Original Article

Quality-of-life impact of diaphragm plication in patients with diaphragmatic paralysis: A retrospective study

Jasmin Valenti[1], Khea Tan[2], Kelly Rubino[2], Ziad Hanhan[3], Dennis Vega[4], Matthew Kaufman[2,5], Thomas Bauer[2,5]

[1]School of Medicine, Hackensack Meridian School of Medicine, Nutley, [2]Department of Surgery, Jersey Shore University Medical Center, Neptune, [3]Riverview Medical Center, Red Bank, [4]Department of Surgery, Hackensack Meridian Ocean University Medical Center, Brick, [5]Department of Surgery, Hackensack Meridian School of Medicine, Nutley, NJ, USA

Address for correspondence:
Jasmin Valenti, 608, 2nd Street Apartment 6, Hoboken, NJ, USA.
E-mail: jasmin.valenti@hmhn.org

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Abstract:

OBJECTIVES: While the overall incidence and prevalence of diaphragmatic paralysis are unknown due to a wide variety of underlying causes, symptomatic patients experience a marked decline in their quality of life. The goal of this study was to measure the impact of diaphragm plication surgery on the quality of life in patients who were diagnosed with diaphragmatic paralysis.

METHODS: A retrospective review of the medical records of 46 patients who underwent diaphragmatic plication surgery was performed. The review included patients who experienced unilateral and bilateral diaphragmatic paralysis. Patients who underwent repeat diaphragm plication surgery were also included in the study. Patients from the retrospective cohort were then contacted by telephone to answer the Dyspnea-12 (D-12) questionnaire. Patients were asked to recall the severity of their symptoms and quality of life preplication, 1-month postplication, and 6-month postplication. Severity of symptoms was ranked as either none, mild, moderate, or severe. Values were then assigned to each rank as follows: none = 0, mild = 1, moderate = 2, and severe = 3. Relative change and statistical significance were calculated with preplication measurements used as the baseline. Scores between preplication versus 1-month postplication and 6-month postplication were then compared by Student's paired *t*-test. All tests were two-sided and statistical significance was set at $P < 0.05$.

RESULTS: Forty-six patients were included in the study, from which 21 answered the D-12 questionnaire. Average scores from each component of the D-12 questionnaire showed improvement in the severity of symptoms from preplication to 1-month postplication. The latter period was then followed by continued improvement in all areas when symptoms 6-month postplication were assessed.

CONCLUSION: In patients with diaphragmatic paralysis, diaphragm plication was effective in reducing patients' symptoms while improving overall quality of life.

Keywords:

Diaphragm plication, diaphragmatic paralysis, video-assisted thoracoscopy plication

Diaphragmatic paralysis is related to injury of the phrenic nerve and its etiology can be either congenital or acquired. The most common causes of diaphragmatic paralysis in adults are idiopathic, related to interscalene block on the phrenic nerve, or injury to the phrenic nerve during cardiac surgery.[1] This is seen more frequently in males, with the left hemidiaphragm more commonly affected.[2-4] While the overall incidence and prevalence of diaphragmatic paralysis are variable due to the wide variety of underlying causes, symptomatic patients experience a marked decline in their quality of life. Patients commonly report dyspnea on exertion, especially with intense exercise and exacerbated when lying supine.[5] Diagnosis is often

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made with chest X-ray and confirmed by fluoroscopy.[2] Conservative management with diaphragmatic physical therapy can be effective in patients who experience spontaneous partial recovery, however, if symptoms fail to improve, surgical treatment is often pursued. The optimal time for intervention varies, with some authors waiting 3–12 months, while others suggest waiting up to 25 months for spontaneous recovery.[2,6,7]

In patients who have the potential for phrenic nerve reinnervation, reconstruction is the preferred method.[8] This requires unilateral paralysis with intact voluntary motor neurons on electrodiagnostic testing. Follow-up studies have demonstrated functional and symptomatic improvement after phrenic nerve reconstruction.[9-11] When this is not feasible, however, due to patient comorbidities, mechanism or acuity of injury, or bilateral involvement, diaphragm plication becomes an appealing treatment solution.[11] Prior studies have primarily used objective measurements to determine the effectiveness of diaphragm plication, pulmonary function tests (PFTs) being the most common measurement of choice.[5,12-16] The utilization of subjective scales has varied greatly between studies and has primarily focused on patients reflecting on the severity of their disease before and after surgery.[2,14,17,18] One study attempted to assess this by tracking expressed satisfaction of improvement postplication.[12]

