An extralaryngeal, extraesophageal surgical approach to a low-grade posterior cricoid chondrosarcoma

Selçuk Güneş[1], Burak Olgun[1], Mustafa Çelik[2], Zahide Mine Yazıcı[1], İbrahim Sayın[1]

[1]*Department of Otorhinolaryngology Head and Neck Surgery, Bakırköy Dr. Sadi Konuk Training and Research Hospital, Istanbul Turkey*
[2]*Department of Otorhinolaryngology Head and Neck Surgery, Kafkas University, Faculty of Medicine, Kars, Turkey*

ABSTRACT

A chondrosarcoma is a malignant tumor originating from cartilage and may occur in any part of the body. It occurs most commonly in the posterior lamina of the cricoid cartilage, followed by the thyroid cartilage, arytenoid cartilage, vocal cords, and epiglottis. In the literature, 25 cases of tracheal chondrosarcoma were reported between 1964 and 2017, but only two cases of cricoid cartilage chondrosarcoma. Currently, no consensus on chondrosarcoma treatment has been established yet, due to the rarity of the condition, and no therapeutic protocol has been shown to be clearly superior to others. Herein, we present the third case of a cricoid chondrosarcoma in the literature. We used a surgical procedure different from those employed earlier; we employed an extralaryngeal, extraesophageal approach. We describe the diagnosis, treatment, and follow-up results in the light of current literature.

Keywords: Chondrosarcoma, extralaryngeal, extraesophageal, approach, cricoid cartilage.

A chondrosarcoma is a malignant tumor originating from cartilage and may occur in any part of the body.[1] Low-grade chondrosarcomas constitute about 1% of all laryngeal tumors and occur most commonly in the posterior lamina of the cricoid cartilage, followed by the thyroid cartilage, arytenoid cartilage, vocal cords, and epiglottis.[1,2] These tumors are usually slow-growing and are only locally invasive, and metastasis is rare.[2] Often, the symptoms include airway obstruction, but may be non-specific.[3]

In the literature, 25 cases of tracheal chondrosarcoma were reported between 1964 and 2017, but only two cases of cricoid cartilage chondrosarcoma. Currently, no consensus on chondrosarcoma treatment has been established yet, due to the rarity of the condition, and no therapeutic protocol has been shown to be clearly superior to others. Herein, we present the third case of a cricoid chondrosarcoma in the literature. We used a surgical procedure different from those employed earlier; we employed an extralaryngeal, extraesophageal approach. We describe the diagnosis, treatment, and follow-up results in the light of current literature.

CASE REPORT

A 54-year-old female patient was admitted complaining of difficulty in swallowing. Routine and systemic examinations revealed non-specific features. On indirect endoscopic laryngeal

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Correspondence: Mustafa Çelik, MD. Kafkas Üniversitesi Tıp Fakültesi Kulak Burun Boğaz Anabilim Dalı, 36100 Kars, Turkey.
e-mail: dr.mcelik@yahoo.com

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examination, a subglottic lesion with a normal mucosal surface was observed just below the left arytenoid cartilage. We made a preliminary diagnosis of a malignancy and scheduled computed tomography (CT) after explaining the harmful effects of radiation to the patient. Contrast-enhanced neck CT revealed a mass featuring punctate calcifications associated with the posterolateral lamina of the cricoid cartilage. The mass was 9×8 mm in size and protruded toward the air column in the posterior segment (at the subglottic level) to the left of the larynx (Figure 1). Contrast-enhanced neck magnetic resonance imaging (MRI) was scheduled for soft tissue evaluation, as no mucosal lesion was evident on laryngeal endoscopic examination. The MRI scan revealed an asymmetric focal lesion 6.5×6×10 mm in size posterior to and under the mucosa at the subglottic level of the left side of the larynx (Figure 2). Based on these findings, a preliminary diagnosis of a cricoid chondrosarcoma and direct laryngoscopy and incisional biopsy were performed under general anesthesia. The biopsy was performed after mucosal elevation; we incised the mucosa over the posterolateral lamina of the cricoid. Histopathological examination revealed a low-grade chondrosarcoma (Figure 3). The possible risks of operation and tracheostomy were fully explained to the patient and a written informed

