OLGU SUNUMU CASE REPORT

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Rare Pathology of the Larynx: Papillary Oncocytic Cystadenoma

Larenksin Nadir Görülen Patolojisi: Papiller Onkositik Kistadenoma

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ABSTRACT Laryngeal cystadenoma (other terminological names; oncocytic cystadenoma, papillary cystadenoma, oncocytic adenoma) is a rare, benign, slow growing tumor predominantly composed of oncocytes. Its epithelial component is similar to Warthin tumors (papillary cystadenoma lymphomatosum) seen in the major salivary glands. We aimed to present a rare case of laryngeal oncocytic cystadenoma in the same case by examining the relationship between the accompanying parotid gland's Warthin tumor. Laryngeal oncocytic cysts are histologically similar to Warthin (papillary cystadenoma lymphomatosum) tumor, but differentiates with not having a two-fold structure consisting of oncocyte and lymphoid stroma. Magnetic resonance imaging is the gold standard method in diagnosis of complex lesions. The optimal treatment is endoscopic or open laryngeal surgical excision. Malignant transformation of the lesion is not clear, close monitoring of the operated area is essential.

Keywords: Laryngeal diseases; systadenoma; adenolymphoma

ÖZET Laringeal kistadenomlar (diğer terminolojik isimler; onkositik kistadenom, papiller kistadenom, onkositik adenom), nadir görülen benign, yavaş büyüyen ve ağırlıklı olarak onkositlerden oluşan tümörlerdir. Epitelyal patolojisi, majör tükürük bezlerinde görülen Warthin tümörlerine (papiller kistadenoma lenfomatozum) benzerdir. Nadir görülen laringeal onkositik kistadenomu olgusunun, aynı olguda ona eşlik eden parotis bezinin Warthin tümörü ile ilişkisini incelemeyi amaçladık. Laringeal onkositik kistler, histolojik olarak Warthin (papiller kistadenoma lenfomatozum) tümörüne benzer, ancak onkosit ve lenfoid stromadan oluşan 2 katlı yapısı olmaması nedeniyle ayrışmaktadır. Manyetik rezonans görüntüleme yöntemi, kompleks lezyonların tanısında altın standart tanı yöntemidir. En uygun tedavi, endoskopik veya açık laringeal cerrahi eksizyondur. Lezyonun maligniteye dönüşümü ile ilgili bilgiler net olmadığından ameliyat edilen bölgenin yakından izlenmesi şarttır.

Anahtar Kelimeler: Larinks hastalıkları; kistadenom; adenolenfoma

Laryngeal cysts are benign lesions associated with the laryngeal mucosa line. Its frequency in non-malignant laryngeal pathologies is around 5-10%. Many classifications have been made about laryngeal cysts. In the classification made by DeSanto, cysts are divided into neoplastic and non-neoplastic. Neoplastic cysts are cystadenoma, congenital cyst, traumatic cyst, vascular malformation and hemangiomas. Nonneoplastic ones are glandular cyst, amygdaloid cyst and lymphatic cyst.[1] In Newman's classification, intralaringeal cysts are divided into three as epithelial, tonsillar and oncocytic.[2]

The two-layer histological structure of oncocyte and lymphoid strode seen in the Warthin tumor is not seen in these cysts.[3] Delayed type hypersensitivity is thought to be the initiating factor in the etiology of oncositic kistadenomas, similar to Warthin tumor.[4] Oncocytes are caused by the metaplasia of the ductal epithelium of the seromucinous gland in response to chronic irritation and cigarette is the most common

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chronic irritant.[5] The location is mostly upper respiratory tract, tongue, pharynx, esophagus and thyroid. Laryngeal oncocytic cystadenomas are rarely seen, 150 cases have been described in the literature.[6] The most common incidence in the larynx is supraglottis, mostly with a single lesion with hoarseness, and seen in the seventh or eighth decad. Less often, it can be diffuse or multifocal. Oncositic cystadenomas can be associated with multiple cysts or relapses, so it is required to follow regularly. The most optimal treatment approach is endoscopic or open laryngeal surgical excision, as the lesion can lead to upper respiratory obstruction when it is large enough.[7]

CASE REPORT

An 88-year-old female patient was admitted to our otolaryngology outpatient clinic with the complaints of increasing hoarseness and a feeling of sning in her throat during swallowing. The patient had no dysphagia, respiratory distress, stridor, weight or loss of appetite. There is a history of 80/pack of cigarettes between the ages of twenty and sixty, two packs a day. In terms of known hypertension disease, it was learned that she had internal medicine follow-up and used her drugs regularly.

