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#### **ORIGINAL ARTICLE**

# Tracheotomy decannulation in children under one year of age

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### Abstract

Introduction: Pediatric tracheotomy indications and outcomes have changed over the last decades. More tracheotomies have been performed in the first year of life than in any other pediatric age group. Information on decannulation for this group is scarce in the literature. **Objective:** To characterize indications, outcomes, and decannulation rates in children under one year of age who have been tracheotomized at a Portuguese tertiary hospital over a 15-year period. Materials and Methods: Data was obtained from 16 newborns and infants and analyzed with respect to age, gender, primary diagnosis, indication for tracheotomy, complications, mortality, and time to decannulation. **Results:** Tracheotomy was required following prolonged positive pressure ventilation in 62.5% of patients and for upper airway obstruction in 37.5%. Congenital disorders were diagnosed in 87.5% of patients. Tracheotomy complications were observed in 56.3% of patients with accidental decannulation rate of 25%. The observed mortality rate was 44% at the median age of 13.7 months. Decannulation was performed successfully after tracheotomy in 25% of surviving patients (mean age of 25.5 months). Congenital anomalies were present in almost all cases and were correlated with higher complication and mortality rates, which reflects and precludes decannulation in those patients. Conclusion: Tracheotomy decannulation after clinical evaluation and serial airway endoscopies in children tracheotomized before one year old is a very safe, controllable procedure. It is more feasible in upper airways disorders, with a 50% successful decannulation rate, two years post-operatively.

Keywords: tracheotomy; pediatrics; child; infant; newborn.

## Introduction

The first successful pediatric tracheotomy was performed by Nicholas Habicot in 1620, in France, on a 14-year-old boy who had swallowed a bag of gold coins. In 1825, Bretonneau exerted an influence over the history of this approach when his practice saved a 5-year-old girl with diphtheria, thus increasing the popularity of tracheotomy. In 1833, Trousseau employed this technique in order to treat 50 patients with diphtheria-associated dyspnea, and tracheotomy became a more routine procedure<sup>1</sup>.

During the 1970s and 1980s, the most common indication for tracheotomy was acute inflammatory airway obstructions, such as diphtheria, croup and epiglottitis<sup>2</sup>. Modern techniques in neonatal and intensive care units, as well as vaccines against *H. influenzae* and *C. diphtheria*, have turned prolonged ventilation followed by airway obstruction into the most common indications for pediatric tracheotomy<sup>3</sup>. Furthermore, recent studies suggest a reversal

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Copyright Cardoso et al. This is an Open Access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited. back to upper airway obstruction (UAO) as the most common indication for pediatric tracheotomy<sup>4,5</sup>.

Children, particularly newborns, are extremely vulnerable after birth and in the following days, presenting the highest concentrated lifetime risk of death. The risk increases in premature infants—particularly vulnerable to temperature variability, feeding problems, low blood sugar, infections, and breathing difficulties<sup>6</sup>. Therefore, performing tracheotomy in the pediatric population is technically challenging; it is associated with higher rates of morbidity and mortality compared to adults<sup>7,8</sup>.

Nevertheless, age at tracheotomy has declined over the last 30 years—most procedures are presently done in children in their first year of life. There is scarce reference in the literature about tracheotomies in children under one year of age, and pediatric decannulation has not been reported in the last 20 years. Therefore, the aim of this study was to characterize indications, outcomes, and decannulation rates in children under one-year-old who have been tracheotomized at a Portuguese tertiary hospital over a period of 15 years.

# **Methods**

Data was obtained retrospectively from patients who underwent tracheotomy from January 2000 to April 2016 (15 years and 4 months) at the Pediatric Hospital of Centro Hospitalar e Universitário de Coimbra, in Portugal. The study followed the Declaration of Helsinki, from 2013, on Biomedical Research Involving Human Participants, and was approved by the Ethics Committee of the Faculty of Medicine at Universidade de Coimbra, Portugal.

Patients under one-year old were selected and analyzed with respect to age, gender, date of tracheostomy, indications to tracheotomy, primary diagnoses, complications, decannulation rate, and mortality. Early complications were defined as those occurred intraoperatively or within 7 days postoperatively. Late complications included those experienced after the first week following surgery.

