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Peri-operative complications in children with Apert syndrome: A review of 509 anaesthetics.

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For Peer Review

Abstract:

Objectives: To perform a retrospective, anaesthesia case note review in children with Apert Syndrome.

Aim: To identify peri-operative complications in this group of patients.

Background: Apert syndrome is a rare autosomal dominant disorder characterized by craniosynostosis, craniofacial anomalies, and severe symmetrical syndactyly (cutaneous and bony fusion) of the hands and feet. Children with this syndrome require general anaesthetics for a number of different operations and procedures. Our institution has records of 71 children with Apert syndrome. Analysis of their general anaesthetic records was undertaken and the incidence of peri-operative complications was investigated.

Methods: A retrospective case note review was performed on 61 children with Apert syndrome over a 14 year period. There were a total of 509 general anaesthetics administered to these children during this period of time.

Results: There were a total of 31 peri-operative respiratory complications occurring in 21 patients (6.1% of the total cases). 23 of these complications were supraglottic airway obstruction (4.5% of total cases).

Conclusions: We found there to be a low incidence of major peri-operative major complications in this group of patients. Nevertheless, a significant proportion of these children have obstructive sleep apnoea and may develop supraglottic airway obstruction on induction and emergence from anaesthesia due to the associated mid-face anatomical abnormalities.

Keywords: Apert syndrome; peri-operative complications; mid-face hypoplasia; obstructive sleep apnoea, airway management

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Introduction

Apert syndrome is a rare disorder which generally occurs sporadically, although an autosomal dominant inheritance has been described[1]. It is evident at birth, and accounts for 4.5% of all cases of craniosynostosis (premature fusion of one or more cranial sutures). Further craniofacial abnormalities include hypertelorism, midface hypoplasia and choanal stenosis. Neurological and intracranial anomalies can include ventriculomegaly, hydrocephalus[2] and developmental delay[3]. The other dominant clinical feature is symmetrical syndactyly of the fingers[4] and feet (cutaneous and bony fusion) generally tending to affect the upper limb more severely than the lower limb[5]. There have also been reports of associated congenital cardiac and renal defects. There is fusion of the cervical spine in 68% of cases of Apert syndrome, mainly at C5-C6, distinguishing it from Crouzons syndrome where the fusion is usually at the C2-C3 level[6].

These features have clinical relevance to anaesthetists as some degree of upper airway obstruction due to reduced nasopharyngeal volume and choanal patency is certain. Obstructive sleep apnoea is present in almost 50%[7] and cor pulmonale may result[8]. There have also been descriptions of complete or partial cartilage sleeve abnormalities of the trachea, which has been postulated to cause lower airway compromise[9].

Children with this syndrome require general anaesthetics for a number of different operations and procedures. It has been suggested that they have a much higher incidence of peri-operative respiratory complications, particularly bronchospasm, than other children[10]. There is very little published data looking specifically at peri-operative outcomes in children with Apert syndrome. Reviews and reports tend to group them together with other craniofacial syndromes[11-13], so specific complications may be masked. Our institution has records of 71 children with Apert syndrome **diagnosed clinically and genetically**. Analysis of their general anaesthetic records was undertaken and the incidence of peri-operative complications was investigated.

Methods

Following ethical and audit committee advice a retrospective case note review was performed on 71 children with Apert syndrome at Great Ormond Street Hospital. Eight case notes were untraceable during the period of time due to filing and tracking errors and therefore omitted from the series and a further 2 sets of case notes had no records of anaesthesia. The anaesthetic charts from the case notes of the remaining 61 children were analysed for the 14 year period from 1995 until 2009.

Details of patient demographics and the procedures undertaken were collated. The patient's pre-operative respiratory status was recorded, specifically looking for a sleep study diagnosis of obstructive sleep apnoea, a history of asthma or wheeze, tracheostomies, and previous respiratory admissions. Intra-operative airway management and complications were documented, and the post-operative period was evaluated.

Results

There were a total of 509 general anaesthetics administered to 61 children (38 males and 23 females) during this 14 year period of time. Their ages ranged from 1 day to 15 years old, and they were classified as ASA II (n=77) and ASA III (n=431), with operation duration ranging from 10 minutes to over 4 hours.

The most frequent reason for a general anaesthetic was an orthopaedic procedure (Table 1.). Within the 224 operations undertaken 115 of these were for a change of dressing. There were 46 major craniofacial operations, the majority of which were posterior vault expansions, frontal remodelling and biparietal distractions (Table 2.). The remaining **minor** craniofacial procedures included adjustments and removals of 'RED (Rigid Extraction Device) frames'.

