Case Report
Clinical characteristics and surgical management of primary carinal tumors

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Abstract: Objectives: The goal of this study was to summarize the clinical manifestations, diagnosis, surgical treatment, and prognosis of primary tracheal carinal tumors. The features of this disease were characterized to improve the quality of life of patients. Method: Clinical information of a patient was collected and presented for those with confirmed primary tracheal carinal tumor and having undergone therapy in our department. The related literature regarding primary carinal tumors (PCT) is also reviewed. Results: The patient was admitted to the hospital because of a chronic cough, which was further diagnosed as a PCT. The tumor was resected and his tracheal carina was reconstructed. Postoperative pathology revealed that the size of the tracheal tumor was 3 × 2.5 × 1 cm, and the microscopic images displayed moderate differentiation of squamous cell carcinoma and partly presented a papillary structure. Postoperative diagnosis was primary tracheal carinal squamous cell carcinoma. The patient recovered well after surgery and has been regularly followed up to date. Conclusions: Tracheal carinal tumors are rare, and the clinical manifestations are not typical; this type of tumor is easy to misdiagnosis. These tumors’ clinical diagnosis mainly relies on CT and bronchoscopy biopsy. Squamous cell carcinoma and adenoid cystic carcinoma are the most common types of trachea tumors. Surgical therapy is the most important treatment, and the main purpose of the treatment is relieving the airway obstruction. According to postoperative pathological staging, the doctors decide whether to offer chemotherapy and other follow-up treatments.

Keywords: Preoperative evaluation, surgical treatment, anesthesia management, postoperative complications, prognosis

Introduction

The incidence of primary tracheal tumors is very low, accounting for approximately 1%-2% of respiratory tumors. The symptoms in the early stage are not obvious and lack specificity. Therefore, it is easy to miss these diagnoses or to misdiagnose these tumors as an acute bronchitis, bronchial asthma, chronic bronchitis, bronchiectasis, or tuberculosis. In these situations, delay of the optimal treatment time is not good [1]. As the tracheal and carina tumors grow, they will cause airway obstruction. It is difficult to maintain oxygen supply and respiratory management during anesthesia. Therefore, the surgical risks are increased, and there is a high complication rate during the operation period. In recent years, owing to the progress made in surgical techniques and anesthesia assisted technology, increasingly complex trachea and carina tumors can be resected but these operations are difficult to handle in the [2]. In this article, we combine information regarding the clinical data of our patient with the reported literature regarding primary carina tumors to promote further understanding of the clinical characteristics with the hope to improve the diagnosis and treatment of patients with this disease.

Clinical case presentation

The patient was a 58-year-old male who was suffered a chronic cough with white sputum for six months who felt chest tightness and shortness of breath. He irregularly took anti-inflammation medication and asthma therapy in the local hospital. The symptoms were slightly
relieved, although they soon relapsed. Later, the patient came to our hospital outpatient department for further diagnosis and treatment. He was a lifelong nonsmoker and denied other basic diseases. We gave him a preoperative examination to exclude contraindications (Figures 1, 2A-C, 3). The patient was given aerosol inhalation (ipratropium bromide 500 μg/Tid and budesonide 1 mg/Tid) for tracheal preparation and he was trained for respiratory physiotherapy.

After the full preparation, a posterolateral thoracotomy of an axillary thoracotomy was performed under general anesthesia. Intraoperatively, adhesions in his right chest were seen, and the neoplasm located in the carina, and the 2nd, 4th lymphadenectomy, with the 7th lymph nodes was partly calcified. The surgical process was as follows: a fourth intercostal incision was made, and the pleural adhesions were separated first and cut off the lower lung ligament. Furthermore, the azygos vein near the vena cava side was ligated and divided, and the loose connective tissue around the lower trachea and bilateral main bronchus (left and right) was dissected. After the 2nd, 4th, and 7th lymph nodes were excised, the bilateral main bronchus was cut down 6 mm away from the carina, and the carina was removed, with the lower section of the endotracheal membrane and the tracheal posterior wall. The tracheal tube was inserted into the left main bronchus on the operating table (Figure 2D) to provide left lung ventilation. The bronchial stump was sent to fast frozen pathology, which suggested that the specimen was negative. Intermittent suture of the tracheal posterior wall was performed with 3/0 absorbable line, and sutured the bilateral main bronchial margin to reconstruct a new carina. The tracheal membranes were continuous stitched with 3/0 absorbable line. The distance between each stitch was 3-4 mm, and each margin was 4-5 mm. All postoperative diagno-

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**Figure 1.** X-ray shows an increased density mass shadow on the left lower lobe; the size of the mass was nearly 2.2 × 3.0 cm. (A: Anteroposterior film, B: Lateral film).

