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Case Report

Transient Esotropia in the Child: Case Report and Review of the Literature

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Keywords

Transient esotropia · Acute acquired concomitant esotropia · Ophthalmoplegic migraine · Accommodative spasm

Abstract

The aim of this report is to investigate the possible causes of acute acquired onset of transient esotropia (AATE) in children and to help to differentiate ophthalmoplegic migraine (OM) from accommodative spasm (AS). A case of an 8-year-old Caucasian female affected by AATE and diplopia is described. The day before AATE onset, the patient complained of slight headache without nausea and vomiting, with spontaneous resolution. AATE diagnosis is challenging. The most likely ophthalmological causes of AATE are AS and OM. In these cases it is important to evaluate the presence of both a familial history of recurrent headaches and an

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AATE associated with migraine, ptosis, nausea, and vomiting. A full ophthalmological evaluation and a thorough refractive examination in cycloplegia are mandatory to exclude ophthalmological causes.

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Introduction

An ophthalmological evaluation is mandatory to get a correct diagnosis in a case of acute acquired onset of concomitant esotropia (AACE) with diplopia [1]. AACE workup implies numerous instrumental examinations with high costs and stress for the child. The absence of clear ophthalmological and neurological causes leads to a diagnosis of exclusion.

We describe a complex case of a child of 8 years affected by a short duration of diplopia and esodeviation (from 2 to 4 h), so that signs are not directly recognized by the ophthalmologist, but just reported by the child, parents, or other health care providers.

The peculiarity of our case is the short duration of strabismus and diplopia and their spontaneous regression. Being transient, it cannot be classified among AACEs; we therefore considered it as a different entity which we have called acute acquired onset of transient esotropia (AATE).

AATE is probably due to ophthalmoplegic migraine (OM) or accommodative spasm (AS). This is the first report in the literature that differentiates AATE from AACE, trying to investigate possible causes, the diagnostic algorithm, and potential treatments.

Case Report

An 8-year-old Caucasian female was evaluated in the outpatient clinic of pediatric ophthalmology of Melegnano Hospital, after she had been hospitalized 1 week before in another hospital for AATE. The child was born at term with vaginal delivery, she had normal mental and motor growth, and she was in good health.

The presentation of symptoms first occurred at school: the patient suddenly complained of double vision, and the teachers noticed esodeviation of the left eye (LE). Three hours later, the patient was accompanied by her father to the nearest emergency department, but on the way, diplopia and strabismus regressed. Symptoms reappeared only when the child was solicited, first by her father in the waiting room and then during the pediatric visit, to fix on a near target. In these cases, diplopia and esotropia regressed in about 15 min.

During the ophthalmological evaluation both diplopia and LE esotropia were no longer present. The patient had a best corrected visual acuity of 20/20 in both eyes using her own spectacles: right eye (RE) -0.75 cylinder (cyl)/ 160° and LE -0.25 cyl/ 20° . Refraction after cycloplegia was not tested. Fundus oculi was performed after dilation and was normal in both eyes. Orthoptic evaluations were normal. The ophthalmologist required further investigation.

The neurological examination and the Simpson test for myasthenia were negative. Brain CT, brain MRI, and electroencephalogram were within the range of normality. Finally, blood tests (complete blood cell count, inflammatory markers, and thyroid function indices) and

neuropsychiatric evaluation were normal. The diagnosis at discharge was of suspected OM, because the patient had reported a slight headache with spontaneous resolution and without nausea and vomiting the day before the AATE attack.

When we evaluated the patient, a week after hospital discharge, the ophthalmological and orthoptic examination was similar to that performed during hospitalization, the only difference being visual acuity of 20/25 in LE with her glasses. We performed refractive examination in cycloplegia half an hour after the administration of tropicamide 1% 3 times in each eye. Refractive cycloplegic values with an autorefractometer (AR600; Nidek, Japan) were RE -1.00 cyl/ 160° and LE $+0.75$ cyl/ 85° . Cycloplegic visual acuity was 20/40 in RE and 20/25 in LE without lens correction, 20/20 in RE and 20/32 in LE with glasses in use, and 20/20 in LE with $+0.75$ cyl/ 85° . The patient's glasses had a low myopic astigmatism (-0.25 cyl/ 20°) in LE, although in cycloplegia she presented a hyperopic astigmatism ($+0.75$ cyl/ 85°) in the same eye. Therefore, in addition to the OM, the AS could be considered as a possible cause.

For 18 months thereafter, the child was instructed to continuously wear glasses with the proper correction, and no episodes of strabismus occurred.

Discussion

AACE is a rare presentation of strabismus and has been classified into 3 forms. The case here described may meet group II criteria, characterized by minimal hypermetropia and diplopia, but in the literature there are only a few case reports and case series about AACE. Moreover, in these studies all patients of group II were referred for strabismus surgery due to persistent esotropia [1–3]. Only Kemmanu et al. [4] described a case of AACE where the patient did not receive surgery. In fact the cause was an AS in LE associated with mild hypermetropia in cycloplegia ($+0.75$ sphere). The esodeviation in LE regressed after 15 days with the use of glasses.

