Research in Developmental Disabilities 34 (2013) 4286–4292

Contents lists available at ScienceDirect

Research in Developmental Disabilities

Pathological trunk motion during walking in children with Amyoplasia: Is it caused by muscular weakness or joint contractures?

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ARTICLE INFO

Article history:
Received 22 July 2013
Received in revised form 10 September 2013
Accepted 10 September 2013
Available online

Keywords:
Amyoplasia
Trunk pathologies
Duchenne gait
Strength
Range of motion

ABSTRACT

The aim was to investigate the causes for pathological trunk movements during gait in children with Amyoplasia. Eighteen children with Amyoplasia were compared with 18 typically developed children. Three-dimensional motions of pelvis, thorax and spine during gait were analyzed. Excessive trunk movements were defined as being above 4 standard deviations of those of typically developed children. Clinical examination of active strength and passive range of motion of the hip, knee and ankle joints were correlated to the parameter that showed the greatest prevalence of pathological trunk motion.

The greatest prevalence of 56% was seen for thorax obliquity range during walking. The spine angles showed the lowest deviations from typically developed children. Significant correlations ($p < 0.001$) between thorax obliquity range and clinical parameters were found for passive hip extension, hip flexion, hip abduction and active hip extension, hip flexion and ankle dorsiflexion strength. The highest correlation coefficients were found for passive hip flexion and active hip flexion strength of rho = −0.73 and rho = −0.69 respectively.

Excessive thorax obliquity during gait in children with Amyoplasia could be mainly caused by reduced strength and mobility of the hip. Therefore both mobility and strength of the hip are equally important and should be increased in the therapy to improve gait in children with Amyoplasia.

1. Introduction

Arthrogryposis multiplex congenita (AMC) is a heterogeneous condition which is characterized by multiple congenital contractures and muscular deficits in multiple body areas (Hall, 1997). The incidence of this syndrome was estimated between 1/3000 live births (Hall, 1997). It is believed that AMC is a result of fetal akinesia due to a number of different causes (Hall, 1997). The term 'Amyoplasia' refers to the most common arthrogrypotic syndrome that accounts for one-third of all patients (Hall, 1997). Amyoplasia is a condition characterized by a generalized lack in the newborn of muscular development

Abbreviations: AMC, Arthrogryposis Multiplex Congenita; ADN, Amyoplasia with natural Duchenne gait; ADE, Amyoplasia with excessive Duchenne gait; TD, Typically developed children; TOR, Thorax obliquity range.

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http://dx.doi.org/10.1016/j.ridd.2013.09.020

and growth with contracture and deformity at most joints; it is typically symmetrical and involves all four extremities with some variation (Bevan et al., 2007). In detail they have clubfeet which are associated with plantar flexor muscle weakness (Hoffer, Swank, Eastman, Clark, & Teitge, 1983), knee flexion contractures the hips are both flexed and external rotated. The shoulders are typically adducted and internally rotated, the elbows are extended with forearm pronation, wrists and fingers are flexed (Bevan et al., 2007). Children with Amyoplasia show maximum deformity at birth (Bevan et al., 2007). To increase their range of motion and to obtain a functional position of the joint, a combination of stretching, casting and often orthopedic surgeries are necessary (Staheli, 1998).

About 78% of patients with Amyoplasia are community level ambulators (Hoffer et al., 1983). However ambulation might require increased movements of the trunk to compensate for the contractures and weaknesses of the hip, knee and ankle joints. In children and adolescents with AMC increased lateral trunk sway, and rotation as well as pelvic lateral elevation, anterior tilt and transversal rotation were observed (Eriksson, Gutierrez-Farewik, Broström, & Bartonek, 2010). In particular the increased lateral trunk sway, also called "Duchenne gait" pattern (Duchenne, 1876) is most noticeable in public and lead to increased energy consumption (Duffels, Hill, Cosgrove, Corry, & Graham, 1996) and in consequence reduced walking distance. In addition Duchenne gait might increase the load on the spinal column (Kumar, 2004). The main reason for Duchenne gait found in the literature is abductor weakness (Duffels et al., 1996; Metaxiotis, Accles, Siebel, & Doederlein, 2000; Krautwurst et al., 2013). Another reason that has been speculated was, that lateral trunk lean assists foot clearance when either hip or knee flexion or ankle dorsiflexion are inadequate (Perry, 2010), which is quite common in Amyoplasia. Most therapeutic interventions have focused on the contractures. However, it has been shown in Amyoplasia that for motor function, muscle strength was more important for mobility than the passive range of motion (Kroksmark, Kimber, Jerre, Beckung, & Tulinius, 2006). Knowing the exact underlying reason for excessive trunk movements is important to find the best therapy to improve gait in patients with Amyoplasia.