**Figure 2.** (A-C) Chest CT shows an oval nodule on the tracheal bifurcation; the size of the nodule was 2.5 × 1.5 cm. The mucus plug was in the left main bronchial, which led to obstructive inflammation in the left lung. (A: Horizontal section of pulmonary window; B: Horizontal section of mediastinal window; C: Coronal section of pulmonary window). (D) Demonstrated that the tracheal tube was inserted into the left main bronchus on the operating table to provide left lung ventilation.
ses were verified pathologically, there were no cancer cells remaining in the residual tracheal end, and the 2nd, 4th, and 7th lymph nodes, which were surgical removed, were all negative (0/1, 0/2, 0/4). The immunohistochemistry result was p40 (+), Programmed cell Death-1 (PD-1) (-), Programmed cell Death Ligand-1 (PDL-1) (20% +), Ki-67 (35% +) (Figure 4). Postoperative therapy contained antibiotic prophylaxis, aerosol inhalation and other basic infusion treatments (Figure 5A, 5B). The patient was discharged on the 7th operative day, and he subsequently visited the outpatient clinic two weeks later (Figures 5C, 5D, 6). His recovery and general situation was good, and the physical examination was normal. He has not received additional radiotherapy or chemotherapy treatment, and telephone and outpatient follow-up to date have shown no recurrence.

Discussion

The term “primary carinal tumors” was used as the MeSH search term in the PubMed data base over the last ten years. The disease could occur at any age, although the most common age is 40-69 years old. The incidence is higher in males than in females. The mainly pathological type is epithelium-derived tumors. The malignant tumors mostly are squamous cell carcinomas and adenoid cystic carcinomas, and the benign tumors are mostly papillomas and fibroids. Malignant tumors are more commonly seen in adults, and benign tumors more common seen in children [3]. Tumors usually occur at the junction of the tracheal cartilage and the membrane, and the most common site is the lower part of the trachea [4]. Gas is likely to cause eddy current around this area. Therefore, some bacteria, viruses and other microorganisms or harmful substances could get stuck in this location.

Preoperative examination

Conventional X-ray results are usually normal in the early period, which is a common cause of misdiagnosis. X-ray examinations are commonly relied upon with attention paid mainly to bronchial lung lesions. Due to its good contrast,
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the common X-ray examination has an advantage over gas-bearing bronchial lung tissue to find lesions. However, the large blood vessels, heart, lymphoid tissue, fat and other soft tissues overlap in the tracheal mediastinal structure. This condition is not clearly presented on X-ray, especially for tracheal lesions. In this case, the patient underwent X-ray many times at the local hospital, but the diagnosis was delayed because he did not undergo timely further examinations. Therefore, for elderly patients who suffer from the above symptoms and for whom medical treatment does not work, we should consider tracheal neoplasms and arrange for detailed examinations. Tracheal CT and 3-D imaging examination could further clarify the details of the tumor and its surrounding areas, such as the size of the endotracheal tumor and mediastinal lymph node metastasis [5]. Electronic bronchoscopy could directly observe the site, shape and traits, etc. of tumor and also could obtain a histopathological diagnosis. It is meaningful to fully understand the scope of lesions before surgery and to determine the surgical program. However, it should be noted that fiberoptic bronchoscopy might lead to several serious adverse reactions, such as hemoptysis, and partial tumor shedding might lead to pneumonia, asphyxia, cardiac arrest and other complications. Another drawback is that fiberoptic bronchoscopy could not be used to confirm if the lesions are the result of invasion. If the tracheal tumor was exogenous growing, it would be difficult to obtain positive results. At present, most scholars believe that if a tracheal tumor is seriously obstructed or has abundant blood supply, fiber axillary bronchoscopy should be very careful [6].

Intravenous anesthesia is undertaken with single lumen endotracheal intubation. If the tumor is small and the lumen obstruction is less than half of the diameter, a small tracheal tube (such as a 6F catheter) is used to cross the tumor site and insert into the distal trachea for ventilation. It may be determined that the catheter could not pass through the tumor in this patient. Therefore, in this case the catheter was left proximal to the mass (2 cm above the tumor) for ventilation. Unilateral bronchial cannula through the incision was used for mechanical ventilation during the operation. The bloody liquid was suctioned off in time to prevent tracheal obstruction. The length of the distal end of the balloon should be trimmed according to the specific results of the intraoperative measurement. This process avoided intubating too deep to affect the side of the upper lung lobe opening, which could not guarantee effective single lung ventilation [7]. When the bronchial intubation obstructs the operation during the anastomosis, we could withdraw the bronchial cannula quickly. After we suture two or three needles, then the catheter was quickly inserted to restore ventilation. A compact and accurate

Figure 4. Postoperative pathology revealed that the size of the tracheal tumor was 3 × 2.5 × 1 cm (A: After surgical resection in vitro; B: When the pathology is drawn, and the microscopic images displayed moderate differentiation of squamous cell carcinoma and partly presented a papillary structure (C: H&E stain; D: The immunohistochemistry result of p40 (+). (Original magnification × 100).

