixation of short duration in one or both temporomandibular joints. Luxation was defined as anterior displacement of the condylar head out of the fossa. The questions related to locking and luxation were addressed to the subjects by asking them whether “their jaw gets stuck or out of place.” Questions not understood were explained. Care was taken not to influence
the subject's answers. Parents were asked only when the child could not answer. In cases of uncertainty, the answers were not recorded.

Differential Diagnosis

Because craniomandibular dysfunction can be a separate entity as well as a manifestation of organic disease, a differential diagnosis was necessary (Schwartz, 1955; Bell, 1969; Morawa et al, 1985; Delbalso et al, 1986). Therefore, children with a history of juvenile rheumatoid arthritis, psoriatic arthritis, muscle diseases, and tumors of the neck and face were not included in this study. Also, children with a current history of toothache (pulpitis, pericoronitis) or upper respiratory infections were excluded. Children who received any type of orthodontic treatment before or during the research examination period were excluded from the sample, since it has yet to be determined definitively whether orthodontic treatment increases (Egermark-Eriksson, 1982) or decreases (Moyers, 1985) the signs and symptoms of craniomandibular dysfunction. None of the 10-year-old subjects (four of them) had had maxillary expansion, nor had any of the younger subjects had orthodontic intervention. Children with a history of head birth injuries, dentofacial trauma, or cranial fractures were also excluded from the study. The information concerning the differential diagnosis was collected by means of a questionnaire distributed to the parents before the examination. At the end of the examination, the answers were checked by the investigator, and additional questions were asked if it was necessary. It should be mentioned, however, that no attempt was made to differentiate between dysfunction produced by developmental abnormalities such as agenesis, aplasia, or hyperplasia of the condyle and craniomandibular dysfunction as a separate entity because a radiographic examination was not included in this study.

Examiners

The examiner attended a 1-day training course, which included clinical examinations of patients without clefts, to standardize the procedure of the study. The examinations were performed in a reclining dental chair that contained a standard dental light source. Each child was examined clinically and subsequently interviewed. The data were recorded on forms specifically designed to ensure consistency of data collection (Vanderas, 1987c).

Data Reduction

The data were computerized, and the SPSS/PC statistical package was used for their analysis. The prevalence of craniomandibular dysfunction was calculated in percentage. Correlations and differences were tested by the chi-square test. A 95-percent level of confidence was used.

RESULTS

Prevalence of Objective Symptoms of Craniomandibular Dysfunction

It is important to note that, while the overall sample size may appear to be small (N=30), the various criteria for inclusion were highly selective in order to minimize sample heterogeneity. The overall prevalence of objective symptoms was 76.6 percent (Table 1). The corresponding values for the subjects with unilateral cleft lip only (N=14) and unilateral cleft lip and palate (N=16) were 57.1 and 93.7 percent, respectively (see Table 1). Because the number of female subjects was small, the sex factor was withdrawn from the statistical analysis. None of the differences, however, was significant. The prevalence of each objective symptom was as follows:

1. The prevalence of temporomandibular joint clicking sounds in the entire group (N=30) was 20 percent (see Table 1). The occurrence of temporomandibular joint clicking sounds in the subjects with cleft lip only was 21.4 percent and in those with cleft lip and palate was 18.7 percent (see Table 1). No statistically significant difference ($\chi^2=0.703$) was found between the group with cleft lip only and the group with cleft lip and palate. Also, there was no statistically significant difference ($\chi^2=0.703$) in the prevalence of clicking sounds between the subjects with right-sided (N=16) and left-sided (N=14) cleft lip and cleft lip and palate. The chi-square test showed no correlation between the prevalence of temporomandibular joint clicking sounds on the right ($\chi^2=0.000$) or left ($\chi^2=0.000$) sides and the presence of clefts on the same side. Crepitation was not found in any of the subjects.

