

A rare large tracheal glomus tumor with postoperative haematemesis

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ABSTRACT

Glomus tumors are uncommon benign tumors which usually arise from the distal portion of the digits. A tracheal glomus tumor with large size is extremely rare. We present a case of a large tracheal glomus tumor that was resected using posterolateral thoracotomy and successful primary reconstruction of the trachea was achieved. Severe haematemesis happened after the patient was discharged. An emergency exploratory thoracotomy was performed but no signs of anastomotic bleeding were observed, while intraoperative gastroscopy revealed plenty of blood and blood clots in the patient's stomach. Medical treatments targeting hemorrhage of upper digestive tract were given and the patient finally recovered.

KEYWORDS

Glomus tumor; trachea; haematemesis

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Glomus tumors are rare, benign but often painful tumors arising from modified smooth muscle cells (SMCs) of the glomus body, generally much less than 1 cm in diameter. These tumors are usually found in the distal portion of the digits, especially under the fingernails. They are morphologically characterized by round, slightly elevated, red-blue, firm nodules (1). However, these tumors can also occur occasionally at sites such as the chest wall, bone, gastrointestinal tract, nerve, eyelid, mediastinum, and trachea, where normal glomus bodies may be few or even absent (2). We reported a rare tracheal glomus tumor which was successfully resected.

Case report

A 15-year-old boy was admitted to our hospital for a 3-month history of cough, hemoptysis and intermittent shortness of breath, without any past medical and smoking history. Physical examination was negative except for dry rales of trachea in auscultation. Radiological examinations revealed a mass at the posterior wall of thoracic trachea, with its size of approximately 2.0 cm × 2.3 cm (Figure 1).

The operation was performed via right posterolateral thoracotomy through the fourth intercostal space. During the operation, a mass with a base about 2.0 cm in diameter was found to be located at the posterior tracheal wall and no pleural adhesion or effusion was found. En bloc resection of the tumor was achieved with 0.5 cm spare of both surgical margins. The tumor was taken out and measured. Maximal and minimal diameter was 2.5 and 2.0 cm respectively. Frozen-section pathological examination confirmed a benign tumor with negative resection margins. Then, tracheal anastomosis was performed by continuous suture using 3-0 prolene stitches. Postoperative bronchoscopy showed that the trachea was successfully anastomosed with no stricture or bleeding (Figure 2).

The tumor was pathologically diagnosed as glomus tumor. The patient's symptoms were relieved and was discharged 11 days after surgery without any main complication.

Three hours later, the boy was sent to the emergency room (ER) with severe hemoptysis, or probably, haematemesis. He was attacked by cardiac arrest and was fortunately resuscitated by CPR. Lots of fresh blood was sucked out via endotracheal suction. Anastomotic bleeding came up as the first diagnosis. An emergency exploratory thoracotomy through the original incision was performed immediately. There was about 400 mL pleural effusion in the right chest, but no active bleeding was found, intraoperative bronchoscopy did not find any bleeding site in the anastomotic stoma either. Then upper gastrointestinal hemorrhage came in to consideration, and intraoperative gastroscopy was assigned. It found plenty of blood and blood clots in the stomach but further exploration was restrained

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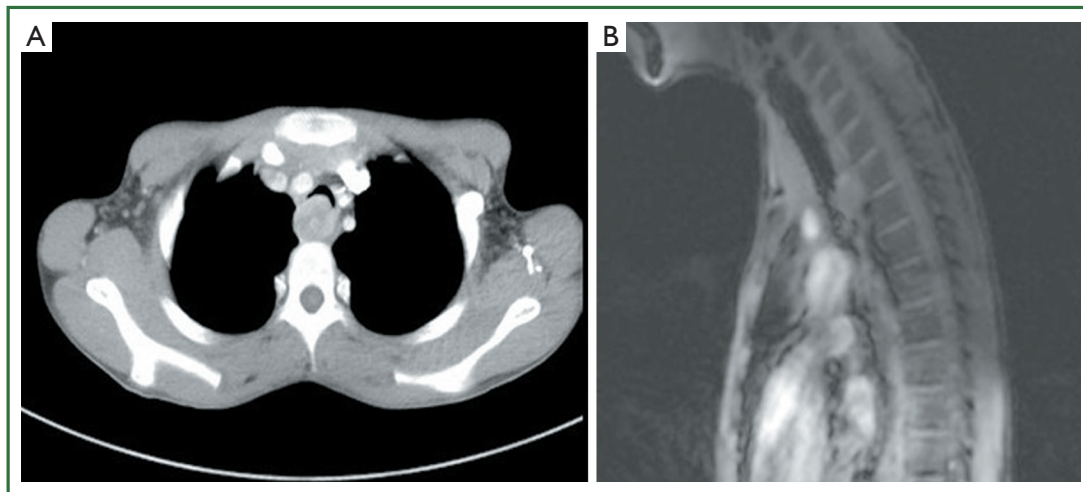


