Case Report

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Neck abscess as the initial manifestation of pharyngeal cancer

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Introduction: Pharyngeal carcinoma and neck abscesses are not uncommon, but neck infection as the initial presentation of primary head and neck cancer is rare, and these patients risk potential misdiagnosis.

Case presentation: We report our experience with two patients who had an unusual first presentation of pharyngeal cancer – a deep neck infection and a relapsing abscess of the thyroid gland.

Conclusion: Our case series demonstrates the need for meticulous physical examination of the head and neck in all patients presenting with neck abscess. It is important to consider malignant tumour as a possible cause of deep neck infection and an abscess of the thyroid gland. Diagnosis and subsequent treatment is delayed if malignancy is not clinically suspected.

Keywords: Deep neck infection; pharyngeal cancer; thyroid gland abscess.

Introduction

Carcinoma of the oropharynx and hypopharynx are relatively common malignancies of the upper aerodigestive tract but bear the worst prognosis of all head and neck tumours. Smoking and alcohol consumption are the major risk factors in the development of pharyngeal cancer. Recently, human papillomavirus, especially types 16 and 18, has been shown to be responsible for 50 % of oropharyngeal squamous cell carcinomas (Döbrossy, 2005; Amar *et al.*, 2013).

Deep neck infection (DNI) is defined as infection in the potential spaces and fascial planes of the neck. This infection usually invades from other primary sites, most often within the pharynx or oral cavity. The predisposing risk factors for DNI include diabetes mellitus, drug abuse, a congenital neck cyst, age, poor oral hygiene and smoking. Immune suppression diseases, such as chronic renal disease and hepatic disease, and chronic steroid therapy for autoimmune disease can also be risk factors for the development of DNI (Liu *et al.*, 2013; Sakarya *et al.*, 2015). Although the incidence of DNI has decreased significantly with the wide use of antibiotics, this condition is still common and presents a constant challenge, as it may lead to lethal complications such as acute respiratory failure, cervical necrotizing fasciitis, descending necrotizing mediastinitis, jugular vein thrombosis, pericarditis, pleural

empyema, arterial erosion, septic shock, dural sinus thrombosis and intracranial abscess (Sakarya *et al.*, 2015; Yang *et al.*, 2015).

We report our experience with two patients who had an unusual first presentation of pharyngeal cancer – a DNI and a relapsing abscess of the thyroid gland. The aim of this study is to raise awareness about this potential pitfall in the management of deep neck abscesses, especially the need of proper physical examination and computerized tomography (CT) investigation of the head and neck in all patients with a neck abscess.

Case report 1

A 65-year-old man, a non-smoker, was referred to the Department of Otorhinolaryngology, Head and Neck Surgery, Comenius University, Jessenius Faculty of Medicine, University Hospital in Martin, Slovakia, from a local hospital where he was treated for a painful left-sided neck mass with a fever of 4 days' duration. He suffered from chronic hypertension, chronic ischaemic heart disease and insulin-dependent diabetes mellitus. An initial ear, nose and throat (ENT) examination showed mild oedema of the inferior part of the left tonsil and a tender, painful 4 cm mass in the left level II region of the neck. Empirical parenteral antibiotic therapy using a combination of clindamycin and metronidazole was administered. A CT scan showed an abscess collection in the left level IIa neck region (Fig. 1).

Abbreviations: AST, acute suppurative thyroiditis; CT, computed tomography; DNI, deep neck infection; ENT, ear, nose and throat.

Fig. 1. CT scan of the neck (axial section on the left, coronal section on the right) showing the abscess collection and necrosis of the lymph node in the left submandibular area (arrows).

