

Continuous Incremental Heavy Halo Traction Combined with Posterior-only Approach for Severe Cervical Kyphosis with Neurofibromatosis-1.

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Abstract

Background Surgical management of cervical kyphosis in patients with NF-1 is a challenging task. Presently, anterior-only (AO), posterior-only (PO) and combined anterior-posterior (AP) spinal fusion are common surgical strategies. However, the choice of surgical strategy and application of Halo traction remain controversial. Few studies have shown and recommended posterior-only approach for cervical kyphosis correction in patients with NF-1. The aim of this study is to evaluate the safety and the effectiveness of Continuous-Incremental-Heavy Halo Traction (CIH-HT) combined with posterior-only approach for treatment of cervical kyphosis with NF-1. **Methods** 19 patients with severe cervical kyphosis due to NF-1 were reviewed retrospectively between January 2010 and April 2017. All the cases underwent CIH-HT combined with posterior instrumentation and fusion surgery. Correction result, neurologic status and complications were analyzed. **Results** In this study, cervical kyphosis Cobb angle decreased from initial 63.0 ± 21.0 degrees to postoperative 10.8 ± 4.0 degrees ($P \leq 0.01$), with total correction rate of 92%, which consist of 44% from CIH-HT and 48% from surgical correction. JOA scores were improved from preoperative 13.6 ± 1.6 to postoperative 16.0 ± 1.0 ($P \leq 0.01$). Neurological status was also improved. There was no correction loss and the neurological status was stable in mean 3.7 years follow-up. The incidence of complications was 36.8% (7/19). Six patients underwent local complications and one patient underwent a second surgery. **Conclusion** CIH-HT combined PO approach is safe and effective method for cervical kyphosis correction in patients with NF-1. A satisfied correction result, and successful bone fusion can be achieved via this procedure, even improvement of neurological deficits can also be obtained. Our study suggested that CIH-HT combined PO approach is another consideration for cervical kyphosis correction in patients with NF-1. **Key words** : Neurofibromatosis-1; Cervical kyphosis; Continuous-Incremental-Heavy Halo Traction; posterior-only approach;

Background

Neurofibromatosis (NF) is an auto somal dominant hereditary disorder that consists of two subtypes: NF-1 and NF-2. NF-1 (von Recklinghausen's disease) which is known as the most common form of neurofibromatosis with an incidence of 1 per 3000-4000 people worldwide, is associated with numerous clinical manifestations[1, 2]. Patients with NF-1 may present with a wide variety of clinical manifestations such as café-au-lait spots(over 90% affected), neurofibromas, Lisch nodules, and various skeletal abnormalities[3]. Spinal deformity is only seen in the subtype NF-1, and can be divided into two categories: dystrophic pattern and non-dystrophic pattern [4, 5].

Kyphosis is the most common deformity that occurs in the cervical spine of patients with NF-1[6]. Although cervical kyphosis are asymptomatic in most patients with NF-1, it can also cause neck pain with occasional neurological complications like nerve root compromise and complete or incomplete spinal cord deficits, with induced life-threatening paralysis[4]. Some patients with the tendency to have progression of kyphosis and deterioration of neurological function were advised to accept early surgical intervention[7]. Surgical management depends on multiple factors such as patient age, kyphotic angle, flexibility, extent of vertebral dysplasia and neurological status. Presently, anterior-only (AO), posterior-only (PO) and combined anterior-posterior (AP) spinal fusion are common strategies for the management of cervical kyphosis in patients with NF-1[8-10]. In recent years, single procedure and combined therapies have also been applied in the treatment of cervical kyphosis in patients with NF-1[8, 9, 11], however, the choice of surgical strategy remains controversial.

In this study, we evaluated the safety and effectiveness of Continuous Incremental Heavy Halo Traction (CIH-HT) combined with posterior-only (PO) approach in the treatment of cervical kyphosis in patients with NF-1.

