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CLINICAL INVESTIGATION

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Clinical characteristics and surgical outcomes of adults with acute acquired comitant esotropia

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Abstract

Purpose To investigate clinical characteristics of adults with acute acquired comitant esotropia and to evaluate the muscle recession amount needed to achieve a favorable outcome after performing medial rectus muscle recession.

Study designs Retrospective study.

Methods Patients diagnosed with acute acquired comitant esotropia, who underwent medial rectus muscle recession with adjustable suture between 2008 and 2016 were included. Surgical outcomes were classified into motor and sensory. The motor outcomes were evaluated at the 1-year postoperative visit and divided into success (orthotropia or esodeviation ≤ 8 PD) and failure (esodeviation > 8 PD). The successful sensory outcomes were defined as elimination of diplopia in primary gaze. Factors including age, sex, refractive error, deviation angle, and surgical amount were compared between groups.

Results Sixteen subjects were included whose mean (± SD) age at the initial visit was 27.5 ± 11.0 years. Mean preoperative maximum angle of deviation was 27.9 ± 9.3 PD at distance and 28.6 ± 12.0 PD at near. Mean refractive error was -2.55 ± 2.92 D. Twelve of 16 subjects (75%) had successful motor and sensory outcomes. Age, sex, refractive error and deviation angle were not different between the two groups. Both success and failure groups required a greater amount of medial rectus muscle recession than those indicated by the Parks' surgical table, with a 40.6 ± 25.8 % augmentation in the success and 7.9 ± 6.9 % in the failure group (P = .028).

Conclusions To achieve better surgical outcomes in adults with acute acquired comitant esotropia, targeting postoperative orthotropia by increasing the amount of medial rectus muscle recession is recommended.

Keywords Acute acquired comitant esotropia · Adult esotropia · Medial rectus recession

Introduction

Acute acquired comitant esotropia (AACE) characterized by the acute onset of esotropia with diplopia, usually occurs in older children and adults. In 1958, Burian and Miller [1] reviewed the features of acute AACE and categorized it into three types. Type I (Swan type) involves binocular vision disruption by monocular occlusion or vision loss secondary to injury or disease [2]. Type II (Franceschetti type) is characterized by no underlying cause other than possible physical or psychological shock [1, 3]. Type III (Bielschowsky type) is associated with uncorrected myopia [4]. Patients with all three types have a large angle of esotropia and no underlying neurological disease. However, some recent studies show that intracranial disease may cause AACE and that neuroimaging should be performed on these patients, especially in older children [5–8].

AACE without an underlying neurological etiology requires surgical treatment because spontaneous recovery is rare. In addition, once patients have achieved orthotropia, they have a good prognosis of binocular recovery [9–11]. However, most of the published reports on AACE have included either children [7, 10, 12, 13], or both children and adults [1, 8, 11, 14]. Very few reports describe clinical findings and surgical outcome only in adults with AACE.

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Here, we investigated the clinical characteristics of adults with AACE and evaluated the recession amount needed to achieve a good outcome after performing medial rectus muscle recession.

Methods

Subjects

We retrospectively reviewed the medical records of patients older than 17 years of age who underwent surgery for AACE at Seoul National University Hospital in South Korea between January 2008 and May 2016. We performed the alternate prism cover test in the 9 diagnostic positions of gaze and Lancaster test to check the comitancy of esotropia. AACE was defined as a sudden onset of concomitant (same deviation in all gaze directions) non-accommodative esotropia without limitation of eye movement. At the initial visit, all patients underwent manifest and cycloplegic refraction and refractive error was converted to the spherical equivalent value (in diopters [D]). Myopia was represented as negative numbers and hyperopia as positive numbers. Patients also underwent stereoacuity testing with the Titmus stereotest (Stereo Optical) and all values were transformed to log arc/sec for statistical analysis. Fusional ability was measured using the Worth four-dot test at distance and near fixation.

