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Original Article

Laryngotracheal stenosis post mechanical ventilation in paediatric burns patients

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Abstract: Introduction: The duration of endotracheal intubation is thought to be the most important factor in the development of acquired laryngotracheal stenosis (LTS); however, there is a paucity of studies examining the incidence of LTS in the paediatric burn population. The aim of this study was to determine the incidence of LTS in paediatric burns patients requiring mechanical ventilation to develop guidelines for consideration of a tracheostomy. Methods: A retrospective review of all children treated at The Children's Hospital at Westmead (CHW) Burns Unit (BU) from December 2009 to December 2019 who required intubation for their burn injury. Results: During the 10-year study period 115 patients required endotracheal intubation after having sustained a burn injury. Of these 11 were excluded. The mean age was 6.2 years (0-16), with the majority of patients being male (65%). The average TBSA was 18.5% with a range of 0.1-70%. Flame was the most common mechanism of burn (n = 59). Burns to the head and/or neck were the most common indication for intubation with the mean duration of intubation 6.1 days (range 0-40). Tracheostomies were performed on two patients (1.9%). LTS was found in two patients (1.9%). Conclusion: LTS in the paediatric burn population post mechanical ventilation appears to be a rare event. Endotracheal intubation can safely be used as the route of airway access in paediatric burns patients. Based on our experience, a definitive recommendation on the timing of tracheostomy in the paediatric burn patient cannot be made.

Keywords: Laryngotrachael stenosis, sub glottic stenosis, tracheal stenosis, paediatric burns, mechanical ventilation

Introduction

Major paediatric burns, particularly inhalational injuries, often require endotracheal intubation and mechanical ventilation. Laryngotracheal stenosis (LTS) is the narrowing of the upper airway, which is comprised of the larynx, glottis, subglottis and trachea, Duration of endotracheal intubation is thought to be the most important factor in the development of acquired LTS [1, 2]. The larynx immediately below the vocal cords represents the narrowest part of a paediatric airway and is at risk of injury in the presence of an endotracheal tube (ETT), leading to LTS after long-term mechanical ventilation [1, 3, 4]. Burn-related oedema may further exacerbate tracheal narrowing, increasing the risk of mucosal damage. Studies have reported an incidence of up to 3% in burns patients following endotracheal intubation or tracheostomy and 5%-23.5% in burns patients with an inhalational injury [3, 5, 6]. For patients requiring long term mechanical ventilation, creation of a tracheostomy has been suggested to reduce the risk of LTS, although the routine use of tracheostomies in burns patients remains controversial with no clear consensus on timing or indications. Reported rates of tracheostomies performed in the adult population range from 0.9% of all burns patients and 14% of burns patients requiring mechanical ventilation within 3 days of admission, continuing until at least day 5 [7-9]. Given the paucity of studies examining the incidence of LTS in the paediatric burn population, we sought to review the contributing factors and evaluate the outcome of LTS in paediatric burns to inform guidelines for consideration of a tracheostomy.

Methods

Study design

A retrospective study was undertaken from December 2009 to December 2019 of children who were treated at The Children's Hospital at

Table 1. Patient data

All patients (n)	104
Male (n)	68
Female (n)	36
Age (mean, range)	6.2 (0.0-16.0)
TBSA (% mean, range)	18.5 (0.1-70)
Mechanism	
- Scald	39
- Flame	59
- Electrical	3
- Chemical	1
- Inhalational	1
- Friction	1
Reason for intubation	
- Head/neck burn	68
- TBSA > 15%	36
- Stridor/hoarse voice/swollen lips	28
- Suspected inhalation injury	4
- Other	4
- Unconscious patient	2
Average duration of intubation (days, range)	6.1 (0-40)
Number of LBOs	17

Westmead (CHW) Burns Unit (BU) for a burn injury and required endotracheal intubation. CHW is part of The Sydney Children's Hospital Network (SCHN) and includes the sole tertiary referral centre for paediatric burns in New South Wales, Australia. The study cohort comprised of children under the age of 16 years old who required intubation due to burn injury. Patients were excluded if the indication for endotracheal intubation was unrelated to their burn injury. Within this cohort, the two groups for comparisons were patients who had a documented history of LTS and those that did not.