## Results

Sixteen patients under one-year-old were selected for the study, where ten were male (62.5%), and six were female (37.5%). The median age of patients at the time of tracheotomy was 3.5 months, ranging from 14 days to 11.5 months.

Tracheotomy was required for prolonged positive pressure ventilation (PPPV) in 62.5% (10/16) of patients, and for UAO, in 37.5% (6/16). Congenital disorders were diagnosed in 87.5% of cases (14/16) (Table 1).

Mortality rate was 44% (7/16) at a median age of 13.7 months (between 3 and 29 months). Only one child died from a tracheotomy-related complication. Decannulation was performed successfully in 25% (4/16) of patients at a mean age of 25.5 months following tracheotomy, and a mean age of 26.5 months at the time of decannulation.

Indication	Patients (%)
Prolonged positive pressure ventilation	10 (62.5)
Bronchopulmonary disease	4
Neurologic disease	1
Congenital central hypoventilation syndrome	2
Laryngeal cleft	1
Muscular disease	1
Cardiac disease	1
Upper airway obstruction	6 (37.5)
Pierre Robin Sequence	2
Craniofacial dysmorphism	1
Laryngeal papillomatosis	1
Bilateral vocal cord paralysis	1
Goldenhar syndrome	1

Table 1. Tracheotomy indications.

# Discussion

Decisions about tracheotomy in neonates and young infants require careful consideration of several aspects, including mortality, potential morbidities, prospects, alternatives, and the impact of tracheotomy on the family.

Tracheotomy in children under one-year-old range from 23-75%<sup>2,7,9-16</sup>. Our study observed results similar to the reviewed literature, with 73% (16/22) of children tracheotomized under one year old.

The main indication for tracheotomy was the need for prolonged ventilation, notably in children born prematurely (56.3% of the sample) who have developed bronchopulmonary dysplasia. Ninety per cent (9/10) of patients in the PPPV group and 83% (5/6) of patients in the UAO group presented congenital disorders. Only two children (12.5%) were considered to have an acquired disease: one case of pneumonia with sepsis and a second one with laryngeal papillomatosis. Over the last decades, advances in vaccination and antibiotics, as well as improvements in surgical management and intensive care units, have contributed to increase survival of neonates and premature infants, including those with congenital anomalies who may not have survived in the past and now receive tracheotomies earlier<sup>10</sup>. Our results corroborate the evidence that the number of premature children with severe congenital anomalies are high and that infection is no longer a notable indication for tracheotomy.

Regional differences also affect which tracheotomy indications are most prevalent. In papers from France (1996 to 2001)<sup>16</sup>, Spain (1999 to 2008)<sup>15</sup>, Portugal (1995 to 2005)<sup>13</sup>, Singapore (1991 to 2001)<sup>8</sup>, and Turkey (2007 a 2010)<sup>2</sup>, most procedures have also been performed for ventilator dependency.

In studies from New Zealand (1987 to 2003)<sup>10</sup>, the United States (2000 to 2011)<sup>4</sup>, and Canada (1982 to 2011)<sup>11</sup>, UAO was the most common indication, showing a reversal back to UAO as the most common indication, most often due to congenital/syndromic conditions. All of them included children above one-year-old, which hinders comparison of results.

Granulation tissue at the stoma or in the trachea is the most common pediatric tracheotomy-related complication, occurring in 18-80% of patients<sup>2,3,5,9-11,14,17,18</sup>. The presence of granulation tissue is so recurrent that some authors do not even classify it as a complication—unless it compromises the airway—and refer it as sequelae/routine findings, due to the absence of a proven technique capable of preventing this occurrence<sup>19</sup>. We considered all granulomas. These can be removed endoscopically; however, we routinely remove them in the operation room after sedating the child. Granulation tissue occurred in 12.5% of cases, having been the second most common complication in our serie (Table 2.). The most common complication was accidental decannulation, in 25% of patients, compared to 0.7-18% of cases in the reviewed articles<sup>3,10,11,20</sup>. Two occurred in hospital in the first week following the procedure, whereas the other took place at the patient's home. Other complications commonly reported, such as emphysema, pneumothorax, severe hemorrhage, and obstructive plugs, have not been recorded<sup>3,10,18</sup>.