All patients underwent laboratory testing to ensure that there were no underlying systemic conditions. Brain and orbit magnetic resonance imaging were also performed on all patients to confirm that there were neither neurological abnormalities nor abnormalities of extraocular muscles and connective tissues such as sagging eye syndrome that could cause esotropia. Patients with abnormal thyroid function measured by T3, T4, free T4, and TSH levels, who were positive for the acetylcholine receptor antibody or had brain abnormalities were excluded from analyses. Patients who had a history of strabismus surgery or congenital or acquired ophthalmic conditions (e.g., optic nerve disease, glaucoma, media opacity, or cataract) were also excluded. This study was approved by the Institutional Review Board of Seoul National University Hospital in South Korea and the study protocol followed the tenets of the Declaration of Helsinki.

Surgical procedures

The alternate prism cover test was performed as part of preoperative examination to measure the angle of deviation in prism diopters [PD] at distant (6 m) and near (33 cm) fixation. Esodeviation was represented as positive numbers and exodeviation as negative numbers. Based on the preoperative maximum angle of deviation, the largest deviation during follow-up before surgery, patients underwent either unilateral or bilateral medial rectus recession by a single surgeon (S-J K) under local or general anesthesia. Patients were considered eligible for surgery under topical anesthesia if they tolerated moderate preoperative pressure over the bulbar conjunctiva overlying the medial rectus muscle of 1 eye applied using a cotton swab stick in the clinic after topical application of 0.5% proparacaine hydrochloride.

Limbal incisions were made in all cases. The medial rectus muscle was isolated on a muscle hook and connective tissue is dissected posteriorly. A central and two locking edge bites of the muscle were taken using a double-ended 6-0 polyglactin (Vicryl) suture. The sutures were tied permanently, and we fixed the muscles on the sclera and sutures were secured using a sliding noose technique on the site of adjustment.

In patients with local anesthesia, surgery was performed after topical application of 0.5% proparacaine hydrochloride. An anesthesiologist monitored the procedure and, upon surgeon request, provided minimal doses of propofol or alfentanil hydrochloride. The dose administered was individually determined for each patient. We recessed the medial rectus muscle according to the surgical amount indicated by Parks' surgical table [15] (Table 1) that the surgeon referred to when treating other types of esotropia, based on the maximum angle of esodeviation. Then, an intraoperative adjustment was performed using distant and near centered targets with the patient in a seated, upright position with any required refractive correction. The intraoperative goal was to eliminate binocular diplopia by making the orthotropia. If diplopia remained, we increased the amount of recessing the muscle, and rechecked the presence of diplopia. If diplopia was eliminated, we completed the surgery and recorded the total surgical amount.

In patients with general anesthesia, the surgical amount of medial rectus muscle recession was augmented. The surgical amount was increased by up to 2 mm (range: 1-2 mm) from

Table 1 Surgical table recommended by Parks et al. for medial rectus muscle recession in patients with esotropia

Angle of esodeviation (PD)	Amount of bilateral MR recession (mm)
15	3
20	3.5
25	4
30	4.5
35	5
40	5.5
50	6.0
60	6.5
70	7.0

Abbreviations: PD prism diopters

the amount of recession (Parks' surgical table [15]) based on the maximum angle of esodeviation. If diplopia persisted following surgery, or only small esodeviation remained immediately after surgery, we checked each patient between 3 and 5 days after surgery and performed the any necessary adjustments within 5 days of surgery. All patients included in the study were followed after surgery for at least 1 year.

Outcome measures

We investigated clinical factors including age, sex, duration of esodeviation between the age of presumed onset and that at initial visit, refractive error, angle of deviation, stereopsis and amount of medial rectus muscle recession. Surgical outcomes were classified into motor and sensory outcomes. The motor outcome was evaluated at the 1-year postoperative visit and divided as follows: success (orthotropia or esodeviation ≤ 8 PD) and failure (esodeviation > 8 PD or exodeviation > 8 PD). The successful sensory outcome was defined as elimination of diplopia in primary gaze. Pre- and postoperative ophthalmologic factors were compared between success and failure groups.

Statistical analyses

Statistical analyses were performed using SPSS software (version 23 for Windows; SPSS). As the data showed abnormal distribution, non-parametric methods including the Mann-Whitney test and Fisher's exact test were used. For all tests, $P < .05$ was considered significant.