Data collection

Data were entered prospectively into the Agency for Clinical Innovation (ACI) New South Wales Statewide Burn Injury Service (NSW SBIS) database. The ACI NSW SBIS database collects information of all patients with burn injuries treated at the three Statewide Burn Injury Units of NSW, with those being CHW, Royal North Shore and Concord Repatriation General Hospital. Information collected included patient information (age and gender), mechanism of injury, location of injury, percentage of total body surface area (TBSA) affected, number of days with intubation, finding of LTS, investigations and management of LTS. The key indicator in this study was a documented incidence of LTS post mechanical ventilation.

Statistical analysis

Descriptive statistics were utilised for the majority of variables.

Ethics

The Sydney Children's Hospital Network Human Ethics Research Committee approved this study (HREC Reference 2019/ETH13748).

Results

Patient data

During the 10-year study period, 115 patients were identified to have endotracheal intubation after having sustained a burn injury. Of these, 11 were excluded: 7 as they were intubated in relation to other injuries, 3 as they were intubated following an adverse drug reaction and 1 as the patient died as a result of their burn injuries on day 38. The remaining 104 patients were all included for analysis (**Table 1**).

Burn characteristics

The mean age was 6.2 years old (0-15), with the majority of patients being male (65%). The average TBSA was 18.5% with a range of 0.1-70%. Flame was the most common mechanism of burn (n = 59), followed by scald (n = 39).

Burns to the head and/or neck were the most common indication for intubation, followed by TBSA greater than 15% and patients displaying stridor, hoarse voice or swollen lips. The mean duration of intubation was 6.1 days (range 0-40). Sixty-eight patients underwent orotracheal intubation initially (of which 3 were changed to nasotracheal), while 31 patients underwent initial nasotracheal intubation. Route of intubation was not recorded for 5 patients. 81 patients were intubated with a cuffed endotracheal tube, although cuff inflation pressures were not recorded for any patients. Of the remaining 23 patients, 14 were intubated with uncuffed endotracheal tubes and in 9 this was not recorded.

Mechanical ventilation characteristics

Of the patients that did not suffer LTS or undergo a tracheostomy the mean duration of intubation was 5.4 days, with a range of 0-38 days. A laryngoscopy, bronchoscopy & oesophagoscopy (LBO) was performed in 17 patients during their admission. Indications for LBO include; stridor post extubation or failed extubation (n = 6), airway oedema noted on intubation (n = 6), concern of airway burns given mechanism of burn and assessment prior to tracheostomy. Tracheostomies were performed on two patients (1.9%): one was performed on day 18 post burn due to ulceration of the vocal cords related to the ETT cuff. The other was performed electively on day 40 post burn given an expected prolonged period of intubation. Both were decannulated prior to discharge, although one patient did later require surgical intervention for the management of a trachea-cutaneous fistula. Two patients were found to have LTS (1.9%). A flame burn was the mechanism for both burns, with both patients' burns involving the head and neck. Patient 1 was intubated for 18 days, while patient 2 was intubated until he underwent a tracheostomy on day 18 post injury - he was decannulated 46 days later. Both these patients required surgical intervention for the management of their LTS. 13 patients were intubated for 18 days or longer, resulting in an incidence of LTS of 15% in patients intubated for at least 18 days. The LBOs identified airway alterations due to then ETT in a further 8 patients for a total of 10 airway changes; subglottic oedema (n = 3), ulcerated vocal cords (n = 2), vocal cord granulation (n = 1), glottic oedema (n = 1), posterior glottic scar (n = 1), posterior glottic ulcer (n = 1) and subglottic ulcer (n = 1). These alterations were noted following a mean duration of intubation of 10.3 days (range 5-18).

Below are case presentations of the two patients that suffered from LTS.

Case description

Patient 1: A 12-year-old boy suffered a 40% TBSA flame burn, including the head and neck, after pouring model car fuel onto a fire. He was intubated orally with a 6.5 cuffed endotracheal tube on presentation to a peripheral hospital due to airway concerns with evidence of inhalational injury noted at the time of intubation. He was extubated on day 18 post injury without complication. Following episodes of worsening shortness of breath, an LBO on day 76 post injury found severe stenosis to the proximal trachea (3 mm lumen, greater than 50% stenosis) requiring dilatation. Following unsuccessful balloon dilatation approximately four weeks later the patient underwent a laryngotracheal reconstruction. Over the subsequent 16 months he required numerous LBOs with a combination of topical mitomycin, corticosteroid injections and radial incisions. Despite this, 22 months later he developed worsening shortness of breath on exertion with an LBO revealing ongoing 40% stenosis requiring a further balloon dilation, radial incisions and corticosteroid injection. There was only some mild improvement post procedure. Given the patients age he was transitioned to adult care.

