



Supraglottic Paraganglioma-Successful Excision with Transcervical and Midline Thyrotomy Approach: A Surgical Challenge

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Abstract

Laryngeal paragangliomas are rare submucosal lesions. They are usually non-secretory in nature. The clinical presentation of the lesion varies from hoarseness, airway obstruction, and/or swallowing difficulty. The mainstay of the treatment for laryngeal paraganglioma is surgical excision of the lesion. The smaller tumors require endoscopic excision while the large tumors require external approach for excision of the lesion. This report discusses the surgical approach and challenges of a transcervical midline thyrotomy approach for excision of the large laryngeal paraganglioma lesion.

Introduction

Paragangliomas are rare neuroendocrine tumors that uncommonly involve the larynx. The laryngeal paraganglioma are reported to derive from non-neural cells of the superior laryngeal nerve or the recurrent laryngeal nerve [1,2]. They are usually non-secretory in nature. The clinical presentation of the lesion varies from hoarseness, airway obstruction, and/or swallowing difficulty [3]. The mainstay of the treatment for laryngeal paraganglioma is surgical excision of the lesion. The smaller tumors require endoscopic excision while the large tumors require external approach for excision of the lesion [2,3]. We report a large laryngeal paraganglioma which was completely resected by transcervical and midline thyrotomy approach. The surgical technique and challenges are discussed.

Case Presentation

A 27 years old female patient, presented to our hospital with hoarseness for 9 months, which was insidious in onset and gradually progressive in nature. She experienced difficulty in breathing for the last 3 months which aggravated to develop stridor. She underwent tracheotomy at a local hospital and referred to us for further management. The patient did not report any dysphagia. There was no history of similar illness in the family. The examination revealed a firm smooth swelling measuring approximately 3 cm × 3 cm in size over the left side of the neck between hyoid and thyroid cartilage. The swelling was pulsatile with a bruit heard over it. A Video-laryngoscopy revealed a mucosa covered hyperemic swelling on the left side of the larynx in the supraglottic region pushing the epiglottis towards the right. The whole of the laryngeal inlet was obliterated and the swelling was seen to reach posteriorly up to the left arytenoid cartilage (Figure 1).

The computerized tomography scans showed a large enhancing lesion measuring around 5 cm × 3.8 cm × 3.7 cm, located in the left supraglottic region with area of an internal necrosis.

The mass was pushing the epiglottis superiorly and to the right and obliterated the laryngeal lumen (Figure 2). The patient underwent 68Ga DOTATATE whole body PET/CT which showed a similar extent of the tumor as in the computed tomography. The Digital subtraction angiography revealed principle arterial feeder to the tumor from the superior thyroid artery and ascending pharyngeal artery on the left side. The tests for secretory nature of the neuroendocrine tumour were negative.

The surgical excision of the lesion was done via an external approach. An inverted hockey stick incision was made over the left side of the neck from the level of the hyoid to the inferior border of the thyroid cartilage. The left common carotid artery along with the internal jugular vein were identified and vascular control achieved using vascular loops. The superior thyroid artery was ligated as it contributed to the vascular supply of the lesion (Figure 3). The lesion was seen to get

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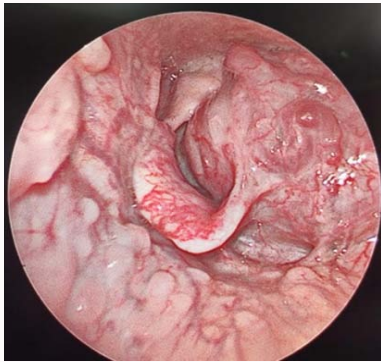


Figure 1: 90° endoscopic view of the submucosal bulge in the left supraglottic region due to the lesion.

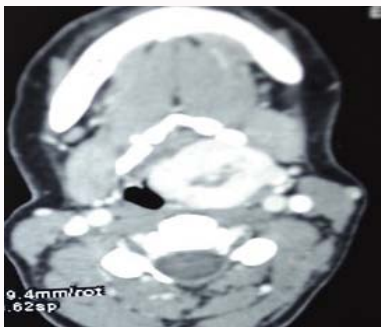


Figure 2: Computerized tomography on the axial plane showing a large enhancing lesion in the supraglottic region with area of necrosis.

