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Radiographic Outcomes of the Short and Intensive Rehabilitation (SHAiR) Program in Patients with Dropped Head Syndrome

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Background: The radiographic outcomes of nonoperative treatment of dropped head syndrome are still unknown. The purpose of the present study was to assess the change in sagittal spinopelvic radiographic parameters after the short and intensive rehabilitation (SHAiR) program in patients with dropped head syndrome.

Methods: This study included 48 consecutive patients with dropped head syndrome who presented with an inability to maintain horizontal gaze and who underwent the SHAiR program during the period of 2018 to 2019. Patients were divided into 2 groups according to their ability to maintain horizontal gaze at the time of final follow-up: those who had regained horizontal gaze (the "effective" group) and those who had not regained horizontal gaze (the "noneffective" group). Sagittal radiographic parameters including the sagittal vertical axis (SVA), the C2-7 angle, the C2-7 SVA, T1 slope, thoracic kyphosis of T1-5 and T5-12, lumbar lordosis, pelvic tilt, pelvic incidence, sacral slope, and curve flexibility, and demographic data and clinical outcomes were compared between the 2 groups using an unpaired t test, chi-square test, and Fisher exact test, as appropriate.

Results: Thirty-five patients in the effective group and 13 patients in the noneffective group were analyzed. The rate of response in regaining horizontal gaze with the SHAiR program was 73%. The C2-7 angle, the C2-7 SVA, T1 slope, and thoracic kyphosis (T1-5) demonstrated significant correction in the effective group ($p < 0.05$). There were no significant changes in other parameters below the mid-thoracic spine—i.e., the thoracolumbar and lumbar spine and pelvis—following the SHAiR program. Scores of the Neck Disability Index and visual analog scale for pain improved significantly in both groups.

Conclusions: The SHAiR program improved horizontal gaze among a large percentage of our patients and reduced cervical pain among patients overall. The correction of thoracic kyphosis (T1-5) might be an important treatment target to restore the appropriate T1 tilt in patients with dropped head syndrome.

Level of Evidence: Therapeutic Level III. See Instructions for Authors for a complete description of levels of evidence.

Dropped head syndrome is a rare condition involving chin-on-chest deformity and the characteristic clinical symptom of the inability to maintain a horizontal gaze[1,2]. Previous reports have shown that several diseases are associated with dropped head syndrome, including Parkinson's disease, amyotrophic lateral sclerosis (ALS), and psychiatric diseases[3-5]. Other reports have described dropped head syndrome in association with idiopathic neck-extensor muscle weakness[1,3]. However, the pathomechanisms of dropped head syndrome without any associated diseases are still unknown, and there is no standardized treatment algorithm.

Several reports have shown the effectiveness of cervical reconstruction surgery for patients with dropped head syndrome, including clinical and radiographic outcomes[6,7]. However, elderly patients are prone to concomitant medical pathologies and are at risk of notable perioperative complications, including severe dysphagia, pulmonary complications, implant failure, and death[1,2]. Therefore, nonoperative treatment has been regarded as a first-line treatment for dropped head syndrome as it is less invasive than surgical treatment. However, the literature on nonoperative treatment of dropped head syndrome is scarce[8,9]. Although radiographic parameters including the C2-7 sagittal vertical axis (SVA), the C2-7 angle, and T1 slope are standard measures for assessing the outcomes of cervical reconstruction surgery[10-12], we are not aware of any previous reports that have evaluated the radiographic and clinical outcomes of nonoperative treatment for dropped head

Disclosure: The **Disclosure of Potential Conflicts of Interest** forms are provided with the online version of the article (http://links.lww.com/JBJSOA/A558).

syndrome in a large number of patients. Although a few case series and case reports showed the effectiveness of nonoperative treatment for dropped head syndrome, those reports showed a low rate of sustained response[13]. Smith et al. reported the reciprocal changes in cervical alignment after surgical correction of lumbar sagittal malalignment[5], and therefore we established and reported a highly effective novel rehabilitation program for improving cervical alignment called the short and intensive rehabilitation (SHAiR) program, which comprises cervical, trunk, pelvic, and walking exercises for dropped head syndrome[14].

The aim of the present study was to investigate the radiographic and pain-related outcomes of the SHAiR program for consecutive patients with dropped head syndrome.

