The Prevalence of Reoperation and Related Risk Factors Among Patients With Congenital Esotropia

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ABSTRACT

Purpose: To determine the prevalence rate and related risk factors of reoperation among patients with congenital esotropia.

Methods: One hundred fifty-seven children with congenital esotropia were divided into two groups after at least one operation: children with deviation within 10 PD (n = 89; success group) and those with deviation greater than 10 PD or history of reoperation (n = 68; failure group). The relationship of risk factors such as age at first operation and primary congenital esotropia of less than 30 or more than 50 PD and accompanying factors such as inferior oblique muscle overaction (> +1), dissociated vertical deviation, lateral rectus muscle underaction, and A-V pattern with reoperation were studied. Final sensory status of children 5 years and older was evaluated by Worth 4-dot and Titmus tests.

Results: Reoperation was indicated in 32.4% of children who had residual esotropia greater than 15 PD after 3 months following their first operation. Congenital esotropia greater than 30 PD ($P = .002$) and lateral rectus muscle underaction of -1 to -2 ($P < .005$), were statistically different between the two groups. Initial operation at younger than 3 years was more likely to achieve gross stereopsis in children 5 years and older ($P = .032$).

Conclusion: Congenital esotropia greater than 30 PD and lateral rectus muscle underaction were found to be risk factors of reoperation.

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INTRODUCTION

Congenital esotropia is the most common eye deviation with an incidence of 1% to 2% of the population[1] and is defined as constant esodeviation with onset prior to 6 months of age. The treatment of choice is surgery, and there is a possibility of reoperation in 19% to 69% of patients according to different reports.[1-6] The risk factors for reoperation are not well known, but age younger than 15 months at first surgery, congenital esotropia greater than 30 or 50 prism diopters (PD), accompanying inferior oblique muscle overaction, dissociated vertical deviation, latent nystagmus, amblyopia, and positive family history of esotropia have been introduced as the risk factors of reoperation, with no consensus on all of them.[1-3]

Although it is generally accepted that final alignment should be achieved before 2 years of age,[1] some surgeons have recommended surgery at a younger age as soon as possible after the onset of stable esotropia for better sensory results. In addition, surgery before 15 months of age has been reported to be associated with higher frequency of reoperation,[1,6] possibly due

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to instability of esotropia, amblyopia, and difficult measurement of deviation.[1] The current recommended age of first surgery for congenital esotropia in the United States is approximately 1 to 1.5 years, whereas in many European countries it is 2 to 3 years of age.[6]

A multicenter European study of early versus late infantile strabismus surgery on 532 children showed 13.5% gross stereopsis with Titmus test at the age of 6 years among those who had surgery at approximately 20 months of age compared with 3.9% of those who had surgery at approximately 49 months of age (P = .001), with reoperation rates of 28.7% and 24.6%, respectively.[6]

There are some standard tables for determining the type and amount of surgery in deviations less than 50 PD with an average success rate of 80%; by increasing the deviation (> 50 PD), this rate will decrease to 37%.[7] Simultaneous surgery on three or four muscles can raise the success rate to 65%, but there is also a 16.7% danger of overcorrection.[8] Large bimedial rectus recession of 7 or 8 mm with the advantages of shorter surgery time and the saving of lateral recti for possible further operations has been reported to have an equal success rate to surgery on three or four muscles,[9] but it may result in adduction limitation and consecutive exotropia after an average of 2 to 3 years of follow-up in 27% of cases.[10]

In this study, we evaluated the rate of reoperation and its main risk factors and final sensory status among patients with congenital esotropia who were referred to our center from 2001 to 2011.

PATIENTS AND METHODS

This retrospective study was performed on 157 children with congenital esotropia from 2001 to 2011. The study was approved by the institutional review board. Informed consent was obtained from the parents or patients who participated in this research. The study and data collection conformed to all local laws and complied with the principles of the Declaration of Helsinki.

All children with esotropia before 6 months of age and a history of at least one operation entered the study. All surgeries were planned within 1 to 2 weeks after stabilization of esotropia, according to the standard Parks table.[7] The first operation was routinely bimedial rectus recession (up to 6 mm) or recession-resection of the amblyopic non-dominant eye if there was strong fixation preference. Patients with congenital cataract, retinopathy of prematurity, ocular anomalies, systemic and neurologic disorders, and those whose previous information had been lost or had follow-up of less than 3 months were excluded.