Therefore the aim of this study is to determine increased trunk movements during gait in patients with Amyoplasia and determine its relation to clinical parameters of strength and passive range of motion in the hip, knee and ankle joints.

2. Materials and methods

2.1. Participants

In a retrospective cross-sectional study 18 children with Amyoplasia were included. Mean age was 8.8 (SD = 2.6) [4–12] years, mean bodyweight 29 (SD = 11) kg, mean body height 131 (SD = 18) cm, mean BMI 16 (SD = 3) kg/m^2, 12/18 were males. Eighteen typically developed children (TD) were measured for comparison. Mean age was 9.4 (SD = 2.3) [5–12] years, mean bodyweight 32 (SD = 10) kg, mean body height 139 (SD = 16) cm, mean BMI 16 (SD = 2) kg/m^2, 10/18 were males. All participants provided written consent, as approved by the local ethics committee. Patients had to be pain free and able to walk barefoot without assistance. Although few children used orthotics on daily basis, all gait studies were performed with the children ambulating barefoot. Exclusion criteria were spinal deformities and previous spinal surgeries, untreated hip dislocations and orthopedic surgeries within the last 24 months. Previous surgical procedures on the patients with Amyoplasia were listed in the following: 17/18 participants had clubfeet, of those, 10/17 were surgically corrected, 7/17 had Ponseti treatment following Achilles tendon tenotomies. 5/18 patients had surgeries to improve knee extension (3/5 supracondylar extension osteotomies, 1/5 knee flexor lengthening, 1/5 ventral epiphysiodesis), 5/18 patients had open hip reposition along with derotation and varisation osteotomies of the proximal femur combined with Pemberton pelvic osteotomy and psoas and adductor tenotomies.

2.2. Data collection

All participants underwent gait analysis with an 8 Camera system (Vicon MX, Oxford, UK) followed by a standardized clinical examination protocol listed in Table 1. The Vicon "Plug-in-Gait" marker set and model was used to determine orientation angles of the thorax and pelvis with respect to the laboratory frame. In addition spine angles were defined as the orientation angles of the thorax with respect to the pelvis. The subjects were asked to walk at comfortable speed down the 12 m walkway. Five consistent walking trials were used for the analysis.

2.3. Data analysis

Trunk motion of pelvis thorax and spine was reported in all 3 planes (sagittal tilt, sideward lean and transverse rotation). Selected parameters for the analysis were mean sagittal tilt, range of frontal lean and range of transverse rotation over the whole gait cycle. Mean sagittal tilt in the frontal plane was chosen instead of the total range, since the range of sagittal motion in these patients is typically low (Eriksson et al., 2010) and adaptive anterior pelvic tilt over the whole gait cycle due to hip flexion contractures (Perry, 2010) might have more relevance in patients with Amyoplasia.

The prevalence of excessive trunk motion was defined as the number of patients being outside 4 times the standard deviation of TD children during the left and/or the right gait cycle. Since most of the patients showed excessive deviations of the frontal plane thorax obliquity (TOR) this parameter was presumed to be most important and was correlated with the ROM and strength data listed in Table 1. Since contractures and muscular deficits in Amyoplasia are typically symmetrical at

Table 1
Clinical examination protocol strength test values were assessed according to Kendall, McCreary, and Provance et al. (1993). Line with the anterior superior iliac spine to femur long axis.