Materials and Methods

Study Design and Participants

This was a retrospective study conducted at a single institution in Japan. The study was approved by the institutional ethics committee review board. One hundred and fifty-three patients with dropped head syndrome who presented with an inability to maintain horizontal gaze and who underwent nonoperative treatment with the SHAiR program between 2018 and 2019 were considered for inclusion. We excluded patients with neuromuscular disease, cerebrovascular disease, inflammatory disease, psychiatric disease, ankylosing spondylitis, diffuse idiopathic skeletal hyperostosis, or a history of cervical spinal surgery or radiation therapy. We also excluded patients with <1 year of follow-up from the end of the hospitalization as well as patients who were not hospitalized. A total of 105 patients with Parkinson's disease, a history of cervical spinal surgery, collagen diseases, depression, cerebral infarction, irradiation, ALS, myasthenia gravis, no hospitalization, and follow-up of <1 year were excluded. We ultimately included 48 consecutive patients who were hospitalized for the SHAiR program. Patients were divided into 2 groups according to their ability to maintain horizontal gaze at the time of final follow-up: those who had regained horizontal gaze (those for whom treatment was effective, or the "effective" group) and those who had not regained horizontal gaze (those for whom treatment was not effective, or the "noneffective" group). We compared the radiographic and clinical outcomes between the 2 groups.

Variables

We recorded measurements of sagittal radiographic parameters including the SVA, the C2-7 angle, the C2-7 SVA, T1 slope, thoracic kyphosis of T1-5 and T5-12, lumbar lordosis, pelvic tilt, pelvic incidence, sacral slope, and curve flexibility as well as demographic data. The C2-7 angle in the extension position was also evaluated. We also recorded clinical outcomes including scores of the Neck Disability Index (NDI) and visual analog scale (VAS) for neck pain. Radiographic and clinical data were evaluated at the initial visit and at the time of final follow-up. We compared these variables both between the 2 groups and within each group between the initial visit and final follow-up.

SHAiR Program

All patients underwent the SHAiR program for 2 weeks with hospitalization[14]. Forty-minute training sessions were performed by each physical therapist and occupational therapist per day. The sessions included cervical paraspinal muscle exercise, range-of-motion exercise with cervical and thoracic mobilization, deep cervical flexor muscle exercise, hip-lift exercise, anterior pelvic tilt exercise, and walking exercise[14]. After discharge, the patients continued to perform the SHAiR program daily at home.

For the range-of-motion exercise with cervical and thoracic mobilization, patients were positioned supine on a stretch pole and guided to raise and lower their arms (Video 1). For the deep cervical flexor muscle exercise, patients were placed in the supine crook-lying position with the neck in the neutral position and were guided through each progressive pressure increment applied to the muscle using the feedback from the pressure sensor. For the hip-lift exercise, patients were instructed to lift their lumbar spine and pelvis while in the supine position. The anterior pelvic tilt exercise was performed as an active exercise wherein patients were instructed to move their pelvis back and forth using their upper extremity while sitting square (sitting on the edge of the bed with one's feet on the floor). A physical therapist assisted the patients in maintaining an upright posture during pelvic anteversion and instructed patients to avoid head and body trunk movement. The rehabilitation of the trunk and psoas muscle by the SHAiR program affected global spinal alignment and improved the clinical outcome of dropped head syndrome[14].

Statistical Analysis

Differences in variables from pre-treatment to final follow-up were compared between the effective group and the noneffective group using an unpaired t test, chi-square test, and Fisher exact test, as appropriate. Differences in variables from pre-treatment to final follow-up were also compared within each group using the same tests. P values of <0.05 with a confidence interval (CI) of 95% were considered significant. All statistical analyses were performed using the Statistical Package for the Social Sciences (SPSS Statistics, version 25.0.; IBM).

Source of Funding

No external funding was received for this study.

Results

Descriptive Statistics

In total, 48 patients with a mean age (and standard deviation) of 76.9 ± 6.5 years and a mean follow-up of 28.2 ± 17.1 months were included in this study. Patient characteristics and radiographic parameters are compared between the 2 groups in Table I. All patients showed impaired horizontal gaze (Video 2). The radiographic assessment showed a kyphotic C2-7 angle and large C2-7 SVA before treatment. Fulcrum bending radiographs showed flexibility of thoracic kyphosis (T5-12).

