Perioperative adverse airway events in cleft lip and palate repair

Background
Cleft lip and palate is one of the most common congenital anomalies requiring surgical treatment. Its aetiology is complex and largely unknown, but usually attributed to genetic and environmental factors in isolation or in combination. Cleft lip and palate is also associated with an increased incidence of congenital abnormalities in other organ systems, including craniofacial disorders, which may pose challenges in airway management. Airway-related problems have been identified as the major cause of anaesthetic morbidity in cleft lip and palate surgery for many years. Fatality due to airway compromise has also been reported following cleft lip and palate surgery. There are a few studies on perioperative complications in cleft lip and palate surgery, but these are largely from developed nations.

In most developed countries, cleft lip and palate care has evolved into organised cleft teams based in regional or supraregional centres. The situation is different in most developing nations, where lack of trained medical personnel still hinders a high-quality dedicated team approach to delivery of care to orofacial cleft patients. Many of these patients from resource-poor nations are managed through surgical outreach programmes funded by donor organisations around the world. This has resulted in an increase in the number of cases managed in the hospital at which this study was conducted.

The authors sought to determine the incidence of perioperative adverse airway events in cleft lip and palate surgery managed by a local team at their centre, to identify the associated factors, and to analyse the management of the patients with a view to make appropriate recommendations for the anaesthetic management of cleft lip and palate patients in developing economies.

Method
Approval for this study was obtained from the Hospital Ethics Committee. One hundred and sixteen patients who...
had cleft lip and palate repair under general anaesthesia with endotracheal intubation at the centre during a five-year period (May 2005 to April 2010) were prospectively studied. The patients’ biographical data, diagnosis and weight and the procedure performed were documented. Records of their American Society of Anesthesiologists (ASA) physical status, preoperative medical and airway problems, associated congenital anomalies, intubation outcomes, intraoperative anaesthetic complications and duration of surgery were taken. The associated postoperative surgical complications, airway problems and treatment were also recorded. For the purpose of this study, perioperative adverse airway event was defined as an airway complication requiring intervention that occurred in the operating room (OR), post-anaesthesia care unit (PACU), or in the ward until discharge after surgery.

Anaesthesia was administered by the two anaesthetists (co-authors). Inhalational induction with halothane in oxygen or intravenous induction with ketamine or thiopentone sodium was used. Laryngoscopy and endotracheal intubation was performed using a straight or curved blade, as appropriate, under deep inhalational anaesthesia or muscle relaxant (suxamethonium). Intubation outcomes were recorded as: easy if successful with one or two attempts, difficult if requiring three to four attempts, and failed when intubation was not possible with four attempts by an experienced anaesthetist. Duration of surgery was defined as the time from skin/mucosal incision to the last stitch. Statistical Package for the Social Sciences™ 16.0 software was used for data management and analysis. All the quantitative parameters were expressed as proportions and mean ± standard deviation (SD). To test for the difference in the proportions between different groups, a chi-square test of significance or Fisher’s exact test was employed with a confidence interval of 95%.

Results

One hundred and sixteen patients were included in the study, consisting of 53 (45.7%) males and 63 (54.3%) females, who had 131 surgeries. The age at the time of surgery ranged from two months to 50 years, with a mean of 6.1 years (± 9.4 years). The patients’ distribution by age is shown in Figure 1.

The mean weight was 17.4 kg (± 17.4 kg) with a range of 3-75 kg, while the mean duration of surgery was 96.1 minutes (± 52.6 minutes). All the patients except one had an ASA physical status of 1 or 2 (Figure 2).

The distribution of the cleft types are shown in Table I, while Table II shows the associated congenital anomalies seen in five (4.3%) of the patients. Seventy-five per cent had Millard cheiloplasty, with or without rhinoplasty, and 25.2% had palatoplasty.

