A mediastinal tracheostomy is a highly complex procedure. Despite presenting great morbidity and mortality, it is not proscribed and could be indicated for selected cases. Maneuvers such as transposition of the trachea under the innominate artery and pectoralis major flap rotation have been developed to reduce the risk of complications. In some cases, it is necessary to reconstruct the alimentary tract with several options depending on the extent and the co-morbidities of the patient. Relevant articles were identified from a literature search using the Pubmed database and Google Academic site using the keywords: Mediastinal tracheostomy, cervical exenteration and pharyngeal reconstruction. There were no restriction on publication data. A 52-year-old male patient with squamous cell carcinoma of the hypopharynx submitted to total laryngopharyngoesophagectomy with colon transposition. A colon-pharyngeal fistula and dehiscence of the tracheostoma with tracheal retraction into the mediastinum were presented as complications. In reoperation, a mediastinal tracheostomy was confectioned for better access to the airway and to perform a fistula repair. Patient had no new complications and is in outpatient control with no recurrence of cancer and only one episode of pneumonia after hospital discharge. The principal articles with the largest series found about mediastinal tracheostomy are presented with discussion about the indications, complications, digestive reconstruction and results. We present a case in which the mediastinal tracheostomy presented an unusual indication and was conducted in a successful manner. Thus, according to the discussion, it is a feasible procedure in well-selected cases.

Keywords: Mediastinal tracheostomy, Colon transposition, Tracheostomy

INTRODUCTION

Mediastinal tracheostomy (MT) is the construction of a stoma on the anterior chest by using the intrathoracic trachea when there is insufficient length for re-anastomosis with the remaining trachea or for a traditional suprasternal tracheostomy. This procedure requires a laryngectomy (if not done previously) associated with the removal of the upper sternum, the
medial third of the clavicles, and eventually the proximal third of the first and second ribs, which provides access to the intrathoracic trachea (Orringer, 1992; Grillo et al., 1990; Gomez et al., 2007). Few surgeons or institutions have extensive experience due to the rarity and complexity of this procedure and its association with high morbidity. In the literature, there are some case series found with small sample sizes, but with acceptable results (Conti et al., 2010).

In 1942, Watson (1942) planned a procedure for the treatment of a squamous cell carcinoma 4cm above the carina. The patient had undergone a laryngectomy followed by radiotherapy 15 years prior. A "V" portion of the sternum was resected and skin flaps were mobilized to allow the closure of the tracheostomy margins.

In 1952, Kleitsch (1952) removed the upper sternum and inserted a polyethylene tube for a mediastinal tracheostomy. In the same year, Minor (1952), after removal of a recurrent carcinoma in a tracheostoma, fashioned skin flaps in the shape of a tube through a sternal opening to connect with the trachea. In 1959, Waddell et al. (1959) pulled a trachea segment on the right of the ascending aorta and anastomosed it with a skin tube created with flaps of the anterior chest region, through an opening in the sternum. Of the four patients who have undergone this procedure, two died of massive bleeding.

In 1962, Sisson et al. (1962) resected a large portion of skin along with the tumor and removed the notch and the heads of both clavicles in a surgery for recurrence of laryngeal carcinoma in stoma. Skin flaps were mobilized to the superior closure of the stoma and the lower failure was covered with skin grafts. After the procedure, 2 patients died from postoperative bleeding by the innominate artery and the pectoralis muscle (PM) flap began to be interposed between the trachea and the innominate artery.

In 1966, Grillo (1966) fashioned a large bipedicled total thickness flap from the skin of the anterior chest region through two main horizontal incisions in an effort to eliminate the tension in the tracheocutaneous anastomosis responsible for the poor healing and the threat of bleeding from the innominate artery. This flap found the terminal stump of the trachea in the mediastinum, which was accessed by removing the notch and sternal portions of both clavicles and the first and second costal cartilages. The stoma emerged at the center of the flaps, requiring only a simple suture.

According to Conti et al (2010), the main indications for mediastinal tracheostomy are malignant neoplasms of the subglottic region extending to the proximal trachea, a recurrence in stoma after laryngectomy, and a well-differentiated thyroid carcinoma with tracheal invasion, after either curative or palliative resection. The same authors recommend chemotherapy and radiation associated with tracheal stent for cases of cervical esophageal cancer with tracheal invasion, once 3-years survival rates is just 11% after radical procedure. Some authors suggest that the minimum length of the residual trachea should be 5.0cm (Orringer, 1992; Gomez et al., 2007). The presence of metastases is not an absolute contraindication for this procedure. However, in these cases, other methods for palliation with less morbidity should be considered, such as radiotherapy or stent placement (Conti et al., 2010).