Patient 2: An 8-year-old boy suffered a 70% TBSA flame burn, including the head and neck, following an unwitnessed event with acetone as the accelerant. He was intubated nasally with a 5.5 cuffed endotracheal tube at a peripheral hospital prior to transfer to our BU. On day 15 post his burn, an LBO identified vocal cord ulceration and a granuloma around the ETT in the larynx. The decision was made to perform an open tracheostomy three days later with a 5.5, long cuffed tracheostomy tube. There was no evidence of airway burn found on LBO. The patient was decannulated on day 64 post burn, but subsequently developed stridor whilst agitated 8 days post decannulation (day 72 post the burn). An LBO revealed elliptical stenosis immediately distal to the tracheostoma with granulation tissue in the previous stoma site. Following removal of the granulation tissue his symptoms improved. The patient was followed up for 29 months following the injury before relocating with his family. He experienced no further symptoms and required no further interventions for his LTS during this time.

Discussion

Given children's airways are comparatively narrow they can become compromised with even minor decreases in airway diameter. In burn related trauma this can be associated with inhalation, ingestion and flame injuries or anterior neck burns and may require intubation to manage [11, 12]. The larynx, immediately below the vocal cords, represents the narrowest part of a paediatric airway and is at risk of injury in the presence of an ETT, with long term intubation the most common cause of acquired LTS [1-3].

LTS has a reported incidence of 2.8%-12% in the paediatric population post intubation (irrespective of the indication for intubation) [1, 3, 13]. In our cohort an incidence of LTS of 1.9% was found in paediatric burn patients requiring endotracheal intubation or tracheostomy for airway management, which is similar to the reported incidence of 3% in adult burns population [3].

The true incidence of LTS is difficult to determine as patients with LTS can be symptom free at rest until the lumen is less than 30% of normal [14]. As a LBO is not typically performed unless there are symptoms, a number of patients with mild LTS may go undiagnosed. In our series only 16% of patients (n = 17) underwent an LBO, meaning our true incidence may be much higher. Similarly, our rate of other airway alterations, 7.7% (n = 8) is significantly lower than 38.2% of moderate to severe laryngeal alterations reported in a prospective study, where all paediatric patients who were intubated for longer than 24 hours underwent flexible fibre-optic laryngoscopy within 8 hours of extubation [2]. This further suggests that the true incidence of LTS in our cohort may be higher given the airway alterations have the potential to develop into LTS - with almost 30% of all patients with laryngeal alterations in the prospective study developing LTS [2]. Another limitation of our study was that the focus remained the issue of LTS following burn injury: we did not examine the requirement for long-term ventilation and any additional complications that may have arisen from this intervention.

While the two cases described in our study are categorised as LTS, given one case occurred post endotracheal intubation (patient 1) and the other post tracheostomy (patient 2) their aetiology is likely quite different.

LTS post endotracheal intubation is reported to occur at the level of the endotracheal cuff in one third of reported cases [15]. Mucosal ischemia is induced when cuff pressure exceeds capillary blood flow pressure, which can result in subsequent necrosis. As the mucosa heals by secondary intention scaring can occur resulting in stenosis [3, 14, 15]. This process occurs over several weeks with patients often not becoming symptomatic many weeks or months post initial injury [3, 14, 16]; patient 1 did not become symptomatic until approximately 8 weeks post extubation. The use of high volume, low pressure cuffs and maintaining cuff pressures between 20-30 cm H_2O, which is below the tracheal mucosa capillary pressure of 25-30 mmHg, has been shown to reduce the incidence of LTS in adults [14, 16]. In children, tracheal mucosa capillary pressure is assumed to be less, given their lower mean arterial blood pressure, with recommended safe cuff pressures between 15-20 cm H_2O [17, 18].

In contrast, cases of LTS post tracheostomy typically result from excessive granulation tissue formation around the tracheal stoma site, as was the case for patient 2. Excessive granulation tissue can result from fractured cartilage during the initial procedure, mechanical leverage of the tracheal tube leading to pressure necrosis, wound sepsis or previous cervical or tracheal trauma [15, 19].

