International Journal of Otorhinolaryngology and Head and Neck Surgery
Luna-Ortiz K et al. Int J Otorhinolaryngol Head Neck Surg. 2022 Dec;8(12):977-980
http://www.ijorl.com
pISSN 2454-5929 | eISSN 2454-5937

DOI: https://dx.doi.org/10.18203/issn.2454-5929.ijohns20223047

Case Series

Cricotracheal resection: a rare procedure in adults

Kuauhyama Luna-Ortiz[1]*, Cesar O. Cabrera-Ochoa[1], Luis C. Zacarias-Ramon[1], Zelik Luna-Peteul[3], Dorian Y. García-Ortega[2]

[1]Department of Head and Neck Surgery, [2]Department of Surgical Oncology, Universitatea de Medicinâ si Farmacie Grigorie T. Popa, Rumania
[3]Universitatea de Medicinâ si Farmacie Grigorie T. Popa, Rumania

Received: 23 August 2022
Revised: 06 November 2022
Accepted: 07 November 2022

***Correspondence:**
Dr. Kuauhyama Luna-Ortiz,
E-mail: kuauhyama@yahoo.com.mx

ABSTRACT

Benign or malignant tumors originating in the cricotracheal region (cricoid cartilage and first tracheal ring) are considered rare events in adult patients. Although there are several treatment options for benign tumors, cricotracheal resection remains the gold standard treatment for malignant tumors. The objective of the present study is to describe the cases that have been treated at our institutions. We selected five cases from a database of 800 clinical records. Three cases were treated at the national cancer institute, Mexico, and two in other institutions. All cases were treated by the same surgeon. Out of four women and one man, with a median age of 52 years, there were two cases of adenoid cystic carcinoma, two of idiopathic stenosis, and one of hemangiopericytoma. All underwent cricotracheal resection with laryngotracheal anastomosis and are still alive. Stenosis or primary tumors of the cricotracheal region can be initially treated with endoscopy. However, cricotracheal resection remains the gold standard treatment as it is a safe surgical procedure with minimal and acceptable sequelae. It also improves disease-free period, overall survival, and quality of life, albeit with a low and acceptable morbidity rate.

Keywords: Subglottis, Idiopathic stenosis, Cricotracheal resection, Adenoid cystic carcinoma, Hemangiopericytoma, Larynx

INTRODUCTION

Cricotracheal resection is considered a surgical option in the management of pediatric patients, in which prolonged intubation results in stridor and dyspnea secondary to stenosis.[1] Conversely, it is a procedure rarely performed in adults. The indications for benign and malignant entities vary, including post-intubation stenosis, extra-laryngeal tumors such as those of the thyroid carcinoma invading the trachea (5-8%) or with stenosis (0.5-1.5%), idiopathic progressive subglottic stenosis, endoluminal primary tumors of the larynx, and, to a lesser extent, laryngeal trauma.[2,3] Endoscopy (with or without laser) or open surgery can be performed to treat benign or malignant laryngotracheal stenosis lesions.[3] The objective of this study is to describe a series of patients treated with cricotracheal resection, a safe surgical option.

CASE SERIES

A retrospective, descriptive, observational, cross-sectional study of a series of cases obtained from a database of 800 clinical records of laryngeal neoplasms treated from 1990 to 2018. This study was reviewed and approved by the institutional review board. We found five patients (0.6%) with a complete clinical record, computed tomography of the larynx, and preoperative nasofibrolaryngoscopy. The same surgeon performed cricotracheal resection and laryngotracheal anastomosis on all five patients, who were followed-up to assess the presence of dyspnea, voice quality, and recurrent/ persistent disease.

Subglottic neoplasms are extremely rare. They accounted for 0.6% at our institution.