Methods

Clinical characteristics

19 patients with cervical kyphosis due to NF-1 treated in our institution were reviewed retrospectively between January 2010 and April 2017 (table 1) including 8 males and 11 females, with average age of 15 years(range 7-26 years). Each patient presented with typical café-au-lait spots, and were definitely diagnosed with NF-1. Dystrophic changes were observed in 15

patients and 4 patients presented non-dystrophic. The involved cervical segments were from C2 to C7. In this study, 12 patients reflected cervical kyphosis while 7 patients presented additional neurological deficits such as neck pain, asthenia of limbs and positive pathological signs.

Imaging procedure

The deformity evaluation was performed on the anterior/posterior and lateral cervical radiographs. Dynamic lateral flexion and extension x-ray images and CT scans were also taken to evaluate the overall flexibility of the cervical spine. Magnetic resonance imaging (MRI) of the cervical spine was also obtained in all patients for further investigation of the intraspinal contents and compressive pathological features.

Clinical evaluation

All patients in this study underwent CIH-HT combined with posterior fixation and fusion. The indications for surgical intervention in these patients were severe cervical kyphosis impairing movement, mechanical neck pain, different degrees of neurological deficit, or progression of cervical deformity. The neurological function evaluation was based on the JOA scores.

Traction procedure

All patients underwent local anesthesia for halo placement. Patients were supine in the bed and halo gravity traction (HGT) applied in patients and started with a parallel traction. Then, a blanket roll was placed under the shoulders and the height was increased gradually. The traction direction was gradually changed from parallel traction to hyperextension traction. The initial traction weight was 2 kg; however, it increased subsequently by 0.5 kg every 3 days reaching maximum traction efficiency. It can be implied that a maximum traction efficiency has been obtained because there was no improvement of kyphotic angle with increasing traction weight or a maximum traction weight that the patient can tolerate has reached (Generally, maximum traction weight is generally less than 1/6 of the patient's weight.). After obtaining maximum traction efficiency, traction was maintained for 2 weeks and then one stage posterior internal fixation and correction were performed under HGT. Neurological examinations were performed 2 times per day, and if the patient had any complaint during the traction, the traction weight was reduced temporarily with the traction maintained. The traction should be continued according to the above method once the symptoms disappear. Lateral cervical radiograph was obtained every week to evaluate the effect of the traction and the improvement of kyphosis.

Surgical procedure

After maintaining maximum traction weight for 2 weeks, posterior spinal fusion and correction were performed under general anesthesia. The operative procedure was performed under maintained maximum traction weight. The lateral mass screws and/or pedicle screws, hooks were placed at the levels of fixation via a middle incision. Generally, patients with osteoporosis and lower bone mineral density can cause a reduction of screw holding force. It is necessary to insert more mass and/or pedicle screws, hooks, to provide more anchor points, disperse correction force, and stabilize the correction result. It is also necessary to loosen facet joints and posterior column osteotomy (PCO) before correction, including SPOs and Ponte osteotomy. After posterior fixation and kyphosis correction, abundant bone graft (allograft bone and / or autogenous iliac bone) was performed to create conditions for posterior column fusion. Somatosensory evoked potentials (SEP), motor evoked potentials (MEP) and wake-up test were used to evaluate the spinal cord function during operative process. All the patients wore cervicothoracic orthosis for three months postoperatively.

Evaluation methods

In order to evaluate the efficiency of CIH-HT combined with PO approach for cervical kyphosis correction in patients with NF-1, correction result was measured with Cobb angle and following parameters were used.

Traction correction rate = $\frac{\text{Pre-Tc Cobb} - \text{Post-Tc Cobb}}{\text{Pre-Tc Cobb}} \times 100\%$

Total correction rate = $\frac{\text{Pre-Tc Cobb} - \text{Post-Op Cobb}}{\text{Pre-Tc Cobb}} \times 100\%$

Surgical correction rate = Total correction rate – Traction correction rate

(Pre-Tc=Pre-Traction, Post-Tc= Post-Traction, Post-Op=Postoperative)

In order to evaluate the safety of CIH-HT combined with PO approach for cervical kyphosis correction in patients with NF-1, JOA scores were recorded to assess neurological functions.

Statistical Analysis

Data were managed and analyzed using SPSS 17.0. Data were presented with mean \pm SD. A paired sample t-test was used to test for significant differences. A $P < 0.05$ was considered statistically significant.