Surprisingly, the SHAiR program was effective for 35 (73%) of the patients, who regained horizontal gaze immediately after 2 weeks of rehabilitation and had preserved horizontal gaze

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TABLE I Comparison of Patient Characteristics and Radiographic Measurements

		Group		
	Total (N = 48)	Effective (N = 35)	Noneffective (N = 13)	P Value
Patient characteristic				
Age* *(yr)*	76.9 ± 6.5	76.1 ± 6.6	79.2 ± 5.9	0.14
No. female/male	42/6	29/6	13/0	0.17
Height* *(m)*	1.52 ± 0.07	1.52 ± 0.08	1.50 ± 0.04	0.15
Weight* *(kg)*	48.4 ± 8.0	49.3 ± 8.2	45.9 ± 7.4	0.19
BMI* *(kg/m^2)*	21.0 ± 3.0	21.2 ± 3.0	20.5 ± 3.1	0.45
Follow-up duration* *(mo.)*	28.2 ± 17.1	26.2 ± 16.7	33.7 ± 17.7	0.21
Radiographic parameter*				
C2-7 angle (positive value indicates kyphosis)				
Baseline	13.7° ± 27.0°	9.6° ± 27.7°	25.5° ± 21.6°	0.06
Extension	–26.5° ± 23.8°	–31.7° ± 21.9°	–18.4° ± 20.4°	0.10
Final	–2.9° ± 30.3°†	–8.8° ± 30.1°†	12.9° ± 25.5°	**0.02**‡
C2-7 SVA				
Baseline	62.0 ± 16.2 mm	60.5 ± 16.3 mm	66.0 ± 15.7 mm	0.30
Final	40.8 ± 17.2 mm†	37.8 ± 16.3 mm†	48.6 ± 17.5 mm†	0.07
SVA				
Baseline	0.6 ± 50.0 mm	1.2 ± 53.7 mm	–0.9 ± 39.8 mm	0.88
Final	7.0 ± 42.8 mm	11.6 ± 45.3 mm	–5.3 ± 33.7 mm	0.17
T1 slope				
Baseline	38.6° ± 15.5°	39.3° ± 15.1°	36.6° ± 17.0°	0.62
Final	32.0° ± 12.6°†	31.8° ± 13.1°†	32.5° ± 11.5°	0.85
Thoracic kyphosis (T1-5)				
Baseline	18.6° ± 10.6°	19.4° ± 11.2°	16.5° ± 8.8°	0.35
Final	13.2° ± 15.4°†	12.5° ± 16.3°†	15.2° ± 13.0°	0.55
Thoracic kyphosis (T5-12)				
Baseline	40.7° ± 15.0°	40.3° ± 14.7°	41.7° ± 16.3°	0.79
Fulcrum bending	25.6° ± 13.2°	25.8° ± 14.1°	25.0° ± 11.2°	0.83
Final	38.1° ± 17.2°	38.0° ± 17.7°	38.4° ± 16.5°	0.94
Lumbar lordosis				
Baseline	46.0° ± 16.4°	46.4° ± 15.5°	44.9° ± 19.3°	0.81
Final	46.6° ± 17.3°	46.7° ± 18.0°	46.2° ± 16.1°	0.92
Pelvic tilt				
Baseline	27.0° ± 9.3°	26.0° ± 9.6°	29.8° ± 8.4°	0.20
Final	25.4° ± 10.0°	24.9° ± 9.9°	26.7° ± 10.3°	0.59
Sacral slope				
Baseline	25.3° ± 10.6°	26.0° ± 10.4°	23.4° ± 11.1°	0.48
Final	27.2° ± 9.6°	28.1° ± 9.4°	25.1° ± 10.2°	0.37
Pelvic incidence				
Baseline	52.0° ± 9.4°	52.1° ± 9.6°	51.6° ± 9.1°	0.87
Final	52.0° ± 9.4°	52.1° ± 9.6°	51.6° ± 9.1°	0.87

*The values are given as the mean and standard deviation. BMI = body mass index, SVA = sagittal vertical axis. †Significant difference between baseline and final follow-up within the indicated group. ‡Bold indicates a significant difference between the Effective and Noneffective groups.

