Indications and clinical outcome in pediatric tracheostomy: Lessons learned

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Objective: Indications for tracheostomy have changed over the last decades and clinical outcome varies depending on the indication for tracheostomy. By gaining more insight in the characteristics and outcome of the tracheostomized pediatric population, clinical care can be improved and a better individual prognosis can be given. Therefore, we studied the outcome of our pediatric tracheostomy population in relation to the primary indication over the last 16 years.

Methods: We retrospectively included children younger than 18 years of age with a tracheostomy tube in the Erasmus Medical Center, Sophia children’s hospital. The primary indication for tracheostomy, gender, age at tracheostomy, age at decannulation, comorbidity, mortality, closure of a persisting tracheocutaneous fistula after decannulation, surgery prior to decannulation and the use of polysomnography were recorded and analyzed.

Results: Our research group consisted of 225 children. Reasons for a tracheostomy were first divided in two major diagnostic groups: 1) airway obstruction group (subgroups: laryngotracheal obstruction and craniofacial anomalies) and 2) pulmonary support group (subgroups: cardio-pulmonary diseases and neurological diseases). Children in the airway obstruction group were younger when receiving a tracheostomy (3.0 months vs. 31.0 months, p < 0.05), they were tracheostomy dependent for a longer time (median 21.5 months vs. 2.0 months, p < 0.05) and they required surgery more often (74.5% vs. 8.3%, p < 0.05) than the children in the pulmonary support group. The decannulation rate of children with a laryngotracheal obstruction is high (74.8%), but low in all other subgroups (craniofacial anomalies; 38.5%, cardio-pulmonary diseases; 34.6% and neurological diseases; 52.9%). Significantly more children (36.7%) died in the pulmonary support group due to underlying comorbidity, mainly in the cardio-pulmonary diseases subgroup. Surgery for a persisting tracheocutaneous fistula was performed in 34 (37.8%) children, with a significant relationship between the duration of the tracheostomy and the persistence of a tracheocutaneous fistula. No cannula related death occurred during this study period.

Conclusion: Main indications for a tracheostomy were airway obstruction and pulmonary support. Children in the airway obstruction group were younger when receiving a tracheostomy and they were tracheostomy dependent for a longer period. Within the airway obstruction group, the decannulation rate for children with laryngotracheal stenosis was high, but low for children with craniofacial anomalies. In the pulmonary support group, the decannulation rate was low and the mortality rate was high. Surgery for a persisting tracheocutaneous fistula was frequently needed.

1. Introduction

Indications for tracheostomy have changed over the last decades. The introduction of vaccines against Haemophilus Influenza and Corynebacterium Diphtheria has decreased severe morbidities of these infections, reducing the number of emergent tracheostomies. In contrast, improved management of critically ill children has led to an increase in survival, thereby increasing the number of children in need of prolonged ventilation and tracheostomies [1–3]. Nowadays the indications for a pediatric tracheostomy have shifted towards cardio-pulmonary and neurological diseases and towards (congenital or acquired) upper airway anomalies [2–4].

Pediatric tracheostomies are associated with serious complications ranging from 10% up to 58% [5] and overall mortality of up to 59% has been described, with a tracheostomy-related mortality up to 6% [4,6,7]. Moreover, the impact of a tracheostomy on caregivers is high, with a

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negative effect on quality of nearly all aspects of life. This makes early decannulation desired by both caregivers and physicians [8–11].

Before considering decannulation the overall health of the patient, medical comorbidities, functional status, and need for long-term pulmonary clearance have to be taken into account. Different decannulation protocols have been described. Most protocols include downsizing the cannula, placement of speaking valve or daytime capping, followed by in-hospital nighttime capping with monitoring and eventual decannulation. In addition, a polysomnography has shown to be a helpful additional tool in predicting successful decannulation outcomes [4,7,12–15].