In addition, very few studies have attempted to assess the quality of life changes and the perceived impact of the disease. Only one study used a tool to measure physical and emotional impact using the St. George's Respiratory Questionnaire (SGRQ).[19] However, the questionnaire contains 50 items, making it questionable whether this survey is appropriate to use for recall and in clinical practice.[20] Currently, no standard quality-of-life assessment has been established for patients who underwent diaphragm plication. However, establishing such a scale may be of benefit, since alterations in quality of life are the reason, many patients seek treatment, and how success is measured from a patient perspective.[21] In addition, postoperative quality-of-life measurements can serve as a guide to identify those who need additional interventions.[22] Thus, the goal of this study was to measure the impact of diaphragm plication surgery on quality of life with a simpler scale that can be more easily utilized.

Methods

Study population

A retrospective review of the medical records of 46 patients who underwent diaphragmatic plication surgery between July 2015 and June 2021 was performed. Data collected from the retrospective review were sex, age, etiology of diaphragmatic paralysis, comorbidities, utilized surgical methods, and complications. The review included patients who experienced unilateral and bilateral diaphragmatic paralysis, as well as those who underwent repeat diaphragm plication surgery. Patients presented with significant dyspnea and underwent preoperative evaluation that included chest radiography. They subsequently underwent diaphragmatic plication either through thoracotomy, laparoscopy, thoracoscopy, or robotic-assisted thoracoscopy. Patients from the retrospective cohort were then contacted by telephone to answer the Dyspnea-12 (D-12) questionnaire.

All patients who responded had their answers recorded on a de-identified document. Patient responses were tracked by assigning individual patients a subject number. This study was approved by the Institutional Board of Review. All mandated orders were followed.

Dyspnea-12

The D-12 questionnaire is a valid and reliable patient-reported survey to evaluate the effects of dyspnea on quality of life.[23] It contains 12 statements related to respiratory function and can be used to evaluate dyspnea at baseline and how it changes over time [Figure 1]. Each statement inquires about a specific component related to the patient's chief complaint of dyspnea that can be rated on a scale of none, mild, moderate, to severe. Patients were asked to rate each statement individually as they answered the questionnaire.

The D-12 questionnaire was selected as it is easy to complete and does not factor in a patient's activity levels, age, or sex, which are components of other scoring systems frequently used to assess health impairments in respiratory diseases. In addition, the D-12 questionnaire has been proven to be a reliable measure of breathlessness in other respiratory-related illnesses such as interstitial lung disease, chronic obstructive

The questionnaire is designed to help us learn more about how your breathing is troubling you. Please read each item and then tick the box that best matches our breathing these days. If you do not experience an item tick the "none" box. Please respond to all items.

	None	Mild	Moderate	Severe
My breath does not go in all the way				
My breathing requires more work				
I feel short of breath				
I have difficulty catching my breath				
I cannot get enough air				
My breathing is uncomfortable				
My breathing is exhausting				
My breathing makes me feel depressed				
My breathing makes me feel miserable				
My breathing is distressing				
My breathing makes me agitated				
My breathing is irritating				

Figure 1: Dyspnea-12 Questionnaire

pulmonary disease, asthma, and heart failure, making it a reasonable measurement for symptomatic diaphragmatic paralysis.[23,24]

Data analysis

Patients were asked to recall the severity of their symptoms and quality of life based on the criteria of the D-12 questionnaire. It should be noted that three patients in the sample population required multiple plications. One patient had two right-sided plications. One patient had a right-sided plication and a left-sided plication. One patient had three right-sided plications. These patients were asked to answer a D-12 questionnaire separately for each surgery they underwent. As a result, while 46 patients were included in the study, there were 50 separate surgeries that were analyzed. Results of all the survey sets were included in the final analysis.

Data from the time periods of preplication, 1-month postplication, and 6-month postplication were collected. Severity of symptoms was ranked as either none, mild, moderate, or severe. Values were then assigned to each rank as follows: none = 0, mild = 1, moderate = 2, and severe = 3.

Mean values of each D-12 item and the total D-12 score were calculated for comparative analysis of the selected time periods. Mean values were calculated by dividing the compiled score of an individual D-12 item or the total score of the entire questionnaire by the number of completed survey sets. A completed survey set is classified as completed D-12 questionnaires for the preplication, 1-month postplication, and 6-month postplication time periods for a single surgery.

Relative change and statistical significance were calculated with preplication measurements used as the baseline [Table 1]. Scores between preplication versus 1-month postplication and 6-month postplication were then compared by Student's paired *t*-test. All tests were two-sided and statistical significance was set at $P < 0.05$.

