An extremely rare case of large Delphian node metastasis preceding primary laryngeal cancer

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Abstract

Objectives: To present an extremely rare case of large Delphian node metastasis preceding primary laryngeal cancer.

Materials and methods: A 74-year-old male who noted a mass on the lower anterior neck and consulted our department immediately. The mass rapidly grew to 6 cm from 2 cm in diameter within 2 months after the initial presentation.

Results: Fiberoptic laryngoscopy was unremarkable. Surgical excision of the lesion showed well differentiated squamous cell carcinoma with invasion into the surrounding tissues. Postoperative radiotherapy was added. During follow-up after those treatments, thickening of the right vocal cord was observed, which gradually became more apparent. Total laryngectomy was performed 13 months after the initial operation.

Conclusion: Delphian node metastasis is included in the differential diagnosis in a case of rapidly increasing mass in the anterior neck.

Keywords: Delphian node; Laryngeal cancer; Thyroglossal duct carcinoma

1. Introduction

The Delphian or cricothyroid lymph node is stressed in many reports as an important site to which laryngeal carcinoma spreads. The node is located on the fascia above the thyroid isthmus and lies between the cricoid and thyroid cartilage. The Delphian node is also called the prelaryngeal or prercicoid node. Usually there is only one lymph node, which receives lymphatic drainage from the larynx, pyriform sinus and thyroid gland. Therefore, an enlarged Delphian node is usually due to metastasis from carcinoma of the larynx, hypopharynx or thyroid gland [1]. However, metastasis in the node is usually proven by pathology as Oslen et al. demonstrated that only one of 20 patients with node involvement had a palpable enlarged Delphian node [2]. We present an extremely rare case of large Delphian node metastasis without an apparent primary tumor at the initial diagnosis, while squamous cell carcinoma (SCC) subsequently developed in larynx.

A 74-year-old male patient noted a mass on the lower anterior neck and consulted our department immediately. Physical examination demonstrated an elastic soft nontender mass about 2 cm in diameter in the anterior midline neck at the level of the cricoid cartilage. There were no other palpable cervical masses. Fiberoptic nasopharyngoscopy and laryngoscopy were unremarkable. Ultrasonography at the initial presentation demonstrated a round solid but partly low echoic mass measuring 27 mm × 23 mm, which was not continuous with the thyroid isthmus. There were no other remarkable lesions in either thyroid lobe. Fine needle aspiration cytology performed at that time demonstrated a round solid, but partly low echoic mass measuring 27 mm × 23 mm, which was not continuous with the thyroid isthmus. There were no remarkable lesions in either thyroid lobe. Fine needle aspiration cytology performed at that time demonstrated inflammatory lesion with squamous cells, which was diagnosed as class II. There were no antibiotics administered because infection of the lesion was not obvious. The patient had been under medication for hypertension, chronic renal failure and old myocardial infarction with coronary stent insertion. Plain computed tomographic (CT) scan was...
scheduled for 1 month after the initial presentation because this case did not seem to be an emergency. CT scan demonstrated a round, low density mass with no invasion of the surrounding structures (Fig. 1A), although there was no contrast medium administered for enhancement because of the poor renal function in this patient. CT scan did not demonstrate an obvious lesion in the larynx (Fig. 1A). He was strictly followed considering the possibility of tumor rather than thyroglossal duct (TGD) cyst. The mass rapidly grew to 6 cm in diameter within 2 months after the initial presentation. Since plain CT scan did not seem to be useful for an evaluation of invasion by this lesion, magnetic resonance image (MRI) was scheduled. A MRI at that time demonstrated a relatively round mass in front of the thyroid and cricoid cartilage. The mass was iso-intense on T1-weighted image and hyper-intense on T2-weighted image (Fig. 1B). Additional fine needle aspiration cytology was not performed at this time because the patient was taking anticoagulant medication.

Fiberoptic laryngoscopy was unremarkable again at this time. After close examination of the systemic condition, surgical excision of the lesion was performed in accordance with Sistrunk’s procedure under general anesthesia 3 months after the initial presentation (Fig. 2A). The endolarynx was investigated by direct laryngoscopy prior to resection, but there were no obvious tumorous lesions. En bloc resection of the lesion with the overlying strap muscles was completed although the mass was densely adhesive to the thyroid cartilage, cricoid cartilage and especially to the cricothyroid membrane. However, there was no damage to the laryngeal frame after removal. A frozen section of the bilateral thyroid lobes adjacent to the mass did not show any sign of malignancy.

The specimen measured 6.6 cm × 5.8 cm × 5.0 cm, and consisted of cystic tumor with thickened wall and a shaggy inner lining containing necrotic caseous material. Histological examination showed well differentiated squamous cell carcinoma with invasion into the surrounding connective tissues (Fig. 2B). Almost the whole tumor consisted of squamous cell carcinoma and there was no normal epithelia of TGD remnant, dysplastic epithelia, or lesion of carcinoma in situ. Very limited exposure to the tumor cells was observed at the resected margin between the tumor and the cricothyroid membrane.
A plain CT scan 2 weeks postoperatively did not show any residual tumor at the surgical site, however, a small right cervical lymphadenopathy, measuring less than 1 cm, was detected. Fluoro-deoxy-glucose positron emission tomography (FDG-PET) performed around the same time demonstrated an accumulation in the lymph node without any other sites of accumulation including the larynx (Fig. 3A). Postoperative radiotherapy with a dose of 60 Gy was added, but there was no chemotherapy administered.