Figure 1. (A) Enhanced CT showed a heterogeneously enhanced mass arising from the posterior wall of thoracic trachea and an undistinguished margin to the esophagus; (B) Enhanced MRI showed the tumor was iso-signal intensity on T1WI and high signal on T2WI, the tumor locates at the lower one-third of thoracic trachea and approximately 2.7 cm to the carina (Right).

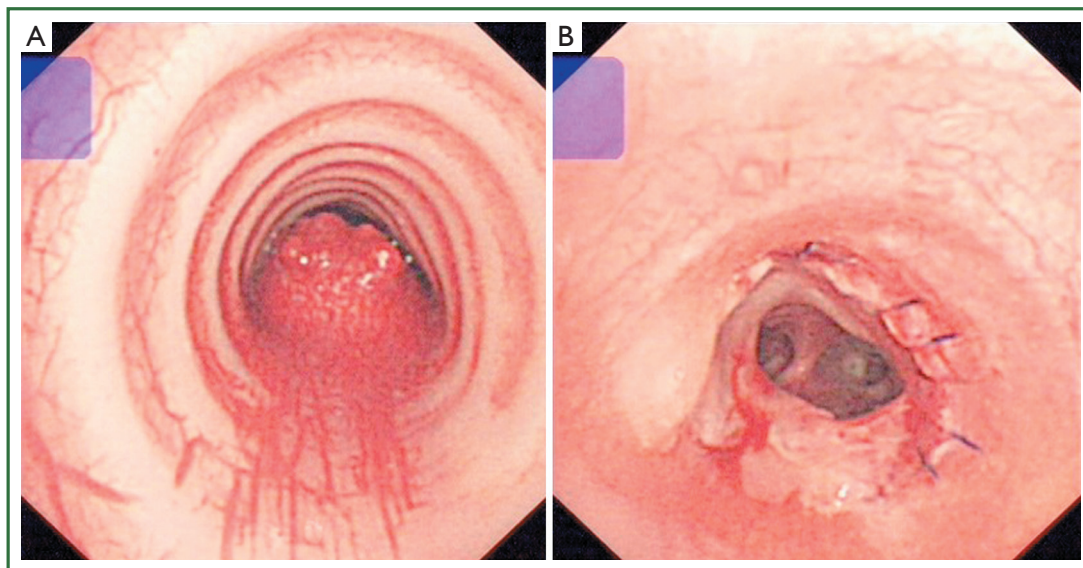


Figure 2. (A) Preoperative bronchoscopy showed a red tumor blocking the trachea; (B) Postoperative bronchoscopy showed that the tumor was removed integrally and the trachea was successfully anastomosed.

because of poor imaging field. Thus, the patient was treated by hypovolemic therapy, hemostasis therapy with somatostatin and Carbazochrome Sodium Sulfonate, gastrointestinal decompression, anti-infection therapy and nutrition support. Postoperative enhanced abdominal CT scan with vascular reconstruction showed nothing abnormal. Another gastroscopy 2 weeks later found nothing special either. The patient finally recovered and was discharged 3 weeks after surgery. The symptoms disappeared, and no sign of postoperative stenosis was found at the patient's 1-year follow up, neither was recurrence or hemorrhage.

Discussion

Glomus tumors are biologically benign but often painful tumors arising from modified SMCs of the glomus body, histologically, these tumors are aggregates, nests, and masses of specialized glomus cells associated with branching vascular channels, all surrounded by connective tissue matrix.

In the sections of the tumor, plenty of small round or oval cells, vessels and smooth muscle were seen by microscope. Immunohistochemistry stain supported the diagnosis of glomus tumor (Figure 3).

