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Bilateral Laryngoceles in Association with Squamous Cell Carcinoma: A Case Report and Literature Review

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ABSTRACT

Coexistence of laryngoceles and laryngeal carcinoma is still being debated, and there are several suggested theories about the pathophysiological relationship between these two entities. We present the case of a 66-year-old male patient with bilateral laryngomucoceles and laryngeal squamous cell carcinoma. A systematic histological examination of whole organ sections showed that the submucosal spreading of cancer around the saccular necks on both sides caused stenosis which probably created a one-way valve mechanism allowing air to enter the saccule but not to exit. Progression of the tumor completely obstructed the laryngeal opening, leading to glandular secretion stagnation and formation of laryngomucoceles.

Key words: larynx, mucocele, laryngeal cancer, etiology

Introduction

The laryngeal ventricle is the space between the vocal and ventricular fold. The bilateral upward extension arising from the anterior end of the laryngeal ventricle is the laryngeal saccule¹. The laryngeal saccule is lined with respiratory epithelium containing serum-mucus-secreting glands, which provide lubrication for the vocal folds. When the saccule is abnormally sized it becomes laryngocela. The laryngocela is also lined with respiratory epithelium and is connected to the laryngeal ventricle by a saccule neck². According to the site of presentation, three types of laryngoceles have been described: internal, external and mixed. The internal laryngocele is confined to the interior of the larynx, and the external laryngocele extends superiorly through the thyrohyoid membrane to the neck. Simultaneous existence of internal and external laryngoceles, medial and lateral to the thyrohyoid membrane, is termed a mixed or combined laryngocele^{3,4}.

Laryngoceles are normally air-filled, but may be filled with mucous to form laryngomucocele or can be filled

with pus and form laryngopyocela. Laryngomucoceles result from the proliferation and secretion of mucous glands within an obstructed saccule neck of laryngocele⁵. Normally, the glandular serum-mucus secretion is evacuated through the saccule neck. After laryngeal trauma, neoplastic disorder or chronic inflammation, which can cause obstruction of the saccule neck, these secretions can stagnate in the laryngocele^{4,6}. The correlation between laryngocele and laryngeal carcinoma is still being debated, and there are a few suggested theories about the pathophysiological relationship between laryngomucoceles and laryngeal carcinoma^{3,7,8}.

Case Report

A 66-year-old male approached our department with a three month history of progressive hoarseness, bilateral neck swelling, dysphonia and difficulty breathing. He had been a heavy smoker but had no other relevant medical history. A clinical examination revealed a 3.3 cm,

soft, painless mass that was extending on both sides of his neck beyond and lateral to the thyroid lamina. No lymphadenopathy was detected during the neck examination. A videolaryngoscopic examination of the larynx revealed an exophytic mass that extended from the laryngeal surface of the epiglottis to both ventricular folds, anterior commissure and anterior part of the vocal folds, with decreased mobility on both sides. A direct laryngoscopy was carried out under general anesthesia for biopsy, and a histopathologic examination revealed the lesion to be well-differentiated squamous cell carcinoma. Computerized tomography (CT) scans assessed the extent of tumour involvement and neck masses more precisely, and at the supraglottic and glottic level showed a bilateral tumour mass with erosion of the ossified thyroid cartilage. The scans also detected the presence of fluid-filled cavities in both laterocervical regions delimited by regular walls, about 2.5 cm in diameter, extending up to the level of the hyoid bone (Figure 1). There was no evidence of enlarged cervical lymph nodes. A total laryngectomy and bilateral selective anterolateral neck dissection was performed and both laryngomucoceles were excised.

Macroscopic examination showed a large tumour on the median laryngeal surface of the epiglottis, ventricular and vocal folds and the deep structures of the larynx (Figure 2). Ossified thyroid cartilage was destroyed by a carcinoma. Macroscopically it seems that a carcinoma involves saccule neck on both sides. Hystological examination showed well-differentiated squamous cell carcinoma which infiltrated the deep structures of the larynx and invaded ossified thyroid cartilage. Carcinoma has extended subepithelially around the both sacculae necks, infiltrating submucosa of the internal parts of both laryngoceles. Overlaying pseudostratified ciliated columnar epithelium was intact (Figure 3). Two cervical lymph node metastases in the right side were detected. The tumour was graded at T4N1M0⁹.

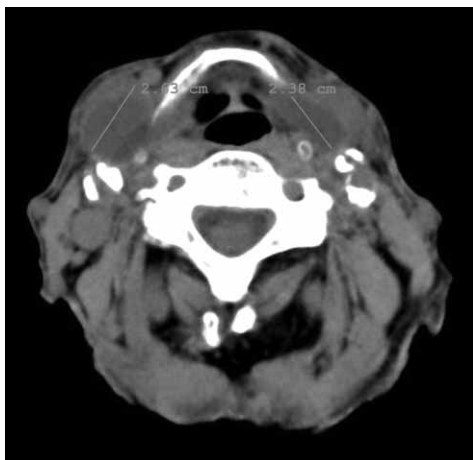


Fig. 1. Axial CT scan showing fluid-filled cavities in the both lateral neck regions. Laryngomucoceles are passing through the thyreohyoid membrane at the level of the hyoid bone.