Results

The retrospective chart review revealed that all 46 patients were symptomatic before diaphragm plication. Thirty-two subjects were male and 14 subjects were female. Patients were anywhere from 32 to 89 years old when they underwent plication. The etiologic causes of diaphragmatic paralysis varied. In many cases, the cause was not specified (n = 15). However, the most common documented cause of paralysis was cardiac surgery (n = 11), followed by intrathoracic surgery (n = 4). Patient comorbidities were also analyzed, the two most common being hypertension (n = 23) and hyperlipidemia (n = 17).

Of the 46 patients that were included in the study, 21 answered the D-12 questionnaire. Since three patients needed repeat surgeries, there were a total of 50 surgeries performed. Of the 50 surgeries, 24 have an associated D-12 questionnaire survey set. The patient who underwent three right-sided plications only provided

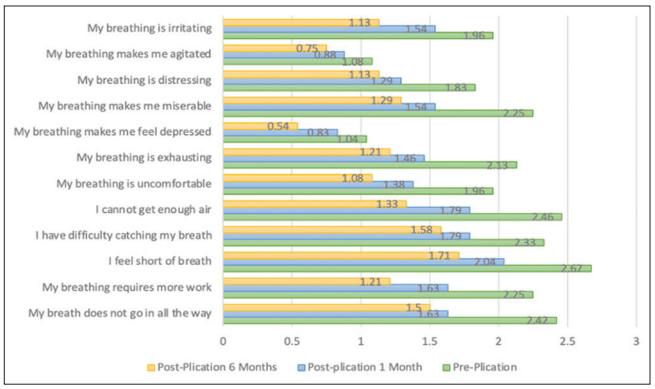

Chart 1: Average Dyspnea-12 questionnaire scores pre- and postplication

Table 1: Comparative Analysis of Dyspnea-12 Questionnaire Averages

Dyspnea-12 Score Item	Pre-Plication	Post-plication 1 Month	Post-Plication 6 Months
1. My breath does not go in all the way	2.42	1.63 (-33%) *	1.5 (-38%) *
2. My breathing requires more work	2.25	1.63 (-28%) *	1.21 (-46%) *
3. I feel short of breath	2.67	2.04 (-24%) *	1.71 (-36%) *
4. I have difficulty catching my breath	2.33	1.79 (-23%)	1.58 (-32%) *
5. I cannot get enough air	2.46	1.79 (-27%) *	1.33 (-46%) *
6. My breathing is uncomfortable	1.96	1.38 (-30%)	1.08 (-45%) *
7. My breathing is exhausting	2.13	1.46 (-31%) *	1.21 (-43%) *
8. My breathing makes me feel depressed	1.04	0.83 (-20%)	0.54 (-48%)
9. My breathing makes me miserable	2.25	1.54 (-32%) *	1.29 (-43%) *
10. My breathing is distressing	1.83	1.29 (-30%)	1.13 (-38%)
11. My breathing makes me agitated	1.08	0.88 (-19%)	0.75 (-31%)
12. My breathing is irritating	1.96	1.54 (-21%)	1.13 (-42%) *
Total Average Dyspnea Score	24.375	17.71 (-27%) *	14.46 (-41%) *

*Represents a P<0.05 when compared to pre-plication value

survey responses for the first and last surgery. Average scores from each component of the D-12 questionnaire showed improvement in the severity of symptoms from preplication to 1-month postplication. The latter period was then followed by continued improvement in all areas when symptoms 6-month postplication were assessed [Chart 1]. The statistical significance between average scores of individual D-12 items when compared to their respective preplication averages varied. A summary of these findings is organized in Table 1.

There was a significant difference between the overall preplication scores (standard deviation [SD] =9.03; mean = 24.375) and the overall 1-month postplication scores (SD = 10.63, mean = 17.71); $t = 0.025$, $P < 0.05$. Significant differences expanded further with the comparison between the overall preplication scores and the overall 6-month postplication scores (SD = 11.41, mean = 14.46); $t = 0.002$, $P < 0.05$.

Most surgeries performed in this study were video-assisted thoracoscopy (VATS) (60%), due to surgeon preference. The next most performed operation was thoracotomy (19%), followed by robotic-assisted VATS (18%), mini thoracotomy (4%), and laparoscopy (2%). In all cases, thoracotomy was performed when adhesions impaired video-assisted attempts at dissection. Operative time tended to be longer, as well as length of hospital stay for those who underwent thoracotomy [Table 2].

