## CASI CLINICI, STUDI, TECNICHE NUOVE CASE REPORT, STUDIES, NEW TECHNIQUES

# Tracheal necrosis, oesophageal fistula: unusual complications of thyroidectomy. Report of two case and literature review.



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## Tracheal necrosis, oesophageal fistula: unusual complications of thyroidectomy. Report of two case and literature review.

BACKGROUND: Thyroidectomy is considered a low-risk operation. The authors report a case of tracheal necrosis after total thyroidectomy for multinodular goiter with bilateral adenomas, and a case of oesophageal fistula after total thyroidectomy for papillary cancer.

METHODS AND RESULTS: The patient with tracheal perforation was treated by a non operative management after clinical stabilization. The patient with oesophageal perforation underwent surgical treatment consisting of neck drain placement. Both patients are alive after 12 months of follow-up, although the patient who had surgery for papillary cancer of the thyroid gland was found to have multiple diffuse liver and lung metastases.

CONCLUSIONS: Thyroidectomy is a safe surgical procedure, but in some patients major complications may arise. In cases of iatrogenic tracheal or oesophageal perforation, conservative non-surgical or conservative surgical treatment, in specialized centers, results in a favourable outcome. The authors discuss the risk factors and management of these two rare complications.

KEY WORDS: Esophageal fistula, Esophageal perforation, Pneumomediastinum, Thyroidectomy, Tracheal necrosis.

### Introduction

Thyroidectomy is considered to be a safe surgical procedure associated with very low mortality and morbidity rates. Transient or permanent hypoparathyroidism, and transient or permanent recurrent laryngeal nerve damage are the most serious complications of the procedure. Less common complications include hemorrage, infections, or injuries to the brachial plexus due to malpositioning of the patient on the operating table. Tracheal necrosis with pneumomediastinum and oesophageal perforation are both very rare complications and few cases of either are reported in the literature. There is no consensus regarding the benefits of surgical versus conservative non-surgical treatment for patients suffering from this complication and various factors must be considered. In patients with oesophageal perforation immediate surgical treatment associated with wide-spectrum antibiotic therapy and total parenteral or enteral nutrition is recommended.

The authors present two cases of these rare complications, and after a review of literature discuss the pathogenetic factors involved and the treatment of the lesions.

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#### Case reports

## Case n. 1

In July 2008, a 65-year-old man underwent elective total thyroidectomy for multinodular goiter with bilateral adenomas. He was a non-smoker, with no significant medical history. Preoperative physical examination and laboratory tests did not reveal any significant abnormalities. There was no specific problems during intubation with a 7.5 mm tracheal tube. An uneventful total thyroidectomy was performed by an experienced senior surgeon (> 1000 thyroidectomy performed) and a routine air leak test performed at the end of the operation was negative. The patient was extubated uneventfully and bilateral vocal cord movements were present. His postoperative course was unremarkable and he was discharged on postoperative day 3. There was no evidence of wound infection or abnormal swelling of the operative site. On post operative day 15, the patient felt a sudden pop in his neck following an episode of vigorous coughing and experienced diffuse bilateral swelling of the neck with rapid progression. In an outpatient setting a multifenestrated, 22 gauge neck drain was positioned through the previous cervicotomy, and air leaked out. The patient was not in respiratory distress, his pulse and blood pressure were stable, and there were no distended neck veins, cyanosis, or other signs of circulatory failure and airway obstruction. A chest X-ray showed subcutaneous emphysema in the neck but no pneumothorax. A computed tomography (CT) scan revealed pneumomediastinum, possibly a consequence of tracheal rupture despite the fact that no defect in the trachea was seen (Fig. 1). On bronchoscopy a 1.5 mm x 1.5 mm perforation was found on the right side of the anterolateral tracheal wall at distance of 4.5 cm above the carina (Fig. 2). Because the respiratory condition of the patient was not critical and the tracheal defect was small, the authors opted for conservative treatment (bed rest, antibiotics, and cough suppressants) rather than surgical repair. After 3 days the cervical drain was removed. The subcutaneous cervical emphysema and the pneumomediastinum gradually resolved. Spontaneous air absorption was complete with-



Fig. 1: Tracheal necrosis: CT scan showing pneumomediastinum.



Fig. 2: Tracheal necrosis: bronchoscopy - a perforation on the right side of the anterolateral tracheal wall was present.



Fig. 3: Tracheal necrosis: CT scan showing spontaneous air absorption after 2 weeks.