Epidemiologic characteristics such as age, gender, age at first operation, family history of eye deviation, and the results of their eye examinations, including cyclorefraction (in cases with > +3.5 D hyperopia, prescription of corrective glasses were considered at least 2 to 4 weeks preoperatively), amblyopia (best-corrected visual acuity < 20/30 or fixation preference), deviation angle (in primary position at far or near with Krimsky or prism alternate cover test), lateral rectus muscle underaction (-1 to -3), inferior oblique muscle overaction (> +1), dissociated vertical deviation, A- or V-patterns (> 15 PD difference between up and down gaze deviation), latent nystagmus, fundus examination, type and amount of surgery in previous operation(s), and follow-up, were recorded from their previous files.

In a follow-up period of 3 months after the first operation, children with deviation within 10 PD were considered as the success group (n = 89) and those with deviation greater than 10 PD or history of reoperation were regarded as the failure group (n = 68). Only surgeries on horizontal rectus muscles were regarded as reoperation in this study. If postoperative alignment was between 10 and 15 PD, we did not recommend further surgery. Postoperative deviation greater than 15 PD was an indication for reoperation. Final sensory status of children 5 years and older was evaluated by Worth 4-dot and Titmus tests.

The relationship of factors such as age at first operation, deviation size of congenital esotropia, amblyopia, inferior oblique muscle overaction, dissociated vertical deviation, A-V pattern, and lateral rectus muscle underaction with reoperation were studied.

Statistical Analysis

To describe data, we used mean ± standard deviation, median (range), and frequency (%). To evaluate the differences between groups, we used the chi-square, Fisher exact, Mann–Whitney U, and t tests. To consider the bilaterally of eyes in comparisons, we used the generalized estimating equation. All statistical analysis was performed using SPSS software (version 17.0; SPSS, Inc., Chicago, IL).

RESULTS

This study was conducted on 157 patients with congenital esotropia coming to Imam Hossein Med-

TABLE 1

Preoperative Characteristics of Patients With Congenital Esotropia in the Success (89 Patients, 178 Eyes) and Failure (68 Patients, 136 Eyes) Groups[a]

Variable	Total	Success	Failure	*P*
Mean age, y	5.4 ± 8	4.5 ± 7.1	7.3 ± 9.5	.099[b]
Median age, y (range)	2.7 (0.3 to 46.2)	2.3 (0.3 to 46.2)	2.9 (0.3 to 39.7)	
Sex				
Female	86 (54.8)	48 (53.9)	38 (55.9)	.808[c]
Male	71 (45.2)	41 (46.1)	30 (44.1)	
Family history of strabismus	34 (21.7)	19 (21.3)	15 (22.1)	.915[c]
Prematurity	15 (9.6)	9 (10.1)	6 (8.8)	.785[c]
Plano (-1 to +1 D)	70 (22.3)	36 (20.2)	34 (25.0)	.615[c]
Hyperopia (D)				
> +3.5	71 (22.6)	53 (29.8)	18 (13.2)	.012[d]
> +2	154 (49.0)	94 (52.8)	60 (44.1)	.265[d]
>+1	230 (73.2)	135 (75.8)	95 (69.9)	.374[d]
Myopia (> -1 D)	14 (4.5)	7 (3.9)	7 (5.1)	.576[e]
Astigmatism (> ± 1.5 D)	41 (13.1)	19 (10.7)	22 (16.2)	.044[d]
Anisometropia (S or C > 1.5 D)	12 (7.7)	6 (6.8)	6 (8.8)	> .641[e]
Amblyopia (VA < 20/30)	60 (19.1)	35 (19.7)	25 (18.4)	.236[c]
Inferior oblique overaction	111 (35.4)	58 (32.6)	53 (39.0)	.134[c]
Dissociated vertical deviation	8 (5.1)	6 (6.7)	2 (2.9)	.467[e]
Latent nystagmus	5 (3.2)	4 (4.5)	1 (1.5)	.390[e]
V-pattern	20 (12.7)	8 (9.0)	12 (17.6)	.107[c]
A-pattern	2 (1.3)	2 (2.2)	0 (.0)	.506[e]
Lateral rectus underaction (-1 to -2)	73 (23.2)	27 (15.2)	46 (33.8)	.005[d]

