Alveolar cleft and maximum cleft width as predictors for difficult laryngoscopy and intubation in patients with unilateral complete cleft lip and palate

Gamal A. Abdelhameed 1*, Wael A. Ghanem 2, Simon H. Armanios 1 and Tamer Nabil Abdelrahman 1

Abstract

Background: Cleft lip and palate is one of the commonest congenital anomalies, which have an impact on feeding, speech, and dental development away from the significant psychosocial sequel. Early surgical repair aims to restore appearance and function, and the modern techniques can leave many defects undetectable. Therefore, the anesthetic challenge facing the pediatric airway with such abnormalities is still of a great impact. The aim of our study among 189 patients enrolled is to correlate alveolar gap and maximum cleft width measurements as predictors of difficult laryngoscopy and intubation in infants with unilateral complete cleft lip/palate aging from 1 to 6 months. As a secondary outcome, their weight is to be correlated too as another parameter.

Results: The alveolar gap and maximum cleft width are both of equal high predictive power \((p \leq 0.001)\) with 100% sensitivity for both and specificity of 76.10% and 82.39% respectively, with a cut off value of \(\leq 10 \text{ mm}\) and \(11 \text{ mm}\) for these dimensions respectively, and odds ratio of incidence of difficult intubation is 4.18 and 5.68 respectively, while body weight \(\leq 5.75 \text{ kg}\) has an odds ratio of 2.32.

Conclusion: Alveolar cleft and maximum cleft width can be used as predictors for anticipation of difficult laryngoscopy and intubation infant patients with unilateral complete cleft lip and palate, while body weight \(\leq 5.75 \text{ kg}\) increases the risk more than twice.

Keywords: Cleft lip/palate, Pediatric anesthesia, Difficult laryngoscopy, Intubation

Background

Children with cranio-facial abnormalities get great concern from both anesthetic and surgical point of view. Cleft lip and palate are common congenital deformities, which are relevantly present in our country with prevalence near 1:500 to 1000 live births, and will result in a child affected by cleft lip and/or palate who will require early intervention (Mairaj et al. 2017).

Unilateral clefts occur twice as often on the left side than on the right and are nine times more common than bilateral cleft (Derijcke et al. 1996).

The severity of a unilateral complete cleft lip/palate varies from the microform to a complete cleft extending all through into the nasal sill, which definitely will distort the airway (Huang and Lee 2009), and various degrees of nasal deformity and alveolar deficiency may also be present (Pool 1966). The rising trend to proceed for lip repair in the early infancy period increases the anesthetic and surgical challenge.

As for the surgical technique, Millard conceptualized his rotation advancement technique while serving in Korea and first published in the fifties (Millard 1958).
His technique is the most widely used by cleft surgeons, but few modifications have been applied since its commencement. Its principles are still the foundation of many unilateral repairs and it is our applied technique in repair with marvelous results (Fig. 1).

The anesthetic procedures performed to these patients have multiple considerations. Difficult intubation is the main concern in anesthesia practice, especially that early surgical repair starts after 1 month of age, and the pediatric airway combined with cleft lip and palate anomaly themselves may further increase difficulty in laryngoscopy and intubation (Liau et al. 2010).

Sample size calculation
Sample size was calculated using MedCalc® version 14.8.1, setting power at 0.8 and the significance level (α) at 0.05 in order to possess a clinically significant predictive power for detection of the 7.06% prevalence of difficult intubation in patients with unilateral cleft lip and palate in 1–6 months’ age group as estimated by Xue et al. (Xue et al. 2006), an alternative area under ROC curve (AUC) of at least 0.75 will be assumed, with a null value of 0.5. Calculation according to the previous data produced a minimal sample size of 170 patients with unilateral cleft lip and palate. Assuming a drop out of 10%, this produced a drop out inflated sample size of approximately 188 participants increased to 189 children enrolled.

Methods
After ethical committee approval and written signed guardian consent, this prospective cross-sectional study including 189 patients of unilateral complete cleft lip with cleft palate aging from 1–6 months, ASA I or II presenting for first stage repair of cleft lip, was conducted in tertiary university hospital. Patients admitted and who underwent first surgery between January 2017 and 2020 were included in this study.

Syndromic cases other than the specified anomaly, as those with gross neurological deficit, associated craniofacial malformations, other complex syndromes, and redo cases, were all excluded from the study except for simple non cyanotic cardiac anomalies.

Patients’ demographic data including age, gender, and weight were recorded.

