ORIGINAL ARTICLE

CLINICAL RESULTS AFTER INTRA-CAVITARY DRAINAGE OF GIANT BULLAE IN PATIENTS WITH POOR PULMONARY RESERVES

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Background: Giant bullae (GB) are space occupying lesions associated with chronic respiratory symptoms. The aim of this study is to evaluate outcome of intra-cavitary tube drainage procedures (ITDP) in terms of clinical and radiological benefits. **Methods:** A prospective study was conducted in The Department of Thoracic Surgery, Jinnah Postgraduate Medical Center, Karachi, from February 2021 to April 2022 after ethical approval. Patients above 12 years, with poor reserve and GB underwent clinical, radiological and laboratory assessment before and after ITDPs to document various studied parameters. **Results:** A total of 48 patients were included; thirty-two (66.7%) were males. Mean age was 46.7±12.14 years. Most common aetiology was COPD (28; 58.3%). GB were ≥10 cm in size in 36 (75%) with right upper lobe involvement in 20 (41.7%). Preoperative dyspnoea score of IV was seen in 41 (85.4%) and chest pain in 42(87.5%) patients. In 34(70.8%) patients, Monaldi procedure and in 14 (29.2%) Brompton technique was used. Dyspnoea score improved from grade IV to II (24/41; p=0.004) along with reduction in pain and cough (p=0.012; p=0.002), respectively. Improvement post operatively in oxygen saturation, forced vital capacity, forced expiratory volume in 1 sec (6.08±1.36%, 0.73±0.516 L and 0.57±0.07 L, respectively, p<0.001) was seen. Partial pressure of oxygen (PaO2) and carbon dioxide improved by 40.6±4.82 (p=0.009) and 13.22±3.62mmHg (p=0.7). Improvement of PaO2 was associated with reduction in the size of bullae (9.33±5.13cm; p=0.006). Radiographical resolution was seen in 41 (87.5%) majorly within 2 months (21; 51.2%). Duration of hospital stay was 4.20±0.92 days with no mortality. Complications were seen in 25 (52.1%) patients. **Conclusion:** Intra-cavitary tube drainage procedures allow both clinical and physiological improvement in patients with GB. They allow resolution of bullae in patients with poor reserves and help in expansion of underlying compressed lung, improving both the clinical symptoms and radiological picture.

Keywords: Giant bullae; Monaldi; Brompton; Dyspnoea; FEV1; FVC; Intra-cavitary drainage; Bullous lung disease

Citation: Lodro MS, Mazcuri M, Ahmad T, Abid A. Clinical results after intra-cavitary drainage of giant bullae in patients with poor pulmonary reserves. J Ayub Med Coll Abbottabad 2023;35(1):21–6.

DOI: 10.55519/JAMC-01-10948

INTRODUCTION

Bullae are air filled spaces in the lung parenchyma. When such spaces occupy more than 30% of the volume within the thoracic cavity, they are referred to as Giant Bullae (GB).[1–4] The development of bullae is a chronic process where destruction of alveolar tissue results in areas of dead space that do not participate in gaseous exchange.[1,3–6] GB is a rare aetiology with a population base study reporting 0.21 cases per 100000 each year.[7]

A wide range of aetiologies have been proposed for the development of bullae including smoking or drug abuse and genetic disorders such as Marfan Syndrome, α1 antitrypsin deficiency, etc.[4–8] GB in some patients may not exhibit any symptoms while others may complain of pain in chest and difficulty in breathing.[4,6,8,9] Symptoms appear as a result of bulla occupying space, increasing size, lack of gaseous exchange, compression of underlying lung tissue and interference with the diaphragmatic function.[3,5,6] Few may present with complications such as infection, pneumothorax or hemoptysis.[8,9]

Chest x-ray (CXR) and Computed tomography (CT) are radiological investigations that are frequently carried out.[4,6] CT chest allows differentiation from pneumothorax, identification of any underlying lung disease and function of both the compressed lung and that of the bullae.[4-6,8–10]

Surgery (plication, excision, and resection) has been advocated as the best management option for improving the quality of life and avoiding unwanted complications in fit patients.[1,4-6] Intracavitary tube drainage procedures (ITDP) have been done by others in unfit patients with poor pulmonary reserves for the improvement of symptoms.[1,5,6]

Major surgeries are contraindicated for patient with poor reserves and ITDP such as Monaldi or Brompton technique have gained

importance.[2,5,10] However, there are only few significant regional studies available on intra-cavitary drainage procedures so far.[3]

The purpose of this study is to the outcome of ITDPs in terms of improvement in symptoms, exercise tolerance, bed side oxygen saturation (SaO2), partial pressure of oxygen (PaO2), partial pressure of carbon dioxide (PaCO2), and clinical and radiological evidence of lung expansion. We also aim to add to the scarce data in both regional and international literature on management of giant bullae in patients with poor cardiopulmonary reserve.[3,4,9]

MATERIAL AND METHOD

This prospective interventional study was conducted in the Department of Thoracic surgery after approval of the ethical review committee from 09-02-21 to 09-4-22.

