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A case of herpes zoster-related phrenic nerve palsy: Effect of home-based pulmonary rehabilitation

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Abstract

Here, we report the case of a 62-year-old man with herpes zoster-related phrenic nerve palsy, which was evident in clinical, radiological, and electrodiagnostic studies. To aid clinical recovery and preserve pulmonary function, we designed a home-based pulmonary rehabilitation program consisting of air-stacking exercises using ambu-bagging, respiratory muscle training to improve the strength and endurance of inspiratory and expiratory muscles, and aerobic and resistance exercises of the upper and lower limbs to improve and prevent the decline of cardiorespiratory fitness. The patient was monitored, and the exercises were adjusted through regular outpatient follow-up. During the overall follow-up period of 26 months, pulmonary rehabilitation led to an increase in forced vital capacity from 2,020 to 3,160 mL, and improved the diaphragmatic height index from -4.46 to 0.28. Clinical symptoms also showed notable improvement, as the Borg Rating of Perceived Exertion scale score decreased from 17 to 7. We found that a home-based, self-performed pulmonary rehabilitation program could promote recovery from herpes zoster-related phrenic nerve palsy. Not only is this important for the rapid restoration of respiratory function and symptomatic relief, it can also protect patients from respiratory complications.

Keywords: Phrenic nerve palsy, pulmonary rehabilitation, case report

INTRODUCTION

Patients with diaphragmatic palsy commonly exhibit dyspnea on exertion and orthopnea.[1] The diaphragm is crucial to inspiration, and longstanding diaphragmatic palsy could lead to declined pulmonary function, as unilateral diaphragmatic palsy is associated with a substantial loss of oxygen pressure in arterial blood and decreased breathing capacity.[2] To assess the severity of diaphragmatic palsy, Pornrattanamaneewong *et al*. (2017)[3] suggested utilizing the diaphragmatic height index (DHI), which is negative when the left diaphragm is higher than the right. A DHI of < 0.2 was suggested as the cutoff value for diagnosing left diaphragmatic palsy.

Herpes zoster is characterized by varicella-zoster virus reactivation in the dorsal root ganglia. The virus commonly affects the sensory nerves but could also involve motor nerves.[4] To our knowledge, herpes zoster-related hemi-diaphragmatic palsy is rare, and pulmonary rehabilitation for the condition has not been discussed. In this case, a patient with postherpetic phrenic nerve palsy (PNP) underwent a 26-month home-based pulmonary rehabilitation, which led to clinically and radiologically improved diaphragmatic function without pulmonary complications.

CASE REPORT

A 62-year-old man with a history of hypertension, diabetes, cerebrovascular accident, pulmonary tuberculosis, chronic obstructive pulmonary disease (COPD), and retinal disorder, who had been diagnosed with herpes zoster virus at a local hospital, came to our emergency room on July 9, 2020, for shooting pain in the left occipital region and posterior neck region corresponding to C3 dermatome.

During the initial assessment, chest radiography showed that the left diaphragm was approximately 5 cm higher than the right, with a DHI of -2.52 (Figure 1A).

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Two months after the first diagnosis of herpes zoster virus, the patient complained of progressing exertional dyspnea, with extreme dyspnea after five steps of stair climbing and a Borg Rating of Perceived Exertion scale (Borg RPE scale) of 17. On follow-up chest radiography, the left diaphragmatic palsy had worsened (DHI, -3.77; Figure 1B).

The patient was referred to the rehabilitation clinic for pulmonary rehabilitation. On nerve conduction study, the compound muscle action potential (CMAP) of the left phrenic nerve was not obtained using a surface electrode. Considering this result, the exertional dyspnea, and aggravated DHI, we concluded that the patient had left PNP, which had led to left diaphragmatic palsy.