2. The prevalence of temporomandibular joint tenderness in the entire group was 26.6 percent (see Table 1). Subjects with cleft lip only had higher frequency of temporomandibular joint tenderness than did those with cleft lip and palate (see Table 1). The difference was

| TABLE 1 Prevalence (Overall and by Cleft Type) of Each Objective Symptom* |
|---------------------------------|---------|---------|---------|---------|---------|
|                                | TMJ Sounds | TMJ Tenderness | Muscle Tenderness | Limited Maximal Opening | Total One or More Symptoms |
| Prevalence                      | %        | %        | %        | %        | %        |
| Overall                        | (6/30) 20.0 | (8/30) 26.6 | (18/30) 60.0 | (4/30) 13.3 | (23/30) 76.6 |
| Cleft lip                      | (3/14) 21.4 | (4/14) 28.5 | (8/14) 57.1 | (2/14) 8.6 | (6/14) 42.9 |
| Cleft lip and palate           | (3/16) 18.7 | (4/16) 25.0 | (10/16) 62.5 | (4/16) 25.0 | (15/16) 93.7 |

* The ratios in parentheses are the number of subjects with the symptom divided by the total number of subjects in each group.
not statistically significant ($\chi^2 = 0.000$). The chi-square test showed no statistically significant difference ($\chi^2 = 0.584$) in the prevalence of temporomandibular joint tenderness between the subjects with right-sided and left-sided cleft lip and cleft lip and palate. Also, no correlation was found between the prevalence of temporomandibular joint tenderness on the right ($\chi^2 = 0.026$) or left ($\chi^2 = 0.000$) sides and the presence of clefts on the same side.

3. The prevalence of muscle tenderness in the entire group was 60 percent (see Table 1). The frequency of muscle tenderness in the group with cleft lip only was 57.1 percent and 62.5 percent in the group with cleft lip and palate (see Table 1). The difference was not statistically significant ($\chi^2 = 0.000$). There was also no statistically significant difference ($\chi^2 = 0.005$) in the prevalence of muscle tenderness between the subjects with right and left cleft lip and cleft lip and palate. In addition, no correlation was found between the prevalence of muscle tenderness on the right ($\chi^2 = 0.005$) or left ($\chi^2 = 0.005$) sides and the presence of clefts on the same side.

4. Six subjects had values of mandibular movements smaller than those defined in this study as minimal. Four of these patients had smaller values in maximal mouth opening, one in right lateral movement, and one in mandibular protrusion. Of the six patients, three were found to have no objective symptoms, two had temporomandibular joint and muscle tenderness, and one had muscle tenderness. All subjects with limited maximal opening had cleft lip and palate. The difference in the frequency of limited maximal opening by cleft type ($\chi^2 = 2.16$) was not statistically significant.

5. The prevalence of deflection found on opening was 33.3 percent. There was no statistically significant difference ($\chi^2 = 0.000$) between the subjects with cleft lip only and those with cleft lip and palate. In all subjects but one, the deflection occurred to the left side. There was no correlation between the frequency of deflection on the right ($\chi^2 = 0.000$) or left ($\chi^2 = 0.312$) sides and the presence of clefts on the same side.

Correlations Between Objective Symptoms

The correlation between muscle tenderness and temporomandibular joint tenderness was significant ($p = 0.022$). No other significant correlations were found between objective symptoms.

**Prevalence of Subjective Symptoms of Craniomandibular Dysfunction**

The overall prevalence of subjective symptoms was 53.3 percent (Table 2). The prevalence by cleft type was 50 percent for the subjects with cleft lip only and 56.2 percent for those with cleft lip and palate (see Table 2). Clicking sounds were reported by 13.3 percent of the subjects. Three subjects reported clicking sounds that were not detected clinically. Locking was not reported by any of the subjects, although luxation was reported by one. The prevalence of headaches was 16.6 percent in the entire group (see Table 2). The corresponding values for the subjects with cleft lip only and cleft lip and palate were 28.5 and 6.2 percent, respectively. The chi-square test showed no significant differences for cleft type with respect to any of the subjective symptoms.

