

Case Report

Concurrent Operation Revealing Concurrent Spontaneous Pneumothoraces in Monozygotic Twins within 24 Hours

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Abstract

We describe the cases of concurrent pneumothoraces in monozygotic twin required Video-assisted thoracoscopic surgery (VATS). The two cases were experienced within 24 hours. The first case of right-sided pneumothorax was experienced in a 15-year-old male born as a second twin brother. The patient presented with a right chest pain, cough and discomfort that started a week prior hospital admission. The patient was diagnosed using chest radiographs and computed tomography. Trocar insertion was performed for aspiration and drainage, and the air leak was prolonged. The VATS at the beginning of the subsequent week during the afternoon, and the bulla was resected using a stapler. The next pneumothorax patient, who was the first twin brother, arrived with the same symptoms after the end of first operation. VATS was performed in the next morning, and the bulla was resected in the same manner as in the case of his brother.

Keywords: Identical twins; Monozygotic twins; Pneumothorax; Video-assisted thoracoscopic surgery

Introduction

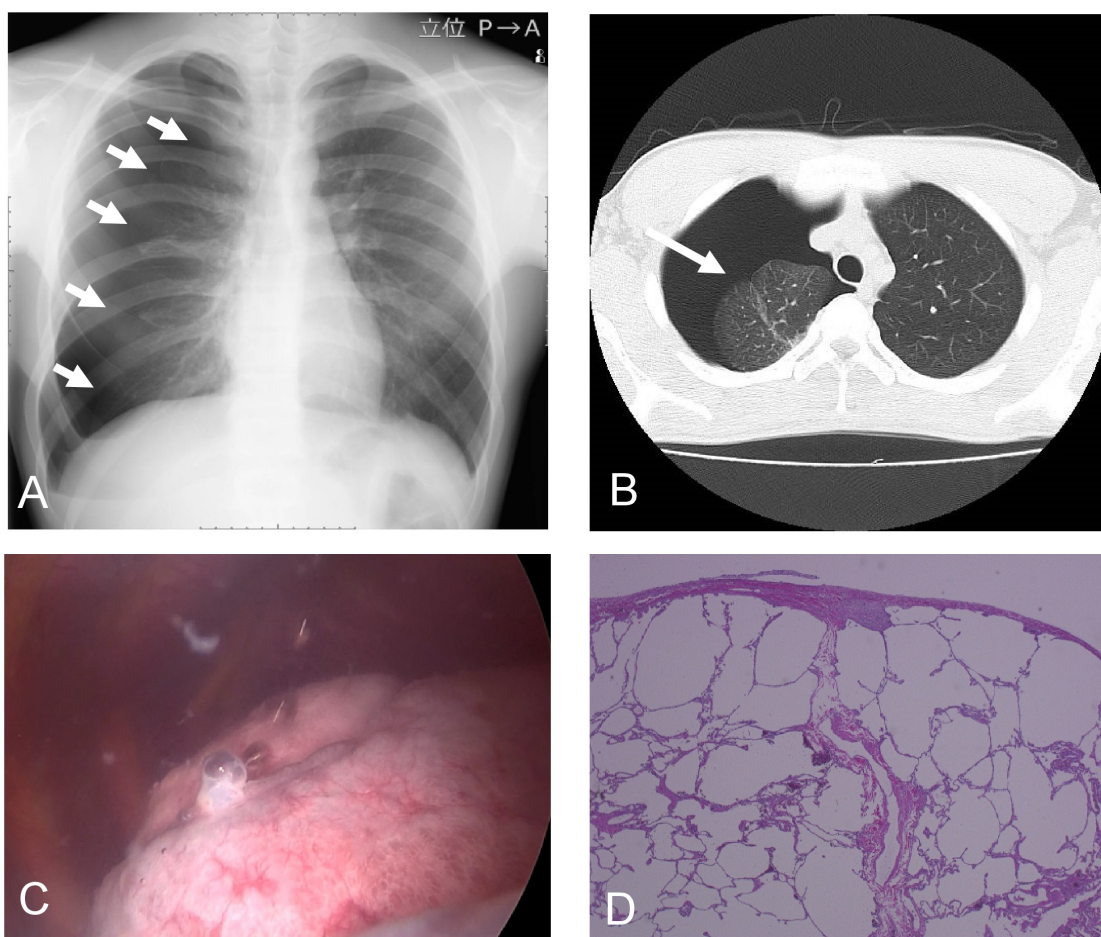
Primary spontaneous pneumothorax occurs at a frequency of 7.4-18 cases (age adjusted incidence) per 100,000 population per year in men and in 1.2-6 cases (age-adjusted incidence) per 100,000 population per year in women [1]. The pneumothorax typically occurs in young tall adults (peak age incidence 20-30 years), and the main risk factors include male gender, cigarette smoking and an asthenic physiognomy. Pneumothorax usually presents with sudden chest pain or discomfort, and erect posteroanterior chest radiographs are generally used to conclude the diagnosis. An early thoracoscopic intervention (within 3-5 days) is highly recommended in the case of persistent air leak [2]. The number of performed operations for spontaneous pneumothorax in Japan was 11,948 in the categories of general thoracic surgery during 2014 [3]. Among these cases, 3,410 patients (28.5 %) were reported to undergo bullectomy only, while 7,625 patients (63.8 %) required additional procedures in their operations. The simultaneous appearance of pneumothorax in twins is very unusual. Rashid et