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Thirty six children had a sleep study diagnosis of obstructive sleep apnoea pre-operatively. **Although we were unable to clarify the exact age of the children when they were diagnosed, their ages when presenting for surgery ranged from 6 weeks to 14 years. Two of these children, aged 11 and 12, were receiving home CPAP. Eight out of the nine adenotonsillectomy patients in this series were having the procedure in an attempt to ameliorate OSA.** Only 5 of the patients had a history of asthma or being ‘wheezy’ in the past. Three of these children were taking regular inhalers for the condition. Six children had a tracheostomy, at some point during their lives. **The indication for four of these children was temporary insertion because of craniofacial surgery. One child had a tracheostomy at the age of 2 weeks to assist prolonged ventilatory requirements. One child had a definitive tracheostomy for severe obstructive sleep apnoea.** Fifteen children had known choanal atresia, and one child had **congenital** subglottic stenosis. Eight of the children had been admitted to hospital with respiratory symptoms since birth, and on sixteen occasions the patients had experienced a recent upper respiratory tract infection prior to their anaesthetic (Table 3.).

Atropine was prescribed before 216 anaesthetics and sedation was given before 44 procedures. The majority of times, no premedication was given (n=293). A majority of the anaesthetics were initiated with a gas induction (251 vs. 58 intravenous). Maintenance of anaesthesia was with volatile agents in all but one of the anaesthetics where total intravenous anaesthesia (TIVA) was used. The patients received positive pressure ventilation for 225 of the anaesthetics, and in the remaining 284 cases the child breathed spontaneously throughout the procedure.

The airway was managed in a variety of ways - face mask (n=68, SV=66, PPV=2), laryngeal mask (n=206, SV=206), tracheal tube (n=199, SV=2, PPV= 197) and tracheostomy (n=36, SV=10, PPV=26). The children were easy to intubate in the majority of cases with a Cormack and Lehane grade I (n=181), grade II (n=15) and grade III (n=3). There were no children with a grade IV view. There were 2 cases where the laryngeal mask did not provide a good seal; one was managed with tracheal intubation and the other with a face mask.

Nasal prongs were in situ prior to 19 anaesthetics in 4 children. Another 6 children had a nasal prong placed at the end of their operations.

In 98 of the 199 occasions, an age appropriate tracheal tube size was used according to the formula: **tracheal tube size (internal diameter in millimetres) = (age/4) +4**. In 68 instances a larger tube size was required, and a smaller tracheal tube was placed in 28 cases. There were 5 anaesthetics where the tube size was not documented (Table 4.).

Peripheral access was documented in 457 of the anaesthetics. On 52 occasions there was no intravenous access. There were 45 arterial lines and 30 central venous catheters placed.

The majority of orthopaedic operations were upper limb procedures and brachial plexus blocks (n=80) were commonly performed. Limited **abduction** of the upper arm and 'winging' of the scapula in Apert syndrome makes axillary brachial plexus block difficult to perform and ultrasound guidance is helpful. The remaining regional blocks were caudal epidurals (n=3), and an ankle block (n=1).

There were a total of 31 peri-operative respiratory complications occurring in 21 patients (6.1% of the total cases). 23 of these complications were supra-glottic airway obstruction (4.5% of total cases), which were easily resolved with simple airway manoeuvres, adjuncts or positive end expiratory pressure (PEEP) (**Table 5.**). There was only one documented incidence of wheeze, which followed fentanyl and erythromycin administration and was clinically felt to be allergy related although no treatment was required. **The child was 3 years old, having syndactyly release surgery and was breathing spontaneously through a laryngeal mask.** The child had a recent URTI but subsequent anaesthetic charts documented this event as an antibiotic allergy. Other problems were: desaturation due to secretions down the tracheal tube, resolved with suctioning (x1); Laryngospasm requiring re-intubation (x1); Self resolving stridor in recovery (x1); Post-operative upper airway clot after micro-laryngo-bronchoscopy (x1); Coughing on induction (x1); Obstruction of nasal prong on postoperative ward leading to respiratory arrest and PICU admission (x1); Desaturation and

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drowsiness in recovery after monoblock distraction (x1), which was successfully treated with naloxone and furosemide.

Five children had a past medical history of asthma/wheeze and 3 of these were taking regular inhalers for the condition; the patient who experienced wheeze under anaesthesia was not one of this cohort. Thirty-six patients had a formal sleep study diagnosis of obstructive sleep apnoea but only 9 of these had documented peri-operative supraglottic airway obstruction. Only 3 children with a recent URTI had documented peri-operative respiratory complications; 1 (described previously) associated with wheeze and allergy; 1 with tracheal tube secretions and the other with supraglottic airway obstruction.

