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Case Presentation

Exercise Therapy for a Patient With Persistent Dyspnea After Combined Traumatic Diaphragmatic Rupture and Phrenic Nerve Injury

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Abstract

We present a case report of a patient with a history of diaphragmatic rupture who had persistent dyspnea for 9 months after primary surgical repair of a right diaphragmatic rupture caused by a car accident. A phrenic nerve conduction study was performed, which demonstrated a rare accompanying ipsilateral phrenic nerve injury with resultant hemidiaphragmatic paralysis. Aerobic exercise therapy for the purpose of improving endurance and dietary modification for weight reduction were prescribed and continued for 6 months. The exercise intensity was prescribed based on the percentage of maximum heart rate as confirmed by an exercise tolerance test. The duration of exercise was gradually increased. In this case, the long-persistent dyspnea was successfully alleviated via nonoperative management to the point that the patient could resume regular activities of daily living.

Introduction

Diaphragmatic paralysis due to diaphragmatic rupture or phrenic nerve injury is a common cause of dyspnea. In cases of diaphragmatic rupture, regardless of the generally positive prognosis after primary surgical repair, complete recovery is achieved in only 43.9% of cases [1]. Moreover, the prognosis for phrenic nerve injury is known to vary with the underlying causal disease or condition. The symptoms of phrenic nerve injury are transient and generally of no clinical significance when they occur after cardiac surgery [2]. However, some cases of hemidiaphragmatic paralysis with persistent respiratory compromise might require diaphragmatic surgery [3]. Reports on separate cases of traumatic diaphragmatic rupture and phrenic nerve injury have been published; however, to the best of our knowledge, this is the first reported case of combined diaphragmatic rupture and phrenic nerve injury. The patient experienced 9 months of continuous dyspnea after a motor vehicle accident. The dyspnea symptoms improved after a regimen of aerobic exercise, dietary modification, and weight reduction, which enabled the patient to resume daily living activities that had been seriously curtailed.

Case Presentation

A 52-year-old woman without any known underlying diseases, including hypertension, heart problems, or metabolic disease, underwent a thoracotomy and primary repair for a right diaphragmatic rupture, a fracture of the seventh rib, and a hemothorax sustained in a car accident. She was hemodynamically stable. Initially, the rupture was successfully repaired without postsurgical complications, and thus satisfactory progress was expected. The initial posteroanterior chest radiograph showed an elevated right diaphragm, and the diaphragmatic height index (DHI) [4] was 2.7 (Figure 1A). One month after surgery, she reported dyspnea. Her Medical Research Council (MRC) dyspnea score was 4. A subsequent contrast-enhanced chest computed tomographic scan revealed an elevated right diaphragm, and no malignant lesion or pulmonary embolism was found. Similarly, an echocardiogram revealed no specific abnormalities. However, posteroanterior chest radiography showed that the right hemidiaphragm was persistently elevated. At that time, the patient's DHI was 2.5. Furthermore, a pulmonary function test revealed a forced vital capacity (FVC) of 2.03 L (67% of the predicted normal value) and a forced

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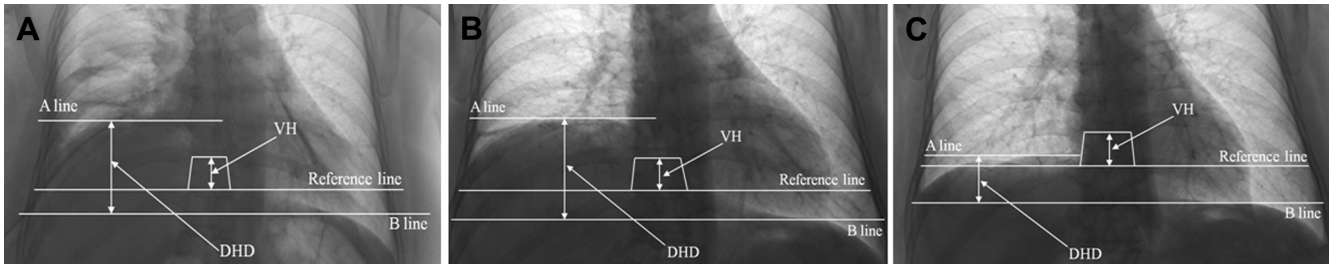

Figure 1. Posteroanterior chest radiograph. Diaphragmatic height index (DHI) = diaphragmatic height difference (DHD)/vertebral height (VH). Upon the initial chest radiograph (A), DHD = 60.30, VH = 22.3, and DHI = 2.7. At the time of rehabilitation evaluation (B), DHD = 49.55, VH = 20.81, and DHI = 2.4. After treatment (C), DHD = 32.45, VH = 21.52, and DHI = 1.5.

expiratory volume in 1 second (FEV1) of 1.55 L (66% of the predicted normal value; Table 1).