According to Maipang et al (1996), the main contraindications for mediastinal tracheostomy are the invasion or involvement of the great vessels, distal trachea invasion of 3.0 to 4.0 cm above the carina, column or prevertebral fascia invasion, and the presence of disease in the bronchial lumen.

The patient must be carefully selected for this procedure due to its complexity, potential for complications and degree of deformity, and postoperative sequelae. The reconstruction of the alimentary tract should be taken into consideration, as, according to the necessity, there will be a considerable increase in the complexity of the surgery. Thus, a good preoperative evaluation becomes crucial. The patient needs a good performance status, good heart and lung function, must be well nourished and psychologically prepared for the deformities and sequelae of resection and reconstruction (Gomez et al., 2007). A history of abdominal surgery influences the type of reconstruction. A history of cervical radiotherapy compromises the tracheal circulation, which increases the risk of ischemia, and makes it more difficult to access the large vessels of the neck for a microsurgical anastomosis (deVries, 1989; Carlson, 1992).

The most commonly used incisions are the collar incision and the bipedicled/apron incision. In cases of recurrence in a previous tracheostoma, the peritracheostoma skin should be resected. Local flaps are used to cover the defect generated by resection and to avoid the stress of structures, such as the deltopectoral flap, PM flap, and bipedicled flap (Orringer, 1992; Grillo et al., 1990; Berthet et al., 2014).

In the case of a collar incision, a platysmal subcutaneous flap is made to expose the larynx, trachea, sternocleidomastoid, and carotid sheaths. The lower flap is made for exposure, and resection is made with a surgical saw of the middle third of the clavicle, sternal notch, and cartilaginous limit of the first two ribs, with preservation of bilateral internal thoracic arteries. The need to extend the second rib resection depends on the length of the remaining trachea. With this, we can have a good view of the upper mediastinal structures. Prior to the use of the saw, the digital dissection of the posterior wall of the sternum is recommended (Orringer, 1992; Berthet et al., 2014).

The trachea is divided obliquely to facilitate the skin suture. The inferior portion is re-intubated and the superior portion is resected along with the affected structures. After that, the trachea is released from the
esophagus by blunt dissection. When the tracheal stump causes compression on large vessels, it is transposed inferiorly through the innominate artery to decrease the distance from the skin and to prevent the anastomosis tension, reducing the risk of peristomal dehiscence, mediastinitis, and rupture of large vessels (Orringer, 1992). Sisson et al. (1962) described the importance of separating the innominate artery from the trachea by using a PM flap, and thereby, reducing the occurrence of vessel rupture and fistula. For Orringer (1992), the most important factor to avoid this disastrous complication was considered to be a tension-free reconstruction, and thus, a myocutaneous flap can be used, such as the deltopectoral flap and thoracoacromial flap (“nipple flap”).

Grillo et al (1990) described modifications in the MT and cervical exenteration techniques. They performed a prophylactic ligation of the innominate artery in all patients with some degree of tension between the trachea stump and the mediastinal vessels under electroencephalographic monitoring. Prior to surgery, all patients underwent arteriography to assess the cerebral vasculature and the patency of the Willis Polygon. The authors also used the omentum to cover the artery stumps and to separate them from the trachea, and gave preference to a bipedicled flap for cutaneous coverage of the tracheostoma.

The main ways to reconstruct the alimentary tract are the transposition of intra-abdominal organs (stomach, jejunum, colon) or the use of microsurgical flaps (jejunum or myocutaneous flaps). The decision must take into account several factors: the main one is the extent of reconstruction. In patients who are submitted to total esophagectomy in addition to laryngotraceal resection, it is necessary to transpose the intra-abdominal organs. The elevation of the stomach is often preferred, due to the reduction in surgical time and fewer anastomoses. In patients who have already undergone some gastric surgery, such as a gastrostomy, the next choice may vary between a long colon segment or jejunum. When only a pharyngectomy is performed, without the need for full esophagectomy, a microsurgical jejunal flap or a tubed myocutaneous flap (radial forearm or anterior thigh) can be used (deVries, 1989; Carlson, 1992; Goligher, 1954; Goldberg, 1989).

