South Afr J Anaesth Analg. 2022;28(5 Suppl 1):S162-169
https://doi.org/10.36303/SAJAA.2022.28.5.2908

South Afr J Anaesth Analg
ISSN 2220-1181 EISSN 2220-1173

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Syndromic paediatric airway

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The syndromic paediatric airway poses a potentially difficult airway due to the anatomical and functional difficulties that are seen in the management of this paediatric airway.[1] A difficult airway is described as one where a trained anaesthesiologist experiences difficulty with face mask ventilation or difficulty with tracheal intubation, or both.[2] The Mallampati scoring system for airway assessment is not always reliable in the paediatric population, therefore some clinicians prefer to use the Colorado Paediatric Airway Score (COPUR) since it is more detailed and possibly more reliable.[2] The syndromes can be divided into conditions associated with hypoplasia of the mandible (micrognathia), which can result in a difficult intubation; midface hypoplasia, which can result in difficult bag-mask ventilation; macroglossia, which can cause difficult bag-mask ventilation and difficult intubation. Syndromes related to cervical spine abnormalities may cause difficulties with laryngoscopy and intubation.[1]

Syndromes associated with:

Micrognathia – include Pierre Robin sequence, Treacher Collins, Goldenhar

Midface hypoplasia – Apert syndrome, Crouzon, Pfeiffer

Macroglossia – Hurler's syndrome, Beckwith-Wiedemann syndrome and Down syndrome

Cervical spine abnormalities – Klippel-Feil, Turner and Noonan's syndrome

In the syndromic patient, proper planning and preparation are key in order to reduce the mortality and morbidity associated with the management of the difficult airway.[3] The entire theatre team needs to be briefed on the plan for managing the airway. A thorough history and examination should be conducted on the patient, taking special note of any upper airway obstruction and its severity. Different airway equipment such as supraglottic devices, video laryngoscopes and fibreoptic bronchoscopes should be available in theatre depending on the cases, available resources and expertise of the airway manager.[4] The primary goal for tackling the difficult airway is to keep the patient breathing spontaneously and to achieve a deep plane of anaesthesia before airway instrumentation. Knowledge of the difficult airway algorithm is key, as it allows one to manage the difficult airway in a stepwise approach.

Keywords: syndromic paediatric airway, upper airway obstruction, difficult paediatric airway

Introduction

The paediatric patient has significant anatomical and physiological differences in their airway compared to an adult.[5] This impacts the technique and tools the anaesthesiologist might use to provide safe anaesthesia to the patient. In addition, there are several pathological processes, particularly in the paediatric patient, which present anatomical as well as functional difficulties in the management of the paediatric airway. The presence of these syndromes can predict a difficult airway.[5]

The ASA Task Force on the Management of the Difficult Airway defines a difficult airway as "the clinical situation in which a conventionally trained anaesthesiologist experiences difficulty with face mask ventilation of the upper airway, difficulty with intubation or both".[2] In a retrospective review of 11 219 paediatric patients, the risk of difficult laryngoscopy was estimated at 1.35%.[2] The smaller the child, the higher the risk of a difficult airway, with neonates and infants having the highest risk.

Airway assessment in the paediatric population is not always easy, and even more difficult the younger the child, due to the child's inability to communicate or cooperate, inherent anxiety, and possibly developmental delay. Scoring systems that have been traditionally used in adults, such as the Mallampati score, cannot be reliably used in children, thus some clinicians prefer to use the Colorado Paediatric Airway Score (COPUR), see Table I.[2]

Certain features that are predictors of a difficult airway in syndromic children are noted in Table II. These features include the presence of dysmorphic features, limited mandibular space (retrognathia, micrognathia, and mandibular hypoplasia), which are features of Pierre Robin sequence and Treacher Collins syndrome. Other features may include limited neck extension as seen in Klippel-Feil syndrome as a result of fusion of the cervical vertebrae, or macroglossia, limited mouth opening or restricted mobility of the temporomandibular joint which are features of Beckwith-Wiedeman syndrome.