**Correlations Between Objective and Subjective Symptoms**

The chi-square test showed no correlations between any of the objective and subjective symptoms.

**Discussion**

The composition of the sample with regard to sex reflects the frequency of clefts in males and females. Cleft lip with or without cleft palate is twice as prevalent in males as in females (Green, 1963; Fraser, 1970). A differential diagnosis was made in this study with respect to organic diseases, toothache, upper respiratory infection, dentofacial injuries, and orthodontic treatment. In other studies conducted on children of corresponding ages without clefts, subjects with these characteristics were included in the samples. Therefore, the ability to compare those results with the results of the present study was not possible. In addition, only white children were included in the sample because there is evidence (Dibbens et al, 1985; Ogura et al, 1985) that skeletofacial pattern might affect the function of the masticatory system.

The methods used in this study (clinical examination, questionnaire, and interview) showed high reproducibility in another study conducted by the same investigator in chil-

**TABLE 2** Overall and by Cleft Type Prevalence of Each Subjective Symptom

<table>
<thead>
<tr>
<th>Subjective Symptom</th>
<th>Headaches</th>
<th>Difficulties Opening Wide</th>
<th>Pain in Temple Region</th>
<th>Pain in Opening Wide</th>
<th>Pain in Chewing</th>
<th>Reported Clicking</th>
<th>Total One or More Symptoms</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Prevalence</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Overall</td>
<td>(5/30)</td>
<td>16.6%</td>
<td>(8/30)</td>
<td>26.6%</td>
<td>(3/30)</td>
<td>10.0%</td>
<td>(11/30)</td>
</tr>
<tr>
<td>CL*</td>
<td>(4/14)</td>
<td>28.5%</td>
<td>(3/14)</td>
<td>21.4%</td>
<td>(2/14)</td>
<td>14.3%</td>
<td>(6/14)</td>
</tr>
<tr>
<td>CLP†</td>
<td>(1/16)</td>
<td>6.2%</td>
<td>(5/16)</td>
<td>31.2%</td>
<td>(1/16)</td>
<td>6.2%</td>
<td>(5/16)</td>
</tr>
</tbody>
</table>

* CL = cleft lip
† CLP = cleft lip and palate

Note: The ratios in parentheses are the number of subjects with the symptom divided by the total number of subjects in each group.
of muscle and temporomandibular joint tenderness might not be long enough to create temporomandibular joint clicking sounds.

In this study, only recurrent headaches of unknown etiology were recorded. Most of the headaches of this nature were attributed to the contraction of the muscles of the head and neck caused by parafunctional activities (Okeson, 1985). No significant correlation was found between muscle tenderness and headaches in this group. A possible explanation for this finding was that the muscle tenderness was slight enough to cause imbalance of the muscles of the head. A number of subjects reported temporomandibular joint clicking sounds that were not detected clinically. A possible explanation may be either the intermittent nature of the sounds (Nilner and Lassing, 1981; Brandt, 1985) or the occurrence of sounds, when the mandible was moved laterally, that consequently could not be detected during mouth opening (Vanderas, 1987c). The other subjective symptoms were occasional and mild. Statistically significant differences in the prevalence of any subjective symptom between the subjects with cleft lip and cleft lip and palate were not found. This finding shows that the severity of cleft did not affect the prevalence of subjective symptoms of craniofacial dysfunction. Also, the lack of correlation between objective and subjective symptoms can be explained by the fact that the severity of objective symptoms was mild enough to cause subjective symptoms.

