

# Slide Tracheoplasty for Severe Congenital Long Segment Tracheal Stenosis in Infants : Surgical and Anesthetic Management

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## Abstract

Congenital long segment tracheal stenosis is the rare occurrence of an intrinsic narrowing of the trachea due to a complete cartilaginous ring. It is difficult to manage and can be life threatening especially when these patients who are usually neonates or infants have the pathological pattern of a long segment and have to come for surgical correction. Despite many technical reports on how to correct this anomaly, currently, the technique of "slide tracheoplasty" is claimed to be the most successful with the good immediate and long-term outcomes. But because of the rare and life threatening disease, so we were encouraged and write this report about the disease and its management.

These are the case series reports of 4 infants with a history and diagnosis of severe long segment congenital tracheal stenosis who needed a definite surgical repair. The authors decided to use the surgical technique of "slide tracheoplasty" with successful outcome. In two of the cases, patients needed cardiopulmonary bypass support during the surgical repair. All of these patients did well after the operation except one patient with a history of congenital heart disease (tetralogy of Fallot) who needed an emergency surgical repair and was reoperated upon with pericardial patch. In this reports the authors did not find any benefit from tracheostomy. Also, details of surgical and anesthetic procedure were discussed with the conclusion that the surgical technique of slide tracheoplasty should be the surgical of choice for the management of congenital long segment tracheal stenosis.

**Key word :** Long Segment Tracheal Stenosis, Anesthesia, Surgery, Slide Tracheoplasty

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Congenital tracheal stenosis is the rare occurrence of an intrinsic narrowing of the trachea due to a complete cartilaginous ring. It is difficult to manage and can be life threatening. It was generally fatal before the early 1980s<sup>(1)</sup>. This anomaly can be found as a single lesion or associated with others anomalies especially anomalies of the cardiovascular system<sup>(2)</sup>. The primary symptoms are the manifestation of a large airway obstruction including: inspiratory-expiratory stridor, recurrent pneumonia, dyspnea and cyanosis. The onset may start at birth or a few months later. Because of the narrowed diameter of the infant trachea and the long segmental involvement, repair is made more difficult and dangerous.

Several treatments of congenital tracheal stenosis have been described including a conservative treatment<sup>(3)</sup>, resection with end to end anastomosis<sup>(4)</sup>, various type of surgical tracheoplasty including the technique of sliding tracheoplasty<sup>(5-11)</sup>, which was claimed to be one of the most successful treatment with excellent immediate and late outcome<sup>(12)</sup> and endoscopic balloon tracheoplasty<sup>(13,14)</sup>. Because of the rare occurrence of this anomaly more information of the treatment and outcome is still needed despite the reported success of the current treatment.

The purpose of these case series report is to describe our experience in the treatment of severe congenital long segment tracheal stenosis in four infants, including the diagnosis, investigation, surgical and anesthetic techniques.

## PATIENTS

### Case 1

A 6-month-old girl who was referred from another tertiary medical center after a diagnosis of long segment congenital tracheal stenosis had been made. She had a history of inspiratory-expiratory stridor since birth but no dyspnea or cyanosis. She then presented with viral croup that progressed to severe dyspnea and respiratory failure needing positive pressure ventilation. The CT scan showed a long segment tracheal stenosis of the lower trachea with early bifurcation of the right upper lobe bronchus from carina (Fig. 1 and 2). Echocardiogram could not find any abnormalities.

### Case 2

A 3-month-old girl with a diagnosis of tetralogy of Fallot and congenital tracheal stenosis which were made during her post natal period. She was discharged from the hospital with a conservative plan.



**Fig. 1.** CT scan of the first patient showed long stenotic segments of lower trachea with early bifurcated of right upper lobe bronchus from carina.

She came back to our hospital in a serious condition of large airway obstruction after suffering from upper respiratory tract infection for a few days. Because of failure to assist her ventilation even with a very high positive pressure ventilation, she underwent the immediate emergency operation after the flexible bronchoscopic examination confirmed her diagnosis. With the severe hypoxemia and hypercarbia and unsuccessful ventilatory support despite the high peak airway pressure of 60-70 cmH<sub>2</sub>O. This patient was put on cardiopulmonary bypass right after her arrival in the operating room.