Fig. 4: Tracheal necrosis: complete closure of the tracheal defect after two weeks.

in 2 weeks as confirmed by clinical and CT scan evaluations (Fig. 3). Endoscopic examination showed complete closure of the tracheal defect (Fig. 4). An air leak test was performed at the site of the initial injury and no escape of air was noted. At one-year follow-up, the patient was well with no symptoms of tracheal damage.

#### Case N. 2

In September 2009 a 67-year-old man, underwent elective total thyroidectomy for papillary thyroid cancer 4 cm in diameter in the right lobe of thyroid gland. He was on Dicumarol therapy for recently diagnosed atrial fibrillation, and had no other significant medical history. The following preoperative diagnostic procedures were performed: high resolution neck ultrasound associated with fine needle biopsy (FNB) (positive for papillary cancer), ear, nose and throat examination that revealed right vocal cord paralysis, a chest X-ray, and an echocardiogram that showed an aneurysmal dilatation of the ascending aorta 4.5 cm in diameter. The patient was hyperthyroid and with medical therapy became euthyroid before surgery. Preoperative serum thyreoglobulin was >1500 ng/ml. After intubation with an 8 mm tracheal tube the tracheal cuff was inflated until no-leak ventilation was achieved. The right lobe of the thyroid gland was found to be fixed to the overlying strap muscles, and the underlying trachea, and to have a posterior nodule about 4 cm in diameter, densely adherent to the right cricopharyngeus muscle and oesophagus. Total thyroidectomy with en bloc removal of the posterior nodule (R0 resection), was performed by the same experienced senior surgeon (case 1). A perioperative oesophagoscopy showed that the oesophageal mucosa was intact. Since preoperative diagnostic procedures had not identified any lymphadenopathy, and surgical exploration of the relevant lymph nodes was negative, prophylactic lymphadenectomy was not performed. The patient was extubated uneventfully and left vocal cord movements were present. Definitive histological examination identified a papillary thyroid carcinoma (pT2). On postoperative day 6, a clinically evident collection of serosanguineous fluid and air was found in the neck.



Fig. 5: Oesophageal perforation: gastrografin swallow reaveling an oesophageal fistula at pharyngo-oesophageal junction.

Investigation with a gastrografin swallow revealed an oesophageal fistula at the level of the right lateral pharyngo-esophageal junction (Fig. 5). The patient underwent reoperation and a neck drain was positioned. He was given total parenteral nutrition (TPN) and broad-spectrum antibiotic therapy with Tazocin<sup>®</sup> (amoxicilline plus clavulanic acid), 2 g three times a day for seven days. After 15 days a new gastrographin swallow showed that the fistula had healed and oral feeding was resumed. A follow-up total body CT scan revealed locoregional recurrence in the right side of the neck, and multiple diffuse liver and lung metastases. The patient underwent metabolic radiotherapy with I-131 (5500 MBq; 150 mCi).

#### Discussion

When thyroid surgery is performed by an expert surgeon mortality is very rare and the morbidity rate is low, about 5% <sup>1,2</sup>. Due to close collaboration between endocrinologists and surgeons, the increasing availability of specialized treatment centers, and a more frequent use of FNB, the number of indications for thyroid surgery has increased over the years and more patients with early stage malignant tumors have undergone surgery.

The most greatly feared surgical complications are transient or permanent lesions of the parathyroid glands and the recurrent laryngeal nerves. Hypocalcemia due to either type of injury to the parathyroid glands is the most common complication and is reported up to 63% of cases <sup>3</sup>, most often after total thyroidectomy or any surgery for thyroid cancer. Lesions of the recurrent laryngeal nerve, whether transient or permanent, are a severe complication and often the cause of legal proceedings and great frustration for both patient and surgeon. The incidence of complications reported after total thyroidectomy and conservative bilateral procedures is the same.

Hemorrhage is the third most important complication but is rare with an incidence of 1.2%<sup>3</sup>, and typically associated with Graves disease. Other extremely uncommon complications are infection and lesions of the brachial plexus due to malpositioning of the patient on the operating table.

The two rare complications presented, tracheal necrosis with subcutaneous emphysema in the neck and pneumomediastinum after total thyroidectomy for multinodular goiter, and oesophageal fistula after total thyroidectomy for a large papillary carcinoma, occurred in patients the authors treated in a regional center for endocrine surgery where 250-300 thyroidectomies are performed each year.

