

## Management of a Case with Isolated Oculomotor Nerve Palsy

### İzole Okülomotor Sinir Felci Olan Bir Olguda Tedavi Yaklaşımı

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**ABSTRACT** The oculomotor nerve has a divisional topographic arrangement at the brain stem and extends with fascicular, cisternal, cavernous sinus and orbital portions. The lesions, at any site along this course, can cause nerve paralysis and the etiologies may differ in adult and child age groups. Congenital reasons, craniopharyngioma, trauma, inflammatory and infectious causes, ophthalmoplegic migraine, malignancy (lymphoma, leukemia), cranial aneurysms are the possible underlying diagnoses in the childhood. The most of the divisional oculomotor nerve palsy reports in the literature are linked to post-viral etiology in the child age group. The current clinical picture resolved spontaneously in these cases. We presented a case of an 8-year-old girl with left eye superior division oculomotor palsy. The clinical investigations for clarifying underlying causes were unremarkable. After a certain follow-up period for spontaneous resolution, surgical treatment was performed and this case is the first one who treated successfully with surgery in the child age group.

**Keywords:** Oculomotor nerve; child; surgery

**ÖZET** Okülomotor sinir, beyin sapında topografik olarak fasiküler, sisternal, kavernöz sinüs ve orbital dallara ayrılacak şekilde düzenlenmiştir. Kafa içinde, belirtilen bu dalların seyri boyunca herhangi bir yerdeki lezyonlar sinir felcine neden olabilir ve etiyolojik sebepler yetişkin ve çocuk yaş gruplarında farklılık gösterebilir. Konjenital nedenler, kraniyofarenjiyoma, travma, inflamatuvar ve enfeksiyöz nedenler, oftalmoplejik migren, malignite (lenfoma, lösemi), kraniyal anevrizmalar çocuk yaş grubundaki olası sebeplerdir. Literatürde çocuk yaş grubundaki okülomotor sinir dal felci vakalarının çoğu post-viral sebeplerle ilişkilendirilmiştir. Takiplerde bu olgularda mevcut klinik tablo kendiliğinden düzelmiştir. Bu vaka raporu sol göz okülomotor sinir üst dal felci bulunan 8 yaşında bir kız çocuğuna ait idi. Altta yatan muhtemel nedenlerin araştırıldığı muayene ve tetkiklerde herhangi bir patoloji saptanmadı. Literatürdeki diğer vakalar dikkate alındığında hernekadar klinik araştırmalarda bir patoloji saptanmasa da spontan düzelmeye için hasta belli bir süre takip edildi. Fakat klinik bulgularında düzelmeye olmayan hastaya cerrahi tedavi uygulandı. Bu vaka literatürde çocuk yaş grubunda cerrahi ile başarılı şekilde tedavi edilen okülomotor sinir dal felci olan ilk çocuk hastadır.

**Anahtar Kelimeler:** Okülomotor sinir; çocuk; cerrahi

The oculomotor nerve is divided into superior and inferior divisions in the region of the anterior cavernous sinus and superior orbital fissure. The superior division oculomotor palsy is usually seen in sphenocavernous region or orbital apex. Oculomotor superior division innervates levator palpebrae superioris and superior rectus muscles. Therefore, mild ptosis and upgaze restriction could be seen in patients with superior division palsy, besides pupillary reactions have been preserved.

In children, congenital reasons, trauma, inflammatory causes, neoplasms, cranial aneurysms, are the most common responsible etiologies.<sup>1</sup> The priority audits must focus on underlying causes in these cases. We present a case to reveal surgical treatment option for the treatment of an superior division oculomotor palsy in a child.