Case 1

A 46-year-old female admitted in September 2012 with a six-month history of progressive stridor. Naso-fibrolaryngoscopy revealed a 2×1 cm cricoid cartilage lesion. Cricotracheal resection was performed without complications. The histopathological analysis showed lymphoid hyperplasia, chronic inflammation and ulceration, and idiopathic subglottic stenosis. The patient did not show signs of recurrence or sequelae.

Case 2

A 65-year-old female with two-year h/f progressive dyspnea who developed stridor. An otorhinolaryngologist had performed transoral partial subglottic resection in May 2017, but symptoms progressed. Patient then came to our hospital, we performed endoscopy and found tracheal stenosis. Consequently, she underwent cricotracheal resection without tracheostomy. Histopathological analysis showed idiopathic subglottic stenosis. She is alive and with mild dysphonia.

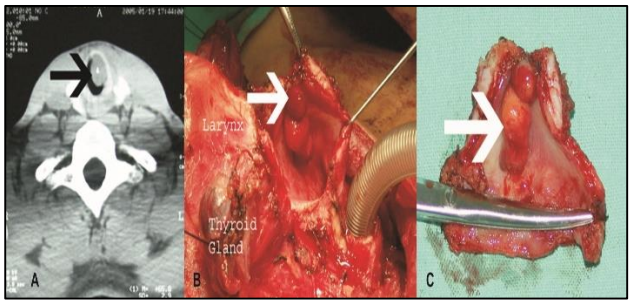

Figure 1 (A-C): CT scan showing the tumor in the cricotracheal region. Tracheostomy up to the carina after cutting into the thyroid gland. Resected tumor.

Case 3

A 24-year-old male developed hematemesis and dyspnea in December 2004. He was referred from another institution, where nasofibrolaryngoscopy and computed tomography of the larynx revealed a 3×2 cm polypoid lesion in subglottic region of the anterior part of cricoid cartilage. Residual tumor remained after endoscopic laser resection in March 2005. Thus we performed crico-tracheal resection without tracheostomy in May 2005. Patient developed a hematoma day after surgery and underwent cervical examination on third postoperative day. A nasogastric tube was placed for 10 days, and patient discharged eight days later. Histopathological report described a residual tumor of 1.7×0.7×0.6 cm. Tumor composed of polygonal and spindle-shaped cells arranged around blood vessels. Positive immunohistochemical reactions were found for vimentin, while they negative for actin, CD34, synaptophysin, chromogranin A, CD21/35, cytokeratins AE1/AE3, epithelial membrane antigen, HMB45, ALK-1, BCL-2, and S-100. Diagnosis malignant hemangiopericytoma. Follow-up showed no signs of functional sequelae.

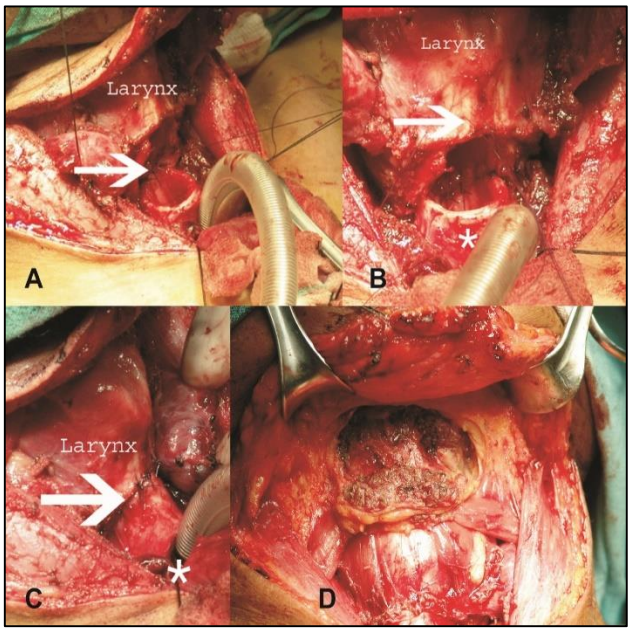

Figure 2 (A-D): Larynx and anastomosis of the posterior subglotis to the membranous trachea with anterior sutures; the asterisk shows the trachea. Anastomosis using simple sutures. If needed, strain is reduced by cutting the thyrohyoid muscle.