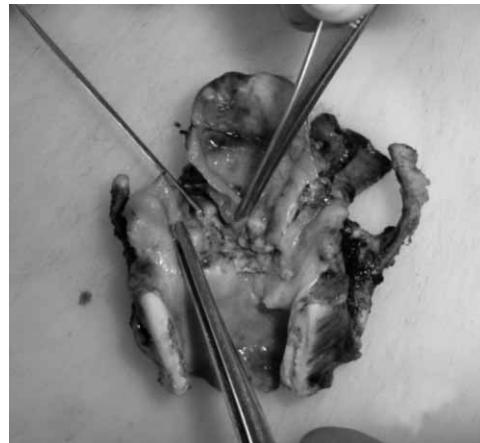


Fig. 2. Macroscopically it seems that a carcinoma involves saccule neck on both sides.

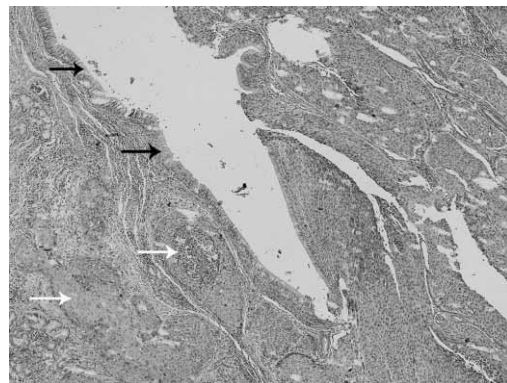


Fig. 3. Histological examination showed pseudostratified, ciliated columnar epithelium of the narrow stalk (black arrow). In submucous there is a well-differentiated squamous cell carcinoma (white arrow) which narrows laryngeal opening.

Four weeks after surgery, the patient underwent post-operative radiotherapy. Two years after treatment, the patient remained free of tumour recurrence.

Discussion and Conclusion

The term laryngocele was introduced by Virchow in 1867, but the condition is thought to have first been reported in 1829 by the French military surgeon Dominique Larrey¹⁰. Incidence is 1:2,500,000 population per year^{4,11}. Peak incidence is in the sixth decade of life. Laryngoceles are unilateral in 85%, with no right- or left-side predominancy, and can be congenital and acquired¹¹. It seems that they develop under conditions of prolonged and pronounced increase in intralaryngeal pressure, which occurs with glass blowing, wind instrument playing, singing and persistent coughing⁶. They can also occur with conditions that produce a flap-valve mechanism, such as tumours, inflammatory lesions and trauma^{4,6,12}.

Diagnosis of laryngocele was based on clinical findings, endoscopic examination of the larynx and imaging

studies. A CT scan of the neck provided a definitive diagnosis of laryngocele^{2,12}.

Coexistence of laryngoceles and laryngeal carcinoma is still being debated and has been reported by several authors. It was first described by Marschik in 1927⁸. The frequency of laryngoceles in laryngeal cancer specimens varies between 4.9% and 28.8%, and in the normal larynx is about 2%^{13–15}. This difference demonstrated the role of cancer in the genesis of laryngoceles. In patients with laryngocele, because of the high frequency of coexistent carcinoma, detailed examination of the larynx is necessary¹⁴. However, in patients with metastatic neck nodes with unknown primary carcinoma, detailed examination of the larynx and looking for laryngocele as a potential site of a primary carcinoma is recommended¹³.

There are several suggested theories about the pathophysiological relationship between laryngomucoceles and laryngeal carcinoma. The first theory describes the distortion of the saccule neck by a carcinoma, which may create a one-way valve that inflates and distends the saccule. It seems that the laryngocele provides a pathway of least resistance for a tumor to spread. This theory does not explain the existence of contralateral laryngocele. The second theory is that carcinoma may arise from the lining epithelium of a large saccule. A preexisting laryngocele might provide a preferential site for tumor development. There is no scientific evidence to support this hypothesis. The third theory postulates an alteration of laryngeal physiology due to carcinoma, which may increase intralaryngeal pressure, possibly as a result of fre-

quent coughing, phonatory misuse or changes in laryngeal neuromuscular mechanics, resulting in the enlargement of the congenitally large saccule^{3,4,8}.

In our case laryngeal cancer spread to the medial laryngeal surface of the epiglottis, ventricular and vocal folds, the deep structures of the larynx and both saccular necks. Although it is thought that the tumour spread into the laryngoceles through the saccular necks, a systematic histological examination of whole organ sections showed that the respiratory epithelium of saccular necks and internal parts of the laryngoceles were intact. Submucosal spreading of cancer around the saccular necks on both sides caused stenosis which probably created a one-way valve mechanism allowing air to enter the sacculus but not to exit. Prolonged forced coughing, difficulty in expectoration and phonatory alterations might be additional causative factors in enlargement of large sacculus. Progression of the tumor completely obstructed the laryngeal opening, leading to glandular secretion stagnation and formation of laryngomucoceles. These findings suggest the possibility of the first theory of laryngocele formation and explain that the saccular neck was not a pathway of low resistance for the tumor to spread.

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OBOSTRANE LARINGOKELE UDRUŽENE S PLANOCELULARNIM KARCINOMOM: PRIKAZ SLUČAJA I OSVRT NA LITERATURU

S A Ž E T A K

O istovremenom postojanju laringokela i karcinoma larinksa se još uvijek raspravlja. Postoji nekoliko predloženih teorija o patofiziološkim odnosima između ova dva entiteta. Predstavljamo slučaj 66-godišnjeg muškarca s bilateralnim laringomukokelama i planocelularnim karcinomom larinksa. Patohistološka analiza pokazala je submukozno širenje karcinoma oko vrata mukokele na obje strane što je vjerojatno prouzročilo stenozu i stvorilo jednosmjerni ventil mehanizam koji omogućava ulaz zraka u laringokelu, ali ne i izlaz. Progresija tumora potpuno zatvara laringealni otvor, a što dovodi do stagnacije žljezdane sekrecije i stvaranja laringomukokele.