Table I: Details of the COPUR airway scoring system[2]

		Points
C – Chin	From the side view, is the chin	
	• Normal size?	1
	• Small, moderately hypoplastic?	2
	• Markedly recessive?	3
	• Extremely hypoplastic?	4
O – Opening	Interdental space between the front teeth	
	• > 40 mm	1
	• 20–40 mm	2
	• 10–20 mm	3
	• < 10 mm	4
P – Previous intubation or obstructive sleep apnoea (OSA)	• Previous intubations without difficulty	1
	• No past intubations, no evidence of OSA	2
	• Previous difficult intubations, or symptoms of OSA	3
	• Difficult intubation, extreme or unsuccessful; emergency tracheostomy, cannot sleep supine	4
U – Uvula	Mouth open, tongue out, observe palate	
	• Tip of uvula visible	1
	• Uvula partially visible	2
	• Uvula concealed, soft palate visible	3
	• Soft palate not visible at all	4
R – Range	Observe line from ear to orbit, estimate range of movement, looking up and down	1
	• > 120°	2
	• 60–120°	3
	• 30–60°	4
	• < 30°	
Modifiers: add point for		
• Prominent front 'buck' teeth		1
• Very large tongue, macroglossia		1
• Extreme obesity		1
• Mucopolysaccharides		2

Prediction points	Intubation difficulty	Glottic view
5–7	Easy, normal intubations	1
8–10	More difficult, laryngeal pressure may help	2
12	Difficult intubation, fibreoptic less traumatic	3
14	Difficult intubation, requires fibreoptic or other advanced methods	3
16	Dangerous airway, consider awake intubation, advanced methods, potential tracheotomy (Patients with hypercarbia awake, severe obstruction)	4
16+	Scores > 16 are usually incompatible with life without an artificial airway	

Table II: List of possible factors complicating airway management in several congenital syndromes[5]

Syndrome	Airway implication
Pierre Robin sequence	Micrognathia, glossoptosis, cleft palate
Goldenhar syndrome	Micrognathia (unilateral), cervical dysfunction
Treacher Collins syndrome	Micrognathia, small oral opening, zygomatic hypoplasia
Apert syndrome	Limited cervical motion, macroglossia, micrognathia, midface hypoplasia
Hunter and Hurler syndromes	Cervical dysfunction, macroglossia
Beckwith-Wiedeman syndrome	Macroglossia
Freeman-Sheldon syndrome	Circumoral fibrosis, microstomia, limited cervical motion
Down syndrome	Atlanto-occipital abnormalities, small oral cavity, macroglossia
Klippel-Feil syndrome	Cervical fusion
Hallermann-Streiff syndrome	Microstomia
Arthrogryposis	Cervical dysfunction
Cri-du-chat syndrome	Micrognathia, laryngomalacia
Edwards syndrome	Micrognathia
Fibrodysplasia ossificans progressive	Limited cervical motion

It is important to note that as well as having airway problems, the syndromic child may also have other associated conditions that may involve other organs, such as renal or cardiac. They may also have developmental delay, susceptibility to malignant hyperthermia and intracranial abnormalities, which may all affect your approach to the difficult airway. It is thus imperative to do a thorough preoperative assessment where a history is taken, a physical examination is conducted, and any investigations are done which may assist you in planning your anaesthetic.[1] Evaluation of the syndromic child should focus on a potentially difficult airway. Signs and symptoms of airway obstruction should be sought, history of apnoea episodes and daytime somnolence should be noted. One should also look for any stridor, snoring, and increased work of breathing. The clinical examination should focus on facial anomalies (micrognathia or midface hypoplasia), anomalies of the palate and mandible, the extent of mouth opening, as well as the assessment of the thyromental distances.[2]