The patient was transferred to our department and was immediately operated on. Revision of the affected cervical space under general anaesthesia was made. Abscess collection in the submandibular area that had spread to the omohyoid muscle was identified and drained. Due to the oedema of the supraglottic area, tracheostomy was performed at the end of the operation. There was no bacterial growth on culture. Systemic combined intravenous antibiotic (clindamycin and metronidazole) and antifungal therapy together with symptomatic anti-oedematous therapy was administered. After this treatment, the clinical condition of the patient gradually improved, inflammatory activity decreased and follow-up ultrasonography showed no fluid collection in the cervical space. An ENT examination showed regression of oedema of the supraglottic structures, but asymmetry of the left lingual tonsil persisted. Therefore, the patient was re-examined under general anaesthesia 6 days after drainage of the neck abscess. Rigid hypopharyngolaryngoscopy revealed a mild mucosal irregularity in the left part of the lingual radix, which showed a poorly differentiated squamous cell carcinoma on biopsy. The patient was staged as stage III (T1N1M0) oropharyngeal carcinoma. The patient subsequently underwent curative chemoradiotherapy.

Case report 2

A previously healthy 62-year-old male, a smoker, suffered from mild dysphagia and painful swelling of the neck with fever of 5 days' duration. An outside CT scan showed a complex of solid and cystic masses in the right

lobe of the thyroid gland with peripheral enhancement. The patient was referred to our department. An ENT examination showed a smooth oedema of the right aryepiglottic fold, retention of saliva in the right pyriform sinus, slight impaired movement of the right vocal cord and a 3 cm tender mass at the right front part of the neck. Levels of thyroid hormones were normal. Empirical parenteral antibiotic therapy (aminopenicillin + clavulanic acid and metronidazole) was administered. The day after admission to our department, the patient was operated on under general anaesthesia. A fluid collection of $4 \times 3 \times 2$ cm in the cranial part of the right lobe was identified and evacuation of the abscess was carried out. Subsequently, a hemithyroidectomy of the right lobe of the thyroid gland was performed. Histological examination of the thyroid gland showed only inflammatory changes, with no atypical or malignant cells. Microbiological investigation from the abscess identified polymicrobial growth of bacteria (*Streptococcus agalactiae* and *Peptostreptococcus*, both sensitive to aminopenicillin). Peroral antibiotic therapy was continued for 16 days following surgery. Follow-up ultrasonography of the neck showed no fluid collection, and 12 days after surgery the patient was released to outpatient care.

Two weeks after discharge from the hospital (26 days after surgery), the patient was admitted to our department for dysphagia, hoarseness and a painful right-sided neck mass of 2 days' duration. The patient was afebrile. An ENT examination showed oedema of the right lateral wall of the hypopharynx and right aryepiglottic fold, and reduced movement of the right vocal cord. A painful

fluctuant mass at the right anterior part of the neck extending from the submental region to the thyroid gland was present. The rest of head and neck examination was normal. The abscess cavity was immediately drained surgically under local anaesthesia. The area was irrigated and a drain was placed in the incision. Aerobic and anaerobic bacteria (*Streptococcus agalactiae*, *Peptostreptococcus* and *Enterobacter* species) were isolated from the abscess. Antibiotic therapy (aminopenicillin + clavulanic acid and ciprofloxacin) was given parenterally. Despite the drainage of the relapsing abscess at the right anterior part of the neck and parenteral combined antibiotic and anti-oedematous treatment, the condition did not regress. The oedema of the right hypopharyngolaryngeal area as well as reduced mobility of the right vocal cord persisted. A CT scan performed 4 days after drainage showed an abscess collection of $30 \times 17 \times 28$ mm in the area of the previous operation. A new fluid collection of $16 \times 14 \times 38$ mm was identified in the right posterior part of the larynx. The right pyriform sinus could not be differentiated, with its lateral wall being a part of the abscess cavity (Fig. 2). The patient was subsequently operated on under general anaesthesia. Revision of the right cervical spaces with evacuation and drainage of abscess collection was made. Panendoscopy showed a tumorous infiltrate in the right pyriform sinus. Biopsy of the lesion revealed invasive squamous cell carcinoma. The patient was staged as stage III (T3N0M0) hypopharyngeal cancer. The patient subsequently underwent curative chemoradiotherapy.

