Laryngeal Chondrosarcoma of the Arytenoid Cartilage Presenting as Bilateral Vocal Fold Immobility: A Case Report and Literature Review

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Summary: Objectives. To describe an atypical case of laryngeal chondrosarcoma of arytenoid cartilage presenting as bilateral vocal fold immobility and to avoid potential missed diagnosis.

Methods. Our case study included a detail history, physical and radiological examination, laryngeal electromyography (LEMG), and surgical treatment and pathology analysis. We compared it with the previously discussed cases of chondrosarcoma of arytenoid cartilage in the literature.

Results. Chondrosarcomas of the arytenoid cartilage is rare, and to date only approximately 10 cases have been reported. We reported a case of a 51-year-old man with 1 month of persistent dyspnea presenting with bilateral vocal fold immobility without neoplasms in larynx. The LEMG showed no obvious abnormality. The cervical-enhanced computed tomography (CT) found no significant signs of a mass except for localized high-density areas in arytenoid cartilage. Right arytenoidectomy and biopsy were performed under general anesthesia with CO2 laser with the pathological diagnosis of chondroma. A total laryngectomy was performed 2 years later, and low-grade chondrosarcoma was the final diagnosis.

Conclusions. Laryngeal chondrosarcomas of the arytenoid cartilage are rare. It is easily neglected, especially in those cases presenting with idiopathic vocal fold immobility without any obvious signs of neoplasms. The LEMG and laryngeal CT are necessary. Sometimes, a biopsy of the arytenoid cartilage is essential.

Key words: Larynx–Chondrosarcoma–Vocal fold immobility–Arytenoid cartilage–Laryngeal electromyography.

INTRODUCTION
Laryngeal chondrosarcoma is the most common nonepithelial malignant tumor of the larynx. It constitutes less than 1% of laryngeal tumors and 0.07–2% of malignant laryngeal tumors. Approximately 600 cases of laryngeal chondrosarcoma have been reported in the literature.1–4 The major site of origin is the cricoid cartilage. The arytenoid cartilage is quite exceptional as a primary site location, with only 10 cases being reported previously.5–12 Owing to its hidden and atypical symptoms, long duration, and other features, the diagnosis of laryngeal chondrosarcoma is easily overlooked. Moreover, it is sometimes difficult to distinguish between low-grade chondrosarcoma and chondroma because of the similarity of their histological features. Thus, the choice of surgery is controversial. We reported a case of a 51-year-old man with an atypical history of continuous dyspnea, but did not have a significant mass sign in the laryngeal findings and radiological presentation. However, the final pathological diagnosis was low-grade chondrosarcoma of the arytenoid cartilage, and the patient underwent a total laryngectomy. The clinical characteristics and treatment of this patient are summarized.

CASE REPORT
A 51-year-old man was admitted to our department in November 2007 with a chief complaint of continuous inspiratory dyspnea and mild hoarseness for 1 month after a cold infection. There was no history of dysphagia, laryngeal pain, or asthma. He had been in good health previously and claimed only occasional tobacco use and no alcohol use.

