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Case Report / Olgu Sunumu

A rare manifestation of Crohn's disease in an adolescent: Optic neuritis

Adolesan Crohn hastasının nadir bir bulgusu: Optik nörit

Ayhan Balkan[1], Sabit Kimyon[2], Alper Mete[2], Murat Taner Gülşen[1]

[1]Department of Gastroenterology, Gaziantep University School of Medicine, Gaziantep, Turkey

[2]Department of Ophthalmology, Gaziantep University School of Medicine, Gaziantep, Turkey

ABSTRACT

Crohn's disease (CD) affects the small and large intestines with focal transmural granulomatous inflammation. Some patients with inflammatory bowel diseases (IBD) also show ocular involvement. Optic nerve involvement in patients with IBD may present as neuroretinitis, papillitis, optic neuritis (demyelinating), or ischemic optic neuropathy. There is limited information on ocular involvement in adolescent and pediatric patients. We present a case of a 17-year-old male who applied to the gastroenterology department with abdominal pain, diarrhea, and sudden vision loss in the right eye and was eventually diagnosed with CD, optic neuropathy, and neuroretinitis. To best of our knowledge, this is the second report regarding optic nerve involvement in CD in the pediatric and adolescent population in literature.

Keywords: Adolescent, Crohn's disease, extraintestinal manifestations, optic neuritis, vision loss

ÖZ

Crohn hastalığı (CD) ince ve kalın barsaklarda fokal transmural granülomatöz inflamasyon yapan bir hastalıktır. İnflamatuvar barsak hastalığı (IBD) olan bazı hastalarda göz tutulumu olabilmektedir. Optik sinir tutulumu olan IBD hastalarında, neuroretinit, papillit, optic (demyelinizan) neurit veya iskemik optik nöropati saptanabilir. Bununla birlikte adolesan ve pediatrik hastalarda oküler tutulum ile ilgili çok az bilgi mevcuttur. Biz Gastroenteroloji bölümüne karın ağrısı, ishal, sağ gözde ani görme kaybı şikayeti ile başvuran ve yapılan incelemelerde Crohn hastalığı, optik nörit ve nöroretinit teşhisi konulan 17 yaşında erkek hastayı vaka sunumu olarak rapor ettik. Bildiğimiz kadarıyla, pediatrik ve adolesan dönemde, Crohn hastalığında optik sinir tutulumu hakkında şu anki literatürde ikinci vakayı rapor ettik.

Anahtar kelimeler: Adolesan, Crohn hastalığı, extraintestinal bulgular, optik nörit, görme kaybı

INTRODUCTION

Crohn's disease (CD) affects the small and large intestines with focal transmural granulomatous inflammation (1). Inflammatory bowel diseases (IBD) mostly present with gastrointestinal symptoms; however, 20%-40% of patients also have extraintestinal findings (2). Previous studies have shown that 1%-2% of the patients with IBD also have ocular involvement (3). The extent of bowel involvement does not correlate with ocular complications, and ocular involvement generally occurs in the early years of the disease. Ocular findings may precede IBD diagnosis in some patients (4).

Optic nerve involvement in patients with IBD may present as neuroretinitis, papillitis, optic neuritis (ON, demyelinating), or ischemic optic neuropathy (5). ON can be present in up to 4% of adult IBD patients (6, 7). Symptoms include blurry or decreased vision for few hours to several days and retrobulbar pain (5).

We report a case of a 17-year-old male who applied to the gastroenterology department with abdominal pain, diarrhea, and sudden vision loss and was eventually diagnosed with CD, ON, and neuroretinitis.

CASE PRESENTATION

A 17-year-old male Syrian refugee applied to another hospital with diarrhea, abdominal pain, hyperemia in his left eye, and was hospitalized. On the third day of hospitalization (DoH), patient described floaters in both eyes and blurry vision in the right eye but he was not referred to an ophthalmologist. On the sixth day, he was referred to another hospital due to unknown reasons. On the eight day, the patient had a temporary vision loss in his right eye, which lasted for 1 hour, and his ophthalmologic examination was evaluated as normal. On the tenth DoH, the patient had fever and permanent vision loss in his right eye. The two hospitals could not diagnose the condition and hence the patient did not receive any specific treatment. He was referred to our hospital on the twelfth DoH. Patient had yellow-colored watery diarrhea 10–15 times a day without blood, abdominal pain, high fever and vision loss in his right eye at his admission to gastroenterology department of our hospital. He was dehydrated and had lost 8 kg weight by this time. He had tenderness in the epigastric region and lower quadrant of the abdomen without defense or rebound. Laboratory tests revealed white blood cell (WBC), 11.000/μL; erythrocyte sedimentation rate (ESR), 40 mm/h; C-reactive protein (CRP), 130 mg/dL; and albumin, 2.7 g/