ROM	Subject position	Goniometer position
Hip extension	Prone, knee extended, test leg brought over edge of bed	Horizontal support to femur long axis
Hip flexion	Supine, contralateral hip extended	Horizontal support to femur long axis
Hip abduction	Supine, knee extended	
Knee flexion/extension	Supine, hip neutral	Femur long axis to tibia long axis
Ankle dorsi/plantar flexion	Supine, knee extended, foot prevented from supinating	Tibia long axis to calcaneus
Strength		
Hip extension/flexion	Seated, pelvis stabilized, ankle neutral	–
Hip abduction	Side-lying, pelvis stabilized knee extended	–
Knee extension/flexion	Seated, pelvis stabilized, ankle neutral	–
Ankle dorsi/plantar flexion	Seated, thigh stabilized, knee flexed 90°	–

Table 2
Prevalence of increased trunk motion during walking in children with Amyoplasia. Excessive motion exceeding 4 standard deviations above the mean value observed in typically developed children during walking.

	Sagittal mean flexion (%)	Frontal obliquity range (%)	Transversal rotation range (%)
Thorax	39	56	44
Spine	11	11	0
Pelvis	6	39	6

both limbs (Hoffer et al., 1983; Bevan et al., 2007) the trunk movements of the left and right gait cycle were pooled together. The strength of the correlation was defined 0.5–0.75 as moderate to good and above 0.75 as good to excellent relationships (Portney & Watkins, 2009).

To further describe the clinical parameters responsible for increased trunk motion, patients with Amyoplasia were divided into two groups. One group with natural Duchenne gait (ADN) and the other with excessive Duchenne gait (ADE) greater than 4 standard deviations of TD children. Wilcoxon rank-sum test on a significance level of $p < 0.001$ was performed to test differences in TOR, gait velocity, strength and ROM between ADN and ADE.

3. Results

The prevalence of increased trunk motion above 4 standard deviations of TD children is shown in Table 2. Most of the patients 10/18 (56%) showed an excessive range in thorax obliquity (TOR). Only 2/18 patients (11%) showed an excessive spine extension angle in the sagittal plane (hyperlordosis) and one of these showed in addition excessive frontal movement of the spine. Excessive deviations of the pelvis were shown mostly in the frontal plane. Independent of the selection of the threshold of 4 standard deviations, the prevalence for all 9 parameters was greatest for TOR also on lower thresholds of 3, 2 and 1 standard deviations resulting in 61%, 67% and 72% of prevalence in excessive TOR respectively.

Significant correlation coefficients between TOR and clinical parameters are shown in Table 3. Good correlation coefficients of rho = −0.73 and rho = −0.69 were found for passive hip flexion, and active hip flexion strength respectively. Passive hip extension, hip abduction, hip extension strength and ankle dorsiflexion showed only moderate correlation coefficients of rho = −0.57, −0.56, −0.57 and −0.53 respectively. The negative relation in all correlations indicates that a greater TOR was related to smaller passive ROM or strength. Significant correlation between strength and ROM parameters exists for the hip and knee in extension and flexion as well as for the ankle in dorsiflexion. The positive relation indicates better strength at a greater ROM.

In TD children average TOR was 4.8° (SD = 2.0°) therefore the threshold of excessive TOR, to differentiate ADE and ADN patients was set to 12.8° (mean plus 4 times SD). Median and range of the parameters measured on ADE, ADN and TD children were shown in Table 4. Walking speed normalized to leg length (Hof, 1996) was smallest for ADE and greatest for TD. Significant differences ($p < 0.001$) between ADE and ADN were observed for TOR, passive hip flexion and passive hip flexion strength. Fig. 1 shows the pelvic spine and thorax angles over the whole gait cycle for ADN, ADE and TD participants. Noticeable differences between TD children were observed for patients with excessive Duchenne gait in pelvic and thorax anterior tilt as well as for pelvic and thorax obliquity. For patients with natural Duchenne gait small deviations outside the normal range were found in pelvic obliquity and thorax tilt. For both groups the spine motion in all planes and pelvis and thorax in the transverse plane did not show considerable differences compared to TD children.

4. Discussion

A considerable amount of patients with Amyoplasia (56%) showed excessive Duchenne gait during walking. Significant ($p < 0.001$) correlations with good correlations coefficients between Duchenne gait and clinical parameters were found for

Table 3
Spearman correlation coefficients between TOR, range of motion (ROM) and strength parameters and within all parameters of all patients with Amyoplasia. Only significant results $p < 0.001$ were shown.