Results

Demographic and clinical characteristics of subjects

Of 98 adult patients with esotropia who underwent either unilateral or bilateral medial rectus recession, 16 (10 men), were included in this study. The mean age at the initial visit was 27.5 ± 11.0 years (range 17.0–63.5 years). All patients suddenly developed binocular diplopia without loss of vision or interruption of fusion and no definite systemic or ophthalmic causes for diplopia were identified. There was no patient with type I (Swan type) and two patients developed esotropia after definite physical or psychological shock, considered as type II (Franceschetti type).

The duration of esodeviation was 2.4 ± 2.5 months. Mean preoperative maximal angle of esodeviation was 27.9 ± 9.3 PD (range 12.0–45.0 PD) at distance and 28.6 ± 12.0 (range 12.0–50.0) PD at near fixation. Mean cycloplegic refraction was -2.66 ± 3.20 D (range -9.75~+0.75 D) in the right eye and -2.44 ± 2.63 D (range -6.75~+1.75 D) in the left eye. No patient had pseudomyopia, which was defined as more than 1 D difference between the cycloplegic and manifest refractions. Seven of 16 patients had diplopia at distance fixation and 6 of 16 patients had diplopia at near fixation by Worth-four dot test.

Mean age at the time of surgery was 28.2 ± 11.0 years (range 17.6~64.4 years). In all patients, the results of intraoperative forced duction test were negative. Surgery was performed under general anesthesia in 7 subjects and under local anesthesia in 9 subjects. Two patients with general anesthesia had a postoperative adjustment because of persistent diplopia. The mean angle of esodeviation at 1 week and 1 year was 0.1 ± 2.0 PD, 4.3 ± 6.5 PD, respectively. No subjects were overcorrected and 12 of 16 subjects (75.0%) had successful motor and sensory outcomes (Table 2, 3).

Comparison of surgical outcomes following medial rectus muscle recession between the two groups

Of 16 subjects, 12 (75%), presented both successful sensory and motor outcomes and 4 (25.0%), presented failed sensory and motor outcomes. Mean age at the initial visit was 28.1 ± 12.7 years in the success group and 25.6 ± 3.8 years in the failure group ($P = .709$). The duration of esodeviation was 2.3 ± 2.5 months in the success group and 2.4 ± 2.5 months in the failure group ($P = .975$). Mean preoperative maximum angle of esodeviation at distance was 29.5 ± 9.4 PD in the success group and 23.1 ± 8.5 PD in the failure group ($P = .254$). Mean preoperative maximum angle of esodeviation at near was 31.3 ± 11.3 PD in the success group and 20.5 ± 11.6 PD in the failure group ($P = .123$).

The postoperative deviation at 1 week was -0.4 ± 2.0 PD in the success group and 1.8 ± 1.3 PD of esodeviation in the failure group ($P = .048$). The postoperative deviation at 1 year was 1.0 ± 2.0 PD in the success group and 14.3 ± 4.3 PD in the failure group ($P < .001$, Table 4).

There were two patients who had a postoperative adjustment because of persistent diplopia. One patient in the success group presented orthotropia immediately after surgery, and 6PD 5 days after surgery and complained of diplopia. We further recessed the medial rectus muscle about 1mm in this patient. Another patient was in the failure group. This patient also presented 8PD 5 days after surgery complaining of diplopia, therefore, we further recessed the medial rectus about 1.5mm. There were no complications of postoperative adjustment in these two patients.

Amount of medial rectus muscle recession

Patients in both the success and failure groups needed more surgical medial rectus muscle recession than those indicated by the Parks' surgical table (formulated for typical esotropia correction) to achieve orthotropia. The success group needed more recession than the failure group (Fig. 1). On average,