at the time of final follow-up (Video 3). When comparing radiographic measurements from the initial visit between these 35 patients (the effective group) and the remaining 13 patients (the noneffective group), we found no significant differences. The C2-7 angle, C2-7 SVA, T1 slope, and thoracic kyphosis (T1-5) showed significant correction between baseline and the time of final follow-up in the effective group ($p < 0.05$), while only the C2-7 SVA was significantly corrected in the noneffective

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TABLE II Comparison of Clinical Outcomes by Study Group*

	Group		
	Effective (N = 35)	Noneffective (N = 13)	P Value
NDI			
Baseline	15.0 ± 7.2	15.9 ± 7.0	0.70
Final	7.8 ± 4.7†	11.2 ± 5.5†	0.08
VAS			
Baseline	44.9 ± 27.3	58.8 ± 24.5	0.10
Final	12.8 ± 14.8†	17.9 ± 23.5†	0.49

*NDI = Neck Disability Index, and VAS = visual analog scale for pain.
†Significant difference between baseline and final follow-up within the indicated group.

group. There were no significant changes in the parameters below the mid-thoracic spine—i.e., thoracolumbar and lumbar spine and pelvis—following the SHAiR program.

In terms of the clinical outcomes, NDI and VAS scores improved significantly in both groups. There were no significant differences in NDI and VAS scores between the 2 groups at the initial visit or at the time of final follow-up, although the NDI score tended to be lower in the effective group than in the noneffective group at the time of final follow-up (Table II).

Case Presentation

A 77-year-old male patient with dropped head syndrome underwent the SHAiR program. At the initial visit, the patient showed moderate chin-on-chest deformity with cervical rotation. From baseline to final follow-up, correction was demonstrated in the C2-7 angle, from 35° to 26°; the C2-7 SVA, from 88 to 71 mm; T1 slope, from 41° to 25°; and T1-5 kyphosis, from 22° to 12°. The NDI score improved from 15 to 1 and the VAS pain score improved from 50 to 0 mm from baseline to final follow-up. The patient regained the ability to maintain horizontal gaze and was included within the effective group (Fig. 1).

Discussion

The literature on nonoperative treatment for dropped head syndrome is scarce and limited to case reports or case series[9]. Deneufgermain et al. reported the effectiveness of rehabilitation for cervical muscle strength in 2 cases[13]. We previously established the SHAiR program, which is a new