Success = deviation ⩽ 10 prism diopters with one operation; failure = deviation > 10 prism diopters or more than one operation; D= diopters; S = sphere; C = cylinder; VA = visual acuity.
[a]Results are presented as mean ± standard deviation, median (range), frequency (%).
[b]Mann–Whitney *U* test.
[c]Chi-square test.
[d]Based on generalized estimating equation.
[e]Based on Fisher exact test.

ical Center during the past 10 years (2001 to 2011). Eighty-nine children were in the success group and 68 in the failure group.

Surgery types were bimedial rectus recession (140 patients), recession-resection (13 patients), three muscles or more (3 patients), and not specified (1 patient). Seventeen patients (10.8%) underwent an inferior oblique weakening procedure simultaneously.

The rate of reoperation in our study was 32.4% (51 of 157). Two patients underwent three operations. A third operation was also indicated in six other patients with last eye deviation greater than 15 PD, but their parents were satisfied with the results of the second surgery. Therefore, the rate of clinically needed third operation was 5.1% (8 of 157) with a mean follow-up period of 38.3 ± 40 months and median of 24 months.

Epidemiologic findings related to parents and children were not statistically significant in this study (Table 1).

Hyperopia +3.5 D or less, myopia greater than -1 D, astigmatism greater than 1.5 D, anisometropia greater than 1.5 D, and amblyopia were similar between the two groups. Approximately 22% of our cases demonstrated hyperopia 3.5 D or greater (*P* = .012) (Table 1). Lateral rectus muscle underaction ranging from -1 to -2 was seen in 33.8% and 15.2% of patients in the failure and success groups, respec-

TABLE 2

Operative Characteristics of Patients With Congenital Esotropic Patients in the Success (89 Patients, 178 Eyes) and Failure (68 Patients, 136 Eyes) Groups

Variable	Total	Success	Failure	*P*
Age at first operation (mo)				
Mean ± SD	55 ± 82	42 ± 46	72 ± 111	
Median (range)	28 (5 to 554)	28 (6 to 301)	33 (5 to 554)	.456[a]
⩽ 12	37 (23.6)	18 (20.2)	19 (27.9)	.259[b]
⩽ 24	68 (43.3)	38 (42.7)	30 (44.1)	.859[b]
⩽ 36	91 (58.0)	53 (59.6)	38 (55.9)	.645[b]
⩽ 48	108 (68.8)	62 (69.7)	46 (67.6)	.787[b]
⩽ 60	119 (75.8)	70 (78.7)	49 (72.1)	.339[b]
Deviation of first operation (PD)				
Mean ± SD	46 ± 13	44 ± 11	49 ± 14	.010[c]
Median (range)	45 (10 to 86)	40 (18 to 80)	50 (10 to 86)	
< 30	7 (4.5)	5 (5.6)	2 (2.9)	.700[d]
< 50	89 (56.7)	60 (67.4)	29 (42.6)	.002[b]
⩾ 50	68 (43.3)	36 (34.0)	32 (62.7)	.002[b]
BMR recession in first operation	140 (89.7)	83 (93.3)	57 (85.1)	.059[b]

Success = deviation ⩽ 10 prism diopters with one operation; failure = deviation > 10 prism diopters or more than one operation; PD= prism diopters; SD = standard deviation; BMR = bimedial rectus.
[a]Mann–Whitney *U* test.
[b]Chi-square test.
[c]Based on *t* test.
[d]Based on Fisher exact test.

tively (*P* = .005). The rates of inferior oblique overaction, V- or A-pattern, dissociated vertical deviation, and latent nystagmus were also similar in both groups. There was no difference between the groups regarding age at first operation, and the reoperation rate among the failure group was constantly reduced with increasing age (Table 2). The esotropia angle was higher in the failure group compared to the success group (49 ± 14 vs 44 ± 11 PD, *P* = .010) and esotropia greater than 30 PD was considered a risk factor for reoperation (*P* = .002) (Table 2). Finally, 79% of all patients found motor success within 10 PD, 15.9% showed borderline results between 10 and 15 PD, and 5.1% of patients showed deviation greater than 15.