Anesthesia and airway management

The correct surgical process and skillful cooperation of the anesthesiologist are critical to the success of this operation.
operation would not lead to the occurrence of hypoxemia.

**Operation technique**

As soon as a trachea and carinal tumor is confirmed, the condition of the patient needs to be evaluated for a surgical operation. Different surgical procedures are based on different location of lesions [1, 8]. A benign tracheal tumor should be removed with a local excision fenestration or end-to-end suturing after a routine tumor resection. Malignant tracheal tumors are mainly treated with tracheal resection followed by an end-to-end anastomosis [9]. There are several main reconstruction methods after carina resection: 1) Suturing the trachea and the right main bronchial residue, and then anastomosis of the right main bronchus and left main bronchial end. 3) Reconstructing a new carina by suturing the bilateral bronchial sides, and then anastomosis with the main bronchus [10]. The third method was chosen in this patient because the angle of the new carina was more similar to the physiological anatomy. This operation is relatively simple and relatively easy to avoid the formation of an angle or a narrow area. The patient had preoperative left lung obstructive pneumonia, but the left lung inflammation subsided postoperatively.

**Postoperative complications**

The risks associated with surgical carinal resection are high, and anastomotic bleeding and fistula are the most serious [11]. Therefore, focus on prevention, anastomotic tension, and anastomotic blood supply are the key factors that affect the healing of anastomosis. First, the resection range has a direct effect on the tension of the anastomosis. The resection range of the upper and lower incision edge should include the removal of at least one normal cartilage ring (the length is approximately 5 mm or more). Studies have shown that the tracheal resection length generally should not exceed 50% of the total length. A resection length less than 4 cm could be directly sutured [12]. Otherwise, care is needed for some measures to facilitate a low-tension anastomosis, including cervical tracheal mobilization, suprahyoid laryngeal release, bilateral hilar release, and division of the inferior pulmonary ligaments [13]. On some occasions, it has been necessary to make a U-shaped incision in the pericardium around the lower pulmonary vein. This process will extend the length of the trachea resection within safe levels. If the preoperative evaluation determines that the lesion is more than 6 cm in length, it is necessary to fully assess the condition. To be able to remove longer tracheal

![Figure 5. X-ray showing a postoperative chest change of left lower lobe with a right rib phrenic angle disappeared, and there were no performance after 48 hours (A: Anteroposterior film; B: Lateral film). Two weeks after discharged, the right pleural effusion decreased and without any significant pneumothorax compared with the latest X-ray (C: Anteroposterior film; D: Lateral film).](image-url)
tumors, an artificial trachea is undergoing further development. Perhaps one day, this artificial trachea could be better applied to serve clinical patients [14]. Surgical specimens are routinely sent for frozen pathology, and a negative stump could undergo one-stage anastomosis and the resection range is close to 5 cm during the intraoperative period, the local residual end would be retained and the patient would undergo follow-up therapy.

Furthermore, because of the anatomical particularity of the tracheal bronchial blood supply, one should not extensively dissect the tracheal surrounding tissues. In our experience, the surface tracheal and bronchial membrane tissues were not stripped excessively. The free edge of the anastomotic lower end is generally not more than 1 cm to guarantee good blood supply. During the anastomosis, we should ensure the tracheal and the main bronchial edge are neat, the connected joint matches well, and the suture margin and needle pitch are moderate. Intermittently sutured tracheal posterior wall with 3/0 absorbable line is done first, and then the tracheal membranes were continuously stitched by 3/0 absorbable line and knotted outside the lumen. The continuous suture has several advantages, such as stitching that is close and reliable, and the inner wall of the tube remains smooth. Adjustment of the edge distance of the anastomosis was done when the bilateral ends of the pipe diameter are inconsistent. After the anastomosis of the outer wall were complete, the outer wall of the tube was relatively smooth compared to the intermittent suture because fewer wire splices could avoid excessive damage to the surrounding tissue. At times, the anastomosis was wrapped with the Mediastinal pleura. This process ensures the anastomotic blood supply and effectively prevents the occurrence of anastomotic fistula, especially for unsatisfac-

Figure 6. Compared with the preoperative CT, the oval nodules at the tracheal bifurcation disappeared, and the left main bronchus was fluent, whereas exudative lesions increased on both sides. A few new pleural effusions could be observed on the right side and considered the possibility of right lower pulmonary atelectasis 5 days after surgery (A: Horizontal section of mediastinal window; B: Horizontal section of pulmonary window; C, D: Three-dimensional airway reconstruction revealed that the lower right lobe of the bronchus were not developing, while the other trachea and bronchus were normal. E: Coronal section of mediastinal window; F: Coronal section of pulmonary window, without any obvious luminal stenosis or signs of compression).
tory anastomosis, severe inflammatory response or poor blood circulation. Other impact factors of anastomosis include stitch length, margin, and the match of anastomosis. In addition, there are reports of several other complications such as pneumonia atelectasis, hoarseness caused by the recurrent laryngeal nerve injury, tracheal diaphragmatic pleural fistula, pleural cavity infection and tracheal anastomotic stenosis [10, 12, 15-17].