The different indications for a tracheostomy, together with a variety of co-morbidities, results in a highly heterogenic pediatric tracheostomy population [5,7]. Counseling patients and parents on their expectation and prognosis of the tracheostomy is challenging and has to be made on individual basis. Outcome of pediatric tracheostomy in previous literature differs, depending on period of publication and the origin of the study [1,16–19]. By reviewing our 16 years’ experience, we present supplementary data regarding clinical characteristics and outcome of this heterogenic population. Our large sample size complements current literature, which will improve the insight in clinical care and provide a better individual prognosis.

2. Material and methods

2.1. Data

We retrospectively collected data from our electronic patient system of all children (younger than 18 years of age) with a tracheostomy tube in the Erasmus Medical Center-Sophia Children’s Hospital between January 2004 and December 2019.

The primary indication for tracheostomy was first divided in 2 major groups: the ‘airway obstruction’ group which was further subdivided in the subgroups ‘laryngotracheal obstruction’ and ‘craniofacial anomalies’ and in the ‘pulmonary support’ group, which was subdivided in the subgroups ‘cardio-pulmonary diseases’ and ‘neurological diseases’.

Medical records were reviewed and data on primary indication for tracheostomy, gender, age at tracheostomy, age at decannulation, comorbidities, mortality, closure of a persisting tracheo-cutaneous fistula after decannulation, surgery prior to decannulation, the use of polysomnography and capping the cannula were recorded.

Since this is a purely retrospective study, this study did not meet the requirements of a study that is subject to the Medical Research Involving Human Subjects Acts.

2.2. Statistical analysis

The Kruskal Wallis test, the Mann Whitney test and the Fisher (-Freeman-Halton) exact test were used to compare the characteristics of the different groups. A p-value of <0.05 was considered statistically significant. Patient baseline characteristics were described as median with range or as number and percentage. All statistical analyses were calculated using Statistical Package for Social Sciences (SPSS) version 25.0 for Windows (2017, SPSS Inc., Chicago, IL, USA).

3. Results

Two hundred and thirty-four children with a tracheostomy tube between January 2004 and December 2019 were eligible for inclusion. Nine patients were transferred to other hospitals, without further follow-up, therefore 225 children were included. Baseline characteristics of the study group are shown in Table 1. Comorbidity other than the primary indication for tracheostomy was present in 118 (52.4%) children and 61 (51.7%) of these children had two or more concurrent comorbidities.

Table 2 shows the different medical conditions which necessitated a tracheostomy in detail. Of the 225 children, 165 (73.3%) needed a tracheostomy in the airway obstruction group and 60 (26.7%) in the pulmonary support group. Eleven (32.4%) children in the neurological diseases subgroup needed a tracheostomy for recovery after a high-energy (head) trauma.

Data presented in number, number (percentage) or median (range).

Table 1
Baseline characteristics of the study group.

| Characteristic                        | Total (n = 225) |
|---------------------------------------|----------------|
| Male                                  | 133 (59.1%)    |
| Age at tracheostomy                  | 3 months (0–224) |
| Referred by other academic hospital  | 87 (38.7%)     |
| Comorbidity                           | 118 (52.4%)    |
| Pulmonary pathology                  | 64             |
| Congenital syndrome                  | 29             |
| Neurological or mental impairment     | 23             |
| Cardiac abnormalities                | 19             |
| Other                                 | 4              |

Data in parentheses range from 0 to 224.

Table 2
Indications for tracheotomy.