Syndromes associated with micrognathia or mandibular hypoplasia

Pierre Robin sequence

Pierre Robin sequence (PRS) has an incidence of 1:5 000 to 1:85 000. The clinical triad of micrognathia (small mandible), glossoptotsis (backward, downward displacement of the tongue), and airway obstruction, defines PRS, see figure 1.[7] A cleft palate is common in these patients, but it does not occur in all patients with PRS. A sequence is a pattern of congenital anomalies that result from a single defect. In PRS, the abnormal mandible displaces the tongue into the nasopharynx, which may prevent fusion of the palate, hence the cleft palate.[8] Glossoptosis gives rise to airway obstruction. Due to its combination of features, PRS is commonly viewed as a classic anticipated difficult airway scenario.[4] At birth, neonates typically have respiratory distress, some have gastro-oesophageal reflux, aspiration and feeding difficulties. Placing the neonate prone or lateral tends to relieve the airway obstruction if severe. A nasopharyngeal airway can also be placed to bypass the obstruction.[8] PRS can be associated with other syndromes, commonly Treacher Collins and Velocardiofacial syndrome. These patients can present for various surgeries, including plastic surgery such as mandibular distraction surgery, where the mandible is brought forward, or

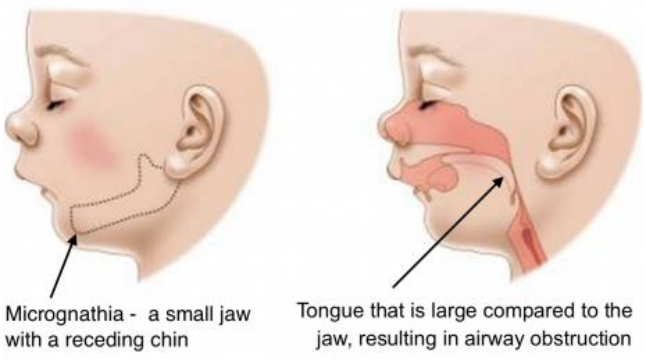

Figure 1: Micrognathia in PRS

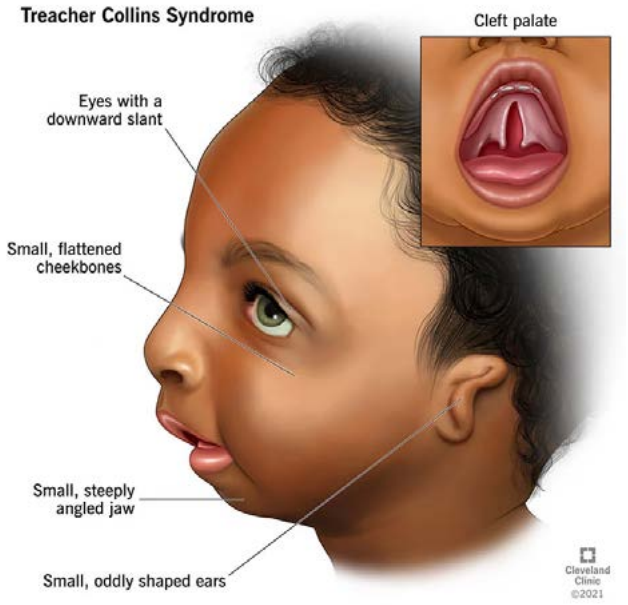

Figure 2: Features of Treacher Collins syndrome[13]

tongue lip adhesion surgery where the tongue is lifted from the posterior aspect of the pharynx to prevent obstruction.[7]

Issues of concern:[8]

- A challenging airway which is difficult to ventilate and intubate due to craniofacial dysmorphology.
- Postoperatively patients can develop spontaneous airway collapse due to pre-existing airway obstruction, possible obstructive sleep apnoea and increased sensitivity to opioids.
- Aspiration is a potential problem.

Treacher Collins syndrome

Treacher Collins syndrome (TCS) is a rare autosomal-dominant disorder; the hallmark features include micrognathia and abnormal development of the zygomatic arch, all of which may result in upper airway obstruction.[9] Features that can impact airway management include bony hypoplasia of the maxilla, zygoma and mandible, small oral aperture, and temporomandibular joint abnormalities (Figure 2). Other findings include hypoplasia of the soft tissues of the malar bone.[10] Cleft lip and palate are present in approximately 40% of cases as well as a high arched palate.[11] As they age, the upper airway obstruction may worsen. It has an incidence of 1 in 50 000 live births. It is often associated with deafness due to meatal atresia and malformation of the inner ear.[11] Patients with TCS often require multiple surgeries to correct facial abnormalities.