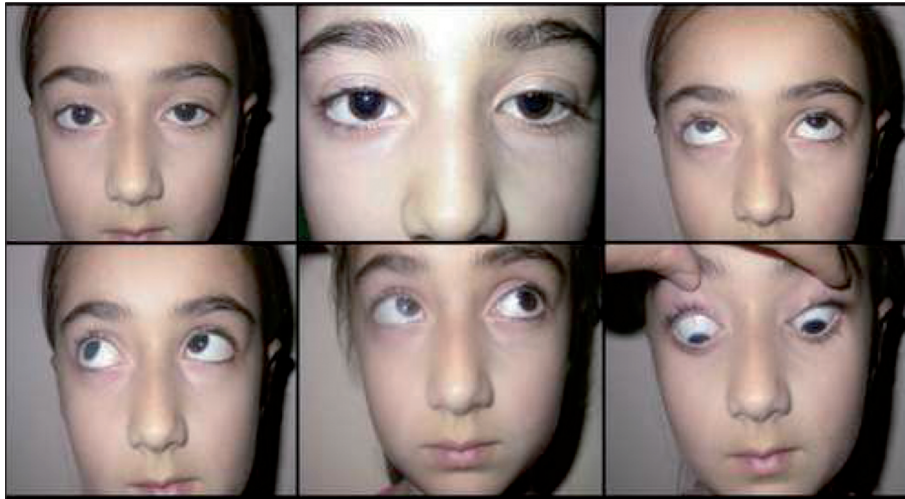
## CASE REPORT

8-year-old girl patient was presented with left eyelid ptosis and upgaze restriction starting one month ago. Her recent medical history was unremarkable including no common cold and flu-like illness. Besides, the patient has no known systemic disease. The initial ocular examination was as follows: Best corrected visual acuities (BCVAs) were 20/20 in both eyes, bilateral anterior segment, fundus examinations were normal, intraocular pressure (IOP) values were 14 mmHg (OD), 13 mmHg (OR). A moderate pseudoptosis, -3/-4 degree elevation limitation and also -3 degree up/right and up/left gaze restriction, 45 prism hypotropia (alternate on and off test and prism cover test) were determined in the left eye and light reflexes were normal in both eyes (Figure 1). Our patient has borderline mental capacity and difficulty in attracting self-expression. So she did not complain a clear diplopia and due to her insufficient coordination, we could not perform Worth's Four Dot test. With the pre-

liminary left oculomotor superior division palsy, necessary examinations and tests were performed for the investigation of possible etiologic factors. Neurological examinations were normal and there was no neck stiffness. Cranial and orbital magnetic resonance imaging (MRI) revealed no intracranial and orbital space-occupying lesions (aneurysms, craniopharyngioma, malignancy, sphenothmoidal sinusitis). The patient had no history of trauma and ophthalmoplegic migraine. The blood tests for the investigation of diabetes, connective tissue diseases, malignancy, viral and other infectious diseases were also unremarkable. Considering possible unnoticed temporary inflammatory and infective causes, the patient was followed up for six months. But the patient's current clinical status did not improve and patient scheduled for the surgery. Superior rectus and inferior rectus insertion from limbus were 7.5 mm and 6.5 mm respectively in the left eye. Traction tests were unremarkable in both eyes. 5 mm superior rectus resection and 4 mm (total 10,5 mm from limbus) inferior rectus recession surgeries were performed to the patient in the left eye under general anesthesia. In the postoperative sixth month examination; 4 prism minimal hypotropia in the left eye, bilateral symmetrical eyelids were present. There were free eye movements to all directions in the right eye while there was a slight restriction in up, up/right, up/left gazes (-1 degree) in the left eye (Figure 2).



**FIGURE 1:** Preoperative pictures of patient demonstrate hypotropia, -3/-4 degree elevation limitation and also -3 degree up/right and up/left gaze restriction, and pseudoptosis in the left eye.



**FIGURE 2:** Postoperative sixth months A 4 prism minimal hypotropia, slight restriction in up, up/right, up/left gazes (-1 degree) in the left eye.