Table 2 Data of individual patients with acute acquired comitant esotropia

Patients (age/sex)	Surgical amount of recessed MR muscle	Method of anesthesia	Laterality	Preoperative angle of esodeviation	Postoperative angle of deviation
1 (17/M)	R) 5.5mm, L) 3mm	Local	Both	15PD	Ortho
2 (20/F)	R) 4mm, L) 4mm	General	Both	23PD	10PD eso
3 (64/M)	R) 5.5mm, L) 6.5mm	Local	Both	28PD	2PD eso
4 (23 /M)	R) 6mm, L) 6mm	Local	Both	30PD	Ortho
5 (17/M)	R) 5mm, L) 5mm	General	Both	30PD	4PD eso
6 (27/F)	R) 4.5mm, L) 6mm	General	Both	37PD	20PD eso
7 (20/M)	R) 7mm, L) 7mm	General	Both	35PD	Ortho
8 (33/F)	R) 6.5mm, L) 5mm	General	Both	35PD	5PD eso
9 (25/M)	R) 8mm, L) 5mm	General	Both	50PD	1PD eso
10 (27/F)	L) 5mm	Local	Left	16PD	15PD eso
11 (17/M)	R) 8mm, L) 6.5mm	Local	Both	43PD	Ortho
12 (30/F)	R) 8mm, L) 8mm	Local	Both	25PD	Ortho
13 (31/F)	R) 6mm, L) 5mm	Local	Both	20PD	2PD eso
14 (27/M)	R) 7mm, L) 6.5mm	Local	Both	25PD	2PD exo
15 (19/M)	R) 8.5mm, L) 7.5mm	Local	Both	45PD	Ortho
16 (19/M)	L) 6.5mm	General	Both	15PD	12PD eso

Abbreviations: MR medial rectus, PD prism diopters

Table 3 Demographic and clinical characteristics of adult patients with acute acquired comitant esotropia

	Total (n=16)
Age at presumed onset (years)	25.3 ± 9.2 (range 17.0~53.5)
Duration of esodeviation (months)	2.4 ± 2.5 (range 0.3~10.0)
Age at initial visit (years)	27.5 ± 11.0 (range 17.4~63.5)
Age at surgery (years)	28.2 ± 11.0 (range 17.6~64.4)
Sex (male:female)	10 : 6
Cycloplegic refraction (diopters)	
Right eye	-2.66 ± 3.20 (range -9.75~+0.75)
Left eye	-2.44 ± 2.63 (range -6.75~+1.75)
Preoperative stereopsis (Log arcsec)	2.68 ± 0.89 (range 1.60~3.78)
Preoperative maximum angle of deviation (PD)	
Distant	27.9 ± 9.3 (range 12.0~45.0)
Near	28.6 ± 12.0 (range 12.0~50.0)
Methods of anesthesia (general : local)	7 : 9
Postoperative deviation (PD)	
Immediate after surgery	1.1 ± 3.7 (range -5.0~12.0)
After 1 week	0.1 ± 2.0 (range -4.0~4.0)
After 3 months	1.8 ± 2.8 (range -2.0~8.0)
After 6 months	2.9 ± 4.3 (range -2.0~14.0)
After 1 year	4.3 ± 6.5 (range -2.0~20.0)
Sensory outcome: elimination of diplopia (n)	12 (75.0%)
Motor outcome: orthophoria or esodeviation ≤ 8 PD (n)	12 (75.0%)

Continuous variables are reported as mean ± standard deviation

Abbreviations: PD prism diopters, N number

adults with acute acquired comitant esotropia required an additional 1.4 ± 1.1 mm of medial rectus muscle recession than listed in the Parks' surgical table, 32.4 ± 26.6% augmentation. The success group augmented the surgical amount by 40.6 ± 25.8 % and the failure group augmented by 7.9 ± 6.9 % (*P* = .028, Table 5).

Table 4 Comparison of clinical factors of adult patients with acute acquired comitant esotropia between success and failure group

	Success (n=12)	Failure (n=4)	*P* value
Age at presumed onset (years)	26.0 ± 10.4	23.2 ± 4.4	.613[a]
Duration of deviation (months)	2.3 ± 2.5	2.4 ± 2.5	.975[a]
Age at initial visit (years)	28.1 ± 12.7	25.6 ± 3.8	.709[a]
Age at surgery (years)	28.7 ± 12.7	26.5 ± 2.7	.743[a]
Sex (male:female)	9 : 3	1 : 3	.118[b]
Cycloplegic refraction (diopters)			
Right eye	-3.06 ± 3.17	-1.44 ± 3.40	.397[a]
Left eye	-2.71 ± 2.74	-1.63 ± 2.42	.495[a]
Preoperative maximum deviation angle (PD)			
Distance	29.5 ± 9.4	23.1 ± 8.5	.254[a]
Near	31.3 ± 11.3	20.5 ± 11.6	.123[a]
Methods of anesthesia (general : local)	4 : 8	3 : 1	.262[b]
Postoperative deviation (PD)			
Immediate after surgery	1.0 ± 4.0	1.5 ± 3.0	.824[a]
After 1 week	-0.4 ± 2.0	1.8 ± 1.3	.048[a]
After 3 months	0.7 ± 2.0	5.0 ± 2.5	.021[a]
After 6 months	1.0 ± 1.9	8.8 ± 3.9	.014[a]
After 1 year	1.0 ± 2.0	14.3 ± 4.3	<.001[a]

