

# Results of the surgical treatment of pulmonary bleb and bullous disease: A retrospective study

Rastin Hosseinzadeh Asli<sup>1,2,3</sup>, Manouchehr Aghajanzadeh<sup>3</sup>, Mahtab Raji Lahiji<sup>1,3</sup>, Hossein Hosseinzadeh Asli<sup>2</sup>, Ali Alavi Foumani<sup>3</sup>, Yousha Pourahmadi<sup>3</sup>

<sup>1</sup>Department of Thoracic and General Surgery, Guilan University of Medical Sciences, Rasht, Iran, <sup>2</sup>Department of General Surgery, Aria Hospital, Guilan University of Medical Sciences, Rasht, Iran, <sup>3</sup>Department of Internal Medicine, Inflammatory Lung Diseases Research Center, Razi Hospital, School of Medicine, Guilan University of Medical Sciences, Rasht, Iran

## ABSTRACT

**Objective:** Bullous lung disease is characterised by the development of bullae within the lung parenchyma. Smoking is considered as a main risk factor of bullae formation. The clinical manifestation varies from asymptomatic bullae to severe respiratory distress. The exact medical approach is still controversial. Here, we aimed to evaluate the patient clinical characteristics and the indication of surgery in the patients. **Methods:** This study was an analytic cross-sectional study in Razi hospital, Rasht, Iran, in 2016–2020. A total of 110 patients with newly diagnosed lung bullae underwent evaluation based on a checklist, which assessed the variables as clinical characteristics, bullae features, the medical treatment, and its complication. Eventually, statistical analysis was performed by using SPSS v21.0. **Results:** A total of 110 patients were enrolled. All the patients were smokers. In 72.9%, the primary presentation was respiratory distress, which needed surgical resection with no recurrence over 8 months. The clinical manifestation varies from asymptomatic bullae in 4.3% and pneumonia in 34.3% to respiratory distress in 61.4%. There was no statistical correlation between clinical manifestation and all the studied variables excluding respiratory distress (p value: 0.659). Seventy patients underwent surgical treatment, in which 20 cases were complicated by air leak that was managed by the chest tube in 16 patients and the Heimlich valve in four patients. No evidence of recurrence was detected. **Conclusion:** The main risk factor of bulla formation is smoking. Because of various clinical presentations, the appropriate medical approach is of paramount importance.

**KEY WORDS:** Bleb, pulmonary bullae, respiratory distress, smoke

**Address for correspondence:** Dr. Rastin Hosseinzadeh Asli, Guilan University of Medical Sciences, Parastar Steet, Rasht - 41937-1311, Iran.  
E-mail: rastinhz@gmail.com

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## INTRODUCTION

A bulla is an air-filled space within the lung parenchyma that is about 1–2 cm in size with a fibrotic wall that is bordered by the remnants of alveolar septae.<sup>[1]</sup> Smoking is considered as a main risk factor of bullae formation.<sup>[2–4]</sup> Despite the high prevalence of this disorder, the exact

role of bullae in pathogenesis of pneumothorax is still unclear.<sup>[5,6]</sup> Lung bullae are categorised into two main groups: bullae within otherwise a normal lung parenchyma which is usually single and bullous formation within an emphysematous lung which are usually multiple.

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A single huge bulla which occupies more than 30% of the hemi-thorax is considered a giant bulla.<sup>[1]</sup> Although the bullous lung disease often occurs in the context of pulmonary disorder as chronic obstructive pulmonary disease (COPD), it could occur in otherwise healthy lungs as an incidental finding on imaging.<sup>[7]</sup> The pathophysiology of bullous formation is a parenchymal weakness resulting in localised air trapping. Consequently, the pressure induced by the elasticity of the adjacent lung leads to build-up of pressure within bullae, contributing to their progressive enlargement and compression and atelectasis of the adjacent lung. Identification and re-construction of the intact parenchyma are the main aims of treatment.<sup>[1]</sup>

A bullae is seen as an avascular, high-radiolucent area with a thin or poorly defined wall. Standard chest X-rays could estimate the size, location, and volume of the bullae. However, to determine the exact size, chest computed tomography (CT) is required to demonstrate detailed characterisation of the size, number, and location of bullae. CT is a sensitive tool which reveals the exact anatomic location of the bullae, a feature which might be easily missed in the anteroposterior or lateral view of chest X-rays. Furthermore, it is capable of differentiating an emphysematous bullae from a pneumothorax.<sup>[8-10]</sup> Lesur *et al.*<sup>[9]</sup> showed that by the use of CT scan, the discovery of bullae had been on the rise. The prevalence of incidental bullae was observed to be 0–15% in a healthy population. There are a few reports of sonography application to recognise an emphysematous bullae from the pneumothorax.<sup>[11]</sup> Regarding modern diagnostic methods, it is noted that 75–100% of patients with a pneumothorax have simultaneous bullae. However, no study has clearly mentioned these changes as etiologic factors in pneumothorax pathogenesis. Furthermore, the exact acquired or genetic basis of these bullae formation is not clear.<sup>[12-14]</sup>

As mentioned before, affected patients tend to have significant exposure to tobacco smoke.<sup>[2-4]</sup> Gaensler EA *et al.*<sup>[15]</sup> showed that all their patients with bullous lung disease had a history of smoking and smoke cessation improved lung changes which secondarily contributed to better response to surgery.