The peculiarity of AATE with respect to AACE is the short duration of strabismus and diplopia (about 3 h) and their spontaneous regression. Being transient, it cannot be classified among AACEs (Table 1).

AATE could be caused by OM, a rare neurological syndrome characterized by recurrent attacks of headache and ophthalmoplegia. This disorder is frequently familial and it usually presents in children. The oculomotor nerve is the most commonly involved cranial nerve, and diplopia, ptosis, and mydriasis are common; also the other cranial nerves may be affected (and, of note, in the case of OM an incomitant deviation may be rarely present). Most patients recover completely within days to weeks, but a minority could show persistent neurological deficits [5]. In our case, the patient had a mild headache the day before the onset of symptoms, and the AATE could have been caused by paralysis of the sixth cranial nerve; but she had no abdominal pain, nausea, and vomiting. Moreover, a family history of migraine headaches was not reported (Table 2).

During the ophthalmological evaluation both diplopia and esotropia in LE were no longer present, and it was not possible to assess whether the deviation was concomitant or incomitant. The latter may be present in the case of OM. However, the short duration of

symptoms and spontaneous remission of AATE could support the OM diagnosis, especially if such attacks had recurred in the future.

AS could be another diagnostic hypothesis for AATE. It is an involuntary condition occurring in the presence of greater than normal accommodative response than accommodative stimulus. AS is characterized by transient episodes of variable esotropia, pseudomyopia, and miosis. It may begin suddenly, is more likely to be bilateral, disappears with cycloplegia, and may resolve spontaneously (Table 2) [6].

In our case, aspects which may be compatible with a diagnosis of AS are as follows: (a) the patient had a hypermetropic astigmatism in LE but the left lens presented a myopic astigmatism; (b) symptoms reappeared when the father and the pediatrician showed fixation aims for a near target; (c) longer reading/study sessions in the last 2 weeks; (d) the short duration and spontaneous regression of symptoms; and (e) the absence of AATE after 18 months, using spectacles with proper correction in a continuous manner. Factors limiting the correct diagnosis were (a) features of AATE were just reported by the patient, father, and pediatrician; as symptoms regressed before the ophthalmological evaluation, the characteristic signs of AS (miosis and pseudomyopia) could not be evaluated; (b) the disorder was unilateral; and (c) a slight hypermetropic defect was present in LE. It will be important to assess future recurrence of the condition, with proper lens correction, with long follow-ups.

Conclusion

The AATE diagnosis is complex. It is necessary to exclude nonophthalmological causes through the execution of neurological examination, test for myasthenia, brain CT, blood tests (complete blood count, inflammatory markers, and thyroid function indices), and neuropsychiatric evaluation. If all tests are negative, full ophthalmological (including cycloplegic refraction) and orthoptic evaluation are mandatory to avoid the unnecessary execution of expensive tests (such as electroencephalogram and brain MRI) which may be a source of anxiety for children and their families.

Acknowledgment

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Statement of Ethics

The parents of the patient (the patient was under 18 years of age) signed a comprehensive consent form according to Good Clinical Practice guidelines before proceeding with all examinations and treatments. It satisfied all the requirements of the Declaration of Helsinki and the Italian national law for the protection of personal data. The local ethics committee decided that no formal ethics approval was required in this particular case.

Disclosure Statement

The authors declare no interests in any instrument/product mentioned in the manuscript.

Author Contributions

D.A., E.N., G.M., and L.R. contributed to the conception and design of the study. D.A., A.P., E.P., S.C., M.S., and P.F. contributed to data acquisition. All authors contributed to data interpretation. All authors drafted the article and approved its final version.

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Table 1. Comparison of the characteristics of AACE and AATE

	AACE (type II)	AATE
Age	childhood	childhood
Signs	sudden diplopia and comitant esotropia	sudden diplopia and esotropia (concomitance has not been evaluated)
Mean time to resolution	from 2 weeks to unresolved	from 15 min to 3 h
Causes	idiopathic, preceding debilitating illness or other physical or psychological stress	ophthalmoplegic migraine or accommodative spasm

Table 2. Comparison of the characteristics of ophthalmoplegic migraine and accommodative spasm

	Ophthalmoplegic migraine	Accommodative spasm
Headache	present	mild or absent
Family history	positive	negative
Systemic symptoms	abdominal pain, nausea, and vomiting	absent
Ophthalmic signs	diplopia, ophthalmoplegia, ptosis, mydriasis, and incomitant esotropia	diplopia, variable esotropia, pseudomyopia, and miosis
Recurrence of AATE	yes	no, in the presence of an appropriate dioptric correction
Therapeutic approach	neurological evaluation and treatment of headache	cycloplegic assessment and prescription of appropriate dioptric correction; annual follow-up in childhood