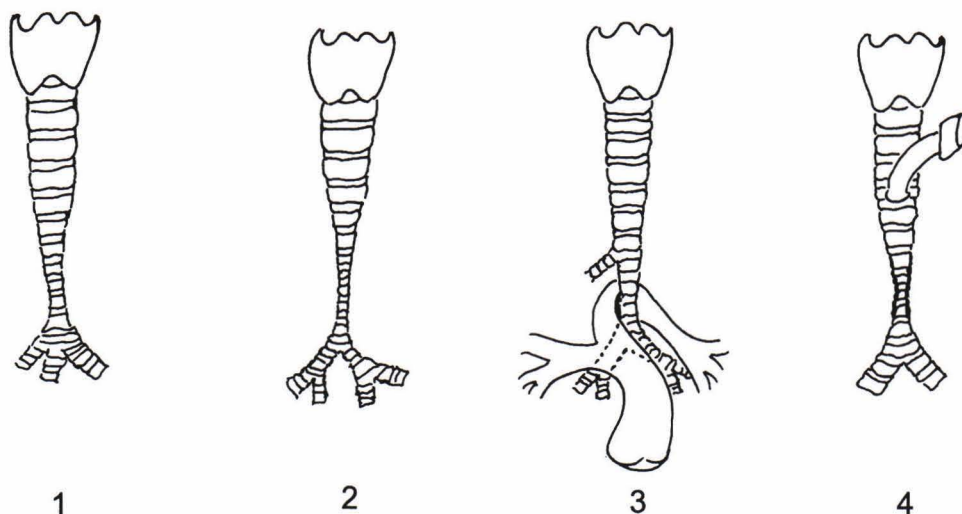
### Case 3

A 1-year-old girl who had a history of recurrent pulmonary infection with noisy respiration since 5 months old. Her last attack caused respiratory failure and needed intubation with positive pressure ventilation. CT scan revealed long segment congenital tracheal stenosis with pulmonary artery sling. Others investigations including flexible bronchoscopic examination, echocardiography and spiral CT scan of the air way which showed an aberrant left pulmonary artery curved behind the narrowing segment of trachea and below the right upper lobe bronchus that separated at a high level of the trachea (Fig. 2).

### Case 4

A one and a half years old boy who was a known case of Kippel-feil syndrome. The physical





**Fig. 2.** The characteristic of trachea anomalies in our patients. Patient 1, stenosis of the lower trachea with anomalous of right upper lobe bronchus. Patient 2, tetralogy of Fallot, stenosis of lower trachea and main stem bronchi. Patient 3, stenosis of lower trachea with pulmonary artery sling. Patient 4, stenosis of lower trachea with intraluminal granulation from MRSA infection.

signs of airway obstruction occurred after extubation on day 7 postoperatively following plication of the left hemidiaphragm for diaphragmatic eventration. The bronchoscopy and CT scan revealed long segment tracheal stenosis with granulation tissue at the lower trachea. The boy was referred to our hospital with tracheostomy tube and respiratory failure requiring positive pressure ventilation. On his arrival, we repeated bronchoscopy and took tissue culture from the lesion. The culture grew a heavy growth of MRSA. The operation was deferred until this organism was clear from his airway.