Anaesthetic concerns include a difficult airway as bag-mask ventilation as well as direct laryngoscopy can be challenging. In a retrospective review of 59 patients with TCS[12] who had multiple anaesthetics for various procedures, 54% had a grade 4 Cormack–Lehane and 5% had failed intubations.[12]

Edward syndrome or Trisomy 18 may also be included here as it is associated with micro/retrognathia, and these patients often

have a small mouth. All these features may result in a difficult direct laryngoscopy.

Syndromes associated with craniofacial synostosis

Craniosynostosis refers to the abnormal premature fusion of the skull bones.

Apert syndrome (acrocephalosyndactyly)

Apert syndrome was named after the French paediatrician Eugene Apert, who described the signs. It has an incidence of 1 in 160 000 live births. It accounts for 4.5% of all craniosynostosis cases. Brachycephaly is common, whereby the coronal sutures fuse prematurely, resulting in a reduced anterior-posterior distance of the head. Other common features include midface hypoplasia, choanal atresia, hypertelorism and syndactyly of the hands and feet.

Associated anomalies, although rare, include cardiac defects, polycystic kidneys and pyloric stenosis.[14,15] These patients usually present for surgery for syndactyly release, dental surgery, craniosynostosis and orthopaedic procedures.

Fused cervical vertebrae are found in two-thirds of patients which may cause problems with intubation.[2] They may also have fused tracheal rings which may require a smaller than normal endotracheal tube.[2] They may also have a problem clearing secretions due to a stiff trachea, thus they are prone to increased secretions and wheezing.

Pfeiffer syndrome

Pfeiffer syndrome has an incidence of 1 in 100 000. In addition to craniosynostosis, the patients often have midface hypoplasia, a narrow nasopharynx or choanal atresia. They may also have a cleft palate, all of which can potentially lead to a difficult airway.

Crouzon syndrome

Crouzon syndrome has an incidence similar to Apert syndrome, they have similar airway problems of midface hypoplasia, flat foreheads, and narrowed nasopharynx – all of which lead to upper airway obstruction and possible development of obstructive sleep apnoea. They may have cervical spine fusion at C2–C3.

The major anaesthetic concern in this group of syndromes relates to the bag-mask ventilation due to midface hypoplasia and potential proptosis, thus a mask may not sit properly. Direct laryngoscopy may not typically be difficult unless there are cervical spine abnormalities. They may also have obstructive sleep apnoea.

Syndromes associated with hemifacial microsomia

Goldenhar syndrome

Goldenhar syndrome is a craniofacial disorder characterised by hemifacial microsomia. Associated anomalies include cleft palate, malar and mandibular hypoplasia, and cervical vertebral hypoplasia.[16] The aetiology is multifactorial, or possibly sporadic. It can be associated with other conditions such as cardiac defects. The main anaesthetic concerns are difficult intubation and difficult bag-mask ventilation. The facial asymmetry may make mask ventilation difficult. Difficult intubation is due to an asymmetrical mandibular hypoplasia, hemifacial microsomia and tracheal deviation to one side.[2] Vertebral anomalies may include fused vertebrae which may make neck extension difficult.[17] The laryngeal mask airway, video laryngoscopes and airtraq have been used successfully to secure airway.

