Subglottic chondrosarcoma treated with transoral laser microsurgery: Case series and literature review

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ABSTRACT

Introduction: Subglottic chondrosarcoma (SGC) is a rare entity that is traditionally treated with open resection. We detail two cases treated with CO₂ transoral laser microsurgery (TLM) and present a literature review of minimally invasive approaches to this disease. Retrospective chart review and search of English language literature are important methods of data collection. Case Series: Two patients were identified. Both presented with worsening dyspnea and stridor. One patient had intermediate grade disease and the other had low grade disease. Both patients were successfully treated with TLM and remain disease free at 42 and 17 months respectively with good voice and functional outcomes. Conclusion: We present two cases of SGC treated with curative intent minimally invasive TLM. In both cases, there is no evidence of disease recurrence or functional limitation at follow-up. These findings are in line with the favorable oncological and functional outcomes demonstrated in the literature describing SGC treatment with TLM.

Keywords: Chondrosarcoma, Conservative surgery, Larynx, Subglottic, Transoral laser microsurgery

INTRODUCTION

Primary laryngeal chondrosarcoma is rare, comprising 0.2% of all head and neck malignancies [1–5]. Despite this, it is the third most common laryngeal malignancy following squamous cell carcinoma and adenocarcinoma [5]. The typical age at presentation is between 60 and 65 years, suggesting age-related inflammation and changes in the ossification process contribute to development of dysplasia [6]. Presenting symptoms depend on the anatomic location of the lesion, the most common being hoarseness (74%), dyspnea (56%), dysphagia (21%), and neck mass (21%) [6, 7].

The majority of laryngeal chondrosarcomas are low grade, well differentiated lesions and originate from the cricoid cartilage [8, 9]. Typically, cricoid lesions grow posterolaterally and invade the glottis and supraglottis [6, 10]. More rarely, lesions can involve the subglottic space [10]. Given the small number of cases of subglottic chondrosarcoma, there is a relative paucity in the literature regarding the optimal treatment approach. The existing literature suggests surgical excision to be the most effective modality, with radiotherapy reserved for non-resectable, recurrent or high-grade lesions, and...
no clear role for adjuvant therapies [10, 11]. Several authors show no survival advantage in total versus partial laryngectomy with low to intermediate grade tumors [2]. Here, we present two cases of low to intermediate grade subglottic chondrosarcomas successfully treated with minimally invasive transoral laser microsurgery (TLM), delineating the role of TLM in conservative management of laryngeal chondrosarcoma.

This case series was performed under the Nova Scotia Health Authority Research Ethics Board guidelines for case reports. No formal research ethics board approval was necessary and therefore no reference number was generated. This paper was accepted for presentation at the Canadian Society of Otolaryngology–Head and Neck Surgery AGM, poster presentation, June 1–3rd, 2019, as a poster presentation.

CASE SERIES

This report was prepared in accordance with the CAseREport (CARE) guidelines. A single reviewer (JM) systematic literature review was carried out using MEDLINE and Google Scholar. The search was performed with the terms “chondrosarcoma” and “laryn*” and (“laser” or “conservative”) from database inception to 18 June 2019. Snowballing and reference review techniques of relevant articles were performed in order to identify additional studies that could potentially meet inclusion.

Case 1

The patient presented with worsening stridor and dyspnea with minimal exertion. There were no other upper aerodigestive tract symptoms at presentation. Past medical history was unremarkable. Computed tomography (CT) with contrast showed a 2 cm mass originating from the left aspect of the cricoid cartilage, nearly obstructing the subglottic airway (Figure 1). There was no lymph node involvement. Awake tracheostomy was performed on an urgent basis for upper airway obstruction. Concurrent biopsy was performed, and the pathology report indicated low grade chondrosarcoma. The patient subsequently underwent excision of the tumor via a TLM approach with CO₂ laser. Intraoperatively it appeared that the lesion originated from the left aspect of the cricoid cartilage and extended superiorly under surface of the left vocal cord but did not involve the cord itself. The entirety of the left side of the cricoid cartilage was removed and an incision on the undersurface of the left vocal cord was required, with preservation of the thyroarytenoid muscle. Due to the nature of the excision, margins could not be taken, and pathological report of the specimen revealed a subglottic chondrosarcoma, intermediate grade (WHO grade 2). Repeat exam under anesthesia and re-excision was planned and pathology report for these specimens was negative for recurrence or persistent disease. There were no peri- or postoperative complications associated with either surgery. The patient was successfully decannulated and had satisfactory patient perceived voice outcomes. Ongoing surveillance via in-office bronchoscopy has revealed no evidence of recurrence at the three year visit.

