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**MAIN ARTICLE**

# Tracheostomy in neurologically compromised paediatric patients: role of starplasty

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## Abstract

*Objectives*: Starplasty tracheostomy is an alternative to traditional tracheostomy. This paper reviews neurologically compromised paediatric patients with tracheostomies and discusses the role of starplasty tracheostomy.

*Method*: A retrospective review was conducted of paediatric patients with a neurological disorder who underwent tracheostomy between 1997 and 2011.

*Results*: Forty-eight patients, with an average age of 7.3 years, were identified. The most common indications for tracheostomy were: ventilator dependence (39.6 per cent), an inability to tolerate secretions or recurrent aspiration pneumonia (33.3 per cent), and upper respiratory obstruction or hypotonia (12.5 per cent). The most common underlying neurological diagnosis was cerebral palsy. There were no early complications. Eighteen (43 per cent) of 42 patients with follow up experienced at least 1 delayed complication. Only 12 patients (28.6 per cent) were decannulated.

*Conclusion*: Patients with primary neurological diagnoses have low rates of decannulation; starplasty tracheostomy should be considered for these patients. Patients with seizure disorder or acute neurological injury tended to have a higher short-term decannulation rate; traditional tracheostomy is recommended in these patients.

**Key words:** Pediatrics; Tracheostomy; Tracheostomy; Nervous System Diseases; Central Nervous System Diseases; Neurologic Manifestations

## Introduction

In paediatric patients, tracheostomy is indicated when: airway obstruction necessitates bypass, access is required for prolonged mechanical ventilation, or airway protection or improved tracheobronchial toilet is required. There are a number of classification systems for tracheostomy indications described in the literature. In general, cases can be divided into five groups: upper airway obstruction, trauma and its sequelae, craniofacial abnormalities, long-term ventilation requirement, and a neuromuscular deficit. Furthermore, airway obstruction can be congenital (e.g. bilateral vocal fold paralysis), acquired (e.g. neoplastic lesions) or infectious (e.g. epiglottitis).

Over the past 40 years, there has been a decrease in tracheostomies performed for infectious causes, associated with the use of vaccines against *Haemophilus influenzae* and *Corynebacterium diphtheriae*.[1] In modern neonatal intensive care units, long-term intubation is now an acceptable alternative. In contrast, the incidence of tracheostomy for chronic medical problems has increased, as paediatric and neonatal diagnosis and treatments improve and children with disabilities survive longer.[2]

Starplasty is a technique based on the geometry of three-dimensional Z-plasty. It consists of generating triangular flaps in the skin from an X-shaped incision, followed by the removal of subcutaneous fat and midline division of the underlying strap muscles. A '+' (plus symbol) shaped incision is then created in the anterior tracheal wall, and the skin and tracheal flaps are then interdigitated and sewn together using a mattress stitch.

The ideal tracheostomy technique possesses the following qualities: technical ease, separation of the wound from tracheal secretions and airflow, postoperative replaceability, rapid maturity, minimal stomal maintenance, minimal tracheal deformation, reversibility, and minimal scarring.[3] Starplasty has been shown to meet these criteria, and in some cases

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it is superior to traditional tracheostomy. The major drawback is that of an increased chance of persistent tracheocutaneous fistula following decannulation. Starplasty has been recommended over traditional tracheostomy for patients with likely long-term tracheostomy requirements or when decannulation is not anticipated.[4]

To date, many studies have analysed the indications, complications and outcomes associated with paediatric tracheostomy. However, in the neurological subgroup, the disease-specific indications for tracheostomy have not been well described. In addition, data on the duration of tracheostomy, often necessary for this subset of patients, and the long-term complications seen with prolonged tracheostomy, are limited. This study analysed the indications and type of tracheostomy performed, outcomes, need for revision surgery, bronchoscopic findings, and complications in a specific subgroup of paediatric patients with a tracheostomy because of a neurological diagnosis.

## Materials and methods

Institutional review board approval to conduct a retrospective chart review was obtained. The electronic medical records of all patients younger than 18 years who underwent tracheostomy at the Cleveland Clinic from 1997 to 2011 were reviewed. All patients with a documented neurological diagnosis were included. Within this subset, the specific neurological diagnoses, indication for tracheostomy, surgical technique, complications, duration of tracheostomy, endoscopic findings and post-operative chest radiograph findings were evaluated.

## Results

Forty-eight patients who met the study criteria were identified. There were 24 male and 24 female patients, with a mean age of 7.3 years. Six patients were lost to follow up and there were no records to document the duration of tracheostomy; these patients were eliminated from the statistical analysis of late complications and rate of decannulation.