Discussion

Primary head and neck cancer and neck abscesses are not uncommon, but DNI as the initial presentation of primary head and neck cancer is rare, and these patients risk potential misdiagnosis. The incidence of head and neck cancers in patients with an initial presentation of DNI is unclear and may be underestimated (Wang *et al.*, 2006; Lin *et al.*, 2012). In a retrospective study (K. Mlcakova, P. Hanzel & A. Hajtman, unpublished data), 58 patients treated for DNI from 2007 to 2013 in the Department of Otorhinolaryngology, Head and Neck Surgery, Comenius University, Jessenius Faculty of Medicine, University Hospital in Martin, Slovakia, were analysed. In 16 patients (28 %), the origin of DNI remained unclear. The most common known cause was pharyngeal (28 %) and dental (20 %) infection. Of the 58 patients with DNI, only in two patients (3 %) was DNI the first manifestation of malignant tumour. Similar results for DNI origin have been observed by other authors (Ridder *et al.*, 2005; Eftekharian *et al.*, 2009; Bakir *et al.*, 2012). The incidence of head and neck cancer initially manifested as DNI was found to be increased in patients aged over 40 years (Lin *et al.*, 2012).

Head and neck squamous cell carcinomas often present with cervical metastases. Metastases from certain primary head and neck sites, such as the nasopharynx, tonsil and thyroid, may give rise to cystic nodal metastases. These cystic metastases may become infected and present as a

Fig. 2. CT scan of the neck (axial section on the left, coronal section on the right) showing a relapsing abscess of the thyroid gland. The right pyriform sinus could not be differentiated, with its lateral wall being a part of the abscess cavity (arrows).

neck abscess. Similarly, other metastases may undergo central necrosis and may appear as suppurative lymphadenitis on imaging. Direct tumour extension that undergoes necrosis can also cause DNI. Therefore, the initial diagnosis of a neck abscess in these cases is entirely reasonable and the malignancy remains undetected (Daramola *et al.*, 2009; Wang *et al.*, 2010; Soon *et al.*, 2012).

The bacteriology of DNI is polymicrobial, including both aerobic and anaerobic bacteria. In a retrospective study of 634 patients with DNI by Celakovsky *et al.* (2015), aerobic and anaerobic bacteria were isolated from 39 % and 10 % of cultures, respectively. Both aerobic and anaerobic bacteria were isolated from 32 % of cultures. This culture rate, especially for anaerobic bacteria, could be underestimated owing to their fastidious nature and the difficulty of isolating these organisms. *Streptococcus pyogenes* and *Staphylococcus aureus* were the most commonly identified aerobic bacteria. The most common isolated anaerobic bacteria were *Peptostreptococcus* spp., *Prevotella* spp. and *Propionibacterium* spp. *Streptococcus agalactiae* and *Peptostreptococcus* spp. were isolated from a patient with a relapsing abscess of the thyroid gland. No bacteria were identified in a patient with a DNI. No organism was isolated from 20–30 % of pus samples (Lee & Kanagalingam, 2011; Celakovsky *et al.*, 2015). This high proportion of no growth was probably due to the prompt use of high-dose antimicrobials early in the course of the disease. Empirical antibiotic therapy was also administered in both patients in our case series. These results showed that, although culture-guided antimicrobial therapy is advocated, empirical antibiotics play a critical role in alleviating the clinical course of the disease.

Acute suppurative thyroiditis (AST) is a rare clinical event and an uncommon form of thyroiditis. The progression of the condition to a thyroid abscess is equally unusual. Both AST and a thyroid abscess represent 0.1–0.7 % of thyroid lesions managed surgically (Tien *et al.*, 2007; Adeyemo *et al.*, 2010). A tender thyroid lesion is the hallmark of AST, but other causes of a tender thyroid gland include de Quervain's thyroiditis (the commonest cause of a painful thyroid gland), acute haemorrhage into a cyst or thyroid nodule, a rapidly enlarging thyroid carcinoma or radiation thyroiditis (Rohondia *et al.*, 1995).