Case 2

The patient presented with a six month history of progressive dyspnea, worse on exertion. Computed tomography scan was suspicious for a chondroid lesion. Biopsy was performed and showed low grade chondrosarcoma involving the cricoid cartilage. The patient underwent a right partial laryngectomy and right arytenoidectomy via TLM. The tumor appeared to originate from the right posterolateral aspect of the cricoid cartilage. Pathologic examination of the specimen demonstrated a low grade, well differentiated chondrosarcoma with increased cellularity, binucleation, nuclear enlargement, and hyperchromasia (Figure 2).

Again, due to the nature of the excision, oncological margins could not be determined. There were no perioperative complications and the patient returned to their baseline diet and breathing. There remains a
small amount of reduced right vocal cord mobility due to arytenoid resection, but no subjective functional complaints indicating a satisfactory patient perceived voice outcome. Current follow-up shows no evidence of disease at the one year surveillance period.

DISCUSSION

Laryngeal chondrosarcoma is a rare malignant neoplasm typically treated with open laryngectomy or endoscopic palliative debulking procedures [1, 6]. Herein we have presented two cases of subglottic chondrosarcomas treated with minimally invasive, curative intent TLM. Both patients have had excellent oncological and functional outcomes.

The primary treatment for laryngeal chondrosarcoma is complete surgical excision [6, 10]. Until recently, all laryngeal chondrosarcomas were traditionally treated with total laryngectomy based on the aggressive nature of cartilaginous malignancies at other sites. Mounting evidence suggests that laryngeal chondrosarcomas are distinct from those at other sites. Typically, laryngeal chondrosarcomas are well differentiated, low grade tumors with five year survival rates reported between 91% and 95% and five year disease free rate approximated at 72% following conservative treatment [12, 13]. Histologic grading of chondrosarcomas is the most important predictor of clinical behavior and prognosis [14, 15]. The World Health Organization (WHO) 2013 criteria use nuclear size, staining pattern (hyperchromasia), mitotic activity, and degree of cellularity to histologically differentiate lesions into Grades 1 (low), 2 (intermediate), and 3 (high) [15]. Given that total laryngectomy is associated with significant morbidity, including dysphagia and change in speech, organ sacrificing treatment is reserved for high grade chondrosarcomas involving more than half of the cricoid cartilage, invading surrounding tissues or involving the external perichondrium, and lesions refractory to conservative interventions [6, 7, 10, 12].

Establishing the optimal conservative modality for low to intermediate grade chondrosarcoma has been challenging given the small number of cases [7]. Endoscopic surgery with TLM has been proposed as a first line modality as patients require shorter postoperative hospitalizations and have improved potential for rehabilitation of speech and swallowing function when compared to open approaches [10, 12, 13]. Consequently, TLM improves quality of life and reduces years of morbidity associated with the treatment of laryngeal chondrosarcomas [12, 13].

Transoral laser microsurgery has equivalent locoregional control and overall survival outcomes, as well as reduced morbidity, when compared to open partial resection in the treatment of epiglottic and arytenoid originating chondrosarcomas [13]. Given cricoid lesions are more challenging to access transorally, TLM was introduced more recently for these lesions [13].

In a case series of seven low grade laryngeal chondrosarcomas (involving less than 50% of the cricoid cartilage, with extension to the glottis and cricoarytenoid joint) resected with TLM and endoscopic curettage, Pelliccia et al. (2014) found 100% survival and 14.3% recurrence at mean follow-up of 80 months [16]. One patient required re-resection for a limited recurrence, occurring at 21 months [16]. In a case series of 13 (6 chondromas, 7 chondrosarcomas) treated with KTP/YAG laser, Merrot et al. (2017) found 95% overall five year survival and 72% disease free five year survival [12]. Overall recurrence rate, requiring re-resection, was 54% [12].

Given the low incidence of subglottic laryngeal chondrosarcoma, there are fewer reports describing the use of TLM. The current literature describes 17 cases of CO₂ TLM resection of subglottic chondrosarcomas, as shown in Table 1. Most cases were performed for debulking purposes. However, primary resection was often the only intervention required. For lesions less than 2 cm in the largest dimension, TLM was used with curative intent [7, 10, 17]. In 13 of 17 documented cases, there was no evidence of disease recurrence or progression at follow-up (3 months to 6 year) (Table 1). In three of these cases re-resection was required, including one case that required three subsequent resections (Table 1).