A preoperative dental impression of the upper jaw was taken using specified polyvinyl siloxane impression compound for making the impression of the cleft in infants; the materials can be supported with the fingers and placed in the patient’s mouth till the material sets. Cleft measurements from the dental impression were done by a specified odontologist (Fig. 2). The dental cast dimensions were used and the specified dimensions (alveolar gap and maximum cleft width) were recorded.
If not available or possible, as in young age group children ranging from 1 to 2 months of age, the specified dimensions were measured in the operation room after inhalational anesthesia just prior to the surgery using any caliper or tool by the operating surgeon or senior pediatric anesthesiologist.

**Alveolar gap and maximum cleft width were measured as the following:**
- Alveolar gap was measured as the distance between the medial and lateral segments of the alveolar margin
- Maximum cleft width was measured as the maximum gap distance along the hard palate (Fig. 3).

### Statistical analysis

Data were analyzed using SPSS version 24.0 (SPSS Inc, Chicago, IL, USA). Parametric numerical data were presented as mean ± standard deviation, whereas non-parametric numerical data were presented as median with interquartile range. Categorical data were presented as number and percentage. Two-group comparisons for numerical data were done using the Student t test and the Mann–Whitney test for parametric and non-parametric data respectively. Categorical data were compared using the chi-square test or Fisher exact test. Association between variables was assessed using logistic regression analysis and expressed as odds ratio (OR) and its 95% confidence interval. ROC analysis was used for assessment and comparing of predictive validity and determining the best cut-off value for each predictor. Significance level was set at $P \leq 0.05$.

### Results

A summary of the demographic and clinical characteristics of the study participants is shown in Table 1.

Body weight was significantly lower in the difficult intubation group ($P < 0.001$), whereas the alveolar gap and maximum cleft width were not significantly different between the two groups.

### Table 1 Summary of the demographic and clinical characteristics of the study sample

| Characteristic                  | Value (mean ± SD or median (IQR)) |
|---------------------------------|-----------------------------------|
| Age (months)                    | 3.91 ± 1.37                       |
| Weight (kg)                     | 5.86 ± 0.95                       |
| Sex                             |                                   |
| Male                            | 102 (54.0%)                       |
| Female                          | 87 (46.0%)                        |
| Alveolar gap (mm)               | 11.0 (10.0–13.0)                  |
| Maximum cleft width (mm)        | 13.0 (11.0–15.25)                 |
maximum cleft width were statistically of higher significant differences in the difficult intubation group \( (P < 0.001) \) while no significance was found when comparing the age \( (P < 0.14) \) or sex \( (P < 0.32) \) of both groups (Table 2).

As depicted from the calculated odds ratio, every unit elevation in body weight, and decrease in the alveolar gap and maximum cleft width lowers the incidence of difficult intubation by about 70%.

This relation held the same trend after adjustment for the effect of other significant independent variables.

For prediction of difficult intubation, ROC analysis of the performance of alveolar gap and maximum cleft width revealed a robust predictive power with high validity (100% sensitivity for both and specificity of 76.10% and 82.39% respectively). No statistically significant differences could be detected when comparing the predictive power of alveolar gap with that of maximum cleft width (Table 3, Fig. 4).

**Discussion**

Patients with cranio-facial anomalies are still a challenge to secure their airway during anesthesia. When anesthesia is administered to cleft patients, there are numerous considerations that should be taken into account: the most serious are those that could lead to a difficult intubation as the airway malformations and the disrupted anatomy restricting easy laryngoscopy.

Difficult intubation probably depends upon multiple factors, as timing of repair, extent of deformity, or even the pediatric anesthesiologist own experience, etc.

We investigated two major parameters of the cleft dimensions and the patient’s body weight in a cohort of 6 months of age to find a predictive value for any of these parameters.

Along with literature, lower age group and weight are more predisposed to the incidence of difficult intubation, but they assumed weight < 10 kg is their cut of value as they investigated larger cohort of age (Huang et al. 2016), while for our cohort was weight < 5.75 kg only.

This study did not find any relationship between sex of the patient and the incidence of difficult intubation \( (p = 0.32) \), although the overall predominance of males’ incidence. However, the percentage of males to females in our study was 54 to 46% respectively.

Aycan et al., after investigating one hundred and twenty two patients aged 4 months and 10 years who underwent plastic surgery due to cleft lip/palate, stated that they found significant association between combined cleft lip-palate and higher mallampati scores \( (p \leq 0.001) \) but they did not specify age group differences or discuss intubation easiness.