According to the literature and considering all infants and children having undergone tracheotomies, mortality is about 12-20%; only 10-25% of those died from complications related to the tracheotomy. Overall mortality usually increases to 6-32% when infants under one year of age are considered alone<sup>17,18</sup>. Mortality after tracheotomy depends on initial indication, progression of underlying condition, and associated comorbidities. Mortality is lower in potentially reversible conditions, such as airway obstruction, and higher in neurologic impairment, congenital heart disease, and genetic syndromes. The survival rate of children with bronchopulmonary dysplasia who required tracheotomy is around 80-90%, compared to 50-70% for those undergoing tracheotomy following congenital heart disease<sup>17</sup>. Twenty-five per cent of the children (4/16) presented very low birth weight (<1500g) and extremely

Complications	Patients (%)
Early	3 (19%)
Accidental decannulation	2
Air leak	1
Late	6 (37.5%)
Accidental decannulation	2
Stromal granulation	2
Tracheocutaneous fistula	1
Tracheal granuloma	1

#### Table 2. Tracheotomy complications.

low birth weight (<1000g), which are associated with increased risk of death, growth retardation, and delayed neurodevelopment<sup>21</sup>. Multiple severe comorbidities in children with debilitating and low birth weight illnesses led to a high overall mortality rate: 44% in our study. Only one child (6%) died of a tracheotomy-related complication after accidental decannulation at home. All other patients died from disease-related progression or complications. Among the seven children who died, 86% required tracheotomy for prolonged intubation. In addition, six patients presented bronchopulmonary disease associated with cardiac malformations, neurologic disease or multicore myopathy; one child presented a Pierre Robin malformation associated with cardiac malformation.

Children can undergo ward decannulation satisfactorily by progressively downsizing, blocking, and finally removing the tube; alternatively, they can be decannulated in the operating room under meticulous observation. Decannulation rates in children who have undergone tracheotomy before one year of age range from 30 to 75%, with a median interval of 22-48 months<sup>17</sup>. In cases where airway obstruction was the original indication for tracheostomy, the child is considered ready for decannulation if the original obstruction has been surgically corrected or if the natural airway has become adequate over time. The great number of patients in the prolonged intubation group, the severity of the primary disease with multiple comorbidities, and the elevated mortality were associated with our lower decannulation rates: 25%.

Children were submitted to serial endoscopy of the airway before decannulation was attempted. Intraluminal suprastomal granuloma in the anterior wall were excised; cases of subglottic stenosis, corrected; and the opening in trachea and skin, closed. Because of the smaller lumen, underdeveloped inspiratory muscles, increment of dead space, and increased proportional oxygen requirements of children compared to adults, a relatively small tracheal luminal reduction may be highly significant and preclude decannulation. Decannulation success has been associated with age at tracheotomy<sup>22</sup>. Our personal experience reflects this data, and our team tries to perform decannulation only when it seems more probable. This may lead to a prolonged time to decannulation with the associated morbidity; however, decannulation panic has not been a problem among us.

We have only observed successful decannulations in the UAO group: two Pierre Robin malformations, one craniofacial dysmorphism, and one laryngeal papillomatosis. The Pierre-Robin children presented the lowest decannulation time: an average of 6.9 months, and average age of 12.5 months; the decannulation occurred after surgical correction of the cleft palate. The children with craniofacial dysmorphism presented the longest time with tracheotomy; they have been decannulated 63 months after tracheotomy at 66 months of age and following surgical mandibular correction. The children with papillomatosis also presented tracheolaryngomalacia—which precludes earlier decannulation even after laryngeal permeabilization surgery—having only been decannulated 25 months after tracheotomy.