Patients more than 12 years of age with radiological evidence of GB occupying 1/3rd or more of hemithorax were included in this study. Other factors taken into account for inclusion were De Vries classification of BLD (bullous lung disease) III-V [5] and Modified Hugh-jones dyspnoea (MHJD) scale grade of IV and V.[2,5,7,10] Patients with good pulmonary reserve evident by MJHD grade I to II, those below 12 years of age and De Vries BLD grade I, II were excluded from this study.

All patients underwent careful history-taking, physical and radiological assessment. Patients underwent pulmonary function tests including bed side spirometry, six-minute walk test (6MWT), and arterial blood gases prior to surgery. All were repeated at different intervals in post-operative (PoP) phase for comparison. Before intervention all patients were counselled for PoP complications. Informed consent was obtained.

All procedures were performed under aseptic measures. The patients were kept in supine or semi supine position under sedation with midazolam and local anaesthesia was infiltrated. The site of incision was determined according to site of the bulla and underlying compressed lung guided by pre-procedure radiological investigations. A 5–10 cm incision was made with up to 2.5 cm rib resection. In Brompton technique (single staged procedure), pleura was incised and bulla was opened and inspected for septa which were divided for free communication. Talc was liberally insufflated into the bulla cavity followed by placement of 24 Fr foleys catheter (FC). Balloon of the catheter was filled with 20–30 ml of air or distil water and purse-string sutures were placed on the visceral pleura. Wound closed in standard method. A chest tube was placed and secured in the pleural cavity from separate stab wound in chest wall, kept for average three days (range from 24 hours to 3 weeks). Later on the chest tube was placed on suction for 24–36 hours and removed as soon as air leak disappeared, whereas the FC was placed on water seal or Heimlich valve® for two weeks or above.[11] The two staged procedure also known as Monaldi procedure was performed in a manner that in the first stage chemical pleurodesis performed to avoid pneumothorax.[5] In the second stage FC is placed within bulla after application of double purse string suture ensuring inclusion of pleura and bulla together.[5] In each case after passing FC gush of air was noticed along with air leak observed in chest drain. Patient was assessed subjectively and objectively for improvement in symptoms, SaO2 and for any radiological changes. All patients underwent arterial blood gases and SaO2 assessment following procedure during hospital stay and at three months. Spirometry was performed post-operatively at one month and three months.

Relevant clinical and demographic data were systematically documented in the formulated data form. Procedural details, radiographic findings, pulmonary function values and complications encountered were recorded. Statistical analysis was undertaken via SPSS for Windows version 22.0 (IBM SPSS, Chicago, IL, USA). Mean with standard deviation (SD) and frequencies with percentages were utilized per variable need. Chi square was applied for correlation.

RESULTS

A total of 48 patients were included in this study, 32 (66.7%) were males and 16 (33.3%) females. Mean age was 46.7±12.142 (range 46–71) years and duration of symptoms was 26.23±14.03 months. Co-existing pathologies included tuberculosis (Tb) in 12 (25%), chronic renal failure in 5 (10.4%), rheumatoid arthritis and systemic lupus erythematosis 4(8.3%) in each respectively. Twenty-eight (58.3%) were smokers and chronic steroid use was seen in 21 (43.8%). Table -1 represents stratification on basis of size and site of bullae.

Pre-operative dyspnoea score (PRDS) was grade IV in 41 (85.4%), six-minute walk test (6MWT) less than 100 meters in 36(75%) patients and chest pain was seen in 42 (87.5%). Post-operative dyspnoea score (PODS) improvement was seen from grade IV to II (24/41; p=0.004) and III (8/41; p=0.034). Grade V patients dyspnoea

improved to grade III (3/7; *p*=0.034). Those patients who were unable to walk pre operatively were seen walking ≥100m but ≤1000 m in all four cases (*p*=0.001). Those covering distance of ≤100m were able to walk ≥1000 m (28/36; *p*=0.441). Table-2 shows comparison of symptoms and performance status between preoperative (PrP) and PoP group.

