

# THE DIAGNOSIS AND MANAGEMENT OF AN IATROGENIC ESOTRACHEAL FISTULA IN A PEDIATRIC PATIENT – CASE REPORT

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**Abstract:** Iatrogenic esotracheal fistula in pediatric patients is an extremely rare complication, and appears either because of the pressure exercised by the tracheostomy cannula or in patients that are long-term bearers of esogastric tubes, which will cause, together with the cannula, ischemic injuries and necrosis of the tissue. The treatment of these lesions is controversial. The paper presents the management of a pediatric patient with an acquired esotracheal fistula, secondary to a tracheostomy.

**Key words:** iatrogenic; fistula; tracheostomy; esotracheal; pediatric

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## INTRODUCTION

Tracheostomy is one of the oldest surgical interventions, the first such operation being reported in Egypt in 3600 B.C. In children, the first tracheostomy was realized by Habricot in 1620 on a 16-year-old patient; since then this procedure was widely used in the 1800s as treatment of acute respiratory insufficiency caused by diph-

theria. Currently, the indications of tracheostomy in pediatric patients include affections that require intubation and prolonged mechanical ventilation, different conditions that involve the upper airway obstruction or the necessity of a correct pulmonary toilet.

The rate of early and late tracheostomy complications in children is higher than in adults,

