



ACUTE ACQUIRED COMITANT ESOTROPIA

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ABSTRACT

Acute acquired comitant esotropia (AACE) is an infrequent presentation of esotropia, characterized by sudden onset eye misalignment and diplopia. The purpose of this article is to review four cases of children diagnosed with AACE. We present 4 cases (children 6-10 years) of AACE. All of them suddenly developed diplopia and esotropia, with large-angle. After complete ophthalmological and neurological examinations in three cases out of four, strabismus surgery was conducted. In one case heterogenous neoplasm was detected and sent to an oncologist and neurosurgeon for evaluation and morphological verification. Bilateral medial rectus recession in two cases and unilateral recession of the medial rectus and resection of the lateral rectus in one case were performed. After strabismus surgery fully recovered binocular vision and was no longer noticeable diplopia. Neurological examinations and neuroimaging should be performed to exclude any potential intracranial disease. Surgery should be taken into consideration 6 months after the onset of the esotropia, if the deviation is stable. Strabismus surgery of AACE has good motor and sensory results and can successfully restore good binocular function. The excessive use of smartphones may be associated with developing and increasing AACE.

Acute acquired concomitant esotropia (AACE) is a rare, distinct subtype of strabismus. [4] It is not associated with accommodative effort, characterized by a sudden onset and diplopia. [6] Spontaneous recovery of AACE is unusual. [1]. AACE occurs in around 0.3% of children with strabismus. [12]

Historically, the precipitating aetiological factors observed among children and adults have suggested three categories of AACE (Burian & Miller 1958) [4]; AACE caused by interruption of fusion (type I) (Swan 1947) [17]; idiopathic AACE thought to be caused by physical and psychological stress (type II), (Franceschetti 1952) [18];

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and AACE caused by myopia (type III) (Bielschowsky1922). [2]. Case studies of selected groups of children and adults have suggested that hyperopia and intracranial disease may be additional causes of AACE. [4] Later, Helena Buch and Troels Vinding retrospectively investigated acute esotropia in children and reported that it could be classified into seven types according to the causes. In all, 48 cases were recorded. The mean age at onset was 4.7 years, being significantly higher among children with intracranial disease. Seven cause-specific types of AACE in childhood were identified: acute accommodative (n = 15, 31%), decompensated monofixation syndrome or esophoria (n = 13, 27%), idiopathic (n = 9, 19%), intracranial disease (n = 3, 6%), occlusion related (n = 3, 6%), AACE secondary to different aetiologic disease (n = 3, 6%) and cyclic AACE (n = 2, 4%). Intracranial disease included hydrocephalus, pontine and thalamic glioma. Of the children with intracranial disease, 2 of 3 had no obvious neurological signs at onset. Four significant risk factors for intracranial disease were identified as follows: larger esodeviation at distance, recurrence of AACE, neurological signs (papilledema) and older age at onset (>6 years). [7] The adoption of mobile technologies and wireless communication infrastructure is a global phenomenon. Among the existing technologies, smartphones have been one of the most prominent success stories of the last decade. In a relatively short period of time, smart mobile technology has significantly penetrated society in the Western world and globally. [8] The association between prolonged near work and esotropia has been noted

for more than 100 years, but has recently received more attention with digital device-related AACE. [13] The first case series on acute esotropia associated with excessive smartphone use was reported in 2016 by Lee *et al.*. [8] However, despite the large number of reported cases and data gathered, the clinical findings, etiology, management, and classification remain controversial. [9]

The main goal of the research was to analyze four cases of acute acquired concomitant esotropia in children.

We present four cases that were referred to our clinic with acute onset comitant esotropia. Comprehensive medical histories were taken, and we recorded onset, precipitating factors and associated symptoms. We assessed angle of the deviation using prismatic cover/ uncover and by synoptophore test, also by Hirschberg test, and performed a motility exam on all patients, to detect paresis and nystagmus also visual acuity and fusion potential was measured in all patients.

Case 1: An eight-year-old boy, with acute strabismus and diplopia that appeared as his mother said after using gadgets for at least 8 hours a day. Clinical features: visual acuity was 20/20 in each eye. The Angle of deviation by Hirschberg test is 20 degrees and by Krimsky test is 40PD, testing with a synoptophore +25 degrees. Cycloplegic refraction: a small accommodation spasm: OD sph -0,25 - cyl 0,5 ax 89; OS sph -0,5 - cyl 0,25 ax90. Binocular vision was evaluated by worth 4 dots, Synoptophore and Titmus test. Underwent fundus examination by direct and indirect ophthalmoscopy. The patient was evaluated by a pediatric neurologist and pediatrician. The patient

underwent vision therapy. After 6 months patient underwent surgical treatment; a recession/ resection procedure was performed in the right eye. His diplopia disappeared the first day after surgery.