The thyroid gland possesses some characteristics that help to make a thyroid abscess an uncommon clinical event. The thyroid gland is relatively resistant to infection due to its encapsulation, its secluded anatomical position, an iodine-rich environment, extensive lymphatic drainage, and good blood flow from the bilateral anastomosing superior and inferior arteries (Adeyemo *et al.*, 2010; Bravo & Grayev, 2011). These provide protection by hindering the invasion of bacteria and their subsequent growth. Haematogenous spread from a distal site of infection is believed to be a common cause of thyroid infection; however, the exact infectious source or pathway is frequently unknown. Congenital thyroid gland pathology

such as a pyriform sinus fistula resulting from the third or fourth branchial pouch can also lead to AST (Rohondia *et al.*, 1995; Ilyin *et al.*, 2007). These anomalies have multiple presentations, including thyroid abscesses, recurrent sinuses, neck cellulitis and compressive symptoms, including dysphagia and odynophagia. A pyriform sinus fistula often becomes symptomatic during childhood and adolescent years, and thus it is important to consider this anomaly in a young patient with such a presentation (Sai Prasad *et al.*, 2007). Other causes of AST and an abscess of the thyroid gland include trauma such as fine-needle aspiration and foreign bodies. Another important risk factor for the development of suppurative thyroiditis and thyroid abscess is a decreased immune status. There is evidence that the incidence of thyroid abscesses may be increasing, particularly in the setting of human immunodeficiency virus infection. Other predisposing factors include multinodular goiter, as well as autoimmune thyroiditis and thyroid cancer (Jacobs *et al.*, 2003; Adeyemo *et al.*, 2010; Bravo & Grayev, 2011). In our patient with a relapsing thyroid abscess, the infection spread from penetration through the pyriform sinus.

Interestingly, most patients suffering from suppurative thyroiditis remain euthyroid during the course of the disease. Yu *et al.* (1998) reviewed the thyroid function tests in 95 cases of suppurative thyroiditis and 83.1 % were euthyroid, including euthyroid sick syndrome. These results were also confirmed in our patient, where the levels of thyroid hormones were normal.

A painful enlarging mass of the neck was the dominant clinical symptom in our cases. A CT scan together with elevated inflammatory markers (leukocyte count $>10.0 \times 10^9$ cells l^{-1} , C-reactive protein >40 mg l^{-1}) aroused a suspicion of inflammatory disorder. In both cases, the primary tumour in the pharynx was not obvious by initial examination. Pharyngeal carcinoma is known to be insidious, and patients may be asymptomatic in the early stages. Compounded with excessive pooling of secretions and surrounding oedema, there was thus difficulty in diagnosing pharyngeal carcinoma in these two patients. The CT examination did not show specific signs of tumour infiltration in the pharynx, and malignancy was not suspected.

As initial treatment, revision of the cervical spaces with incision and drainage of the abscess was performed. After managing the neck abscess by surgery combined with antibiotic therapy, the oedema in the pharynx diminished but did not disappear completely. Therefore, re-examination under general anaesthesia was indicated. Panendoscopy showed a pharyngeal tumour in both patients. Subsequently, they underwent curative chemoradiotherapy.

Pharyngeal carcinoma can initially manifest as a DNI and an abscess of the thyroid gland. Therefore, meticulous physical examination including panendoscopy under general anaesthesia and CT examination of the head and neck are indicated in patients who present with a neck

abscess. In patients at high risk of head and neck squamous cell carcinoma, such as smokers, alcohol drinkers and older patients, the index of suspicion must remain high, even in the presence of an initial benign cytology.

Conclusion

Our case reports demonstrate the need for meticulous physical examination of the head and neck in all patients who present with neck abscess. It is important to consider a malignant tumour as a possible cause of DNI and an abscess of the thyroid gland. Diagnosis and subsequent treatment is delayed if malignancy is not clinically suspected.

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