The patient underwent a right superficial parotidectomy operation five years ago with the diagnosis of Warthin tumor and the vulva operation with the diagnosis of squamous carcinoma in situ the same year. No pathological finding was observed in the right parotid site and neck. Systemic examinations were otherwise normal.

In indirect laryngoscopic examination performed in the outpatient clinic with a 70 degree endoscope, a 2×1 cm cystic lesion was detected in the right ventricle, it was not fragile, well-circumscribed, covered with normal mucosa. In the bilateral vocal cords, other laryngeal structures were released other than reinked edema. In other otorhinolaryngological examination, no pathological finding was observed.

In the neck magnetic resonance (MR) examination of the patient before the operation, a 12*11 mm mass extending towards the supraglottic region, which partially interrupted the air column in the right ventricle of the larynx, was observed. Malignant diagnoses could not be excluded in the radiological evaluation of the mass showing hyperintense feature in T2 imaging.

Although amyloid, condrome or other benign cystic lesions were considered in pre-liminary diagnosis due to the history of heavy smoking and clinical findings direct laryngoscopy and surgical excision were planned under general anesthesia in the operating room. A written informed consent was obtained from the patient.

In the operating room, it was considered that the lesion did not have malignant properties in the direct larynchoscopic view. When we looked at other laryngeal structures, pathological findings were not observed in bilateral vocal cords other than reinke edema. The lesion was endoscopically exclused after the frozen biopsy from the cyst was not reported as malignant. The cyst, which was monitored as originating from the front of the right ventricle, was removed in one piece without rupture and sent to pathology (Figure 1).

The pathology result of the lesion was reported as oncocytic papillary cystadenoma. The patient returned to her normal life after three weeks. In the fourth month control, no lesions were observed in favor of recurrence in the right parotid and right ventricle. The patient was regularly followed up (Figure 2).

DISCUSSION

Laryngeal oncocytic cysts are histologically similar to Warthin (papillary cystadenoma lymphomatosum)

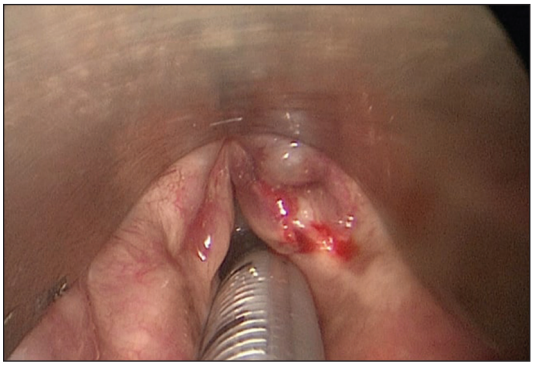

FIGURE 1: Papillary cystadenoma located in the front of the right ventricle in the direct laryngoscopic examination.

FIGURE 2: The area that was operated after the operation and the view of the cyst completely cleaned from this area.

tumor, but differentiate due to the fact that it does not have a two-fold structure consisting of oncocyte and lymphoid stroma.[3] In our patient, Warthin tumor developed in the right superficial parotid gland five years ago. Later, papillary cystadenoma developed in the right ventricle of the larynx, again in the same anatomic half of the body. In this respect, the ability to see these two diseases together especially in advanced age patients should be kept in mind and should be taken into consideration in the long-term follow-up of patients.