about 33-49%. Early complications include intra-operative events: hemorrhage, injuries of adjacent anatomical structures, pneumothorax, acute pulmonary edema, difficulties in inserting a cannula. Late complications include decannulation, hemorrhage, infection, inflammatory granuloma, aphonia, and deglutition disorders.

This paper presents the management of a pediatric patient with an acquired esotracheal fistula, secondary to a tracheostomy.

## CASE REPORT

We present the case of a 3-year-5-month-old male patient, hospitalized in the Pediatric Surgery Department of our hospital presenting a tracheostomy orifice in the cervical area.

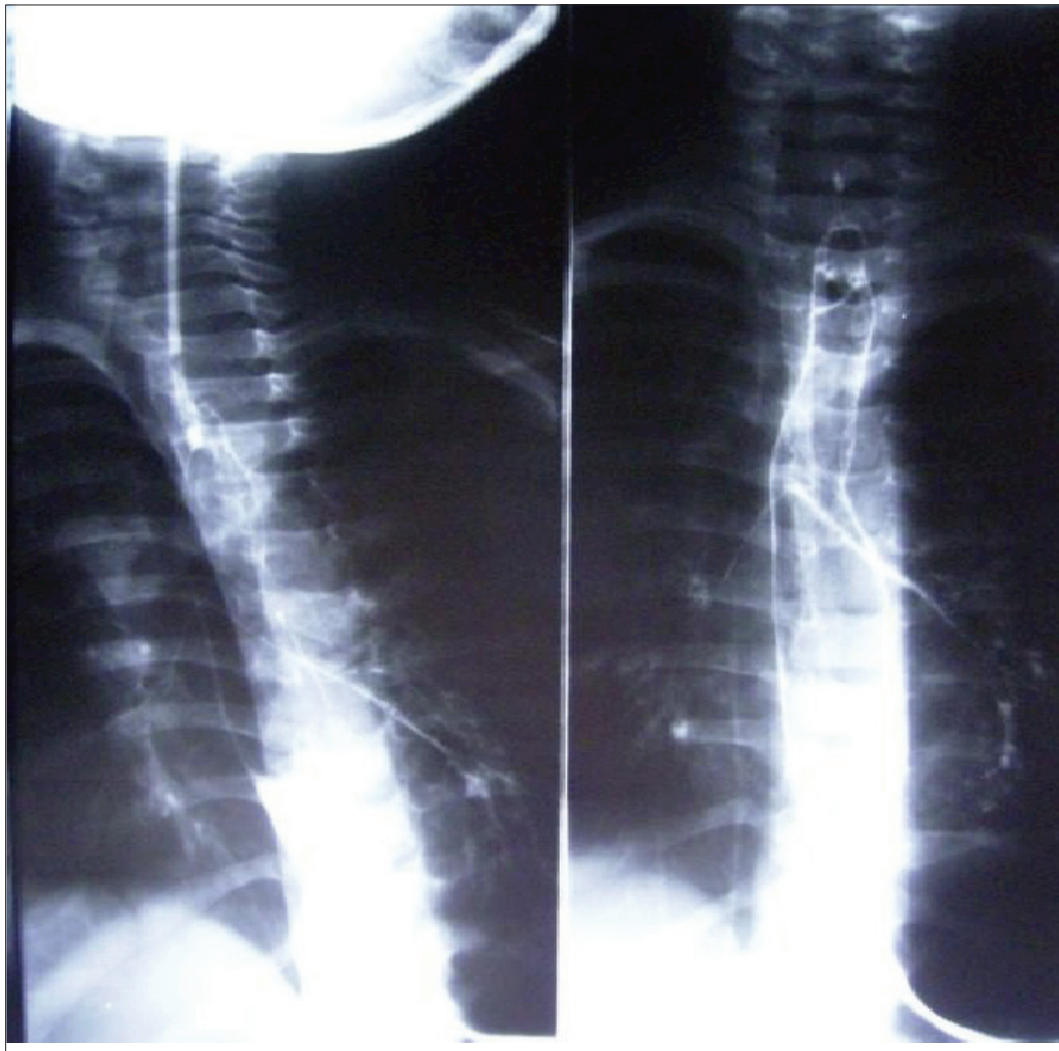
The patient's history revealed the beginning of the affection 10 days before his hospitalization, with high fever (40°), cough, dyspnea and tachypnea. He was admitted to a pediatric unit in a county hospital, where his respiratory status worsened with a decrease in O<sub>2</sub> saturation, important dyspnea, stridor and cyanosis. Phenomena of severe acute respiratory insufficiency were observed requiring orotracheal intubation, but this procedure failed because of massive glottic edema. The ENT surgeon was requested to perform an emergency tracheostomy but during the procedure, with the patient on spontaneous breathing under general anesthesia, cardiopulmonary arrest occurred. Resuscitation procedures were initiated and orotracheal intubation succeeded with the recovery of cardiorespiratory parameters; the tracheostomy was completed. About 8 h after