Results

Improvement of Cobb angle

In this study, Cobb angle decreased from initial 63.0 ± 21.0 degrees to Post-Op 10.8 ± 4.0 degrees. $P < 0.01$. Total correction was 92%. All the patients underwent CIH-HT for 10-61 days. (mean 21.8 ± 12.4 days) with maximum weight of 5 kg (mean 3.7 ± 0.7 kg). Cobb angle decreased from Pre-Traction (Pre-Tc) 63.0 ± 21.0 degrees to Post-Traction (Post-Tc) 35.1 ± 11.2 degrees. $P < 0.01$. Traction correction rate was 44%. Posterior fixation and correction was performed after maintaining maximum traction weight for 2 weeks. Postoperative Cobb angle was 10.8 ± 4.0 degrees. Surgical correction rate was 48%. All the patients were followed up for mean 3.7 years. Cobb angle was 10.3 ± 5.6 degrees in last follow up. There was no correction loss in follow-up. (table 2).

Improvement of JOA scores

In this study, 12 patients presented only cervical kyphosis while 7 patients displayed additional neurological deficits. JOA scores improved from preoperative 13.6 ± 1.6 to postoperative 16.0 ± 1.0 , $P = 0.01$. JOA scores were 16.3 ± 0.6 in 2 years follow-up, the neurological status was stable in follow-up. (table 3)

Complications

In this study, the incidence of complications was 36.8% (7/19). Loose halo frame (pin loosening) occurred in 3 patients during traction. Pre-traction external fixation pin tract infection occurred in 1 patient while 2 patients contacted pin site infection. 1 patient underwent a second surgery because of unilateral upper limb paralysis (Muscle force of right triceps brachii dropped from preoperative grade 5 to postoperative grade 3 and muscle force of right deltoid and biceps brachii dropped from preoperative grade 5 to postoperative grade 0). There were no systemic complications, pneumonia, thromboembolism, sepsis and peptic ulcers, which can arise due to prolonged bed rest.

Discussion

Cervical abnormalities associated with NF-1 include enlarged neural foramina, cervical kyphosis, and gross cervical kyphosis with subluxation or dislocation. Kyphosis is the most common deformity and surgical management of this deformity has received little attention in literature reviews. For cervical kyphosis in NF-1, successful managements requires early recognition, a more aggressive and reliable intervention to prevent disastrous worsening of the deformity. Several factors complicate the treatment and they are: i) a potential high risk of spinal cord injury during the correction. ii) difficulties in placing stable anchors in dystrophic vertebrae. iii) difficulty in obtaining solid bone fusion and iv) manipulation of the extreme degree of deformity in the presence of compromised cord may lead to severe cord damages and ischemia.[7]

Three approaches were recommended to manage cervical kyphosis; Anterior-only(AO) approach, Posterior-only(PO) approach and combined anterior and posterior(AP) approach[10, 12]. Most previous studies have recommended AP approach, while a

few studies have suggested anterior fusion or posterior fusion alone for cervical kyphosis associated with NF-1 because of some reasons, such as fusion failure, pseudoarthrosis and correction loss in follow-up[7, 11]. A successful spinal fusion via single approach is fraught with difficulties in NF-1. Winter and workmates[13, 14] and Sirois and Drennan[15] reported a 64% and 72% incidence of fusion failure. In a study from Parisini et al[16], fusion failures affected 53% of patients who received posterior instrumented fusion alone in comparison with 23% who underwent sequential anterior and posterior fusion. Hsu et al reported a 7.5% incidence of failure in dystrophic curves managed by AP[17]. Therefore, how to manage the fusion failure is a challenge for kyphosis correction in NF-1. It is well known that NF-1 affects bone quality (osteoporosis) and quantity (vertebral body dysplasia)[18]. It is extremely difficult to place stable anchors where there are severe dystrophic changes in the cervical spine [19, 20]. Without stable anchors, the correction will eventually be suboptimal. Moreover, the other surgical challenge seen in NF-1 is lower bone mineral density (BMD). It has been reported that decreased bone BMD in both sexes at an early age is up to 50% of individuals with NF-1[19]. Therefore, dystrophic and osteoporotic vertebral bodies may be insufficient to hold screws. Furthermore, preoperative HGT can also have an impact on the BMD. Long traction duration may bring more bone mineral loss[21]. In this study, CIH-HT combined PO approach was used for the treatment of cervical kyphosis in patients with NF-1. In order to achieve stable fixation and successful bone fusion via PO approach, two points, more anchor points and abundant bone graft, in the operation are particularly important. More lateral mass and/or pedicle screws, hooks, even short screws, could provide more anchor points, which could disperse correction force and stabilize the correction result. Additionally, Abundant bone graft(allograft bone and / or autogenous iliac bone) was adopted to provide enough solid support for a successful bone fusion. As a result, the correction result was stable and bone fusion was successful in follow-up. After the operation, all the patients wore a cervicothoracic orthosis for three months, postoperative external fixation is mandatory to maintain the correction and to obtain solid bone union. Our study showed that with support of more anchors, abundant bone graft and postoperative external fixation, a stable fixation and successful bone fusion can be achieved via PO approach for cervical kyphosis correction in patients with NF-1.