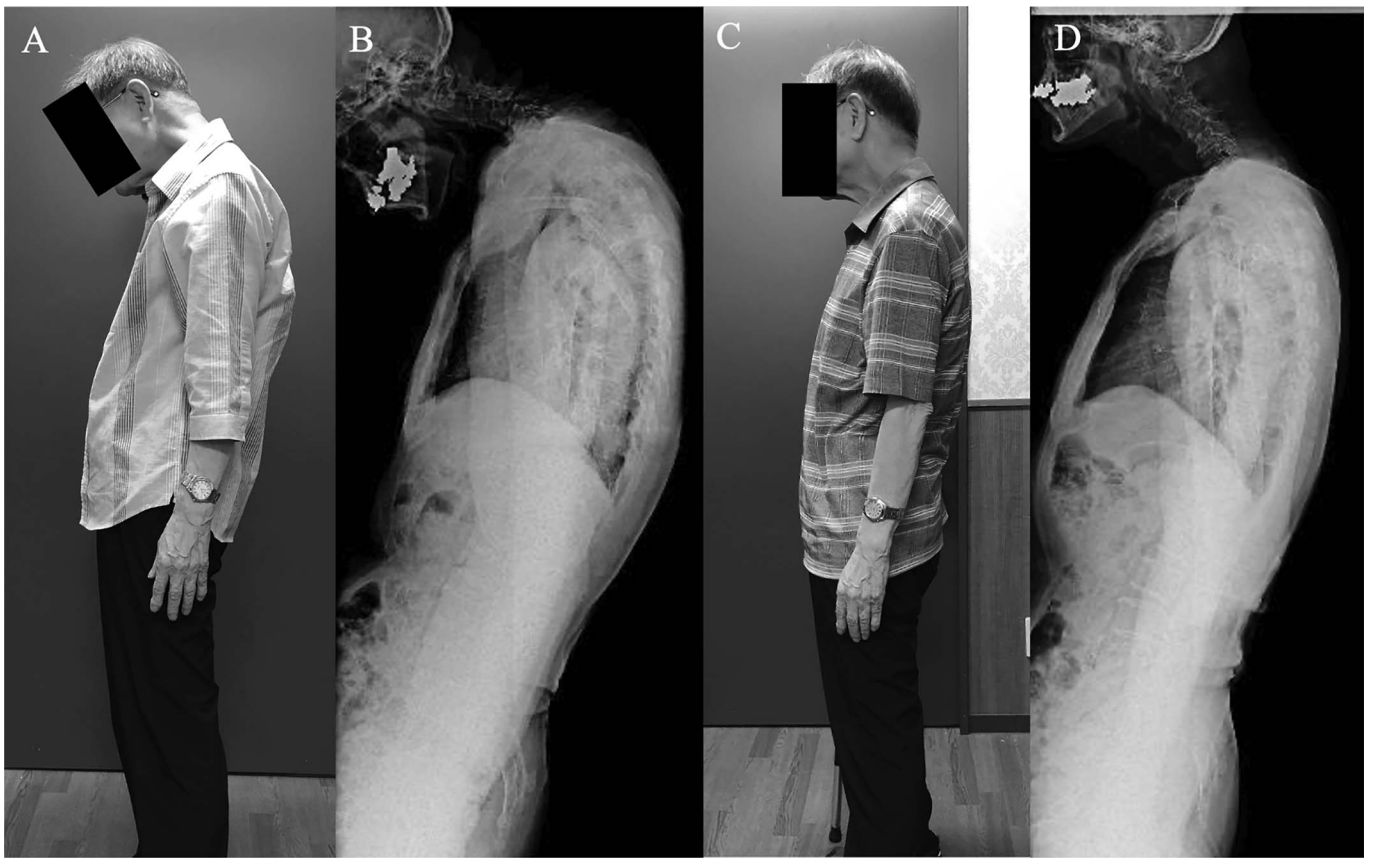

Fig. 1

A 77-year-old male patient with dropped head syndrome. **Figs. 1-A and 1-B** The patient presented with impaired horizontal gaze due to chin-on-chest deformity at the initial visit. **Figs. 1-C and 1-D** The patient regained horizontal gaze, and the C2-7 angle, the C2-7 SVA, T1 slope, and T1-5 kyphosis were corrected after the SHAiR program.

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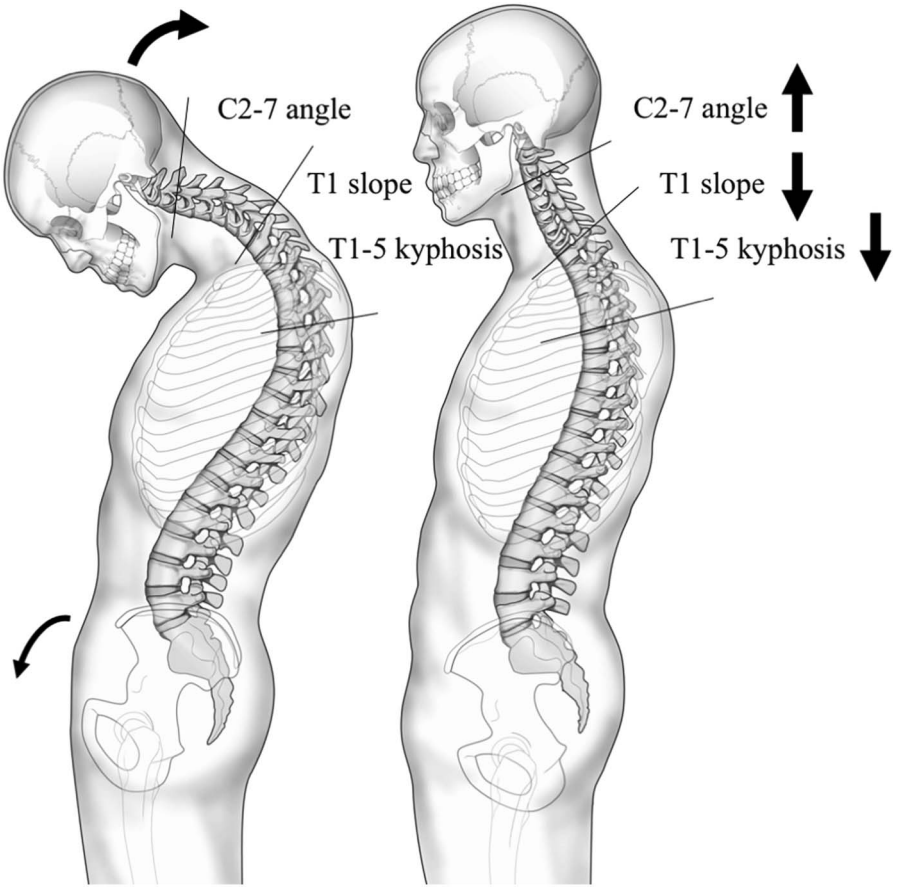