Nine months after surgery, the patient's MRC dyspnea score was, surprisingly, still 4, and she was referred to the Department of Rehabilitation Medicine for a phrenic nerve conduction study and further management. Posteroanterior chest radiography showed that the right hemidiaphragm was still elevated. The DHI was 2.4 (Figure 1B), the FVC was 1.99 L (66% of the predicted normal value), and the FEV1 was 1.55 L (66% of the predicted normal value; Table 1). The phrenic nerve conduction study was performed according to the method detailed by Markand et al [5] and revealed that a right compound muscle action potential (CMAP) was not evoked; needle electromyography of the diaphragm was not performed. Based on these results, the diagnosis of accompanying right diaphragmatic paralysis due to phrenic nerve injury was confirmed. Although an exercise tolerance test conducted at that time revealed a maximal oxygen consumption (V_{O_2} max) of 14.9 mL/kg/min, which was close to the normal value (18.7 $\pm$ 5.5) specified for the patient's age group [6], the metabolic equivalent of task score was 4.3 and indicated severely restricted daily living activities. The peak respiratory exchange ratio was 0.95 beats/min (Table 1), which might indicate a submaximal effort. She was able to perform self-care, such as lightly washing herself and dressing, but she found it difficult to climb the stairs and go shopping. Moreover, she was not able to participate in any recreational or leisure activities.

To alleviate the patient's symptoms and enable the resumption of daily living activities, aerobic exercise therapy and dietary modification were prescribed. Accordingly, the patient exercised 5-6 times per week on a treadmill preceded and followed by 5-minute warm-up and cool-down periods under a health professional's supervision at the local public health center. The exercise intensity was prescribed based on the percentage of maximum heart rate as confirmed by an exercise tolerance test. The level of intensity began at 60% to 85% of the maximal heart rate and increased by 5% every month. The exercise durations were tolerable and were gradually increased. The initial 10- to 15-minute exercise duration was, after the first 4 weeks, augmented to a minimum of 30 minutes (Table 2). The patient was instructed to stop the exercise if she experienced dizziness, dyspnea, chest pain, or lower extremity pain. The patient exhibited very good compliance, performing the exercise as scheduled

Table 1
Physiological changes before and after exercise therapy

	1 Month After Surgery	Before Exercise Therapy	6 Months After Exercise Therapy
MHR, beats/min		143	179
Peak RER		0.95	1.25
V_{O_2} max, mL/kg/min		14.9	28.6
MET		4.3	8.2
CMAP of right phrenic nerve		not evoked	not evoked
Pulmonary function test			
FEV1, L	1.55	1.55	1.77
FEV1, % predicted	66	66	76
FVC, L	2.03	1.99	2.36
FVC, % predicted	67	66	80
FEV1/FVC	0.76	0.78	0.75
FEV1/FVC, % predicted	101	104	100
DHI	2.5	2.4	1.5

MHR = maximum heart rate; RER = respiratory exchange ratio; V_{O_2} max = maximal oxygen consumption; MET = metabolic equivalent of task; CMAP = compound muscle action potential; FEV1 = forced expiratory volume in 1 second; % predicted = percentage of predicted normal value; FVC = forced vital capacity; DHI = diaphragmatic height index.

Table 2
Physiological changes before and after exercise therapy and exercise prescription

	1 Month After Surgery	Before Exercise Therapy	1 Month After Exercise Therapy	3 Months After Exercise Therapy	6 Months After Exercise Therapy
Body weight, kg	68	67.6	63.4	59.5	60.5
BMI, kg/m^2	29.4	29.2	27.4	25.8	26.3
MRC dyspnea scale	4	4	3	1	1
Exercise intensity, percentage of MHR			60%	70%	85%
Exercise duration, min			10-15	Minimum of 30	Minimum of 30

BMI = body mass index; MRC = Medical Research Council; MHR = maximum heart rate.

without exception. Over the course of outpatient follow-up at intervals of approximately 4 weeks, the patient's dyspnea status, exercise intensity and duration, and body weight were monitored.