al. were the first to report a concurrent pneumothorax in twins [4]. The authors presented the cases of simultaneous pneumothoraces in monozygotic twins within 24 hours, which were subsequently treated by surgery.

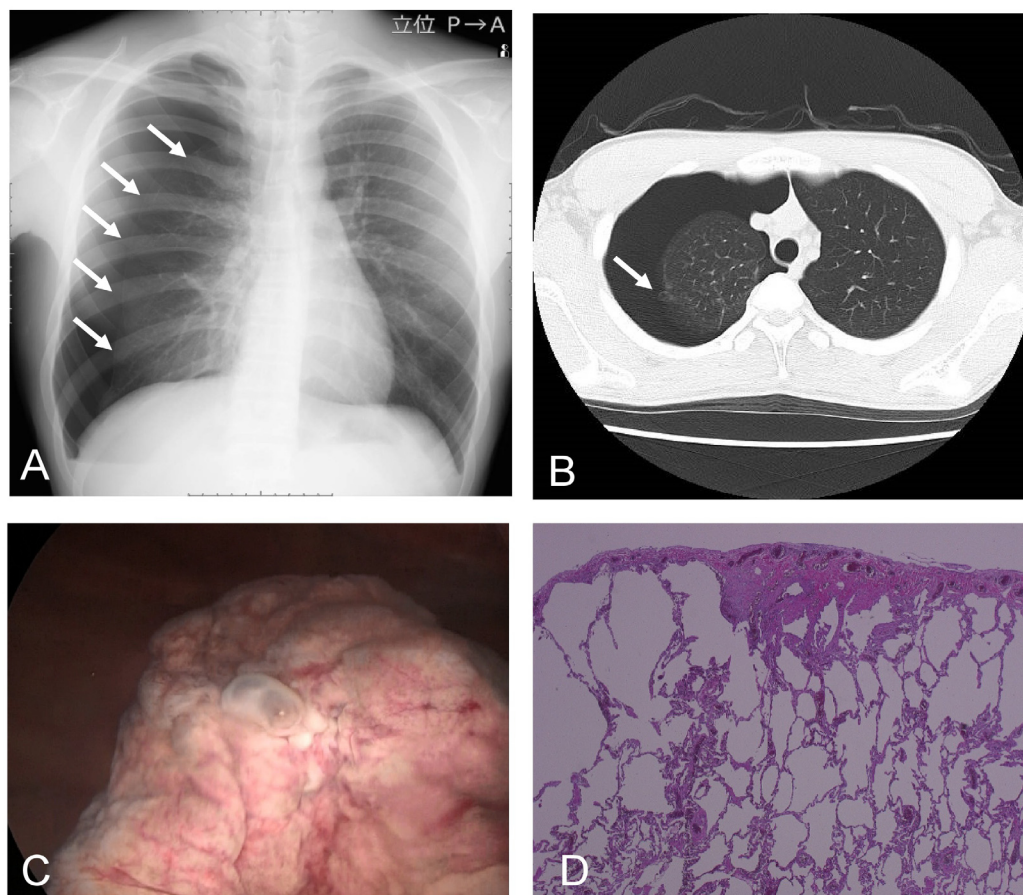
Case Report

The first patient was a 15-year-old male who presented with a right-sided chest pain, cough and discomfort that had started one week prior to the hospital admission. He was the second twin, and his identical twin brother was born with a birth weight of 1,080 g and periodic limb movement disorder from perinatal trouble. The patient was also born prematurely weight 900 g. The chest radiographs confirmed a pneumothorax on the right side (Figure 1A). The computed tomography (CT) of the lung also showed the pneumothorax on the right side but did not reveal the definite and not presence of Bulla (Figure 1B). We inserted a trocar for aspiration and drainage of the pneumothorax. The patient took his junior high school exams for 2 days at the hospital during the weekend. The air leak was prolonged, we then performed a video-assisted thoracoscopic surgery (VATS) at the beginning of the subsequent week during the afternoon. When the thoracoscope

was inserted into the thoracic cavity, an anterolaterally apical bulla was obtained. The leakage was confirmed by water seal test, and then the bulla was resected using the staplers (Figure 1C). When no more leakage was observed by the water seal test, a chest tube was inserted and the operation was completed. Another pneumothorax patient, who uses a wheelchair, directly arrived (Figure 2A). His face greatly resembled the face of the first patient, and so he was identified as the monozygotic twin of the first patient. The new patient also presented the right-sided chest pain, cough, and discomfort that had started a week prior to his arrival to the hospital. The CT of the lungs showed definitively the pneumothorax on the right side as well as a bulla at the same position as the one