**Fig. 1.** Tracheostomy orifice. Nasogastric tube.

surgery, dyspnea and cardiorespiratory failure appeared again, the thoracic X-ray showing bilateral pneumothorax. Immediately, a bilateral pleurostomy at anterior axillary level in the fourth intercostal space was performed. Subsequent evolution attested remission of the respiratory status and the patient's decannulation after 4 days. Three days after the extraction of the tracheostomy cannula, saliva and alimentary scraps externalized at the level of the cervical anterior wound, through the stitches, and consequently the patient was sent to our hospital.

On admission, the patient had fever, productive cough, and evacuated an important quantity of aerated saliva and even milk at the level of the tracheostomy orifice. The thoracic radiography revealed an interstitial infiltrate. Taking into account the patient's clinical status and history, the existence of an esotracheal fistula was suspected and esophagus repose was decided by feeding through a nasogastric tube, as well as by gentle aspiration of the secretions from the proximal esophagus. In order to mark out the fistula level and also



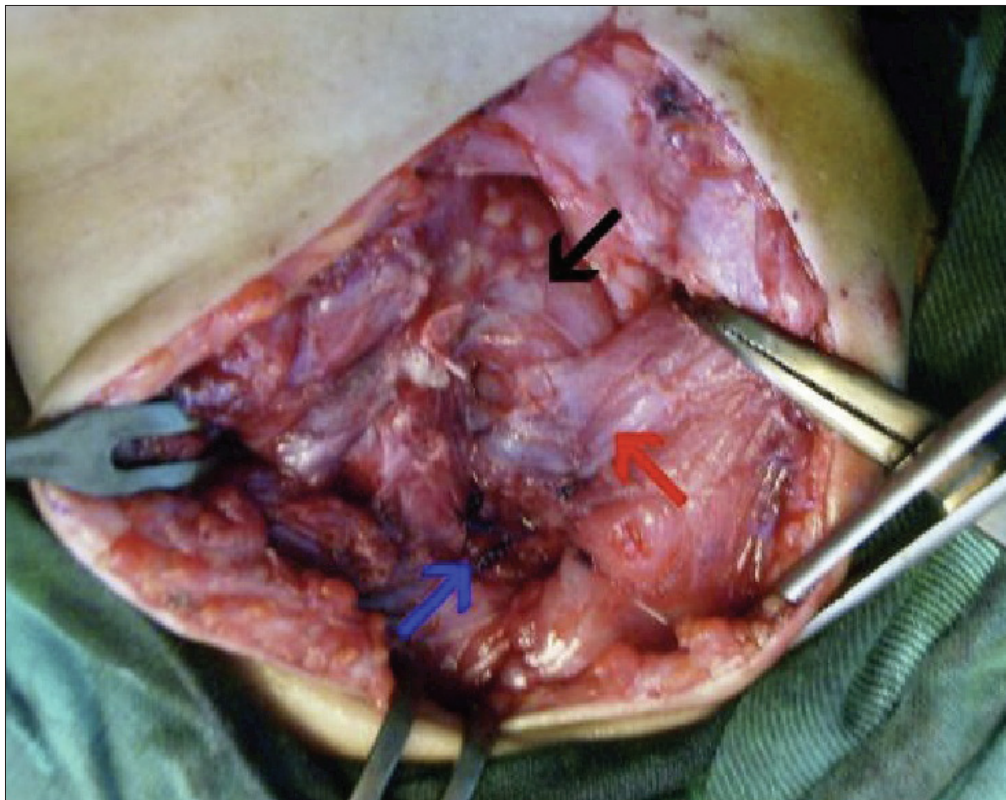
**Fig. 2.** Esogastric transit and bronchogram.



to exclude a possible tracheobronchial foreign body as the cause of the initial episode of acute respiratory failure, a bronchoscopy was done one week after admission without identification of a foreign body but only multiple, purulent tracheal secretions and mucosal edema. Methylene blue is injected into the esophageal tube with its gradual withdrawal, highlighting the colored substance on the posterior wall of the trachea at about 5 cm from the glottis, confirming the clinical suspicion. A multidrug resistant strain of *Staphylococcus aureus* was isolated from the bronchial aspirate. Conservative treatment with antibiotics (teicoplanin), nasogastric tube feeding and continuous aspiration of the superior esophagus was applied. Slow favorable evolution was obtained with a gradual decrease of secretions at the level of

the tracheostomy orifice, and subsequently with its closure. (Fig. 1)

Four weeks after hospitalization, under complete cicatrization of the tracheostomy cutaneous orifice, opacification of the esophagus was performed with a contrast substance, which revealed its passage in the bronchial tree, and the persistence of the esotracheal iatrogenic fistula (Fig. 2). Subsequently, an upper digestive endoscopy was performed with the identification of erythematous inflamed esophageal mucosa in the upper third, and the presence of a fistulous orifice on the anterior esophageal wall, about 10 cm from the dental arch. In these conditions, surgery was performed; a left laterocervical approach was performed with identification and isolation of the esophagus and trachea. Inflammatory phenomena were identified



**Fig. 3.** Intraoperative aspects. Fistulous orifice at the tracheal (blue arrow) and esophageal (black arrow) level; sternocleidomastoid muscle flap (red arrow).

together with the communication area between the trachea and esophagus at this level. The fistulous tract is sectioned and excised, residual defects being sutured, in a transversal single plan at the tracheal level, and in two plans of the esophagus wall. In order to prevent recurrence, a muscle flap removed from the left sternocleidomastoid muscle and from the prethyroid muscles was interposed by

suture to the prevertebral fascia. (Fig. 3). Operative loge was drained through an aspiration tube. The outcome was favorable, the esophagus opacification ten days after surgery revealing the integrity of its wall. (Fig. 4). The patient was examined after one month, six months and one year, respectively, and each time he was free of any symptomatology or radiological modifications.