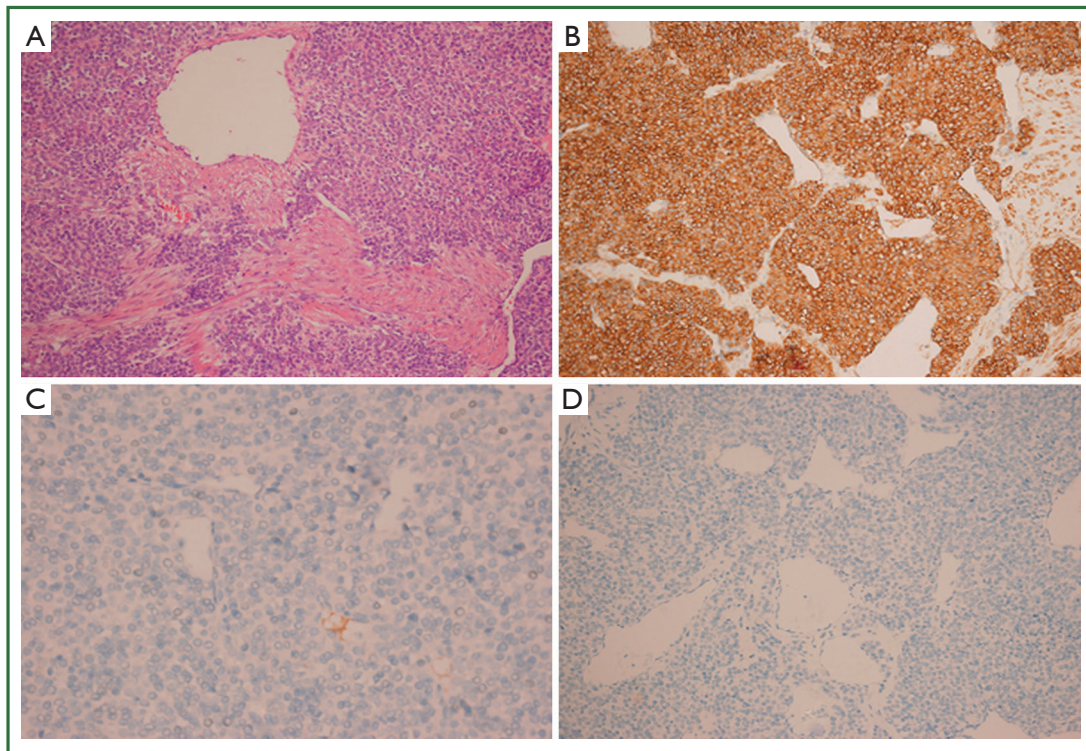


Figure 3. (A) The tumor consists of large amount of vessels and smooth muscle tissue (H&E 200×); (B) SMA(+) reveals the smooth muscle portion of the tumor (SMA stain, 200×); (C) CK(−) suggests the tumor is not epithelium-derived (CK stain, 200×); (D) CD56(−) suggests that there is no neural/neuroendocrine tissue (CD56 stain, 200×).

Surgical removal of the tumor with primary reconstruction of trachea was usually the first choice for treatment (3). Successful removal via flexible bronchoscope has been reported (4), but in their case, the tumor was 2.0 cm × 1.0 cm in size and there seemed to be no tight adhesion with tissue surrounding though it locates at the lower tracheal segment. Cervical collar incision has also been reported recently (5), and in their case, the tumor was 1.3 cm × 1.2 cm in size, located at the upper portion of the trachea. On the other hand, posterolateral thoracotomy, instead of bronchoscope, was performed on our patient because the tumor located at the lower portion of trachea, had a relatively large size, the base of the tumor was wide and risk of severe bleeding was high.

To our knowledge, this is the first report of postoperative severe hemorrhage of upper digestive tract after removal of tracheal glomus tumor. However, the reason that caused the severe hemorrhage after the first surgery remains unclear.

Glomus tumor of the stomach has been reported (6), as well as multiple lesions (7). With regard to our case, nothing abnormal was found through radiological and endoscopic examinations. Therefore, we made two hypotheses as following: first, the patient simultaneously suffered from another mini glomus tumor, which caused severe hemorrhage but could hardly be detected by CT scan or gastroscopy; second, the patient suffered from severe stress ulcer due to the surgery and

the ulcer had been cured when postoperative gastroscopy was performed. When we reviewed the treatment strategies, the second thoracotomy seemed to be unnecessary since it only played a role in excluding problems of anastomotic stoma, which could be manifested by bronchoscopy, a simpler and less invasive test. If we chose to apply the bronchoscopy to check whether the anastomotic bleeding existed at the beginning, the second thoracotomy could be avoided.

Conclusions

Glomus tumor is rare benign tumor derived from soft tissue with good long-term prognosis (8). While coping with patients with tracheal glomus tumor, potential multiple lesions should be intentionally evaluated. Tracheal resection via thoracotomy should be taken in order to remove the tumor of lower portion of the trachea or with a relatively large size (maximum diameter >2.0 cm) safely and integrally. In addition, the misdiagnosis of the case reminds us to confirm the anastomotic bleeding carefully as long as the situation is not extremely urgent so that unnecessary invasive procedures can be avoided.

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