



# Surgery for simultaneous bilateral primary spontaneous pneumothorax: when and how?

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**Abstract:** Although primary spontaneous pneumothorax (PSP) is common in adolescents and young adults, simultaneous bilateral primary spontaneous pneumothorax (SBPSP) is an unusual and potentially life-threatening condition with limited data on its incidence in the literature. There are evidence-based guidelines for the management of PSP, whereas the specific treatment strategy is not clearly defined in SBPSP despite clinical practice recommendations that surgery should be offered at the first episode. Currently, for most thoracic surgeons worldwide, video-assisted thoracic surgery (VATS) has taken a leading role in the treatment of PSP, due to its well-known advantages including less pain, better cosmetics, and faster recovery. In this article, we aim to explore the aspects of epidemiology, pathophysiology, diagnosis, management, and evolution of surgical approaches related to SBPSP.

**Keywords:** Simultaneous bilateral spontaneous pneumothorax (SBSP); video-assisted thoracic surgery (VATS); one-stage

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## Introduction

Primary spontaneous pneumothorax (PSP) is a common benign disease affecting adolescents and young adults. In Taiwan, a national population-based cohort study in 2013 documented an incidence of 7.18/100,000 (1), which is comparable with the data (1.2 to 18/100,000) reported in Western countries (2). However, simultaneous bilateral primary spontaneous pneumothorax (SBPSP) is a relatively rare clinical event with limited data on its incidence in the literature (3). Its clinical symptoms and signs vary and include chest pain, mild shortness of breath, and even tension pneumothorax (4). Patients with SBPSP may deteriorate rapidly if left untreated. Therefore, early and prompt management is recommended when SBPSP is recognized. Although surgery is advocated for SBPSP,

based on international consensus mentioned in the leading guidelines (2,5), relevant studies of SBPSP are scarce and reported mostly as cases. The aim of this article is to provide a literature review of SBPSP, mainly focused on its epidemiology, diagnosis, management strategy, and future perspectives.

## Epidemiology

The incidence of simultaneous bilateral spontaneous pneumothorax (SBSP) has been reported to be around 7.8–20% of all cases of spontaneous pneumothorax (6). Studies by Sayar *et al.* and Akcam *et al.* reported that approximately 58% and 68% of patients with SBSP, respectively, had causative underlying pulmonary disease (3,7). Several medical conditions known to be associated

with SBSP include lung metastasis, tuberculosis, histocytosis X, and COPD (3). SBSP in a patient without an underlying medical condition is regarded as SBPSP, shown to account for 1.6% of first presentation in PSP patients in one large cohort study (6). In other large case series, SBPSP constitutes approximately 2-2.4% of patients operated for PSP (8,9).

### Pathophysiology

In PSP, air leaks through the visceral pleura, possibly induced by a rapid increase in transpulmonary pressure or defects of visceral pleura (10). While the exact pathogenesis of PSP is not completely understood, rupture of blebs/bullae that cause air to leak into the pleural space remains the significant factor explaining the occurrence of PSP. In fact, apical blebs/bullae are found in the majority of PSP patients during video-assisted thoracic surgery (VATS) and in nearly all patients during thoracotomy (11,12). These subpleural air-containing lesions are associated with other factors such as patient height, distal airway inflammation, hereditary predisposition, low body mass index, which may predispose to PSP (13). Additionally, some authors reported that patients with SBPSP tend to be younger, with lower body mass index, and have abnormal blebs/bullae (6,14).

Moreover, in extremely rare circumstances, SBPSP with congenital pleuro-pleural communication has been reported (15,16). The reported sites of congenital pleuro-pleural communication were the anterior mediastinum and the lower middle mediastinum. Therefore, the aforementioned mediastinal communication should be carefully inspected during surgery for SBPSP, and closure of the pleuro-pleural communication is essential to prevent future recurrence (16).

### Clinical manifestation and diagnosis

SBPSP, which is similar to PSP, occurs most often at rest. A typical physical examination tends to reveal decreased breath sounds and reduced chest expansion bilaterally. Although hemodynamic compromise or significant hypoxemia is uncommon in PSP, the simultaneous occurrence of PSP carries potentially more severe clinical symptoms such as chest pain, shortness of breath, and even tension pneumothorax (4). In light of the variability of presented clinical features, the diagnosis of SBPSP is often confirmed with radiographic imaging such as thoracic radiography or computed tomography. Since rupture of the blebs in the

lung is the most common cause of PSP, the presence of these air-containing lesions on high-resolution computed tomography (HRCT) has been regarded as a significant predictor for PSP recurrence (17). The reported incidence of blebs/bullae on HRCT ranged from 47% to 94% in ipsilateral lung (17-23) and 55% to 70% in contralateral lung (21-23). Further, Lee *et al.* mentioned that patients with SBPSP had a significantly higher incidence of blebs/bullae observed on HRCT compared with non-SBPSP (88.5% *vs.* 63.5%) (6). Therefore, in patients of SBPSP potentially requiring surgery, HRCT may be useful to identify more occult pulmonary lesions such as blebs/bullae, as well as guiding surgical management.