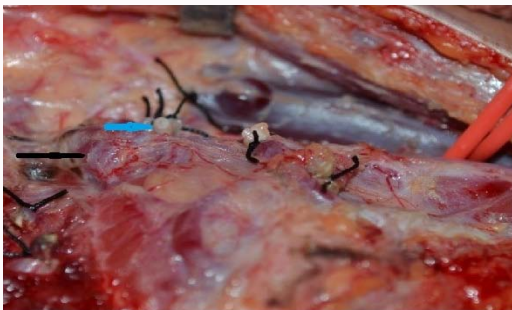


Figure 3: Showing the ligated superior thyroid artery (blue arrow) and the tumour (Black arrow). The IJV and CCA can be seen.

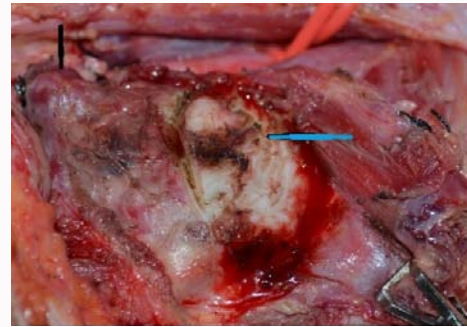


Figure 4: Showing the tumour at the thyrohyoid membrane (black arrow) and exposed thyroid lamina (blue arrow).

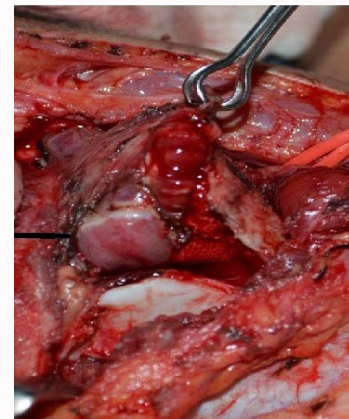


Figure 5: Showing the split thyroid lamina with the tumour in the laryngeal inlet (Black arrow).

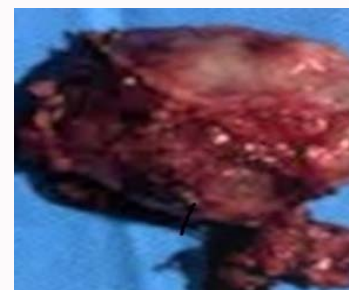


Figure 6: Showing the well lobulated excised specimen.

into the larynx through the thyrohyoid membrane and had splayed the area (Figure 4). At this stage of dissection, a midline thyrotomy was performed which enabled the full view of the intra laryngeal part of the lesion (Figure 5). The median thyrotomy provided proper and head-on exposure of the lesion. The tumor was extending inferiorly reaching just above the left true vocal cord and had obliterated the ventricle. The thyrotomy also enabled the surgical team to visualize the tumor precisely and have an enough working space for the preservation of the normal vital structure in the larynx. Ryle’s tube was inserted under vision to avoid injury to the esophagus while removal of the lesion.

The specimen measured 4 cm × 2 cm × 2 cm in size (Figure 6). The tumor was a firm vascular mass superiorly attached to the left half of epiglottis pushing rest of epiglottis toward the right, laterally attached to left aryepiglottic fold and extending inferiorly to the upper border of left true vocal cord. The lesion was attached to

internal surface and superior cornu of the thyroid cartilage breaching the thyrohyoid membrane. It was also attached posteriorly to the arytenoids cartilage. The split thyroid cartilage was repaired using 4-0 prolene with proper alignment and opposition. The wound was closed in layers with a corrugated open drain. The patient remained on nasogastric feeds for two days. A video-laryngoscopy seven days later showed bilateral vocal cords mobile and there is no sign of aspiration. He was decannulated on day 8, with satisfactory voice quality. To improve further, the patient started on speech therapy. There is no complication observed in short term follow up. The histopathological examination showed tumour cells arranged in nests or trabecula separated by thin fibro vascular septate, giving a zellballen pattern and suggestive of Paraganglioma (Figure 7).