Fig. 2

Illustration of the typical change in global alignment from pre- to post-rehabilitation in effective cases. The regaining of horizontal gaze and substantial correction of the C2-7 angle, the C2-7 SVA, T1 slope, and upper-thoracic kyphosis were observed. Pelvic tilt and sacral slope tended to improve after treatment.

rehabilitation program for dropped head syndrome, and found significant improvement in cervical alignment[14-16]. In the current study, we evaluated 48 consecutive patients with dropped head syndrome who underwent nonoperative treatment using the SHAiR program. To the best of our knowledge, this is the largest study reporting the clinical and radiographic outcomes of nonoperative treatment for dropped head syndrome.

The SHAiR program includes several muscle and range-of-motion exercises for the trunk and lower limbs; in contrast, a conventional rehabilitation program consists of cervical paraspinal muscle and range-of-motion exercises[1,13]. Petheram et al. reported a low response rate for a program of strengthening the cervical and trunk extensor musculature and use of cervical collar in dropped head syndrome[1]. According to their report, 3 patients showed no improvement, 2 patients had mild improvement, and 1 patient showed marked improvement. In the current study, the SHAiR program was effective in restoring the ability to maintain horizontal gaze for 35 (73%) of 48 patients with dropped head syndrome, and the clinical response rate of the SHAiR program was much higher than that of other nonoperative treatment in the previous report. Therefore, the SHAiR program, comprising cervical, pelvic, and hip exercises to improve global spinal alignment, in addition to patient education for preventing stress on the cervical extensor muscle, might be a possible breakthrough for the treatment of dropped head syndrome.

On radiographic analysis, the C2-7 angle, the C2-7 SVA, T1 slope, and thoracic kyphosis (T1-5) were significantly corrected in the effective group. Other spinopelvic parameters, below the mid-thoracic spine, showed no significant changes at the time of final follow-up in either group. Murata et al. reported that the C2-7 angle exhibited a strong correlation with global balance among 41 patients with dropped head syndrome[17]. Hyun et al. reported that the T1 slope minus the C2-7 angle was associated with the outcomes of surgical treatment of cervical deformity[12]. Therefore, the radiographic treatment target for dropped head syndrome might be to restore the appropriate C2-7 angle and T1 tilt. However, we are not aware of any previous reports that have revealed how to improve T1 tilt. From our results, we believe that the correction of thoracic kyphosis (T1-5) might be an important enough target to restore the appropriate T1 tilt for the treatment of dropped head syndrome. An illustration of the typical change in global alignment from pre- to post-rehabilitation in effective cases is shown in Fig. 2.

Caruso et al. reported that neck pain of varying severity was a characteristic symptom of dropped head syndrome[18]. In the current study, NDI and VAS scores significantly improved,

suggesting that the SHAiR program may dramatically improve cervical pain and clinical outcomes.

There were several limitations to this study. First, this was a retrospective study at a single institution, and the level of evidence is low accordingly. Second, as the follow-up duration was short, the long-term clinical and radiographic outcomes of the SHAiR program were not evaluated. Third, compliance with the home regimen of the SHAiR program after hospitalization was not evaluated in this study. Additional study is therefore warranted to clarify the long-term clinical and radiographic outcomes of the SHAiR program for patients with dropped head syndrome.

In conclusion, we believe that this is the largest study to have investigated the outcomes of nonoperative treatment for dropped head syndrome, with a particular focus on radiographic parameters. The correction of thoracic kyphosis (T1-5) might be an important target in the treatment of dropped head syndrome. Our novel SHAiR program improved horizontal gaze among a large percentage of our patients and reduced cervical pain among patients overall and may potentially be a breakthrough for the treatment of dropped head syndrome. ■

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