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Original Article

Post Intubation Tracheo Esophageal Fistulae - Critical Analysis of Its Presentation and Surgical management

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ABSTRACT

Aim:To evaluate the Presentation, Surgical management and outcome in Patients referred with Post intubation tracheoesophageal fistula to a tertiary care hospital. Methods: We report our experience with Six patients (male: female - 1:5) who presented with Post intubation tracheoesophageal fistula managed in a single center over a period of January 2012 - October 2014. The mean duration of intubation days was 21.8 days . At presentation patients were debilitated from either pulmonary complications of aspiration (n=3, 50%) and malnutrition (n=3, 50%). Patients were evaluated with Upper GI endoscopy, bronchoscopy, MRI neck and CECT neck. Preoperative nutritional support was managed with PEG, Nasogastric tube and feeding jejunostomy. The TEF was closed with primary repair of the oesophageal defect in single layer in all cases. Primary tracheal defect repair in three cases, segmental resection of the trachea in three with single stage reconstruction in two patients, revision tracheostomy and delayed reconstruction of the trachea in one case. Sternocleido mastoid muscle flap interpostion in 5 cases. Results: Preoperative nutrition optimisation were given for all patients for a mean of 13.6 weeks. Postoperative complications developed in 33.3 % of patients (n=2). Mortality in two cases due to aspiration pneumonitis (n=2, 33.3%). Mean post operative hospital stay was 20 days (range, 8 - 56 days). Fistula closure was thus ultimately successful in 5 out of 6 patients (83.3%) and no long term complications during our mean follow-up of 2 years. Conclusions: Proper evaluation of Tracheoesophageal fistula is needed to assess the location, size and the adjacent tracheal lumen before planning definitive procedure. When there is no luminal compromise of the tracheal lumen, primary repair of the tracheal and oesophageal defects can be done. Interposition Muscle flap prevents re-fistulisation. Management is complex and the outcome depends on multiple factors.

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Introduction

PITEF is reported to be the most common cause of Adult Benign TEF despite advances in anaesthesiology care. This is attributed to the prolonged need for mechanical ventilation even with the use of the usual cuff inflation volumes. Factors responsible for morbidity and mortality need to be analyzed as this is a complex disease, critical clinical evaluation and decision making is necessary for optimal management.

Materials and methods:

Retrospective analysis of prospectively collected data was

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MATERIALS AND METHODS

done between January 2012 to October 2014 and analyzed six cases of PITEF who were referred to our department for definitive management. The patient's demographic data and accurate history taking was done and recorded systematically, written informed consent for Diagnostic and therapeutic procedures were obtained. The investigative modalities and treatment decisions were taken by multidisciplinary team .Type of surgery, Postoperative course and complications and mortality were evaluated. Complications were defined as immediate when occurs within 30 days and as long term after 30 days after primary definitive surgery. Patient was reviewed after discharge and re evaluated if symptomatic.

Surgical procedure

The patient is nursed in head-up position. Aggressive chest physiotherapy and other measures for pulmonary toileting are liberally utilized. These aforementioned measures are certainly helpful in most, if not all patients. Still, a small amount of saliva enters the respiratory passage which is amenable to tracheal suctioning.

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The optimum surgical therapy is individualized based on the etiology and the patho-anatomy .Surgery was done with adherence to all the principles of surgery of TEF as outlined by Grillo et al [1]

The steps we followed are

- a. Complete exploration & division of fistulous tract
- b. Primary repair of esophageal defect
- c. Harvesting of muscle flap
- d. Tracheal repair or Resection with single staged or delayed reconstruction

