An Infant with Benign Isolated Abducens Palsy After Vaccination

Aşılama Sonrası Gelişen Benign İzole Abdusens Paralizili Bir İnfant

Aşılama Sonrası Benign Abdusens Paralizisi / Benign Abducens Palsy After Vaccination

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Özet

Benign izole abdusens paralizisi, başka nörolojik bulguların eşlik etmediği, abduksiyon kısıtlılığına bağlı gelişen içe şaşılık ve çift görmeyle karakterize kendi kendine düzelen bir klinik antitedir. Bu antite hafif ateş epizotlarından veya viral enfeksiyonlardan sonra ortaya çıkabilir. Hücre zedelenmesinin patofizyolojik mekanizması bilinmemektedir. Sinir paralizilerine hassasiyete neden olabilen genetik yatkınlık zemininde, otoimmün aracılıklı veya direk viral invazyona bağlı demiyelinizasyon ve lokal arteritten kaynaklandığı düşünülmektedir. Bu vaka raporunda difteri, asellüler boğmaca, tetanoz, inaktif polio ve Haemophilus influenzae type b (DTPa-IP-Hib) aşı uygulamasından iki hafta sonra sağ gözde aniden başlayan içe şaşılık şikayetiyle bizim polikliniğimize başvuran ve benign izole abdusens paralizisi tanısı alan 19 aylık bir kız hasta sunuldu.

Anahtar Kelimeler

Abdusens Sinir Paralizisi; Aşılama; Çift Görme; Paralitik Şaşılık

Abstract

Benign isolated abducens palsy is a self-improving clinical entity characterized by esotropia and diplopia led by the deficiency of abduction, and accompanied by no other neurological findings. The entity may occur after experiencing minor fever episodes, viral infection. The pathophysiological mechanism of cellular injury remains unclear. Hypotheses involve damage arising from autoimmune mediation or direct viral invasion causing demyelination, localized arteritis or genetic predisposition, which could increase susceptibility to such nerve palsies. Diagnosed with benign isolated abducens palsy, a 19-month-old girl infant admitted to our outpatient clinic with an acute onset of esotropia in the right eye developing two weeks after the vaccination of diphtheria, acellular pertussis, tetanus, inactivated polio and Haemophilus influenzae type b (DTPa-IP-Hib) was presented in this report.

Keywords

Abducens Nerve Palsy; Vaccination; Diplopia; Paralytic Strabismus

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Introduction

The abducens nerve, also known as the sixth cranial nerve, innervates the lateral rectus muscle of the eye and is involved in lateral horizontal ocular movement An abduction deficit results in esotropia, ipsilateral abduction deficiency and double vision [1]. Characterized by esotropia and diplopia led by the deficiency of abduction, benign isolated abducens palsy (BIAP) is a self-improving clinical entity and accompanied by no other neurological findings [1-3]. The exact pathophysiological mechanism of cellular injury in BIAP remains unclear. Hypotheses include damage arising from autoimmune mediation or direct viral invasion leading to demyelination, localized arteritis or genetic predisposition, which may enhance the susceptibility to such nerve palsies [4].

In this article, we present a 19-month-old girl infant with benign isolated abducens palsy, which developed four weeks after the vaccination of diphtheria, acellular pertussis, tetanus, inactivated polio and Haemophilus influenzae type b (DTPa-IP-Hib)

Case Report

A healthy 19-month-old girl was admitted to our outpatient clinic with an acute onset of esotropia in the right eye. The case revealed no history of head trauma, nausea, vomiting, confusion, aphasia or other symptoms related to cranial nerve dysfunction. However, the patient had been vaccinated with diphtheria, acellular pertussis, tetanus, inactivated polio and Haemophilus influenzae type b (DTPa-IP-Hib) four weeks earlier. Her prenatal and natal history was unremarkable. Developmental milestones were normal. No cases of neurodegenerative diseases were noted in the family members.

On physical examination, the patient failed to gaze at right lateral side with right eye (Figure 1A and 1B). Pupillary light reflex, anterior segment and fundoscopic examination were normal in both eyes. The patient could not be evaluated for diplopia. The remaining physical examination was unremarkable.