In the first case reported by the authors, it seemed the perforation occurred when an eschar that formed due to improper use of electrocautery was cast off. It should be noted that there were no particular difficulties in mobilizing the trachea, and that the surgeon was an experienced endocrine surgeon who had performed >1000 thyroidectomies but not yet observed this complication. Due to the small size of the perforation, <2mm, the paucity of symptoms, and the respiratory stability of the patient, conservative treatment was adopted, and the patient recovered within 2 weeks. This agrees with the reports in the literature of similar cases which show that when a tracheal perforation is <2cm non operative management is possible and leads to complete recovery.

The most common causes of tracheal perforation after surgery are improper use of electrocautery, neck infection, evascularisation of the tracheal wall, prolonged and improper intubation, and type III tracheomalacia. The incidence of tracheal perforation after thyroidectomy is extremely low, although it is sometimes underestimated because it is not always reported; in a review article covering 11,917 cases in a period of 45 years, J.E.Gosnell reports that the incidence is 0.06%<sup>4</sup>. The main risk factors are considered to be female sex, thyrotoxicosis, inadequate size of the tracheal tube, excessive pressure from the tracheal cuff, extensive and improper use of electrocautery, hematoma and associated infection, and frequent bouts of severe coughing 5-8. In most cases the initial lesion is iatrogenic and perforation occurs after some time has elapsed, (in the case reported by the authors it occurred after 2 weeks), and is most often due to a thermic lesion, often of the anterolateral wall of the trachea. Immediate perforation is rarer, and normally recognized and repaired during surgery. After a variable period of time the patient develops subcutaneous emphysema in the neck as well as pneumomediastinum. The patient's clinical picture differs depending on the size of the lesion. Cervical abscess, mediastinitis, pneumothorax, tension pneumomediastinum, and cardiac tamponade, all of which require emergency surgery, are not systemic conditions, and only rarely cause death 9. In these cases direct repair, consisting of tracheostomy or even tracheal resection and anastomosis if there is extensive necrosis, is advisable and long-term results vary <sup>5,6,10</sup>. When the lesion is < 2 cm in diameter, conservative treatment with clinical resolution and good long-term results is possible, and is also reported in cases of traumatic tracheal perforations of the same size 11,12.

In the second case reported, a fistula in the right lateral wall of the cervical esophagus formed after the removal of a large papillary carcinoma with local extension and tenacious adhesions to the right cricopharyngeus muscle and the right lateral wall of the esophagus. When the operation was over an oesophagoscopy was performed to rule out injury to the oesophagus, and the oesophageal mucosa was shown to be intact.

It seems, therefore, that in this case, as well as the previous one, the perforation was the result of surgical manoeuvres, and that it was devascularization of the oesophageal wall that led to the development of the fistula on postoperative day 6. The possibility that the fistula arose due to neoplastic infiltration can be excluded

because of the patient's rapid recovery after cervical drainage and TPN. In most cases like this, placement of a cervical drain and administration of TPN lead to recovery and probably constitute the treatment of choice when the perforation is small. Mechanical or manual suturing which is required if the perforation is large, is associated with a high risk of dehiscence and a high incidence of oesophageal stenosis <sup>13</sup>.

Perforation of the oesophagus is characterized by a mortality rate of 5.5-20% and high morbidity rates that vary depending on the pathogenesis of the defect, its dimensions, and its location, (perforation of the cervical oesophagus is the less dangerous than that of the mediastinal or abdominal oesophagus) 14-17. Perforation of the cervical oesophagus has been reported to occur most commonly after orotracheal intubation, dilatation of stenosis, surgery of the oesophagus, neck, or chest, less frequently after positioning of a laryngeal mask airway or a nasogastric tube, and in rare cases after thyroidectomy, especially reoperations <sup>18,19</sup>. The treatment protocols followed for esophageal perforation range from conservative to surgical management 20,21 which involves procedures that are more complex and associated with significant mortality and morbidity. Choice of treatment should be based on various factors; the cause and dimensions of the perforation, the general condition, age, comorbidities, and the nutritional status of the patient. In cases of cervical oesophageal perforation conservative treatment is probably the most appropriate. Mortality and morbidity rates are low and long-term results are good.

Given that fistulization of the cervical esophagus is, per se, a low-risk condition, the treatment protocol requires, in most cases, drain placement and administration of parenteral nutrition as well as broad- spectrum antibiotics. If conservative treatment fails, various solutions are possibile; suturing of the oesophageal wall, repair with a muscle flap, insertion of a prosthesis, or resection and jejunal or colon interposition. The method chosen depends on various factors, and resection, which is associated with significant mortality and morbidity, is indicated mainly to control sepsis.