Complete exploration of the fistula

Neck was opened by 7-8 cm left cervical incision or transverse incision. Platysma muscle and the fascia along the anterior edge of the sternocleidomastoid muscle is incised in the direction of the wound. Gentle finger retraction is used to separate the sternocleidomastoid muscle from underlying cervical fascia medial to it. Retraction of the sternocleidomastoid muscle laterally helps identification of the underlying omohyoid muscle. The central tendon of which is divided tenses the contiguous omohyoid fascia layer is tensed by traction of the medial belly of the divided omohyoid muscle. Omohyoid fascia is incised in the direction of the wound cranially and caudally. The omohyoid fascia layer leads to the carotid sheath and its contents, which are gently retracted laterally. The larynx and the trachea are simultaneously retracted medially by the assistant's index finger and the risk of recurrent laryngeal nerve injury is minimized by avoid the use of metal retractor against the tracheo-esophageal groove during any portion of the operation. The middle thyroid vein is often identified by retraction of interior jugular vein and clamped at either end with fine right-angled clamps, divided and ligated. Esophagus was identified by palpating the nasogastric tube and hooked with finger from posterior to medially. Umblical tape was used to hold the esophagus. The fistulous tract is exposed with care taken to prevent recurrent laryngeal nerve injury.

In case of small size fistula with a long tract it can be hooked out and divided but if it is large with small tract, the fistulous site may be opened directly or seen after opening the trachea while doing tracheal resection.

Primary repair of esophageal defect:

After division of the fistula, the margins of the esophageal defect was trimmed until a healthy tissue is seen and the edges closed in single layers with 3-0 viacryl interrupted sutures with 4 mm distance.(Figure 1). A nasogastric tube is left as stent in esophageal lumen by passing the repaired site. In case of any minor leak enteral nutrition can be started through nasogastric tube.

Harvesting of muscle flap:

After the repair of opening in the esophagus ,as a part of preventing the overlap of ischemic zone over other ischemic zone which will lead to invariably fistulisation which is prevented by a superiorly based sternocleido mastoid pedicled flap was harvested without causing local functional disability or Aesthetic deformity. Fascia of lesser supraclavicular fossa between sternal and clavicular head of sterno cleido mastoid is dissected. Sternal head is isolated and detached from sternoclavicular joint. (Figure 2).Dissection proceeded cranially by carefully separating the

myofibers leaving behind transverse cervical artery branch to sternal head. In case a long muscle flap is needed for the retrosternal elongation of fistula closure ,muscle insertion with periosteum from the elongation is harvested from the retrosternal area in order to get a length. Sternohyoid muscle is carefully retracted and dissection is continued proximally upto the sterno cleido mastoid branch of superior thyroid artery and also will stop at and maintain intramuscular cock screw anastomotic vessel between transverse cervical artery and superior thyroid artery is maintained.(Figure 2)

The excursion of the raised flap with the easy interposition between repaired stoma between trachea and esophagus is examined (Figure 3). The muscle flap is fixed using 4-0 viacryl to contralateral pre tracheal fascia and carotid sheath enclosing thyroid by interrupted sutures.

Tracheal Repair, Resection and Reconstruction:

After division of the fistula the margins of the opening in the membranous trachea is trimmed. In fistula of size upto 2 cms and absence of any tracheal stenosis a primary repair of trachea is done with single layer interrupted 3-0 viacryl sutures.

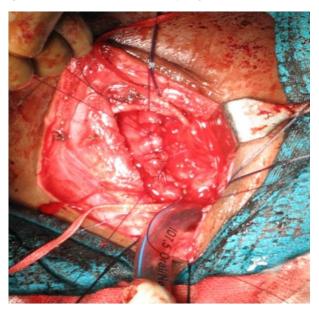
In fistula of size larger than 2 cm and presence of tracheostomy stoma stenosis or tracheal cuff stenosis with significant narrowing a segmental tracheal resection is done.