Physical and radiological examinations
Under the stroboscope, the right vocal fold was fixated at the paramedian position, whereas the left vocal fold was incompletely limited. But no neoplasms were observed in the larynx (Figure 1). Laryngeal electromyography (LEMG) characteristics were nearly normal. No abnormal results were found in the chest X-ray, thyroid ultrasound, or cranial computed tomography (CT) examination. The esophageal barium meal examination demonstrated slightly narrowing of the right pyriform sinus and a compression-filling defect of the upper esophagus. The cervical-enhanced CT found that there were no significant signs of a mass except for an area of localized high density in the arytenoid cartilage, especially on the right side (Figure 2). The patient was followed up without any special treatment. Six months later, the patient developed obvious dyspnea for 1 week after a cold infection. The stroboscopy showed signs similar to those of the previous examination except that the laryngeal mucosa was swollen and had acute hyperemia. The cervical-enhanced CT showed no changes.
Surgical treatment and biopsy
We reviewed the results of all the patient’s examinations and considered that the abnormal high density of the arytenoid cartilage may be the cue for diagnosis, that is, to say the lesion of the arytenoid cartilage itself may have led to the immobility of vocal folds. Thus, we decided to perform a CO2 laser right arytenoidectomy and biopsy under direct laryngoscope with general anesthesia to relieve dyspnea and define the characteristics of abnormal arytenoid. During the surgery, we found that the right arytenoid cartilage was calcified and harder than usual. The histopathology of arytenoid showed that the chondrocytes were abnormally hyperplastic with the pathological diagnosis of chondroma. However, some cells even showed obvious signs of hyperchromatism and cellular atypia, whose nucleus became round, polygonal, or irregularly shaped, which may suggest a tendency to become malignant (Figure 3A). Regular follow-up was highly recommended. After the surgery, the patient experienced relief of dyspnea and refused further treatment. The patient did not have respiratory difficulties or other symptoms until 22 months after the surgery. In April 2010, the patient was hospitalized again for a gradual return of dyspnea. We attempted a residual right arytenoidectomy with a CO2 laser for the biopsy, but the residual right arytenoid cartilage was like sand. At this time, the pathological diagnosis was identified as chondrosarcoma. Finally, we performed a total laryngectomy for the patient, and the pathological diagnosis was low-grade chondrosarcoma (Figure 3B), and bilateral arytenoids and interarytenoid were involved, which may also explain the left vocal fold limitation, whereas the vocal folds, ventricular folds, thyroid cartilage, epiglottic cartilage, and cricoid cartilage were not involved.

The patient was followed up regularly for more than 2 years and 8 months and is free of disease and in good health at present.

DISCUSSION
Chondrosarcoma is a type of slow-growing nonepithelial malignant tumor that most commonly originates in the long bones of limbs, pelvis, spine, and shoulder blades. Chondrosarcomas of the head and neck are infrequent, accounting for 10% of all chondrosarcomas.1 The first case of laryngeal chondrosarcoma was reported by New13 in 1935. To date, approximately 600 cases have been reported. The cricoid cartilage is the most commonly affected location (69–80%), followed by the thyroid cartilage (9–20%). The epiglottis and the arytenoid cartilage account for less than 5% of the cases. The diagnosis of laryngeal chondrosarcoma is very easily neglected because of its slow growing feature. Most patients in the previous studies had a long history, ranging from 3 to 216 months. The symptoms of hoarseness, dyspnea, dysphagia, and some other symptoms can be found when the chondrosarcoma grows large or aggressive; nevertheless, the prognosis at this point is even worse owing to the long period before treatment.

To the best of our knowledge, only 10 cases have been reported regarding the arytenoid cartilage (Table 1). Chondrosarcomas of the arytenoid cartilage most often occurred in elderly men according to previous reports,5–12 eight males
versus two females ranging in age from 46 to 78 years. Their main symptoms were hoarseness (eight cases\(^5\),\(^6\),\(^10\),\(^12\)) and/or dyspnea (four cases\(^5\),\(^10\),\(^12\)). Using laryngoscopy and radiology, obvious neoplasms can be found in the larynx in eight cases,\(^5\),\(^8\)–\(^12\) and not been mentioned in the other two cases.\(^6\),\(^7\) However, our case had an atypical history and laryngeal signs. The patient experienced dyspnea only for 1 month after a cold infection. The stroboscopy only showed that the right vocal fold was fixated and the left vocal fold was incompletely limited, but no neoplasm can be found. In this case, the laryngeal CT and LEMG played very important role in the diagnosis. The cervical-enhanced CT showed no typical signs of a mass, just instead of a limited region of high density in the arytenoids, especially on the right side. In addition, no significant abnormality of bilateral recurrent laryngeal nerves was found from LEMG examination, which demonstrated that the arytenoid lesion itself may be the real cause to limit vocal fold mobility. So, we decided to perform CO\(_2\) laser arytenoidectomy and biopsy to identify the pathological changes of arytenoid. We found that the texture of right arytenoid was harder than usual, and the pathology showed chondroma. So, we firstly reported that LEMG and laryngeal CT are necessary to the patients with idiopathic vocal fold immobility, and a biopsy of the arytenoid cartilage is indispensable at times.

