Rare disease

Swyer–James–McLeod's syndrome and pneumothorax on same side: delay in chest drain removal despite full expansion

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Summary

Swyer–James–McLeod's syndrome (SJMS) occurs as a result of childhood recurrent respiratory infections. As this condition presents with few symptoms, if any, it may pass unnoticed until adulthood. The authors are presenting a case of a 17-year-old male admitted with spontaneous pneumothorax. Even though his lung expanded, his chest x-rays still mimicked a pneumothorax. Chest CT pulmonary angiogram was needed to confirm the diagnosis of unilateral SJMS. Diagnostic difficulty arose because both conditions were on the same side.

BACKGROUND

Swyer–James–McLeod's syndrome (SJMS) is a rare condition characterised by unilateral hyperlucency of a part of, or the entire, lung, which was first described in 1953 by Swyer and James and further detailed by McLeod. It is considered to be an acquired disease secondary to viral bronchiolitis and pneumonitis in childhood. The striking feature of our case is that pneumothorax and SJMS were on the same side, which needed CT chest to ascertain the resolution of the pneumothorax, diagnosis of SJMS was made and chest drain was subsequently removed. He did not have any history of childhood infections or hospitalisations.

CASE PRESENTATION

A 17-year-old male presented with an acute left sided chest pain and shortness of breath. He had a three-pack-year cigarette smoking history as well as significant history of passive cigarette smoking since birth. He also inhaled cannabis for 2 years. He had no history of childhood infections or hospitalisations. Chest x-ray (CXR) showed a large area of left-sided hyperlucency (figure 1), and a diagnosis of a large left-sided pneumothorax was made. Chest drain was inserted, but a CXR after 24 h still showed hyperlucency on the left hemithorax even though the chest drain was still in situ, neither swinging nor bubbling and kinked.

Figure 1 CXR with chest drain. **Figure 2** CXRt without chest drain.

Figure 3 CTPA with hyperlucency.

Low-grade suction was applied, but the CXR appearance did not change. Respiratory examination revealed reduced chest expansion and breath sounds on the left side. We did post chest drain removal CXR which still did not show full lung expansion (figure 2),^{1–6} and it was very difficult to decide that pneumothorax has resolved or not so we arranged chest CT pulmonary angiogram (figure 3). It demonstrated a hyperlucent left lung which was fully expanded and diminished vascularity in the left lower lobe with hyperinflation of the pulmonary parenchyma and no endobronchial lesions or bronchiectasis identified hence a diagnosis of SJMS was made.

Clinical as well as radiological findings suggested the diagnosis of SJMS. Subsequently his pulmonary function tests were normal.

OUTCOME AND FOLLOW-UP

Surgical treatment was offered to patient, but he refused and was followed up in respiratory clinic regularly.

Learning points

- ▶ SJMS can present without history of childhood recurrent chest infections.
- ▶ SJMS and pneumothorax can persist together.
- ▶ The most important learning point is the sensible further investigation of cases when the unexpected happens or appears to have happened.
- ▶ Easy pleural disease is easy. When things do not go according to plan, careful imaging is mandatory like CT chest.
- ▶ It can be so tempting to put another chest drain in such cases as we had problems, so it is very wise to reconsider the diagnosis if things are not going in the right direction.

DISCUSSION

To the best of our knowledge, only seven cases of SJMS have been reported, while only one case was reported as having pneumothorax and SJMS together so we are presenting the second case where pneumothorax and SJMS presented together.

Competing interests None.

Patient consent Obtained.

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