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(CASE REPORT)

Chronic myeloid leukemia revealed by bilateral proliferative retinopathy: A case report and literature review

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Abstract

Bilateral proliferative retinopathy is a rare complication of chronic myeloid leukemia (CML) as a few case reports have been published to date. In this case report, a 32-year old diabetic female presented with history of bilaterally decreased vision. Ophthalmologic examination showed bilateral proliferative retinopathy (i.e., retinal detachment, vitreous hemorrhage, pre-retinal fibrosis and the presence of bilateral peripheral capillary dropout with multiple retinal sea fan neovascularization) for which panretinal laser photocoagulation and vitrectomy were planned. During the preoperative workup, complete blood count revealed hyperleukocytosis. Later on, the karyotype analysis identified Philadelphia chromosome, confirming the diagnosis of CML. Hence, it was an interesting case where bilateral proliferative retinopathy directed to the diagnosis of CML. Therefore, proliferative retinopathy may be the first presentation of CML.

Keywords: Proliferative retinopathy; Chronic myeloid leukemia; Case report.

1. Introduction

Chronic myeloid leukemia (CML) is a myeloproliferative disorder, contributing to 15% - 20% of all adult leukemias [1]. Globally, the annual incidence of CML is reported to be 1-2 cases per 100,000 adults [2]. Only in United States, 8490 new cases and 1090 deaths were estimated to occur from CML in the year 2018 [3]. Similarly, the prevalence of CML is estimated to be 181,000 in the year 2050 worldwide [4]. Although the prevalence of CML is rising; however, its survival rate has improved since mid-1970s owing to the development of advanced therapeutic strategies [3]. About half of the patients with CML remain asymptomatic, often being suspected and diagnosed on routine investigations [5]. Most common symptoms are signs of the patients with CML are fatigue, malaise, weight loss, anemia and splenomegaly [5]. Reciprocal Philadelphia chromosome translocations are manifested by 90% cases of CML [6]. Current treatment of CML includes tyrosine kinase inhibitors (TKIs) such as Imatinib [5].

At the back of eye, visual pathway starts with retina that possesses photoreceptors for visual transduction. Pathological alterations in retina caused by a variety of conditions results in retinopathy– unilateral or bilateral. Diabetes mellitus and hypertension are major causes of retinopathy [7]. However, bilateral proliferative retinopathy is a rare complication of CML as a few cases have been reported in the literature to date. Retina is the most common intraocular part affected by CML in terms of vascular abnormalities such as tortuous vessels, retinal or vitreous hemorrhage, microanevrysms and neovascularization [8]. This is an interesting case report as the visual manifestations of bilateral proliferative retinopathy led to the diagnosis of CML.

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2. Case presentation

A 32-year old diabetic female presented to us with history of gradual but progressive onset of decreased vision in both eyes. The decreased vision in right eye started a year ago while in the left eye it was marked for the last three months. She was on Insulin therapy since her diagnosis of diabetes type 1 (T_1DM). On examination, the patient was vitally stable. Ophthalmologic evaluation revealed a Best Corrected Visual Acuity (BCVA) of 2/10 in the left eye and hand motion in the right eye. Fundoscopy showed retinal detachment, vitreous hemorrhage, pre-retinal fibrosis and the presence of bilateral peripheral capillary dropout with multiple retinal sea fan neovascularization. The ophthalmic surgery was planned and preoperative work was advised. Complete blood count (CBC) revealed hyperleukocytosis at 165 x 10^9/L, thrombocytosis at 557 x 10^9/L, and hemoglobin at 10 g/dL. Random blood sugar (RBS) and glycosylated hemoglobin (HbA1c = 6.0%) levels were within normal limits. Liver function tests (LFTs), renal function tests (RFTs) and serum electrolytes (S/E) were normal.

Peripheral blood smear showed marked leukocytosis (neutrophilic polynucleosis) with granulocytes in various stages of maturation with an increased number of immature myeloid cells at 31% (myelemia), as well as, excess of basophils and eosinophils. Circulating blasts were only 1% (Figure 1). The bone marrow smear confirmed the chronic phase by showing only 2% of blasts (Figure 2). The patient received bilateral panretinal laser photocoagulation and a vitrectomy was performed. On further investigation, karyotype identified Philadelphia chromosome and bone marrow aspirate showed only 3% of blasts. Hence, ophthalmologic manifestations of bilateral proliferative retinopathy led to the diagnosis of CML. The patient was put on tyrosine kinase inhibitor, Imatinib.

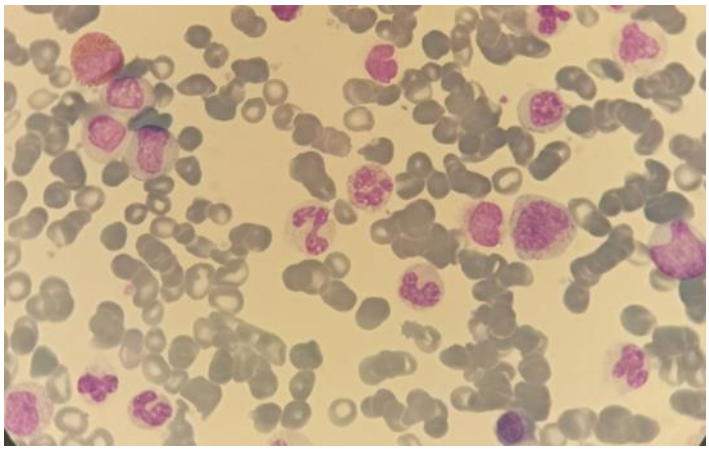

Figure 1 Peripheral blood smear (100×)