There was one patient with a major bleed following a bipartition distraction, who required unexpected admission to the PICU for ongoing management. Two children vomited, one on induction and the other on emergence, but neither resulted in aspiration.

The majority of patients returned to the ward/ HDU environment after their operations (n=494) with only 15 admissions to the PICU/NICU post-operatively.

Discussion

Although a retrospective case note review may well under-report complications, we are confident that major problems requiring treatment will have been captured. Overall, we found there to be a low incidence of major peri-operative respiratory complications in this group of patients and in contrast to a previous publication[10], we found the incidence of bronchospasm to be extremely low at 0.2% and there were no cancellations of surgery due to respiratory complications.

The lower airway abnormalities described in the literature did not cause excessive problems in our cohort, tracheal tube size was either age appropriate or greater in 83.4% of the group and only one

child had a documented **congenital** sub-glottic stenosis[9]. In one child, there was desaturation related to partial obstruction of the tracheal tube by secretions and although this was easily resolved, we note that this complication has been reported previously[14]. Tracheobronchial secretions may also cause lower airway obstruction and mimic bronchospasm, which may account for some of the cases previously described.

Our results showed that our group of patients were far more likely to experience supra-glottic rather than infra-glottic obstruction. It may be that this was under-reported in our data, as minor degrees of supra-glottic obstruction may not have caused the anaesthetist or patient substantial problems and may have seemed unworthy of reporting. Simple airway manoeuvres and adjuncts usually rectified any upper airway obstruction documented. **The majority of upper airway obstructions occurred on induction of anaesthesia. There were only four incidences of postoperative obstructions. A common supposition would be to assume the incidence of postoperative problems occurs more frequently with longer intubations and prolonged surgery. Our results do not support this statement. The majority of the supraglottic obstructions occurred when the child was scheduled to have 'shared airway' surgery such as Ear, Nose and Throat (ENT) and dental procedures. Perioperative airway complications were encountered infrequently in the major craniofacial operations. Just over half of the incidents of supraglottic obstruction arose in children under the age of 2 years, which is not unexpected.** We find it re-assuring to have collected data which demonstrates that if supra-glottic airway obstruction does occur, laryngoscopy and intubation is generally easy. It should be noted however, that although surgical mid-face advancement improves the cosmetic appearance, it results in a **marked** deterioration of laryngoscopic view and this may **cause unexpected difficulties**[15, 16].

Our data demonstrates that patients with Apert syndrome may require many anaesthetics of relatively short duration. A large proportion of the anaesthetics were of less than 30 minutes and

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this explains the large proportion of cases where the patient was breathing spontaneously either with a face-mask or laryngeal mask airway.

In 52 of the anaesthetic charts there was no peripheral vascular access. This was a deliberate policy intended to preserve valuable vascular access sites during hand dressing changes. An intraosseous device was immediately available for all these cases but was never required.

In our cohort of patients the peri-operative problems occurred in 21 children (34%), which is a similar incidence to Elwood et al. who reported respiratory complications in 33% of patients[10]. Very few of the complications were serious and none resulted in cancellation of the procedure so it is important that a population of patients are not incorrectly labelled as ‘high risk’. Nevertheless a significant proportion of these children have obstructive sleep apnoea and may develop supraglottic airway obstruction on induction and emergence from anaesthesia due to the mid-face anatomical abnormalities associated with this syndrome and the anaesthetist must be prepared to deal with this.

The authors have no financial or other interests to declare.

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Table 1. Type of surgery.

Type	Number of Anaesthetics
Orthopaedics	224
ENT	79
Neurosurgery	62
Radiology	51
Craniofacial	46
Dental	18
Plastics/Cleft surgery	10
General	10
Eyes	7
Urology	1
Cardiac	1

Table 2. Major craniofacial reconstructions.

Procedure	Number of cases
Bipartition distraction	7
Monoblock distraction	5
Frontal remodelling	8
Cranial vault expansion	5
Posterior vault expansion	16

Table 3. Patient demographics.

Operation duration		Ages		Weights (kg)		Pre-operative respiratory status	
Duration (mins)	No. of cases	Age	No. of patients	Weight (kg)	No. of patients	History	No. of patients
0-30	261	Neonates	4	<3	3	Obstructive sleep apnoea	36
31-60	65	4wk-6mths	69	3.1-6	51	CPAP	2
61-120	119	7-12mths	107	6.1-10	141	Asthma/wheeze	5
121-240	49	13mth-5yrs	235	10.1-15	140	Regular inhalers	3
>241	15	6-11yrs	52	15.1-20	87	Tracheostomy	6
		>12yrs	40	20.1-40	52	Choanal atresia	15
				>41	83	Subglottic stenosis	1

Table 4. Age appropriate tracheal tube sizes (based on the formula: Tracheal tube internal diameter (mm) = age/4 + 4).

