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Elastic Band Training Improves Adiposity and Physical Performance in Adults with Prader-Willi Syndrome: A Pilot Study

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OBJECTIVE: The aim of this study was to investigate the changes in the regional fat distribution of body composition and the physical performance of adults with Prader-Willi syndrome (PWS) after progressive elastic band (EB) training.

METHODS: Participants were trained three times per week, performing three sets of 15-20 repetitions for each of the exercises with EB for 24 weeks. Changes in body composition were measured using dual-energy X-ray absorptiometry. Physical performance was assessed by determination of hand grip strength, the timed up-and-go test, the 30-second chair stand test, the 2-minute step test, and the Berg Balance Scale.

RESULTS: Six participants were enrolled in this pilot study. Following 24 weeks of progressive EB resistance exercise training, significant reductions were observed in body weight (p = 0.028), body mass index (BMI) (p = 0.043) and regional extremity fat mass and percentage (p < 0.05). Regarding physical performance, significant improvements were observed after 8 weeks of training in the timed up-and-go test (p = 0.027), the 30-second chair stand test (p = 0.028), and the 2-minute step test (p = 0.028). Balance function improved after 16 weeks of training (p = 0.027).

CONCLUSION: Progressive EB training for 24 weeks could provide an additional simple and safe training, with improvements in body weight, BMI, regional fat mass and physical performance observed in individuals with PWS. (Tw J Phys Med Rehabil 2019; 47(2): 133 - 143)

Key Words: body composition; exercise; Prader-Willi syndrome

INTRODUCTION

Prader-Willi syndrome (PWS) is a multisystem genetic disorder caused by a loss of paternally inherited expression on a region of chromosome 15q11-13, with an
estimated prevalence of 1 in 10,000-30,000 births.\[^1\] It is the most common genetically caused obesity disorder, mainly due to hypothalamic dysfunction and growth hormone (GH) deficiency, which impairs satiety regulation, increases adipose tissue while decreasing lean mass, and consequently leads to morbid obesity if not strictly controlled. It presents as a unique genetic model associated with cognitive impairment, hypotonia, hyperphagia, and sarcopenia,\[^2\] different from the characteristics of subjects with simple obesity,\[^3\] and predisposing them to higher energy intake, reduced physical activity and motor skills, functional limitations, and disability.\[^4-6\]

Abnormal body composition with higher adiposity and lower lean mass\[^5,6\] results in lower muscle strength,\[^7\] cardiopulmonary fitness,\[^8\] and total energy expenditure.\[^9\] Individuals with PWS showed lower levels of physical activity than did age-matched peers.\[^4\] Obesity complications, such as type 2 diabetes mellitus, obstructive sleep apnea and pulmonary hypertension, are common, as are higher risks of morbidity and mortality.\[^1\] Two systematic reviews\[^6,10\] showed that in adults with PWS, exercise-training interventions of aerobic\[^11\] or resistance\[^12-14\] exercises or a combination of both\[^15-17\] were safe and effective as means to improve cardiopulmonary fitness,\[^11\] fat mass\[^11,17\] and muscle strength\[^11,13\] and were suggested to be incorporated as a coadjuvant therapy to caloric diet restriction and GH therapy. However, the majority of the studies were mainly designed for children or adolescents. Progressive resistance intervention alone performed twice a week for 10 weeks in community gymnasiums near where the PWS participants lived, using pin-loaded weight machines, was shown to be feasible for people with PWS and could increase muscle strength.\[^18\] However, not all individuals with PWS have adequate money or transportation to access equipment in community gymnasiums, and there are not enough sports centers in the country for them to access; thus, another more economical and practical exercise-training mode should be developed.

The elastic band (EB) is inexpensive and easy to use, and can be used as home-based resistance training programs. It has been used among elderly people to increase muscular strength, and its physiological benefits have also been shown to be as effective as conventional weight machines or free weights for improving both upper limb strength and lower limb strength.\[^19,20\] However, no previous studies have investigated the effects of EB training on body composition and physical function among individuals with PWS. We hypothesized that a specific EB physical training program adapted for adults with PWS would decrease body fat and improve physical performance to prevent the further development of obesity. If EB training could improve physical performance and body composition, it would represent an additional simple and convenient treatment option for individuals with PWS for home-based program training.