Syndromes associated with macroglossia

Down syndrome

Down syndrome is the most common genetic abnormality and has an incidence of 1 in 700 live births. The features most often seen are macroglossia, midface hypoplasia and a narrow hypopharynx. Decreased pharyngeal tone together with macroglossia can lead to upper airway obstruction. This is compounded by an increased incidence of tonsillar and adenoid hypertrophy.[2] The trachea and nares may be smaller than average, and there is an increased incidence of subglottic stenosis. There are other associations such as cardiac anomalies, and they are at increased risk of diseases such as Hirschsprung disease and leukaemia.[18] They may also have atlantoaxial instability which is caused by laxity of the transverse ligament or malformation of the odontoid process. Airway concerns include potentially difficult mask ventilation, and the use of an oral airway may be needed. However, laryngoscopy tends to be straightforward, as the soft tongue can be displaced by the laryngoscope blade. The presence of any neurology should be investigated preoperatively. Cervical spine x-rays are not routinely done as they do not reliably rule out instability. Instead neck flexion-extension should be kept to a minimum and a soft collar can be placed to maintain the neck in a neutral position.[2]

Beckwith-Wiedemann syndrome

Beckwith-Wiedemann syndrome is mainly characterised by macroglossia, omphalocoele and gigantism. It has an incidence of 1 in 14 000 live births.[2] Macroglossia is seen in 95% of patients, and early treatment includes surgical reduction.[19] Anaesthetic concerns include upper airway obstruction and difficult laryngoscopy. During induction, placing the patient prone or lateral can help relieve the obstruction. Intubation can be made easier by having an assistant pull the tongue out of the way with forceps. The tracheal diameter can be bigger than average, thus it is usually recommended that a cuffed tube be used.[2] It is important to note that tongue oedema can occur postoperatively and these patients need to be monitored in a high dependency unit.[19]

Mucopolysaccharides

The mucopolysaccharides (MPS) are a group of hereditary disorders that lead to organ failure and a shortened lifespan.

They are caused by deficiencies of different lysosomal storage enzymes creating a build-up of glycosaminoglycans (GAGs) in various tissues throughout the body. Hurler syndrome (type 1) is the most severe and best-known form. Airway management is of concern in patients with MPS. Deposition of GAGs submucosally in the tongue results in macroglossia, nasopharynx and larynx deposition of GAG can lead to upper airway obstruction.

Other concerns include difficult bag-mask ventilation and difficult direct laryngoscopy, the incidence of difficult airway can be up to 25%.[17] They may also have a short neck, and limited range of movement, intubation or video laryngoscopy is often needed and a supraglottic device should be available as a rescue tool.

Syndromes associated with limited cervical mobility

Klippel-Feil syndrome

Klippel-Feil syndrome is due to fusion of the cervical vertebrae, and it appears as if the head is sitting on the shoulders. Type 1 patients have fusion of C2 and C3, type 2 patients have long fusion below C2 with an abnormal occipital-cervical junction, and type 3 have thoracic and lumbar spine anomalies together with type 1 and type 2. It is characterised by a short, sometimes webbed neck, low and posterior hairline and reduced motility of the cervical spine. Other associated anomalies include facial asymmetry, cleft lip and micrognathia.[20] Direct laryngoscopy and tracheal intubation may be difficult due to limited mobility of the cervical spine. There is a risk of spinal cord injury during laryngoscopy and intubation, thus careful neck movement is necessary. LMA and fibreoptic-assisted intubations have been used with success.[2]

Turner syndrome

Turner syndrome is a genetic disorder characterised by an abnormal X chromosome and patients are 45X karyotype. The main anatomical changes relating to the airway include a short neck, maxillary and mandibular hypoplasia. They may also have contracture of several joints including the temporomandibular joint, all of these problems may pose a difficult airway. The shorter length of the trachea, as well as its higher bifurcation, can predispose to inadvertent bronchial intubation.[21]

Noonan's syndrome

Noonan's syndrome is a multisystem congenital disorder. The main airway anaesthetic problems include a short neck micrognathia, limited mouth opening, high arched palate and macroglossia, all of which pose a potentially difficult airway to the anaesthesiologist.[22]

The above syndromes, with limited cervical mobility, can make direct laryngoscopy difficult.[17]