### Operative and anesthetic techniques

The operations were performed under general anesthesia and carefully controlled ventilation with the technique of slow respiratory rate to allow the air to pass through the obstructive lesion. Anesthetic technique was planned according to the lesion and the condition of each patient. Two patients (case 1 and case 4) who had isolated tracheal stenosis without associated cardiovascular anomalies were operated on without cardiopulmonary bypass. Ventilation during tracheoplasty was maintained by insertion of a sterile endotracheal tube (no. 3.0) into the distal trachea then



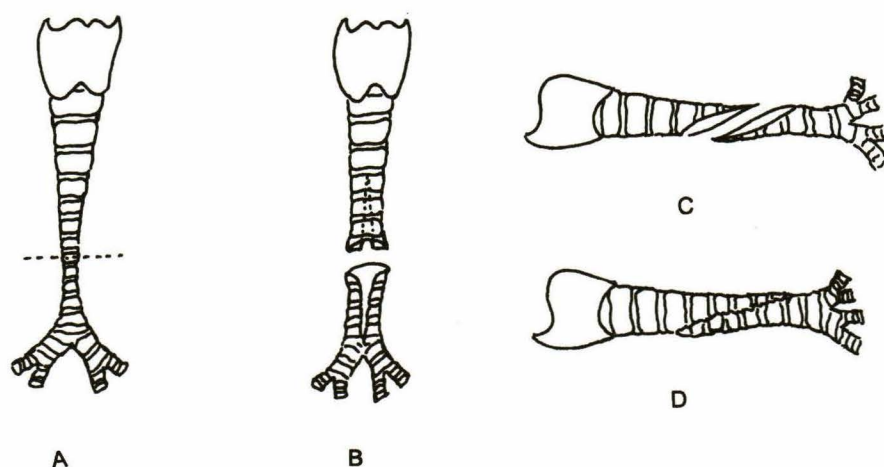
**Fig. 3.** Complete cartilaginous ring of upper part of trachea (arrow). Endotracheal tube in the lower trachea (double arrow) (patient 1).

inserted into the left main bronchus. This was frequently adjusted and fixed in position with great care and connected across the operative field (with the new sterile anesthetic circuit) (Fig. 3). The other two

patients who had associated cardiovascular anomalies (case 2 with tetralogy of Fallot and case 3 with pulmonary artery sling) were put on assisting cardiopulmonary bypass. This technique permitted the simultaneous correction of cardiovascular anomalies.

The patients were positioned with cervical extension. The incision of the first patient was a cervical incision with a minimal upper median sternotomy extension. The other three patients needed median sternotomy incisions. The anterior surface of trachea was exposed as long as possible to access the extent of the lesion. The midpoint of the lesion was dissected circumferentially and encircled (Fig. 4 and 5). An anchoring suture at both sides of the carina was performed for traction. The midpoint of the lesion was divided transversely (Fig. 3 and 4), then the anterior surface of the lower trachea was cut longitudinally to increase the luminal diameter. The sterile endotracheal tube was inserted into this new opening and adjusted until the appropriate ventilation was achieved. The proximal end of the trachea was dissected upwards. To minimize disturbing blood supply, the circumferential dissection should be kept as short as possible. But the posterior dissection might be high up to the normal trachea in order to

provide enough space for posterior splitting of all of the complete tracheal rings (Fig. 4, 6 and 7). The distal trachea needed only minimal mobilization because the vertical splitting was on the anterior surface. It is important to split all of the stenotic lesion that sometimes extends over the carina to the main bronchus. With cervical flexion, the proximal and distal tracheal flaps were slide over each other and sutured together with multiple interrupted 6-0 polydioxanone stiches (knot outside the lumen), using parachute technique (Fig. 4 and 8). When the sutures were tied nearly complete, the cross-field endotracheal tube was withdrawn and the remaining defect was closed. Then ventilation went back to depend on the primary endotracheal tube that was inserted into the upper trachea at the beginning of the operation (Fig. 9). In all patients, ventilation after the repair was much better and needed much lower peak airway pressure to ventilate. The integrity of anastomosis was tested by increasing peak airway pressure up to 30 cmH<sub>2</sub>O. Any leaking points was reinforced with 6-0 prolene double arm simple stiches. In the second and third patients simultaneous shunts for tetralogy of Fallot and the left pulmonary artery relocation for pulmonary sling were performed respectively. At the



**Fig. 4.** Slide tracheoplasty. A. Division of midpoint of stenotic part. B. Vertical split of the complete tracheal ring, along the posterior wall of proximal trachea and anterior wall of distal trachea. C. Lateral view of the spatulated segment of the trachea. D. Sliding of proximal and distal segment to fix over each other.