Figure 1. Before and after surgery

He was orthotropic, regained binocular single vision for near and distance and stereopsis was normal (Fig. 1). Results were stable 6 months after surgery.

Case 2: A six years old boy, with acute onset of strabismus and diplopia. His mother says 1-2 weeks ago he had viral infection and also spend a lot of time with gadgets. Clinical Features: Movements of the eyeball is free in all directions. VA - RE was 20/20, diplopia. Angle of deviation by Hirschberg test is 25 degrees and by Krimsky test 45 PD, testing with a Synoptophore +25 degrees. Binocular vision was evaluated by worth 4 dots; Synoptophore and Titmus test. Dilation

was performed with cyclopentolate 1% , refraction: OD sph +1,25 cyl +0,25,25ax 89; OS sph +1,25 +cyl 0,5ax90. The patient underwent fundus examination by direct and indirect ophthalmoscopy. The patient was evaluated by a pediatric neurologist and pediatrician. After six months bilateral medial rectus recession was performed. After surgical intervention eyes position became symmetrical. (Fig2) No longer noticeable diplopia after surgical treatment , Stereopsis recovered.



Figure 2. Before and after surgery

Case 3: A 7 years old boy, myopic, with acute onset of strabismus and diplopia.

Clinical features: Muscle movements are not limited in all directions. Visual acuity: 20/20, Angle of deviation by Hirschberg test 25 degrees / by Krimsky test 50 PD and Synoptophore + 25 degrees. Dilation was performed with cyclopentolate 1% Refraction: OD sph

– 6,0 cyl – 1,25 ax180 OS sph– 5,5 cyl – 1,5ax170. Patient underwent fundus examination by direct and indirect ophthalmoscopy. The patient was evaluated by a pediatric neurologist and pediatrician. The participant, also underwent vision therapy program. After 6 months of conservative treatment, bilateral medial rectus recession was

performed. After surgical intervention eyes position became symmetrical by Hirschberg, Krinsky methods and

Synoptophore test. No longer noticeable diplopia after surgical treatment (Fig3). Stereopsis full recovered



Figure 3. Before and after surgery

Case 4: An 8 year-old boy with AACE and diplopia. Clinical features: Muscle movements are not limited in all directions. Visual acuity: 20/20 , Diplopia , Angle of deviation by Hirschberg test 25 degrees / by Krinsky test 55 P/ Synoptophore + 25 degrees. He had of a cycloplegic refraction OD sph +0,5 cyl +0,25 ax180 OS sph+1,0 cyl-0,25 ax170. Patient underwent fundus examination by direct and indirect ophthalmoscopy. Eye fundus was normal. In one week after the manifestation of strabismus the patient had episodes of vomiting and headache, he was evaluated as asthenic by neuropathologist and prescribed MRT scanning by which detected heterogenous neoplasm in the parasagittal right front lobe, primary neoplastic process of the brain. The patient was sent to the neurosurgical oncologists and neuro-oncologist, and for morphological verification.

Discussion

AACE is a special subtype of esotropia characterized by acute onset of comitant esotropia with diplopia and equal deviation in all gaze directions. [4,11, 16] Ocular motility is generally normal without evidence of paralysis of the extraocular muscles. [16] AACE is more

common in older children and adults. [5,11,16] The specific pathogenesis, mechanism and etiology of strabismus of this type is still not clear. [5, 10, 11, 15] Neurological diseases are considered to be associated with this type of strabismus [5] It was classified into three types by Burian and Miller. They are generally considered as having performed the initial pivotal work in the field of AACE. [4] Recently, AACE was associated with excessive smartphone use. [3, 8, 10] The mechanism was similar to that proposed by Bielschowsky.[2] In our study the first and second case of AACE was associated with excessive smartphone use. The third case was Bielschowsky type followed by Burian-Franceschetti, and the forth case was developed after intracranial disease. We can confirm that AACE of childhood has a small but significant association with intracranial disease. This calls for further studies with higher levels of evidence to fill the current gap in this area of science. [14] AACE should be investigated by the ophthalmologist and the neurologist. MRI should be performed to rule out intracranial disease, however, there is still controversy regarding the best timing for neuroimaging, since there is no single sign or symptom that can predict the presence of underlying intracranial pathology.