How to manage the anticipated difficult paediatric airway

The anaesthesiologist must always anticipate a potentially difficult airway in children who are syndromic. It may be a difficult bag-mask ventilation, a difficult intubation or both. The possibility of a difficult cricothyroidotomy should also be considered, especially in a "can't intubate, can't ventilate" situation. Proper planning is paramount for managing a difficult paediatric airway. Although the anaesthetic plan will be guided by the physical examination of the patient, history, planned surgical procedure and the anaesthesiologist's experience, the available resources will also impact the plan. Since paediatric patients desaturate quickly, each patient needs to be evaluated thoroughly and plan for unforeseen circumstances.[4] A history of any upper respiratory tract infections should be noted as this may increase the morbidity in the difficult airway. The most experienced anaesthesiologist in dealing with paediatric airways should be available; other specialities, such as ENT surgeons and general surgeons, should be available immediately in theatre if deemed necessary. The anaesthetic plan should be discussed with the patient, the parents and the entire surgical team.

The LEMON approach has been used in adults to identify the difficult airway. The tool has not been used in children but can be extrapolated for use in older cooperative children.[6]

L: Look externally for indicators of a difficult airway (facial anomalies)

E: Evaluate mouth opening, and thyromental distances

M: Mallampati score

O: Obstruction, look for signs of airway obstruction

N: Neck mobility, congenital anomalies that limit neck movement

As part of the preparation plan, alternative airway techniques for providing oxygenation and ventilation should be considered in the child who might be difficult to intubate with direct laryngoscopy.

Preparation of airway equipment should also include:[4]

- Various face masks, nasal and oropharyngeal airways, and introducer stylets.
- Two laryngoscopes and a straight blade (McCoy may be more suitable).
- Multiple endotracheal tubes of varying sizes.
- Supraglottic devices for airway rescue and conduit for tracheal intubation (Fastrach™ LMA).
- Other appropriate intubating devices, e.g. Glidescopes, airtraq devices, video laryngoscopes and flexible fibreoptic bronchoscope.
- Optical stylets, this device combines features of the lighted stylet with features of a bronchoscope to create a device to be used for blind intubation with the added advantage of a camera to visualise the glottis.
- Equipment for surgical cricothyroidotomy should be available for potentially difficult cases. This should only be attempted as a last resort and in experienced hands.