Inhalational injury has previously been reported to be associated with an increased risk of developing LTS [3, 13, 26]. The cumulative trauma sustained by both an inhalational injury and endotracheal intubation/tracheostomy has been postulated to be the cause of this increased risk [11]. Only one of the two patients with LTS suffered from a documented concurrent inhalational injury. We do note however that he required more interventions than the other patient. The significance of this in the context of such a small sample size is uncertain. Given the mechanism and distribution of patient 2's injury, an inhalational burn may have been possible but not recognised. The trend for patients with LTS who have had a previous inhalational injury requiring more exhaustive management than those without an inhalational injury has been previously reported. This highlights that burns clinicians should have a lower threshold to investigate for LTS in patients with known inhalational injury [3, 13, 16].

Determining the optimal route of prolonged airway management in children with severe burns remains a difficult clinical dilemma. Long term endotracheal intubation is associated with LTS, vocal cord ulcerations, sinusitis, unplanned extubation and main stem intubations. Tracheostomies have been reported to be safe and effective in severely burned adults and children, offering benefits such as improved patient comfort, oral feeding, earlier ambulation and facilitating pulmonary toileting, which is particularly beneficial to patients with inhalation injury who are at greater risk of pneumonia [8, 10, 20]. Conversely, tracheostomies also have a reported short-term complication rate of between 3-10% [21] and long-term morbidity such as LTS, dysphagia, cosmetic deformities, voice changes, trachea-oesophageal fistulas and tracheomalacia [8, 20, 22]. The results of a limited meta-analysis of critically ill adults and children found that percutaneous tracheostomy techniques offer some benefits in terms of wound infections and unfavourable scaring, when compared to open techniques, however given the limitations of the study the findings cannot be applied to all critically ill patients [23].

The ongoing controversy surrounding the timing and indications of tracheostomies has meant their routine use among paediatric burns patients is still unclear, although a retrospective review by Palmieri et al. (2002) found early tracheostomy in severely burned children to be safe and effective. Tracheostomies were performed at a mean of 3.9 days post admission and they reported no cases of tracheal stenosis, tracheoesophageal fistulas or dysphagia. While in our cohort of the two patients that underwent tracheostomies one developed tracheal stenosis and one developed a trachea-cutaneous fistula. Conversely, a retrospective review of prolonged trans laryngeal intubation in 98 critically ill children (79% of which had burns) who required mechanical ventilation for a minimum of 7 consecutive days (mean 20 days), found trans laryngeal intubation to be safe and effective, with only one incidence of subglottic stenosis and an unplanned extubation rate of 5.1% [24]. The similarly low incidence of LTS post endotracheal intubation in our cohort supports the finding that long term endotracheal intubation of severely burned children is a safe method of prolonged mechanical ventilation.

In terms of timing of a tracheostomy in burns patients, current evidence suggests there is no improvement in outcomes between early or late tracheostomies. A randomised control trial comparing early (mean day 4) or late (day 14-16) tracheostomies in adult burns patients found no difference in outcomes [8]. A nationwide observational study in Japan also reported no improvement in 28-day mortality when it compared adults with severe burns who required mechanical ventilation within 3 days of admission, continuing until at least day 5, who underwent a tracheostomy [7]. The TracMan study, while not specifically looking at patients with burn, found no mortality benefit from performing an early tracheostomy, comparing day 4 to day 10, in mechanically ventilated adult patients with a high risk of prolonged ventilation [21]. This is in contrast to a meta-analysis of randomised controlled trials comparing early tracheostomy (within 7 days of intubation) to later tracheostomy (any time after 7 days) or no tracheostomy in critically ill patients that found early tracheostomy is associated with lower mortality in the intensive care unit. This review did acknowledge that further studies are needed to examine the long-term mortality and potential complications with tracheostomy [25].

Conclusion

Our series found that LTS in the paediatric burn population post mechanical ventilation is a rare event. Large percentage flame burns, inhalational injury and prolonged intubation should heighten suspicion of the potential for LTS to develop. As such, these patients require close surveillance and appropriately long follow-up to facilitate early diagnosis and timely management. The low incidence of LTS in our series demonstrates that long term endotracheal intubation can safely be used as the route of long-term airway access in paediatric burns patients. Given the available data we are unable to provide a clear recommendation on the role and timing on tracheostomies in paediatric burns patients; rather our study highlights the need for a multi-centre, randomised control trial to further examine these questions.

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Disclosure of conflict of interest

None.

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