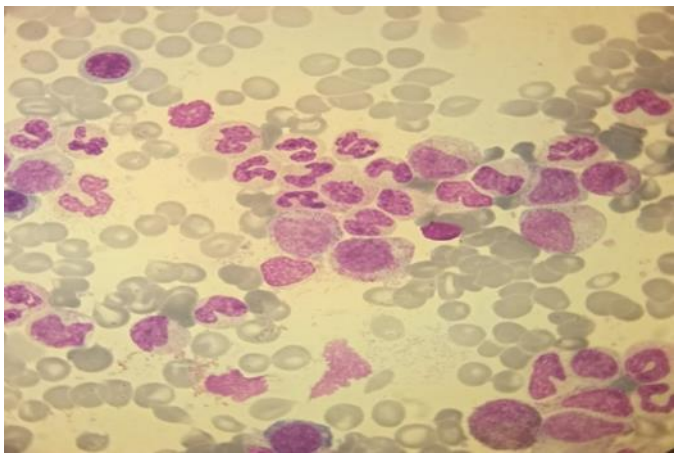

Figure 2 Bone marrow smear (100×)

Following treatment, the proliferative retinopathy completely regressed in both eyes. The patient has been stable for the last 6 months and her BCVA is 20/40 in both eyes at the most recent visit.

3. Discussion

In the present case report, CML presented with bilateral proliferative retinopathy– a rare finding in chronic leukemias. Although retinopathy is a rare complication of CML; however, retina is the most affected intraocular structure in this lethal hematological malignancy [8]. Most often, CML remains asymptomatic, and up to half of the patients have ocular manifestations at the time of diagnosis [9]. Therefore, ophthalmologists may be the first examiners to encounter leukemic retinopathy, suspecting and directing the patients for the evaluation of leukemia.

As mentioned earlier, a few case reports on bilateral proliferative retinopathy have been published so far. Macedo et al. [10] reported a 48-year-old Caucasian male with bilateral proliferative retinopathy whose diagnosis of CML was confirmed on cytogenetic analysis later on. The present case report had similar ophthalmic findings to their study. Rane et al. [11] reported a 51-year old female with CML who initially presented with sudden decrease in vision in left eye. Ophthalmic examination revealed bilateral retinal hemorrhages with multiple retinal infiltrates. They confirmed the diagnosis of CML with Philadelphia chromosome through cytogenetic analysis and bone marrow biopsy. Similarly, Mandava et al. [12] reported a 27-year, otherwise healthy man, with bilateral proliferative retinopathy. Fluorescein angiography revealed severe bilateral retinal fibrovascular proliferation, tractional retinal detachment and vitreous hemorrhage, and bone marrow biopsy identified BCR-ABL translocation.

Huynh et al. [13] reported bilateral proliferative retinopathy in terms of mild vitreous, sub hyaloid, intraretinal hemorrhage and segmental perivascular white infiltrates in a 46-year-old man who was recently diagnosed with CML. Recently, Mohamed et al. [14] reported two cases of CML who initially presented with ocular manifestations. In the first case, a 31-year old Egyptian male presented with the complaint of blurred vision in right eye, and Fundoscopy revealed bilateral retinal hemorrhage. In the second case, a 35-year-old male presented with sudden loss of vision. In both the cases, hematology routine tests, peripheral blood film, bone marrow and cytogenetic analysis were suggestive of CML. From India, Priya et al. [15] reported a 58-year-old male who presented with blurred vision and floaters in his right eye. Fundoscopy revealed bilateral retinal and vitreous hemorrhages, perivascular sheathing and retinal infiltrates. On further examination, fundus fluorescein angiography (FFA) revealed bilateral multiple hyperfluorescent dots, neovascularization of the disc and capillary no perfusion areas. They demonstrated that FFA offers a clinical clue for leukemic proliferative retinopathy in terms of a peculiar bumpy appearance of arteries, arterioles and capillaries called as “string of beads” appearance. Furthermore, CBC and bone marrow confirmed the diagnosis of CML. It has been reported that CML disrupts the integrity of retinal and choroid vasculature, hemorrhage, neovascularization, and neoplastic infiltrate [16]. Unfortunately, ocular manifestations in CML are associated with poor overall prognosis [16].

In the present case report, it is necessary to discuss that the patient was also suffering from T_1DM, and diabetes is one of the most common causes of proliferative retinopathy. Previously, it has been studied that CML accelerates proliferative retinopathy in the patients with co-existent diabetes [17]. Undiagnosed CML in a patient with preexisting controlled DM is rarely, the significant fundus findings overlap with DM. However, optimal management of diabetes in terms of normal ranges of RBS and HbA1c suggest the leukemic proliferative retinopathy. In addition, the central mechanism of leukemic proliferative is attributed to retinopathy hyperleukocytosis leading to leukocytosis retinopathy [18].

4. Conclusion

Bilateral proliferative retinopathy can be the first manifestation of undiagnosed CML. Hence, the patients presenting with proliferative retinopathy should necessarily be looked for CML. Therefore, ophthalmologists should be careful when dealing with proliferative retinopathy.

Compliance with ethical standards

Acknowledgments

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Disclosure of conflict of interest

The authors declares that there is no conflict of interest regarding the publication of this article.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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