Conclusion: Neurological examinations and neuroimaging should be performed to exclude any potential intracranial disease. Surgery should be taken into consideration 6 months after the onset of the esotropia, if the deviation is stable. Strabismus surgery of AACE has good motor and sensory results and can successfully restore good binocular function. The excessive use of smartphones may be associated with developing and increasing AACE.

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ABSTRACT

ОСТРАЯ ПРИОБРЕТЕННАЯ СОДРУЖЕСТВЕННАЯ ЭЗОТРОПИЯ

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Острая приобретенная содружественная эзотропия (ААСЕ) – редкий тип эзотропии, который обычно встречается у детей старшего возраста. В статье проанализировано четыре случая ААСЕ у детей 6-10 лет. У всех детей внезапно развилась диплопия и эзотропия с большим углом косоглазия. В результате полного офтальмологического и неврологического обследования у одного пациента было обнаружено гетерогенное новообразование. Проведена морфологическая верификация опухоли. Трое пациентов были прооперированы. Двум пациентам была выполнена двусторонняя рецессия медиальной прямой мышцы одному – односторонняя рецессия медиальной прямой мышцы и резекция латеральной

прямой мышцы. В результате операции полностью восстановилось бинокулярное зрение; диплопия не наблюдалась. Результаты позволяют сделать вывод о высокой вероятности выздоровления стереопсиса (до 100%). Кроме того, чтобы исключить внутрочерепное заболевание всем пациентам с острой приобретенной содружественной эзотропией необходима консультация невролога и неврологическое обследование. Хирургическое вмешательство рекомендуется проводить не ранее 6 месяцев после возникновения косоглазия, при условии стабильного угла отклонения. Причиной острой приобретенной эзотропии называется интенсивная нагрузка на близком расстоянии.

Ключевые слова: острая приобретенная содружественная эзотропия, использование смартфона, бинокулярное зрение, хирургия косоглазия

რეზიუმე

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მწვავე შეძენილი შეუღლებული ეზოტროპია სიელმის ერთერთი იშვიათი ტიპია, იგი ძირითადად ვითარდება ბავშვთა და მოზარდთა ასაკში. სტატისტიკაში განხილულია მწვავე შეძენილი შეუღლებული ეზოტროპიის ოთხი შემთხვევა. ყველა აღნიშნულ შემთხვევაში სიელმე განვითარდა მოულოდნელად, დიდი გადახრის კუთხით, რასაც თან ერთვის დიპლოპია. სრული ოფთალმოლოგიური და ნევროლოგიური კვლევის შედეგად ერთ პაციენტს დაუდგინდა ჰეტეროგენული ნეოპლაზია, ჩატარდა მორფოლოგიური ვერიფიკაცია. 3 პაციენტს ჩატარდა ქირურგიული ჩარევა, აქედან ორს შიგნითა სწორი კუნთების რეცესია, ხოლო ერთს შიგნითა სწორი კუნთის რეცესია და გარეთა სწორი კუნთის რეზექცია. ქირურგიული მკურნალობის შედეგად სრულად აღგა ბინოკულური მხედველობა და დიპლოპია აღარ შეინიშნებოდა. ჩვენი დაკვირვებით ყველა პაციენტი საჭიროებს ნევროპათოლოგის კონსულტაციას, იმისათვის რომ გამოირიცხოს ინტრაკრანიალური დაავადება. ქირურგიული ჩარევა სასურველია ჩატარდეს სიელმის დაწყებიდან არა ნაკლებ 6 თვისა, იმ შემთხვევაში, როდესაც გადახრის კუთხე სტაბილურია. აღსანიშნავია რომ, ქირურგიული მკურნალობის შედეგად სტერეოფისის აღდგენის პოტენციალი მაღალია, თითქმის 100% ში. ლიტერატურულ მონაცემებზე დაყრდნობით და ჩვენი დაკვირვებით მწვავე შეძენილი ეზოტროპიის მიზეზი შესაძლოა გახდეს ინტენსიური დატვირთვა ახლო მანძილზე.

საკვანძო სიტყვები: მწვავე შეძენილი შეუღლებული ეზოტროპია, სმარტფონის გამოყენება, ბინოკულარული მხედველობა, სიელმის ქირურგია