The application of HGT for treatment of cervical kyphosis in NF-1 is still controversial despite the use of halo spinal traction being widely used for treating severe spinal deformity[22-26]. Ward et al[27] reviewed 2 adult patients with cervical kyphosis in NF-1 and argued that traction can have a positive effective or lead to a disastrous consequence. Steinmetz et al believed that the time limit for traction is 3-5 days. There is no improvement of kyphosis if traction was more than 5 days[28]. However, some authors believed that traction was very important adjuvant treatment for cervical kyphosis. Recent studies have further shown the safety and efficacy of HGT as an operative adjunct procedure for cervical kyphosis correction. Kawabata et al[7] described preoperative HGT as a safe means to straighten cervical kyphosis before surgery in 3 patients with NF-1. HGT could provide a slow and gradual correction while the patients were awake, which typically decreased the amount of corrective force that needed to be applied to the cervical spine. With the help of preoperative HGT, partial correction of the deformity can be achieved to make the surgical procedure easier with less operative risk[21]. In this study, each patient underwent CIH-HT prior to the correction surgery. 44% traction correction rate was achieved eventually and the cervical kyphosis was corrected to safe preoperative levels. Our study further proved that CIH-HT was safe and an effective adjuvant management for cervical kyphosis correction in NF-1.

Compared with several previous studies[7, 8, 10, 11, 29, 30](table 4), CIH-HT combined PO approach had a better correction. CIH-HT provided the first level correction (44%), and surgery provided the second level correction (48%). CIH-HT provided safe partial correction, and typically decreased the amount of corrective force that needed during the surgery. Some factors contribute to the high traction correction rate. In this group, most of patients were presented with flexible kyphosis without a rigid facet joint and they underwent prolonged traction (an average of 20.9 days, the maximum of 61 days). The soft tissue and facet joint can be released to the maximum as a result of long time and heavy traction. Additionally, there is no strong muscles and ligaments around cervical vertebrae and long time hyperextension traction, which provided a continuous forward force in the cervical spine, and contributed to partial correction. Subsequently, intraoperative traction and PCO(posterior column osteotomy) was performed to provide a safe surgical correction rate and surgical correction rate eventually reached up to 48% via PO approach.

The complication rate for the management of cervical kyphosis associated with NF-1 has not been well defined. It was considered that surgical correction of cervical kyphosis in patients with NF-1 has one of the highest rates of complications in cervical spine surgery [31]. Postsurgical complications included cutaneous infection, junctional kyphosis, kyphosis progression, fusion failure and pseudarthrosis at final follow-up. However, a previous study from Helenius[31] stated that the risk of complications did not differ significantly according to the surgical approach. Additionally, Preoperative halo traction was not associated with a lower risk of complications (44% compared with 69%; $p = 0.24$). In this study, the incidence of complications was 36.8% (7/19). Although this complication rate is high, most of the complications (6/7) were traction-related as a result of being in bed for a long time and only 1 case underwent surgery-related complication. Our study indicates that CIH-HT combined PO approach for the treatment of cervical kyphosis in patients with NF-1 did not increase postsurgical complications.