| Airway obstruction (n = 165; 73.3%) | Pulmonary support (n = 60; 26.7%) |
|------------------------------------|----------------------------------|
| Laryngotraheal obstruction         | Craniofacial anomalies           | Cardio-pulmonary diseases       | Neurological diseases            |
| (n = 139; 84.2%)                  | (n = 26; 15.8%)                 | (n = 26; 43.3%)                 | (n = 34; 56.7%)                 |
| Acquired                           | Acquired                         | Acquired                         | Acquired                         |
| laryngotraheal stenosis (n = 73; 52.5%) | Pierre Robin/retrognathia       | Pulmonary hypertension/lung hypoplasia | High-energy trauma             |
| Congenital                         | Treacher                         | 10 (38.5%)                      | 11 (32.4%)                      |
| laryngotraheal stenosis (n = 24; 17.3%) | Collins                          | Prematurity, BPD                 | Neuromuscular disease           |
| Vocal cord palsy/paralysis (n = 14; 10.1%) | Crouzon                        | 5 (19.2%)                       | 5 (14.7%)                       |
| Tracheomalacia (n = 12; 8.6%)      | Nager                            | Cardiomyopathy                   | Central respiratory dysfunction |
| Head/neck tumor (n = 8; 5.8%)      | Apert                            | 2 (7.7%)                        | 3 (8.8%)                        |
| Subglottic hemangioma (n = 3; 2.2%) | Saethre-                         | Extensive burn                   | Arnold-chiari malformation      |
| Battery erosion (n = 2; 1.4%)      | Chotzen                          | 2 (7.7%)                        | 2 (5.9%)                        |
| Neck abscess (n = 1; 0.7%)         | Bilateral                        | 1 (3.8%)                        | Tetraplegia                     |
| Dog bite (n = 1; 0.7%)             | Goldenhar                       | 1 (3.8%)                        | 2 (5.9%)                        |
| Carotis blow out due to myotic aneurysm (n = 1; 0.7%) |                          |                                  |                                  |

* Single median maxillary central incisor syndrome.
Table 3a
Clinical characteristics in relation to the main indication for tracheostomy.

| Airway obstruction (n = 165; 73.9%) | Pulmonary support (n = 60; 26.7%) | Total (n = 225) |
|-----------------------------|----------------------------------|-----------------|
| Boys                        |                                  |                 |
| Age at tracheostomy         | 3.0 months (0–216)               | 3.0 months (0–224) |
| Comorbidity                 | 92 (55.8%)                       | 26 (43.2%)      |
| Death                       | 6 (3.6%)                         | 22 (36.7%)      |
| Successful decannulated     | 114 (69.1%)                      | 27 (45.0%)      |

Data presented in number (percentage) or median (range).
#Significantly (p < 0.05) different when compared to the other group.
*Corrected for children that had immediate decannulation and surgical closure of the fistula after a laryngotracheal reconstruction or microscopic laryngeal surgery.

was significantly shorter, these were mostly patients recovering from a head trauma. Chronically ill neurological patients that were not decannulated, had a median age of 33.0 months (1–191 months).

Twenty-eight (12.4%) children died due to the underlying pathology or co-morbidity, there were no tracheostomy-related deaths. The mortality rate in the pulmonary support group was higher when compared to the airway obstruction group, mainly in the cardio-pulmonary diseases subgroup.

In 141 (62.7%) children decannulation was successful. Children in the laryngotracheal obstruction subgroup had a significantly higher rate of decannulation (74.8%) when compared to the other subgroups, who had decannulation rates between 34.6% and 52.9%.

The laryngotracheal obstruction subgroup also showed the highest rate of decannulation after surgery; 87.9% of the children were successfully decannulated, especially when a reconstruction for an acquired laryngotracheal stenosis was performed (93.7%). In the craniofacial anomalies subgroup only 56.3% of the children were decannulated despite multiple surgeries (61.5% of the children). In the pulmonary support group only in 8.3% surgery was done to correct upper airway obstructions mainly for a suprastomal granuloma or removal of granulation tissue.

Table 4 shows the steps taken prior to decannulation in relation to the primary indication of the tracheostomy. Fifty-six (39.7%) children were decannulated by capping the cannula followed by polysomnography, 51 (36.2%) children were decannulated directly after a laryngotracheal reconstruction or microscopic laryngeal surgery with immediate surgical closure of the fistula and 30 (21.3%) children were decannulated by only capping the cannula. In 2 children (1.4%) an accidental but successful decannulation occurred. In 33 (23.4%) of the decannulated children, an endoscopy was not performed prior to decannulation, these were mainly children from the neurological diseases subgroup.