Tube size	Numbers	Percentage
Age appropriate	98/199	49.2%
Larger size	68/199	34.2%
Smaller size	28/199	14.1%
Unclear	5/199	2.5%

Table 5. Details of patients who experienced supraglottic airway obstruction.

Patient	Age	Known preoperative Upper Airway obstruction?	Surgery	Duration of surgery	Timing of obstruction	Manipulation of airway to overcome obstruction	Definitive airway for procedure
1.	3 yrs	Yes - OSA	Tonsillectomy and Adenoidectomy	60 mins	Induction	Oropharyngeal airway	Tracheal tube
2.	19 mths	No	Fronto-orbital remodelling	6 hrs	Induction	Oropharyngeal airway	Tracheal tube
3a.	8 mths	Yes - OSA	Change of dressing	20 mins	Induction	Oropharyngeal airway and PEEP	Face mask and oropharyngeal airway
3b.	9 mths	Yes - OSA	Change of dressing	20 mins	Induction	Oropharyngeal airway	Face mask and oropharyngeal airway
3c.	9 mths	Yes - OSA	Tonsillectomy and Adenoidectomy	30 mins	Induction	Oropharyngeal airway	Tracheal tube
4a.	12 yrs	Yes – OSA. Nocturnal CPAP	Dental extractions	2 hrs 45 mins	Induction	Jaw support and suction	Tracheal tube
4b.	9 yrs	Yes - OSA	Tonsillectomy and Adenoidectomy, EUA ears	1 hr 15 mins	Induction	Jaw support	Tracheal tube
5.	14 yrs	No	Correction of fixed flexion deformity of hand	40 mins	Induction	PEEP	Tracheal tube
6.	4 mths	Yes – OSA/choanal atresia	Adenoidectomy, EUA postnasal space	60 mins	Induction	Jaw support	Tracheal tube
7.	5 mths	Yes - OSA/choanal atresia	Frontal remodelling	4 hrs	Induction	LMA	Tracheal tube
8.	5 yrs	Yes - OSA	Adenoidectomy	40 mins	Induction	Jaw thrust	Tracheal tube
9.	14 yrs	Yes - OSA	Syndactyly repair	1 hr	Emergence	Oropharyngeal airway until awake	Tracheal tube
10a.	9 mths	Yes – OSA/choanal atresia	Nasal prong and dilation	20 mins	Induction	Oropharyngeal airway and nasal prong	LMA
10b.	2 yrs 10 mths	Yes - OSA	CT Head	10 mins	Induction	Oropharyngeal airway	LMA
11a.	4 mths	No	Cleft palate repair	1 hr 30 mins	Induction	Oropharyngeal airway	Tracheal tube
11b.	9 mths	No	Syndactyly repair	3 hrs 30 mins	Induction	Oropharyngeal airway	Tracheal tube
12.	12	No	Grommets	10 mins	Induction	Oropharyngeal	LMA

	mths	(subsequently diagnosed OSA)				airway and PEEP	
13.	6 weeks	Yes - OSA	MLB and nasal prong	60 mins	Induction	Nasal prong	Nasal prong
14.	14 yrs	Yes - OSA	Cranial cyst enucleation	1 hr 30 mins	Emergence	Jaw thrust	Tracheal tube
15.	4 yrs	No	Division of fingertips	60 mins	Induction	Jaw thrust and PEEP	Tracheal tube
16a.	7 mths	No	Cranial remodelling	2 hrs	Emergence	Oropharyngeal airway	Tracheal tube
16b.	4 yrs	No	Change of dressing	10 mins	Induction	PEEP	Face mask
16c.	9yrs	No	Dental extractions	30 mins	Emergence	Chin lift	Tracheal tube

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Legends to tables:

Table 1. Type of surgery.

Table 2. Major craniofacial reconstructions.

Table 3. Patient demographics.

Table 4. Age appropriate tracheal tube sizes (based on the formula: Tracheal tube internal diameter (mm) = age/4 + 4).

Table 5. Details of patients who experienced supraglottic airway obstruction.

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*Conversions from US\$ or € will be made at our monthly rate.

Please debit my credit card number:

Verification ID Number: This can be found on your credit card (Visa/MasterCard have 3 numbers and Amex has 4)

Name on card _____ Expiry Date _____ / _____

With the sum of NIL

Signature Sarah Samuels Date 12/9/10

Note: payment by credit card can only be accepted if **all** sections are completed.

Please use BLOCK CAPITALS where possible.

Payment by Purchase Order

If you are asking your university or institution to pay we require purchase order details

I attach order number _____ dated _____

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