obtained in the previous case of the second twin (Figure 2B). However, this patient was belonephobic, and refused the insertion of chest tube, demanding a VATS instead. The emergency operation was scheduled in the morning of the next day. When the thoracoscope was inserted into thoracic cavity, the antero-lateral apical large bulla was obtained as in the case of second twin (Figure 2C). The resection of the bulla was performed, and a chest drain was inserted. Both of the twin brothers removed the chest drain on the second postoperative day. The twins were also discharged from the hospital together during same day. Regarding the pathological findings, the first twin had a bulla with pleural fibrous thickness (Figure 2D), whereas the second twin suffered from so-called emphysema-like changes with pleural fibrous thickness (Figure 1D). The second twin had a recurrent mild pneumothorax two months later, but he was treated by our observation. The cases of concurrent pneumothoraces, which required VATS intervention, were experienced in monozygotic twins within 24 hours.



Figures 1 (A-D): **A)** The chest radiographs confirmed a pneumothorax on the right side of the second twin. **B)** The CT showed the pneumothorax on the right and did not definitively reveal a bulla. **C)** The leakage was confirmed by water seal test, and bulla was then resected using the staplers. **D)** The pathological findings of the second twin revealed so-called emphysema-like changes with pleural fibrous thickness.



Figures 2(A-D): **A)** The first twin brother, considered as the monozygotic twin given the high level of resemblance of the faces, and had a right-sided pneumothorax. **B)** The CT showed the right-sided pneumothorax as well as a bulla at the same position as in the case of the second twin. **C)** The antero-lateral apical large bulla was obtained similarly as in the case of the second twin. **D)** The pathological findings showed that the first twin had a bulla with pleural fibrous thickness.