Figure 2: (A) Low power microscopy showing hypercellular, ossified cartilage, with myxoid changes. (B) High power microscopy showing disordered cartilage with chondrocytes demonstrating binucleation, nuclear chromasia, and mild nuclear atypia, consistent with a low grade chondrosarcoma.
Four patients had progressive disease following TLM resection and required total laryngectomy, two of which had extensive disease at presentation and underwent debulking initially to delay radical surgery. Three of the patients only required total laryngectomy following symptomatic disease recurrence 18 months after initial endoscopic management (Table 1). Of those requiring total laryngectomy, two were found to have intermediate grade disease [18, 19]. There were no reported perioperative complications [7, 10, 17–20].

Table 1: Summary of current literature describing treatment intent/course and outcome of sub-Glottic laryngeal chondrosarcoma cases managed with CO₂ TLM

| Authors             | Treatment                                                                 | Treatment intent | Outcomes                                                                 |
|---------------------|---------------------------------------------------------------------------|------------------|--------------------------------------------------------------------------|
| Sauter et al. (2007) [7] | (1) Initial partial resection using CO₂, re-resection 1 month following.  (2) R side cordectomy using CO₂ TLM. | (1) Debunking    | (1) NED 3 m F/U.                                                          |
| Cattaneo et al. (2016) [10] | Complete resection of subglottic lesion using CO₂ TLM, with temporary tracheostomy. | Curative         | (2) NED 3 m F/U.                                                          |
| Damiani et al. (2014) [17] | (1) Complete resection of cricoid lesion with extension to inferior paraglottic space using CO₂ TLM.  (2) Complete resection of subglottic lesion using CO₂ TLM, with temporary tracheostomy. Remodeling with YAG. (3) Complete resection of cricoid lesion using CO₂ TLM, with temporary tracheostomy. Remodeling with YAG. (4) Complete resection of cricoid lesion with extension to inferior paraglottic space using CO₂ TLM. (5) Complete resection of cricoid lesion with extension to inferior paraglottic space and C-A joint using CO₂ TLM. (6) Complete resection of cricoid lesion with extension to C-A joint using CO₂ TLM. | (1) Curative  (2) Curative  (3) Curative  (4) Curative  (5) Curative  (6) Curative | NED in all patients at 31–60 m F/U. |
| Rinaldo et al. (2000) [19] | (1) Partial resection using CO₂.  (2) Two resections using CO₂ TLM to improve airway. (3) Partial resection using CO₂. (4) Four consecutive resections using CO₂. | (1) Debunking  (2) Debunking  (3) Debunking  (4) Debunking | (1) No evidence of progression at 18 F/U.  (2) Partial laryngectomy, NED at 1 y 6 m F/U.  (3) Recurrence at 10 m F/U. Total laryngectomy performed.  (4) Died of other causes at 3 y F/U. NED progression. |
| Bon et al. (2014) [20] | (1) Partial resection of cricoid lesion using CO₂.  (2) Partial resection of cricoid lesion using CO₂. | (1) Debunking  (2) Debunking | F/U at 28 m. Both patients radiologically stable disease, with no evidence of progression. |
| Windfuhr (2003) [18] | (1) Two resections of subglottic lesion using CO₂.  (2) Resection of subglottic cricoid lesion using CO₂. | (1) Debunking  (2) Debunking | Both patients underwent total laryngectomy to obtain clear margins. One immediately following endoscopic management, one following disease recurrence at 8 m causing narrowing of tracheal diameter. |

NED: No evidence of disease; F/U: Follow-up; C-A joint: Cricoarytenoid fixation.
We present two cases of low to intermediate grade subglottic chondrosarcomas treated with curative intent TLM. At 1 and 3 year follow-up there is no evidence of disease recurrence or treatment associated morbidity. Neither patient reports loss of function or subjective voice complaints.

**CONCLUSION**

We present two cases of low to intermediate grade subglottic chondrosarcoma treated using TLM with curative intent. Neither patient shows evidence of disease recurrence or long-term complications, such as loss of function or subjective voice complaints. Traditionally, TLM in this setting has been reserved for debulking procedures. However, this work adds to a growing base of research reporting promising oncological and functional outcomes following the use of TLM with curative intent for subglottic chondrosarcomas. These findings are similar to those reported in the literature, further adding evidence to the effectiveness of TLM for curative treatment in select patients. Given these results, we suggest that TLM may be a viable alternative to open partial laryngectomy for removal of low to intermediate grade chondrosarcomas in the subglottic region with excellent functional outcomes.

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**Author Contributions**

Jenna MacDonald – Conception of the work, Interpretation of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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