### Management

According to the British Thoracic Society (BTS) guidelines and American College of Chest Physicians (ACCP) Delphi consensus statement (2,5), SBPSP remains one of the indications for definitive treatment of PSP, such as pleurodesis or surgery. Nevertheless, no clear decision strategy regarding SBPSP has been established, possibly due to the scarcity of such case events.

In order to obtain the relevant treatment experiences for SBPSP, we identified case series over the last three decades in the literature (excluding single case report and studies with incomplete clinical feature and outcome) (shown in *Table 1*) (6-9,24-27). The following are the associated treatment strategies extracted from the literature with regards to treatment of SBPSP.

#### Chest drainage

Based on the aforementioned guidelines, supplemental oxygen therapy, percutaneous needle aspiration, or a more aggressive procedure like intercostal drain placement are recommended, depending on the clinical symptoms and the size of the pneumothorax (2,5). Following the same suggestions, unilateral or bilateral chest drainage should be administered for patients of SBPSP involving larger or symptomatic pneumothorax. For those PSP patients with mild-to-moderate extent of simultaneous occurrence, surgical intervention can be offered directly if not physiologically contraindicated (8). However, from a practical point of view, it could be difficult to make therapeutic recommendations according to these guidelines in developing countries with restricted facilities. Instead of VATS treatment, Flores-Franco *et al.* reported their

**Table 1** Patients with SBPSP in literature

First author	Year	Patient, n	Mean age (year)	Male, n (%)	One-stage VATS treatment, n (%)	Surgical approach	Mean follow-up (month)	Recurrence, n (%)
Graf-Deuel (24)	1994	5	22.8	4 (80%)	3 (60%) (pleurodesis only)	Multiportal, BS approach	73	0 (0)
Aye (25)	2002	4	21.5	4 (100%)	4 (100%)	Multiportal, BS approach	40	0 (0)
Chen (9)	2008	4	22.1	4 (100%)	4 (100%)	Multiportal, BS approach	46	1 (25%)
Lee (6)	2008	13	20.9	13 (100%)	7 (54%)	Multiportal, BS approach	44	2 (15%)
Song (26)	2013	2	18	2 (100%)	2 (100%) (no contralateral pleurodesis)	Multiportal, unilateral anterior TM approach	11	1 (50%)
Kim (27)	2017	2	17.5	2 (100%)	2 (100%) (no pleurodesis)	Uniportal, BS approach (supine position)	22	0 (0)
Cho (8)	2017	25	16.3	24 (96%)	25 (100%) (no pleurodesis)	Multiportal, unilateral apicoposterior TM approach (11 pts)	62	11 (44%)
Akcam (7)	2018	5	22.4	5 (100%)	3 (60%)	Multiportal, BS approach	36	2 (40%)

BS, bilateral sequential; TM, transmediastinal; pts, patients.

successful experience using bilateral catheter drainage with Heimlich valves for one patient of SBPSP. They considered that this alternative approach could be useful in places with limited resources for VATS (28).

### **One-stage operation**

With the advances in minimally invasive techniques, bilateral surgery for pneumothorax via conventional approach including sternotomy or thoracotomy was replaced by VATS in the early 2000s (24,25,29). Nowadays, one-stage bilateral VATS has been advocated as a standard method in managing patients with ipsilateral PSP and contralateral blebs/bullae (21,22,30,31). In the relevant literature regarding surgery for SBPSP, similarly, one-stage VATS treatment has been considered the preferable choice through multiportal and bilateral sequential (BS) approach (6,7,9,24,25,27).

### **Uniportal VATS**

In recent decades, since uniportal VATS has gained increasing feasibility and popularity, and its employment in the treatment of PSP and bilateral pulmonary lesions has been reported to be effective (32,33). In 2017, Kim used the one-stage uniportal VATS technique in the supine position to treat SBPSP patients, and succeeded in achieving less