### Table I: Cleft type cross-tabulated against adverse peri-operative airway events

| Cleft type                        | Adverse perioperative airway event | Total |
|-----------------------------------|-----------------------------------|-------|
|                                  | No  | Yes |                  |
| Bilateral cleft lip               | 6   | 0   | 6 (5.2%)         |
| Bilateral cleft lip and palate    | 3   | 1   | 4 (3.4%)         |
| Bilateral transverse facial cleft | 1   | 0   | 1 (0.9%)         |
| Isolated cleft palate             | 12  | 1   | 13 (11.2%)       |
| Left cleft lip                    | 43  | 0   | 43 (37.1%)       |
| Left cleft lip and palate         | 14  | 3   | 17 (14.7%)       |
| Right cleft lip and palate        | 10  | 1   | 11 (9.5%)        |
| Right cleft lip                   | 21  | 0   | 21 (18.1%)       |
| Total                             | 110 | 6   | 116 (100%)       |

Fisher’s exact test  
p=0.02  (significant)

### Table II: Associated congenital anomalies

| Congenital anomalies | Frequency (%) |
|----------------------|---------------|
| None                 | 111 (95.7%)   |
| *ASD with hypertelorism and glandular hypospadias | 1 (0.9%) |
| Low-set ears with orbital hypertelorism and mid-face retrusion | 1 (0.9%) |
| Microcephaly, micrognathia with low-set ears and hypertelorism | 1 (0.9%) |
| Spina bifida occulta | 1 (0.9%)      |
| Membranous *VSD     | 1 (0.9%)      |
| Total               | 116 (100%)    |

a= Atrial septal defect  
b= Ventricular septal defect
Seven (6%) patients had mild cough and catarrh or chronic nasal discharge preoperatively. One of these (14.3%) had a perioperative adverse airway event (difficult intubation associated with desaturation), compared to five (4.5%) of the remaining 109 patients who had no preoperative airway symptoms. The relationship between the presence of mild cough and catarrh or chronic nasal discharge at the time of surgery and the risk of perioperative adverse airway event was not statistically significant ($p=0.26$). All the cases with adverse perioperative airway events had a combined cleft lip and palate, except for one who had an isolated cleft palate repair. Three (10%) of the 33 palatoplasty cases had perioperative adverse airway events. These perioperative adverse airway events are described in Table III, along with the clinical features of the six patients (4.6%).

Postoperative surgical complications (palatal fistula in three patients and wound dehiscence in two patients) were not significantly associated with perioperative adverse airway events ($p=0.28$). There was significant association between perioperative adverse airway events and the type of cleft lip and palate ($p=0.02$) (see Table I), but not between perioperative adverse airway events and the age of the patient at the time of surgery ($p=0.38$).

**Discussion**

Anaesthesia for cleft lip and palate surgery is known to carry a high risk of adverse airway events.\(^3\) In a review of perioperative airway complications following pharyngeal flap palatoplasty, Peña et al reported an incidence of 10% in the 88 patients studied,\(^2\) while Antony and Sloan recorded an incidence of 5.7% in their study of airway obstruction following 247 consecutive palatoplasties.\(^12\) About 5% of the patients in this series experienced perioperative adverse airway events. However, the current review included patients for both lip and palate surgery in isolation or as combined procedures, and three (10%) of the 33 palatoplasty cases had postoperative airway complications.

Late presentation of patients with cleft deformities is very common in developing countries.\(^11\) Only forty per cent of the patients in this study had their repair done before the age of one. Similar findings were reported by Adeyemo et al in Lagos, Nigeria: only 71.3% of the patients in their study had their repair by the age of six years.\(^3\) Younger age has been associated with increased incidence of perioperative adverse airway events in orofacial cleft and paediatric anaesthesia in general.\(^7,12,14\) Xue et al\(^7\) and Gunawardana\(^14\) showed that the frequency of difficult laryngoscopy and intubation is higher in orofacial cleft patients younger than six months of age. The relatively lower incidence of perioperative adverse airway events in the current study is attributable to the age at the time of surgery (mean 6.1 years), compared to 1.5 years noted by Antony and Sloan.\(^12\) This factor has to be considered when drawing up an anaesthetic plan for orofacial cleft patients, particularly during surgical outreach programmes.