### MATERIALS AND METHODS

#### Subjects

Individuals with a genetically confirmed diagnosis of PWS, aged over 18 years old, were recruited through a single hospital’s outpatient pediatric and genetic medicine clinic. The participants needed to have sufficient command of the Mandarin language to understand the study information, be motivated to complete the training program and have no contraindications to exercising according to the American College of Sports Medicine guidelines. Participants who could not follow simple instructions due to severe cognitive impairment, had severe musculoskeletal osteoarthritis or deformities that could interfere with exercising, or had participated in previous resistance exercises in the past 6 months were all excluded. In consideration of mild cognitive impairment, informed consent was obtained from both the participants and their parents. The study followed the principles of the Declaration of Helsinki, was approved by the Research Ethics Committee (04-XD33-100).

#### Procedure and intervention protocol

A 24-week training program using EBs (Thera-Band®, The Hygenic Corporation, Akron, OH, USA)\[^21\] was performed in a small group of participants with PWS. The EB resistance program was performed three times a week (once supervised and guided by an experienced physical therapist in a community day center near the hospital and twice supervised by teachers of the community day center who received instruction on how to perform the exercises) over 24...
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weeks for a total of 72 sessions. Each session lasted 60 minutes, including a 5-minute warm-up consisting of light-intensity aerobic and dynamic stretching exercises, followed by 50 minutes of EB exercise and a 5-minute period of cool-down and static stretching exercises. The nine EB exercises were designed to train specific major muscle groups, strengthen the motivation of participants, and facilitate home-based compliance, which included five exercises for the upper extremities (shoulder flexion, abduction, adduction, lateral raise, and elbow flexion), two for the trunk (side bending and bridging), and three for the lower extremities (squat, lateral quick kicks, and hip abduction plus external rotation) (Figures 1A and 1B).

Participants used the lighter Thera-band (yellow color) and were instructed to progressively increase resistance every two to four weeks or when they could perform more than 20 repetitions of a given motion by shortening the EB with more grip wraps at the handles. The initial resistance exercise intensity level was instructed at the level of 11 (light) to 13 (somewhat hard) on the Borg scale of perceived exertion and then progressively increased to the level of 15 (hard) to 17 (very hard). Three repetitive sets were performed for each exercise. An active recovery period of 60 to 90 seconds was allowed between exercises. Diet control was not specifically restricted, as family members were controlling the participants’ total caloric intake per day as usual.

Measurement

Functional motor performance was evaluated before the start of the 24-week training program (ERT0), at weeks 8 (ERT8) and 16 (ERT16), and at the end of the program (ERT24).

Height and body weight were measured and body mass index (BMI) was calculated. Changes in body weight, body mass index (BMI), regional body fat percentage, fat mass, and lean body mass between baseline and after the 24-week intervention were measured using dual-energy X-ray absorptiometry (DXA) (Hologic Densitometer Discovery A, USA). In order to reduce confounders, data of the left non-dominant upper and lower limbs are considered.

Physical assessments consisted of the following: (1) three 3-second-sustained handgrips using a Baseline hydraulic dynamometer with a 1-min interval between each measurement (the highest of the three handgrip measurements was included) to measure upper extremity muscle strength;[14,23] (2) the 30-second chair stand test and the timed up-and-go (TUG) test to measure lower extremity muscle strength, endurance and walking speed;[24,25] (3) the 2-minute step test to measure physical fitness;[24] and (4) the Berg Balance Scale (BBS) to assess balance.[26]

The 30-second chair stand test assesses the endurance in PWS individuals by counting the number of sit-to-stand repetitions achieved and is an important lower body strength clinical test.[23] The TUG test measures the time a PWS patient requires to rise from a chair, walk a 3-meter-long line on the floor, turn around, walk back, and sit down again.[25] Adults without balance problems can complete the test in less than 10 seconds, but those with limited mobility skills take more than 30s to complete the task.[25] Static and dynamic activities of varying difficulty were assessed using the BBS. Examined items varied from simple mobility tasks, such as transferring, standing up unsupported, and sit-to-stand movements, to more difficult tasks, such as turning 360 degrees, reaching forward with an outstretched arm, tandem stand, and single-leg stand.[26]

Statistical analysis

Data were analyzed using SPSS 20.0 (IBM SPSS Statistics). The results are presented as the means and standard deviations (SDs) or medians and ranges. The Wilcoxon signed-rank test was used to compare the functional motor performance and body composition at baseline and after the training program. In all tests, values of p < 0.05 were considered significant.