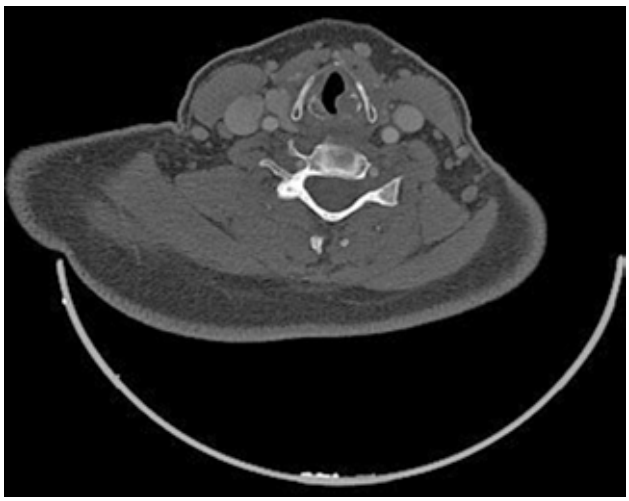

Figure 1. *Contrast-enhanced neck computed tomography showing a mass featuring punctate calcifications associated with posterolateral lamina of cricoid cartilage. The mass was 9×8 mm in size and protruded toward air column in posterior segment (at subglottic level) to left of larynx.*

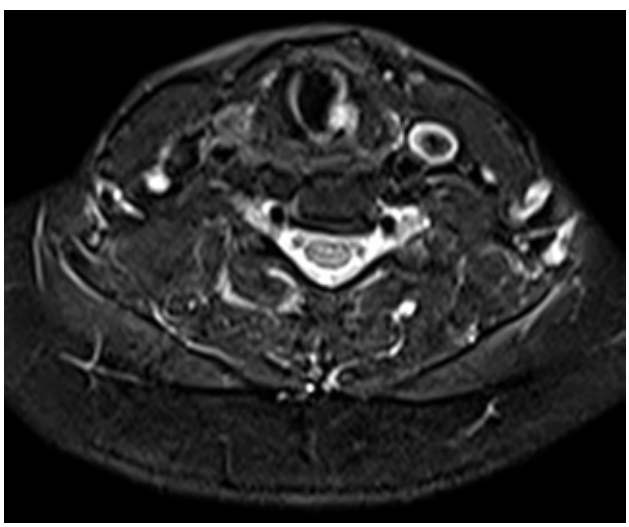

Figure 2. *Contrast-enhanced neck magnetic resonance imaging showing an asymmetric focal lesion 6.5×6×10 mm in size posterior to and under mucosa at subglottic level of left side of larynx.*

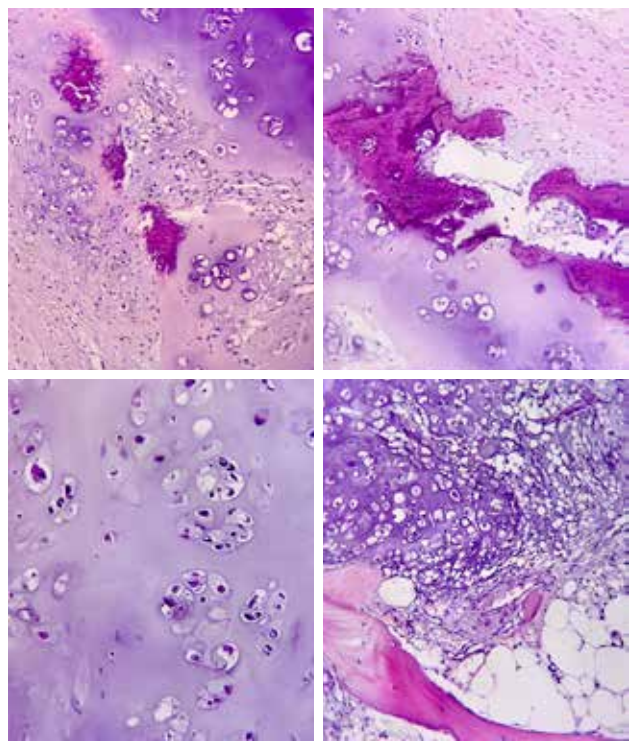

Figure 3. *Histopathological examination of an incisional biopsy specimen showing pleomorphism, atypical chondroid neoplasia featuring cells with multiple nuclei, and increased cellularity of the chondroid matrix, compatible with a low-grade chondrosarcoma (H-E ×100).*

consent was obtained. We planned extralaryngeal, extraesophageal partial cricoidectomy, and tracheostomy.