During decannulation follow-up, it is normal to observe a tracheal collapse due to weakening of the cartilage in the tracheotomy region. Although a small stridor is observed for a few days or weeks afterwards, this has not been a problem in our series. We recommend surgically closing the trachea fistula at decannulation in order to attain a better laminar flow in the airways. Rogers *et al.* recommended the same after prolonged tracheotomy<sup>23</sup>.

Some papers describe the use of nasotracheal intubation following decannulation<sup>23</sup>. It allows the child to become accustomed to increased airway resistance prior to definite decannulation. We haven't had such cases. On the other hand, this study presents the limitations usually found in retrospective studies, including the lack of standardized tracheotomy indication and procedures, and perioperative care. Although the study's sample includes only 16 patients, it is the largest sample of the last 20 years addressing the decannulation issue; nevertheless, it is very small. We suggest that a multi-center study be carried out. The varying classifications for indications, as well as the inclusion of children above one year old in most of the papers on pediatric tracheotomy, further encumbers comparisons.

# Conclusions

The most common indication for pediatric tracheotomy in children under one-year-old was prolonged intubation, particularly due to bronchopulmonary disease. Congenital anomalies were present in most of the cases in both the PPPV ventilation group and the UAO group. Mortality was high and related to disease progression or complications, reflecting the severity of the primary disease and precluding decannulation in most of the patients. Pediatric tracheotomy is a relatively safe procedure associated with a low incidence of procedure-related mortality.

Tracheotomy decannulation after clinical evaluation and serial airway endoscopies in children tracheotomized before one year of age is a very safe, controllable procedure. It is more feasible in upper airways disorders with a successful decannulation rate of 50% two years post-operatively.

# References

- Wtmore R. Pediatric tracheotomy. In: Bluestone C, Simons J, Healy G, editors. Bluestone and stool's: pediatric otolaryngology. 5th ed. USA: PMPHouse; 2014. 1565 p.
- Atmaca S, Bayraktar C, Aşilioğlu N, Kalkan G, Özsoy Z. Pediatric tracheotomy: 3-year experience at a tertiary care center with 54 children. Turk J Pediatr. 2011;53(5):537-40. PMid:22272455.
- Özmen S, Özmen ÖA, Ünal ÖF. Pediatric tracheotomies: a 37-year experience in 282 children. Int J Pediatr Otorhinolaryngol. 2009;73(7):959-61. http://dx.doi. org/10.1016/j.ijporl.2009.03.020. PMid:19395057.
- Lawrason A, Kavanagh K. Pediatric tracheotomy: are the indications changing? Int J Pediatr Otorhinolaryngol. 2013;77(6):922-5. http://dx.doi.org/10.1016/j. ijporl.2013.03.007. PMid:23537927.
- 5. Nobre S, Roda J, Félix M, Estêvão MH. Traqueostomia em idade pediátrica: experiência de um quarto de século. Acta Pediatr Port. 2011;42(6):269-73.

