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Case Report

Bilateral Retrobulbar Optic Neuritis Revealing Type 1 Diabetes Mellitus

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Abstract

Introduction: Optic nerve involvement is exceptional in diabetes mellitus with an estimated frequency of 1.58%. It is largely dominated by ischemic neuropathy and optic atrophy, while retrobulbar optic neuritis (RBON) remains exceptional and unusual. We report a bilateral and simultaneous RBON inaugural of type 1 diabetes mellitus.

Case Report: A 29-year-old man with no medical history was explored for bilateral and sudden decrease in visual acuity. The somatic examination was without abnormalities. The ophthalmological examination noted a visual acuity at 4/10 on the left eye and 7/10 on the right eye and a normal eye fundus. Ophthalmologic explorations concluded to a bilateral RBON. The etiological investigation of this RBON was negative. The pre-corticosteroid check-up showed fasting glycemia at 1.80 g/l, postprandial at 3.2 g/l and HbA1c at 8%. Anti-GAD and anti-ICA antibodies were positive confirming type 1 diabetes. The patient was put on intensive insulin therapy normalizing his fasting and postprandial glycemia. The outcome was favorable with gradual improvement in vision parallel to the equilibration of blood sugar level. Ophthalmologic check-up at one month noted visual acuity at 7/10 on the left eye and 8/10 on the right eye. At 3 months visual acuity was 10/10 on both sides; eye fundus, visual field and visual evoked potential test were normal.

Conclusion: To the best of our knowledge, bilateral RBON associated with a chronic imbalance of type 1 diabetes mellitus was previously reported only once in the literature. Our observation is distinguished by the male sex, the bilateral and simultaneous involvement, and its inaugural character of the disease.

Keywords: *Retrobulbar Optic Neuritis; Type 1 Diabetes Mellitus; Optic Neuropathy*

Introduction

Optic neuritis is a rare condition. The global incidence of unilateral forms of this neuropathy is estimated to be between 0.94 and 2.18 per 100 000 habitant and seems to be similar in all countries and ethnic groups [1]. Simultaneous bilateral involvement remains exceptional and often described as "unusual" during optic neuritis [2,3]: only 0.4% of patients with optic neuritis had bilateral involvement in the Morrow MJ., *et al*. series [3].

Optic neuritis may be the first symptom revealing a demyelinating central nervous system disease (most often multiple sclerosis), or more rarely systemic, dys-immune, or infectious pathology [4].

Bilateral involvement is particularly suggestive of an "unusual" or less classical underlying etiology, such as diabetes [3]. Indeed, optic neuritis can exceptionally complicate some endocrinopathies, especially diabetes and dysthyroidism [5,6]. This involvement can be uni- or bilateral [7] and is often associated with poorly controlled diabetes [8].

Retrobulbar Optic Neuritis (RBON) remains exceptional and unusual as a complication during diabetes mellitus. We report a bilateral RBON inaugurating a type 1 diabetes mellitus.

Case Report

A 29-year-old man, with no medical history consulted in ophthalmology for bilateral and sudden decrease in visual acuity evolving for a week. The somatic examination was without abnormalities. Ophthalmological examination noted visual acuity at 4/10 on the left eye and 7/10 on the right eye and a normal eye fundus. The visual field showed a slight limitation of the nasal field with a bilateral exclusion of the blind spot.

Further specific investigations (electroretinogram, optical coherence tomography, retinal angiography, and visual evoked potential test) confirmed the diagnosis of bilateral RBON.

The etiological assessment of this RBON was negative (inflammatory tests, blood count, liver enzymes, phosphocalcic balance, angiotensin converting enzyme, serum protein electrophoresis, immunological tests, tumor markers, systemic vasculitis markers, viral, bacterial and parasitic serologies, HLA B51 and B27 typing, myelogram, lymphocyte phenotyping, standard radiographs and body-scan, orbitocerebral and medullary MRI, lumbar puncture, salivary gland biopsy, upper and lower digestive endoscopy, dosing of vitamin B12 and folic acid).