RESULTS

Six participants were recruited through the Prader-Willi Syndrome Association (Taiwan) and attended the community day work center located in the North of Taiwan during the daytime. Participants enrolled in this study included two females and four males, ages ranged from 19 to 31 years, with a mean age of 24.5 years old. The mean body weight was 72.33 ± 20.14 kg, and the mean BMI was 30.1 ± 6.73 kg/m². Subject characteristics
and physical performance scores at baseline are shown in Table 1. All adults with PWS showed less hand grip strength, fewer sit-to-stand repetitions during the 30-s chair stand test, a longer average time when performing the TUG test and fewer up-counting numbers during the 2-minute step test than did the healthy adults.\textsuperscript{[23-25]} Significant improvements were observed in body weight, BMI, regional fat mass, and body composition percentages and in all measures of physical performance after EB training.

After training, the adults with PWS exhibited significant reductions in body weight ($p = 0.028$) and BMI ($p = 0.043$). In addition, the fat amounts and percentages in the left upper extremity ($p = 0.048$; $p = 0.046$) and left lower extremity ($p = 0.028$; $p = 0.008$) also decreased significantly after the 24-week EB resistance training (Table 2). Although not significant, the amount of trunk fat also decreased.

### Table 1. Baseline physical characteristics of subjects with Prader-Willi syndrome

<table>
<thead>
<tr>
<th>Age (years)</th>
<th>Case 1</th>
<th>Case 2</th>
<th>Case 3</th>
<th>Case 4</th>
<th>Case 5</th>
<th>Case 6</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sex</td>
<td>M</td>
<td>M</td>
<td>M</td>
<td>F</td>
<td>M</td>
<td>F</td>
</tr>
<tr>
<td>Height (m)</td>
<td>1.60</td>
<td>1.51</td>
<td>1.65</td>
<td>1.44</td>
<td>1.53</td>
<td>1.52</td>
</tr>
<tr>
<td>BMI (Kg/m$^2$)</td>
<td>42.5</td>
<td>28.9</td>
<td>27.9</td>
<td>27</td>
<td>31</td>
<td>22.9</td>
</tr>
<tr>
<td>Grip (lbs)</td>
<td>65</td>
<td>55</td>
<td>31</td>
<td>35</td>
<td>60</td>
<td>40</td>
</tr>
<tr>
<td>TUT (s)</td>
<td>14</td>
<td>17</td>
<td>13</td>
<td>19</td>
<td>9</td>
<td>10</td>
</tr>
<tr>
<td>ST (times)</td>
<td>9</td>
<td>9</td>
<td>7</td>
<td>5</td>
<td>10</td>
<td>10</td>
</tr>
<tr>
<td>SP (times)</td>
<td>55</td>
<td>46</td>
<td>51</td>
<td>18</td>
<td>56</td>
<td>48</td>
</tr>
<tr>
<td>BBS (/56)</td>
<td>51</td>
<td>44</td>
<td>49</td>
<td>35</td>
<td>48</td>
<td>53</td>
</tr>
</tbody>
</table>

Abbreviations: BMI, body mass index; TUT, 3 meter timed up-and-go test; ST, 30 seconds sit to stand test; SP, step in place test; BBS: Berg balance scale.

### Table 2. Comparison of body composition before and after intervention

<table>
<thead>
<tr>
<th>Variables</th>
<th>Before intervention</th>
<th>After intervention</th>
<th>p value$^a$</th>
</tr>
</thead>
<tbody>
<tr>
<td>BW (kg)</td>
<td>72.35 ± 20.14</td>
<td>65.55 ± 15.89</td>
<td>0.028*</td>
</tr>
<tr>
<td>BMI (kg/m$^2$)</td>
<td>30.1 ± 6.73</td>
<td>26.48 ± 4.58</td>
<td>0.043*</td>
</tr>
<tr>
<td>LAF (kg)</td>
<td>20.34 ± 6.91</td>
<td>16.79 ± 7.55</td>
<td>0.046*</td>
</tr>
<tr>
<td>LAF (%)</td>
<td>54.63 ± 7.28</td>
<td>49.73 ± 7.53</td>
<td>0.046*</td>
</tr>
<tr>
<td>LAL (kg)</td>
<td>15.43 ± 33.41</td>
<td>14.88 ± 3.28</td>
<td>0.917</td>
</tr>
<tr>
<td>LLF (kg)</td>
<td>62.63 ± 24.40</td>
<td>52.34 ± 21.18</td>
<td>0.028*</td>
</tr>
<tr>
<td>LLF (%)</td>
<td>46.35 ± 4.02</td>
<td>41.63 ± 6.45</td>
<td>0.008*</td>
</tr>
<tr>
<td>LLL (kg)</td>
<td>67.51 ± 17.18</td>
<td>67.07 ± 14.49</td>
<td>0.753</td>
</tr>
<tr>
<td>TF (kg)</td>
<td>140.80 ± 63.28</td>
<td>129.49 ± 71.27</td>
<td>0.116</td>
</tr>
<tr>
<td>TF (%)</td>
<td>39.83 ± 6.00</td>
<td>26.62 ± 21.60</td>
<td>0.075</td>
</tr>
<tr>
<td>TL (kg)</td>
<td>183.30 ± 61.60</td>
<td>200.42 ± 43.58</td>
<td>0.075</td>
</tr>
</tbody>
</table>