All patients under went spirometry for assessment of lung mechanics. Figure 1 and 2 represent changes of FVC (forced vital capacity) and FEV1 (forced expiratory volume in one second) in all patients respectively.

In 34 (70.8%) patients Monaldi procedure was utilized, whereas Brompton technique was used in 14 (29.2%). Improvement of symptoms was seen in 43 (89.6%) patients and statistically significant with both procedures (*p*=0.008). In all patient's significant improvement were seen post operatively in terms of SaO2 at room air, FVC, FEV1 (*p*<0.001 respectively). Reduction of size of bullae and PaCO2 was also noted though not statistically significant (*p*=0.291, 0.7 respectively). Table-3 represents changes and difference in various parameters at different time intervals.

Following procedure, significant improvement of PaO2 (*p*= 0.009) and cough (*p*=0.002). Improvement of cough correlated with reduction in size of bullae (*p*=0.006). Chest pain reduced significantly in 19 patients (*p*=0.012). Resolution of bulla on radiology was considered when size was reduced to 80 percent of original size seen in 41 (85.4%). Out of 41 resolution was seen in 21 (51.2%) in two months, 12 (29.2%) in four weeks and 8 (19.5%) within three months. No change in radiology was seen in 7 (14.6%) patients. Figure-3 shows CXR with different time interval of reduction in size of bulla.

During the study mean duration of hospital stay was 4.20±0.92 days with no mortality. Complications were seen in 25 (52.1%) including catheter block in 12 (48%), subcutaneous emphysema in 8 (32%), accidental removal of catheter in 7 (28%), wound infection 6 (24%) and prolonged air leak in 4 (16%). Various complications were found coexisting with each other out of which accidental removal of catheter was related to wound infection in three cases (*p*=0.009). Recurrence of bulla was seen in 5(12.1%) patients. All patients were planned a follow up at four intervals, i.e., 15 days, 1st month, 2nd month and 3rd month. Twelve (25%) patients had additional visits other than the proposed program.

Table-1: Location and Size of Bullae

SITE	
Right Upper Lobe	20(47.7%)
Left Upper Lobe	16(33.3%)
Right Lower Lobe	8 (16.7%)
Left Lower Lobe	4 (8.3%)
Size of bullae	
>10 cm	36 (75%)
5-10 cm	12(25%)

Table-2: Stratification of pre-operative and post-operative group in symptoms and performance status

	Pre-operative	**Post-operative**
MHJD Scale[8]		
Grade I	Nil	12(25%)
Grade II	Nil	24(50%)
Grade III	Nil	12 (25%)
Grade IV	41 (85.4%)	Nil
Grade V	7 (14.6%)	Nil
6MWT *		
Unable to perform	4 (8.3%)	Nil
≤100m	36 (75%)	4 (8.3%)
>100m < 1000 m	8 (16.7%)	12(25%)
≥ 1000m	Nil	36 (75%)
Chest pain		
Present	42 (87.5%)	23 (47.9%)
Cough		
Present	28 (58.3%)	8(16.7%)

*MHJD= Modified Hugh Jones dyspnoea, 6MWT= six-minute walk test

Table-3: Changes in parameters in pre-operative and post-operative cases

	Pre-operative	**Postoperative**			**Difference**
		Within 15 days	1 month	3 months	
Sao2 at room air (%)	86.66±2.52	89.93±2.39	-	92.87±2.63	6.08±1.36
PaO2(mmHg)	44.47±2.26	76.45±4.49	-	85.03±4.91	40.6±4.82
PaCO2(mmHg)	56.53±3.41	47.87± 3.11	-	43.04±3.69	13.22±3.62
Size of Bullae (cm)	12.88±3.94	-	-	3.47±4.94	9.33±5.13
FEV1 (L)	Unable to perform	-	0.81±0.12	1.38±0.17	0.57±0.07
FVC (L)	Unable to perform	-	1.90±0.43	2.64±0.61	0.73±0.516

SaO2= oxygen saturation, PaO2=partial pressure of oxygen, PaCO2= partial pressure of carbon dioxide PaCO2, FVC= forced vital capacity , FEV1=forced expiratory volume in one second

