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Is an isolated cleft lip an isolated anomaly?☆

J.D. Deelder [a], C.C. Breugem [b,*], I.A.C. de Vries [b], M. de Bruin [a], A.B. Mink van der Molen [b], C.M.A.M. van der Horst [a]

[a] *Department of Plastic, Reconstructive and Hand Surgery, Academic Medical Centre, University of Amsterdam, Amsterdam, The Netherlands*
[b] *Department of Paediatric Plastic Surgery, Wilhelmina Children's Hospital, University Medical Centre, University of Utrecht, Utrecht, The Netherlands*

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Summary *Introduction:* It is well known that patients with cleft lip/palate or cleft palate can have associated anomalies. However, there is a relative paucity of information about the possible anomalies associated with an isolated cleft lip. A recent study (Vallino et al., 2008) showed that children with cleft lip and/or alveolus often develop cleft palate-related issues. This inspired us to investigate our population.
Methods: A questionnaire was sent to the parents of 214 children with cleft lip and/or alveolus; 161 questionnaires were returned (response rate (RR): 75%) and included in our study. The study consisted of 91 boys and 70 girls (0.3–13.1 years: mean 6.8 ± 3.5 years).
Results: Speech and/or language problems were reported in 34% and ventilation tube insertion in 21% of children with ≥6 years' follow-up. Of the children in that group, 33% reported to have undergone an episode of acute otitis media and 11% reported five episodes or more. Additional congenital anomalies were found in 4% of children with a cleft lip and in 16% of children with a cleft lip/alveolus.
Conclusion: Our results demonstrate that an isolated cleft lip can often be described as an isolated anomaly, although children with cleft lip and/or alveolus develop cleft palate-related issues more often than anticipated. Therefore, we suggest an intensive monitoring and treatment of children with these types of clefts.

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* Corresponding author. Cleft Palate Surgeon, Department of Pediatric Plastic Surgery, PO Box 85500, 3508 GA Utrecht, The Netherlands. Tel.: +31 87555555.
E-mail address: c.c.breugem@umcutrecht.nl (C.C. Breugem).

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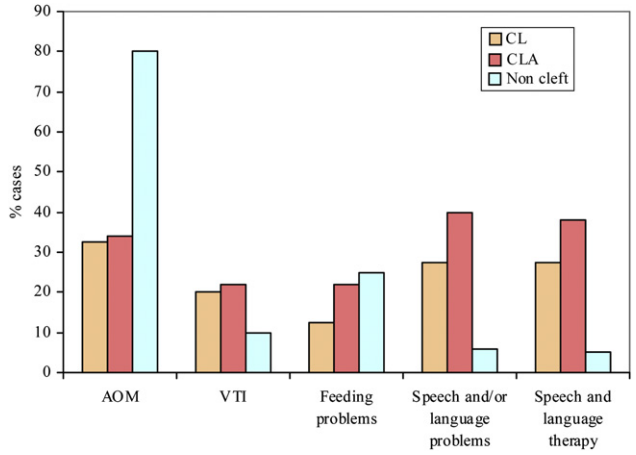

Figure 1 Accompanying anomalies (% of group total).

Orofacial clefts include a range of disorders, which affect the lips, alveolar ridge and/or palate. These defects arise in about 1—7 per 1000 live-born babies, with ethnic and geographic variation.[1]

Many studies have shown that children with clefts that include the palate are at high risk of developing hearing, feeding, speech, language, cognition and dental problems.[2–7] These children are therefore recommended to be included in multidisciplinary team care.[8] Less research, however, has been performed solely on children with clefts that donot involve the palate. Many clinicians presume that the development of these children approaches that of their non-cleft peers after the cleft lip (CL) is surgically repaired; (parental) guidance and further treatment is often adjusted to that prospect.

Vallino et al. (2008) show that children with cleft lip and/or alveolus (CLA) only do develop cleft palate-related issues. They report a higher prevalence of speech and language problems than in the general population and conclude that children with CLA need to be monitored and followed more rigorously with regard to possible speech and hearing problems.[9]

This outcome prompted us to examine our cleft population. We studied children with isolated CLA and explored the prevalence and nature of cleft palate-related anomalies and accompanying congenital defects in this underexposed subgroup.

Methods

This research was approved by the Medical Ethics Review Committee (METC) at the University Medical Centre, Utrecht, the Netherlands.

Participants

In the Academic Medical Centre (AMC), Amsterdam, the Netherlands, and the Wilhelmina Children's Hospital (WKZ), affiliated with the University Medical Centre (UMC), Utrecht, the Netherlands, children with clefts are treated by a specialised multidisciplinary cleft team. Children treated for CLA between 1997 and April 2009 were eligible for study. Children were excluded, if they had any cleft palate involvement.

A questionnaire was sent to the parents of 214 children: 78 with CL and 83 with CLA. Of the 214 questionnaires sent, 161 were returned (response rate: 75%) and included in this study. The group consisted of 91 boys and 70 girls. Children ranged in age from 0.3 to13.1 years; mean 6.8 $\pm$ 3.5 years.

Questionnaire

Questions concerned history of acute otitis media (AOM), ventilation tube insertion (VTI), feeding problems and speech and language problems. Other questions regarded the extent of the cleft, accompanying congenital defects and if genetic counselling or DNA analysis was performed.

Analysis

We used descriptive statistics to summarise the responses to the questionnaire items.

Results

A summary of the most important findings is shown in Figure 1.

Extent of CL

Children with CL (n = 78) and children with CLA (n = 83) reported to have a cleft as shown in Figure 2(a) in, respectively, 22% and 6%. The cleft shown in Figure 2(b) was reported in 35% and 13%; that in Figure 2(c) in 32% and 36% and that in Figure 2(d) in 12% and 45%.

Genetics and additional congenital defects

Seventy-seven percent of patients were seen by a clinical geneticist. In the AMC, 5.4% of children received genetic (DNA) analysis, while in the WKZ, genetic analysis was performed in 11% of CL patients and 15% of CLA patients. Three (3.8%) children with a CL had additional anomalies, while 13 (16%) of patients with a CLA had additional congenital defects. The specific defects are listed in Table 1.

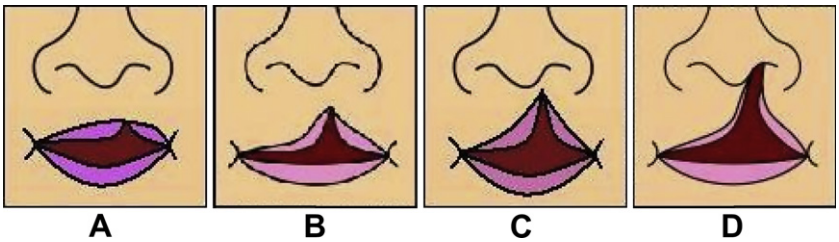

Figure 2 Extent of cleft lip or cleft lip and alveolus.