Abbreviations: BW, body weight; BMI, body mass index; LAF, left arm fat; LAL, left arm lean; LLF, left leg fat; LLL, left leg lean; TF, trunk fat; TL, trunk lean.

Values are expressed as mean ± SD.

$^a$ Calculated by Wilcoxon rank test.
Figure 1A. Legend: Elastic band resistance exercises, including flexion of shoulders (A), flexion of elbows (B), abduction of shoulders (C), adduction of elbows (D), and trunk lateral bending (E).
Figure 1B. Legend: Elastic band resistance exercises, including trunk extension (A), external rotation of hips (B), abduction of the hip (C), and wide squat stance (D).
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Figure 2. Legend: Changes in the physical performances, including 30 seconds sit to stand (A), 3-meter timed-up-and-go test (B), step in place test (C), and Berg balance scale (D).
Abbreviations: ERT0, before elastic band resistance training intervention; ERT8, 8 weeks after elastic band resistance training intervention; ERT16, 16 weeks after elastic band resistance training intervention; ERT24, 24 weeks after elastic band resistance training intervention. TUT, 3 meter timed up-and-go test; ST, 30 seconds sit to stand test; SP, step in place test; BBS, Berg balance scale.
*p<0.05 with Wilcoxon rank test.

Regarding the physical performance assessments, the timed up-and-go test (p = 0.027), 30-second chair stand test (p = 0.028), and 2-minute step test (p = 0.028) all showed significant improvements as early as week 8 of training. The balance function (p = 0.027) improved significantly after 16 weeks of training. Comparisons of baseline and follow-up data regarding functional performance are shown in Figure 2.

**DISCUSSION**

The primary outcome of this pilot study suggests decreases in body weight (p = 0.028) and fat amounts and percentages in the left upper extremity (p = 0.048; p = 0.046) and left lower extremity (p = 0.028; p = 0.008) and improvements in physical performance after resistance EB training in adults with PWS. From an obesity risk factor perspective, our findings confirm the effects of EB training for the treatment to reduce adiposity, avoid muscle strength decline, and improve physical performance, including walking speed (p = 0.027), number of full stand-ups (p = 0.028), endurance (p = 0.028) and balance (p = 0.027). These findings are an important addition to the limited evidence of strengthening exercise alone in
PWS individuals. Previous studies also demonstrated positive effects on BMI and fat mass,\cite{11,17} but were conducted using aerobic exercise alone\cite{11} or a combination of aerobic and resistance exercises.\cite{16,17}

In contrast to previous studies,\cite{12,13,16,18} which showed improvements in lean body mass after resistance exercise training, our trial showed no significant increments in upper and lower extremities and trunk lean mass, but mild decreases in muscle mass, similar to other previous studies, including physical assessment of grip strength.\cite{11,17} Those studies were performed on children and adolescents with PWS who had been under growth hormone replacement treatment.\cite{12,13} As lean mass volumes detected by DXA also involve the contained extracellular water, our results could be possibly be explained by the impaired water metabolism identified in individuals with PWS, which indicate that the loss could be due to water loss rather than real lean mass loss.\cite{27} Other possible explanations could be attributed to lack of an additional protein supplement after resistance training\cite{28} or to GH deficiency, which increases adiposity while decreasing muscle mass.\cite{6} Another possible study limitation was that only one training session was supervised by the physical therapist, which might reduce the quality of the posture and intensity during the resistance training.