The percentages of patients with undercorrection (esotropia > 10 PD) and overcorrection (exotropia > 10 PD) were 17.1% and 3.9%, respectively. Comparison of these two groups did not show any statistically significant difference regarding the age at first operation (esotropia: 53 ± 82 vs exotropia: 51 ± 35 months, *P* = .328), initial congenital esotropia angle (esotropia: 50 ± 12 vs exotropia: 47 ± 1 PD, *P* = .484), and amblyopia (esotropia: 11 vs exotropia: 3 patients, *P* > .999).

Postoperative gross stereopsis with Titmus test (30%) and fusion with Worth 4-dot test (63%) had similar results between the two groups (Table 3). All patients 5 years or older at their last visit who were expected to be able to respond to the Titmus test were compared in terms of stereopsis. The results were similar if the age at first operation was younger than 3 years, whereas fewer older patients with gross stereopsis (66.1%) were seen without considering the number of operations (Table 4). As can be seen in the decision tree, the most appropriate age for the first operation is 37 months, if a minimum number of operations and achieving gross stereopsis in patients older than 5 years old is desired (Figure 1).

DISCUSSION

According to our results, motor success was achieved in 79% of children with undercorrection and overcorrection of 17.1% and 3.9%, respectively.

Reoperation was needed in 32.4% of patients, with risk factors being congenital esotropia greater than 30 PD and lateral rectus muscle underaction. Age at first surgery younger than 3 years would result in gross stereopsis in 5-year-old children.

Trigler and Siatkowski reported a 34% rate of reoperation in 149 patients with congenital esotropia[1] and Castro et al. had a 35.4% reoperation rate in their study,[11] both of which were similar to our findings.

Vroman et al. reported 19% and 25% reoperation rates in 56 patients with congenital esotropia less than 50 PD and esotropia greater than 50 PD, respectively, which is better than our results.[2] The amount of bimedial rectus recession in patients with esotropia greater than 50 PD was 6.5 to 7 mm in their study, which was higher than the Parks standard table used in our study; therefore, it is expected that they would have lower reoperation rates.[2]

In Simonsz et al.'s study, 60% to 80% of children who had surgery at 20 months of age and 25% of children who had surgery at 49 months of age needed reoperation.[6] This rate was 44% in patients younger than 24 months and 42% in patients younger than 48 months in our study.

In our study, the mean deviation of congenital esotropia was different between the two groups (*P*

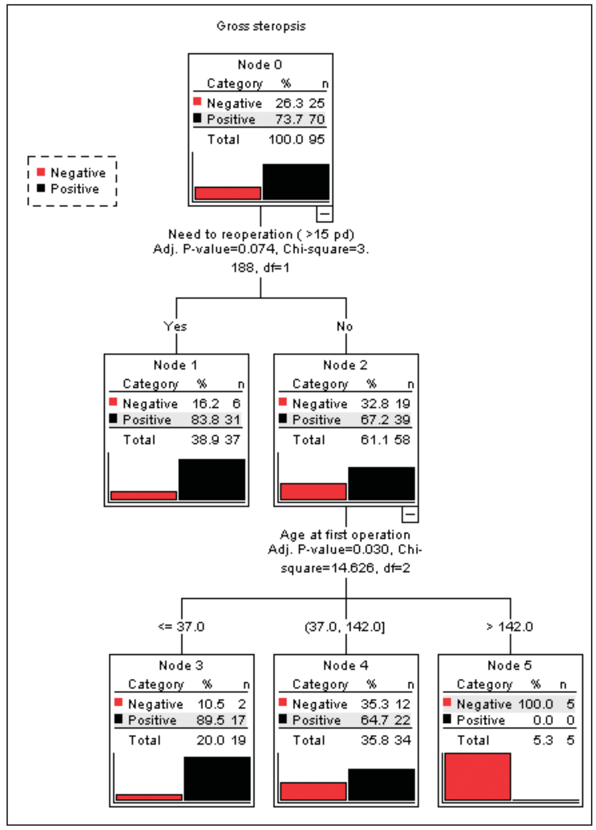

Figure 1. The most appropriate age at operation to achieve gross stereopsis at 5 years old was 37 months or younger with minimum number of operations in this study.