Although electrodiagnostic studies showed no response in CMAP, as the phrenic nerve conduction study used surface electrodes, the subthreshold movement may have been misinterpreted as no response. Furthermore, it was previously described that baseline CMAP does not necessarily correlate with functional recovery[5], and studies indicate that PNP could be alleviated through pulmonary rehabilitation.[6] Therefore, pulmonary rehabilitation program was conceptualized to maximize rehabilitation potential and prevent pulmonary complications. Because the patient could not make frequent long-term visits to the center, home-based rehabilitation was initiated. Self-exercises were educated at the rehabilitation center until the patient could execute each exercise independently, and he was monitored at the outpatient clinic every 3 months.

First, the patient started an air-stacking exercise using ambu-bagging for three sessions daily to prevent atelectasis and maintain lung volume. Maximal inhalation was initiated, and additional air was infused via 3 ambu-bagging without exhalation. After an additional air infusion, the patient held his breath for >5 s before exhalation. This procedure was repeated 20 times per session. Moreover, an incentive spirometer was used to improve inspiratory muscle strength and endurance, prevent atelectasis, and encourage diaphragmatic movements.[7] The inspiration exercises using the spirometer consisted of inspiration lasting 5 s, with 10 repetitions per session, for 3 sessions daily.

Second, aerobic exercises, such as the treadmill, indoor cycling, and stair climbing, were performed for >150 min weekly. Intensity of at least a 'moderate' degree was recommended, in which the patient could have a conversation but could not sing a song.

Third, the patient was prescribed strengthening exercises for both upper and lower extremities. The strengthening exercises were recommended to be carried out for > 60 min weekly. For each movement, 3 sets of 8-12 repetitions per set were carried out and for each session at least one movement from upper and one from lower extremities were carried out. (Figure 2)

The aerobic and strengthening exercises were based on the 'outpatient self-exercise program' at our institute. The upper extremity strengthening exercises mainly targeted shoulder flexion, abduction, extension, elbow flexion and extension. The lower extremity strengthening exercises focused on hip flexion, abduction, extension,

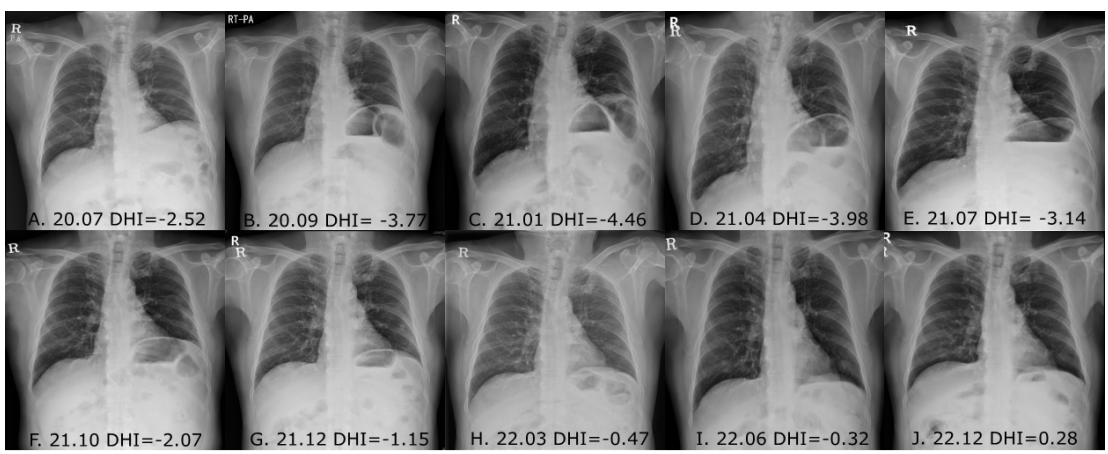

Figure 1. Serial follow-up chest radiographs showing changes in DHI. (DHI=DHD/VH[3]). The year-month and DHI at each time point are shown in the figure panels. (A–I) The left diaphragm is higher than the right diaphragm. (J) The right diaphragm is higher than the left diaphragm. Normal DHI is DHI > 0.2.[3]
* DHI: diaphragmatic height index