Discussion

Familial spontaneous pneumothorax is extremely uncommon. Although specific spontaneous pneumothorax is not reported, different syndromes such as Marfan syndrome, Ehlers-Danlos syndrome, and Birt-Hogg-Dubé syndrome have been shown to present spontaneous pneumothorax with other associated pathologies. To date, the phenotypic characterization has failed to demonstrate any associated connective tissue disorder or genetic pattern in these diseases. Several genetic subtypes of the human leukocyte antigen (HLA) haplotypes appear more frequently in familial cases -specifically the HLA A2 and B40. Alpha-1-antitrypsin has also been demonstrated to play a role in familial pneumothorax among Alpha-1-antitrypsin-deficient patients.

Considering appendicitis, monozygotic twins with simultaneous acute appendicitis are quite rare [5]. The literature

shows that only four cases of acute appendicitis have been identified in the world.

On the other hand, five cases of pneumothoraces in twins have been reported, and the simultaneous cases were four cases including the present case (Table 1) [4, 6-9]. Concurrent VATS bullectomies performed within 24 hours have not yet been reported. The probability of an identical twin pregnancy is about 1 in 250. Hence, considering the population of identical twins, if simultaneous pneumothorax were a genetic phenomenon in this patient group, the incidence of this entity would be expected to be significantly higher than the one already reported.

Although genetics may play a role in the development of pneumothoraces, environmental factors are probably the main causes of these clinical conditions. Therefore, the occurrence of simultaneous pneumothoraces in monozygotic twins is coincidental.

Author	Year	Age	Sex	Side	Concurrent	Surgery
Rashid A et al. [5]	1971	71	Male	Right	○	○
Tsukadaira A et al. [6]	2001	17	Male	Left	○	Not described
Castillo C et al. [7]	2005	17	Male	Left	○	○ First twin only
Inoue N et al. [8]	1982	15	Male	Bilateral		○
Hofmann LJ et al. [9]	2012	15	Male	Bilateral		○
Ours	2017	15	Male	Right	○	○

Table 1: Reported cases of pneumothorax in identical twin.

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References

1. Baumann MH, Noppen M (2004) Pneumothorax. *Respirology* 9: 157-164.
2. Mikroulis D, Lukman AL, Didili V (2005) Familial spontaneous pneumothorax. *Respirology* 10: 403.
3. Masuda M, Okumura M, Doki Y, Endo S, Hirata Y, et al. (2016) Thoracic and cardiovascular surgery in Japan during 2014. *General Thoracic and Cardiovascular Surgery* 64: 665-697.
4. Rashid A, Sendi A, Al-Kadhimi A (1986) Concurrent spontaneous pneumothorax in identical twins. *Thorax* 41: 971.
5. Gurbuz AT, Muckleroy SK, Davis-Merritt D (1996) Simultaneous acute appendicitis in monozygotic twins: coincidence or genetic?. *Am Surg* 62: 407-408.
6. Tsukadaira A, Okubo Y, Ota M, Hotta J, Kubo K (2001) Concurrent left-sided spontaneous pneumothorax in Japanese monogerminal twins. *Respiration* 68: 625-627.
7. Castillo C, Baquerizo A, Rodriguez P, Rodríguez JC, Iwamoto G (2005) Simultaneous left-sided, primary, spontaneous pneumothorax in identical twins. *Med Sci Monit* 11: Cs33-35.
8. Inoue N, Kitano I, Murakami T (1982) [Surgical treatment of spontaneous pneumothorax occurring in monovular twins--report of two cases]. *Kyobu Geka* 35: 447-450.
9. Hofmann LJ, Hetz SP (2012) Pediatric bilateral spontaneous pneumothoraces in monozygotic twins. *Pediatr Surg Int* 28: 745-749.