Difficult intubation is known to be a main factor in deaths associated with anaesthesia in surgical patients.\(^7\) A case of difficult intubation with desaturation, which proved easier when the patient returned without an upper respiratory tract infection (URTI), was noted in this study. The ongoing infection and inflammation could have contributed to the difficulty in intubation and desaturation experienced during

| Table III: Perioperative adverse airway events and patient characteristics |
|-----------------|-----------------|-----------------|-----------------|
| **Patient**     | **Biodata and characteristics** | **Airway complications** | **Treatment**    |
| 1               | 4.5 months, 6.5 kg, right cleft lip and palate, cough | Difficult intubation and desaturation | Rescheduled, uneventful two weeks later |
| 2               | 11 months, 6 kg, left cleft lip and palate, associated microcephaly, micrognathia and hypertelorism | Best laryngoscopic view was Cormack and Lehane III: failed intubation | Postponed until child is 10 kg |
| 3               | 2.5 years, 13 kg, cleft soft palate with bifid uvula, had palatoplasty | Post-extubation bronchospasm | Halothane in 100% oxygen, IV aminophylline, discharged POD 3 |
| 4               | 1 year, 8.6 kg, left cleft lip and palate, had palatoplasty | Post-extubation laryngeal oedema | Reintubation in the OR, nursed in the ICU with ETT for 72 hours, hydrocortisone, nebulised adrenaline, oxygen, antibiotics and analgesic, discharged POD 7 |
| 5               | 15 months, 9 kg, left cleft lip and palate, had palatoplasty, IV diazepam 1 mg stat in PACU, then 5 mg every 8 hours for 24 hours for sedation | Postoperative chest infection 24 hours after surgery | Oxygen, hydrocortisone, antibiotics, improved after 48 hours, discharged POD 13 |
| 6               | 8 months old, 5.5 kg, bilateral cleft lip and palate, had lip repair | Postoperative chest infection 24 hours after surgery | Oxygen, nebulised salbutamol, IV furosemide, digoxin, antibiotic, hydrocortisone and supplemental oxygen, improved after 72 hours, discharged POD 7 |

\(^{a}p=\) postoperative day
the initial presentation of this patient for anaesthesia and surgery. A case of failed intubation in which anaesthesia was safely reversed was also recorded. The current study suggests an association between perioperative adverse airway events and combined cleft lip and palate compared to isolated cleft lip. Takemura et al.10 and Xue et al.11 noted a similar finding in their reports on infants with more severe cleft lip and palate: those who had bilateral cleft lip and palate had a significantly higher incidence of perioperative respiratory complications than those with simple cleft lip.

Early feeding difficulties associated with cleft lip and palate are known to result in inadequate weight gain, and nasal cavity irritation from food and saliva, coupled with impairment of the nasal filtration function, can cause infections, such as rhinitis, sinusitis and typhlitis. Thus, infants with orofacial cleft may present with recurring respiratory infections. Surgical repair promotes an improvement in these conditions and therefore all efforts should be made to avoid undue cancellation of surgery. However, the risks of anaesthesia and perioperative adverse airway event, should be individually balanced against the benefits of surgery within the limit of safety. One of the eight patients with mild URTI at the time of surgery had a perioperative adverse airway event (difficult intubation with associated desaturation postintubation). This resulted in longer hospital stay, but the association between URTI and anaesthetic complications was not statistically significant. All the patients were assessed preoperatively and those with moderate to severe URTI were treated with antibiotics and were not operated on until two to three weeks later, when the infection had subsided.

Following cleft palate repair, factors that may predispose patients to upper airway obstruction include critical reduction in size of a previous difficult airway, excessive sedation, so that the infant fails to adequately protrude the tongue, and laryngeal oedema due to a large endotracheal tube, resulting in stridor. Tongue suture can be used to manage airway obstruction caused by the tongue falling back until the patient resumes total control of the airway. Few prospective series exist, but a 5% rate of immediate upper airway obstruction on extubation has been reported, and occurs particularly in children with an associated syndrome, especially Pierre Robin syndrome.12 Only one of the patients in this series had obstruction due to laryngeal oedema after an apparently uneventful surgery with a well-selected plain endotracheal tube with pharyngeal packing. Extensive pharyngeal packing could also have contributed to airway oedema in this patient.