Figure 1. Lateral gazing failure on right eye of the patient with BIAP (A,B)

Laboratory investigations revealed the following findings: hemoglobin 11.9 g/dL; leukocyte count, 12,200/mm3 with a differential of 40% neutrophils, 50% lymphocytes, 6% monocyte and 4% eosinophils; and platelet count 442,000/ mm3. C-reactive protein and erythrocyte sedimentation rates were 3.19 mg/dL and 4 mm/h, respectively. Serum electrolytes, renal and liver function tests were normal on admission. Non-contrast-enhanced and contrast-enhanced MR imaging were normally determined. No viral agent was identified in cerebrospinal fluid by polymerase chain reaction.

The patient was diagnosed with BIAP, based on clinical and laboratory findings. Part-time occlusion therapy for the left eye was conducted by ophthalmologist to block the inhibition of the affected eye. She completely recovered six weeks after the diagnosis. No further treatment regimes such as botulinum toxin or a surgical intervention were needed. Additionally, no recurrence sign was observed during a 12-month follow-up period.

Discussion

The abducens nerve is the longest cranial nerve originated from pons and so being affected often by intracranial pathologies such as neoplasia, hemorrhage, increased intracranial pressure, vascular disease and secondary to surgery [2]. It innervates only lateral rectus muscle whose pathologies cause the deficit of eye abduction [1].

BIAP, firstly described by Knox et al. in 1967, is an entity of unknown pathophysiology, but can occur after minor febrile illness, viral infection [1,5]. Cases of BIAP after vaccination of influenza, diphtheria-pertussis-tetanus (DPT) and mumps-measles-rubella (MMR) have been reported in the literature [6]. On medical history of our case, vaccination was noted. Therefore, precipitating factor arising from the vaccination was considered to lead to BIAP. Patients with BIAP mostly complain of a visual disturbance and esotropia with sudden onset. Among the complaints, the chief one is binocular horizontal diplopia occurring only with both eyes open [7]. Our case was also admitted with acute onset esotropia in right eye, but diplopia could not be evaluated because of her young age.

BIAP usually resolves fully and spontaneously in a few months, but it can recur following before mentioned precipitating factors. The recurrence rate was reported as 30% by Mahoney and Liu [2]. In another study, Yousuf and Khan reported that the mean age in the recurrent cohort was younger than that in the non-recurrent cohort, and that while a tendency was present toward both non-recurrence and the involvement of right-sided palsy in boys, the same was in favor of both recurrence and the involvement of left-sided palsy in girls [8]. In contrast to report of Yousuf and Khan, our case was female, and her right eye had been affected. The average interval between recurrences are reported as 12 months by Knapp et al [3]. In our case, no recurrence was observed during a 12-month of follow up, and the improvement period was six weeks, which was shorter than those of literature data.

The purpose of the treatment is to prevent the development of amblyopia secondary to double vision. For this purpose, the vision of non-affected eye must be blocked with part-time occlusion therapy. If no improvement is seen in the patient's condition at least after a 3-month follow-up, the chemodenervation of the ipsilateral medial rectus muscle with botulinum toxin or surgery, including full temporal transposition of the vertical rectus muscles, may be considered as a therapeutic option. Our case did not require chemodenervation with botulinum toxin or surgery because she was completely improved with part-time occlusion therapy within six weeks.

Other reasons causing abducens palsy must be excluded in or-

der to diagnose BIAP accurately. Therefore, findings such as papilledema, movements of eye, the signs of meningeal irritation and other systemic signs must be looked for carefully. Brain magnetic resonance imaging is a beneficial modality used for anatomic pathologies. If there are signs related to meningitis like fever, vomiting or seizure, a lumbar puncture should be performed, and cerebrospinal fluid should be examined.

In conclusion, based on our report, we would like to emphasize that BIAP should be taken into consideration in infants with sudden onset esotropia due to vaccination, and part-time occlusion therapy can successfully be used in the treatment modality.

Competing interests

The authors declare that they have no competing interests.

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