This study showed high prevalence of objective and subjective symptoms of craniofacial dysfunction in children with unilateral cleft lip or cleft lip and palate. In the current study of children with clefts, objective symptoms were found in 76.6 percent of the sample as compared with studies reported previously in children without clefts in which objective symptoms were reported to range from 27 to 74 percent. Additionally, subjective symptoms in the cleft sample studied occurred in 53.3 percent, as compared with studies reported previously in children without clefts in which subjective symptoms were reported to range from 19 to 85 percent. The fact that the prevalence of objective symptoms in the cleft sample studied is higher than the prevalence reported in any of the studies of children without clefts should serve to alert the clinician to the potential for craniofacial dysfunction in children with clefts, and, further, should serve to encourage the clinician to include a diagnostic evaluation for this disorder as an adjunct to developing comprehensive treatment plans for children with clefts. In general, the purpose of an epidemiologic study is to identify not only individuals in need of treatment but also individuals with subclinical signs (Vanderas, 1987c). Because mild symptoms of craniofacial dysfunction are present at very young ages for the cleft sample in the current study, dental examination should include an evaluation of the masticatory system to identify and follow up these patients.

REFERENCES

Commentary

Because of the recent surge of interest in treating temporomandibular joint disorders (TMDs) in dentistry, it was inevitable that investigations of this type applied to children with cleft palate be initiated. Thanks to Drs. Vanderas and Ranalli, we have our first course of what will soon be a banquet of reports on the subject. However, before we partake of this repast, a few words of caution are in order.

The preceding article represents a well conceived and executed study, in keeping with sound epidemiologic standards set by Helkimo (1974) and others, but there are some problems. Inherent in any of these studies is the assumption that only one objective and/or subjective sign or symptom is indicative of a disorder. This is not necessarily the case. Moreover, the authors also assume that “any of the symptoms might be the initiation of a more serious situation later in life.” In other words, there may be an evolutionary process involved in TMD. This assumption has yet to be proven by any sound scientific research. Anyone involved with the treatment of patients with TMD knows that the vast majority of patients with acute TMD symptomatology often improve with or without professional intervention. Those of us involved in adult dental care have often been surprised at how many of our patients with marked crossbites, joint sounds, and major malocclusions function very well over the years, even with these abnormalities. If anything, the preponderance of growth and development research today suggests that the child, and to some extent the adult, is remarkably adaptive in nature, particularly in the temporomandibular joint region.

The objective findings in this study center on muscle and joint tenderness, joint sounds, and mandibular movements. Muscle and joint tenderness usually is determined by digital palpation, which varies depending on the examiner. Care must be taken not to apply too much or too little pressure during muscle palpation. The muscles most often reported on this examination may be more inherent in the myofascial system. To palpate the pterygoids, one has to perform an intraoral examination. The high frequency of tenderness excursions can be easily attributed to the frequency of crossbites present in children with clefts. Our own center’s data suggest that 88 percent of patients with cleft lip and palate have crossbite present. In the general population, crossbites are the most common cause of mandibular functional shifts. Regarding joint sounds, these are rather commonplace, and their significance in the pediatric population is still not understood.

The subjective data in this study derived from the interview and questionnaire format are certainly interesting. Of course, these data must be interpreted within the framework of all such studies. Voice intonation and word phrasing vary and could change the interpretation of a given question. It is...
difficult to determine from the Vanderas and Ranalli report what type of training was given to the examiners, their level of training, or the level of supervision. Perhaps more important, questions were "explained" to the respondents. This may have introduced a bias.

The criticisms offered in this commentary are not meant to question the results. Rather, the intent is to complement the authors' work and perhaps to add another perspective to the interpretation of the data. I have been involved in a similar study on a broader sampling of craniofacial patients. I support Drs. Vanderas and Ranalli in their recommendations to include a temporomandibular joint functional examination within the dental evaluation administered to these patients. But, as a clinician who treats children and adults with and without clefts, I am particularly impressed with the fact that, of the hundreds of patients with cleft lip and palate and craniofacial anomalies that I have seen over the years, I cannot recall one of them complaining about their temporomandibular joint to me. However, in the so-called normal population of patients, the frequency of temporomandibular joint symptomatology is far greater. This situation is ironic, as the authors point out, because the inherent dysmorphology in cleft and craniofacial patients should be potentiating factors for TMD. If anything, this may bring to attention the psychosocial aspects inherent in the study of TMD, but that's a tale to be told at another time.

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