Thirty-four (37.8%) of the 90 children who did not have primary closure of the tracheostomy had a persistent tracheocutaneous fistula for which a surgical closure was necessary. There was a significant relationship between the duration of the tracheostomy and the persistence of a tracheocutaneous fistula after decannulation; the longer the tracheostomy was in situ, the larger the risk of a persisting tracheocutaneous fistula.

Table 5 shows the reasons for failed decannulation in relation to the primary indication for tracheostomy. Overall, the main reason for failure in the airway obstruction group was multiple level obstruction (45.1%). In the pulmonary support group, death was the main reason for failure (66.7%). Two patients of the ‘airway obstruction group’ could not be decannulated as a result of persisting nocturnal ventilation and one patient as a result of persisting neurological impairment. The two patients had persisting night time ventilation because of obstructive sleep apnea. The persisting neurological impairment of the other patient is explained by swallowing problems and aspiration after airway surgery.

In 67 children a capped polysomnography was performed prior to decannulation. Eighteen children were not decannulated because of respiratory abnormalities seen on the capped polysomnography. Additional surgery was done in 5 children, followed by a successful decannulation. In 2 children decannulation was done one year after the initial polysomnography, without any additional treatment. Eleven children remained tracheostomy dependent; 6 children due to higher multi-level obstruction, 3 children are scheduled for surgery or our decannulation protocol again and 2 children due to persisting aspiration.

Of note, despite a successful capping trial and adequate polysomnography results, 2 children presented a few hours after decannulation with acute airway problems due to a suprastomal granuloma in 1 and higher multi-level collars in the other child. One child was successful decannulated after removal of the granuloma, the other child was re cannulated and additional surgery has not led to decannulation yet.

Table 3b
Clinical characteristics in relation to the subgroups of indication for tracheostomy.

| Airway obstruction | Craniofacial anomalies (n = 26; 11.6%) | Pulmonary support |
|--------------------|---------------------------------------|-------------------|
| Laryngotracheal obstruction (n = 139; 61.8%) | 14 (53.8%) | 16 (61.5%) |
| Age at tracheostomy | 0.5 months (0–20) | 4.5 months (0–167) |
| Comorbidity         | 10 (38.5%) | 19 (73.1%) |
| Death               | 1 (3.8%) | 17 (65.4%) |
| Successful decannulated (with/without surgery) | 10 (38.5%) | 9 (34.6%) |

Data presented in number (percentage) or median (range).
#Significantly (p < 0.05) different when compared to the other group.
*Corrected for children that had immediate decannulation and surgical closure of the fistula after a laryngotracheal reconstruction or microscopic laryngeal surgery.
higher multi-level airway obstruction. These results are in agreement with current literature that shows that complex congenital craniofacial anomalies pose a great challenge for clinicians which is reflected in lower decannulation rates [21,22].

The oldest group of children who received a tracheostomy were children in the neurological diseases subgroup and the time the tracheostomy tube remained was significantly shorter for these patients. The decannulated children of this group were mainly post-traumatic patients. The median age and period of tracheostomy is consistent with literature [1,8] and can be explained by the fact that many children (32.4%) received a temporary tracheostomy after a high-energy head trauma during the recovery phase. On the other hand, chronically ill children in the neurological diseases and the cardio-pulmonary diseases subgroups are less likely to be decannulated. This low decannulation rate of 45.0% in the pulmonary support group is explained by the need for persistent nocturnal ventilation, persisting neurological or mental impairment and also because a relatively large number (36.7%) of children died in this group.

As a result of underlying pathology and comorbidity, the mortality rate in the pulmonary support group was high, especially in the cardio-pulmonary diseases subgroup. In contrast, the mortality rate in the airway obstruction group was only 3.6%. In a literature review on tracheostomy related mortality, Dal’Astra et al [6] showed that reported overall mortality ranges from 2.2% to 59.0%. Most of these deaths were associated with underlying comorbidity and not the tracheostomy itself, especially among children under one year of age, as well as premature or extremely underweight newborns.