Improvement of neurological deficits via PO approach is a challenge in cervical kyphosis correction. NF-1 may present multiple levels of involvement and is likely to form complicated paraspinal and/or intraspinal tumors. A intraspinal tumors or tumors with nerve root invasion should be resected to achieve nerve decompression, and this procedure should be performed before a manipulation for deformity correction is done[32]. In addition, a progressive deformity can also lead to severe neurological impairment. When a neurological deficit is present in a young patient with NF-1, it is usually caused by increased kyphosis[33]. In this study, neurological deficits were improved to some degree via CIH-HT combined with PO approach. We inferred the following reasons to project the improvement of neurological deficits. Firstly, the neurological deficits caused by cervical kyphosis in NF-1 are mainly compression in front of the spine. It could be improved simultaneously with cervical kyphosis through a long time standard traction. Secondly, the anterior column of the cervical spine is prolonged and the posterior column is relatively shortened via long time hyperextension traction. As a result, the compression in the front of spine can be improved. Thirdly, compression in cervical kyphosis is caused by apical vertebra and adjacent intervertebral disc. Traction can expand the intervertebral space and make intervertebral disc recovery to mean decompression. Also, the tolerance of spinal cord to ischemia and hypoxia was increased via a CIH-HT to reduce the risks of intra-operative neurological injury. Lastly, Posterior Column Osteotomy (Ponte osteotomy and SPOs) in apical region under intra-operative traction further released facet joint and shortened the posterior column, which decreased traction in the rear of spinal cord. Moreover, postoperative neurological deficit is another issue for cervical kyphosis correction in patients with NF-1. In a systematic literature review, Guzman et al[34] reported that the prevalence of C5 nerve root palsy was 7.7% after anterior cervical procedures and 7.8% after posterior procedures; however, most of the deficits were resolved spontaneously during the 2-year follow-up period. In this study, postoperative neurological deficits occurred only in 1 patient (5.3%, 1/19) who recovered after a revision surgery. We found that CIH-HT combined PO approach can also reduce the risk of postoperative neurological deficits.

Conclusion

This present study revealed that CIH-HT combined PO approach is a safe and effective method for cervical kyphosis correction in patients with NF-1. Our data indicated that satisfied correction result and successful bone fusion can be achieved via this procedure, and improvement of neurological deficits can also be obtained. Our study suggested that CIH-HT combined PO approach is another consideration for cervical kyphosis correction in patients with NF-1.

Abbreviations

Neurofibromatosis - NF

Neurofibromatosis-1 - NF-1

Anterior-only - AO

Posterior-only - PO

Combined anterior and posterior - AP

Continuous-Incremental-Heavy Halo Traction - CIH-HT

Three-dimensional computed tomography - CT

Magnetic resonance imaging - MRI

Japanese Orthopaedic Association - JOA

Posterior Column Osteotomy - PCO

Somatosensory evoked potentials - SEP

Motor evoked potentials - MEP

Pre-Traction - Pre-Tc

Post-Traction - Post-Tc

Postoperative - Post-Op

Bone mineral density - BMD

Declarations

Ethics approval and consent to participate

This study was approved by Ethic Committee of Central South University.

Consent for Publication

Written informed consent was acquired from each of the patients (or their parents and legal guardians) to authorize treatment, imageology findings, and photographic documentation. The patients (or their parents and legal guardians) consented to the publication of their pictures as well as their anonymous and clustered data.

Availability of data and material

The datasets analyzed during the current study are not publicly available but are available from the corresponding author on reasonable request.

Competing interests

The authors declare that they have no competing interests.

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Authors' contributions

ZZ contributed to the designs and drafted the manuscript. HZ and CG helped the design of the study and participated in the surgeries. AD,JL did the acquisition, analysis, and interpretation of data of the work. All authors read and approved the final version of the manuscript.