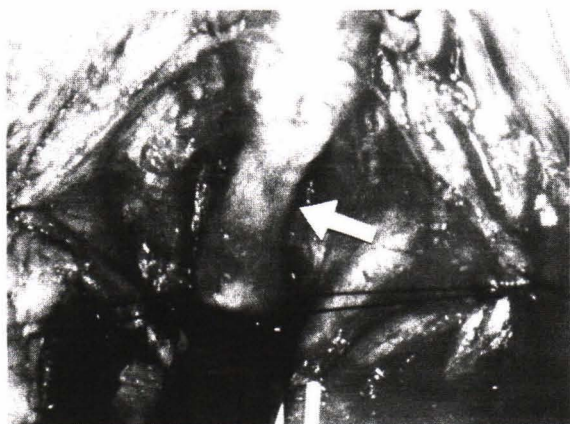


Fig. 5. The stenotic segments of lower trachea in patient 1 (arrow).



Fig. 6. Vertical splitting of posterior surface of the proximal stenotic part (arrow), (patient 1).

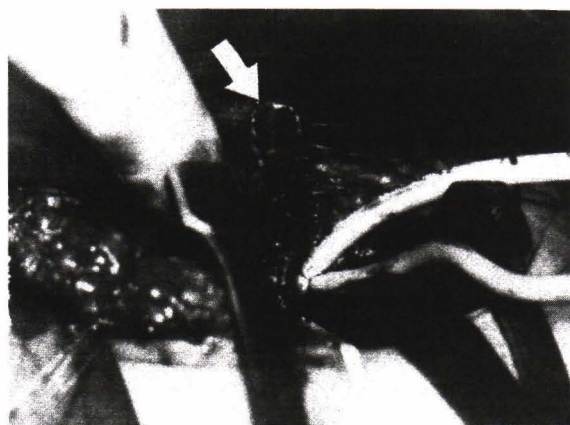


Fig. 7. The spatulated proximal stenotic part (arrow), (patient 1).

end of the operation, the wound was irrigated with warm normal saline and closed over a low pressure suction drain.

All of the patients were kept paralysed with neck flexion for at least 24 h. The chin-chest suture was used only in the first patient.

## RESULTS

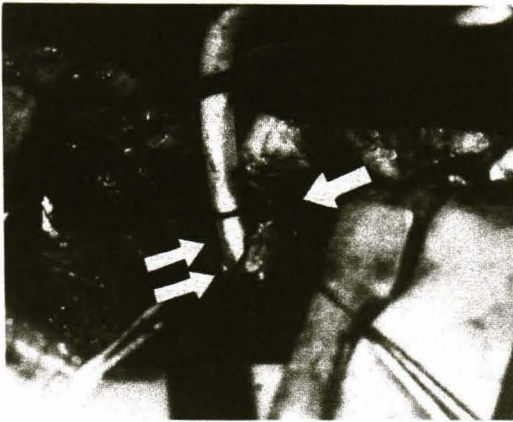
There was no operative death but some complications did occur. The first patient developed partial atelectasis of the right lung, requiring vigorous

suction and ventilatory support for 3 days. After that her progress was uneventful. At her last follow-up, 6 months after the operation, she was in good condition without any sign of airway obstruction. The second patient was reoperated upon to create a larger lumen at the bifurcation of right and left main bronchi with free pericardial patch, four days after the first operation. Then a tracheostomy was performed for tracheal toilet. She still needs repeated bronchoscopy to remove granulation tissue that is growing on the pericardial patch and dilatation of the anastomotic site. The third patient recovered uneventfully, completely asymptomatic after her operation and was extubated on the next day after the operation. The fourth patient had a large amount of air leakage from the tip of the posterior aspect of the anastomosis after the anastomosis was finished. It was not possible to access this point by rotation. So the anastomosis was taken down and reanastomosed. His postoperative period was remarkably uneventful and ventilatory support could be withdrawn two days after the operation. Follow-up bronchoscopy showed adequate diameter of the anastomosis without any granulation tissue.

