Hypopharyngeal fistula complicating difficult thyroidectomy for invasive papillary cancer

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ABSTRACT

Thyroidectomy is associated with certain known complications, including recurrent laryngeal or superior laryngeal nerve injury and hypocalcaemia due to hypoparathyroidism. Other much rarer complications include oesophageal injury with oesophageal fistula formation. In this report, we describe the clinical course of a patient with an invasive papillary thyroid carcinoma who underwent total thyroidectomy and developed hypopharyngeal fistula in the immediate postoperative period, a complication that has never been reported previously following thyroidectomy. Under conservative treatment, the fistula closed within two weeks and the patient was referred, in good condition, to the oncologist for completion of therapy.

KEYWORDS

Thyroidectomy – Hypopharyngeal fistula

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Papillary carcinoma is the most common form of thyroid cancer, comprising about 60% of cases, followed by follicular cancer with a rate of 20%.¹ The remaining cases are distributed between medullary and anaplastic cancers as well as lymphoma. While thyroidectomy is the treatment of choice for papillary cancer, there is controversy regarding its extent, from lobectomy to total thyroidectomy. Known surgical complications of thyroidectomy include injury to the recurrent or external laryngeal nerves, leading to voice changes and swallowing abnormalities, as well as injury to the parathyroid glands, leading to hypocalcaemia.

Similar to spontaneous oesophageal perforation, described by Herman Boerhaave almost three decades ago,² spontaneous pharyngeal perforation has also been reported.³ More commonly, however, this rare condition is brought about by a variety of causes, not including thyroidectomy.

Case History

A 38-year-old woman, with no past medical history, presented with a neck mass that had been noticed 5 years earlier. There was no history of thyroid dysfunction or obstructive symptoms and the systemic review was unremarkable. On examination, her vital signs were normal and there was a marked enlargement of the right thyroid lobe, notably in an upward direction. There were no palpable lymph nodes and no signs of thyroid dysfunction. Her blood count was normal, as well as liver, renal and thyroid functions.

Ultrasonography showed a large complex nodule, measuring 4 cm × 5 cm, occupying the entire right thyroid lobe, with areas of calcification. A smaller hypoechoic nodule

was also found in the left side. No abnormal cervical lymph nodes were noted. Ultrasonography guided fine needle aspiration (FNA) of the right nodule revealed papillary carcinoma, so the patient was consented for total thyroidectomy.

During surgery, the enlarged, firm right thyroid lobe was densely adherent to the surrounding structures including the strap muscles and trachea, which was also pushed to the left. Unusually tedious and sharp dissection was needed to free the lobe and after completion, the abnormally high level of its upper extent became evident (Fig 1). A left thyroid lobectomy then followed and, conversely, this procedure was simple. At the end of surgery, suction drains were inserted.

On the first postoperative day, thyroxine replacement was started and, unfortunately, hoarseness of voice due to laryngoscopically proven paralysis of the right vocal cord was noted. Additionally, turbid discharge came along the drain in postoperative day 2, which became purulent and included food particles in the following days. About that time, a Gastrografin® (Bracco, Princeton, NJ, US) swallow confirmed the presence of hypopharyngeal fistula on the right side (Fig 2), for which conservative treatment was started. The suction drains appeared to hinder the free egress of food debris containing discharge, so they were removed and replaced by a rubber drain, with the aim of keeping the track open (Fig 3).

The patient was kept on nil by mouth, and a nasogastric tube was inserted and used for feeding. Piperacillin/tazobactam (4g every 8 hours) and metronidazole (500mg every 8 hours) were administered intravenously, together with daily dressings and saline irrigation along the track.

TC = thyroid cartilage; CB = cricoid bone; T = trachea

Figure 1 In order to release the gland from the surrounding tethered structures, the dissection was carried out both in an upward (arrow) and outward direction, far beyond what is customary in such conditions.

Figure 3 The cut end of a Foley's catheter was used as a rubber drain to allow egress of the thick pus containing particulate material from the wounds of the original suction drains.

Figure 2 Leaking Gastrografin® high in the neck above the level of the cricoid cartilage (circle)

Under this treatment, the discharge cleared gradually and stopped two weeks later. A Gastrografin® swallow confirmed closure of the fistula and the drain was removed. Following this, the patient was referred to the oncologist for further management.