Continuous variables are reported as mean ± standard deviation

Abbreviations: PD prism diopters

[a]Mann-Whitney test, [b]Fisher's exact test

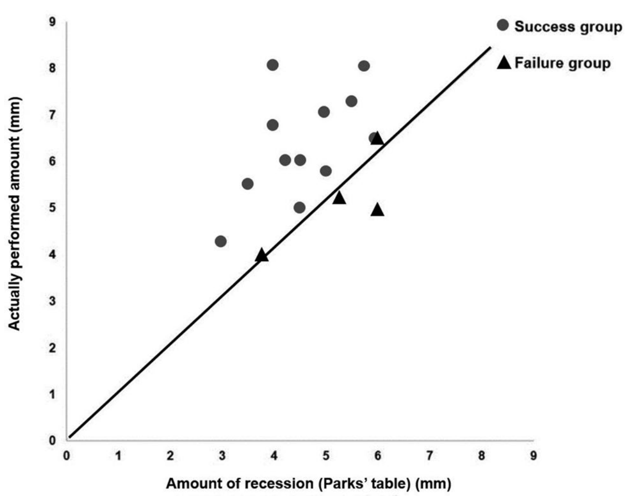

Fig. 1 Comparison of surgical amount of medial rectus muscle recession between two groups. Patients in both the success and failure groups needed a greater surgical amount of medial rectus muscle recession than that indicated by the Parks' table (formulated for typical esotropia correction) to achieve orthotropia. The success group increased more surgical amounts than those in the failure group

Discussion

In patients with AACE, surgery is attempted and the most common procedure is medial rectus muscle recession. However, following bilateral medial rectus recession or medial rectus recession/lateral rectus resection recurrence is common [16, 17]. In the present study, we evaluated 16 adult patients with AACE. Twelve of these 16 patients (75.0%) had successful motor and sensory outcomes.

Spierer [9] analyzed acute concomitant esotropia in patients older than 16 years of age. The 10 subjects had a mean refractive error of -4.1 ± 3.2 D, and almost all had myopia. He, therefore, concluded that myopia is a certain feature of acute concomitant esotropia in adulthood. In the present study, all subjects complained of sudden onset of binocular diplopia in the absence of any definite systemic or ophthalmic causes. Our subjects had a mean refractive error of -2.55 ± 2.92 D (range, 9.75 to +1.75 D), similar to that found by Spierer. Of the total 16 subjects examined here, only 1 could be considered to have high myopia (subject refractive error = -9.75 D). High myopia can cause esodeviation, but brain magnetic resonance imaging on this patient did not reveal any evidence of myopic strabismus fixus, ocular abnormalities, or extraocular muscle abnormalities.

Fresina et al. [18] report that AACE in myopic patients (Bielschowsky type) may be caused by a convergence spasm that is often secondary to a myopic overcorrection. Myopic overcorrection can increase induced hyperopia for near vision, which would then be dominated by an excess in accommodation and, subsequently, convergence. Because convergence cannot be relaxed during distance fixation, esodeviation remains. In addition, diplopia is often worse

Table 5 Surgical amount of medial rectus muscle recession in adults with acute acquired comitant esotropia. The amount of recession listed in the Parks' surgical table[a] (A) and actually performed recession amount in adults with acute acquired comitant esotropia (B) were compared

	Total	Success (n=12)	Failure (n=4)	*P* value
Response to surgery (PD / mm)	4.9 ± 1.5	5.0 ± 1.3	4.5 ± 2.1	.611[b]
Additional surgical amount (Difference between A and B, mm)	1.4 ± 1.1	1.8 ± 1.0	0.4 ± 0.4	.025[b]
Augmentation ((B-A) / A (%))	32.4 ± 26.6	40.6 ± 25.8	7.9 ± 6.9	.028[b]

