Case Report

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Two cases of benign abducens nerve palsy in children and their long-term follow-up

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Abstract

Our aim was to explore the clinical characteristics and diagnostic methods of benign abducens nerve palsy in children. The clinical and laboratory characteristics, treatment approach, and prognosis of two children with benign abducens nerve palsy were retrospectively evaluated. The main clinical symptoms of the two children were limited binocular movement accompanied by double vision, and the positive physical signs were limited binocular abduction accompanied by diplopia. No abnormalities were found in laboratory examinations or in imaging of the head, chest, and abdomen. Both children were treated with B vitamins, methylprednisolone, and gamma globulin, and the clinical symptoms disappeared within 3 months of starting treatment. The cause of benign abducens nerve palsy in children is unknown, but may be related to immune factors. In the two cases presented here, the patients recovered after treatment with immunomodulators.

Keywords

Abducens nerve palsy, sixth nerve palsies, benign, child, immunomodulators, diplopia

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Introduction

The abducens nerve nucleus is located in the middle dorsal side of the pontobulbar junction. Its nerve fibers project to the external rectus muscle, which serves as the abductor of the eyeball.[1] Because the abducens nerve is long at the base of the skull, it is particularly vulnerable to increases in intracranial pressure or skull base fractures.

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The main manifestations of abducens nerve palsy are internal obliquity and limited ipsilateral abduction, and some children may have diplopia. Lee et al.[2] analyzed the causes of abducens nerve palsy in children and concluded that tumors and related neurosurgery were the most common causes (45%), followed by elevated intracranial pressure (15%), traumatic brain injury (12%), congenital causes (11%), inflammation (7%), other causes (5%), and idiopathic causes (5%). It has also been reported that Guillain–Barre syndrome[3] and upper respiratory infections[3] can lead to abducens nerve palsy. Benign abducens nerve palsy has a low incidence in children, and its diagnosis relies on the exclusion of other diagnoses.[4] The age of onset of the disease varies, and the minimum age of onset is reported to be 6 months.[4] It has been reported[5] that this disease occurs more frequently on the left side, and there is no significant difference in the incidence between men and women.[3] Patients tend to recover spontaneously within 6 months, and while some cases may experience recurrence, the long-term prognosis is generally good.

This study summarized the clinical data of two patients, and followed them long term to provide a reference for the diagnosis, treatment, and prognosis of benign abducens paralysis in children.

Clinical data

Case 1

A 10-year-old boy was admitted to the department of neurology of the Children's Hospital affiliated with Soochow University in July 2018 because of binocular limited outreach and double vision. There was no special history, personal history, or family history, and no history of head trauma. Physical and neurological examinations were normal except for double abducens nerve palsy: when the left and right eyeballs tried to move outward, the sclera was exposed for 4 mm, with diplopia. No abnormalities were found in laboratory examinations, or in imaging of the chest and abdomen. Magnetic resonance imaging (MRI) of the brain revealed an arachnoid cyst at the bottom of the right temporal lobe. Cerebrospinal fluid examination findings were as follows: leukocyte count: 0.004×10^9/L, glucose: 3.52 mmol/L, and protein: 0.451 g/L. Oxiracetam, B vitamins (B1, B6, mecobalamin), methylprednisone, and gamma globulin were administered, and the patient was hospitalized for 3 weeks. At discharge, there was no significant improvement in diplopia and the range of binocular abduction. After discharge, the patient continued to take methylprednisone and B vitamins, and during the first 2 weeks after discharge, the range of binocular abduction began to improve: when the left and right eyeballs tried to move outward, the sclera was exposed for 3 mm, with diplopia. One month after discharge, the patient still presented with limited abduction in both eyes, but the degree of abduction was reduced, and sclera was exposed for 1 to 2 mm, with diplopia. Two months after discharge, all symptoms had disappeared; there was no double vision or abduction limitation. Six months after discharge, the patient still showed no symptoms, and had a good long-term prognosis without sequelae.

Case 2

An 11-year, 5-month-old boy presented with binocular limited outreach in the left eye, first, and was admitted to the department of neurology of the Children's Hospital affiliated with Soochow University in February 2019. He had a respiratory infection 1 week prior to palsy symptom onset, and there was no other special history, personal history, or family history, and no history of head trauma.