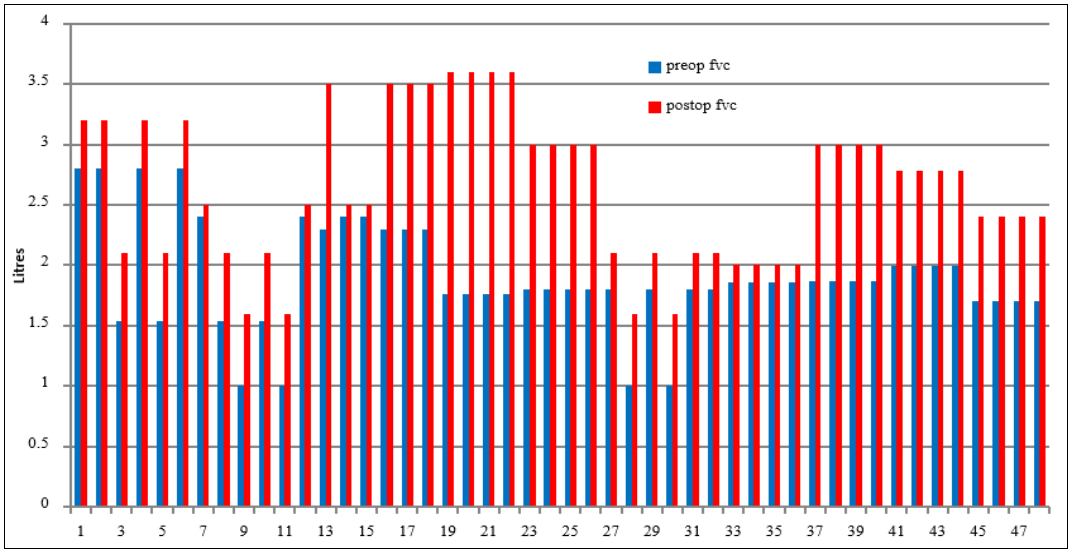

Figure-1: Preoperative and Post operative FVC

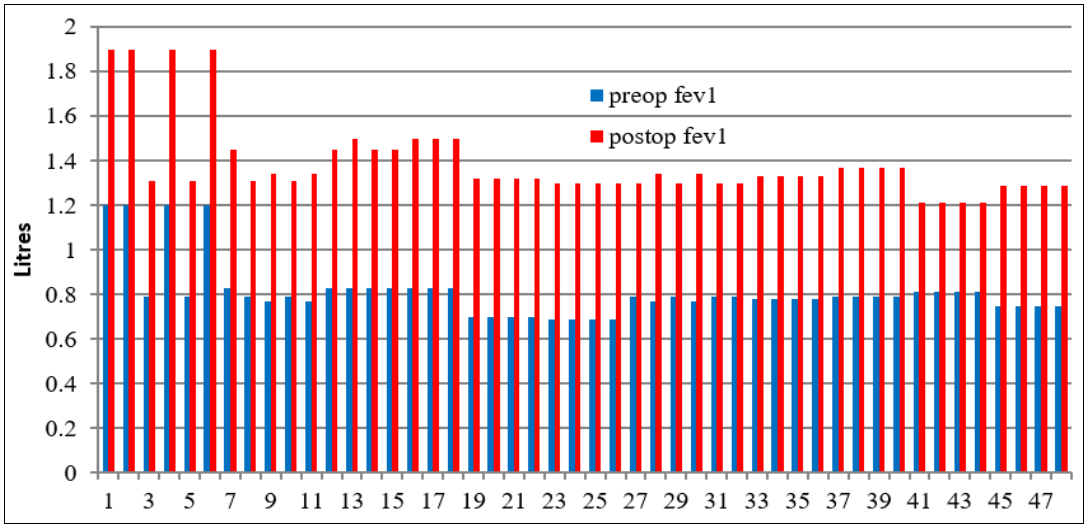

Figure-2: Pre and Postoperative FEV1

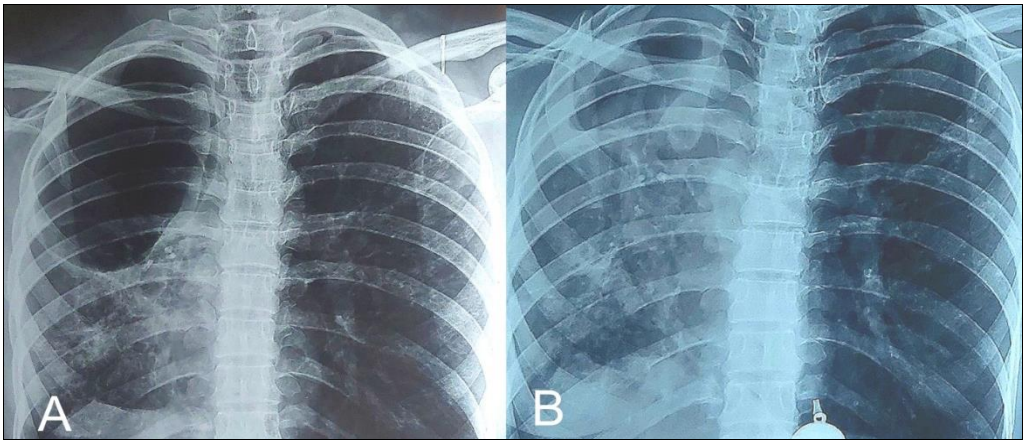

Figure-3: CXR representing different intervals of reduction in size of bullae.
A= Preoperative Xray B= four weeks

DISCUSSION

Bullous lung disease is a chronic aetiology that requires some form of surgical intervention. Patients not fit for general anaesthesia require intracavitary drainage under sedation. Lone *et al* reported an incidence of 21.6 cases per year.[12] Comparatively a larger prospective study reported up to 13 patients suffering from BLD requiring surgery each year.[13] BLD with GB has a lower incidence and is often mistaken for pneumothorax.[2,7,8] For GB, literature reports 80–90% male predominance.[2,7,14,15] In our study we found GB affecting 66.7% males. The mean age of presentation according to our study was 46.7±12.142 (range 46–71) years which coincides with various studies reporting its presentation between ages 40–70 years.[7,15,16] In our study 28 (58.3%) patients were smokers which is lower than reported in literature that is 63-100%.[2,4,12] Tuberculosis was seen in 12 (25%) patients which is higher than reported by Lone *et al.*[12] In literature, idiopathic aetiology for GB has been reported in 36% patients[12] but we do not report any idiopathic or genetic cases. GB is seen mostly in the upper lobe.[4,8] Lone *et al* confirmed that three fourth of his patients had upper lobe involvement which is similar to our study (75%).