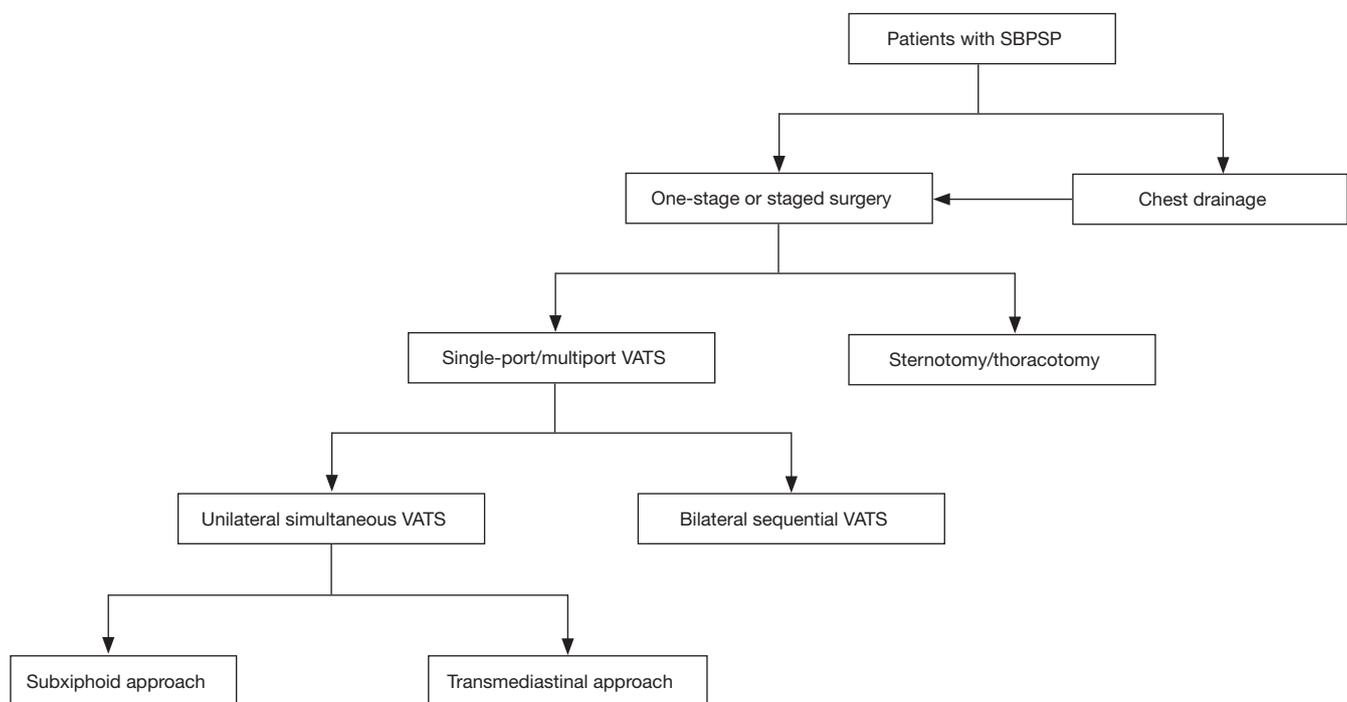
operative time and better cosmetics (27).

### **Unilateral trans-mediastinal approach**

Although one-stage VATS using BS approach has been mostly undertaken for treatment of SBPSP, it is associated with chronic incisional pain, poor anesthetic outcomes, and increased operative time. In the literature, Wu *et al.* reported the very first study treating ipsilateral PSP with contralateral bullae using anterior transmediastinal (TM) approach through unilateral incision (34). Later, several reports discussed this unilateral TM approach, which can also be performed in SBPSP, and showed its efficacy and low morbidity (8,26). Despite the favorable outcomes brought by this TM approach, including decreased operative time, duration of drainage, hospital stay, and potential cosmetic effect, this procedure has certain contraindications, such as a history of sternotomy and intolerance to single-lung ventilation. Notably, the authors also claimed that, in their experience, VATS bullectomy through this unilateral TM approach is more suitably performed on the right side.

### **Subxiphoid approach**

Moreover, in pursuit of reducing acute and chronic postoperative wound pain, subxiphoid VATS has been adopted in a wide array of thoracic operations, including



**Figure 1** Algorithm for surgical approach in SBPSP. SBPSP, simultaneous bilateral primary spontaneous pneumothorax; VATS, video-assisted thoracic surgery.

major lung resections (35,36). However, subxiphoid approach for SBPSP is not mentioned in the literature. To date, subxiphoid approach for patients of ipsilateral PSP with contralateral blebs/bullae only has been reported in limited studies (37,38). Their results showed that significantly lower postoperative pain scores and longer operative time were observed in the subxiphoid VATS group compared with the transthoracic VATS group. Interestingly, perioperative data including duration of drainage and hospital stay did not differ significantly.

## Summary

Although SBPSP is rare and its clinical features variable, ranging from mild dyspnea to tension pneumothorax. Based on guideline recommendations, once early diagnosis is established and/or urgent chest drainage is administered, such SBPSP patients who are inherently young and physiologically fit are considered suitable to undergo one-stage VATS treatment.

The current trend in VATS is to utilize fewer ports and less invasive access to reduce postoperative wound pain, chronic wound paresthesia, operative time, duration of

drainage, and hospital stay. Even though the aforementioned innovative VATS techniques using TM or subxiphoid approach have demonstrated satisfactory results for simultaneous resection of bilateral pulmonary bleb/bullae, the long-term outcomes including recurrence rate have not been validated due to the limited number of cases and studies. For example, we know that the role of pleurodesis is essential in the prevention of postoperative recurrence after VATS bullectomy. Because contralateral pleurodesis cannot be efficiently performed through the unilateral TM approach, this may have contributed to the high recurrence rate (44%) in the largest case series of SBPSP to date (8). In addition, subxiphoid VATS may potentially be an alternative way to manage patients with SBPSP in selected conditions, but this necessitates further clinical studies to verify its efficacy and feasibility. An algorithm for the surgical approach in SBPSP has been proposed and is demonstrated in *Figure 1*. In conclusion, all the above-mentioned novel techniques should be used appropriately in carefully selected patients. Surgical access should be safe and easily reproducible in order to be employed by thoracic surgeons on a routine basis. Otherwise, one-stage VATS using BS approach remains the reliable and effective procedure in the

treatment of SBPSP.

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