Postoperative care should be intense. Immediate postoperative intensive care unit (ICU) placement is recommended in patients undergoing esophagectomy and microsurgical anastomoses. The major cause of mortality after surgery is the primary rupture of the innominate artery with mortality rates ranging from 33%–55%. (Waddell et al., 1959; Sisson et al., 1962; Terz et al., 1980) Other complications are alimentary tract fistula from anastomosis, ischemia of the trachea, skin flap loss, chyllothorax, and hypoparathyroidism. Nevertheless, MT can offer acceptable terms of palliation and quality of life, similar to that of patients undergoing routine laryngectomy (Orringer, 1992; Grillo et al., 1990; Conti et al., 2010). Bone resection (manubrium, clavicle, sternum, first and second ribs) can create an area of instability in the anterior chest with pulmonary herniation and an increased susceptibility to pneumonia (Berthet et al., 2014).

In this paper, we describe a case where the indication of MT was a consequence of postoperative complications in a patient submitted to laryngopharyngectomy associated to a total esophagectomy. In addition, the reconstruction transit was performed by colonic transposition, which is not routinely used.

**CASE REPORT**

The patient was a 52-year-old male and a smoker who presented in August 2013 with otalgia, odynophagia, dysphagia, and hoarseness that started 6 months prior to admission. Laryngoscopy revealed a tumor affecting arytenoids, interarytenoid space, post-cricoid region, aryepiglottic left fold, and left pyriform sinus, with extension to the superior esophageal sphincter and cervical esophagus confirmed by MRI scan and computed tomography (CT). The left hemilarynx was paralyzed, and there was destruction of the cricoid and thyroid cartilages. Cervical lymph nodes were not clinically affected. PET CT did not establish disease in other sites. Clinically staged as T4a N0 M0. As the patient refused radical surgical treatment, he was submitted to an indication scheme with 3 doses of Taxol/platinum/5—fluorouracil in days 1, 22, and 43, with excellent response. As a result, we decided to continue conservative treatment with intense modulated radiotherapy (IMRT) (7000 cgy) associated with cetuximab.

In week twelve after treatment, the laryngoscopy and PET CT scan showed complete response. Patient remained in monthly control, but did not attend regular consultations.

Fourteen months after his initial treatment, the patient presented again with otalgia and noisy breathing, and with associated dysphagia. The laryngoscopy showed ulceration throughout the post-cricoid region. A digestive endoscopy showed a 3-cm long involvement of the cervical esophagus with tracheal invasion.

Endoscopic gastrostomy was performed, and a rescue surgical treatment was indicated, which was again refused. Cetuximab was initiated in a maintenance scheme. After two months, the patient returned with hoarseness and dyspnea, and an urgent tracheostomy was required. In August 2015, the patient was submitted to a total laryngopharyngoesophagectomy, with colonic 3esion3ernal transposition for alimentary tract reconstruction, associated with resection of the affected trachea and confection of a tracheostomy (Figures 1–3). A left deltopectoral flap was used to cover the colon and to avoid cutaneous tension.
In the postoperative period, the patient presented with a colon—pharyngocutaneous fistula and severe cervical infection, which caused a tracheostomy dehiscence and collapse of the tracheal stump to the mediastinum. The fistula was primarily closed, but without success.

After extended hospitalization in the ICU, the administration of a large variety of antibiotics and infectious-process resolution, a great difficulty of cannula exchange and airway patency remained due to the depth of the tracheal stump position.

In November 2015, MT was indicated. It occurred with resection of 50% of the manubrium, removal of the clavicular heads, and transposal of the tracheal stump inferiorly through the innominate artery. The colon—pharyngocutaneous fistula was again primarily closed. The left PM flap was used for mediastinal vessel coverage and a right deltopectoral flap was used to reinforce neck coverage (Figures 4–5).
The patient was evaluated with a colon-pharyngocutaneous fistula that needed to be closed twice during hospitalization with re-suturing of the edges, but with no complications of MT. Patient received discharge for ambulatory control in the third postoperative month from the first surgery.

Currently, 10 months after the rescue surgery and 7 months from MT, there is no evident disease or surgical complications. The patient presented just a single episode of moderated pneumonia that needed a short hospitalization for systemic antibiotic therapy.

**DISCUSSION**

In our reported case, the first treatment planned was a radical surgical resection because there was locally advanced cancer. However, the patient refused this treatment plan, leading to completion of a chemotherapy regimen of induction (TPF) associated with concurrent radiotherapy with cetuximab. This treatment consisted of three cycles 21 days apart (75mg/m² Taxol d1; 35mg/m² platinum d1 and d2; and 750 mg/m² 5-FU d1–5) as an option to radiotherapy with the intent to preserve organs.
with control comparable to surgical treatment. Another advantage to this treatment is a better laryngeal preservation rate than with the exclusive use of radiotherapy (Jordan et al., 2010; Pfister et al., 1991; Karp et al., 1991). In addition, the replacement of cisplatin by cetuximab has lower levels of toxicity when performed concurrently with radiation therapy (Lefebvre, 2009).