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Tables

Table 1: Demographic, Imaging, and Functional Data

NO.	Sex	Post-Tc Cobb(°)	Max Weight(kg)	CIH-HGT Time(days)	Post-OP Cobb(°)	Follow- up Cobb(°)	Pre- Tc JOA	Post- OP JOA	Follow- up JOA	Involved vertebra	Neurological symptoms	Dystrophic kyphosis
1	M	28	3	20	11	11	14	16	17	C3-C6	No	No
2	F	20	3.5	12	16	15	15	16	16	C3-C7	No	Yes
3	F	30	4	16	8	6	15	17	16	C4-C7	No	No
4	F	52	5	61	17	15	14	16	17	C2-C5	No	Yes
5	F	39	3.5	24	12	14	13	16	16	C3-C5	Yes	Yes
6	F	27	3.5	16	13	16	10	14	15	C3-C6	Yes	No
7	M	32	4.5	10	15	16	16	16	16	C3-C6	No	No
8	M	20	3	13	8	10	14	16	16	C2-C6	No	No
9	F	24	4	16	10	16	12	17	17	C3-C7	Yes	No
10	M	40	5	15	17	20	13	15	17	C2-C5	Yes	No
11	F	46	3.5	20	11	9	11	14	16	C2-C6	Yes	No
12	F	33	3	24	7	5	14	16	16	C3-C6	No	Yes
13	F	54	4.5	46	8	6	15	17	16	C3-C7	No	No
14	M	45	4	26	9	8	16	17	17	C3-C6	No	No
15	F	36	4	26	6	6	13	16	16	C2-C6	Yes	Yes
16	M	34	3	19	12	8	15	17	16	C3-C7	No	No
17	M	17	2.5	12	2	-4	14	15	17	C2-C6	No	Yes
18	F	52	3.5	20	14	10	13	17	17	C3-C6	Yes	No
19	M	37	4.5	18	9	8	14	16	17	C2-C5	Yes	No

Table 2 Improvement of kyphosis and statistical results

Parameters	Mean±SD	T value	P value
Pre-Tc vs. Post-Tc	63.0±21.0 vs. 35.1±11.2	9.90	0.01
Post-Tc vs. Post-Op	35.1±11.2 vs. 10.8±4.0	9.88	0.01
Pre-Tc vs. Post-Op	63.0±21.0 vs. 10.8±4.0	11.4	0.01
Post-Op vs. Follow-up	10.8±4.0 vs. 10.3±5.6	0.80	0.44

Table 3 Improvement of JOA score and statistical results

Parameters	Mean±SD	T value	P value
Pre-Tc vs. Post-Op	13.6±1.6 vs. 16.0±1.0	-8.26	0.01
Post-Op vs. Follow-up	16.0±1.0 vs. 16.3±0.6	-1.68	0.11

Table 4 Results of several previous studies

Authors	Year	Case	Pre-OP Cobb	Post-OP Cobb	Traction	Surgical procedure	Correction rate(%)
Yonezawa, I. et al	2003	1	72	35	No	ASF+PSF	51.40
Laohacharoensombat, W. et al	2010	1	120	55	Skull traction	ASF	54
J.M. Ma et al	2011	8	58.5 (mean)	2.5 (mean)	No	540° comb procedure	95.7
F.L. Wu et al	2012	1	125	30	Cervical suspensory traction	ASF+PSF	76
Kawabata, S. et al	2013	3	140/81/72	50/15/27	Halo-gravity traction	ASF+PSF	64.3/81.5/62.5
Kevin R. Choksey et al	2015	1	46	28	No	ASF	39.1

Figures

Figure not provided with this manuscript version.

Figure 1

Fig. 2 case 14 This case presented progressive NF-1 cervical kyphosis without neurological deficits. His preoperative neurological function was reflected by a JOA score of 16. His photos showed severe cervical kyphosis and many coffee spots on skin (Fig.1A). Radiography showed severe cervical kyphosis of 80° (Fig.2B). CT revealed there was no dystrophic changes in the cervical vertebra (Fig. 2C). MRI demonstrated a mild dural compression (Fig.2D).He received CIH-HT(Fig.2E) and the kyphotic angle gradually improved from 80° to 45° (Fig.2F).The posterior surgery was performed by placing pedicle screws and lateral mass screws from C2 to C7, Autogenous iliac bone and allogeneic bone were grafted at posterior column for fusion(Fig.1G).Cervical kyphosis improved to 17° after surgery (Fig.1H,I). Two years after surgery, CT showed a solid bone union and no significant correction loss (Fig.2J).