Trachea is opened, fistula opening into trachea is visualized (figure 4) and segmental resection done.ET tube is removed per orally and inserted into the cut end of the distal tracheal segment for ventilation. Traction sutures placed lateral part of cut ends with Polyglycolic acid (2-0 viacryl) sutures. The interrupted single layer 3-0 viacryl sutures with 4 mm distance is taken from posterior midline in membranous trachea and proceeded laterally and then completed anteriorly and all sutures left without tying. Discrepancy in size of proximal and distal trachea is corrected by proportionate suture placement.ET tube in distal trachea is removed and placed per orally from proximal trachea crossing the anastomotic site. Neck is flexed and lateral traction sutures are held by assistant surgeon to facilitate the close approximation of the resected ends ,starting to tie the knots anteriorly and lateral traction sutures are tied on one side and further securing the sutures towards posteriorly upto midline and this is proceeded in the other site to complete the tracheal anastomosis (figure 5).wound is flooded with saline and ET tube deflated and anesthetist transiently produces 30 cms of water pressure by continuous forced ventilation so that any air leak is noticed in wound help to check the anastomotic integrity. The wound is closed in layers with a suction or corrugated drain. Guardian sutures or 'Grillo' O silk stitch placed between the chin and anterior chest to achieve head flexion (35°) and this remain for 6 or 7 days following operation in those who underwent tracheal resection .This stitch prevents hyper extension of neck so that traction in the anastomotic region is prevented. When a tension free end to end anastomosis is not possible after a laryngeal release alternative procedure like fashioning a temporary tracheostomy helps to come out of this situation and Tracheal reconstruction can be done after 6 weeks as a delayed procedure.

Result:

Six patients (male: female -1:5) presented with PITEF. Median age of presentation was 30 years (range, 23 to 43). Causes for prolonged intubation were consumption of organ phosphorus poison with suicidal intent in 4 cases and following tuberculous

Figures 1: Primary repair of Esophagus



 $\label{lem:continuous} \textbf{Figure 2: line diagram-harvesting of sternocleidomastoid interposition \ muscle \ flap}$

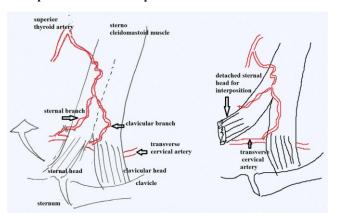


Figure 3: Muscle interposition flap over esophageal repair

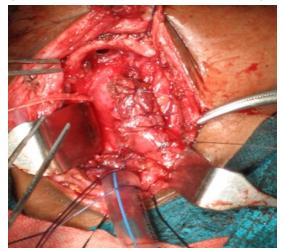


Figure 4: fistula opening seen through tracheostomy stoma



Figure 5: completed tracheal reconstruction



Figure 6:

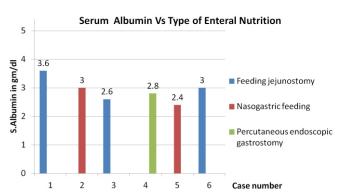
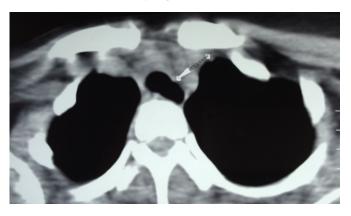


Figure 7: UGI scopy shows large communication of trachea and esophagus visualized as common channel at site of fistula



Figure 8: CT chest shows a large fistulous communication between trachea and esophagus



 $Figure \ 9: Duration \ of \ Mechanical \ ventilation \ Vs \ Size \ of \ Fistula$

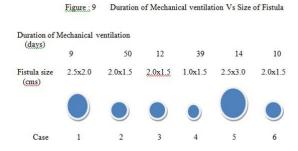


Figure 10:

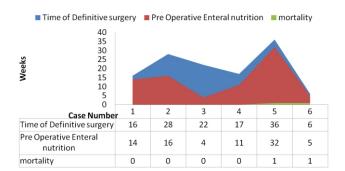


Figure 11:

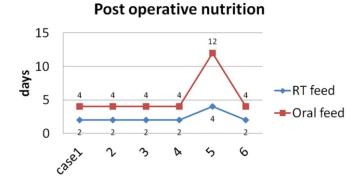


Figure 12:

