Subglottic Leiomyoma

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ABSTRACT

Educational Objective: At the conclusion of this presentation, participants should be able to discuss soft tissue tumors of the subglottis and their management.

Objectives: To present a case with near complete obstruction of the subglottis and trachea by a leiomyoma.

Study Design: Case report and literature review.

Methods: Chart and literature review. Characteristic computed tomography, gross and pathologic images are demonstrated.

Results: The patient presented with a 3 year history of worsening of her shortness of breath and decline in her mental status over the past few days. She had previously been diagnosed as having asthma and was found to have a large obstructing mass in the subglottis and superior trachea on CT scan, and she required an emergent awake tracheostomy. Initial biopsies were suggestive but not diagnostic of leiomyoma. Endoscopic excision through a transcervical approach was performed.

Conclusions: Leiomyoma of the subglottis is an extremely rare clinical entity. Management should focus on establishing a safe airway and subsequent complete excision of the leiomyoma as recurrence is possible when sub-totally removed. This case illustrates the danger that so-called benign masses may pose to the airway.

INTRODUCTION

Leiomyoma is a benign smooth muscle tumor that can occur anywhere in the body where smooth muscle is found. They are most commonly associated with smooth muscle of the uterus or GI tract. Airway leiomyomas are quite rare and thought to arise from either smooth muscle fibers surrounding the airway or smooth muscle fibers in blood vessels giving rise to a so-called angioleiomyoma. Tracheal leiomyomas are thought to represent about 1.5% of all tracheal tumors (1). Airway leiomyomas may arise anywhere from the supraglottic larynx to lung parenchyma (2,3) though most commonly in the distal third of the membranous trachea (1).

Presenting symptoms depend on the location and size of the tumor, but cough, wheezing and dyspnea on exertion are the most common presenting complaints. (4) This case illustrates a “benign” clinical entity presenting at a very late stage with near fatal consequences.

CASE PRESENTATION

A 40 year old female was taken to an outside hospital for altered mental status and insomnia for the past few days. The woman reportedly had been diagnosed with asthma 3 years ago, which had been poorly responsive to medical therapy. Her shortness of breath had been gradually getting worse over the past 6 weeks. A CT of the chest at the outside hospital showed the inferior aspect of a tracheal mass. At this point she was transferred to our institution.

On examination in the emergency room she was initially extremely drowsy. She had no audible stridor or wheezing and had decreased breath sounds bilaterally. ABG revealed a PCO2 of 111. A CT of the neck showed near complete obstruction of her trachea and subglottis by a round, homogenous mass. The mass was slightly greater than 2 cm in all dimensions and located 2 cm inferior to the true cords. The mass was isointense compared to the membranous trachea. There was no peripheral or central enhancement.

She was taken to the operating room urgently for an awake tracheostomy. A tracheostomy was place between the 2nd and 3rd tracheal rings. Microinyangoscopy and rigid bronchoscopy at that time showed a smooth, firm, broad-based mass originating from the superior membranous trachea extending into the subglottis. The mass showed prominent superficial vasculature. Multiple biopsies were obtained using biopsy forceps.

Two days after the awake tracheostomy the patient was taken back to the OR for excision of the lesion. The mass was addressed using an anterior transcervical approach through the previous tracheotomy incision. The 2nd tracheal ring was removed for better access to the tumor. The leiomyoma was removed in a piecemeal fashion until it was completely removed. The mass appeared to extend through the membranous trachea, though did not appear to involve the esophagus. The wound was then left to heal by secondary intention. She was decannulated on postoperative day 3.

The patient is doing well post-operatively she reports doing well with no return of her shortness of breath and no signs of recurrence or stenosis.

DISCUSSION

This case illustrates an unusual presentation of a rare clinical entity. The patient was otherwise very healthy allowing her to compensate despite her severely narrowed airway. Progression to severe hypercapnea with altered mental status has not previously been reported in tracheal or subglottic leiomyomas.

Airway tumors involving the subglottis or trachea occur in less than 0.04/100,000 (5) with 90% of those being malignant (6). Adenoid cystic carcinoma and squamous cell carcinoma represent the vast majority of malignant tumors. Benign tumors arise from respiratory epithelium, salivary glands, neuroendocrine tissue, mesenchymal structures. Squamous papillomas, granular cell tumors, chondromas, pleomorphic adenomas, schwannomas, hemangiomas, and leiomyosarcomas are the most commonly reported benign tumors of the trachea (1,6). Appropriate diagnosis is perhaps the most important step when deciding the most appropriate treatment for these lesions. Differentiation between a leiomyoma and a leiomyosarcoma is extremely important, as leiomyomas are not thought to have any malignant potential.

Surgical approaches for resection of tracheal and subglottic leiomyomas are widely varied depending on the size and location of the tumor and previous experience with the lesions. Recommendations are typically for either tracheal resection or endoscopic excision depending on the size of the base of the lesion and location within the airway. Endoscopic excision is associated with a higher rate of recurrence (7). When performing endoscopic resection of leiomyoma, cauteterization of the base of the lesion with electrocautery or laser is generally recommended to prevent this. Significant bleeding resulting in death, though rare, has also been reported with endoscopic resections. (8) Transcervical approaches have generally been well tolerated with no recurrence reported. The gold standard for definitive removal according to most authors is a sleeve tracheal resection. The risk of anastomotic break down, granulation or stenosis leads many authors to reserve this for very broad-based lesions or for incidences of recurrent lesions. (9) In our patient the inferior aspect of the lesion was visible through the trachotomy thus providing good access without the need to create any new incisions. This provided better visualization and access than a trans-oral endoscopic approach would have afforded.

REFERENCES