Eighteen patients (42.8 per cent) had cerebral palsy, which was the most common underlying neurological diagnosis. The remaining patients underwent tracheostomy for: intractable seizures (6 patients, 14 per cent), brain tumours (5 patients, 11.9 per cent), mitochondrial myopathy (3 patients, 7 per cent), anoxic encephalopathy (3 patients, 7 per cent), neurofibromatosis (2 patients, 4.7 per cent), leukodystrophy (2 patients, 4.7 per cent) and other diagnoses (9 patients, 21 per cent) (Table I).

The indications for tracheostomy were: ventilator dependence (39.6 per cent), inability to tolerate secretions or recurrent aspiration pneumonia (33.3 per cent), upper respiratory obstruction or hypotonia (12.5 per cent), vocal fold paralysis or laryngeal stenosis (10.4 per cent), and intractable seizures (4.2 per cent). The average duration of tracheostomy for patients still cannulated at the time of this study was 7.6 years. Starplasty tracheostomy was utilised in 75 per cent of patients and the remainder underwent traditional tracheostomy. The average follow-up period was 6.9 years.

TABLE I
CHARACTERISTICS OF PATIENTS WITH NON-CLASSIFIABLE NEUROLOGICAL DIAGNOSES

| Age at tracheostomy | Gender | Neurological diagnosis |
|---|---|---|
| 1 year | M | Intracranial tuberculosis |
| 17 years | M | Traumatic subdural haematoma |
| 2 years | F | Myelomeningocele & Arnold–Chiari malformation |
| 5 years | F | Septo-optic dysplasia |
| 9 months | M | Pompe disease |
| 1 year | F | Hydrocephalus |
| 8 years | F | Pontine cavernoma, seizures |
| 2 years | M | Tuberous sclerosis, seizures |
| 1 year | F | GM1 galactosidase deficiency with neurological decline, seizures |

M = male; F = female

Only 12 (28.6 per cent) of the patients with long-term follow up were decannulated. The average duration of tracheostomy in this group was 2.7 years. Of the 12 patients that were decannulated, 3 had undergone traditional tracheostomy and 9 had undergone starplasty tracheostomy. None of the traditional tracheostomy patients and five (56 per cent) of the starplasty tracheostomy patients developed persistent tracheocutaneous fistulas ($p = 0.20$). Six of the 12 patients were decannulated within 18 months of their original tracheostomy. The underlying diagnoses of these patients, which were acute or temporary neurological morbidities, are outlined in Table II.

Of the 36 patients that underwent starplasty tracheostomy, there were no documented early complications such as pneumothorax or haemorrhage. There were 33 starplasty tracheostomy patients with long-term follow-up data; 16 of these patients (48 per cent) experienced delayed complications. Complications included: the development of suprastomal granulation tissue, in 11 patients; persistent tracheocutaneous fistula, in 5 patients (Table III); and tracheal or subglottic scarring, in 3 patients. Of the 12 patients that underwent traditional tracheostomy, there were no early complications. Two traditional tracheostomy patients (22 per cent) had the late complication of suprastomal granulation development ($p = 0.05$).

Results from at least 1 laryngoscopy were available for 14 patients (29 per cent) and results from at least 1 bronchoscopy were available for 18 patients (37.5 per cent). Laryngoscopy findings were normal in 10 patients; vocal fold immobility was evident in 1 patient, mild laryngomalacia in 1 patient and paradoxical vocal fold movement in 1 patient. One patient was noted to have posterior glottic stenosis, which was the original indication for tracheostomy. Two patients were

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TABLE II
CHARACTERISTICS OF PATIENTS WITH TRACHEOSTOMY OF LESS THAN 18 MONTHS' DURATION

| Diagnosis | Indication for tracheostomy | Age at tracheostomy (years) | Tracheostomy duration (months) |
|---|---|---|---|
| Intractable seizures, brain malformation | Seizures | 12 | 13 |
| Intracranial tuberculosis | Ventilator dependence | 1 | 5 |
| NF type 1, macroglossia | Upper airway obstruction – OSA | 2 | 5 |
| Ependymoma | Ventilator dependence | 4 | 4 |
| Status epilepticus | Ventilator dependence | 17 | 3 |
| Seizures, developmental delay | Inability to manage secretions | 2 | 6 |

NF = neurofibromatosis; OSA = obstructive sleep apnoea

found to have mild tracheomalacia on bronchoscopy. Thirty-nine patients (81.3 per cent) underwent post-operative chest radiography; no post-operative complications were detected on the radiographs.