The physical assessments in our study included the determination of hand grip strength, the 30-second chair stand test, the timed up-and-go test, and the 2-minute step test, and all of which showed different degrees of improvement after 8 weeks of EB training, except grip strength. No previous studies have evaluated the physical performances, including walking speed, upper and lower extremity strength, endurance and balance, among individuals with PWS before and after exercise training. One study evaluated hand grip strength and dexterity before and after training, but did not reach statistical improvements after treatment, similar to our study results.\cite{14} At baseline, our subjects all showed decreased physical performance levels when compared with healthy adults, and their scores were similar to those among older populations.\cite{23-26} However, after training, significant improvements were found only in the physical performances which mainly involved the trunk and lower extremities. Poor motor coordination, agility, balance, and stamina have been described in the PWS literature.\cite{5,29}

An increased BMI not only has negative effects on physical activity but also leads to functional impairments, which can lead to impaired balance and increased risk of falls. Benefits from posture programs designed to improve balance and muscle strength and weight reduction programs have shown to improve posture instability.\cite{15,30} Paolo Capodaglio et al. found that individuals with PWS had poorer balance capacity than nongenetically obese counterparts and normal-weight individuals, and weight reduction, muscle strengthening and balance training were suggested to improve the condition, although the results of their subsequent study showed no obvious improvements in balance.\cite{15} In contrast, our study showed significant improvements after as early as 16 weeks of EB training. This improvement could be attributable to the EB resistance exercise program, which used nine activities designed to train different major muscle groups over the trunk and upper and lower extremities as opposed to focusing on only the distal lower extremities. Our results indicate that EB training improve the sedentary behavior of individuals with PWS through easy home training programs and could be considered as another training program.

This study is the first to investigate the effect of progressive EB training on the body composition and physical performance of individuals with PWS. However, the study has several limitations, such as the small sample size and the nonrandomized control design, which limit its generalizability to all people with PWS. In addition, the psychological profile of PWS individuals may represent an obstacle to adherence rates during training sessions, which were not recorded during this study. A further limitation is the lack of factors associated with diet consumption records, such as additional protein supplementation or diet restriction. Future studies with a larger sample size, a randomized control group, and longer-term follow-up are warranted to reduce data heterogeneity and to generate a better understanding of the effects of the training program on muscle strength.

**CONCLUSION**

Our study reports a simple, safe, low-intensity and effective EB training method for adults with PWS that is
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performed three times per week over a 24-week period and supervised by physical therapists or community educators. The results show that this method can improve physical performance and body composition in these individuals. In the future, studies with larger sample sizes, adequate control groups, higher load intensities, adequate diet control and longer-term follow up are warranted.

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Elastic band training improves adiposity and physical performance

普瑞德威利氏症候群成人患者彈力帶訓練對於肥胖症及身體功能表現之改善研究：前導性研究

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慈濟醫學大學醫學系 復健科 小兒科 4

本研究目的為調查與分析普瑞德威利氏症候群成人患者彈力帶運動訓練後之身體組成、肌力、肌耐力與平衡等動作表現的變化。受試者執行一週三次，每組 15-20 下，一次做三組的彈力帶訓練，總共訓練 24 週。利用雙能量 X 光吸收儀分析身體組成的變化。身體功能表現的測試包括：握力、坐站起走測試、30 秒坐站測試、以及柏格氏平衡量表。研究共收取了 6 位普瑞德威利氏症候群成人患者。經過 24 週的彈力帶漸進式訓練後，結果顯示體重(p = 0.028)、體脂(p = 0.043)、及局部肢體脂肪量及比率(p < 0.05)皆有顯著改善。至於身體功能的表現，經過 8 週的訓練，坐站起走測試(p = 0.027)、30 秒坐站測試(p = 0.028)、以及二分鐘登階測試(p = 0.028)皆有顯著進步。平衡功能則於訓練 16 週後才達顯著改善(p = 0.027)。經過 24 週的漸進式彈力帶訓練可提供普瑞德威利氏症候群成人患者另一種簡單又安全的訓練方法，初步顯示可改善身體脂肪量以及身體功能的表現。(台灣復健醫誌 2019; 47(2): 133 - 143)

關鍵詞：身體組成(body composition)、運動(exercise)、普瑞德威利氏症候群(Prader-Willi syndrome)