Although the majority of patients benefit from a medical treatment which is the same as COPD management, the large emphysematous bullae do not have an adequate response to standard conservative treatment and further surgical therapy is needed. Based on the patients' clinical status, diverse surgical approaches such as bullectomy, lobectomy, and pneumonectomy might be considered.<sup>[16-18]</sup>

Regarding diverse clinical manifestations as asymptomatic bullae in one side of the spectrum compared to serious features such as the pneumothorax, hemoptysis, or compressive effects, it is of paramount importance to design an appropriate medical guideline in approach to lung bullous disease.<sup>[19]</sup>

In this study, we aimed to assess the factors such as characteristic features, the possible complication, the selected diagnostic and therapeutic method, surgery indication, and prognosis of the patients with bullous lung disease who were referred to our center since 2016–2020 in Razi hospital, Rasht, Iran.

## METHODS

This study was an analytic cross-sectional study in Razi hospital, Rasht, in 2016–2020.

A total of 110 patients of any age and sex with newly diagnosed bullous lung disease underwent evaluation. The diagnosis was based on clinical suspicion and chest CT scan.

The data collection tool was a checklist prepared by referring to the patients' file which included the patients' characteristics such as age, sex, past medical history, habitual history, and clinical manifestation and bullae features such as size, location, and the site of involvement. Furthermore, the medical therapeutic approach command in the case of surgery, the complication and prognosis, and the time of hospitalisation were recorded. It should be noted that the patients with incomplete information were excluded.

After data collection, statistical analysis was performed by using SPSS v21.0. Normal data distribution was performed by Kolmogorov–Smirnov test. For the comparison of the quantitative variables, independent two-sample t-test was performed, and for the qualitative variables, Chi-square test was performed. A *P* value less than 0.05 was considered significant.

## Ethical statement

This study has been accepted by the Inflammatory Lung Disease Research Centre of Guilan University of Medical Sciences, Rasht, Iran, and was conducted in accordance with the Declaration of Helsinki.

## RESULTS

A total of 110 patients were enrolled in the study, of whom 70 underwent surgical management. The mean age ( $\pm$ SD) of study participants was  $50.9 \pm 12.83$ . The least age and the highest age of the patients were 18 and 80 years, respectively. 70% of the patients were male, and in 93%, the past medical history was negative. 72.9% had a history of cigarette smoking. The clinical manifestation varied from asymptomatic bullae in 4.3% and pneumonia in 34.3% to respiratory distress in 61.4%. In 55.7%, there was a single bullae compared to multiple bullae in 27.1%. The right and left lungs were involved in 54.3% and 18.6%, respectively. In addition, in 27.1%, both sides were involved. The 70 patients underwent surgical treatment, in which 20 cases were complicated by air leak

that was managed by the chest tube in 16 patients and the Heimlich valve in four patients. The later follow-up revealed complete recovery in these patients over a month. Furthermore, the mean hospitalisation time was 11 days. No evidence of recurrence was detected. The patients' characteristics are brought in Table 1.

There was no statistical correlation between clinical manifestation and all the studied variables excluding respiratory distress, which showed a meaningful statistical relationship based on the Cramer's index (p value: 0.659) [Table 2].

The chest X-ray pattern and different views of patients' lung CT scans consisting of bullae are shown in Figure 1. The surgical resection of pulmonary bullae including processes of resecting and the resected bullae are displayed in Figure 2.

**DISCUSSION**

The fluid level in a bullae has an exclusive pathology which presents itself in various types. The exact etiology of lung bullous diseases is unknown. However, the probable hypothesis is inappropriate bronchial connection leading to incomplete drainage of the fluid and consequent fluid collection, which results in inflammation and infection.<sup>[1,7,19,20]</sup>

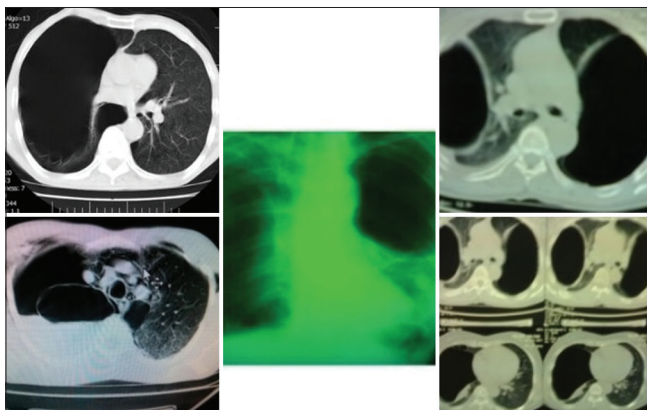
In our study, we evaluated 70 patients with newly diagnosed bullous lung disease. The majority of the patients were male, similar to the study of Gunnarsson *et al.*,<sup>[21]</sup> who observed 12 patients with giant bullae over 18 years. They revealed that 11 patients were male with an average age of 60 years. In contrast, in the study of Schipper *et al.*<sup>[18]</sup> on 43 patients with giant emphysematous bullae, they found that the majority of the patients were female, and in the study of Amjadi *et al.*,<sup>[22]</sup> nine out of 15 patients with bullous lung disease were female.