			ROM							Strength						
		TOR	Hip ext	Hip flex	Hip abd	Knee ext	Knee flex	Ankle df	Ankle pf	Hip ext	Hip flex	Hip abd	Knee ext	Knee flex	Ankle df	Ankle pf
Gait	TOR		−0.57	−0.73	−0.56	–	–	–	–	−0.57	−0.69	–	–	–	−0.53	–
ROM	Hip ext			–	–	–	0.56	–	–	0.53	0.55	–	–	–	–	–
	Hip flex				0.73	–	–	–	–	–	0.65	–	–	–	–	–
	Hip abd					–	–	–	–	–	–	–	–	–	–	–
	Knee ext						–	–	–	–	–	–	0.68	0.59	–	–
	Knee flex							–	–	0.71	0.70	–	0.57	0.74	–	–
	Ankle df								–	–	–	–	–	–	0.78	–
	Ankle pf									–	–	–	–	–	–	–
Strength	Hip ext										0.82	–	0.69	0.76	–	–
	Hip flex											–	0.68	0.75	–	–
	Hip abd												–	–	–	–
	Knee ext													0.69	–	–
	Knee flex														–	0.56
	Ankle df															0.64
	Ankle pf															

The first line shaded in gray indicates the important correlations with the range of thorax obliquity (TOR). The diagonal line shaded in gray shows possible correlations between strength and the associated range of motion.

Table 4
Median and range of clinical and gait parameters of patients with Amyoplasia subdivided into excessive (ADE) and natural (ADN) Duchenne gait. Values for typically developed children (TD) were shown for comparison. Wilcoxon rank-sum test between ANE and ADN were shown in the last column.

Parameter	ADE	ADN	TD	ADE vs. ADN
Thorax obliquity range [°]	24.1 [13.0, 47.0]	5.9 [3.7, 10.9]	4.8 [1.9, 10.9]	$p < 0.001$*
Velocity [non-dimensional]	0.3 [0.2, 0.6]	0.4 [0.2, 0.5]	0.5 [0.4, 0.6]	$p = 0.003$
Hip extension [°]	0.0 [−20, 5]	2.5 [−10, 15]	10.0 [0, 20]	$p = 0.021$
Hip flexion [°]	90.0 [40,130]	120.0 [90,130]	125.0 [120,140]	$p < 0.001$*
Hip abduction [°]	30.0 [10, 60]	40.0 [20, 50]	45.0 [30, 60]	$p = 0.003$
Knee extension [°]	−10.0 [−30, 10]	0.0 [−20, 15]	0.0 [0, 15]	$p = 0.001$
Knee flexion [°]	90.0 [30,110]	137.5 [20,150]	147.5 [140,155]	$p = 0.007$
Ankle dorsiflexion [°]	0.0 [−15, 20]	5.0 [−5, 20]	20.0 [5.0, 30.0]	$p = 0.130$
Ankle plantarflexion [°]	15.0 [5, 40]	10.0 [0, 30]	40.0 [20.0, 50.0]	$p = 0.396$
Hip extension strength	3.0 [1.0, 4.5]	4.0 [3.0, 5.0]	5.0 [4.0, 5.0]	$p = 0.003$
Hip flexion strength	2.0 [1.0, 4.0]	4.8 [2.0, 5.0]	5.0 [4.0, 5.0]	$p < 0.001$*
Hip abduction strength	4.0 [1.5, 5.0]	4.5 [3.5, 5.0]	5.0 [4.0, 5.0]	$p = 0.127$
Knee extension strength	3.8 [1.0, 4.5]	4.5 [1.0, 5.0]	5.0 [5.0, 5.0]	$p = 0.007$
Knee flexion strength	2.8 [1.0, 4.0]	4.5 [2.0, 5.0]	5.0 [4.0, 5.0]	$p = 0.001$
Ankle plantarflexion strength	2.5 [0.0, 4.0]	3.3 [1.0, 5.0]	5.0 [5.0, 5.0]	$p = 0.027$
Ankle dorsiflexion strength	1.0 [0.0, 4.0]	3.3 [1.0, 5.0]	5.0 [5.0, 5.0]	$p = 0.001$

* Significant results $p < 0.001$.