Postoperative management

Postoperative care is important for patients undergoing carinal resection. The deformation of the carina would affect the distribution of gas dispersion and effective expectoration. The angle of the new carina is changed, and the postoperative anastomosis caused by edema from suturing. Therefore, the bronchial and pulmonary secretion discharge resistance is increased. The patient usually is unable to cooperate to cough effectively because of fear of pain. Unsatisfactory expectoration is common, which could cause atelectasis and even pneumonia. Expectoration is even more difficult when the anastomosis complicated with stenosis or fistula. The patient was provided with ventilator-assisted breathing until he restored spontaneous breathing after anesthesia recovery. This process would prevent atelectasis and play a positive role in lung function recovery. There is a benefit for respiratory secretions discharge that routinely uses aero-sol expectoration and increased use of bronchial drugs the postoperative period. However, this patient appeared to have right lower pulmonary atelectasis and right pleural effusions due to his cough and sputum fatigue. It is important that provide the patient with sufficient nutrition and maintain the postoperative internal environment balance. It is widely known that the trachea moves up and down with the neck flexion. It is better to reduce the anastomotic tension by keeping the neck in flexion.

Prognosis

In the past, there was a high risk of surgery and a high rate of postoperative complications; therefore, in the past, conservative treatment was employed. With the development of surgical and assistive techniques, the surgical indications of those cases were enlarged and the results proved that surgery is an effective treatment. Spaggiari reported a series of 37 carina resections; the overall 5-year survival rate was 27%, with a 5-year survival rate of 32% in down-staged patients and 12.5% in not down-staged patients. Very good survival results could be obtained, especially with highly selected patients (pN0 or pN1) [2]. Liu summarized that the patients with primary tracheal and carinal tumors experienced a 5-year survival of 55.0% compared to 16.7% for those with locally advanced lung cancer directly infiltrating the carina (P<0.05) where the patients who underwent surgical resection had a significantly better 5-year survival than the patients with inoperable disease (P<0.01) [4]. Gonfiotti presented that the 5-year overall survival after carinal resection ranged from 26% to 44% in various series. In their experience, survival is significantly affected by endobronchial extension of the primary tumor and by nodal status (pN0 patients have a 5-years survival rate of 50%). Clearly, the most important factor affecting survival is adequacy of the tumor resection [11].

Patients with wide range lesions, which are difficult to surgically resect, should consider radiation therapy. The situation is the same as those with residual postoperative tumor, positive margins, or for patients with contraindications to surgery. The majority of malignant tracheal malignant, especially adenoid cystic carcinomas and squamous cell carcinomas are more sensitive to radiotherapy, although the effect of radiotherapy on airway adenocarcinoma is unknown. Carvalho reported the application of high-dose and short-range radiotherapy in the airway [18]. These researchers used a bronchoscope to implant the radioactive material in the tumor, which proved to inhibit local tumor growth. There were two cases of squamous cell carcinoma, one case of recurrent adenoid cystic carcinoma and one case of recurrent plasmacytoma in that article. The results showed that of all patients had local tumor control at the time of the first assessment. Only one case of squamous cell carcinoma experienced a recurrence in two years and one case of adenoid cystic carcinoma experienced airway stenosis after two years. However, this therapy has some disadvantages, such as high cost, long operation time, pain, and need to be carried out several times. The conditions of patients who are unable to tolerate this therapy are generally poor. Therefore, the indications
for this therapy should be strictly controlled [19]. In this situation, we should not achieve to cure the patient’s disease as a goal, although we should commit to reduce the patient’s airway obstruction, improve his ventilation and the quality of life. In our case, the patient proved negative margins and no lymph node metastasis, so he just regularly followed-up in the outpatient department or by telephone.

Conclusions

With the extensive use of CT and fiberoptic bronchoscopy in recent years, clinicians have improved the understanding of primary tracheal cancer. The treatment of primary tracheal tumors should follow the principles of early detection, early diagnosis, and early treatment. Surgical resection is the first treatment choice for primary tracheal cancer. Safe and stable anesthesia is essential for patients. Clinicians aimed to eradicate the symptoms of airway obstruction, while the safety of anastomosis and the eradicative resection should also be taken into account. The preoperative and postoperative aerosol inhalation and physiotherapy sputum or other measures would assist with the maintenance and recovery of airway functions.

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Disclosure of conflict of interest

None.

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