In the literature, these cysts can also be referred to in different terms such as oxyphilic adenoma, eosinophilic granular cell cyst, oncocytoma, oncocytic cyst and oncocytic papillary cystadenoma.[8] Since it is rare, it should be kept in mind in differential diagnosis. The common view in the etiopathogenesis of laryngeal oncocytic lesions is the metaplasia of the seromucinous gland ductal epithelium due to chronic irritation. Cigarette is the most common chronic irritant especially in the elderly population who smoke excessively.[5-7] It is more common in decades. Although it is common in men, it is more symptomatic in women.

Oncocytic cystadenoma can develop from anywhere on the larynx except the free edge of the true vocal cords without glandular epithelium. The most common location in the larynx is the supraglottic region. It is mostly seen as a solitary isolated cyst. It may be pedicular, sessile or polypoid in character.[9] It is very rare to be seen as a diffuse or multiple cyst.

The most frequent complaints of patients are depending on the localization of the cyst, hoarseness or voice roaring.[10] Patients' voice problems are often present chronically for months rather than being acute. In supraglottic cysts, swelling sensation during swallowing (globus), dysphagia, odynophagia, referral otalgia, snoring symptoms may be encountered. Complaints like pain, stridor or laryngeal obstruction are much rarer. Rarely, fast-growing cysts are infected by occlusing the respiratory tract or lesions around the epiglottis, causing an increased risk of mortality by epiglottitis. Some cases are asymptomatic and diagnosed incidentally.

Computed tomography is the first method that should be used as a imaging method in defining the disease. MR imaging is the gold standard method in diagnosis of complex lesions. Endolarengeal ultrasound and optical coherence tomography can improve the preoperative evaluation of laryngeal cysts, but should be correlated by the clinician. Temporary symptomatic control can be achieved in the treatment of laryngeal cysts with aspiration.[11]

Although histopathologically, oncocytic cystadenomas are defined as the primary neoplasic lesion that develops on the basis of degeneration or is a primary neoplasic lesion, it has not been clearly reported in the literature. Since oncocytic metaplasia is strongly associated with smoking, it is a pre-stimulating condition for the risk of dysplastic neoplasia in the larynx. In the literature, it has been reported that squamous cell carcinoma develops from common oncocytic laryngeal tissue.[4] Histopathological uncertainty continues in this regard.

In diffuse or multiple oncocytic cysts, the important condition to be considered in follow-up is recurring. Relapse is more common, especially in cases of biopsy that are repeated frequently in a short period of time, or in diffuse laryngeal cysts.[12] Surgical manipulations can cause a faster and more of relapse cysts than precystic metaplastic areas.[13]

Local surgical excision is recommended with a conservative approach, since oncocytic cysts may tend to relapse and their relationship with malignant lesions is still uncertain. Surgical excision can be performed endoscopically by the transoral method, as

well as open laryngeal procedure in diffuse or recurrent lesions.

In the endoscopic transoral method, carbon dioxide (CO_2) laser can be more advantageous in reducing the frequency of recurrence compared to the cold knife method.[14]

Today, transoral robotic procedures reduce the total cost and prevent relapses more because it reduces hospital stay and operation time compared to open surgical technique.[15]

One of the important points that clinicians should pay attention to is close follow-up in the post operative period. Since the information about the malignant transformation of the lesion is not clear, close monitoring of the operated area is essential. Since neither malignant transformation of oncocytic papillary cystadenomas nor true clinical laryngeal oncocytic metaplasias can still be revealed, there is no numerical data about the risk of recurrence in the literature.[11]

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Hüseyin Avni Ulusoy, Filiz Gülüstan;* ***Design:*** *Hüseyin Avni Ulusoy;* ***Control/Supervision:*** *Filiz Gülüstan;* ***Data Collection and/or Processing:*** *Filiz Gülüstan, Hüseyin Avni Ulusoy;* ***Analysis and/or Interpretation:*** *Hüseyin Avni Ulusoy;* ***Literature Review:*** *Hüseyin Avni Ulusoy;* ***Writing the Article:*** *Hüseyin Avni Ulusoy;* ***Critical Review:*** *Filiz Gülüstan;* ***References and Fundings:*** *Hüseyin Avni Ulusoy;* ***Materials:*** *Hüseyin Avni Ulusoy.*

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