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Case Report / Olgu Sunumu

Laryngeal inflammatory pseudotumor: a case report

Larengeal enflamatuvar psödotümör: Olgu sunumu

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Inflammatory myofibroblastic pseudotumor (IMP) is a rare neoplasm usually found in the lower respiratory tract, pulmonary system, and abdominal cavity. Conservative surgical procedures are often performed in the management of the tumor. In this article, we present a 64-year-old male with a local IMP of the larynx. The clinical presentation, diagnosis, histopathology, and management of this uncommon tumor were also discussed.

Key Words: Inflammatory myofibroblastic pseudotumor; larynx; neoplasm; vocal cord.

Enflamatuvar miyofibroblastik psödotümör (EMP), genellikle alt solunum yolu, pulmoner sistem ve abdominal kaviteyi tutan, ender rastlanan bir tümördür. Tümörün tedavisinde sıklıkla konservatif cerrahi girişimler uygulanır. Bu makalede, larenksi tutan lokal EMP'li 64 yaşında bir erkek hasta sunuldu. Makalede bu ender rastlanan tümörün klinik tablosu, tanısı, histopatolojisi ve tedavisi de irdelendi.

Anahtar Sözcükler: Enflamatuvar miyofibroblastik psödotümör; larenks; tümör; vokal kord.

Inflammatory myofibroblastic pseudotumors (IMPs) are known as plasma cell granulomas or atypical fibromyxoid nodules. The disease is named because of the histopathological wide spectrum from plasma cell to myofibroblastic cell dominancy. Inflammatory myofibroblastic pseudotumors may involve different systems such as the pulmonary system, abdominal cavity, tracheobronchial tree, nasopharynx and mastoid cavity.[1] Inflammatory myofibroblastic pseudotumors rarely occur in the larynx. Laryngeal IMPs was first described in 1992 by Manni et al.[2] Here, we report a case of laryngeal inflammatory pseudotumor and review the literature.

CASE REPORT

A 64-year-old male was admitted to our clinic in January 2008 with complaints of hoarseness and voice crackles for two months. The patient had no other medical history except for antireflux therapy for a few previous months. He had no systemic comorbidity but had a smoking history of 45 pack/years of cigarettes.

Indirect laryngoscopic examination revealed a mass in the right vocal cord involving the anterior commissure. Both vocal cords were mobile with normal opening of the glottis. Direct suspension laryngoscopic evaluation and excisional biopsy were planned. The patient underwent the surgical procedure in January 2008. Under general anesthesia, direct microlaryngoscopy revealed a mass with fibrotic, regular surface and minimal infraglottic extension in the anterior $^1/_3$ portion of the right vocal cord (Figure 1). The excisional biopsy was performed for the lesion. The histopathological

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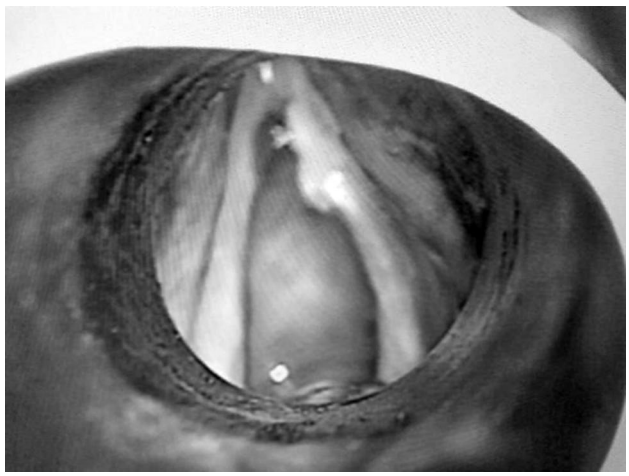

Figure 1. *Direct microlaryngoscopy showed a mass with fibrotic, regular surface, minimal infraglottic extension in the anterior 1/3 portion of the right vocal cord.*

diagnosis was reported as idiopathic IMPs. A 1.5x0.8x0.7 cm biopsy specimen was examined. The surface of spherical specimen was totally ulcerated except for a millimetric hyperplastic squamous fragment. Under the epithelium, stroma with edematous myxoid connective tissue was observed. With medium magnification myofibroblastic cell-blocks could be seen (Figure 2).

Although necrosis could be seen in the stroma, neither atypia nor mitosis were observed. On immunohistochemical study, the squamous epithelium was stained with pancreatin. A focal cytoplasmic stain pattern was observed with smooth muscle actin (SMA) and S-100 (Figure 3).

Therefore, under general anesthesia in the second direct microlaryngoscopy, the lesion was resected totally with cold surgical procedure and no complication in January 2008. The histopathological examination was also confirmed as IMP.

The patient was informed and advised for regular follow-up, and also medicated with pantoparazole 40 mg/day, calcium carbonate 2040 mg - magnesium carbonate 240 mg/day for six months for his reflux complaints. Over two years, the patient consulted twice with the same complaints, and IMP recurrences were suspected related with new masses in the anterior 1/3 of the right vocal cord each time. The patient underwent three revision surgeries (July 2008 - December 2008 - June 2009) under general anesthesia each time in the last three years. All two histopathological examinations were also reported as IMP. No recurrences were observed since the last surgery (June 2009). Only minimal localized atrophy and scar tissue remain in the anterior right vocal cord, with no synechia, web or other complications during our follow-up of one and half year.