## DISCUSSION

The oculomotor nerve is divided into superior and inferior divisions within the anterior third portion of the cavernous sinus. Therefore, the superior division palsy is usually occurred secondary to lesions in front of one-third of the cavernous sinus or in the posterior orbit.<sup>2</sup> Otherwise, the oculomotor nerve has a divisional topographic arrangement at the brain stem and extend with fascicular, cisternal, cavernous sinus and orbital portions. Thus, the divisional oculomotor nerve palsy may occur because of lesions anywhere along its course.<sup>3-5</sup>

Congenital reasons, trauma, inflammatory causes, neoplasms, cranial aneurysms, craniotomy, diabetes mellitus and sphenoid sinusitis were the possible etiologies causing oculomotor nerve palsy in childhood.<sup>1,5-7</sup> Also, cases with divisional oculomotor nerve palsy associated with ophthalmoplegic migraine have been reported in the literature in children.<sup>8,9</sup>

Our patient had no history of trauma and ophthalmoplegic migraine. Cranial and orbital MRI imaging revealed no findings related to an aneurysm, malignancy (craniopharyngioma, lymphoma, leukemia) and sphenoidal sinusitis. Blood tests investigating the connective tissue diseases, infective causes and diabetes did not show any pathology.

To our knowledge, only three oculomotor superior division palsy cases have been reported in the literature due to a post-viral infective cause.<sup>10-12</sup> One of these cases reported by Engelhardt, 5-year-old boy had superior division palsy just after a common cold. Increased titers of the influenza-A virus have been detected in the serological tests in this case and symptoms resolved spontaneously after four months follow-up.<sup>11</sup> Other cases were 39 years old man and 10 years old boy had a history of the common cold. Any specific viral marker tests were not performed in these cases. The clinical findings resolved spontaneously after two months later in both patients. The mechanism of oculomotor nerve palsy due to a post-viral etiology is unclear. Autoimmune reactions and post-infectious cranial neuropathy of childhood may be responsible causes.<sup>11,13</sup>

There were limited cases in the literature for surgical correction of isolated oculomotor nerve palsy.<sup>14-16</sup> Flanders and colleagues presented a case series comprising 12 adult patients whose chief complaint of diplopia.<sup>16</sup> But all of these cases had underlying etiologic reasons such as an aneurysm, tumor, and cerebrovascular accident.

Failure to find any evidence at the end of the diagnostic tests and imaging may be insufficient to reveal the main underlying cause. So at this point, we believe that a subclinical infection and a persistent unnoticed autoimmune reaction may have affected nerve functions.

Considering cases in the literature with spontaneous improvement due to viral etiology, we followed our patient for six months. But patient's clinical findings did not resolve during this time. Regarding patients age and low risk for amblyopia the time before surgery could be extended for a minimum 12 months. But at this point the patient and parents requested were also considered. Therefore patient underwent surgery, 5 mm superior rectus resection and 4 mm (total 10,5 mm from limbus) inferior rectus recession surgeries were performed in the left eye. 4 prism minimal hypotropia, bilateral symmetrical eyelids were determined in postoperative follow-up (Figure 2). There is no case of a child who had undergone surgery because of an idiopathic oculomotor superior division palsy in the literature. Priority should be the investigation of the possible underlying causes in these cases. But, if the reason could not clarify as in our patient surgery may be considered as an alternative treatment option.

### Informed Consent

The written informed consent was obtained from the patient.

### Source of Finance

During this study, no financial or spiritual support was received neither from any pharmaceutical company that has a direct connection with the research subject, nor from a company that provides or produces medical instruments and materials which may negatively affect the evaluation process of this study.

### Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

### Authorship Contributions

**Idea/Concept:** Mustafa Gök, Mehmet Aslan, Özgül Altıntaş; **Design:** Mustafa Gök, Özgül Altıntaş; **Control/Supervision:** Mustafa Gök, Mehmet Aslan, Özgül Altıntaş; **Data Collection and/or Processing:** Mustafa Gök, Mehmet Aslan; **Analysis and/or Interpretation:** Mustafa Gök, Özgül Altıntaş; **Literature Review:** Mustafa Gök; **Writing the Article:** Mustafa Gök; **Critical Review:** Mehmet Aslan, Özgül Altıntaş; **References and Fundings:** Mustafa Gök; **Materials:** Mustafa Gök.

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