Swyer-James syndrome is characterized by a unilateral hyperlucent lung and sometimes causes pneumothorax. We present here a case of pneumothorax in a patient with Swyer-James syndrome who required three separate operations.

Case Report

A 21-year-old man with a history of bronchial asthma during childhood presented with left recurrent pneumothorax. Chest x-ray showed collapse of the left lung. Computed tomography showed hyperlucency and some bullae in the left upper lobe. Thoracoscopic bullectomy and pleurodesis were performed. Pneumothorax recurred twice, for which thoracoscopic bullectomy and pleurodesis were performed. During the third operation, pleurodesis of almost the entire left upper lobe was performed. Since this third operation, the patient has been free from recurrences of pneumothorax for three years. The lung perfusion scintigram performed during the absence of pneumothorax showed a lack of accumulation in the left upper lobe. The lung ventilation scintigram performed during the absence of pneumothorax showed low accumulation and delay of washout in the left upper lobe. These findings are compatible with Swyer-James syndrome. To prevent the recurrence of pneumothorax related to Swyer-James syndrome, some kind of procedure, such as extensive pleurodesis, is necessary.

Keywords: pneumothorax, thoracoscopy, reoperation

Introduction

Swyer-James syndrome is characterized by a unilateral hyperlucent lung and sometimes causes pneumothorax. We present here a case of pneumothorax in a patient with Swyer-James syndrome who required three separate operations.

A 21-year-old man with a history of bronchial asthma during childhood presented with left recurrent pneumothorax. He has no history of smoking or other diseases.

Chest x-ray showed collapse of the left lung (Fig.1a). Computed tomography showed hyperlucency and some bullae in the left upper lobe (Fig.1b).

Thoracoscopic surgery was performed for a persistent air leak. Some bullae were found in the left upper lobe; one of which was determined to be the cause of the air leak. The surface of the left upper lobe appeared normal. Bullectomy and ligation of bulla were done for the bullae in the left upper lobe. To reinforce the suture line, pleurodesis with a polyglycolic acid sheet (Neovil®, Gunze, Tokyo, Japan) was performed (Fig. 2).

Left pneumothorax recurred 10 months after the first operation. The cause of pneumothorax was a newly generated bulla in the left upper lobe. Bullectomy, ligation of bullae, and reinforcement with a polyglycolic acid sheet were performed again.

Five years and 8 months after the second operation, left pneumothorax occurred again. The cause was again a newly generated bulla in the left upper lobe. Bullectomy and ligation of bulla were done for the bullae on left upper lobe. To prevent recurrence of pneumothorax, we applied a polyglycolic acid sheet to almost the entire

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surface of the left upper lobe. Since this third operation, the patient has been free from recurrences of pneumothorax for three years.

Lung ventilation and perfusion scintigraphy were performed during the absence of pneumothorax (Fig. 3A). The lung perfusion scintigram showed a lack of accumulation in the left upper lobe. The lung ventilation scintigram showed low accumulation and delay of washout in the left upper lobe (Fig. 3B). Pathological findings of resected lung parenchyma showed emphysematous alveoli with thin walls (Fig. 4). These findings are compatible with Swyer-James syndrome.

Discussion

Swyer-James syndrome, which was first reported by Swyer and James in 1953, is characterized by a unilateral hyperlucent lung. Macleod described the clinical features of this disease in 1954, which is why Swyer-James syndrome is also called Macleod’s syndrome.

This syndrome is defined as unilateral pulmonary hyperlucency on chest X-ray and emphysematous changes in the diseased lung. This syndrome is now considered a postinfectious manifestation of childhood bronchiolitis obliterans. Clinical features usually include repeated respiratory infections.

Several cases of spontaneous pneumothorax in patients with Swyer-James syndrome have been reported. In every surgical case, air leakage was found from the bullae in the diseased lung. It is thought that bullae are likely to develop in the diseased lung.

Inoue, et al. reported a case of pneumothorax related to Swyer-James syndrome treated with thoracoscopic bullectomy that recurred 10 months after the initial surgery. In that case, newly generated bulla was found near the previous suture line. In our case, newly generated bullae were found in the same lobe as during the previous operation. It is thought that postoperative recurrence often occurs in patients with pneumothorax related to Swyer-James syndrome due to emphysematous changes...
of the diseased lung. During the third operation, we applied polyglycolic acid sheets to almost the entire surface of the left upper lobe, and the patient has been free from recurrences of pneumothorax for three years. The application of polyglycolic acid sheets to all diseased portions of the lung seemed to be useful. To prevent the recurrence of pneumothorax, some kind of procedure, such as extensive pleurodesis, is necessary.

**Conclusion**

Swyer-James syndrome is one of cause of spontaneous pneumothorax and pneumothorax due to Swyer-James syndrome often recurs. To prevent the recurrence of pneumothorax related to Swyer-James syndrome, some kind of procedure, such as extensive pleurodesis, is necessary.

**Disclosure Statement**

There is no conflict of interest.

**References**