A Hamartoma Located in the Trachea

Hamartoma is rarely found to be localized in the trachea. In the literature, only about ten cases have been reported. A 52-year-old male who was being treated for asthma for 15 years applied to our hospital with a progressive dyspnea complaint. During his physical examination, stridor was heard, after which a computed tomography of his chest revealed a tracheal mass. Fiberoptic bronchoscopy revealed a mass which obstructed 80% of the tracheal lumen attached to the posterior tracheal wall with a broad base. The mass was removed surgically with segmentary resection of the trachea. Histopathological examination of the lesion indicated that it was a hamartoma. Hamartomas can localize in the trachea very rarely, causing serious obstruction.

Keywords: trachea, hamartoma, resection

Introduction

Primary tumors of trachea are usually malignant (90%). Only 10% of them are benign. Amongst the benign tumors, fibroma, schwannoma, leiomyoma and hamartomas can be found. Hamartomas are the most frequently seen benign lung tumors. They frequently present as peripheral intraparenchymal nodules. Histologically, they are composed of cartilage, fat, bone and connective tissue, and smooth muscle cells. Endobronchial hamartomas make up only 1.4% of all hamartomas. Compared to endobronchial hamartomas, tracheal hamartomas are quite rare. In the literature, approximately 10 cases of tracheal hamartoma have been reported.

Case

A 52 years old male patient applied to our hospital with a complaint of progressive shortness of breath that has been going on for 15 years. Based on his asthma diagnosis, the patient had been using a bronchodilator and a steroid inhaler for treatment. He also received immunotherapy treatment for 2 years. In the patient’s history, there was nothing remarkable except a 12 packages/year cigarette use. During his physical examination, a significant stridor was determined. Complete blood count and routine biochemical parameters were in normal ranges. The result from the Pulmonary Function Test (PFT) was consistent with mild obstructive airway disease. The presence of stridor led to postulate that there might be a fixed airway obstruction and Thoracic Computed Tomography (CT) scans were obtained.

In the CT, a low density lesion was observed in the trachea, extending from the posterior to 2.5 cm proximal of the carina, causing a significant obstruction in the tracheal lumen (Fig. 1). Rigid bronchoscopy under general anesthesia was performed to obtain a tissue sample. We used intravenous agents exclusively to provide complete anesthesia (total intravenous anesthesia). The anesthesia induction was achieved using 2 mg/kg propofol, 0.05 mg/kg midazolam, 1 mcg/kg fentanyl and 0.5 mg/kg atracurium, and maintained using 1 mg/kg/h propofol, 0.05 mg/kg/h...
midazolam, 2 mcg/kg/h fentanyl and 0.5 mg/kg/h atracurium infusion, given during the procedure. At bronchoscopic examination, lobular contour lesion covered with smooth mucosa was observed in the distal trachea, located between 13th and 17th cartilaginous rings from the vocal cords, attached to the posterior wall with a broad base, obliterating the tracheal lumen by 80% (Fig. 2). Based on the biopsy obtained, it was reported as lipomatose tissue. Since the lesion was located in the trachea with a broad base, endoscopic treatment was not considered. Based on this, it was decided to remove the lesion using surgical methods. The proximal trachea was intubated with a left double lumen endotracheal tube. The tube was not inserted to distal trachea and tip of the tube was left at proximal of the tumor. After the right posterolateral thoracotomy, location of the tumor was determined at distal trachea. The tumor was not extending to the mediastinum but it was forming protuberance on the wall of the trachea. The trachea and both main bronchi were dissected. After careful examination, the lungs were ventilated with 100% oxygen for three minutes, and ventilation was stopped. Then, distal trachea was incised at proximal level of the tumor and distal level of the tumor was explored. The tracheal segment containing the tumor, approximately 3 cm in long was excised. The proximal and distal ends of the trachea were approximated by suturing its. Suturing was begun from the left side of the ends with 3-0 polypropylene, continued with running sutures, for three minutes after stopping the ventilation. Then the left double lumen tube was pushed forward to intubate the left main bronchus, and ventilation was restarted. During the procedure, a sterile endotracheal tube and sterile connection tubes were kept on the nurse’s table to intubate the left lung via thoracotomy incision, if needed, but were not used. Patient was well tolerated the apnea period and no significant hypoxia was developed (minimum level of oxygen saturation was 84%). Then end-to-end anastomosis was completed with running sutures using the same suture. The suture line was covered with the mediastinal pleura. No air leak was seen and the procedure was terminated. The patient was extubated in the operating room.

During the post-operative period, no complication was observed. The patient was discharged at 4th post-operative day.

As a result of the pathological examination of the resection specimen, although the lipomatose component was in the foreground, the lesion was decided to be lipomatose hamartoma since a cartilage structure was observed (Fig. 3). After the treatment, patient’s dyspnea completely disappeared. In our 1.5 year follow-up, no problems were encountered.

Comment

Primary benign tumors of trachea are neoplasms that
grow slowly which do not have the potential to become malignant. However, based on its localization, they can even cause death through airway obstruction. For this reason, early diagnosis is crucial. In the case where symptoms are non-specific, diagnosis is usually delayed and patients are followed for a long time with asthma diagnosis.

Hamartoma, which is one of the benign tracheal tumors, can also be called “cartilaginous hamartoma” or occasionally, “benign mesenchymoma.”

While the parenchymal ones contain more cartilage pathologically, endobronchial hamartomas consist mostly of fat tissues. Generally cartilage is not present or is found in very small amounts. Besides this, smooth muscle, seromucinous bronchial and chronic inflammatory cells can be seen. Thus, it is hard to arrive at the hamartoma diagnosis based on the small material obtained using bronchoscopic biopsy, as it is the case with our patient. Only lipomatous tissue could be obtained through biopsy. Due to the cartilage component found in the resection material, it was considered to be lesion lipomatous hamartoma. In these tumors, it can be difficult to establish a hamartoma diagnosis without obtaining large biopsy specimens.

Due to inflamed surface of the hamartoma, bronchoscopic appearance can not distinguish it from bronchogenic carcinoma, but thoracic CT, by showing high contrast resolution when the lesion contains an abundance of fat, can be particularly helpful in diagnosis. In the literature, including pediatric and adult patients, 10 cases of hamartoma with tracheal localization have been reported. During their bronchoscopy, most patients were observed to have polypoid lesions, causing lumen obstruction. In only one pediatric case, the lesion was extraluminal, presenting as a neck mass related to the trachea. In a study compiling information from 71 hamartoma cases, tracheal localization was reported in only 2 cases.

The treatment method of benign primary tracheal tumors varies, depending on the size and localization of the lesion. In benign tracheal lesions, more conservative treatments such as laser or endoscopic resection, are recommended. However, in the case where a mass invading the tracheal wall is present, surgical resection should be the initial choice. In our case, since the lesion was found to be attached to the tracheal lumen with a broad base, surgical resection was preferred without the consideration of endoscopic treatment.

As a result, we found that it is appropriate to present this case in order to emphasize that hamartomas, although rare, can be endotracheally located.

References