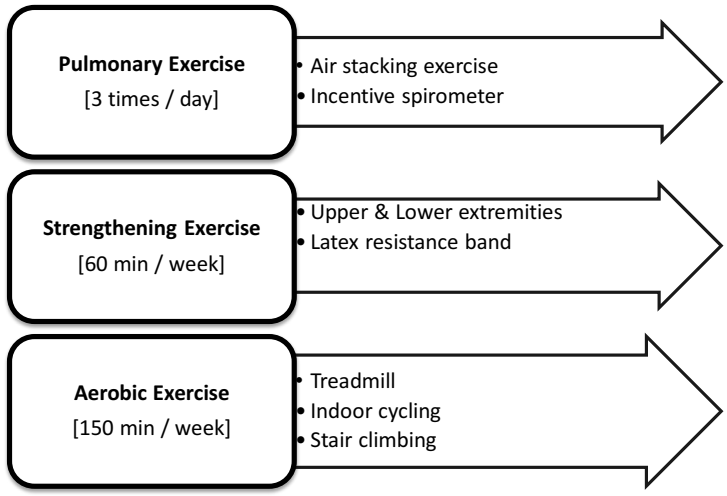

Figure 2. Summary of recommended exercise regimen consisting of pulmonary, strengthening and aerobic exercises

knee flexion and extension. Latex resistance bands were used for the strengthening exercises where the patient started training with red latex resistance band with 1.7 kilogram of resistance at 100% elongation and increased to green latex band with 2.1 kilogram of resistance at 100% elongation upon need. Exercise was considered successful when the patient performed two or more pulmonary exercises daily, >110 min of aerobic exercises weekly, and >40 min of strengthening exercises weekly. The patient was monitored and encouraged at each outpatient visit and via self-report, it was concluded that the patient successfully performed all the required weekly exercises during the follow-up period.

For objective assessment, a pulmonary function test was performed, and during the follow-up period of 26 months, forced vital capacity (FVC) increased from 2020 to 3160 mL, and DHI changed from -4.46 to 0.28. Initially, the left diaphragm was higher than the right; however, by the end of the follow-up, the right was higher than the left (Figures 1 and 3).

By the end of the treatment, considering the patients' subjective symptoms, the patient could climb 10 flights of stairs without stopping and

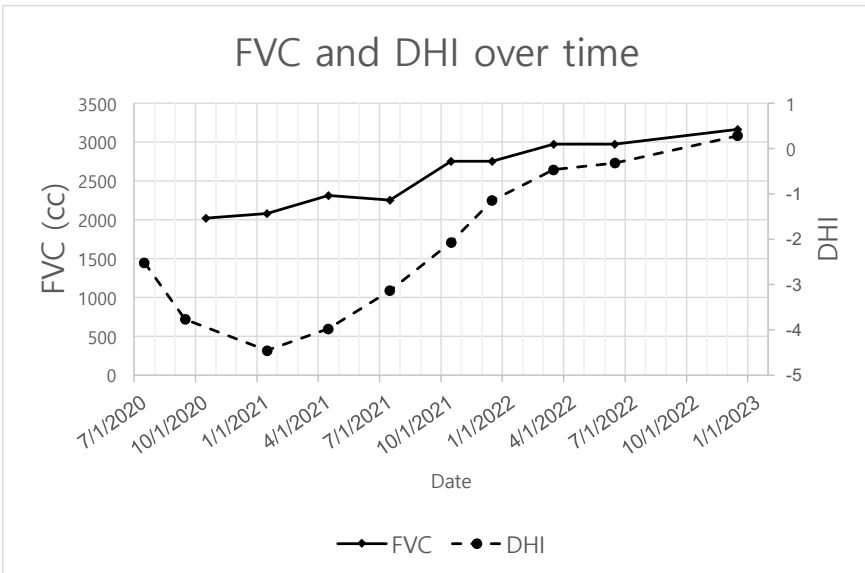

Figure 3. FVC changes are shown in the solid black line. DHI changes are shown in the dashed black line.
*FVC: forced vital capacity; DHI: diaphragmatic height index

did not complain of dyspnea during any daily life activity. The patient reported an improvement in the Borg RPE score from 17 to 7.