Given the negativity of paraclinical explorations, the diagnosis of "idiopathic" RBON was retained and the decision was to treat it with systemic glucocorticoids. As part of the pre-corticosteroid assessment, the blood sugar level was high at 1.9 g/l. The patient was referred to us to control his diabetes before the glucocorticoids treatment.

The clinical examination at admission in our department was without abnormalities. In particular, there were no signs of systemic disease (connective tissue disease or vasculitis). The interrogation found neither consumption of alcohol, tobacco or other toxic substances nor long-term drug intake.

Fasting blood glycemia was 1.80 g/l retested at 1.88 g/l, postprandial at 3.2 g/l, and HbA1c at 8% confirming the diagnosis of diabetes mellitus. Pancreatic imaging was normal. The serum c-peptide level was very much decreased and anti-GAD and islet cell antibodies (ICA) were positive confirming the autoimmune nature of diabetes. Screening for other systemic or organ-specific autoimmune diseases was negative (antinuclear antibodies, anti-soluble antigen antibodies, anti-smooth muscle antibodies, anti-mitochondrial M2, anti-LKM1, anti-thyroglobulin, anti-MPO, anti-phospholipids, and anti-neutrophil cytoplasm (ANCA)).

The patient was treated with intensive protocol combining progressive doses of long-acting and short-acting insulin analogues (complete basal-bolus regimen) to normalize fasting and postprandial blood sugar levels. The outcome was favorable with gradual improvement in vision parallel to the equilibration of blood glycemia. Given these findings, the use of systemic corticosteroids was rejected and the diagnosis of optic neuritis associated with type 1 diabetes mellitus was retained.

Under insulin therapy, ophthalmologic check-up at one month noted visual acuity at 7/10 on the left eye and 8/10 on the right. At three months visual acuity was 10/10 on both sides; eye fundus, visual field and visual evoked potential test were normal.

Discussion

RBON is a very rare clinical form of optic neuropathy: only 5 cases in the Indian series of 83 patients with optic neuropathy of Saxena R., *et al.* 6% with a female predominance [9].

Optic nerve damage is exceptional in diabetes mellitus [10-12]. Its frequency was estimated at 1.58% in the large series of 1700 diabetics of Ignat F., *et al* [6].

Similarly, diabetes is only exceptionally found as an etiology of optic neuritis: 3.7% in the series of 27 cases of optical neuritis of Bee YS., *et al* [13].

The spectrum of optic neuropathies associated with diabetes mellitus remains largely dominated by ischemic optic neuropathy and secondary optic atrophy [6,8]. Papillitis and papillary edema [14], optic neuromyelitis [15], and retrobulbar optic neuritis [8,16] are much rarer.

In diabetics, this optic neuropathy can be isolated or integrated into a more complex presentation of simultaneous damage of several cranial pairs in the same patient [17].

The possible pathogenic mechanisms of this neuropathy during diabetes mellitus are mainly neuro-gluco-toxicity and immunological reactions. Indeed, bilateral optic neuritis was reported concomitantly with a decompensated blood glucose homeostasis in a fourteen-year-old adolescent [18]; and in general, it has been proven in the large review of literature by Callaghan BC., *et al*. that optimal glycemic control can prevent and improve neuropathy in type 1 diabetes [19].

More rarely, an immune-mediated optic neuritis could explain the occurrence of optic neuritis during type 1 diabetes mellitus because of the autoimmune nature of this endocrinopathy (neurological toxicity of diabetes-specific autoimmune antibodies associated with a particular vulnerability of the optic nerve?).

To the best of our knowledge, bilateral RBON associated with a chronic imbalance of type 1 diabetes mellitus was reported only once previously [18]. Our observation is distinguished by the male sex, the bilateral and simultaneous involvement, and its inaugural character of the disease.

The negativity of the etiological investigation as well as the improvement under insulin therapy alone suggests a direct causality.

Conclusion

As rare and unusual as it is, this neuropathy deserves to be known in diabetics given its functional prognostic implications. Our observation is, to the best of our knowledge, the first one to report a bilateral and simultaneous RBON as inaugural symptom revealing type 1 diabetes mellitus.

Conflicts of Interest

No conflicts of interest.

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