## Discussion

In our study, patients with a primary neurological diagnosis had low rates of decannulation and a prolonged duration of tracheostomy. Thirty of the 42 patients with follow-up data (71 per cent) had remained tracheostomy-dependent throughout, until the time of the study. These findings support the low rates in decannulation and the prolonged cannulation duration previously reported in patients with neurological indications.[5]

In our study, starplasty tracheostomy was shown to be safe and efficacious in these patients who need prolonged tracheostomy. Persistent tracheocutaneous fistula, however, is the primary disadvantage of the technique. Fifty-six per cent of our patients who underwent starplasty tracheostomy and were decannulated went on to develop a persistent fistula. This rate is similar to that previously described, of 52 per cent.[6] In comparison, none of the patients in our study who underwent traditional tracheostomy developed a persistent tracheocutaneous fistula. In the literature, the incidence of tracheocutaneous fistula in patients who were decannulated after traditional tracheostomy is as low as 10–30 per cent.[5,7]

As the rate of persistent fistula is comparatively lower and there are fewer long-term complications with traditional tracheostomy, if there is high likelihood of decannulation within a short time interval, traditional tracheostomy should be considered over starplasty tracheostomy. In our study, patients requiring tracheostomy for seizure disorders or acute intracranial injury had high rates of decannulation within less than 18 months of placement, and therefore patients with these diagnoses should be considered for traditional tracheostomy. The remaining patients who were eventually decannulated had cerebral palsy or anoxic brain injury, and required their tracheostomies for more than two years. Such patients are not likely to benefit from traditional tracheostomy over starplasty tracheostomy because of the long duration of tracheostomy dependence.

Endoscopic examination is recommended for paediatric patients undergoing tracheostomy for evaluation of the airway for any lesions contributing to respiratory failure. In our study, findings included mild tracheomalacia, paradoxical vocal fold movement, mild laryngomalacia and unilateral vocal fold immobility. Although the lesions identified in this study were not the only reasons for tracheostomy, they may have contributed to continued dependence.

Although not the main intent of this study, the utility of post-operative chest radiography in our patient population was also evaluated. Studies on adult patients have demonstrated low rates of complications detectable by chest X-ray following tracheostomy, ranging from 2 to 3 per cent, and the complications had already been detected on physical examination before a chest radiograph was obtained.[8] Many studies have therefore concluded that routine chest radiography after tracheostomy in adults is unnecessary. However,

TABLE III
CHARACTERISTICS OF PATIENTS WITH PERSISTENT TRACHEOCUTANEOUS FISTULA AFTER DECANNULATION

| Diagnosis | Tracheostomy technique | Indications for tracheostomy | Age at tracheostomy (years) | Tracheostomy duration |
|---|---|---|---|---|
| Intractable seizures, brain malformation | Starplasty | Intractable seizures – inability to wean from ventilator | 12 | 1 year 1 month |
| Hydrocephalus, post-cardiac surgery, tetralogy of Fallot | Starplasty | Prolonged need for intubation due to respiratory distress | 1 | 6 years |
| Seizures, developmental delay | Starplasty | Ventilator dependency, inability to manage secretions | 2 | 6 months |
| Status epilepticus | Starplasty | Failure to wean from ventilator | 17 | 3 months |
| Intracranial tuberculosis | Starplasty | Inability to wean from ventilator | 1 | 5 months |

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the data on the paediatric population are more limited. In a study of 420 children, published in 1982, Wetmore *et al*. reported an overall complication rate of 49 per cent and a tracheostomy-related mortality rate of 2 per cent.[9] Based upon these high complication rates, many centres today routinely obtain post-operative chest radiographs for children after tracheostomy. In our own study, 39 patients underwent post-operative chest radiography. Of these, none were found to have a major complication such as pneumothorax or pneumomediastinum. This does question the need for routine chest radiography in all cases of paediatric tracheostomies. However, further studies are required to elucidate the need for post-operative radiography.

- **Starplasty tracheostomy consists of interdigitated skin and anterior tracheal wall flaps**
- **Starplasty is efficacious, with less chance of accidental decannulation; however, the chance of persistent tracheocutaneous fistula following decannulation is increased**
- **This study focused on tracheostomy in neurologically compromised paediatric patients**
- **Common indications for tracheostomy were ventilator dependence, secretion intolerance or recurrent aspiration pneumonia, and upper respiratory obstruction or hypotonia**
- **Patients with neurological diagnoses have lower decannulation rates than patients with seizure disorder or acute neurological injury**
- **When long-term tracheostomy is anticipated, starplasty tracheostomy should be considered**

Limitations of our study include the inherent reporting bias present with a retrospective analysis. In addition, a few patients had multiple indications for tracheostomy, and there was likely a degree of subjectivity in deciding which indication was the primary one for classification purposes. Lastly, the study is limited by the number of patients as a result of exclusively using patients with neurological diagnoses.

## Conclusion

Starplasty tracheostomy is our preferred technique in most paediatric patients with a primary neurological diagnosis, given the likelihood of prolonged dependence and low rates of decannulation. Exceptions would be in patients with a seizure disorder or acute intracranial injury who are expected to have high rates of decannulation within a short time interval of less than 18 months. Routine chest radiography may not be necessary in uncomplicated cases.

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