Corresponding Author/Sorumlu Yazar: Ayhan Balkan **E-mail/E-posta:** dr.ayhanbalkan@hotmail.com

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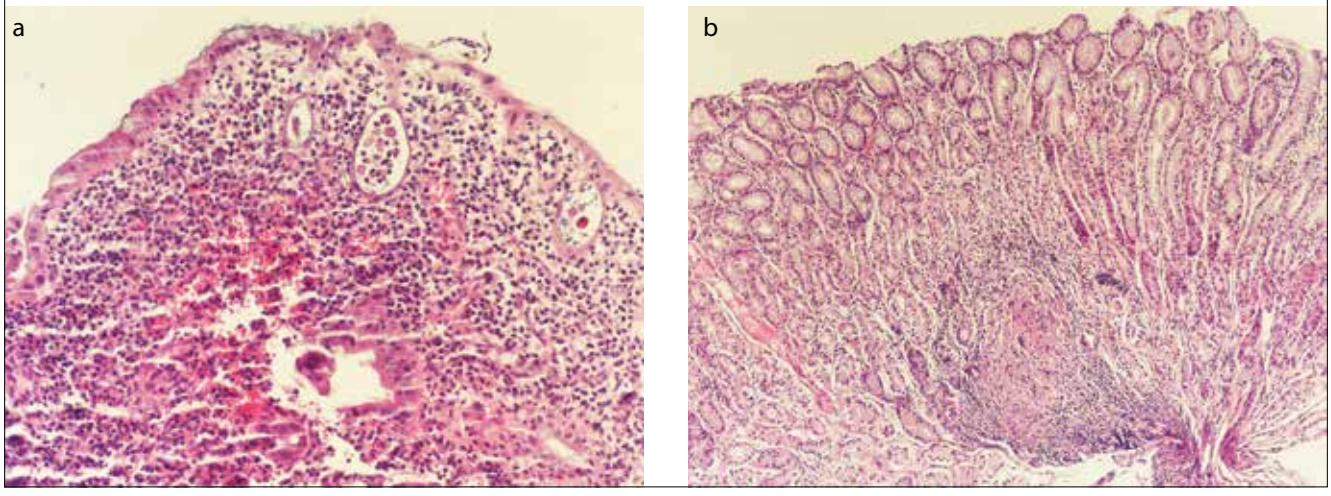
Figure 1. a-c. (a) Macular OCT image at presentation shows submacular fluid in the right eye. (b) Anterior segment image of the right eye shows iris pigments on the lens. (c) Anterior segment image of the left eye shows iris pigments on the lens

Figure 2. a, b. Gastric and colonic involvement of CD. (a) Mixed inflammatory cell reaction attacking surface epithelium and crypts in colon mucosa (H and E ×200). (b) Granuloma at lamina propria of gastric mucosa (H and E ×100)



dL. We excluded infection and infestation but detected hemoglobin in the fecal matter. Posteroanterior chest X-ray, thorax and upper and lower abdominal computed tomographies (CTs) (Somatom Sensation 16, Siemens, Germany), and whole body CT angiography tests showed normal findings. Cytomegalovirus, parvovirus, Epstein-barr virus, and Venereal Disease Research Laboratory tests were negative. The patient was consulted to the ophthalmology department. His best-corrected visual acuities (BCVAs) were "light perception" and 20/20 in the right and left eyes, respectively. There were iris pigments on the lens of both eyes; fundus examination revealed papilledema and macular star and edema in the right eye (Figure 1).

Patient was diagnosed with ON and neuroretinitis in the right eye, and 1 g intravenous (IV) steroid treatment was started. Cerebral and orbital magnetic resonance imaging (MRI; Siemens Medical Solutions, Erlangen, Germany) tests were normal.

Colonoscopy showed focal aphthous ulcers in the rectum and skip areas of deep ulcerations with a cobblestone pattern in the transverse and descending colon. Biopsy revealed chronic active colitis with mixed inflammatory cell reaction. Esophagogastroduodenoscopy showed wide ulcerations at the great curvature of the stomach. Biopsy revealed granulomatous gastritis (Figure 2).