[12] We also saw higher propensity for right side (28 cases; 58.3%) which is comparable to other studies.[2,14] Dyspnoea was the most common presenting complaint with chest pain in 42 (87.5%) and cough in 28 (58.3%) all being higher than reported by Zhu *et al*.[2,12] CXR is the most frequently available investigation on presentation whereas CT scan remains the most valuable investigation.[17,18] CT scan not only aids in defining GB and underlying lung but also helps to differentiate bullae from pneumothorax by the presence of double bubble sign.[17,18] Bullectomy has been advocated as supreme procedure and traditionally open thoracotomy was performed.[2,3,16,17,19]

Over the years it was established that open procedure resulted in worsening of reserves and excessive tissue removal, therefore video assisted thoracoscopic surgery (VATS) was preferred.[2,17,19,20] VATS was preferred which avoids adverse effects of open procedure. However, it is also associated with complication rates of 50–65% and mortality rates 2.5–22%.[3,4,15,16] Intracavitary tube drainage procedures are very beneficial for those with limited pulmonary reserve and unable to undergo a bullectomy under general anaesthesia. It ensures no removal of lung tissue and immediate symptomatic relief with little morbidity and low mortality.[11] In our study immediate symptomatic relief was seen in 43 (89.6%) patients which is supported by literature.[5,11,15,20] Success of both procedures in terms of symptom improvement was noted (p=0.008). Chest pain and cough significantly improved in patients who had resolution of bullae (p=0.012, 0.002, respectively).