The histopathology report showed infiltrating papillary carcinoma, 3cm in its maximum dimension, on the right

side and a smaller papillary microcarcinoma (4mm) on the left. The right-sided tumour elicited a desmoplastic reaction, and extended into the surrounding fibromuscular and fatty tissues. No lymph node involvement was found. Two weeks after discharge, the patient appeared in the outpatient clinic in good health and she was scheduled by the oncologist for whole body imaging in the following days.

Discussion

The occurrence of oesophageal or pharyngeal fistula following thyroidectomy is an almost unknown complication. A MEDLINE® search was conducted using the terms 'pharyngeal fistula' and 'thyroidectomy', 'hypopharyngeal fistula' and 'thyroidectomy', 'pharyngeal fistula' and 'thyroid', 'hypopharyngeal fistula' and 'thyroid', first in the title and abstract, then in all fields and no pertinent results were retrieved. Repeating the same search after replacing the word 'oesophageal' for 'hypopharyngeal' yielded only two pertinent reports.^{4,5} This reflects either the extreme rarity of the condition or the natural reluctance of physicians to expose their own unusual complications.

In our patient, the diagnosis of papillary carcinoma was established by ultrasonography guided FNA preoperatively and despite the operative impression of apparent local extension into the surrounding tissues, no metastatic deposits were found in the retrieved lymph nodes and the extensive tethering of the right lobe was explained by the histopathologically demonstrated desmoplastic reaction surrounding the tumour. This tethering, in addition to the unusual upwards extension of the tumour in the neck, were crucial factors in bringing about this complication.

Generally, the clinical picture, management and prognosis of pharyngo-oesophageal perforation is much influenced by the mechanism of injury and its location as well as the time lapse between the injury and the institution of

treatment. Clinically, the patient may present with neck pain, dysphagia, subcutaneous emphysema, fever, leucocytosis and systemic signs of sepsis.^{6,7}

When suspected, x-ray of the neck in a lateral projection will demonstrate retropharyngeal air when present and a Gastrografin® swallow or computed tomography with oral contrast will clarify the anatomical site of the leak.^{8,9} Additionally, endoscopic examination may reveal the tear, its site and size as well as the presence of oedema or haematoma in the region.¹⁰ However, it may fail to show the abnormality¹¹ if vigilance is not exercised, as in our case.

If not treated properly and with the persistence of infection in the region, certain potentially serious complications may arise, including mediastinitis¹² and cervical abscess.^{13,14} More seriously, carotid pseudoaneurysm may form,¹⁵ which may rupture subsequently with a fatal outcome. Fortunately, no serious complication occurred in our patient, who remained well throughout her hospital stay.

Contrary to oesophageal perforation, no guidelines are present in the literature regarding the management of hypopharyngeal perforation. Owing to this lack of knowledge, and as the hypopharynx and cervical oesophagus are intimately related, the management principles of traumatic fistula in these two locations may be similar and, theoretically, the recommendations applied to cervical oesophageal perforations may be extrapolated to those of the pharynx. At this juncture, emphasis on early diagnosis and management have been stressed,¹⁶ and depending on the clinical scenario, the size of perforation and its timing, a conservative or surgical approach may be chosen. The conservative approach includes keeping the patient on nil by mouth and institution of nasogastric feeding with the administration of intravenous antibiotics.^{9,17–19} On the other hand, if chosen, surgical repair may be carried out through the transoral¹⁴ or transcervical routes and is always accompanied by drainage.^{8,20}

Pharyngocutaneous fistula is a known complication of total laryngectomy, performed for laryngeal or hypopharyngeal carcinoma. Even in these difficult situations with obvious adverse healing factors, spontaneous healing is likely in many cases under conservative measures. Failing this, surgical intervention is called for.¹⁵

In the patient presented here, the history of difficult thyroidectomy, followed by the appearance of turbid discharge along the drain, raised the suspicion of fistula formation, before it declared itself in the following days, when the diagnosis was confirmed with a Gastrografin® swallow. Drainage was provided with saline irrigation of the thyroid bed and intravenous antibiotics were administered. Once the discharge stopped and the Gastrografin® study confirmed the closure of the fistula, the drain was removed. Meanwhile, the patient was kept on nil by mouth and her nutritional status was maintained through nasogastric feeding. Surprisingly, there was no evidence of sepsis at any stage.

Conclusions

To our knowledge, this is the first report on hypopharyngeal fistula complicating difficult thyroidectomy. The condition is potentially dangerous but, fortunately, as in our case, it could be managed conservatively. Early recognition and institution of treatment with keeping the patient on nil by mouth, nasogastric feeding and administration of intravenous antibiotics, may end in success.

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