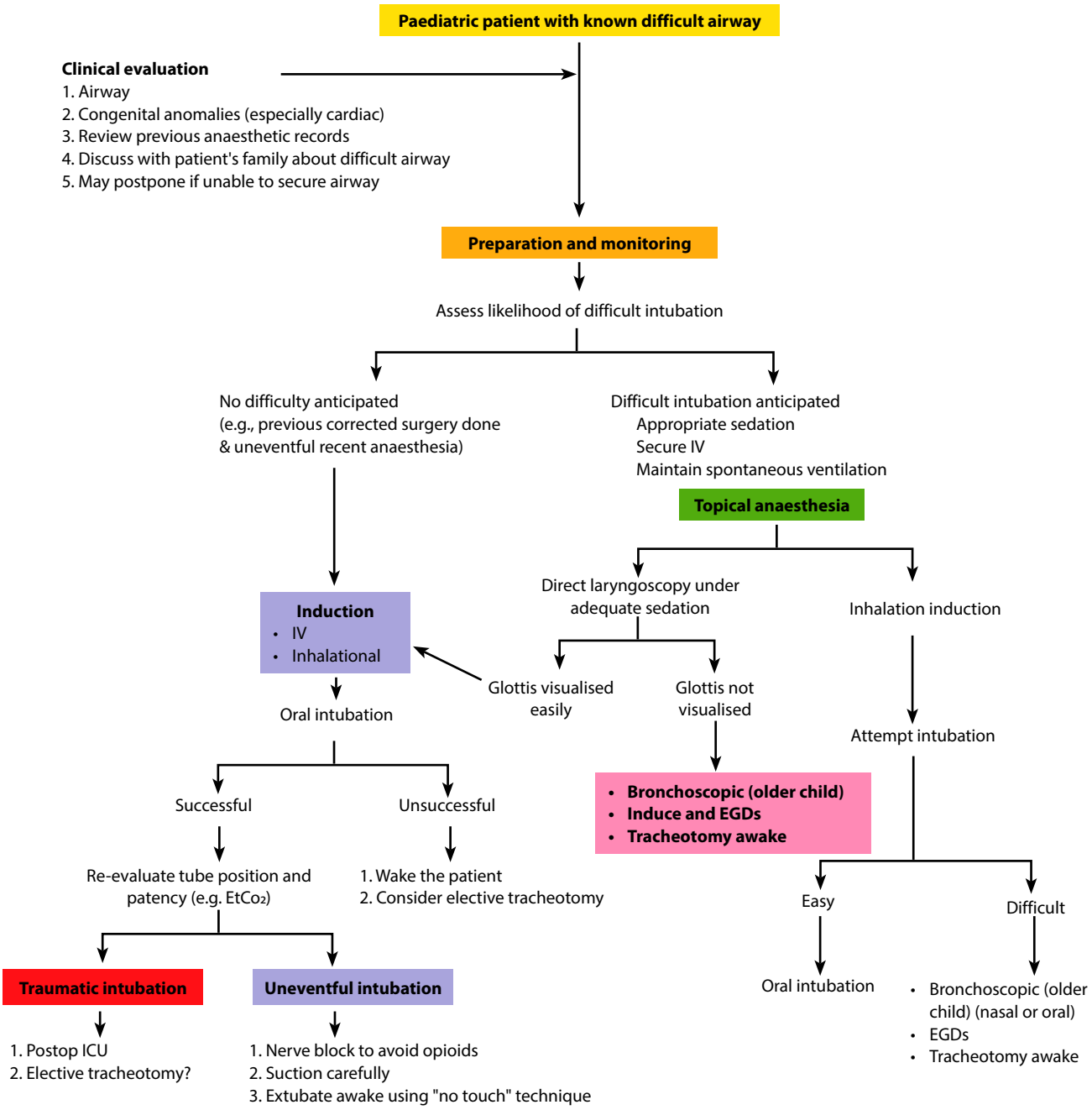

Figure 3: Algorithm for the paediatric patient with a known difficult airway[4]

Induction technique[4]

The primary objective for induction in a difficult airway is maintaining spontaneous ventilation, and achieving a plane of anaesthesia that will allow airway instrumentation.[4]

- Induction should be smooth and gradual.
- Sedatives that are known to cause loss of airway muscle tone should be avoided.
- Atropine or glycopyrrolate can be given to dry secretions.
- Try to obtain intravenous access using EMLA cream prior to induction for difficult airways.
- Inhalation induction remains popular for spontaneous ventilation
- An IV propofol 10–20 ug/kg/min and remifentanil 0.05 ug/kg/min infusion will allow most patients to breathe spontaneously.
- Mask-ventilation technique should be such that the airway is not obstructed.
- If obstructed, various techniques can be employed in a stepwise approach, such as moving the child to the prone or lateral position.
- Do a gentle chin lift/jaw thrust.
- If this fails to adequately oxygenate, use of an appropriately-sized nasopharyngeal or oral airway can be made.
- If this fails, use of a supraglottic device such as an i-gel LMA can be made.
- If surgery requires an endotracheal tube, direct laryngoscopy or some other intubating device can be used, such as a video laryngoscope.
- Minimise attempts at direct laryngoscopy to reduce airway trauma.