TABLE 3

Postoperative Characteristics of Patients With Congenital Esotropia in the Success (89 Patients, 178 Eyes) and Failure (68 Patients, 136 Eyes) Groups

Variable	No. of Available Patients	Total	Success	Failure	*P*
BCVA (LogMAR)					
Mean ± SD	234 eyes	0.19 ± 0.23	0.2 ± 0.2	0.18 ± 0.26	.585[a]
Median (range)		0.1 (0 to 1.6)	0.1 (0 to 0.7)	0.1 (0 to 1.6)	
VA < 20/30		71 (30.3)	41 (34.2)	30 (26.3)	.136[a]
Worth 4-dot test					
Suppression	115 patients	42 (36.5)	21 (35.0)	21 (38.2)	.723[b]
Fusion		73 (63.5)	39 (65.0)	34 (61.8)	
Steropsis (arc sec)					
⩽ 3,000	117 patients	82 (71.1)	39 (65.0)	43 (75.5)	.218[b]
> 3,000		35 (29.9)	21 (35.0)	14 (24.6)	
Amblyopia (< 20/30)					
Yes	117 patients	60 (51.3)	35 (58.3)	25 (43.9)	.117[b]
No		57 (48.7)	25 (41.7)	32 (56.1)	

Success = deviation ⩽ 10 prism diopters with one operation; failure = deviation > 10 prism diopters or more than one operation; BCVA = best-corrected visual acuity; LogMAR = logarithm of the minimum angle of resolution; SD = standard deviation; VA = visual acuity.
[a]Based on generalized estimating equation.
[b]Chi-square test.

TABLE 4

No. of Patients Older Than 5 Years Who Achieved Gross Stereopsis According to Age at First Operation (70 of 95 Patients)

Time of First Operation (Y)	< Age at First Operation	> Age at First Operation	*P*
1	11 (78.6)	59 (72.8)	.653[a]
2	21 (84.0)	49 (70.0)	.172[b]
3	31 (86.1)	39 (66.1)	.032[b]
4	40 (83.3)	30 (63.8)	.031[b]
5	49 (84.5)	21 (56.8)	.003[b]

Success = deviation ⩽ 10 prism diopters with one operation; failure = deviation > 10 prism diopters or more than one operation.
[a]Based on Fisher exact test.
[b]Chi-square test.

= .010). Additionally, esotropia greater than 30 PD was considered a risk factor for reoperation, which is similar to Trigler and Siatkowski's results but lower than Vroman et al.'s cut-off of 50 PD.

Poor infant cooperation, instability of the eye deviation, and accompanying motor disorders might interfere with accurate measurement of esotropia, resulting in different outcomes. In contrast to other studies, many of our patients presented after 2 years of age. The reasons could be limited parental awareness, unavailable strabismus subspecialists, low income, fear of surgery, and a hope for the spontaneous improvement of esotropia.

Undercorrection (17.1%) and overcorrection (3.9%) rates in our study were in line with previous studies, which have reported 10% to 21.4%[2,3] undercorrection and 5% to 27%[3,9,10] overcorrection. In 36 cases with mean deviation of 74 PD of congenital esotropia, Weakley et al. reported 75% motor success by bimedial rectus recession equal to 7 mm with 11% overcorrection,[9] a similar success rate but higher overcorrection rate compared to our study. Stager et al. studied 88 patients with large-angle congenital esotropia by the same method and reported 27% overcorrection.[10] The lower age at first operation was related to the higher rate of overcorrection (38% in 7 months vs 20% in 12 months) with a mean follow-up of 2.3 years.

In Minkoff and Donahue's study, only 30% of 10 patients who simultaneously underwent surgery on three or four muscles for their large-angle congenital esotropia with a mean age of 13 months at the initial operation achieved alignment, with undercorrection and overcorrection of 10% and 60%, respectively.[12] Although better alignment with larger bimedial rectus recession or surgery on three or four muscles may be achieved, there is a danger of overcorrection. These authors believed that acceptable alignment in the sensitive period of binocular vision development is valuable and worth the possibility of further operation, but we believe its advantages and disadvantages should be discussed with the patient's parents before the surgery.

