Case Report

Isolated Oculomotor Nerve Palsy Due to Chronic Inflammatory Demyelinating Polyradiculopathy

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Abstract

Ophthalmoplegia in chronic inflammatory demyelinating polyradiculopathy (CIDP) clinic is very rare. In this report we present a case with acute onset isolated inferior division of oculomotor nerve palsy due to CIDP which has not been reported before in literature. 32 years old female patient was admitted to our department with sudden onset diplopia which has begun five days ago. She had been a CIDP patient under control for 2 years. On neurological examination we detected paresis of medial and inferior rectus with efferent pupil defect on the right side suggesting inferior division of oculomotor nerve involvement. Her orbital MRI investigation disclosed a contrast enhanced hyperintense lesion at the superior orbital fissur on the affected side. After pulse methyl prednisolon treatment for five days, a remarkable improvement was observed in ophthalmoplegia. The clinicians should be aware of the possibility that ophthalmoplegia can be seen in CIDP which shows good response to corticosteroids.

Keywords: Inflammatory polyneuropathy, oculomotor nerve, ophthalmoplegia

Özet

Kronik İnflamatuar Demyelinizan Poliradikulopatiye Bağlı İzole Okulomotor Sinir Paralizisi

INTRODUCTION
Chronic inflammatory demyelinating polyneuropathy (CIDP) infrequently manifested with cranial neuropathy and hypertrophy (1,6,9,10,13). Although there are few cases of cranial nerve hypertrophy in CIDP manifesting with ophthalmoplegia (10,13) proptosis (6,9), and lid retraction (2), isolated inferior division of oculomotor nerve palsy due to CIDP has not been reported in literature before. In this report, we present a case with acute onset partial oculomotor nerve palsy. Our patient had CIDP for last 2 years and she was on remission. Neurological findings suggested that inferior division of oculomotor nerve palsy could cause this clinical picture. Radiological findings demonstrated the nerve involvement at the anterior part of cavernous sinus and superior orbital fissure.

CASE PRESENTATION
Thirty-two years old female patient was admitted to our department with sudden onset diplopia. Her complaint has begun 5 days ago. A mild and short lasting peri-orbital pain was accompanied to diplopia. She was diagnosed as CIDP two years ago and she had been under control in our out-patient department. After methyl prednisolone therapy, her motor, sensorial findings and ataxia were improved. She was on remission with the methyl prednisolone treatment (16 mg/alternated dose regimen). Two years before CIDP diagnosis, she had two diplopia attacks that lasted within several days. Since she did not admitted to our hospital with these complaints and had no reports about her health condition regarding previous attacks, it is impossible to make a comment on them.

Her neurological examination disclosed that a partial oculomotor nerve involvement (right medial rectus, inferior rectus paresis with a dilated pupil which is unreactive to light) (Figure 1). Additionally, deep tendon reflexes were abolished on upper and lower extremities. No ataxia was observed. Evident motor dysfunction was not observed in her motor examination. Vibration sensation was diminished in lower extremities. Visual acuity was 20/20. The pattern of oculomotor nerve involvement was appropriate with lower division of n.oculomotorius involvement (Figure 1). Routine hemogram, blood biochemistry, C3 and C4 levels, thyroid function test, serum protein electrophoresis, immunelectrophoresis and cerebrospinal fluid examination were unremarkable. Brucella, human immunodeficiency virus, cytomegalovirus and Epstein-Barr virus were investigated with the suspicion of vasculitis, but all of them were normal. Antiganglioside antibodies (GM1b IgM, GQ1b IgG, GD1b), antmyelin-associated glycoprotein, and acetylcholine receptor antibodies were also negative.

Histological examination of cerebrospinal fluid was also normal. Slowed peripheral nerve conductions were observed on her electrophysiological investigations. Since clinical and electrophysiological regression was absent, a new exacerbation of CIDP was not considered. Her cranial magnetic resonance imaging (MRI) was normal. Her orbital MRI investigation disclosed that a contrast enhanced hyperintense lesion at the superior orbital fissur on the right side (Figure 2). No abnormality was observed on the cranial magnetic resonance angiography.