The pili structure of the jejunum and ileum was normal in MRI enterography. Endoscopy findings and pathologic investigations were compatible with CD. We started azathiopurine (Excella GmbH, Germany) 2 mg/kg/day and lansoprazole (Sanovel, İstanbul, Turkey) 15 mg/day. Methylprednisolone (Mustafa Nevzat, İstanbul, Turkey) was continued as 48 mg/day orally after 3 days. On the seventeenth DoH, laboratory tests were within normal limits. Patient's abdominal pain and diarrhea were improved but vision loss in the right eye unchanged. The patient was discharged and called for ophthalmologic follow-up after 1 month and gastroenterologic follow-up after 3 months.

At the first month follow-up, BCVA was "no light perception" and 20/20 in the right and left eyes, respectively. Fundus examination revealed optic and macular atrophy (Figure 3).

At the third month follow-up, laboratory tests were within normal limits. The methylprednisolone dosage was decreased to 32 mg/day and tapered 16 mg per week. Azathiopurine 2 mg/kg/day and lansoprazole 15 mg/day were continued. In the ophthalmologic examination, BCVA was no light perception and 20/20 in the right and left eyes, respectively. Fundus examination revealed optic and macular atrophy, which were also confirmed by optical coherence tomography (OCT; Figure 4). Fundus of the left eye was normal.

Figure 3. a-c. (a) Macular OCT of the right eye at the first month follow-up shows foveal atrophy. (b) Fundus photography of the right eye at the first month follow-up shows optic atrophy and maculopathy. (c) Fundus photography of the left eye at the first month follow-up is normal

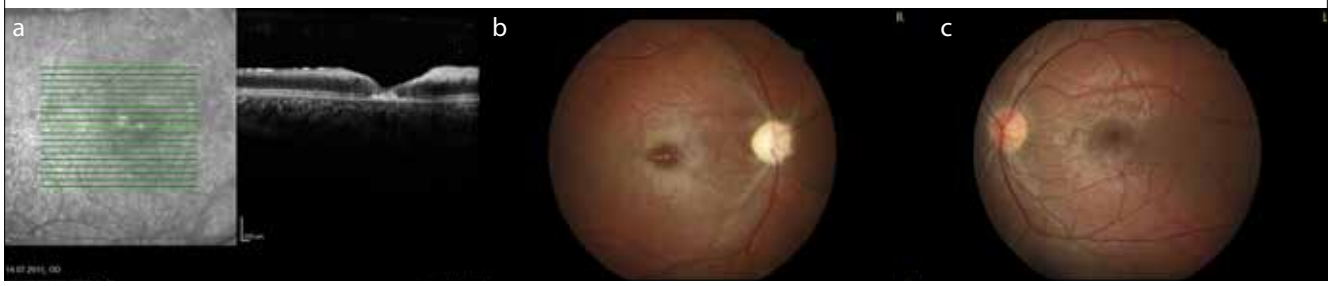
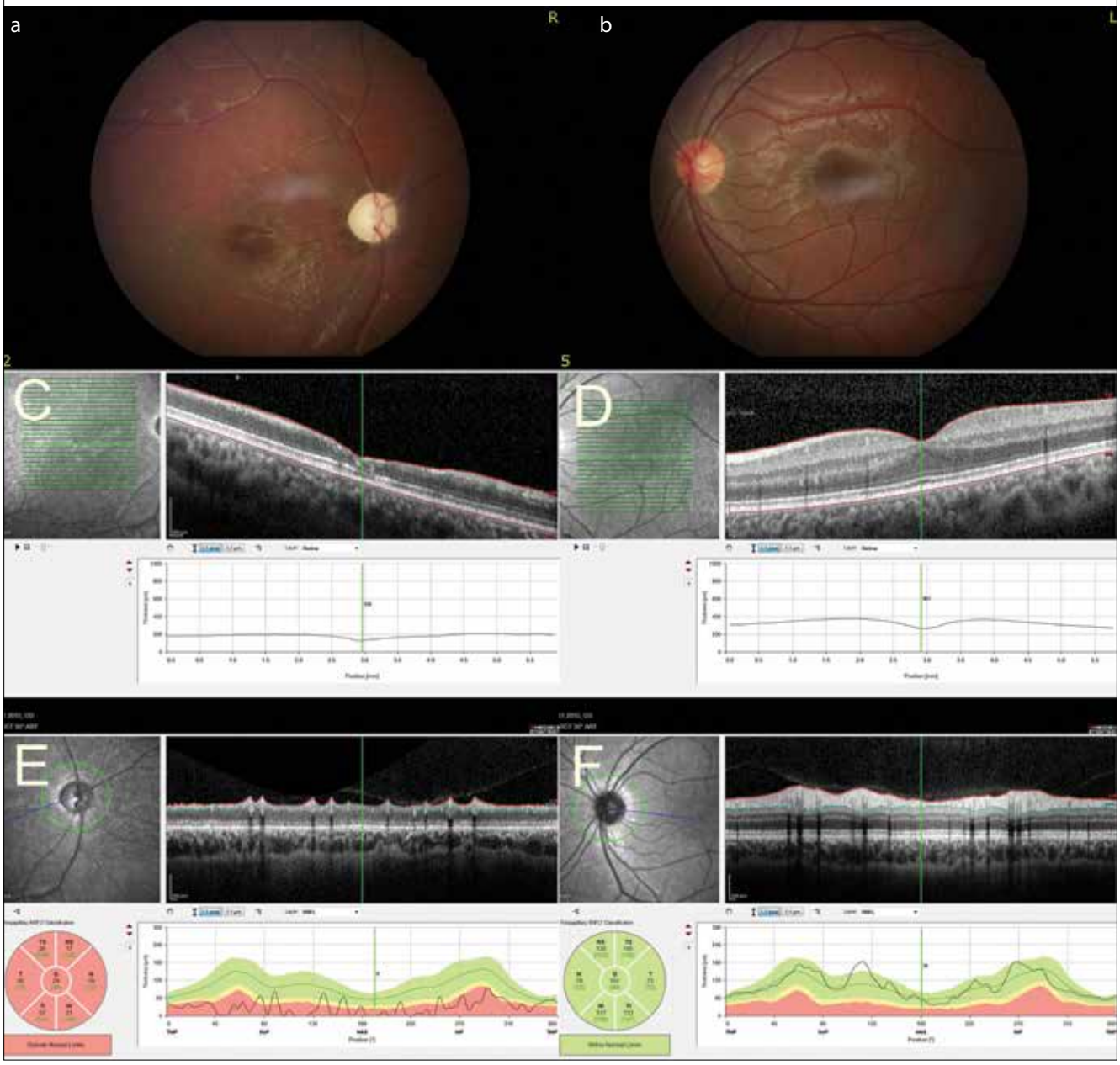