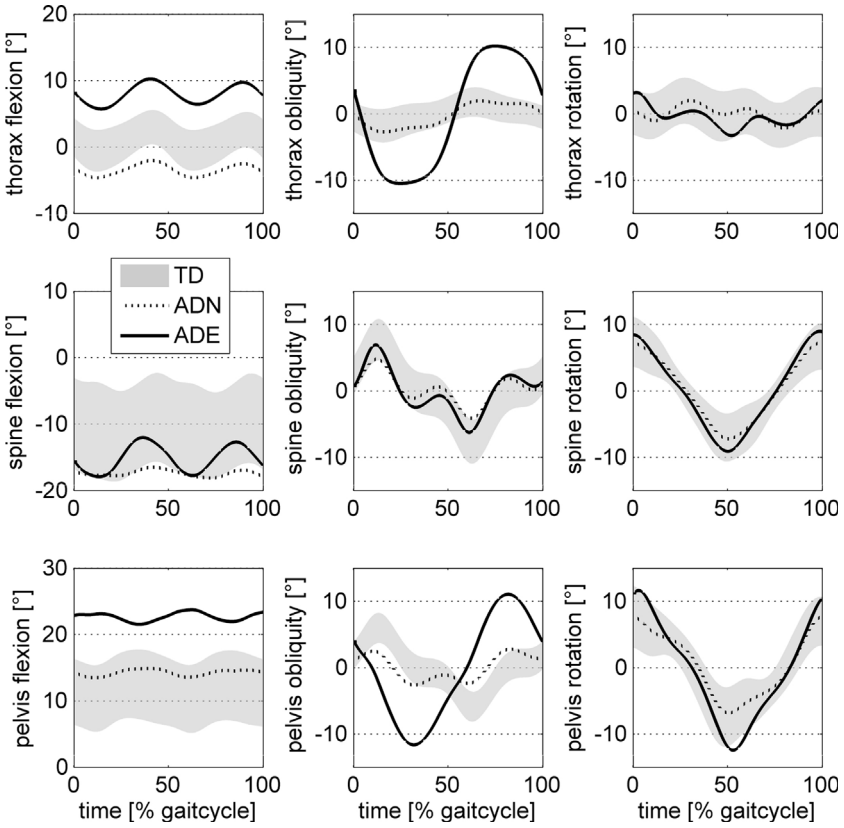

Fig. 1. Mean pelvis spine and thorax angles over the whole gait cycle of patients with excessive (ADE, $n = 10$) and natural (ADN, $n = 8$) range in thorax obliquity. The range and standard deviation of TD participants were shown for comparison. The sagittal, frontal and transverse plane are shown in the first, second and third column respectively.

passive hip flexion and active hip flexion strength. Therefore both limited active strength and limited passive ROM of the hip were suggested to be the main cause for compensatory Duchenne gait.

In the literature thorax movements in children and adolescents with AMC were reported to be inhomogeneous (Eriksson et al., 2010). Increased TOR of ($M = 21°$, SD = 7°) was shown for the most impaired children and adolescents that walk with knee and ankle foot orthoses, compared to ($M = 7°$, SD = 3°) for those walking only with ankle foot orthoses. This corresponds to the median of 22.7° for ADE and 7.3° for ADN children observed in this study. While the thorax was showing an

exaggeration of the natural movement pattern in the frontal plane in Fig. 1, the pelvis showed an unusual movement pattern. The pelvis in patients with ADE showed an excessive elevation of the ipsilateral side during swing phase, the so-called "pelvic hike" gait pattern (Perry, 2010). This pelvic hike gait pattern was also observed for patients with AMC in a previous study (Eriksson et al., 2010).