Case 4

A 61-year-old female was admitted for the first time in January 2007. Her condition had begun with asthenia and adynamia two years earlier. She then complained of headaches and foreign body sensation in the pharynx; she received treatment for bronchial asthma. Progressive dyspnea was added to her list of symptoms. A CT scan showed lumen reduction of the cricotracheal region. Clinical examination revealed dysphonia and laryngeal stridor loud enough to be heard at some distance. Nasofibrolaryngoscopy showed stenosis by exophytic tumor in the cricotracheal region with 90% lumen obstruction. We performed a cricotracheal resection without tracheostomy in January 2007 due to a tumor measuring 1.8×1 cm, located on the right anterior aspect of the subglottic region that extended up to the third tracheal ring. The tracheostomy cannula was removed on the eighth day without complications. The patient was discharged nine days after surgery. The histopathological report showed a subglottic grade II adenoid cystic carcinoma invading the cricoid cartilage, extending to the third tracheal ring and thyroid gland, and exhibiting perineural and perivascular infiltration. Macroscopically, the lesion measured 2.5x0.8 cm, posterosuperior border was in contact with the tumor, and right thyroid lobe was infiltrated. Adjuvant radiotherapy was administered until May 2007 (70 Gy in 35 sessions). Side effects included mucositis and hypothyroidism, which were treated medically. A 10-year follow-up revealed no recurrence.

Case 5

A 66-year-old female with a history of systemic arterial hypertension and controlled heart failure was admitted in June 2012. She had had dyspnea for two years, which had been misdiagnosed as bronchial asthma. She was referred from another institution with a diagnosis of subglottic tumor, for which she had undergone endoscopic resection. The histopathological analysis had shown adenoid cystic carcinoma. The patient underwent cricotracheal resection of the inner aspect of the cricoid cartilage without tracheostomy. The patient was discharged four days after surgery without complications. Histopathological analysis confirmed the diagnosis of grade II adenoid cystic carcinoma with superficial infiltration of the perichondrium and cricoid cartilage. The tumor was 1.1×0.6×0.5 cm with negative surgical margins and Delphian node. The patient remained disease-free after a 7-year follow-up.

DISCUSSION

The two viable options for treating cricotracheal tumors are endoscopy with laser resection, or open surgery. These procedures were first described by Gerwat and Bryce in 1974 and later by Pearson in 1975. They were later refined and popularized by Grillo, especially for cases of benign and idiopathic origin. Moreover, these procedures allow for circumferential excision, preservation of tissue integrity, and restoration of continuity. Despite being associated with a high incidence of preoperative mortality, mainly due to cardiovascular and respiratory complications ranging from 15-39%, these procedures achieve complete resection of the lesion, with success rates of up to 95%.[4] Surgical procedures such as tracheal resection and anastomosis and cricotracheal resection and anastomosis is recommended for patients who are not eligible for endoscopic resection or underwent endoscopic resection but had persistent disease or symptoms secondary to post-intubation stenosis.[5-8] The of primary tracheal tumors is 0.142 per 100,000 inhabitants; most of which are squamous carcinomas (55%), followed by adenoid cystic carcinoma (45%), undifferentiated carcinoma (12.8%), small-cell carcinoma (9.7%), adenocarcinoma (5.9%), large-cell carcinoma (3.8%), sarcomas or secondary tumors (3.8%).[8] In our five patients, adenoid cystic carcinoma accounted for 40% of cases. There were two cases of idiopathic stenosis and one of hemangiopericytoma.[5]