Organ preservation protocols were introduced after a study that observed the effectiveness of induction chemotherapy followed by radiation therapy compared to standard therapy (surgery followed by radiation therapy). This study showed a 64% laryngeal preservation in the first group with similar survival in both groups (D.V.A.L. C.G., 1991). Other studies have found that a concomitant chemoradiotherapy scheme had even better laryngeal preservation rates with survival similar to induction therapy (Forastiere et al., 2003; Prades et al., 2010). Bonner et al. (2006) observed that the combination of cetuximab with radiotherapy produced better oncological results than radiotherapy alone. Finally, the TREMLIN study showed that the use of cetuximab associated with radiotherapy after a TPF-induction regimen could be an alternative with less toxicity when compared to the traditional scheme of concurrent chemoradiation (Lefebvre, 2009). However, in patients with large tumor volume, gross invasion of cartilage, or functional impairment (airway obstruction and difficulty in swallowing), radical surgery followed by adjuvant radiotherapy remains the standard treatment (Rosenthal et al., 2015).

Thus, a laryngopharyngoesophagectomy would be the gold standard treatment in this case but the patient denied it. We therefore chose the TREMLIN scheme with the aim of organ preservation, which obtained a good response for 14 months.

MT consists of a procedure that is usually indicated as a reestablishment of an alternative airway after resection of advanced tumors, especially after laryngopharyngectomy associated with esophagectomy, and after resection of recurrence in the tracheostoma (Chan et al., 2011). In the presented case, the indication was not because of a tumor, but as a way of solving a surgical complication of the first procedure, the anastomotic dehiscence with migration of the remaining trachea into the mediastinum.

Current literature shows a small series of cases, often obtained over decades. However, these studies show the degree of morbidity and mortality associated with this procedure as well as the techniques used to protect the artery and innominate vein (Orringer, 1992; Grillo et al., 1990; Berthet et al., 2014).

Orringer (1992) studied 44 patients who underwent MT, 10 as a palliative procedure and another 34 (72%) for airway reconstruction after cervical exenteration (pharyngolaryngectomy with esophagectomy). Of the latter, 31 had alimentary tract reconstruction by gastric transposition and 3 by transposition of the colon. A perioperative mortality rate of 14% (6 patients) was observed, all of whom submitted to cervical exenteration. Only 9 patients (32%) had a postoperative course without complications. The length of stay ranged from 10 to 51 days (median 26.2 days). Only 1 patient had a rupture of the innominate artery, in which case the trachea was not transposed down the artery. Orringer reported that the critical factor to avoid fatal complications was the elimination of tension between the structures, which could be achieved with the implementation of the trachea below the innominate artery.

In our case, postoperative complications were due to a preexisting colon-pharyngeal fistula and the patient would have evolved without complications from the viewpoint of the MT. The trachea was moved beneath the innominate artery, which guaranteed us a trachea–skin suture without tension. In addition, a PM muscle flap was essential for the protection of large vessels.

In the work of Kamiyama et al. (2015), 40 patients undergoing pharyngolaryngectomy with a total esophagectomy were studied. MT was performed in 9 cases (22.5%), and of these, 4 (44%) had complications related to the tracheostomy. Of the 31 patients who underwent traditional tracheostomy, only 4 (12.5%) had complications related to ostomy. Survival at 5 years was 48.6% and perioperative mortality was 5% (2 cases), 1 by bleeding due to brachiocephalic vein injury and the other due to injury of the innominate artery.

Berthet et al. (2014) evaluated 12 patients undergoing MT, all for recurrence in stoma after laryngectomy. In all 12 surgeries, relocation of tracheal segments remaining below the innominate artery and a myocutaneous flap to cover the tracheostomy were necessary. The reconstruction of the alimentary tract was required in 4 patients (3 with gastric transposition and 1 with primary closure). There was 1 patient death in the postoperative period (8.3%), due to a vascular fistula. The length of hospital stay varied between 13 and 86 days. Survival at 5 years was 53%. The author concluded that despite the surgical risks, the long-term survival after total resection of the lesion is acceptable.