Physical and neurological examinations were normal except for double abducens nerve palsy: when the left and right eyeballs tried to move outward, the sclera was exposed for 4 to 5 mm, with diplopia. No abnormalities were found in laboratory examinations, or in chest and abdominal imaging; head MRI was negative. Routine biochemical, oligoclonal, and ganglioside antibody profiles in cerebrospinal fluid were normal. Mouse nerve growth factor, B vitamins (B1, B6, mecobalamin), methylprednisone, and gamma globulin were administered, and the patient was hospitalized for 4 weeks. At discharge, the binocular abduction limitation was slightly better than at admission: when the left eyeball tried to move outward, the sclera was exposed for approximately 3 mm, and when the right eye ball tried to move outward, the sclera was exposed for approximately 4 mm, with diplopia. After discharge, the patient continued to take methylprednisone and B vitamins, and during the first 2 weeks after discharge, the range of binocular abduction began to improve: when the left and right eyeballs tried to move outward, the sclera was exposed for 2 to 3 mm, with diplopia. One month after discharge, the patient still presented with limited abduction in both eyes, but the degree of abduction was reduced, with only 1 to 2 mm of sclera exposed, with diplopia. Two months after discharge, all symptoms had disappeared; there was no double vision or limited abduction. The patient had a good prognosis without sequelae.

Discussion

In the present study, we report two cases of benign abducens nerve palsy in children, both of whom were over 10 years old. Both patients had bilateral abducens nerve involvement. The second case had an upper respiratory infection 1 week before the disease onset and developed limited left eye abduction 8 days later. The arachnoid cyst at the bottom of the right temporal lobe in case 1 could not have affected bilateral abducent nerves; therefore, we believe the abducent nerve palsy was independent of the cyst. All auxiliary examinations in the two cases showed no abnormalities, which excluded other causes. In the follow-up examinations, the children's symptoms were slightly improved within 5 to 6 weeks, and completely resolved within 12 to 14 weeks, with no sequelae. Given this recovery rate, and the benign and idiopathic characteristics, both of these cases can be diagnosed as benign abducens nerve palsy.

The pathogenesis of benign abducens palsy in children is unclear, but immune-mediated injury or demyelinating lesion is widely considered to play a role. Because neither patient had long-term recurrence or neurological sequelae, this disease can be considered benign;[5,6] however, both patients should be followed closely to ensure that a tumor diagnosis was not missed.[7] Considering that benign abducens nerve palsy in children may be caused by immune-mediated nerve injury, we treated both patients with methylprednisone, gamma globulin, and vitamin B for nerve nutrition, and their clinical symptoms gradually improved. Some studies[3] also claim that the disease itself is self-limited and does not require specific treatment.

In the two cases reported here, no abnormal findings were observed in neurological or other auxiliary examinations, making it an exclusive diagnosis of abducens palsy. Sullivan[8] reported that most children with benign abducens nerve palsy started to recover in 3 to 6 weeks, and symptoms disappeared by 10 weeks, with a good prognosis. The two cases we reported showed slight improvement in symptoms at 5 to 6 weeks and complete recovery by 12 to 14 weeks, which was slightly slower than the cases reported by Sullivan.

Sturm et al.[4] claim that the average recovery time for this disease is 3.6 months, with the longest cases not exceeding 6 months.

In summary, in this paper, we summarized and reported the follow-up clinical characteristics of two children with benign abducens nerve palsy to provide a more comprehensive and in-depth understanding of the disease and to provide a reference for its diagnosis and prognosis.

Ethics statement

This study was approved by the medical ethics committee of the Children's Hospital of Soochow University (approval number: 2019LW015). Written informed consent was obtained from the parents of both participants.

Author contributions

Jihong Tang: study concept and design, and data analysis. Tong Zhang: data collection and assistance in data analysis; drafted the manuscript.

Declaration of conflicting interests

The authors declare that there is no conflict of interest.

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References

1. Nam YS, Kim IB and Shin SY. Detailed anatomy of the abducens nerve in the lateral rectus muscle. *Clin Anat* 2017; 30: 873–877.
2. Lee MS, Galetta SL, Volpe NJ, et al. Sixth nerve palsies in children. *Pediatr Neurol* 1999; 20: 49–52.
3. Teksam O, Keser AG, Konuskan B, et al. Acute abducens nerve paralysis in the pediatric emergency department: analysis of 14 patients. *Pediatr Emerg Care* 2016; 32: 307–311.
4. Sturm V and Schöffler C. Long-term follow-up of children with benign abducens nerve palsy. *Eye (Lond)* 2010; 24: 74–78.
5. Vallée L, Guilbert F, Lemaitre JF, et al. Benign paralysis of the 6th cranial nerve in children. *Ann Pediatr (Paris)* 1990; 37: 303–305.
6. Bourtoulamaiou A, Yadav S and Nayak H. Benign recurrent sixth (abducens) nerve palsy following measles-mumps-rubella vaccination. *Case Rep Pediatr* 2015; 2015: 734516.
7. Mahoney NR and Liu GT. Benign recurrent sixth (abducens) nerve palsies in children. *Arch Dis Child* 2009; 94: 394–396.
8. Sullivan SC. Benign recurrent isolated VI nerve palsy of childhood. *Clin Pediatr (Phila)* 1985; 24: 160–161.