Table 2: Comparison of Surgical Approaches

Surgical approach	Number of surgeries	Number of surgeries with complications	Average length of hospitalization (days)
Laparoscopic	2	0	2
Mini thoracotomy	2	0	5
Thoracotomy	9	2	2.89
VATS	30	7	2.37
Robotic-assisted VATS	8	4	3.63

The most common postoperative complication [Table 3] among all the surgical approaches was urinary retention requiring catheterization. Thoracotomy was also associated with atrial fibrillation, return to the operating room for control of hemorrhage, blood transfusion, atrial fibrillation, and pneumothorax requiring chest tube. Video-assisted thoracotomy was associated with re-intubation, prolonged ventilator time, pleural effusions, atrial fibrillation, sepsis, and new dialysis requirements. Among those who underwent robotic-assisted VATS, two patients had fatal cardiac arrests. One patient had an advanced directive of do-not-resuscitate while the other fatality was due to a failure to achieve return of spontaneous circulation.

Among the patients who responded to the questionnaire, four cases were complicated by urinary retention requiring straight catheterization, one of which was also complicated by return to the operating room for control of bleeding. Average length of hospital stay was 2 days, with the majority of patients remaining in the hospital for 1 day.

Discussion

In symptomatic patients, diaphragm plication can serve as an alternative for treating chronic diaphragmatic paralysis. Plication works by increasing the thoracic space and restoring the diaphragm to its original position, as well as relieving compression on the atelectatic lung and improving the respiratory function of the intercostal, perithoracic, and abdominal muscles.[24] This helps to improve symptoms of diaphragm paralysis, however, it has not been shown to be useful in ventilator weaning.[25] In a retrospective study by Simansky *et al*., only 1 of 4 adults were able to be weaned off the ventilator after plication.[14] In contrast, 37 of 41 patients reported significant improvement in dyspnea after plication in a report by Freeman *et al*.[17]

Table 3: Complications and Associated Surgical Approach

Complication	Thoracotomy	VATS	Robotic-assisted VATS	Total
Urinary retention requiring catheterization	1	6	2	9
Prolonged ventilator time		1		1
Pleural effusion		1		1
Reintubation		1	1	2
Atrial fibrillation	1	1		2
Sepsis		1		1
Dialysis		1		1
Acute Encephalopathy			1	1
Fatal cardiac arrest			2 *	2
Bleeding with return to OR	1			1
Need for transfusion	1			1
Pneumothorax with Chest Tube Placement	1		1	2

*One patient who underwent cardiac arrest was a DNR. The other patient who underwent cardiac arrest expired due to inability to achieve ROSC

Our article demonstrated that plication was effective in reducing patients' symptoms while improving overall quality of life. A statistically significant decrease of the items "my breath does not go in all the way," "my breathing requires more work," "I feel short of breath," "I cannot get enough air," "my breathing is exhausting," and "my breathing makes me miserable" within 1-month postplication suggests that the surgery is highly effective in treating dyspnea related to diaphragm paralysis. Items "I have difficulty catching my breath," "my breathing is uncomfortable," and "my breathing is irritating" did not show statistically significant improvement until 6 months postplication. This suggests that it likely takes an extended period for patients to recondition to activity postsurgery. Discomfort and irritation with breathing may not resolve completely until 6 months postprocedure due to the invasive nature of the surgery.

The findings found from 1-month postplication are consistent with the study conducted by Groth *et al*., which utilized the SGRQ. Since the questionnaire consists of 50 items, answers are divided into two parts. The first part is the symptoms score, which assesses the patient's perception of the frequency and severity of their symptoms. The second part includes an activity score, which assesses the impact of daily physical activities and an effect score, which assesses psychosocial dysfunction. The symptoms score, activity score, and effect score were all found to be significantly improved by 1-month postplication.[19]

These findings align with the items that were found to significantly improve from the D-12 questionnaire. Items, "my breath does not go in all the way," "breathing requires more work," "I feel short of breath," "I cannot get enough air," and "my breathing is exhausting," are like items found in the symptoms and activity section of the SGRQ. Meanwhile, the item "my breathing makes me miserable" addresses the psychosocial effect of diaphragmatic paralysis. Consistency among these studies indicates that the D-12 questionnaire is a suitable replacement for the SGRQ. Since the D-12 questionnaire is much shorter than the SGRQ, there is more promise in utilizing it for future studies and clinical practice, as shorter questionnaires have been found to significantly increase response rates among subjects.[26]

