Case report

Racemose hemangioma revealed by massive intraoperative hemorrhage: A case report

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ABSTRACT

Introduction and importance: Bronchial arterial racemose hemangioma is considered to be a type of pulmonary arteriovenous malformation (PAVM). We encountered an extremely rare case of racemose hemangioma, which was identified because of massive bleeding during lung cancer surgery.

Case presentation: A 71-year-old man was suspected of having lung cancer and underwent a right upper lobectomy. During the dissection of the peribronchial connective tissue around the upper lobe bronchus, a sudden massive hemorrhage occurred after separation of the pulmonary artery and vein. Hemostasis was difficult to achieve with compression hemostasis and the point of bleeding was unknown; thus, we were called in to search for the source of the bleeding, which was identified as the bronchial artery. Postoperatively, a racemose hemangioma was diagnosed via preoperative chest computed tomography and imaging analysis with the Ziosoft® (Ziosoft Inc. Tokyo, Japan).

Clinical discussion: Racemose hemangioma has weaker blood vessel walls than normal blood vessels. Once bleeding occurs, it is very difficult to stop. However, once the bleeding point was identified, hemostasis via suturing and cutting was possible.

Conclusion: As hemorrhaging from a racemose hemangioma is very difficult to stop, preoperative imaging retrieval is very important.

1. Introduction

Bronchial arterial racemose hemangioma is considered to be a type of pulmonary arteriovenous malformation (PAVM). Approximately 4% of PAVMs communicate with the systemic circulation, such as the bronchial arteries [1]. The prevalence of PAVM in Japan has been reported to be 0.038% [2]; therefore, a shunt between the bronchial artery and pulmonary arteries, as described in this case, is extremely rare. In our department, 1073 patients underwent surgery between January 2016 and August 2020; however, this was the only case with a bronchial arterial racemose hemangioma (0.09%).

The work has been reported in line with the SCARE 2020 criteria [3].

2. Presentation of a case

A 71-year-old man exhibited a nodule shadow in the right upper lobe of the lung on a preoperative chest computed tomography (CT) scan for sigmoid colon cancer. A chest CT was performed again postoperatively for sigmoid colon cancer, and the nodule shadow at the same site increased to 11 mm (Fig. 1a). The patient was therefore referred to our department for detailed medical examinations and was identified as a current smoker.

The thoracoscopic surgery was performed with three ports, with the patient in the left lateral decubitus position. Partial resection of the right upper lobe of the lung on a preoperative chest computed tomography (CT) scan for sigmoid colon cancer. A chest CT was performed again postoperatively for sigmoid colon cancer, and the nodule shadow at the same site increased to 11 mm (Fig. 1a). The patient was therefore referred to our department for detailed medical examinations and was identified as a current smoker.
Fig. 1. (a): A chest CT scan revealed a nodule shadow in the right upper lobe of the lung. (b), (c): A preoperative chest contrast CT scan revealed a loop-shaped blood vessel around the upper lobe bronchus. The red markings indicate racemose hemangioma. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)
sudden hemorrhage occurred. Hemostasis was performed for 5 min using the upper lobe lung, and the main pulmonary artery was clamped as the bleeding point was unknown; therefore, this was converted to a thoracotomy procedure. Finally, we detected bleeding near the bronchial wall, away from the pulmonary artery. It was sutured with 4-0 prolene; the upper lobe bronchus was subsequently divided, and the right upper lobe was isolated. Although there was no hemorrhaging, the additional suture was created more centrally on the bleeding bronchial artery. The operating time was 257 min, and the total volume of blood loss was 1380 mL. The pathological diagnosis was a large cell neuroendocrine carcinoma, pT2aN0M0, pStageIIB.

After surgery, reexamination of the preoperative chest contrast CT scan revealed a loop-shaped blood vessel on the dorsal side around the upper lobe bronchus (Fig. 1b and c); image analysis using the Ziostation2® (Ziosoft Inc. Tokyo, Japan) confirmed shunting blood vessels between the bronchial and pulmonary arteries (Fig. 2), and the diagnosis was confirmed as racemose hemangioma. When the same imaging procedure was performed again on postoperative day 7, some racemose hematoma remained (Fig. 3); however, the patient’s course was favorable, and he was discharged 12 days postoperatively. One month postoperatively, adjuvant chemotherapy was performed for sigmoid colon cancer. Moreover, no rebleeding and no recurrence of lung cancer was observed.

3. Discussion

We had previously reported that preoperatively performing a three-dimensional (3D)-CT angiography is important for understanding vascular malformations and branching anomalies [4–6]; however, we were unable to identify this rare vascular abnormality preoperatively. Even if it could be preoperatively diagnosed, careful operation is required, because bronchial arterial racemose hemangioma has weaker blood vessel walls than normal blood vessels [7]. There are reports that it occurs most often in the right middle or lower lobe bronchus, but this is not definitive [8]. Some studies report that preparing for percutaneous cardiopulmonary support when similar abnormal blood vessels are observed is necessary [9]. Although the preoperative CT scan indicated abnormal blood vessels, racemose hemangioma was not recognized; therefore, during the operation, the hemangiomas were damaged, resulting in a 1380 mL loss of blood. Bleeding from the bronchial arteries is sometimes difficult to stop, as the bronchial arteries themselves run deep into the tissue, and the bleeding points are unknown. It could also cause intraoperative death; in fact, we initially believed the hemorrhage to originate from the pulmonary artery, and subsequently clamped the main pulmonary artery. Hemorrhage from a bronchial arterial racemose hemangioma is easily mistaken for damage to the pulmonary artery; however, once the bleeding point was identified, hemostasis via suturing and cutting was possible. After the operation, although a part of the bronchial arterial racemose hemangioma remained, no rebleeding was observed. Therefore, the risk of postoperative rebleeding is considered to be considerably low if the vascular treatment is correctly performed.

In our department, image analysis is currently performed using a medical workstation, called the Ziostation 2®, in cases of suspected abnormal blood vessels. When using a medical workstation in these cases, automatic extraction of the aorta is installed as a standard function, and a separated 3D image of the pulmonary arteries and veins is created by threshold value adjustment using the difference in contrast and automatic recognition; the extraction function is performed semi-automatically. Considering this case to be an educational opportunity, we decided to share the image with all medical staff at the preoperative conference.

4. Conclusion

We encountered a case of racemose hemangioma, which turned out to cause massive intraoperative bleeding. It is very important to share preoperative image retrieval with multiple people. Additionally, preoperative image retrieval showed a racemose hemangioma; thus, careful preparation is necessary when surgery is performed. In particular, more caution is required when loop-shaped vascular findings are found on contrast-enhanced CT.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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