Discussion

The Paragangliomas are neuroendocrine tumors that arise from

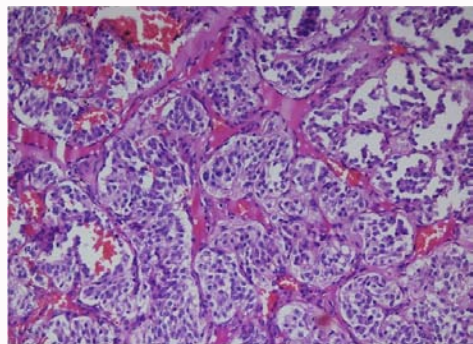


Figure 7: Histopathology specimen of the excised specimen showing a typical Zellballen pattern.

the paraganglion system. There are two pairs of laryngeal paraganglia rests. The first pair is located in the edge of the thyroid cartilage and false vocal fold along the course of the superior laryngeal artery and nerve i.e. superior laryngeal paraganglia. The inferior paraganglia are located around the inferior cornu of the thyroid cartilage to cricoid cartilage [2]. In our case, the site of origin could probably be the superior laryngeal paraganglia cell rest, which accounts for 90% of the laryngeal paraganglioma [4,5]. The laryngeal paragangliomas are uncommon submucosal lesions arising in the supraglottis. It has been reported more in females with female to male ratio being 3:1. The laryngeal paragangliomas occur during the fourth to sixth decades of life while our case was relatively young at 27 years [4]. Usually, the lesion is non-secretory depending upon the size and site, the symptoms associated with lesion are hoarseness, dyspnea, stridor, dysphagia and/or foreign body sensation, and similar reported in our case except for dysphagia and foreign body sensation [3-5]. On physical examination, the superior laryngeal paraganglioma typically appears as a lobulated, smooth mass in the false vocal cord region. The larger lesion herniates through the thyrohyoid membrane, with a presentation similar to our patient [2,6]. A contrast CT and/or gadolinium-enhanced MRI is advised to evaluate the extent and relationship to adjacent structures and the great vessels. With current imaging modality, the Gadolinium-enhanced MRI is imaging modality of choice [7].

Laryngeal paragangliomas need to be differentiated from carcinoid tumors, small cell neuroendocrine carcinoma and medullary carcinoma. The diagnostic imaging by 68Ga DOTATATE PET/CT helps to determine the extent and multifocality of the lesion. A similar approach was selected in our case [7]. However, 111 in pentetreotide scanning can distinguish a neuroendocrine tumor from other submucosal lesions.

The main blood supply to the laryngeal paraganglioma is the superior laryngeal artery, a branch of the superior thyroid artery. The super selective angiography permits delineation of the blood supply and detection of additional synchronous cervical paraganglioma. Some authors advocate selective embolization of the tumors while others advise simple arterial ligation of the feeder vessel during surgery to accomplish a similar result. The above technique was used in our case to control the bleeding intraoperatively [8].

The location and symptomatic presentation necessitate an appropriate planning for the surgical excision. The foremost goal of the treatment is complete excision of the tumor along with the preservation of laryngeal function. The smaller lesions can be subjected to endoscopic removal. However, recurrences have been reported on long term follow up [2,6,7,9].

The CO₂ laser excision is also an alternative modality for smaller lesions [10]. The open surgical approach results in superior disease control and complete excision of the lesion. The surgical steps in our case included the exposure and delineation of the planes up to the entry of the tumor through the thyrohyoid membrane. The ligation of the superior thyroid artery helped to control the vascularity of the tumour adequately. The transcervical access for control of great vessels and the midline thyrotomy ensured a complete exposure of the intralaryngeal component and its attachments. This facilitated the complete excision of the lesion while preserving the normal mucosa of the larynx and minimum disruption of laryngeal function. The open procedure has been reported to have a long disease-free state in subsequent follow up [2,6].

Conclusion

Laryngeal paragangliomas are uncommon tumors. An adequate exposure with transcervical and midline thyrotomy approach ensures complete removal with preservation of laryngeal functions.

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