Pulse methyl prednisolone treatment (intravenous, 1gr/day, 5 days) was instituted for patient. Although a remarkable improvement was observed in the paresis of inferior rectus after this treatment, adduction paresis showed modest improvement. Her control orbital MRI with contrast was normal.
**Figure 1:** Right partial oculomotor nerve involvement (medial rectus, inferior rectus paresis) was detected on neurological examination.

**Figure 2:** Contrast enhanced, fat suppressed axial T1 (A, B), and coronal (C) T1 weighted images showing a contrast enhanced thickened nerve coursing through right superior orbital fissure (arrow) and lateral wall of cavernous sinus. Coronal T2 weighted image (D) through orbit shows asymmetrically enlarged T2 hyperintense right third cranial nerve.
DISCUSSION
Cranial neuropathy is clinically uncommon in patients with CIDP. Several CIDP cases with ophthalmoplegia due to cranial nerve involvement have been reported before in literature\(^1,2,6,9,10,13\). CIDP exacerbation can also be seen with ataxia, areflexia and ophthalmoplegia as an overlap with Miller Fisher Syndrome (MFS)\(^3\). Korkmaz et al. reported a 14 years old age patient who had wall eyed bilateral internuclear ophthalmoplegia (WEBINO) with MFS which occurred during the course of CIDP previously\(^12\). Although it is very rare ophthalmoplegia\(^10,13\), proptosis\(^1,6,9\), and lid retraction\(^5\) may be also manifested in CIDP patients with cranial nerve hypertrophy which can be demonstrated by MRI. In this report, we present a case with acute onset isolated inferior division of oculomotor nerve palsy due to CIDP which has not been reported before in literature.

Many causes of isolated inferior division palsy of third nerve have been described\(^8,11\). The diagnosis may be mistaken for many pathologies. Causes of acquired inferior division of oculomotor nerve palsy include local orbital disease, trauma, vascular abnormality, tumor, and infarction in the cavernous sinus or midbrain. Occasional cases have been associated with vasculitis, demyelination, or viral infection\(^4,5,14,17\). In our case, new neurological features of the patient were suggesting the involvement of inferior division of oculomotor nerve. There was no additional motor or sensory finding. Therefore, an orbital lesion can be responsible for the clinical picture was suspected. However, no structural lesion was demonstrated in the radiological investigations. In addition laboratory tests for the evaluation of thyroid function, infection, and vasculitis were all normal.

Isolated inferior division of oculomotor nerve has also been observed in some patients suggesting an inflammatory process\(^7,15\). In these reports, enhanced third nerve by fat suppression technique of MRI was helpful in the demonstrating of inflammatory process. Although CIDP rarely causes involvement of cranial nerves, it should be considered in the differential diagnosis of cranial nerve hypertrophy. It has been also reported that ophthalmoplegic attacks can be seen related with CIDP. Although our patient had mentioned transient diplopia previously, it is impossible to speculate on this. Nevertheless it should not be forgotten that ophthalmoplegia due to CIDP may reoccur in these patients\(^1\).

Anti-GM1 and antiganglioside antibodies may also be positive. Partial third nerve palsy due to CIDP has been reported previously\(^16\). In this case, contrast enhancement was observed in proximal segment of the oculomotor nerve. However, isolated inferior division palsy of third nerve due to CIDP has not been reported before. Radiological findings disclosed the involvement of more distal segment of oculomotor nerve in our case.

Cranial nerve involvement in CIDP shows good response to corticosteroids and IVIG. The other treatment options for CIDP are plasma exchange, and immunosuppressive agents however they do not cause regression of nerve hypertrophy. The partial recovery after IVIG and corticosteroid treatment suggests a role of humoral mediators but does not clarify their nature. After pulse methyl prednizolon treatment, ophthalmoplegia showed also a remarkable improvement in our patient as expected.

In conclusion clinicians should be aware of the possibility that ophthalmoplegia can be seen in CIDP which shows good response to immun therapies such as corticosteroids and IVIG. CIDP may involve oculomotor nerve more distally causing isolated inferior division palsy of third nerve.
REFERENCES