Chondroma and low-grade chondrosarcoma are difficult to distinguish by histology, which increases the difficulty of their treatment. We considered that noncartilaginous texture of arytenoid may suggest its pathological changes. In this case, although some cells showed signs of cellular atypia, the original pathological diagnosis was chondroma with the solid texture. However, the texture of residual arytenoid was like sand after 22 months, which might potentially indicate its tendency to malignant transformation. The eventual diagnosis was low-grade chondrosarcoma, and apparent karyokinesis and pathological karyokinesis of tumor cells could be seen.

Oneal and Ackerman\(^14\) divided chondrosarcomas into three histological levels, namely low, intermediate, and high grades. An increase in the histological grade of the chondrosarcoma also increased the risk of tumor recurrence. Rozeman\(^15\) stated that the histopathological grade was the most important factor related to chondrosarcoma recurrence, as all prognostic indicators are inseparable from the pathological grades. In the previously reported 10 cases of laryngeal chondrosarcoma of the arytenoid cartilage, six cases were low-grade chondrosarcomas with very favorable prognoses.\(^6\),\(^7\),\(^9\)–\(^12\) Two cases were high-grade chondrosarcoma and had a recurrence after the surgery; one died because of metastases. One case was moderate-grade chondrosarcoma, who also had an unfavorable prognosis.\(^5\) The last case was myxoid chondrosarcoma, a rare type vulnerable to distant metastases that leads to a poor prognosis.\(^8\) The patient in our report had a low-grade chondrosarcoma and had a favorable prognosis after the surgery.

At present, surgery is the primary treatment for laryngeal chondrosarcoma, including endoscopic removal, laryngofissure removal, and partial or total laryngectomy.\(^16\)–\(^18\) The laryngeal chondrosarcoma rarely metastasized to the cervical lymph node; therefore, neck dissection was not a routine procedure. Neither radiotherapy nor chemotherapy was a routine treatment method owing to their insensitivity to chondrosarcoma. In the previously reported 10 cases of laryngeal chondrosarcomas in the arytenoid cartilage, three patients underwent endoscopic arytenoidectomy\(^9\),\(^11\),\(^12\); and of them, two patients ultimately underwent a laryngofissure removal\(^11\) and a total laryngectomy,\(^12\) respectively. Three patients underwent laryngofissure removal\(^5\),\(^10\) and one of them underwent a total laryngectomy\(^5\) finally. One patient underwent a partial laryngectomy,\(^5\) whereas other three patients underwent a total laryngectomy directly.\(^6\)–\(^8\) Our patient underwent endoscopic arytenoidectomy first at the early stage and, ultimately, underwent a total laryngectomy when histologically confirmed as low-grade chondrosarcoma. Neither radiotherapy nor chemotherapy was chosen for him. The surgery was effective, and the prognosis is favorable.