Continuous variables are reported as mean ± standard deviation

Abbreviations: PD prism diopters

[a]Recommendation of Parks et al., [b]Mann-Whitney test

in adults with presbyopia. In these patients, cycloplegics can be used to interrupt accommodation in the early stages of AACE. Surgery can then be performed once symptoms stabilize. Furthermore, they propose a new preoperative assessment for esotropia, which stimulates accommodative convergence, allowing surgeons to plan wider bilateral medial rectus muscle recessions and improve surgical outcomes. Other previous studies also report that accommodative spasm may be related to acute acquired comitant esotropia [19, 20]. Even if subjects in the current study were not classified as having the Bielschowsky type of esotropia, accommodative convergence could be associated with acute acquired comitant esotropia.

Fusional amplitude has also been associated with AACE. Ali et al.[21] report young adults with gradually progressive intermittent, binocular diplopia that suddenly converted to constant, comitant, large-angle esotropia. They considered these cases to be the results of decompensation of simple esophoria, which slowly deteriorates and presents with esotropia only when enhanced divergence fusional amplitudes no longer suffice. Lyons et al.[8] also present 10 patients with AACE reporting that decompensation of a preexisting phoria or monofixation syndrome were the commonest etiologies. In the present study, patients with decompensated esophoria owing to decline in fusional amplitudes could be included.

Brodsky et al. [22] describe it as a tenacious distance fusion and phenomenon of "eat up prisms" which allows the examiner to build the measured esodeviation slowly with prism adaptation. It initially masks esodeviation, which becomes manifest only after a long period. Savino et al. [23] report that the prism adaptation test was positive in 10 of 14 patients (71%) between 4 to 40 years of age with acquired comitant esotropia. They state that there could be a variation in muscle tonus when retinal images were displaced by wearing prisms. Repka et al. [24] demonstrate that prism adaptation significantly improved 1-year motor outcomes in prism responders with acquired esotropia older than 3 years of age. However, they also report that increasing rectus muscle recession amount over that suggested by a previous surgeon's surgical tables helps achieve favorable results by avoiding the time and expense associated with prism adaptation [25]. The prism adaptation test was only performed on one of the subjects in our success group, likely because it is difficult to perform this test in adults with diplopia. Furthermore, wearing prism glasses over several months in adults is uncomfortable [26]. Therefore, we did not perform the prism adaptation test. Instead, we performed a medial rectus muscle recession under local anesthesia, which lead us to conclude that these adult patients required a greater amount of surgical correction than those with other types of esotropia (recession amount needed larger than that listed in Parks' surgical table for treating typical esotropia). Therefore, in patients who underwent the procedure under general anesthesia, we should have increased the amount of surgical correction to achieve orthotropia just after surgery.

In the failure group, the chance of esodrift increased as residual esodeviation immediately after surgery increased. The residual esodeviation just after surgery was also significantly larger in the failure group than in the success group. Therefore, aiming for orthotropia following surgery is an important factor. Additionally, our subjects in both the success and failure groups underwent procedures with a recession amount larger than that suggested by the Parks' table; success group augmented 40.6 ± 25.8 % and failure group augmented 7.9 ± 6.9 %. Therefore, increasing the recession amount was necessary in these patients with AACE to achieve favorable surgical outcomes.

Our study had some limitations, including its retrospective design and small sample size. Additionally, the prism adaptation test was not performed on all subjects as described above. However, there are recent studies reporting that prism adaptation test taking half an hour to several hours is enough. Especially, Akbari MR et al.[26] most recently reported that short prism adaptation test repeated every 20 minutes was sufficient to find the endpoint of motor stability. Therefore, further study including the prism adaptation test and homogenous surgical group would be necessary. Lastly, our subjects were followed for a minimum period of 1 year.

Further studies with longer-term follow-up including other types of esotropia in adults should be performed.

To the best of our knowledge, this is the first study to evaluate clinically relevant factors, surgical amount of medial rectus muscle recession and outcomes in adults with AACE. We recommend increasing the surgical amount of medial rectus muscle recession and targeting postoperative orthotropia to achieve favorable results in adults with acute acquired comitant esotropia.

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