Grillo et al. (1990) performed MT in 14 cases in a series of 18 patients who submitted with cervical exenteration. The innominate artery had to be divided in 7 of these patients. Of these 7, only 1 had hemiplegia, which was treated through a bypass to the left subclavian artery. Alimentary tract reconstruction was performed, preferably by the choice of the colon, transposing it through a substernal tunnel. In this study, the colon was used for the reconstruction in 10 patients, while the stomach was used only in 3. There was 1 patient death in the study due to anastomotic leakage in a reconstruction with the gastric tube, which resulted in mediastinal sepsis. The median survival was 10 months, and 6 patients had survival higher than 4 years. The authors describe functional results equivalent to laryngectomy and recommend this procedure for palliation only in...
cases with a survival prediction longer than 6 months. Chan et al. (2011) studied 38 cases of patients with cervical mediastinal tumors who underwent MT. There was no artery ligation in any case. Of these, 38, 31 (81.6%) required repositioning of the trachea below the innominate artery to prevent tension and 14 patients required a PM flap to tracheostoma closure. Digestive tract reconstruction was necessary in 34 cases, of which 8 had a primary closing of the neopharynx. A jejunum free flap was used in 8 cases, the gastric tube was transposed in 12, and a tubed PM flap was made in 6 patients. The authors describe an in-hospital mortality of 12, and a tubed PM flap was made in 6 patients. The authors describe an in-hospital mortality of 2 cases (5.3%) due to bleeding of the great vessels. There was a leakage of gastrointestinal Anastomosis in 6 patients (3 who underwent gastric transposition and 3 who were reconstructed with a tube-shaped PM). Eleven patients (28.9%) had ischemia or partial necrosis of the terminal portion of the trachea. In follow up, 18 patients (47.4%) had stenosis of tracheostoma. In these cases, a history of leakage of the anastomosis or tracheal ischemia was statistically significantly as a risk factor for tracheostoma stenosis (p = 0.34 and p = 0.26, respectively). The PM flap was considered as a protective factor, inasmuch as tracheostoma stenosis occurred in just one (7.1%) of patients submitted to it. The survival rate was 80.6% at 1 year and 55.6% at 5 years. The authors recommended not to skeletonize the terminal stump of the trachea to prevent its ischemia.

In a study involving 13 patients undergoing MT, Conti et al. (2010) described 2 in-hospital deaths (one for bleeding of the innominate artery), and only 5 cases with no complications. The average hospital stay was 29 days, ranging from 12 to 101 days. Survival at 3 and 5 years were 57% and 43%, respectively. Worse outcomes were observed in patients operated for esophageal carcinoma or for laryngeal carcinoma recurrence.

Maipang et al. (1996) analyzed 12 cases of MT associated to laryngopharyngeal carcinoma and gastro transposition. The average length of hospital stay was 50.6 days (range 27–104 days). Complications occurred in 5 patients. There were 2 hospital deaths, 1 by bleeding due to the innominate artery, and the other by stroke. We chose colonic transposition because the patient was submitted to a previous gastrostomy. The myocutaneous flaps were not suitable due to the need to reconstruct the esophagus. In existing literature, when a complete resection of the esophagus occurs, the main options are the gastric pull-up, colonic transposition, and transplantation of the jejunum, with the choice depending on the service experience and the specific conditions of the patient (previous gastrostomy, prior laparotomy, and previous colonic or jejunal surgery). When the gastric pull-up is chosen, there is the possibility of distal ischemia or gastric stumps ischemia, which rarely happens with the colon. Furthermore, the colon can provide a large segment for reconstruction, but at the expense of increased intra-abdominal anastomoses. In cases where only the cervical esophagus is resected, the microsurgical jejunal flap is a good option, as are tubed myocutaneous flaps (e.g., the anterolateral thigh flap and forearm flap) (deVries, 1989; Carlson, 1992; Goligher, 1954).

The MT is an important option for airway reconstruction where there is an insufficient length of trachea to perform the traditional procedure in the cervical region. There was a decrease in the incidence of innominate artery rupture after the use of flaps for coverage and confection of tension-free trachea–skin Anastomosis. Although it has a high rate of complications, MT continues to be plausible in well-selected patients, as observed in our case report. Despite the tumor volume, the patient presented a good clinical condition and was observed to have a favorable response to the radiotherapy and chemotherapy scheme, thus he had a good prognosis for the radical procedure. The same occurred in accordance with the literature with the passage of the trachea below the innominate artery, and the protection of mediastinal vessels and tracheocutaneous Anastomosis with a PM muscle flap, which was an essential factor for the absence of vascular complications (Orringer, 1992; Berthet et al., 2014; Conti et al., 2010).

To conclude, MT is an exceptional procedure due to the morbidity and risks it presents and should be performed only in patients with an excellent general condition. It may also be indicated as a palliative treatment in selected cases. This report shows the feasibility of its use associated with a laryngopharyngoesophagectomy for a locally advanced tumor. The surgeon should be aware of the possibility of esophagectomy, with several kinds of gastrointestinal transit reconstruction.

REFERENCES