SURGICAL TECHNIQUE

Tracheotomy was performed at the level of the third ring under general anesthesia. Ventilation was performed during tracheotomy. An oblique incision (~8 cm in length) was created in the region anterior to the left sternocleidomastoid (SCM) muscle. The subplatysmal area was revealed and the carotid artery, vagus sinus, and jugular vein were lateralized to create an anterior view of the SCM muscle. The pharyngeal muscles were incised from the posterior margin of the thyroid cartilage and the sinus piriformis was elevated posteriorly. Then, the cricothyroid joint was separated. The posterior cricoid cartilage was located and the pharyngeal mucosa and cricoarytenoid muscle elevated. A tumor originating from the posterolateral lamina of the left cricoid was, then, detected. Disease-free endolaryngeal mucosa was removed from the cricoid cartilage. The chondrosarcoma was excised extralaryngeally and extraesophageally. As the lesion laid very close to the cricoarytenoid insertion, the joint was also included in excision. Frozen-section histopathological examination was performed to ensure adequate surgical margins, which were disease-free. To eliminate protrusion of the left arytenoid cartilage into the laryngeal lumen, the defective region was filled with the left-upper thyroid cartilage horn and the arytenoid vocal process was sutured to the lamina of the left thyroid cartilage. Perioperative endolaryngeal examination revealed that the endolaryngeal mucosa was free of disease. One day later, laryngeal endoscopy showed that the laryngeal mucosa was disease-free, and the right vocal cord was mobilized. The tracheostomy was closed on postoperative Day 3 and oral feeding initiated on Day 4. The patient was discharged on Day 5. Histopathological examination of the definitive specimen revealed a low-grade chondrosarcoma. No recurrence or residue was observed during six months of follow-up.

DISCUSSION

Laryngeal chondrosarcomas (LCs) are rare, being more common in males, and are usually of low grade.[4] They predominantly contain cartilage and are graded in terms of cellularity, extent of pleomorphism, and atypia (Grades 1-3).[5] Of all LCs, 10% occur in the head and neck region, most commonly in the cricoid cartilage.[6] As is true of other head and neck cancers, the etiopathogenesis has not been fully understood yet, but may involve congenital cartilage involution defects, chronic inflammation, and/or abnormalities of cartilage calcification.[7] The rarity of the cancer and the absence of long-term follow-up make it difficult to elucidate the etiopathogenesis.

Head and neck chondrosarcomas may present with different symptoms including non-specific voice disturbance, dyspnea, and dysphagia; however, these vary depending on the tumor location.[8] Routine physical and endoscopic examination of the larynx may be useful in the diagnosis of laryngeal pathologies. Radiological imaging is valuable in cases lacking pathological findings on physical examination. The CT and MRI are preferred for evaluation of soft tissues and anatomical structures, such as bone and cartilage. If radiological biomorphism is apparent accompanied by a punctate pattern in the cartilage matrix and extraosseous soft tissue development, a chondrosarcoma should be considered in the differential diagnosis, although the tumor lacks radiologically-specific features.[9-11]

In the present case who experienced difficulty in swallowing, we found a subglottic lesion under the left arytenoid cartilage on indirect endoscopic laryngeal examination. The laryngeal mucosa over the lesion was of normal appearance. Bilateral vocal cord movements were also normal. Therefore, we performed CT and MRI. A cricoid cartilage chondrosarcoma was suspected, as punctate calcifications were evident. In the differential diagnosis, we considered a laryngeal spinal cell squamous carcinoma, hamartoma, and high-grade sarcoma. Histopathological examination of an incisional biopsy specimen showed pleomorphism, atypical chondroid neoplasia featuring cells with multiple nuclei, and increased cellularity of the chondroid matrix, compatible with a low-grade chondrosarcoma.

Most cases are resectable and various surgical approaches are currently available. Neither chemotherapy nor radiotherapy is effective, even on intralesional application.[12] Guillem et al.[14] performed partial subglottic tracheal excision, cricoidectomy, and tracheostomy for the treatment of a mediastinal extension of a cricoid chondrosarcoma. Ryabov et al.[15] also performed tracheal resection, tracheostomy, and end-to-end anastomosis via open neck surgery to treat a tracheal chondrosarcoma. Choi et al.[1] performed transoral endoscopic resection of an epiglottic chondrosarcoma. Gao et al.[3] performed tracheal resection of a low-grade cricoid chondrosarcoma. Pignataro et al.[8] performed cricoidectomy and tracheostomy with laryngofissure to treat a cricoid chondrosarcoma. Pellicia et al.[2] performed transoral endoscopic resection of seven low-grade cricoid chondrosarcomas; only two patients required tracheostomies. In our case, we performed extralaryngeal, extraesophageal partial cricoidectomy. No gold standard surgical method is available; rather, the method chosen should depend on the experience and preference of the surgeon. Our extralaryngeal, extraesophageal approach seeks to protect the endolaryngeal and esophageal mucosae of patients with low-grade cricoid chondrosarcomas.

In conclusion, cricoid chondrosarcomas are rare malignant tumors. An unusual tumor, such as a laryngeal chondrosarcoma, should be considered in the differential diagnosis of cases with non-specific laryngeal symptoms. Otorhinolaryngologists should be aware of cricoid chondrosarcomas. We believe that our novel extralaryngeal, extraesophageal partial cricoidectomy technique is useful in this patient population.

Declaration of conflicting interests

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