Trigler and Siatkowski had more inferior oblique muscle overaction in their case group (45% vs 35%), but similar to our results the difference was not statistically significant.[1] Twenty percent of patients with esotropia less than 50 PD and no patient with esotropia greater than 50 PD showed inferior oblique muscle overaction in Vroman et al.'s study.[2] In Keen and Willshaw's study, it was detected in 30% and 10% of patients before and after surgery, respectively. Dissociated vertical deviation was observed in 15% of patients, all diagnosed after surgery.[3] Although the detection of inferior oblique muscle overaction in an infant with severe esotropia is difficult, it has been reported in up to one-third of patients with congenital esotropia before surgery,[1-3] which is similar to our results.

Lateral rectus muscle underaction has not been mentioned as a risk factor of reoperation in other studies. Although most lateral rectus muscle underaction was mild and the patients' doll's movements were positive and improved by bimedial rectus recession surgery, it might show more contracture of medial rectus muscles that can induce abduction limitation and may be considered as a risk factor for reoperation.

Mild hyperopia (< +3.5 D) was the most frequent refractive error, consistent with the literature.[11] Hyperopia greater than +3.5 D was seen in 22.6% of our cases. These patients might also have early onset accommodative esotropia. They were not excluded from the study because their esotropia onset was before 6 months of age as reported by their parents, the same timing as congenital esotropia. Twenty-nine percent of patients in Trigler and Siatkowski's study also had hyperopia greater than +2 D, with the same probable diagnosis.[1] Simonsz et al. reported early onset accommodative esotropia in many monkeys with natural esotropia[6] and Gunton and Nelson also mentioned accommodative esotropia as an accompanying eye movement disorder in patients with congenital esotropia.[13]

In our study, increasing the age at first operation decreased the number of patients who achieved stereopsis. Surgeries at younger than 3 years of age had similar stereopsis results, whereas there was a statistically significant decrease if surgeries were performed after 3 years of age (P = .032) without considering the number of operations (Table 4). The appropriate age for the first operation to achieve gross stereopsis at 5 years of age or older with a minimum number of operations was estimated to be 37 months in our study, which is in line with the multicenter European study of early versus late infantile strabismus surgery[6] (Figure 1).

Ing and Rezentes reported 94% fusion in patients with less than 21 months of misalignment who achieved alignment by age 24 months,[14] which is higher than our findings (63%). The reason might be the younger age at operation and shorter duration of misalignment among their patients. Fifty-seven percent and 27% of children who achieved alignment before 6 months and from 7 to 15 months of age, respectively, achieved fusion in Gunton and Nelson's study.[13]

In the European study of early versus late infantile strabismus surgery, 13.5% and 3.9% of patients who had surgery at 20 and 49 months of age achieved gross stereopsis at 6 years of age, respectively,[6] which indicates a lower success rate compared to our result of 30% gross stereopsis at 5 years of age (Table 3).

In Murray et al.'s study of 17 patients with congenital esotropia who had late surgery and aligned within ± 8 PD with no amblyopia, 88% fusion with Bagolini test was seen but there was no fusion using Titmus and Worth 4-dot tests. They concluded that late surgical alignment will result in a lower degree of binocular function.[15] Morris et al. achieved fusion in 100% and 66% of 12 patients with congenital esotropia with the same condition using the Worth 4-dot and Titmus tests, respectively.[16] They suggested that some patients might have a period of alignment that the authors were not aware of, so there is a possibility of achieving fusion even with late surgery.

As was discussed, the amounts of postoperative stereopsis and fusion are not similar in different reports.[6,13-18] Age at first operation, postoperative alignment, amblyopia, accompanying extraocular muscle dysfunction, age, and type of stereopsis test are variables that can affect the degree of postoperative binocular function.

Our study had some limitations. For example, 22.6% of our patients with hyperopia greater than +3.5 D may have had early onset accommodative esotropia, which might cause better binocular function results. In addition, more than half of our patients had their initial operation after 2 years of age, with the advantage of more accurate deviation measurement but the possible disadvantage of less binocular function.

Our reoperation rate of 32.4% is similar to other reports. Congenital esotropia greater than 30 PD and lateral rectus muscle underaction were found to be risk factors of reoperation in the current study. Initial surgery at younger than 3 years of age was more likely to achieve gross stereopsis in children 5 years and older.

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