Before decannulation a laryngo-, trachea-, bronchoscopy with a rigid and flexible endoscopy under general anesthetic is recommended to confirm the presence of a patent airway at all levels, at least one mobile vocal fold, and to rule out any occluding suprastomal granuloma or malacia [10,23]. Waddell et al. [24] found a better outcome in decannulation when there was a shorter time between the final endoscopy and the trial of decannulation. In our study, an endoscopy prior to decannulation was not performed in 33 (23.4%) of our children, mainly children from the neurological diseases subgroup. Two children needed a medical replacement of the cannula a couple of hours after initial decannulation (despite a normal polysomnography). A suprastomal granuloma) because of upper airway collapse and to a suprastomal granuloma. This could have been prevented with an endoscopy performed just prior to decannulation. This finding supports the advice to perform an airway endoscopy shortly prior to decannulation.

As for the use of polysomnography, this procedure has shown to be a useful tool in predicting successful decannulation outcomes, especially in young children or children with complex airway problems [4,7,12–15]. We also found that a polysomnography is helpful in deciding whether to decannulate, especially in the airway obstruction group. In our study, polysomnography showed abnormalities in 18 of the 67 children (mostly children from the airway obstruction group). Decannulation was postponed for 7 children while 11 children remain tracheostomy dependent as a consequence of multi-level obstruction.

Our decannulation procedure varied among the different indication

### Table 4

| Airway obstruction                  | Pulmonary support                  | Total (n = 141) |
|------------------------------------|------------------------------------|----------------|
| Laryngotracheal obstruction        | Cardio-pulmonary diseases          | 56 (39.7%)     |
| (n = 104)                          | Neurological diseases               |                |
| Craniofacial anomalies             |                                    |                |
| (n = 10)                           |                                    |                |
| Capping the cannula and polysomnography |                                    |                |
| 39 (37.5%)                        | 2 (22.2%)                          |                |
| Surgery with immediate decannulation |                                    |                |
| 49 (47.1%)                        | 1 (11.1%)                          |                |
| Only capping the cannula           |                                    |                |
| 14 (13.5%)                        | 5 (55.6%)                          |                |
| Other                              |                                    |                |
| 2 (1.9%)                          | 1 (11.1%)                          |                |
| 2x no information                 |                                    |                |
|                                    | 9 (50.0%)                          |                |
|                                    | 1 (5.6%)                           |                |
|                                    |                                    | 30 (21.3%)     |

Data presented in number (percentage).

### Discussion

We describe the indications, the clinical characteristics and outcome of children with a tracheostomy in relation to two main indications: airway obstruction and pulmonary support. We found that children within the airway obstruction group were younger when receiving a tracheostomy, they require surgery more often and were tracheostomy-dependent for a longer time. The decannulation rate for children in the laryngotracheal obstruction subgroup was high, but low for all other subgroups. As a result of underlying morbidity, mortality in the pulmonary support group was high, especially in the cardio-pulmonary subgroup. Children with a tracheostomy in the neurological diseases subgroup were significantly older and they were tracheostomy-dependent for a shorter period of time.

By dividing the groups in upper airway obstruction and pulmonary support, we distinguished between the need for a tracheostomy in children with a soft tissue obstruction or a structural framework anomaly, as opposed to children who were dependent on chronic ventilation or had pulmonary clearance problems. The most common indication for a pediatric tracheostomy in our study was the management of upper airway obstruction (73.3% of the children), in almost a quarter of the children this was support of pulmonary function. Recent studies describe a shift from emergent tracheostomies because of infectious processes towards tracheostomies in children with neurological diseases, cardio-pulmonary diseases and (congenital or acquired) airway obstruction [1,2,4,8,20]. Compared to these studies, the number of tracheostomies performed for upper airway obstruction in our study is relatively high, which is explained by the fact that our hospital is a national referral center for airway anomalies and rare craniofacial anomalies; almost 40% of patients with these anomalies were referred by other tertiary referral centers in the country.