CONCLUSIONS

Laryngeal chondrosarcomas of the arytenoid cartilage are rare. It is easily neglected, especially in those cases presenting with idiopathic vocal fold immobility without obvious signs of neoplasms. LEMG and laryngeal CT are necessary. Sometimes, a biopsy of the arytenoid cartilage is essential.
<table>
<thead>
<tr>
<th>Authors</th>
<th>Age/Sex</th>
<th>Symptom</th>
<th>Duration (mo)</th>
<th>Laryngoscope/Radiology</th>
<th>Location</th>
<th>Treatment</th>
<th>Pathology Grade</th>
<th>Follow-Up</th>
</tr>
</thead>
<tbody>
<tr>
<td>Huizenga and Balogh</td>
<td>60/M</td>
<td>Hoarseness, dyspnea, and wheezing</td>
<td>6</td>
<td>Right arytenoid mass</td>
<td>Right arytenoid and posterior hypopharynx</td>
<td>Laryngofissure removal</td>
<td>High</td>
<td>NED: 3 y</td>
</tr>
<tr>
<td></td>
<td>63/M</td>
<td>Hoarseness and dyspnea</td>
<td>24</td>
<td>Large hypopharyngeal mass</td>
<td>Left arytenoid and interarytenoid</td>
<td>Radiotherapy Left</td>
<td>Moderate</td>
<td>Died postoperatively</td>
</tr>
<tr>
<td></td>
<td>68/M</td>
<td>Hoarseness, cough, dysphagia, and neck mass</td>
<td>8</td>
<td>A smooth intact laryngeal mucosa distorted by a submucosal mass</td>
<td>Bilateral arytenoids and interarytenoid Metastases: lung, kidney, and neck</td>
<td>Laryngofissure removal</td>
<td>High</td>
<td>DOD: 4 y</td>
</tr>
<tr>
<td>Finn et al</td>
<td>70/M</td>
<td>Hoarseness</td>
<td>36</td>
<td>NR</td>
<td>Right arytenoid</td>
<td>Total laryngectomy</td>
<td>Low</td>
<td>DOC: 12 y, DOD: 15 mo</td>
</tr>
<tr>
<td>Moran et al</td>
<td>61/M</td>
<td>Hoarseness</td>
<td>NR</td>
<td>NR</td>
<td>Left arytenoid metastases: lung and cerebra</td>
<td>Total laryngectomy</td>
<td>Myxoid chondrosarcoma</td>
<td></td>
</tr>
<tr>
<td>Lippert et al</td>
<td>78/M</td>
<td>Hoarseness</td>
<td>24</td>
<td>Left arytenoid and pyriform sinus mass</td>
<td>Left arytenoid</td>
<td>Total laryngectomy</td>
<td>Low</td>
<td>NED: 2 y</td>
</tr>
<tr>
<td>Rinaldo et al</td>
<td>46/M</td>
<td>Hoarseness and dysphagia</td>
<td>6</td>
<td>A smooth swelling on the right aryepiglottic fold</td>
<td>Right arytenoid</td>
<td>Endoscopic removal</td>
<td>Low</td>
<td>NED: 4 y (lost)</td>
</tr>
<tr>
<td>Shinhar et al</td>
<td>53/F</td>
<td>Hoarseness and dyspnea</td>
<td>216</td>
<td>A large supraglottic mass bulging from the posterior right larynx</td>
<td>Right arytenoid and cricoid</td>
<td>Laryngofissure removal</td>
<td>Low</td>
<td>NED: 2 y</td>
</tr>
<tr>
<td>Lee et al</td>
<td>56/F</td>
<td>Low pitched and no laryngeal complaints</td>
<td>—</td>
<td>A thick white plaque on the right arytenoid cartilage</td>
<td>Right arytenoid</td>
<td>Endoscopic removal</td>
<td>Low</td>
<td>NED: 3 y</td>
</tr>
<tr>
<td>Kanotra et al</td>
<td>47/M</td>
<td>Hoarseness and dyspnea</td>
<td>48</td>
<td>A smooth, mucosa-covered mass on the left arytenoid cartilage</td>
<td>Left arytenoid, Left arytenoid, cricoid, and thyroid</td>
<td>Endoscopic removal</td>
<td>Low</td>
<td>NED: 11 y after irradiation</td>
</tr>
<tr>
<td>Present study</td>
<td>51/M</td>
<td>Dyspnea</td>
<td>1</td>
<td>Bilateral vocal fold paralysis</td>
<td>Right arytenoid and interarytenoid</td>
<td>Endoscopic removal</td>
<td>Low</td>
<td>NED: 2 y 7 mo</td>
</tr>
</tbody>
</table>

Abbreviations: NED, no evidence of disease; DOD, dead of disease; NR, not reported; DOC, dead of other causes.
REFERENCES