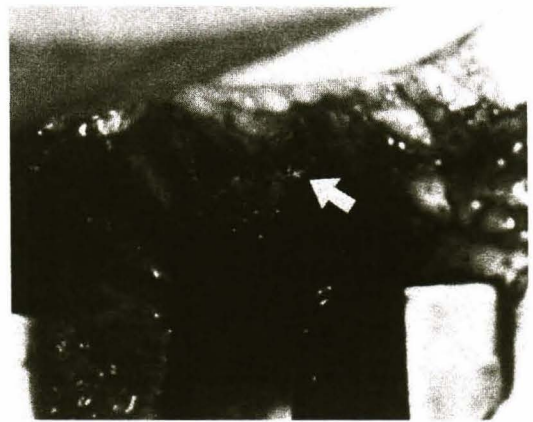
## DISCUSSION

Congenital tracheal stenosis is a rare condition with a high mortality rate. The important questions of the management are when and how the congenital tracheal stenosis should be treated. Before 1980, conservative treatment in long segment tra-





**Fig. 8.** The beginning of approximation of both spatulated parts by sliding one over each other (arrow). Endotracheal tube still remained in lower trachea for ventilation (double arrow), (patient 1).



**Fig. 9.** Finishing anastomosis (arrow). Ventilation went back to depend on endotracheal tube in upper trachea, (patient 1).

cheal lesion was acceptable because the narrow diameter of an infant trachea might prevent the success of corrective operation<sup>(3)</sup>. In the last 20 years, various techniques have been used to enlarge the stenotic trachea in infants. These techniques are divided into two major groups ; operative tracheoplasty and balloon dilatation tracheoplasty. In the operative tracheoplasty group, several techniques have been introduced such as costal cartilage graft tracheoplasty<sup>(5)</sup>, anterior pericardial patch tracheoplasty<sup>(6)</sup>, free tracheal autograft tracheoplasty<sup>(9)</sup>, and slide tracheoplasty<sup>(7,8)</sup>.

We decided to use the slide tracheoplasty technique as described by Goldstraw<sup>(7)</sup> in 1989 and modified by Grillo<sup>(7)</sup> in 1994, as we found that this technique is quite simple. Using a well vascularized trachea satisfactory results and good immediate and long-term outcome can be achieved<sup>(13)</sup>. Because of the high incidence of associated cardiovascular malformations, complete preoperative evaluation and planing should be performed<sup>(15)</sup>. In our second patient, we had no time to complete the investigation because of her serious and life threatening condition. The emergency operation was done immediately after the patient had been stabilized with an assisting cardiopulmonary bypass. So that the most distal stenosis at the left and right main bronchi could not be perfectly corrected. She needed a second operation to correct this problem but the trachea did not

have enough space for further sliding. The pericardial patch was applied to enlarge the lumen of both the main bronchi and caused growth of granulation tissue.

In the past, conservative treatment might have been suitable for long segment congenital tracheal stenosis but it should be abandoned now. Our patients had presenting symptoms several times before respiratory failure developed. The worsening of respiratory symptom were usually precipitated by respiratory tract infection or airway manipulation (e.g., endotracheal intubation). This can be explained by the fact that the small diameter of an infants' airway cannot tolerate swelling of the mucosal lining at the same degree as that of an adult. So it is harmful to leave a lesion like this for a long time. The patient who has a history of tracheal obstruction such as biphasic stridor, recurrent pneumonia, difficult intubation or extubation should be investigated thoroughly.

The preoperative investigation, apart from direct bronchoscopy should include CT scan of the airway and echocardiography because bronchoscopy alone cannot demonstrate the extent of a lesion and associated cardiovascular anomalies. Respiratory and cardiovascular malformations should be managed on a multidisciplinary basis and corrected simultaneously.