In addition, studies have found that improved PFT scores are associated with improved Medical Research Council (MRC) Dyspnea Scale scores, another subjective scale.[2,17] In a prospective study by Freeman *et al*., mean forced vital capacity (FVC), forced expiratory volume at 1 s (FEV_1), functional residual capacity, and total lung capacity all improved by 19%, 23%, 21%, and 19% ($P < 0.005$), respectively, when measured 6 months after surgery, as were mean MRC dyspnea scores ($P < 0.0001$). An additional, long-term retrospective series study by Celik *et al*. found that FVC and FEV_1 improved by 30.6% ($P < 0.001$) and 10.9% ($P < 0.001$), at late follow-up averaging 5.4 years after diaphragmatic plication.

While PFTs were not analyzed in this study, future studies could incorporate the usage of PFTs and the D-12 questionnaire to track improvement postplication. If an improvement is detected in both measurements and a correlation is established, it is possible that the D-12 questionnaire can be used as the main measurement to analyze for improvement in future practices. The potential of implementing this change lies in reducing further need and costs associated with additional PFT testing postoperatively.

Limitations

When utilizing the D-12 questionnaire, there was a varied interpretation of certain items. For example, when patients were asked to answer the item, "My breathing is distressing," many were confused on what to classify as distress. For this study's purposes, distress was described as fear of an acute crisis due to an inability to breathe. This reveals that while the D-12 questionnaire is a simple measurement tool, additional information could be added to certain items to unify interpretation among patients. There was also variance in the interpretation of the terms "mild," "moderate," and "severe." Cultural factors and personal experiences highly influenced the interpretation of these terms and how each individual stratified their symptom severity.

The greatest limitation of this study was its retrospective nature. Patients may not have been able to accurately recall their symptoms at the desired time points, especially if their surgery was several years ago. Recall bias may have altered some of the results, as patients may have not been able to recall details accurately. This is especially the case for patients who required multiple surgeries. As a result, patients may have answered more or less favorably based on their individual long-term outcomes, a factor that influences memory accuracy and significance.[27] For example, patients who felt the surgery was successful years afterward, may have provided responses that showed a greater improvement in their symptoms. In addition, the development of comorbidities after surgery may have impacted the patients' perspective on the effectiveness of their surgery. For example, one individual patient reported a 30-pound weight gain since the time of his surgery, which had greatly impacted his ability to breathe. When asked to recall his symptoms 6-month postplication, the patient kept referencing the time point since after he gained weight, which was over a span of a few years. As a result, his questionnaire demonstrated a lesser improvement from his preplication scores.

Finally, due to the retrospective nature of the study, there was no standardized surgical approach to the patients. The etiology of each patients' diaphragmatic paralysis, prior thoracoabdominal surgeries, and surgeon preference all determined whether patients underwent a minimally invasive approach in the form of robotic VATS, thoracoscopy, or laparoscopy versus a thoracotomy or mini thoracotomy. In general, patients undergoing a minimally invasive approach tend to have decreased pain in the postoperative period and faster return to function. Patients who underwent a thoracotomy may have had increased pain in the postoperative period relatively and a higher likelihood of requiring rehabilitation facilities, contributing to increased time to recover their respiratory function. A minimally invasive approach was the preferred method with or without the robot. Thoracotomy and laparoscopy (for left diaphragm) were reserved for those patients with extensive adhesions that prevented a minimally invasive approach.

A more proper subgroup analysis based on surgical approach-thoracotomy, thoracoscopy, robotic-assisted thoracoscopy, and laparoscopy-may be of benefit regarding future studies. Thoracotomy should be especially investigated, as it is the most utilized surgical technique to address diaphragmatic paralysis overall, making it an ideal comparison group.[2] This would clarify whether a specific approach has short-term or long-term implications in the postoperative period. Subgroups can further be broken down into the outcomes seen in patients with unilateral versus bilateral paralysis and specific comorbidities to see if any factors limit the benefit of plication. Further analyzing these subgroups may inform surgical approaches and recommendations.

All patients in this study were simultaneously added to a prospective database for a future study. The prospective study will seek to minimize the limitations mentioned above.

Conclusion

This study demonstrates the improvement seen in the quality-of-life assessment in symptomatic patients with diaphragmatic paralysis. We are continuing this study with our prospective D-12 questionnaire to further evaluate patients with this disease process. Since all patients in this cohort were symptomatic, it is unclear if asymptomatic patients will achieve any improvement with plication. Currently, asymptomatic patients are not offered plication as an option.

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Conflicts of interest

There are no conflicts of interest.

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