DISCUSSION

Oike *et al*.[8] reported that of 24 cases of herpes zoster-related PNP, only six showed alleviation of diaphragmatic palsy. Even in asymptomatic patients, decreased inspiratory muscle power leads to declined pulmonary function and furthermore, it has been associated with lower-segment atelectasis, which can increase the risk of pneumonia due to secretory stasis.[9]

Therefore, in patients with hemi-diaphragmatic palsy, rehabilitation is imperative to relieve symptoms and even in asymptomatic patients, rehabilitation is required to restore breathing capacity and prevent complications. Due to poor prognosis of herpes zoster-related PNP, the importance of rehabilitation is even more highlighted in these patients.

Herpes zoster infection of the motor nerves causes nerve axonopathy, characterized by demyelination, axon degeneration, and lymphocyte infiltration.[10] A few studies have shown that exercise increases neurite outgrowth, induces neurotrophin release, and increases synaptic conduction velocity, which promotes neuronal activation and regeneration.[11-13]

To alleviate herpes zoster-related PNP, we utilized home-based pulmonary rehabilitation comprising generalized aerobic and strengthening exercises and specific exercises for the respiratory muscles and chest wall. After receiving educational sessions on the above exercises at the hospital, the patient performed self-exercises at home. Although the patient had a history of pulmonary pathologies, such as old tuberculosis and mild COPD, home-based rehabilitation could alleviate PNP symptoms and signs. Furthermore, the patient did not experience pulmonary complications, such as pneumonia, during follow-up.

In this patient, DHI was aggravated for the first 6 months after herpes zoster infection. Recovery after peripheral nerve damage occurs via collateral sprouting and axon regeneration, and 3–6 months of recovery are required.[14] In this case, we can assume that for the first 6 months, phrenic nerve axonopathy occurred, and despite some neural recovery, nerve damage was dominant, leading to worsening DHI. However, the DHI normalized 29 months after symptom onset and 26 months after rehabilitation was initiated. Therefore, we believe continuous pulmonary rehabilitation and monitoring for over 2 years are vital for such patients, as the DHI may not improve immediately after treatment initiation.

In contrast, pulmonary capacity showed a linear increase throughout treatment (Figure 3), and the patient reported similar improvements in clinical symptoms. This may be because the diaphragm is not the only contributor to breathing, and FVC could increase as other accessory inspiratory muscles are strengthened. With the concurrent enhancement of physical fitness via aerobic and strengthening exercises, clinical symptoms may improve before DHI changes. Therefore, clinical symptoms or FVC could serve as early indicators of functional recovery in PNP because radiological recovery may be slower.

Unfortunately, as this was a retrospective study, no information on respiratory muscle strength and functional assessments were reported as these were not readily assessed at the outpatient clinic. In the future, in similar cases, respiratory muscle strength measurements such as maximal inspiratory pressure or maximal expiratory pressure could be assessed. Also for functional assessments, Cybex knee extension could be used to assess the muscular strength of lower extremities and inbody could be used to measure body compositions such as muscle mass and fat contents to evaluate the effect of aerobic and strengthening exercises. Patient compliance is important because the rehabilitation method above is home-based and can be encouraged via education and regular check-up at the outpatient clinic.

The importance of rehabilitation for various PNP types has been highlighted.[6] However, there is no consensus on the optimal rehabilitation method. Whether the rehabilitation is conducted in the inpatient or outpatient clinic, we believe that pulmonary, aerobic, and strengthening rehabilitation exercises are crucial for preventing additional complications and improving clinical and radiologic features of PNP patients. We believe the positive effect of home-based rehabilitation on herpes zoster-related PNP provides a good reference for standard PNP management.

DISCLOSURE

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Conflicts of interest: None

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