In patients who have respiratory failure due to acute upper airway obstruction, technique of respiratory support can be either *via* the endotracheal

tube above the obstructive lesion or by passing this tube through the obstructive lesion. Sitting the end of the endotracheal tube above the obstructive lesion is the much preferred technique to avoid any trauma to the obstructive lesion. Technique to ventilate this kind of patient is also one of the important points. The peak inspiratory pressure should be high enough to pass the obstructive lesion and in the appropriate rate that should not be so fast as to create turbulent flow through this obstructive lesion and enough for the exhalation. Sometimes this peak inspiratory pressure may be as high as 60-70 cmH<sub>2</sub>O and hand ventilation may be more practical in this situation with more plan to the treatment of this upper airway obstruction. Referring to the report "Use of high frequency jet ventilation"<sup>(16)</sup> in this kind of patient with case of severe obstruction might be more dangerous. Team management should plan not only for the treatment but also the long period of preoperative preparation, and during the surgical correction. Care should be taken in attention to both the location of the endotracheal tube that should be rechecked constantly and the pattern of ventilation during the period of transportation of this patient to the operating room, especially in patients with a severe obstructive lesion (as in the second patient) also at the time of arrival in the operating room and during operation. Anesthetic technique should include general anesthesia with appropriate control ventilation and the plan to prepare for the small endotracheal tube to ventilate during the tracheoplasty process or during cardiopulmonary bypass. Care should be paid to clear tracheal secretion before closing the tracheal stump to prevent postoperative complications of atelectasis, (as in the first patient). Appropriate narcotic, sedative and neuromuscular blocking agents should be used to control ventilation in the postoperative period.

Preoperative tracheostomy should be avoided because the common obstructive lesion is below the tip of the tracheostomy tube. So it is not helpful and causes some serious problems. One of our patients had a tracheostomy done at the referral hospital. This is not only useless but also an obstacle in the operative field. Intraoperative tracheostomy is also unnecessary because the slide tracheoplasty provides a non-collapsible airway that does not need any long-term airway stenting. So the only indication for tracheos-

tomy is tracheal toilet in the postoperative period (as in the second patient).

The use of cardiopulmonary bypass during tracheoplasty is still controversial even with reported use of ECMO<sup>(17)</sup> during the preoperative period. We do not use cardiopulmonary bypass routinely. Only two patients who had cardiovascular anomalies needed cardiopulmonary bypass. We found that cardiopulmonary bypass increased the opportunity to correct associated cardiovascular anomalies, but slide tracheoplasty alone can be performed adequately without it.

The technique of slide tracheoplasty requires adequate exposure of the anterior surface of the trachea from the cricoid cartilage to the carina, to identify the exact midpoint of the lesion. The circumferential dissection should be kept minimal, only around the midpoint and some distance away from the proximal part of the trachea. The proximal trachea should be mobilized both the anterior and posterior aspects. Anterior and posterior dissection of the upper trachea will not jeopardize the blood supply that comes along lateral border of the trachea. The distal trachea that was opened vertically on the anterior surface will have no dissection. The upper and lower tracheal flap can be slid together easily with cervical flexion and traction-stay sutures at both sides of the carina. The important point of this technique is the security of the sutures on the posterior aspect of the trachea especially at the tip of the lower flap. This point is inaccessible when the anastomosis is complete. Minor air leakage can be repaired with anterior vertical tracheotomy<sup>(8)</sup> but major leakage needed reanastomosis (as the fourth patient). When the anastomosis is nearly closed, soft suction should be applied within the trachea in order to clear secretions and blood from the airway so prevention postoperative atelectasis (that occurred in our first patient).

Considering the progress of our patients, the result of our operation and the literature review; long segment congenital tracheal stenosis should no longer be treated conservatively. We have no experience about balloon dilatation. The concept of slide tracheoplasty gives us a better way to deal with the previously poor prognosis lesion in our institution. The reconstructed trachea from this technique is uncollapsible and allows for early extubation. It also has no raw surface from any kind of graft. The aims

of healing of the trachea are first intentional healing without over granulation and second, tracheal growth which is not impaired as the infant grows<sup>(18)</sup>. Our

preliminary result shows that slide tracheoplasty is simple, attractive and suitable for infant that will have long life span.