DISCUSSION

Inflammatory pseudotumor (IPT) is a term that is used to describe a benign tumoral process that may affect a variety of organ systems.[3] It is a rare slow growing tumor with distinct histological appearance and usually benign clinical course. It has been described in all age groups in many tissues and in both sexes. First reported in the lung, it can occur anywhere in the body. The lung, spleen, liver, gastrointestinal tract and mesentery, mediastinal and retroperitoneal soft tissues, pancreas, bladder, thyroid, larynx, meninges, orbit, heart, breast,

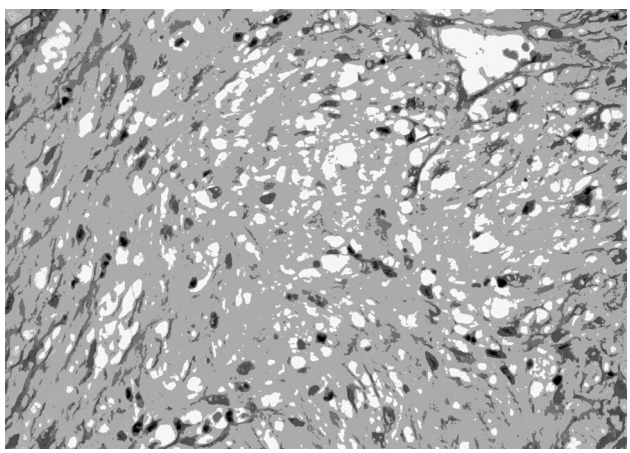

Figure 2. *Myofibroblastic cell groups and pattern of the lesion with medium magnification. Short fascicular alignment of fusiform cells. (H-E x 200).*

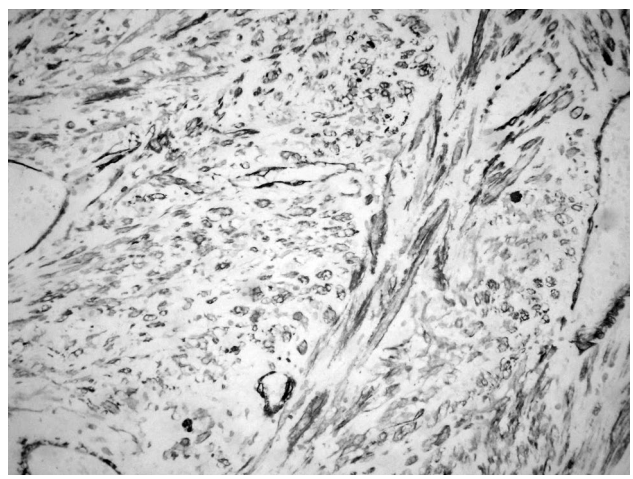

Figure 3. *Immunohistochemical study; myofibroblastic cell groups stained with actin (x 400 IHC Actin).*

epididymis, skin, soft tissues and the lymph nodes have all been reported to be the affected sites.[4]

Laryngeal pseudotumors are benign, highly recurrent and rare tumors.[5-7] Although its pathophysiology has not yet been clarified, misuse of vocal phonation, deep coughing and gastroesophageal reflux have been implicated. The mechanism is supposed to be myofibroblastic proliferation during tissue regeneration due to subclinical trauma.[8] Smoking and gastroesophageal reflux were risks factors for our case report in development of IMP. It may be confused with laryngeal malignancies and/or recurrent laryngeal papillomatosis. Distant metastasis rarely occurs. Vascular invasion and malignant transformation may be seen. Pathologically it may be confused with fibrosarcoma, malignant fibrous histiocytoma and leimyosarcoma. Thus immunohistochemical staining should be considered to make a differential diagnosis. The treatment includes surgical excision, radiotherapy or CO_2 laser resection.[9]

No treatment protocols or modalities have been stated due to the rarity of laryngeal inflammatory pseudotumors. Many authors advice endoscopic excision of the lesion with combination of oral/parenteral steroid therapy.[10,11] We performed microlaryngoscopic surgeries four times within three years, but could not give steroid therapy because of his gastroesophageal reflux disease. Although radiotherapy stands to be a therapeutic option, there have been recurrences reported after this therapy.[10] Therefore, radiotherapy should be used as neoadjuvant therapy for recurrences or for those patients who are not suitable for surgery. There are controversial reports which claim CO_2 laser ablation to be very successful in local control of the tumor and achieving low recurrence rates.[10] For recurrences, wider surgical procedures including laryngectomy with or without radiotherapy have been advised.[2] In our case, the local recurrences involved a limited portion of the right vocal cord, thus radiotherapy or wider surgical procedures were not considered. The diagnosis of these limited local recurrences were achieved with the help of close follow-up of our patient which makes follow-up very important for early diagnosis.

In conclusion, IMP lesions may present as chronic otitis media, chronic sinusitis, laryngeal masses in daily otolaryngologic practice.[12,13] Physicians should keep IMP in mind in the differential diagnosis of fungating laryngeal masses. This lesion can mimic malignancy, and therefore histopathological confirmation is required prior to any intervention. Currently, microlaryngoscopic surgical resection still seems to be the standard of care for the disease.

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