Lone *et al* saw PRDS of 2.25±0.59 which decreased following interventions to 1.30±0.9.[12] We had higher values of PRDS with mean of 3.91±0.49. Following ITDP the mean of dyspnoea score reduced to 2±0.71. Both procedures were affective in upstaging the dyspnoea score by 2 values in 27 (56.25%) cases. PODS improvement was seen from grade IV to II (24/4; p=0.004) and grade V to grade III (3/7; p=0.034). Vigneswaran *et al* found improvement of functional class by one score in 54.5%.[14] Furthermore, we saw significant improvement in 6MWT (p<0.001) but literature lacks studies for comparison. Radiographic resolution was witnessed in 41 (87.5%) patients which is lower compared to previous reports of up to 97% which can be attributed to selection of advanced stages of BLD in our study.[5] Various other variables can identify success of procedures in BLD which include SaO2, PaO2 and PaCO2.[11,12,16] Procedures performed in our study improved SaO2 by 6.08±1.36 % (p<0.001). In literature no significant work correlates the role of surgery in improvement of SaO2 other than an Indian study that failed to report any improvement.[12] Post ITDP, PaO2 improved significantly by 40.6±4.82 mmHg (p=0.009) at three months. Schipper et al reported pre-operative PaO2 levels of 65±12 mmHg followed with six-month value of 73±13mmHg.[15] The author also compared PaCO2 levels which prior to ITDP were 43±9mmHg and at six months was 41mmHg.[15] We saw a decrease of PaCO2 in our patients by 13.22±3.62mmHg at three months which was statistically insignificant (p=0.7) but was higher than reported in literature. Other authors also studied the effect of ITDP in BLD in terms of PaCO2, nevertheless they failed to report any post ITDP values or significant changes.[11,12,16] During our study we saw more than 75% patients having bullae larger than 10cm with median size of 12.9 cm which was similar to another study.[2] Although reduction in size was not statistically significant (p-0.291) a mean reduction of 9.33±5.13 cm was noted. Many authors studied the changes in lung mechanics in terms of FEV1 and FVC.[1,2,12-15] FEV1 is by far the most important factor to define positive outcomes in the PoP.[3,12] In our patients, FEV1 improved by 0.57±0.07 L and FVC by 0.73±0.516 L (p<0.001). In a Chinese study following VATS procedure for GB, FEV1 improved by 0.4 L.[2] In another prospective study, post ITDP values of FEV1 and FVC were 1.47±0.56 L and 2.42±0.71 L, respectively.[12] Lone *et al* reported improvement of both FEV1 and FVC till six months (values were 2.18±0.49 L and 3.15±0.65 L.[12] It is evident on comparison that in earlier cases VATS procedure and later intracavitatory procedures provide near similar results. Greenberg *et al* and another author reported improvement of lung function tests by 83% following Monaldi Procedure and Bullectomy.[5,14]

In our study all patients were discharged and no mortality was reported as compared to other authors reporting a mortality rate of 4–26% for ITDP.[3,5,11,16] Procedures for GB and BLD via VATS or open surgery have high complication rates of more than 60% with prolonged air leak reported as the most common complication.[2,3] A great deal of work has been done with

regard to bullectomy and their complications but there is scarcity of literature on complications of ITDP. Shah *et al* reported morbidity of 15.5%, with wound infection in 5.18% and pneumonia in 6.89% patients.[11] We report complications in 25 (52.1%) patients all of which were manageable and non-fatal. Catheter block remained most common complication followed by subcutaneous emphysema**.** Accidental removal of catheter observed in seven (28%) cases was found to be associated with presence of wound infection (3/7; p=0.009). Recurrence of bulla following ITDP such as Monaldi procedure can be upto 21%.[20] Five (12.1%) of our patients presented with recurrence of bullae. Up to 51.2% of our patients had resolution of bullae by two months with sustained symptomatic relief. Studies including ITDP and Bullectomy report a duration of stay ranging 12–15 days.[11,15] In our study duration of stay was 4.20±0.92 days.

CONCLUSION

Intracavitary tube drainage procedures show a real potential of bringing immediate symptomatic relief to patients who have very poor pulmonary reserve to undergo general anaesthesia and bullectomy. It restores their functional status to carry-out daily routine without getting incapacitated. Improvement is seen in multiple variables like PaO2, PCO2, bed side SaO2 and 6MWT along with FEV1 and FVC. The size of the bulla also regresses allowing the underlying compressed lung to expand. This study is a single center study with a small sample size. Furthermore, study was carried out for a specific duration of time. Therefore, results shown may not depict the full advantage of ITDP. A larger sample size with a prolonged follow up has the potential to further improve results of ITDP in chronically debilitated patients with giant bullous disease of lung.

Acknowledgment: None

AUTHORS' CONTRIBUTION

MSL: Study concept, study design, data collection. MM: Drafting, data interpretation, literature search, data collection. TA: Final approval of version to be published, revising it critically for important intellectual concepts. AA: Data analysis, literature search, reviews of draft.

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Submitted: May 5, 2022	*Revised: August 13, 2022*	*Accepted: August 24, 2022*

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