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## การผ่าตัดแก้ไขหลอดคอตีบแคบอย่างรุนแรงในแนวยาวอย่างรุนแรงที่เป็นมาแต่กำเนิด ด้วยวิธี Slide Tracheoplasty : วิธีการผ่าตัดและการให้ยาระงับความรู้สึก

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ภาวะที่มีการตีบของหลอดคอที่เป็นมาแต่กำเนิดเป็นภาวะที่มีการตีบแคบของหลอดคออันเนื่องจากการที่กระดูกอ่อนของหลอดคามีลักษณะคอบง ทำให้หลอดคามีขนาดเล็ก พบภาวะนี้ได้ไม่บ่อยนัก แต่เป็นภาวะที่อันตรายถึงแก่ชีวิตได้ โดยเฉพาะเมื่อความผิดปกตินี้มีลักษณะเป็นแนวยาว และจำเป็นต้องมารับการผ่าตัดแก้ไข ซึ่งมักพบในผู้ป่วยเด็กเล็กหรือทารกแรกเกิดที่มีขนาดของหลอดคোট่ำๆ ถึงแม้ว่าจะมีวิธีการผ่าตัดชนิดต่าง ๆ ที่ช่วยในการรักษาผู้ป่วยกลุ่มนี้ แต่ปัจจุบัน slide tracheoplasty เป็นที่ยอมรับและมีผลลัพธ์ที่ดีทั้งในระยะสั้นและระยะยาว แต่เนื่องจากจำนวนผู้ป่วยในแต่ละรายงานมีน้อย จึงยังจำเป็นต้องอาศัยการรายงานเพิ่มจากสถาบันต่าง ๆ ในด้านการผ่าตัดและผลการรักษา ทั้งนี้เพื่อใช้เป็นแนวทางในการผ่าตัดรักษาผู้ป่วยกลุ่มนี้ต่อไปในอนาคต

รายงานฉบับนี้เป็นรายงานผู้ป่วย 4 รายที่มีประวัติ การตรวจค้นและวินิจฉัยว่ามีการตีบแคบของหลอดคอที่เป็นมาแต่กำเนิดและจำเป็นต้องได้รับการผ่าตัดแก้ไข โดยคณะผู้รายงานได้เลือกใช้วิธีการผ่าตัด slide tracheoplasty ซึ่งผู้ป่วย 2 รายใน 4 ราย ต้องใช้เครื่องปอด-หัวใจเทียม เนื่องจากมีโรคหัวใจพิการแต่กำเนิด (tetralogy of Fallot) 1 ราย และมี pulmonary sling 1 ราย การผ่าตัดผู้ป่วยทั้ง 4 รายประสบผลสำเร็จ ยกเว้นผู้ป่วยรายที่มีโรคหัวใจพิการแต่กำเนิดร่วมด้วยที่จำเป็นต้องมารับการผ่าตัดอีกครั้ง โดยใช้เยื่อหุ้มหัวใจช่วยขยายส่วนที่ตีบแคบของหลอดลมใหญ่ซ้ายและขวา รายงานนี้กล่าวถึงรายละเอียดของการผ่าตัดและวิธีการให้ยาระงับความรู้สึก รวมทั้งข้อควรระวังต่าง ๆ พบว่าการเจาะคอผู้ป่วยกลุ่มนี้จะไม่ก่อประโยชน์และทำให้เกิดความลำบากในการผ่าตัด

โดยสรุป การผ่าตัดผู้ป่วย 4 รายที่มีการตีบแคบตามแนวยาวของหลอดคอชนิดเป็นมาแต่กำเนิด โดยวิธี slide tracheoplasty พบว่าได้ผลดีและมีผลข้างเคียงน้อย

**คำสำคัญ :** หลอดคอตีบแนวยาวแต่กำเนิด, การตมยาสลบ, การผ่าตัด, slide tracheoplasty

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