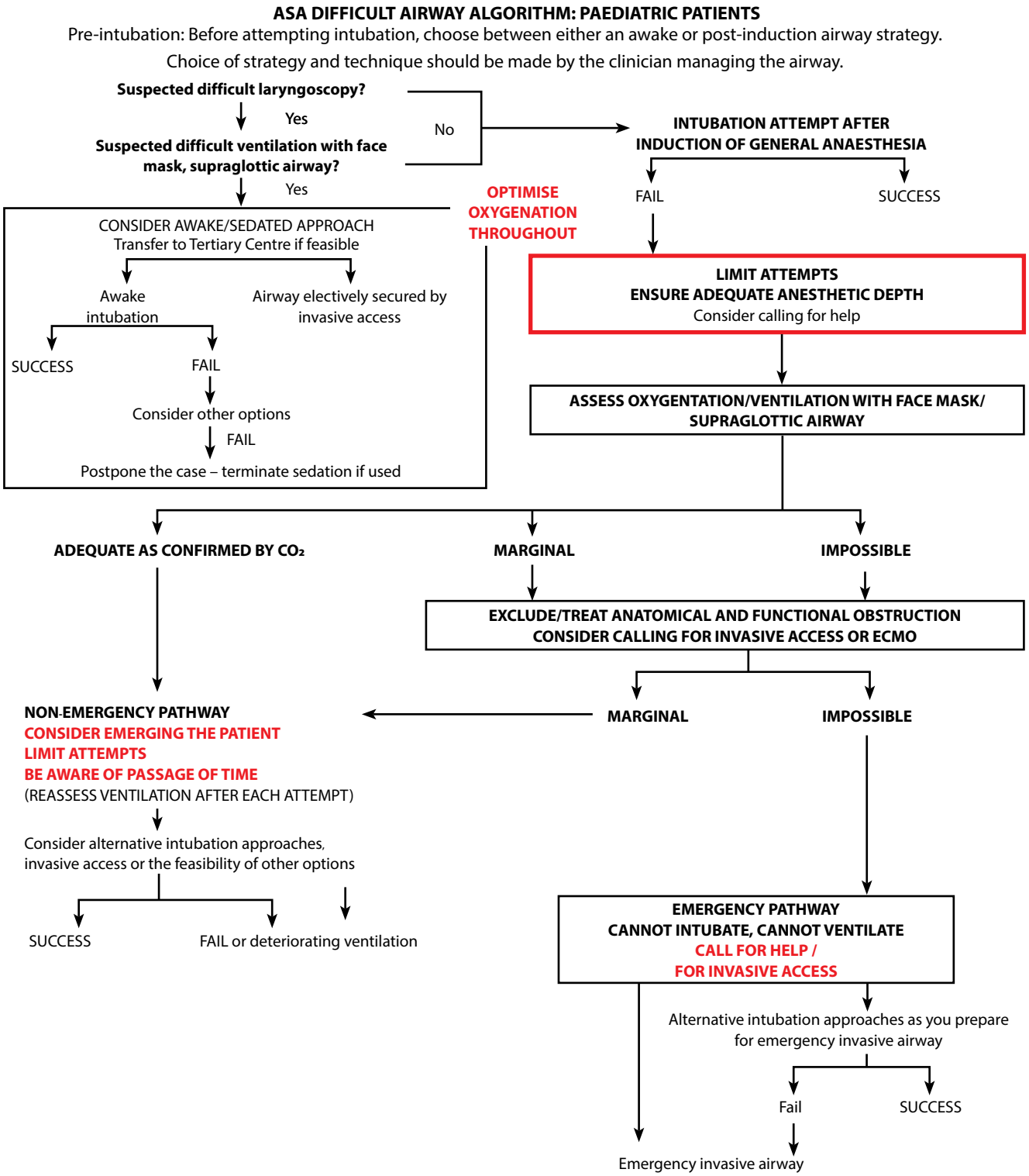

Figure 4: ASA difficult airway algorithm: paediatric patients[23]

Figure 3 and Figure 4 provide algorithms for the difficult paediatric airway.

It must be emphasised that regional techniques, where possible, should be used to avoid instrumentation of the airway and reduce the amount of opioids used. Awake fibreoptic bronchoscope can be attempted in the older child who is cooperative, although this approach is rarely used in children.

At end of surgery, if extubation is the plan, the patient should be extubated fully awake, and in the position least likely to cause airway obstruction. All airway equipment that was present at induction should be available and ready for use at extubation if necessary.

Conclusion

There are multiple syndromes that have an important bearing on the airway in the paediatric population. It is important for the anaesthetist to know how the different syndromes can impact the airway. A multidisciplinary approach to airway management and surgery should be obtained, and a clear and concise plan

should be communicated to the surgical team regarding the airway management. This can reduce morbidity and mortality associated with these syndromes and airway management.

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