During follow-up after those treatments, thickening of the right vocal cord was observed, which gradually became more apparent. Furthermore, the mobility of the right vocal cord became impaired immediately, then finally became fixed. A biopsy specimen was taken, but the pathological diagnosis did not show any malignancy because the thickening was almost covered with normal epithelia. However, FDG-PET showed accumulations in the right vocal cord as well as the lymph node in the right neck (Fig. 3B). Total laryngectomy accompanied by right modified neck dissection after confirmation of SCC with frozen section diagnosis was performed 13 months after the initial operation. Surgical specimen demonstrated well differentiated SCC with papillary appearance extending from the lower surface of the right vocal cord (Fig. 4A). Poorly rather than well differentiated SCC was dominant in the invading portion near the thyroid cartilage. However, the upper surface of the vocal cord was widely covered with normal epithelia (Fig. 4B). Both oral and tracheal margins were microscopically free from carcinoma cells. However, metastasis to the base of the tongue was observed several months later. Furthermore, his renal dysfunction progressed rapidly at that time.

Fig. 3. FDG-PET performed after the initial operation demonstrating there was no accumulation in the larynx (A). FDG-PET performed before the second operation demonstrating accumulation in the right vocal cord (B).

Fig. 4. Sagittal view of the interior of the total laryngectomy specimen demonstrating carcinoma with a papillary appearance in the lower vocal cord showing anterior deep invasion (A). Sagittal histological view demonstrating carcinoma cells invading the thyroid cartilage (arrow heads) as well as an almost normal appearance of the upper surface of the vocal cord (U) in contrast to the lower surface of the vocal cord with partly exposed carcinoma cells (L).
2. Discussion

The name “Delphian” was first suggested by Randall to Means, who subsequently used this term for the first time in a textbook published in 1948 [3], because a positive node may predict the clinical behavior of a thyroid or laryngeal tumor, just as the oracle at Delphi foretold the future. Indeed, a positive Delphian node is a predictor of increased probability of lateral neck metastasis and decreased probability of survival, regardless of the primary site or stage, as Oslen et al. reported [2]. Similarly, patients with a positive Delphian node have a poorer prognosis than those with negative nodes as Resta et al. reported [4].

However, the Delphian node metastasis preceding a visible primary laryngeal cancer is extremely rare. Histological findings of the removed larynx demonstrated SCC mainly spreading in the lower surface of the vocal cord and subglottis. Therefore, the reason why a primary laryngeal cancer was not detected despite a large Delphian node in this case could be that a tiny occult lesion which could not be observed with regular examinations due to its location might exist. Findings on FDG-PET, CT scan and fiberoptic laryngoscopy as well as direct laryngoscopy performed at the initial surgery were in accordance with this explanation. Of course, CT scan or FDG-PET are not definitive, but complementary methods of diagnosing a tiny laryngeal lesion. Whole organ study indicating frequent involvement of the conus elasticus and anterior subglottis in cases with Delphian node metastasis can also support this hypothesis [5].

The TGD cyst is the most common congenital anomaly characterized by a swelling mostly centered in the anterior neck. This remnant rarely becomes malignant. Rapid increase in size could indicate malignant transformation of the benign TGD cyst. The vast majority of carcinomas arising in the TGD cyst are papillary thyroid adenocarcinoma. Only 10 cases of SCC arising from the TGD cyst (TGD-SCC) have been reported [6]. White and Talbert proposed strict criteria for the diagnosis of TGD-SCC [7]. These are:

1. There must be no other carcinoma in the area that could have extended or metastasized to the TGD remnant, accounting for the lesion.
2. The basic lesion must be in the classic location of a TGD remnant.
3. Thyroid tissue not in continuity with the thyroid gland must be present in the cyst wall.
4. Evidence of tumor invasion to the surrounding tissue confirms the malignant nature of the lesion.
5. Evidence of transition from normal TGD epithelium to invasive SCC is proof that the lesion arose from the TGD epithelium.

In this case, the tumor was located in the anterior midline neck, which is compatible with the classic location of TGD cyst. Since there were no apparent primary lesions at the time the initial mass was removed, a rare case of the TGD-SCC could be possible. However, histological findings in this case demonstrated an absence of normal epithelia, suggesting that there were no transitional zones from normal TGD cyst into invasive SCC. This finding did not meet one of the strict criteria for the diagnosis of the TGD-SCC.

3. Conclusion

This case of an atypical large Delphian node metastasis highlights the importance of prompt diagnosis of the lesion as well as microlaryngoscopic examination because of the possibility of occult or microscopic laryngeal cancer. Although the anterior midline neck is the classic location of TGD cyst, Delphian node metastasis as well as carcinoma arising in the TGD cyst is included in the differential diagnosis in a case of rapidly increasing mass in the region.

References