In this study the greatest prevalence of compensatory trunk movements were found for an excessive lateral thorax sway during walking (Duchenne gait). Besides the highest prevalence it has a great clinical relevance because: (1) children are concerned about their distinct physical appearance among their peers, (2) children have a reduced walking distance due to higher energy consumption (Duffels et al., 1996), and (3) excessive spinal lateral tilt might potentiate the risk for low back pain (Kumar, 2004). The latter can be excluded based on the results of this study, since the spine does not have to balance the excessive thorax lean because of the pelvic hike gait pattern mentioned in the previous paragraph. However goal directed therapy by knowing the exact reason for excessive trunk sway is still important for the wellbeing and participation of these patients. For a predictive correlation analysis the heterogeneity of TOR in the patients with Amyoplasia observed in this study was a good prerequisite. The correlation analysis revealed significant correlations for passive hip extension, hip flexion, hip abduction and active hip extension and hip flexion strength. Best correlation coefficients were found for passive hip flexion and hip flexion strength that showed also significant differences between both groups. This is in agreement with the literature which reported reduced hip flexion strength for AMC patients with increased TOR (Eriksson et al., 2010). Hip flexion strength is required during swing to achieve sufficient foot clearance; if hip flexion strength is low compensatory pelvic hike observed in ADE might assist foot clearance. Duchenne gait was suggested as a deliberate action that reduces the abductor moment required to stabilize the trunk mass during walking. Therefore Duchenne gait was typically associated to abductor weakness (Duffels et al., 1996; Metaxiotis et al., 2000; Krautwurst et al., 2013). Contrary to this study abductor strength in patients with ADE was not significantly reduced (Table 4). The reason of the Duchenne gait might therefore be to assist the abductors in performing the pelvic hike gait pattern to elevate the ipsilateral side of the pelvis during the stance phase of gait.

In this study both hip flexion and hip flexion strength were related to TOR, so that both seem to be equally important for walking. In addition hip abduction seemed to be related to TOR however passive hip abduction showed good correlation with passive hip flexion. One reason might be tightness of m. gluteus that act as extensor and partly as adductor and external rotator that will limit passive hip flexion, hip abduction, and internal rotation.

In the literature strength was suggested to be more important than ROM (Kroksmark et al., 2006). In this study ROM and strength were positively correlated with each other for hip flexion, knee extension, knee flexion, and ankle dorsiflexion. This suggests that adequate strength might only be achieved with an adequate range of motion. This has been shown in training science where training over the full range of motion resulted in greater strength gain than training in only parts of the joint range (Pinto et al., 2012). This hypothesis is further supported by the long term follow up study in AMC patients (Fassier, Wicart, Dubousset, & Seringe, 2009) where the progressive muscle activity improvement after the recovery of a passive range of motion was suggested as good prognostic factor for functional ambulation in AMC. The authors further concluded that active hips and knees were most important for ambulation. This agrees with this study where considerable limitations of passive ankle ROM in Amyoplasia compared to TD children did not show significant differences between ADE and ADN.

Hip weakness responsible for increased TOR in ADE might be also a result of previous psoas recession due to open repositioning of the hip. 4/5 patients with previous hip surgeries were included in the ADE group and only 1/5 in the ADN group, which suggest a possible relation with the psoas tenotomies performed.

Dividing patients based on 4 standard deviation of TOR might be a useful way to classify ambulating patients with Amyoplasia into two groups. Excessive TOR is relatively easy to recognize for the clinician without any gait analysis system and consequently suggest therapy for improving predominantly hip mobility and strength.

Besides Duchenne and pelvic hike gait pattern in the frontal plane, patients with ADE showed in addition an increased pelvic and thorax anterior tilt in the sagittal plane (Fig. 1). Pelvic anterior tilt might be caused by the reduced hip extension ability. Because in mid-stance phase when the hip continues to extend, the weight bearing limb moves behind the body, hereby hip flexion contractures cause the extension to be limited. This limitation requires adaptation of increasing anterior pelvic tilt to achieve a sufficient step length. Thorax anterior tilt might be a consequence of the pelvic anterior tilt, since in case of an upright thorax; the spine would have to balance anterior pelvic tilt with increased hyperlordosis that might increase the risk of developing back pain (Beckers & Bekaert, 1991; Keorochana et al., 2011).

The limitations of this study are that subjective manual testing was used to quantify strength. Measurements using handheld dynamometer or isokinetic exercise machines might provide more reliable data. However manual testing might be more relevant in typical clinical examination. Correlation between two variables does not imply that one variable causes the other, but a good correlation is a necessary prerequisite for causation. However in this study a sound biomechanical explanation can be given: increased TOR might compensate for reduced foot clearance caused by impaired hip flexion during walking.

5. Conclusions

The results of this study suggest that compensatory Duchenne gait in children with Amyoplasia could be mainly caused by reduced strength and passive mobility of the hip in flexion. Therefore both passive ROM and strength of the hip muscles are equally important and should be increased helping children to grow into adulthood with an appealing and efficient gait pattern.

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