**Table 2** Comparison between the study groups regarding demographic and clinical characteristics and their association with difficulty of intubation

|                      | Difficult intubation group \( [n = 30] \) | Smooth intubation group \( [n = 159] \) | \( P \) | \( OR \) (95%CI) Adjusted OR (95%CI) |
|----------------------|------------------------------------------|----------------------------------------|-------|-----------------------------------|
| Age (months)         | 3.58 ± 1.09                              | 3.97 ± 1.41                            | 0.14  | 0.81 (0.60–1.08)                  | –                      |
| Weight (kg)          | 5.07 ± 0.81                              | 6.01 ± 0.90                            | <0.001| 0.30 (0.18–0.51)                  | 0.30 (0.15–0.60)       |
| Sex                  |                                          |                                        |       |                                   |                        |
| Male                 | 19 (63.3%)                               | 83 (52.2%)                             | 0.32  |                                   |                        |
| Female               | 11 (36.7%)                               | 76 (47.8%)                             | 1.58 (0.71–3.54)\(^a\) | | |
| Alveolar gap (mm)    | 12.0 (11.0–13.0)                          | 10.0 (10.0–10.0)                       | <0.001| 0.29 (0.16–0.53)                  | 0.53 (0.31–0.90)       |
| Maximum cleft width (mm) | 14.0 (12.0–16.0)                         | 11.0 (11.0–11.0)                       | <0.001| 0.29 (0.16–0.53)                  | 0.37 (0.21–0.65)       |

\(^a\)Calculated with “female” as the reference category

**Table 3** ROC analysis for the validity of alveolar gap, maximum cleft width, and body weight in prediction of difficulty of intubation

|                      | Alveolar gap  | Maximum cleft width | Body weight  |
|----------------------|---------------|---------------------|--------------|
| AUROC (95%CI)        | 0.88 (0.82–0.92) | 0.91 (0.86–0.95)   | 0.76 (0.69–0.82) |
| \( p \) value (AUC = 0.5) | < 0.001      | < 0.001             | < 0.001      |
| Cut-off value        | \( \leq 10.0 \) mm | \( \leq 11.0 \) mm | \( \leq 5.75 \) kg |
| Measures of validity |               |                     |              |
| Sensitivity (%)      | 100%          | 100%                | 83.33%       |
| Specificity (%)      | 76.10%        | 82.39%              | 64.15%       |
| PPV (%)              | 44.1%         | 51.7%               | 30.5%        |
| NPV (%)              | 100%          | 100%                | 95.3%        |
| Positive likelihood ratio | 4.18         | 5.68                | 2.32         |
| Negative likelihood ratio | 0            | 0                   | 0.26         |
They finally concluded that Mallampati scores could be used as predictor for difficult intubation and laryngoscopy, which is not specific to such cohort (Aycan et al, 2016).

Along with our results, in another retrospective single blind trial of total of 145 infants born with cleft lip/palate with various oral measurements, Arteau-Gauthier et al. (2011) suggested that width of the cleft at the hard palate level could have a relationship with difficult intubation potentiality, but without any cleft lip ($p = 0.0323$). We further investigated this point and concluded that a cut of value of cleft width ($\leq 11.0$ mm) has an odds ratio of 5.68 than smaller clefts in patients with unilateral complete clefts (Arteau-Gauthier, 2011).

Another point of difference was that we correlated the alveolar gap distance as a predictor with a cut off value ($\leq 10$ mm) and odds ratio of 4.18 which was not correlated in that study. In addition, the average age of their investigated patients was 10 months while we included a lower age cohort.

In a prospective 800 patients’ study, Gunawardana (1996), correlated this anomaly to the incidence of difficult intubation to be 11% in children between 1 month and 6 months’ age group, but in our study, it reached 15% in the same age group.

Simpson and Wilson also believed that difficult laryngoscopy (grades III or IV) occurs in up to 10% of ASA I patients for cleft lip/palate repair with no associated syndromes, and large alveolar defects may hamper laryngoscopy, as there is a tendency for the laryngoscope to fall into the cleft, but did not specify measurements and advised packing with gauze may help prevent this, as may the use of a straight blade (Simpson and Wilson, 2002).

Somerville and Fenlon in their article postulated that a large alveolar defect, especially on the right side, increases the difficulty of laryngoscopy also for the same reason, but both did not present certain dimensions (Somerville and Fenlon, 2005).

**Conclusion**
This study highlights a significant incidence of difficult airway in patients with unilateral complete cleft lip/palate which reaches 15%, and the alveolar cleft and maximum cleft width parameters can be used as strong equal predictors for anticipation of difficult laryngoscopy and intubation in these patients (100% sensitivity for both and specificity of 76.10% and 82.39% respectively).