It should be noted that smoking is considered as an important risk factor of bullae formation.<sup>[2-4]</sup> Schipper

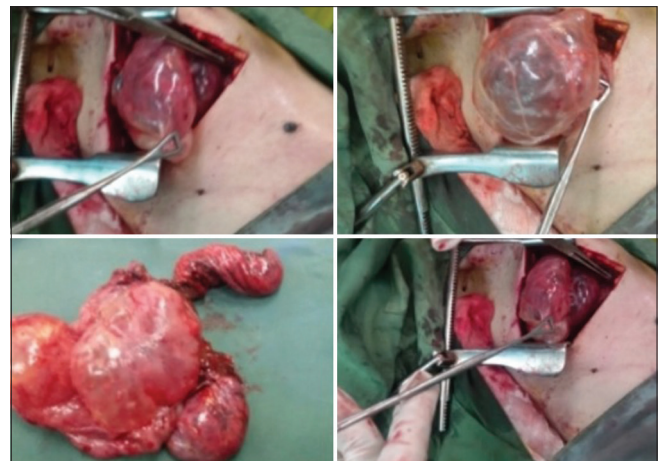
*et al.*<sup>[18]</sup> showed all their patients were smokers with a 50 pack/year consumption. Similarly, Lesur *et al.*<sup>[9]</sup> evaluated the relationship between smoking and bullae in an idiopathic spontaneous pneumothorax in a case-control

**Table 1: Patient characteristics**

Variable	Number	Percentage
Age		
<35 y/o	10	14.3
35-80 y/o	33	47.1
>80 y/o	27	38.6
Gender		
Male	49	70
Female	21	30
Past medical history		
Negative	65	92.9
Positive	5	7.1
Habitual history (smoking)		
Negative	19	27.1
Positive	51	72.9
Clinical presentation		
Incidental	3	4.3
pneumonia	24	34.3
Respiratory distress	43	61.4
Bullae size		
Small (2 to 6 cm)	18	25.7
Large (7 to 20 cm)	52	74.3
Bullae number		
Single	39	55.7
Two	12	17.1
multiple	19	27.1
Bullae location		
Rt side	38	54.3
Lt side	13	18.6
Both sides	19	27.1
Lung involvement		
Negative	51	72.9
Positive	19	27.1
Therapeutic approach		
Conservative	16	22.9
Resection	52	74.3
Resection and conservative	2	2.9
Hospitalisation period		
<5 days	7	10
5-7 days	56	80
>7 days	7	10



**Figure 1:** CT scan with huge right- and left-side bullae



**Figure 2:** Complete and intact bullae during and after resection

**Table 2: Clinical presentation**

	Incidental	Pneumonia	Respiratory distress
Respiratory distress			
Negative	66.7%	4.2%	0%
positive	33.3%	95.8%	100%

Linear-by-Linear Association=4.191 df=1 Sig. = 0.041.Cramer's V=0.659 Sig. = 0.000

study. Their finding revealed that all their patients were smokers. Besides, in 85% of the case group and 155 of the control group, a simultaneous bullae was detected.

In our study, 72.9% of the patients were smokers, similar to the previous studies.

Although lung bullous disease might be asymptomatic and found only incidentally on imaging, it is usually associated with pulmonary malfunction. Sudden and severe dyspnea should raise the suspicion for the pneumothorax because of a ruptured bullae which occurs mostly in the context of underlying emphysema.<sup>[7,23-25]</sup> Our study showed that the most common clinical presentation was respiratory distress, in which case the patients had been under surgical resection with a complete recovery over a month.

It is of paramount importance to take an appropriate medical therapy in patients with severe distress or superimposed infection. These patients can be candidates for surgical treatment. Currently, the classic surgical method for isolated bullae is bullectomy, which involves surgical resection or ablation of one or more bullae.<sup>[16-18,26-30]</sup>

In our study, 52 patients were under bullectomy with a significant response and on recurrence which was similar to other studies, which highlighted the importance of surgical resection in the case of respiratory distress.

**CONCLUSION**

Bullous lung disease is characterised by the development of bullae within the lung parenchyma. Although it usually occurs in the context of pulmonary disorder, it might be seen in isolation. The most common risk factor is smoking, which was also evident in our study.

Clinical manifestation varies from incidental bullae to severe respiratory distress. In our study, most patients were presented with respiratory distress which showed the positive role of surgery in approach to lung bullae.

The limitation of the current study is the lack prolonged follow-up. As a result, there was no chance to evaluate the long-term outcome.

We suggest further studies in a case-control design with a higher sample size and consideration of other variables such as body mass index.

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**Conflicts of interest**

There are no conflicts of interest.

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