Beginning 2 months after the patient started the exercise therapy program, the dyspnea upon exertion was somewhat alleviated. Six months after the commencement of exercise, her body weight was 60.5 kg, which was a 7.1-kg reduction from the original 67.6-kg measurement, and her MRC dyspnea score had improved to 1 (Table 2). A follow-up pulmonary function test revealed an FVC of 2.36 L (80% of the predicted normal value) and an FEV1 of 1.77 L (76% of the predicted normal value). The V_{O_2} max was 28.6, and the metabolic equivalent of task score was 8.2 (Table 1). The DHI, according to a follow-up posteroanterior chest radiography, had been reduced to 1.5 (Figure 1C), although the CMAP still could not be evoked. Because her dyspnea had been completely alleviated, the patient was judged to have recovered to a state wherein regular daily living activities could be resumed.

Discussion

In this case, unilateral diaphragmatic paralysis resulting from right phrenic nerve injury was discovered during a phrenic nerve conduction study performed on a patient who had reported 9 months of continuous dyspnea after surgery for traumatic diaphragmatic rupture. The patient's symptoms were improved by conservative management that included exercise, dietary modification, and weight reduction.

Because diaphragmatic paralysis caused by phrenic nerve injury after blunt trauma is symptomatically very similar to diaphragmatic rupture, the diagnosis is often delayed, particularly because of the statistical incentive for early doubt [7]. Confirmation of an elevated hemidiaphragm by chest radiography and computed tomography is effective for the diagnosis of phrenic nerve dysfunction; however, for a more accurate diagnosis, the sniff test, ultrasonography, and a phrenic nerve conduction study also should be performed. Of these tests, the nerve conduction study is the gold standard [8,9]. In the present case, the phrenic nerve injury was accompanied by a diaphragmatic rupture; however, as previously noted, an elevated hemidiaphragm was identified on chest radiography and computed tomography at the time of injury, and the initial diagnosis was only of diaphragmatic rupture, which delayed the diagnosis of the phrenic nerve injury. The phrenic nerve injury was diagnosed only later as a result of the phrenic nerve conduction study that was performed because the patient's dyspnea persisted for 9 months.

Traumatic diaphragmatic rupture is occasionally associated with high morbidity and high mortality, depending on the associated injury [10].

Reportedly, many cases of traumatic phrenic nerve injury require 6-12 months for full restoration of normal diaphragmatic function [11]. Approximately 50% of the cases of unilateral diaphragmatic paralysis are asymptomatic, and the prognosis is positive when there is no underlying lung disease; thus aggressive treatment is not necessary is such cases [12]. When respiratory compromise is obvious and related symptoms have not been resolved, they can be alleviated by diaphragmatic surgery [3]. In contrast, in asymptomatic cases, clinical observation is sufficient unless a malignancy of the mediastinum or infection is present [13]. For cases in which dyspnea continues, physical therapy and weight reduction have been advocated [14]; however, no examples of this remedial approach could be found in the literature. Similarly, the literature contained no cases of simultaneous diaphragmatic rupture and phrenic nerve injury, which was the situation of the patient reported herein. After having reported 9 months of continuous postoperative dyspnea, the patient's condition was completely alleviated. Her MRC dyspnea score improved from 4 to 1 as a result of a regimen based on progressive aerobic exercise, diet modification, and weight reduction.

The right DHI, as determined by the posteroanterior chest radiography performed 6 months after the commencement of exercise therapy, had been markedly reduced to 1.5 from 2.5 (1 month after surgery) and 2.4 (before the exercise therapy). However, this DHI was still significant, particularly in terms of the DHI cutoff point of 1.1 defined by Pornrattanamaneewong et al [4] for the diagnosis of right phrenic nerve dysfunction. Additionally, considering that the right CMAP was not evoked in a follow-up phrenic nerve conduction test, it seems that the unilateral diaphragmatic paralysis

sustained as a result of the phrenic nerve injury was still present even 6 months after the commencement of exercise therapy, by which time the dyspnea had begun to improve. In the case of this patient, a sniff test was not performed. However, a sniff test would have been helpful in assessing whether there was a restoration or an absence of diaphragmatic function. Both the FVC and FEV1 before the exercise therapy were reduced to values below normal. These reductions were considered to be a mild restrictive pattern that resulted from the diaphragmatic paralysis. Although no evidence related to chronic obstructive pulmonary disease was identified in an interview with the patient, her FEV1/FVC after exercise therapy was reduced to borderline. However, both the FEV1 and FVC were improved after exercise.

Conclusion

In cases of severe persistent dyspnea resulting from unilateral diaphragmatic paralysis caused by combined diaphragmatic rupture and phrenic nerve injury, individualized exercise including aerobic exercise and dietary modification can be very effective.

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Disclosure

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