As regards the body weight, patients $\leq 5.75$ kg have an odds ratio of 2.32 of incidence of difficult airway. These features must be considered before anesthesia of these infants.

**Study limitations**
This study had some limitations. There might be subtle biases as the accuracy of the measurements included in the study as they were done by different personnel. Another limitation encountered was the minimal differences in measurements between awake and inhalation-anesthetized children as it might be considered as a bias.

**Acknowledgements**
Not applicable

**Authors’ contributions**
GA conceived the study and WG shared in its design. GA and WG undertook data collection, data capturing, and handling. SA and TA coordinate data analysis with the assistance and review by GA. TA and GA drafted the manuscript. All authors read and approved the final manuscript.

**Funding**
The study was funded by personal resources.
Availability of data and materials
The data of this article is available from the corresponding author.

Ethics approval and consent to participate
Ethical committee approval was obtained for this study and the study was retrospectively registered under Ethics approval and consent to participate. This study was approved by the Research Ethics Committee at Faculty of Medicine, Ain Shams University Hospital, Cairo, Egypt (FAMSU R 15/2020). This research was registered in the Pan African Clinical Trials Registry (https://pactr.samrc.ac.za/) in the 5 August 2019 with the following ID (PACT R20190849133758). Informed written consent to participate in the study was provided by all guardians according to our local and international research ethical guidelines.

Consent for publication
The locally signed consents are for participation in this research and sharing images of the discussed anomaly or pictures for the postoperative results.

Competing interests
The authors declare no competing interests.

Author details
1Department of Anaesthesia and ICU, Faculty of Medicine, Ain Shams University, Cairo, Egypt. 2Department of Pediatric Surgery, Faculty of Medicine, Ain Shams University, Cairo, Egypt.

Received: 5 September 2020 Accepted: 8 February 2021

Published online: 04 March 2021

References
Arteau-Gauthier IL (2011) Jacques E.; Godbout, Audrey. Can we predict a difficult intubation in cleft lip/palate patients? J Otolaryngol Head Neck Surg 40(5): 413–419.
Aycan İO, Yıldırım MB, Turgut H, Gümrük A, Baysal YZ (2016) Abstract PRS31. Airway management of patients with cleft lip-palate anesthesia & analgesia. 123:p673–p674. https://doi.org/10.1213/01.ane.0000492914.82991.56
Derijcke A, Berens A, Carels C (1996) The incidence of oral clefts: a review. Br J Oral Maxillofacial Surg 34(6):488–494
Gunawardana RH (1996) Difficult laryngoscopy in cleft lip and palate surgery. Br J Anaesthesia. 76(6):757–759
Huang A, Rufland L, Hajduk J, Jagnanathan N (2016) Difficult airway management of children in ambulatory anesthesia: challenges and solutions. Ambulatory Anesthesia 3:37–45. https://doi.org/10.2147/AA.S91983
Huang MHLSL, Lee S (2009) In: KR LJE (ed) Anatomy of cleft lip and palate, 2nd edn. McGraw-Hill, New York, pp 223–239
Koh LKD, Kong CE, Ip-Yam PC (2002) The modified Cormack-Lehane score for the grading of direct laryngoscopy: evaluation in the Asian population. Anaesthesia Intensive Care 30(1):48–51. https://doi.org/10.11177/0310057X0203000109
Liao JY, Sadove AM, van Aslit JA (2010) An evidence-based approach to cleft palate repair. Plastic Reconstructive Surg 126(6):216–2221
Maraj A, Anthony B, Emanuela T (2017) Epidemiology of cleft lip and palate, designing strategies for cleft lip and palate care, Mazen Ahmad Almasri, IntechOpen. https://doi.org/10.5772/67165 https://www.intechopen.com/books/designing-strategies-for-cleft-lip-and-palate-care/epidemiology-of-cleft-lip-and-palate
Millard DR (1958) A radical rotation in single harelip. Am J Surg 95(2):318–322
Pool R (1966) The configurations of the unilateral cleft lip, with reference to the rotation advancement repair. Plastic and reconstructive surgery. 37(6):558–565
Simpson S, Wilson IH (2002) Draw-over anaesthesia review. Update Anaesthesia 15:13–18
Somerville N, Fenlon S (2005) Anaesthesia for cleft lip and palate surgery. Continuing Educ Anaesthesia Crit Care Pain 5(3):76–79
Xue FS, Zhang GH, Li P, Sun HT, Li CW, Liu KP et al (2006) The clinical observation of difficult laryngoscopy and difficult intubation in infants with cleft lip and palate. Paediatric Anaesthesia. 16(3):283–289

Publisher’s Note
Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.