Treatment of subglottic tumors depends on several factors, including but not limited to age, overall health, comorbidities, extent of disease, presence of regional or distant metastases, histologic type, and radiosensitivity. However, in heterogeneous tumors, for which chemotherapy and radiotherapy have poor results or many side effects, surgery remains the gold standard, and conservative surgery should be the treatment of choice.[5] Contraindications include extensive tumors for which safe anastomosis is not feasible, metastatic disease, or resections that would have serious functional sequelae.[9] Several factors must be considered during surgical planning: a) exact location and extension of the tumor, both inside and outside the airway, b) amount of normal trachea that will remain after planned resection, c) age and body complexion-in patients with a short and stiff neck, resection would be limited to no more than 4 cm; whereas young, thin, and tall patients may undergo up to 6 cm resection, d) history of previous cervicothoracic surgery, infection, or irradiation.[9] In a series of 38 patients who underwent cricotracheal resection due to post-intubation stenosis, on average six tracheal rings were resected. All patients with tracheostomy (n=29) were successfully decannulated. No mortality was observed. The type or number of comorbidities was not related to the extent or type of resection.[10] Furthermore, it is worth mentioning that tracheostomy is rarely necessary because the laryngeal lumen is hardly affected in adults. The only factor to consider before performing it would be a bilateral lesion of both recurrent laryngeal nerves. In one case of this series, a recurrent nerve was sacrificed due to the proximity of the tumor. so an adequate location should be performed prior to tumor resection and at the present time this surgery should be considered, under neuromonitoring of the recurrent nerve. However, if the surgeon is not confident to leave the patient without a tracheostomy it will always be an excellent option to do it.

Endoscopic resection is an option for progressive idiopathic subglottic stenosis. Diagnosis is made after excluding infection, inflammation, trauma, gastroesophageal reflux disease, recurrent polychondritis, or Wegener's granulomatosis as causes of stenosis.[3] Open surgery is indicated for persistent and recurrent tumors or that cannot be endoscopically resected.[4]

Pariket et al demonstrated that surgery (cricotracheal resection) improves overall survival in patients with subglottic adenoid cystic carcinoma.[11] The subglottis is a laryngeal subsite that has generated controversy regarding its anatomy, incidence of primary tumors, and histology. There are different views on the proper management of malignant tumors in the subglottic region. Endoscopic surgery is a viable option after assessing whether there is residual or recurrent tumor, in which case a more radical approach would be necessary to achieve R0 margins. For instance, cricotracheal resection remains the treatment of choice for adenoid cystic carcinoma and hemangiopericytoma. In such cases, the objective should be to secure a safe airway and tracheal blood supply in order to resect only the trachea length that can be safely reconstructed to avoid excessive anastomotic tension.[12] Postoperative complications (granulation, restenosis, and dehiscence) are rare, but they can have high morbidity. Dehiscence has been the main complication in some series (up to 4%).[3] We did not observe such complications in our case series, probably because we normally perform two technical procedures.

We resect the anterior trachea, as we usually do in supra-cricoid laryngectomies, and if tension is still high, we release the larynx by cutting the thyroid muscles.

Individuals aged over 70 years or with cardiovascular and pulmonary comorbidities are at higher risk for postoperative complications. However, cricotracheal resection is the best treatment option, and the risk will have to be taken if endoscopy fails.[4] The largest series ever reported showed that 4% of the patients had a permanent tracheostomy or T-tube and a mortality rate of 1%.[13]

CONCLUSION

Stenosis or primary tumors of the cricotracheal region can be initially treated with endoscopy. However, cricotracheal resection remains the gold standard treatment for primary, persistent, or recurrent cases or when the cricotracheal region was secondary infiltrated. Morbidity and mortality rates are low after cricotracheal resection, and tracheostomy is rarely necessary.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Luna-Ortiz K, Cabrera-Ochoa CO, Zacarias-Ramon LC, Luna-Peteul Z, García-Ortega DY. Cricotracheal resection: a rare procedure in adults. Int J Otorhinolaryngol Head Neck Surg 2022;8:977-80.