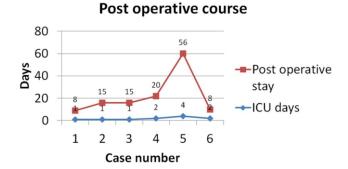


Figure 13: OGD scopy shows healed fistula site in anterior wall of esophagus



Table: 1 Sample characteristics

| characteristic | value |
|--|-------------|
| Age , mean(SD),Y | 30.66(8.11) |
| Age range, No.(%) of patients | |
| 20-30 | 4(66.6) |
| 30-40 | 1(16.6) |
| 40-50 | 1(16.6) |
| Sex , No.(%) of patients | |
| Male | 1(16.6) |
| female | 5(83.3) |
| Perfomance status-ECOG, | |
| No.(%) of patients | 5(83.3) |
| 1-2 | 1(16.6) |
| 3-3 | |
| Cause for prolonged intubation, No.(%) of patients | |
| Organo phosphorus poisoning | 4(66.6) |
| Myasthenia gravis | 1(16.6) |
| Partial hanging | 1(16.6) |
| Days intubated,mean (SD) | 21.8(18.08) |
| Symptom onset -Days after Extubation, mean(SD) | 7.5(4.72) |
| Timing of referral after symptom onset in weeks,mean(SD) | 13.1(9.43) |
| Timing of definitive surgery after symtpom onset in weeks,mean(SD) | 20.8(10.4) |
| Signs & Symptoms, No.(%) of patients | |
| Swallow induced cough | 6(100) |
| Coughing of ingested food | 6(100) |
| Breathlessness | 5(83.3) |
| Headache | 3(50.0) |
| Sinusitis | 2(33.3) |
| Aspiration pneumonia | 4(66.6) |
| Consolidation | 2(33.3) |
| Tracheal stenosis | 4(66.6) |

| Pre operative Nutritional status | |
|--|-------------|
| Haemoglobin, mean(SD) | 9.6(1.55) |
| Serum albumin g/dl, mean(SD) | 2.9(0.41) |
| Pedal edema, No.(%) of patients | 3.0(50.0) |
| Preoperative Enteral nutritional support in Weeks,mean(SD) | 13.6(10.17) |
| Enteral support, No.(%) of patients | |
| Feeding Jejunostomy | 3(50.0) |
| Nasogastric tube | 2(33.3) |
| Percutaneous endoscopic gastrostomy | 1(16.6) |
| Investigations ,No.(%) of patients | |
| Upper GI scopy | 6(100) |
| Bronchoscopy | 6(100) |
| X ray chest | 6(100) |
| X ray neck | 6(100) |
| X ray PNS | 3(50.0) |
| CECT neck | 5(83.3) |
| MRI neck | 2(33.3) |

Table: 2

| Type of Surgical Procedures | N=6 |
|--|--------------------|
| | No.(%) of patients |
| Esophageal repair | |
| Primary repair of the esophagus | 6(100) |
| Tracheal Repair | |
| Primary repair | 3(50) |
| Segmental Excision of trachea & tracheal anastomosis | 2(33) |
| Segmental Excision of trachea & Delayed tracheal anastomosis | 1(16) |
| Interposition of vascularized sternomastoid muscle flap | 5(83) |
| Difficulty in exposure of the fistula | 2(33) |
| Laryngeal release | 2(33) |

