CASE REPORT

A case of pulmonary granuloma associated with mycobacterium avium complex infection arising from the surgical margin after segmentectomy

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Abstract The patient was a 50-year-old man with a history of right S9 + 10 segmentectomy for lung carcinoid at another hospital 4 years and 9 months ago. Two years and nine months after the surgery, a nodule was noted at the surgical margin. Furthermore, 9 months later, the nodule was enlarged and detected a strong accumulation on positron emission tomography–computed tomography. Therefore, the recurrence could not be denied and surgery was performed. The intraoperative pathological diagnosis was inflammatory granuloma, which was then removed by wedge resection. A culture of the tumor content showed mycobacterium avium complex (MAC). The final pathological diagnosis was a pulmonary inflammatory granuloma associated with MAC infection. Although a number of studies showed the risk of foreign body granuloma or an inflammatory pseudotumor after pulmonary resection with surgical staples, in this case the prior segmentectomy was carried out with electrical cautery alone. A nodule detected at the surgical margin after segmentectomy should be suspected to be an inflammatory change, in particular granuloma formation, as well as tumor recurrence and second primary lung cancer, even if the segmentectomy was done without staplers.

Keywords Inflammatory granuloma · Surgical margin · Mycobacteriosis avium complex infection · Post-segmentectomy

Introduction

Segmentectomy has been frequently performed for small-sized lung cancer in recent years [1]. However, it was reported that sublobar resection for primary lung cancer resulted in higher incidence of local recurrence than that of lobectomy. When a nodule is detected and is consequently enlarged near the surgical margin by follow-up radiological examination after surgery, it is often difficult to determine whether to be a recurrence of the malignancy or the pathogenesis of a second primary lung cancer or an inflammatory change, even with positron emission tomography–computed tomography (PET–CT), which is useful for distinguishing between benign and malignant nodules [2, 3]. A number of studies have reported cases in which the nodule at the surgical margin diagnosed as a foreign body granuloma formation or an inflammatory pseudotumor after pulmonary parenchyma resection using surgical stapler [4–6]. Here, we report a case of pulmonary inflammatory granuloma associated with MAC infection after lung segmentectomy using an electric cautery without surgical staplers.

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Case report

The patient was a 50-year-old man underwent right S9 + 10 segmentectomy for primary lung cancer at the other hospital 4 years and 9 months ago. Preoperative computed tomography (CT) imaging had revealed a tumor

(1.5 × 1.4 cm in size) spanning S9b and S10b of the right lung (Fig. 1). According to the operation record obtained from the previous hospital, lung segmentectomy was performed by resecting the lung parenchyma using an electric cautery without using surgical stapler, and then absorbent polyglycol acid (PGA) sheet and fibrin glue spray were applied on the resection surface to manage air leakage from injured pulmonary parenchyma. It was diagnosed as typical carcinoid (pT1aN0M0, p-stage IA) based on histopathological findings, and the sufficient surgical margin was histologically confirmed. Based on the operative record of the previous hospital, the lung parenchyma resection was performed without the use of a stapler. Veins (V6c and V8b) that run between S9 + S10 and S6–8 down to the periphery were exposed. The lung was cut along the line between the inflated S9 + 10 and the deflated S6–8 by an electric scalpel only. Veins were preserved on the resection surface of the remaining inferior lobe. Postoperative course was uneventful. He has been followed up on outpatient basis without adjunctive chemotherapy. Two years and nine months after the surgery, CT revealed a localized infiltrative shadow (12 × 10 mm in size) at the resection margin (Fig. 2a). He was subsequently referred to our hospital. Nine months after the first detection of the shadow, the shadow opacity became denser and expanded to a size of 18 × 14 mm on CT (Fig. 2b). A strong accumulation (maximum standard uptake value: 8.75) of F-18 fludeoxyglucose (FDG) was confirmed on PET–CT (Fig. 2c).

The patient history implied inflammatory changes or a recurrence of the malignancy. According to CT image, inflammatory changes or the pathogenesis of a second primary lung cancer, especially lung adenocarcinoma was suspected, and PET–CT could not rule out malignancy. The final preoperative diagnosis primarily showed

Fig. 1 Chest CT showed a well-demarcated tumor (1.5 × 1.4 cm in size) spanning S9b and S10b of the right lung. V8b was indicated by arrow

inflammatory changes in particular granuloma formation at the surgical margin. The differential diagnosis suggested the pathogenesis of a second primary lung cancer or a recurrence of the malignancy. Surgical intervention was performed. An intraoperative needle biopsy led to a diagnosis of inflammatory granuloma, so wedge resection was performed for the lesion. The resected specimen included pus exudation, and histological examination did not identify a malignancy. There was marked inflammatory cell infiltration along with an epithelioid cell granuloma arising from the surgical margin as located in S6 (Fig. 3). A culture of the tumor content was identified as mycobacterium avium complex (MAC). The postoperative definitive diagnosis was pulmonary inflammatory granuloma associated with MAC infection.