Figure 4. a-f. Ocular findings at the third month follow-up. (a) Fundus photography of the right eye shows optic atrophy and maculopathy. (b) Fundus photography of the left eye is normal. (c) Macular OCT of the right eye shows macular atrophy. (d) Macular OCT of the left eye is normal. (e) Retinal nerve fiber layer (RNFL) analysis of the right eye shows optic atrophy. (f) RNFL analysis of the left eye is normal



DISCUSSION

Ocular manifestation in IBD were reported in 3.5%-43% cases; interestingly, they are more common in colonic or gastric CD cases similar to our case (8). There is limited information on the ocular involvement in adolescent and pediatric patients (9). ON caused by IBDs can be treated with high-dose IV methylprednisolone (10).

We reported an adolescent patient with colonic and gastric CD who presented with severe abdominal pain and diarrhea and developed sudden vision loss in the right eye. The vision loss did not improve despite a 3-day high-dose IV methylprednisolone treatment. In our patient, the most important problem was delayed diagnosis and treatment. Fortunately, all gastrointestinal symptoms were completely resolved with methylprednisolone, azathiopurine, and lansoprazole treatment for CD.

It is unclear if the anti-inflammatory treatment given for IBDs prevents optic nerve inflammation or not; however, a treatment that resolves the gastrointestinal disease may also improve extraintestinal involvement. In our patient regular treatment and follow-up for CD might not be able to improve the vision of the right eye but at least it may be protective for the left eye. Additional studies are needed to clarify the protective effect of anti-inflammatory treatment.

CONCLUSION

Patients presenting with different systemic findings who also have ocular hyperemia, blurry vision, temporary vision loss, retrobulbar pain, or ocular motility disorders require extra attention, and the ophthalmological examination must be performed carefully to ensure that the diagnosis and treatment is not delayed. To best of our knowledge, this is the second report about optic nerve involvement in CD in the pediatric and adolescent population in literature (11).

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