Table 3 : Data of Surgical outcome & complications following Post intubation TEF repair

| Immediate Postoperative complications (<30 days) | N |
|---|--------------------|
| | No.(%) of patients |
| Recurrent Tracheo-esophageal fistula | 1(16) |
| Esophageal leak | 0(0) |
| Air leak | 1(16) |
| Tracheal dehiscence-partial | 1(16) |
| Tracheal dehiscence-complete | 0(0) |
| Tracheomalacia and stridor | 1(16) |
| Wound infection | 1(16) |
| Aspiration pneumonia | 2(33) |
| Vocal cord paresis | 0(0) |
| Respiratory failure | 0(0) |
| Line infection | 0(0) |
| Haemorrhage | 0(0) |
| Quadriplegia | 0(0) |
| Deep vein thrombosis | 0(0) |
| Postoperative long term complications (>30c | lays) |
| Recurrent Tracheo-esophageal fistula | 0(0) |
| Esophageal stricture | 0(0) |
| Esophago-cutaneous fistula | 0(0) |
| Tracheal stenosis in patients after primary tracheal repair | 0(0) |
| Tracheal restenosis after tracheal resection | 0(0) |

Clinical presentation:

The patients' symptom is dominated by the presence of respiratory complaints due to aspiration of esophageal contents. The characteristic symptom of cough induced by swallow of oral liquids is pathgnomonic and is called the Ono's sign.

A bedside test can be performed by making the patient drink water stained with methylene blue which will be seen to appear in the tracheostomy. Nevertheless, aspiration of the swallowed water into the larynx and trachea can still produce the same results.

Hence, this test should be carefully interpreted. Patient can have purulent sputum and coughing out of ingested food can also occur. In certain cases, the fistula may be visible directly through the tracheostomy orifice itself. Mean of onset of symptoms related to PITEF after weaning from ventilator is less than a week in 4 cases .(mean 7days,range 1- 14days).All of the patients was diagnosed after weaning from ventilator and had swallow induced cough and coughing of ingested food. Breathlessness is seen in 5 cases. Headache was present in 3 patients and was associated with sinusitis in those two was on prolonged nasogastric feeding of more than 16 weeks.

The patients were referred to this center after a mean period of 13.1 weeks after onset of symptoms (range, 4 – 30weeks). The built and nutritional status was assessed (Table 1). Three patients had pedal edema and their serum albumin level was <3 g/dl (2.6, 2.8, 2.4 g/dl) and each had different type of enteral nutritional support (feeding jejunostomy, PEG and nasal gastric tube respectively. Timing of referral in those three patients were 12 weeks, 6 weeks and 30 weeks respectively. Preoperative nutritional support was managed with PEG (n=1, 16.6 %), nasogastric tube (n=2, 33.3%) and feeding jejunostomy (n=3, 50%) (Figure 6). Nutritional status depends on multiple factors like type of nutritional support prior to referral and timing of referral since onset of TEF.

Investigations:

Chest x-ray reveals pulmonary infiltrates and the stomach may be filled with air. The pulmonary status of the patients is in Table 1.

Bronchoscopy is the investigation of choice in patients suspected to have TEF. The lengths of the fistula and of the normal $\,$ airway are measured. A PITEF usually lies at or 1 cm below the level of a tracheostomy, since the fistula is located at the cuff site. Upper GI endoscopy also done to visualize the level and extent of the opening on the esophageal side. Patients were evaluated with Upper GI endoscopy (n=6, 100%) (Figure 7), bronchoscopy (n=6, 100%), MRI neck (n=2, 33.3%) and CECT neck (n=5, 83.3%)) (Figure 8). The defect was within 5 cms of the proximal trachea in all patients and fistula size measuring a mean diameter of 2.0 cm (range, 1.0 - 3.0cm) and tracheal stenosis in 4 patients. During follow up symptomatic patient was evaluated with UGI scopy, barium swallow & Bronchoscopy. At presentation all patients were debilitated from either pulmonary complications of aspiration (n=4, 66.6%) and malnutrition (n=3, 50%) (Table 1)Preoperative nutrition optimisation were given for all patients during a mean of 15 weeks (range,4 - 32weeks). Duration of mechanical ventilation and size of fistula formation does not correlated in this study, this indicates the Endotracheal cuff pressure is the main significant factor(Figure 9). The mean duration of intubation days was 21.8 days in this study.