**Fig. 4.** Clinical (a) and radiological (b) postoperative aspects.

## DISCUSSION

Acquired esotracheal fistulae are rare entities, very difficult to treat, especially in children, supervening most of the time after tracheostomy by hyperinflation of the balloon of the cannula after endotracheal intubation or after accidental ingestion of batteries by esophageal and tracheal wall erosion. The symptomatology of these patients include the evacuation of saliva and food at the level of the tracheostomy orifice, cough episodes associated with the moment of deglutition or repeated pulmonary infections. The diagnosis includes esophageal examination with a contrast substance, which will identify 70% of the lesions by the opacification of the tracheobronchial tree (Grillo, 2003). Endoscopy remains the best diagnostic method both for endotracheally intubated patients and for those with spontaneous respiration. Upper digestive endoscopy can leave small fistulae, hidden in the folds of the esophageal mucosa, while a rigid or flexible bronchoscopy ensures a better identification of the lesions, as well as the airway clearing by means of lavage. The instillation of methylene blue in the esophagus during bronchoscopy may identify the exact location of the injury, which is extremely useful for surgery (Couraud et al., 1998).

The management of these situations is not clearly defined, the literature presenting multiple treatment methods according to numerous causes that can lead to the appearance of these fistulae, hindering the definition of a clear treatment strategy. Therefore, the patient has to be evaluated according to the fistula etiology, his general status, duration of symptomatology, any possible previous surgery meant to repair the fistula and the optimal treatment strategy has to be adapted to each case. There is a consensus

regarding the moment of intervention, which has to be done after a stabilizing period, because the surgical treatment of the acquired esotracheal fistulae is not considered an emergency procedure (Biswas et al., 2010). Mechanical ventilation, through the positive pressure exercised in the trachea can also compromise the surgery by reopening the esotracheal communication. These patients' preoperative treatment has to take into account the minimization of saliva and food aspiration in the respiratory tract, the prevention and treatment of pulmonary infection, and the reduction of local inflammation (Chauhan and Long, 2004). In some cases, conservative treatment can lead to spontaneous closure of the fistula, after 4-11 months (Grisel et al., 2008).

The methods of surgical approach include the repositioning of the tracheostomy balloon beneath the fistula level, the exclusion of the esophagus by placing a gastrostomy or a jejunostomy in order to feed the patient, followed by fistula closure through resection of the involved tracheal segment, tracheal anastomosis together with suture of the esophageal defect, with or without interposing some muscular flaps (Dartevelle and Macchiarini, 1996, Grillo et al., 1976; Thomas, 1972). Other authors recommend the use of esophageal stents which do not present the risk of complications or mortality, but which have proved to be only temporary solutions (Eleftheriadis and Kotzampassi, 2005; Freire et al., 2006).

The cases of pediatric patients with acquired esotracheal fistula are extremely rare, most of the information found in literature referring to congenital ones, associated or not with esophageal atresia. In 1993, Rawlings et al. reported two cases of acquired esotracheal fistulae in two patients



with extreme prematurity who required long endotracheal intubation (Rawlings et al., 1993). In 1998, Birman and Beckenham reported two other cases of fistulae resulting from prolonged endotracheal intubation (Birman and Beckenham, 1998).

In our case, the clinical diagnosis of the lesion was realized by identifying the evacuation of saliva through a cutaneous orifice of the tracheostomy. The conservative treatment, including antibiotics, alimentation by nasogastric tube, as well as gentle aspiration of the secretions from the esophagus proximal to the lesion, was initiated in order to facilitate the spontaneous closure of the lesion, but also to reduce the local inflammation and the lesions of pulmonary aspiration. Despite the favorable clinical evolution, with gradual reduction of secretions through the cutaneous orifice of tracheostomy followed by its complete cicatrization, the esogastric opacification and upper digestive endoscopy identified the persistence of the fistulae. We nonetheless think that the initial conservative treatment led to an important reduction of the local inflammation, which insured the success of the surgical treatment together with the interposition of the muscular layer between the esophagus and the trachea.

Although it is rare, an acquired esotracheal fistula secondary to a tracheostomy can be a redoubtable complication. In order to perform a tracheostomy in pediatric patients with minimum risks, good communication between the anesthesiologist and the ENT surgeon is necessary, especially if the indication is that of acute respiratory failure. Tracheostomy is preferably performed on an anesthetized patient, with an already placed endotracheal tube or with a bronchoscope placed in the trachea. The success of

surgical treatment of an acquired esotracheal fistula depends on the immediate recognition of the clinical symptomatology, on imaging diagnostics, especially with endoscopic identification of the dimensions and location of the lesion, as well as on the correct staging of the treatment. The initial conservative therapy is essential for a patient's stabilization and for enhancing the effectiveness of the surgery.

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