In cases of differentiated thyroid carcinoma with suspected locoregional infiltration, preoperative diagnostic testing should include oesophagoscopy and possibly echoendoscopy in order to exclude involvement of the oesophagus. However, in the case the authors present, detailed preoperative staging would not have led to changes in the surgical treatment which consisted of total thyroidectomy followed by oesophagoscopy to establish whether the oesophageal mucosa was intact.

## Conclusions

In conclusion, the complications presented by the authors are potentially lethal but can easily be treated conservatively. In such cases the size of the perforation in the trachea and the esophagus is a prognostic factor that has a major impact on the results of treatment. Total thyroidectomy remains a low-risk procedure. The rare complications described can be effectively treated in multi-disciplinary specialized centers.

#### Riassunto

La tiroidectomia è considerata un intervento chirurgico con scarse complicanze intra e peri operatorie se eseguita da chirurghi esperti.

Ipocalcemia, disfonia transitoria o permanente, emorragia, infezione del sito chirurgico rappresentano le complicanze più frequenti. La necrosi tracheale e la fistola esofagea, descritte dagli AA, rappresentano evenienze post operatorie molto rare che richiedono un trattamento chirurgico o medico immediato in Centri specializzati.

Nel case report-1 descritto dagli AA, il pz, sottoposto ad intervento di tiroidectomia totale per adenoma tiroideo bilaterale con decorso post operatorio privo di complicanze, a distanza di due settimane, in seguito ad improvviso accesso di tosse, presentava rapido e progressivo rigonfiamento del collo. Sottoposto ad esami strumentali presso centro specializzato, veniva diagnosticato imponente enfisema sottocutaneo con pneumomediastino da probabile rottura tracheale. La diagnosi era poi confermata da esame broncoscopico che evidenziava perforazione della parete anterolaterale della trachea. Si posizionava drenaggio con progressiva risoluzione del quadro clinico.

Le cause più comuni della perforazione tracheale, dopo chirurgia tiroidea, sono le infezioni del tratto cervicale, lesioni vascolari della parete tracheale, uso improprio dell'elettrocoagulatore e difficoltose e prolungate intubazioni oro-tracheali per l'anestesia. La perforazione immediata da lesione accidentale intraoperatoria è molto rara e nella maggior parte dei casi viene riparata nel corso dell'intervento chirurgico. Molto più frequente, invece, è la necrosi tardiva da danno vascolare che si verifica dopo un variabile periodo di tempo dalla tiroidectomia. Il quadro clinico dipende dalle dimensioni della lesione. Ascesso cervicale, mediastinite, pneumotorace e/o pneumomediastino, tamponamento cardiaco sono condizioni gravi che richiedono interventi in emergenza, ma raramente sono causa di morte. La scelta del trattamento, conservativo vs chirurgico, dipende dalle dimensioni della lesione.

Nel case report-2 gli AA descrivono un caso di fistola esofagea in paziente sottoposto a tiroidectomia totale per cancro capillifero della tiroide. In sesta giornata post operatoria il paziente mostrava una tumefazione in regione anteriore del collo con fuoriuscita di materiale corpuscolato dalla ferita chirurgica. Veniva praticato RX esofago con gastrographin e tc collo e mediastino che mettevano in evidenza soluzione di continuo della parete laterale dell'esofago associata a enfisema sottocutaneo che si estendeva fino allo stretto toracico superiore. Si procedeva a revisione chirurgica e a posizionamento di drenaggio; a sette giorni dal reintervento il paziente veniva dimesso dopo aver praticato Rx di controllo che risultava negativo per stravasi extraluminali.

La perforazione esofagea è caratterizzata da elevata morbidità che dipende dalla patogenesi, dalle dimensioni e dalla localizzazione della lesione. Si può verificare in seguito a intubazione oro tracheale, dilatazione di stenosi, chirurgia di esofago, collo e torace e raramente in seguito a tiroidectomia specialmente nei reinterventi. La scelta del trattamento dipende dalla sede della lesione e può essere conservativo o chirurgico, con procedure spesso molto complesse e associate a elevata morbilità e mortalità.

In conclusione, le complicanze presentate dagli AA sono potenzialmente letali ma posssono essere facilmente trattate in modo conservativo in centri specializzati multidisciplinari.

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