The youngest group of children who received a tracheostomy were children in the craniofacial anomalies subgroup, who had severe structural anatomical abnormalities of the upper airway necessitating tracheostomy early in life. Together with the children in the laryngotracheal obstruction subgroup, the tracheostomy dependency in these groups is much longer than in the pulmonary support group. Since surgery of the airway is usually performed when a child has grown older, these children must retain the tracheostomy for a longer time.

To achieve decannulation, the majority of the children (77.0%) in the airway obstruction group needed one or multiple surgeries, whereas only 8.3% of the children in pulmonary support group needed ‘minor’ surgery to correct an upper airway obstruction (removal of granulation tissue or a suprastomal granuloma). After surgery, children in the laryngotracheal obstruction subgroup had the highest likelihood of decannulation (87.9%), in particular those who had undergone surgical reconstruction for an acquired laryngotraheal stenosis (93.7%). This is in contrast with children of the craniofacial anomalies subgroup. Almost 62% of these children underwent multiple surgeries, which led to a disappointing decannulation rate of 56.3%, mainly due to persisting higher multi-level airway obstruction. These results are in agreement...
groups. In 36.2% of the children, mostly from the laryngotracheal obstruction group, the tracheostomy was surgically closed during a laryngotracheal reconstruction or microscopic laryngeal surgery. In 39.7% of the children a procedure was followed with downsizing the cannula, placement of speaking valve, capping of the cannula, followed by an in-hospital polysomnography with a capped cannula and decannulation. Children from the craniofacial anomalies subgroup have mainly been decannulated following this protocol. Finally, 21.3% of the children only had a capped cannula without a polysomnography, most of whom were children from the pulmonary support group; a polysomnography was waived because no preexisting upper airway anomalies were present.

In 34 children (37.8%) a tracheocutaneous fistula persisted after decannulation and surgical closure was necessary. A longer duration of tracheostomy was associated with a higher risk on a persistent fistula. This is in agreement with literature, were rates between 3% and 48% are reported [4, 5, 7, 25].

We acknowledge the limitations of this study. The data was extracted from the hospital records retrospectively, carrying the risk of incomplete data. Our study group was a heterogeneous group of children with various indications for tracheostomy and comorbidities; therefore, it is difficult to draw definite conclusions. Nevertheless, despite these limitations we feel that the large sample size of our study, including a large group of children with airway anomalies, provides more insight into clinical characteristics and prognosis in the two main groups of children that have a tracheostomy either for airway obstruction or for pulmonary support.

5. Conclusion

This study demonstrates the clinical outcome of tracheostomies in children in relation to two main indications; airway obstruction and pulmonary support. The mortality rate is high in the pulmonary support group due to underlying comorbidity. Children in the airway obstruction group are tracheostomy dependent for a longer time and they require surgery more often. The decannulation rate for children with laryngotracheal stenosis is high, but low for children with craniofacial anomalies as a consequence of multiple levels of obstruction. Polysomnography is a helpful tool for deciding whether to decannulate, and endoscopy prior to decannulation is recommended, especially in the airway obstruction group. Surgery for a persisting tracheocutaneous fistula is frequently needed.

5.1. Lessons learned

- Children with a tracheostomy as a result of airway obstruction are younger when receiving a tracheostomy, are tracheostomy dependent for a longer time and require surgery more often; the decannulation rate for children with a laryngotracheal obstruction is high, but low for children with craniofacial anomalies.
- Mortality as a result of underlying comorbidity is high in children with a tracheostomy for pulmonary support, especially in the cardiopulmonary diseases subgroup.
- Children with a tracheostomy due to temporary neurological diseases are significantly older and they are tracheostomy-dependent for a shorter time, especially after a high-energy head trauma.
- Chronically ill children in the neurological disease and the cardiopulmonary disease group are less likely to be decannulated.
- An airway endoscopy performed just prior to decannulation is recommended.
- A polysomnography is a helpful tool in determining decannulation outcome, mainly in children with multi-level obstruction.
- Surgery for a persisting tracheocutaneous fistula is frequently needed.
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