Neck was opened by left cervical incision in three patients and transverse incision in three. In those who underwent a transverse incision, two was on tracheostomy status at surgery. Difficulty in intubation was present in 4 patients (n=4, 66.6 %) who also had tracheal stenosis, (Table 1). After the fistula was divided a primary repair of the oesophageal defect was done in all cases (n-6,100%). Tracheal defect was closed primarily in single layer (n=3, 50.0%) in three cases and tracheal stenting was done for one case who also had tracheal stenosis and tracheomalacia. In three cases who had tracheal stenosis the tracheal segments resected were $1.5\,\mathrm{cm}, 1.5\,\mathrm{cm}$ and $2.5\,\mathrm{cm}$ respectively , two underwent primary

reconstruction of the trachea and one after a laryngeal release(n=2,33.3 %) and one underwent a segmental tracheal resection with revision tracheostomy (n=1,16.6%) as there was tension in bringing the tracheal ends together after a laryngeal release[2,3], a delayed repair of Trachea as staged procedure done after 6 weeks .5 patients were buttressed by interposing pedicled sterno-cleido mastoid muscle flap (n=5, 83%) (Table 2). Suction drain was kept in all patients.

Flap cover was not used in one patient who underwent primary repair of trachea who also had tracheomalacia and for fear of extrinsic compression of tracheal lumen by flap, a tracheal stenting was done for stabilization of tracheal lumen. All these procedures are performed in a single stage in 5 cases and one patient delayed tracheal reconstruction done as second stage this study. All our patients was extubated immediately after surgery.

Immediate postoperative complications developed in 33.3 % of patients (n=2), these patients was treated with steroids for two weeks before referral. One had wound infection, tracheal air leak, Partial anastomotic dehiscence and aspiration pneumonia. In the patient with tracheal stent for tracheomalacia the stent was migrated down to carina and developed respiratory distress on 5th post-operative period, the stent was removed. She developed recurrent TEF in 3rd week following primary repair of trachea and esophagus without muscle flap. This patient also developed stridor due to tracheomalacia on 3rd post operative week and aspiration pneumonia (Table 3).

Mortality occurred in two cases. One postoperative death in patient following tracheal resection with laryngeal release and reconstruction as single stage who developed respiratory distress and surgical emphysema on neck and chest on 5th post operative day due to air leak and partial tracheal dehiscence on postero lateral aspect of tracheal reconstruction . Respiratory distress was relieved by removal of skin sutures and resuturing of tracheal dehiscence on the same day. This patient again developed partial anastomotic tracheal dehiscence on 9th post operative day and underwent tracheostomy(Table 3).He tolerated oral feeds on 2nd week and continue to have nutritional support both oral and through feeding Jejunostomy. The cause of death was due to aspiration pneumonitis in the 56 th post-operative day. Timing of definitive surgery may have an influence in mortality and related to debilitation of patient condition due to poor nutritional status and recurrent lung infection (Figure 10).

Another patient with recurrent TEF following primary surgery was readmitted with breathlessness and cough of food which was managed conservatively by Nasogastric tube, antibiotics, bronchodilators and chest physiotherapy. Patient suddenly developed inspiratory stridor which was managed by emergency tracheostomy .Patient died of aspiration pneumonia on 54th day following primary surgery.

Majority of cases (n=5,83.3%) tolerated liquid diet on 4th post operative day and soft solid diet on 6 th post operative day.(figure 11). Mean post operative hospital stay was 20 days (range, 8 – 56 days). (Figure 12). TEF closure was thus ultimately successful in 5 patients (83.3%) in those who all had an interposition muscle flap (Table 2).

The follow-up was of range 12 months to 27 months. No Long term complications was found in the follow up (Figure 13) with no evidence of recurrent TEF, esophageal stricture or tracheal stenosis (Table 3).