- Mangili G, Garzoli E, Tullio DI, Pietro S, Marina R, Alberto C. Feeding of preterm infants and fortification of breast milk. Reprod Health. 2017;39(2, Suppl 1):158. PMid:28673079.
- Corbett HJ, Mann KS, Mitra I, Jesudason EC, Losty PD, Clarke RW. Tracheostomy: a 10-year experience from a UK pediatric surgical center. J Pediatr Surg. 2007;42(7):1251-4. http://dx.doi.org/10.1016/j.jpedsurg.2007.02.017. PMid:17618889.
- Ang AHC, Chua DYK, Pang KP, Tan HKK. Pediatric tracheotomies in an Asian population: the Singapore experience. Otolaryngol Head Neck Surg. 2005;133(2):246-50. http://dx.doi.org/10.1016/j.otohns.2005.03.085. PMid:16087023.
- Nassif C, Zielinski M, Francois M, Van Den Abbeele T. Tracheotomy in children: a series of 57 consecutive cases. Eur Ann Otorhinolaryngol Head Neck Dis. 2015;132(6):321-5. http://dx.doi.org/10.1016/j.anorl.2015.08.035. PMid:26409827.
- Mahadevan M, Barber C, Salkeld L, Douglas G, Mills N. Pediatric tracheotomy: 17 year review. Int J Pediatr Otorhinolaryngol. 2007;71(12):1829-35. http://dx.doi. org/10.1016/j.ijporl.2007.08.007. PMid:17953995.
- Ogilvie LN, Kozak JK, Chiu S, Adderley RJ, Kozak FK. Changes in pediatric tracheostomy 1982-2011: a Canadian tertiary children's hospital review. J Pediatr Surg. 2014;49(11):1549-53. http://dx.doi.org/10.1016/j.jpedsurg.2014.04.014. PMid:25475792.
- Alladi A, Rao S, Das K, Charles AR, D'Cruz AJ. Pediatric tracheostomy: a 13-year experience. Pediatr Surg Int. 2004;20(9):695-8. http://dx.doi.org/10.1007/s00383-004-1277-5. PMid:15449082.
- Sousa A, Nunes T, Roque Farinha R, Bandeira T. Traqueostomia: Indicações e complicações em doentes pediátricos. Rev Port Pneumol. 2009;15(2):227-39. http://dx.doi.org/10.1016/S0873-2159(15)30129-X. PMid:19280071.
- De Trey L, Niedermann E, Ghelfi D, Gerber A, Gysin C. Pediatric tracheotomy: a 30-year experience. J Pediatr Surg. 2013;48(7):1470-5. http://dx.doi.org/10.1016/j. jpedsurg.2012.09.066. PMid:23895956.
- Salcedo C, Martinez M, Reyes E. Pediatric tracheostomy: a ten-year analysis in the Intensive Care Unit of Sancti Spiritus Jose Marti Pediatric Teaching Hospital. Medwave. 2014;14(4):e5949. http://dx.doi.org/10.5867/medwave.2014.04.5949. PMid:25383685.
- Butnaru CS, Colreavy MP, Ayari S, Froehlich P. Tracheotomy in children: evolution in indications. Int J Pediatr Otorhinolaryngol. 2006;70(1):115-9. http://dx.doi. org/10.1016/j.ijporl.2005.05.028. PMid:16169607.
- DeMauro S, Akinbi H, Maschhoff K. Tracheotomy for the neonate and infant: indications, complications, timing and outcomes. In: Lioy J, Sobol S, editors. Disorders of the neonatal airway. New York: Springer; 2015. p. 263-79. http:// dx.doi.org/10.1007/978-1-4939-1610-8\_25.

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- Wootten CT, French LC, Thomas RG, Neblett WW 3rd, Werkhaven JA, Cofer SA. Tracheotomy in the first year of life: outcomes in term infants, the Vanderbilt experience. Otolaryngol Head Neck Surg. 2006;134(3):365-9. http://dx.doi. org/10.1016/j.otohns.2005.11.020. PMid:16500428.
- Carter P, Benjamin B. Ten-year review of pediatric tracheotomy. Ann Otol Rhinol Laryngol. 1983;92(4 Pt 1):398-400. http://dx.doi. org/10.1177/000348948309200422. PMid:6881848.
- Van Buren NC, Narasimhan ER, Curtis JL, Muntz HR, Meier JD. Pediatric tracheostomy: timing of the first tube change. Ann Otol Rhinol Laryngol. 2015;124(5):374-7. http://dx.doi.org/10.1177/0003489414560430. PMid:25432165.
- 21. Tchamo ME, Prista A, Leandro CG. Low birth weight, very low birth weight and extremely low birth weight in African children aged between 0 and 5 years old: a systematic review. J Dev Orig Health Dis. 2016;7(4):408-15. http://dx.doi. org/10.1017/S2040174416000131. PMid:27072315.
- Gray RF, Todd NW, Jacobs IN. Tracheostomy decannulation in children: approaches and techniques. Laryngoscope. 1998;108(1 Pt 1):8-12. http://dx.doi. org/10.1097/00005537-199801000-00002. PMid:9432059.
- Rogers J. Decannulation by external exploration of the tracheostome in children. J Laryngol Otol. 1980;94(5):563-7. http://dx.doi.org/10.1017/S002221510008926X. PMid:7400684.