Orofacial cleft is associated with over 200 syndromes or sequences, and several have significant anaesthetic implications. Craniofacial abnormalities are the most common. The presence of other associated craniofacial anomalies has been associated with a significantly increased difficulty in airway management and risk of airway obstruction.12,15,16 Although the presence of a syndromic disease in the patients in the current series could not be established because of the unavailability of a geneticist’s services, the only child with failed intubation had associated microcephaly, micrognathia and orbital hypertelorism suggestive of a syndromic disorder. A thorough physical examination and the ability to detect anomalies that could impact on the management outcome of cleft lip and palate is routine in the authors’ practice and should be emphasised particularly in resource-poor nations.

Conclusion

Adverse respiratory airway events are not uncommon in orofacial cleft surgery. These complications usually occur immediately following extubation or in the early postoperative period. More severe forms may necessitate admission to an intensive care unit and specialised care. This study suggests that combined cleft lip and palate, palatoplasty, and younger age are associated with an increased incidence of perioperative adverse airway events.

Conflict of interest

The authors declare no financial support or conflict of interest.

References

1. Tremlett M. Anaesthesia for cleft lip and palate surgery. Current Anaesthesia & Critical Care. 2004;15:309–316.
2. Jones RJ. A short history of anaesthesia for hare lip and palate repair. Br J Anaesth. 1971;43(8):796–802.
3. Peña M, Choi S, Boyadjian M, Zalait G. Perioperative airway complications following pharyngeal flap palatoplasty. Ann Otol Rhinol Laryngol. 2000;109:808–811.
4. Hodges SC, Hodges AM. A protocol for safe anaesthesia for cleft lip and palate surgery in developing countries. Anaesthesia 2000;55:636–641.
5. Takemura H, Yasumoto K, Toi T, Hooyamada A. Correlation of cleft type with incidence of perioperative respiratory complications in infants with cleft lip and palate. Paediatr Anaesth. 2002;12(7):585-588.
6. DeMey A, Vadoud-Sayed U, Demol F, Govaerts M. Early postoperative complications in primary cleft lip and palate surgery. Eur J Plast Surg. 1997;20:77-78.
7. Xue FS, Zhang GH, Li P, et al. The clinical observation of difficult laryngoscopy and difficult intubation in infants with cleft lip and palate. Paediatr Anaesth. 2006;16:283-289.
8. Bell C, Oh TH, Loefler JR. Massive macroglossia and airway obstruction after cleft palate repair. Anaesth Analg. 1988;67:71-74.
9. Fillies T, Homann C, Meyer U, et al. Perioperative complications in infant cleft repair. Head & Face Medicine [serial online] 2007;3:9. Available from http://www.head-face-med.com/content/3/1/9
10. Xue FS, Zhang GH, Li P, et al. The clinical observation of difficult laryngoscopy and difficult intubation in infants with cleft lip and palate. Paediatr Anaesth. 2006;16(3):283-289.
11. Bell C, Oh TH, Loefler JR. Massive macroglossia and airway obstruction after cleft palate repair. Anaesth Analg. 1988;67:71-74.
12. Fillies T, Homann C, Meyer U, et al. Perioperative complications in infant cleft repair. Head & Face Medicine [serial online] 2007;3:9. Available from http://www.head-face-med.com/content/3/1/9
13. Adeyemo WL, Ogulwew MO, Desalu I, et al. Cleft deformities in adults and children aged over six years in Nigeria: Reasons for late presentation and management challenges. Clinical, Cosmetic and Investigational Dentistry 2009;1:63-69.
14. Gunawardana RH. Difficult laryngoscopy in cleft lip and palate surgery. Br J Anaesth. 1996;76:757-759.
15. Nargozian C. The airway in patients with craniofacial abnormalities. Paediatr Anaesth. 2004;14(1):53-59.
16. Butler MG, Hayes BG, Hathaway MM, Begleiter ML. Specific genetic diseases at risk for sedation/anaesthesia complications. Anaesth Analg. 2000;90:837-855.