Outcomes of Cleft Palate Repair in Patients with Pierre Robin Sequence: A Matched Case-Control Study

Sir:

We read with great interest the recent article by Hardwicke et al., entitled “Outcomes of Cleft Palate Repair in Patients with Pierre Robin Sequence: A Matched Case-Control Study” published in Plastic and Reconstructive Surgery.1 This retrospective study describes the speech outcomes in children with Robin sequence associated cleft palate who had airway intervention (nasopharyngeal airway) during early infancy. We congratulate the authors for this study. However, although this article is interesting, there are some issues that require clarification.

The authors categorized their Robin sequence neonates into three groups; however, the only group included in this study were the newborns who received airway support with a nasopharyngeal airway and nasogastric tube feedings (group 3). We wonder: why did the authors not compare the speech outcomes in the group with a nasopharyngeal airway to the other two Robin sequence groups of patients (i.e., those who did not require these additional interventions)? Our question pertains to a selection bias, because the newborns that required airway and feeding interventions must certainly have had more severe phenotypic disease. In addition, how long was the airway and/or the feeding intervention used? Was the intervention required for just a few days or for a few weeks to months?

Another important factor the current study did not fully answer is the influence that a long-term nasopharyngeal airway may have on the velopharyngeal musculature. Abel et al.2 have demonstrated that a nasopharyngeal airway is typically necessary for an average of 8 months (range, 3 weeks to 27 months), with more than 10 percent of patients needing the airway intervention for more than 1 year. It has been well demonstrated that mandibular distraction often results in early removal of all other airway interventions and the nasogastric feeding tube.3 Because it has been demonstrated that Robin sequence patients with a nasopharyngeal airway often have a nasogastric tube for a prolonged time,2 this could also influence the oropharyngeal musculature and subsequent speech.

Another point of interest was that the Robin sequence group of patients was subsequently compared to a nonsyndromic group of cleft palate patients. However, we were quite surprised that only 12.5 percent of Robin sequence patients had a recorded syndrome, whether known or unknown. Recently, a cohort of 191 Robin sequence patients were reevaluated by a clinical geneticist, and an isolated Robin sequence was found in only 24 percent of cases.4 Therefore, it is highly likely that a more rigorous genetics assessment would reveal more syndromic patients in the current study, which would bias the speech results. In the study conducted by Basart and colleagues,4 5 percent had a neuromuscular disorder, whereas 47 percent had a multisystem disorder. It is possible that these factors could determine the speech outcome. This study clearly demonstrated that more multicenter studies are mandatory before definitive conclusions can be drawn.

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DISCLOSURE

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REFERENCES


Reply: Outcomes of Cleft Palate Repair in Patients with Pierre Robin Sequence: A Matched Case-Control Study

Sir:

We would like to thank Drs. Breugem and Hong for their commentary on our recently published article.1 In response to their question about a comparison of speech outcomes of infants with Pierre Robin sequence who