Discussion:

Until late 1960s, the largest series of acquired TEF patients did not contain single case related to endotracheal cuff trauma [4]. However, quite early in the next decade, a series by Thomas had reported 46 such cases [5].A PITEF occurs due to cuffed intubation causing excessive pressure on the tracheal mucosa, more than mean capillary perfusion pressure of the mucosa which leads to tracheal damage and pathologic changes such as ischemia, inflammation, ulceration, tracheal necrosis or stenosis and tracheo-esophageal fistula [6-10].

Over inflation of a large-volume cuff by even a small, added volume of air makes it into a high pressure cuff which can potentially erode the entire width of the membranous wall. These fistulas are generally large in size and are termed 'giant fistulas'. However these fistulas rarely have mediastinal leak/ sepsis since it progresses over period of time and does not occur acutely. Spontaneous healing of such fistulae never occurs and prevention is the best method to treat them. Usage of low-pressure, large-volume cuffs has reduced the incidence but still, the potential danger exists with prolonged intubation.

In this study the size of the fistula was not related to the number of days of mechanical intubation. It is recommended to maintain the cuff pressure within a narrow ideal range of 20 to 30 cmH2O to prevent complications. [11-13]. Although some articles propose a pressure as high as 40 cmH2O of ETT cuff pressure (which is equal to tracheal capillary pressure) for initiation of mucosal damage, [14, 15].

High intracuff pressure is probably the single most important factor in the development of post intubation TEF as we have not able to analyze this because all cases are referred after the establishment of fistula. Repair of PITEF should be delayed until the patient is fully stabilized and weaned from ventilation [18]. Any treatment directed towards TEF should principally aim at cessation of spillage of esophageal contents into the respiratory passage. Esophageal diversion is almost never necessary. Often times, the fistula is located so close to the cricopharyngeus that exteriorized esophagostomy is impossible.

Early extubation following major tracheal surgery is a debatable issue. There are reports of routine early extubation and selective early extubation as well [16,17]. As the chance of reintubation is very high in certain group of patients with decreased pulmonary function and tracheomalacia, these patients are better extubated after few days of postoperative ventilation. As all of our patients recovered from the primary cause which mandated prolonged intubation and they did not need further mechanical ventilation in the immediate post op period.

Tracheal resection is usually successful and has a low mortality. Tracheal anastomotic complications are uncommon, and important risk factors are reoperation, diabetes, lengthy resections, laryngotracheal resections, young age (pediatric patients), and the need for tracheostomy before operation[19].

Lengthy (> or =4 cm) resections is one of significant predictors of anastomotic complications with P value of 0.007 in single-institution, retrospective review of 901 patients who underwent tracheal resection in a study by Cameron D Wright et al [19]. In this study mortality in one patient who underwent tracheal resection had previous tracheostomy and length of the segment resected is 1.5cm . Tracheal defect may be too long to permit tracheal

reanastomosis, the esophagus is closed nonetheless to eliminate the fistula with a revision tracheostomy to prevent complications of tracheal dehiscence.

Structural interposition between the trachea and esophagus is advised to decrease the recurrence of TEF. It is generally advised to place a vascularized healthy tissue in between the tracheal closure and esophageal suture line . This is achieved either with a muscle or with a pericardial/pleural flap to separate the lumen of esophagus and trachea. Primary repair of both airway and esophageal defects with tissue flap interposition can safely be performed with good results. [20]. Adequate control of pulmonary and local sepsis is indispensible for good outcomes. Hence aggressive preoperative preparation is necessary. We would like to emphasize that these disorders are best managed in tertiary care hospitals where multidisciplinary team management is possible and helps in achieving the best possible results.

Conclusion:

Proper evaluation of PITEF is needed to assess the location, size and the adjacent tracheal lumen before planning definitive procedure. Nutritional and respiratory status of the patient should be improved prior to surgery. When there is no luminal compromise of the trachea, primary repair of the tracheal and oesophageal defects can be done. Tracheal Resection is attempted only if it appears reasonably possible to make a tension free anastomosis and should have all alternative modes of therapy available in mind. Muscle flap interposition prevents refistulisation.

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