Discussion

Segmentectomy has frequently been performed for small-sized lung cancer in recent years [1]. To secure sufficient surgical margin, it is necessary to resect lung parenchyma with careful detection of the tumor. In addition, veins flowing on the intersegmental surface and pneumatization/collapsing lines should be applied for orientation of intersegmental plane [1–3]. In this case, electric cautery was used for the separation of intersegments to conserve lung function. If a nodule is detected and is consequently enlarged close to the surgical margin after segmentectomy or wedge resection, physicians should first consider a possibility of local recurrence of the primary cancer at the surgical margin. The pathogenesis of a second primary lung cancer or inflammatory changes, in particular granuloma formation, were considered as differential diagnoses. While PET–CT is useful for differential diagnoses of benign and malignant lesions [2, 3], diagnostic imaging provides limited accuracy, so surgical resection is appropriated for a definitive diagnosis.

Stapler-aided lung resection is often used as we can simplify surgical procedure. On the other hand, some studies have reported the cases of a foreign body granuloma formation and an inflammatory pseudotumor, which was firstly detected as a mass-like opacity at the stapled resection margin that was detected during postoperative follow-up period [4–6]. A possible pathogenesis mechanism is that a foreign body reaction against the staples and a non-anatomical dissection lead to poor ventilation and perfusion of the lung tissue, resulting in poor drainage, which in turn contributes to infection. According to the operative record of the previous hospital, the lung parenchymal resection was performed without surgical staplers. In this case, the granuloma developed years after segmentectomy and histological findings demonstrated no

Fig. 2 **a** Chest CT at the first time of our hospital admission showed a small nodule (size, 1.2×1.0 cm) at cut end (S8). **b** Nine months later, the nodule increased in size during follow-up. **c** F-18 fludeoxyglucose (FDG) uptake was seen in the nodule and SUV max was 8.75 on PET-CT

Fig. 3 Pathological findings. The segmental plane ‘postoperative nascent pleura instead of normal pleura’ was indicated by arrow (hematoxylin and eosin staining; low magnification view). There was marked inflammatory cell infiltration along with an epithelioid cell granuloma arising from segmental plane (open square, hematoxylin and eosin staining; high magnification view)

evidence of foreign body reactions, even though absorbent suture reinforcement material and fibrin glue were used. Instead, it might be attributed to a local disturbance in ventilation and perfusion, and resultant poor drainage, which resulted in the MAC infection, although an appropriate anatomical segmentectomy was performed [7, 8].

When a nodule is detected and is consequently enlarged close to the surgical margin after lung cancer surgery, an

examination primarily should be performed based on the possibility of local recurrence. PET-CT, which can efficiently differentiate between benign and malignant lesions, might not distinguish small-sized inflammatory/granulomatous lesions from malignant lesions. The fact that FDG is taken up by macrophages is considered to be mainly involved in FDG accumulation in inflammatory granuloma [2, 9]. Granulomas composed mostly of macrophage-derived epithelioid cells and partly of such as multinucleated giant cells. The level of macrophage activity is reportedly associated with pathological differences in the granuloma. Highly active granuloma has abundant epithelioid cells and multinucleated giant cells [9].

As more cases of pre-invasive lesions of lung adenocarcinoma will be detected with the widespread use of chest CT scanning, and further, the number of patients undergoing segmental resection will also increase along with an increase in reports that segmentectomy is beneficial [10], we anticipate that sublobar (partial) resection will be indicated for more patients in the future. Finally, we anticipate that an increasing number of patients will have a pulmonary parenchymal surgical margin in the residual lobe. Therefore, in cases detected surgical marginal lesions that show a growing nodule with a strong accumulation of FDG on PET-CT, it would be reasonable to perform a surgical resection that could be used as a diagnostic therapy tool.

When an expanding nodular at the surgical margin is observed during the postoperative management of

segmentectomy, recurrence of the malignancy at the margin should be considered primarily. However, diagnostic imaging offers limited accuracy, and therefore, surgical resection should be performed for both a definitive diagnosis and therapy. Inflammatory changes, in particular granuloma formation, should be considered in the differential diagnosis.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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