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Basedowian Orbitopathy without Thyreotoxicosis: A Case Report from Sikasso Hospital

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Abstract

Basedowian orbitopathy is an autoimmune disease representing the most frequent extra-thyroidal manifestation. Its pathophysiology is imperfectly elucidated and is the result of cross-reactivity between thyroid antigens and orbital tissue. It is most frequently seen in the context of hyperthyroidism (basedow's disease or Graves' disease), sometimes hypothyroidism (Hashimoto's thyroiditis), or euthyroidism. Diagnosis is often straightforward, but management remains difficult. The introduction of simple medical treatment exposes the patient to the risk of recurrence. In Sikasso, there have been few studies of Graves' disease. The aim of this study was to describe the clinical, therapeutic and evolutionary features of Graves' disease in Sikasso Hospital. The patient was a 35-year-old woman with no previous history of Graves' disease and no history of alcohol or tobacco intoxication, consulting for bilateral exophthalmos with moderate pain for 1 year. Endocrine tests were normal (euthyroidism), as was orbital-cerebral MRI, which revealed stage II exophthalmos with no sellar or supra-saltar abnormalities. The main complication was exposure to keratitis. Our patient was treated with corticosteroids. The evolution was favorable under treatment, with no recurrence after one year's follow-up. Graves' disease is the most common cause of hyperthyroidism, but this should not obscure the diagnosis of euthyroidism, as demonstrated in this case report. Exophthalmos remains the main ophthalmological clinical manifestation.

Keywords

Euthyroidism, Basedowian Orbitopathy, Orbital-Cerebral, MRI, Corticosteroid Therapy

1. Introduction

The orbit is second only to the thyroid in autoimmune thyroid pathologies, particularly Graves' disease. The term orbitopathy is more appropriate than ophthalmopathy, since the pathology affects the oculomotor muscles and orbital fat, rather than the eye itself. Orbital pathology can be associated with hyperthyroidism in the majority of cases, but also with hypothyroidism or even sometimes with biological euthyroidism (Means or Saint-Yves syndrome) [1].

Orbitopathy is defined by the involvement of the oculomotor muscles and orbital fat. In the vast majority of cases, orbitopathy complicates Graves' disease.

Orbital involvement occurs in 25% of patients with Graves' disease; versus 75% who will not develop orbitopathy [2].

Rarely, orbital involvement may also occur as part of Hashimoto's autoimmune thyroiditis.

Graves' orbitopathy is clinically apparent in less than one in four patients. It may accompany or precede the disease and corresponds pathophysiologically to autoimmune involvement of the retro-orbital tissues. It is usually bilateral. Characteristic signs are exophthalmos and periorbital edema.

The impact of OB (basedowian orbitopathy) is sometimes major and should not be underestimated. Severe forms can lead to loss of visual function through optic nerve damage or exposure keratitis. The aesthetic consequences are often obvious in these forms and can lead to a significant deterioration in quality of life [3]. We report the 1st case of Graves' orbitopathy in Sikasso Hospital, with no thyroid abnormality, and an orbital-cerebral MRI showing grade II exophthalmos in both eyes, with no sellar or supra-saltar abnormality. The aim of this study is to describe the diagnostic procedures in this case of basedow's disease without thyroid anomaly.

2. Observation

This was a 35-year-old patient, with no particular medical or surgical history, gynecological or obstetrical, regular menstrual cycle, no history of alcohol or tobacco intoxication, drug use or contraception; she had been seen in consultation for bilateral exophthalmos with moderate pain for 1 year (**Figure 1(a)**, **Figure 1(b)**).

Examination revealed bilateral axial exophthalmos with moderate pain on mobilization and discomfort in the form of foreign-body sensation and tingling in both eyes. On clinical examination, general condition preserved weight 40 kg, height 1.49 cm, blood pressure 130/90 mm, heart rate 84 beats per minute, no functional signs of dysthyroidism.

(a) (b)

Figure 1. Bilateral exophthalmos (a) face and (b) profile.

On ophthalmological examination, bilateral, reducible exophthalmos with supra-palpebral retraction and moderate sub-palpebral edema were noted, accompanied by moderate pain on mobilization. There were no oculomotor disorders. In the anterior segment, there were a few superficial punctate keratitises in the eyes.

Visual acuity was 6/10, Parinaud 2, and ocular pressure was 12 mm Hg in both eyes. The fundus was also normal in both eyes.

There were no signs of thyrotoxicosis, no goiter, and the rest of the examination was unremarkable.

A referral to an endocrinologist led to thyroid tests based on TSH (thyroid stimulating hormone), T3 (triiodothyronine) and T4 (tetraiodothyronine) assays, the results of which were normal and thus in favor of clinical and biological euthyroidism.

In view of a normal thyroid workup, a sellar MRI was ordered and revealed bilateral stage II exophthalmos (**Figure 2**), with no orbital anomalies or sellar or supra sellar tumors.

The autoimmune work-up was positive for TRAK.

Thyroid ultrasound and Doppler ultrasound of the supra-aortic trunks were unremarkable. In view of these clinical and paraclinical signs, the diagnosis of basedowian orbitopathy without thyrotoxicosis was made.

Our management was multidisciplinary, with medical treatment involving ophthalmologists and endocrinologists. The patient received corticosteroid therapy at 1 g/kg combined with adjuvant corticosteroid therapy, analgesic treatment such as Paracetamol 1 g/6 hours by mouth, local ocular treatments (sunglasses and ocular surface wetting agents) and regular follow-up.

Progression under treatment was marked by an improvement in physical and functional signs, with visual acuity improving to 10/10 P2, a reduction in palpebral retraction and a clear, transparent cornea, thus motivating a gradual reduction in corticosteroid therapy until its total cessation at 2 months. The second thyroid check-up at 2 months showed normal TSH.

3. Discussion

Basedow's disease can be associated with hyperthyroidism in the majority of cases, but also with hypothyroidism or even sometimes with biological euthyroidism (Means or Saint-Yves syndrome) [1].

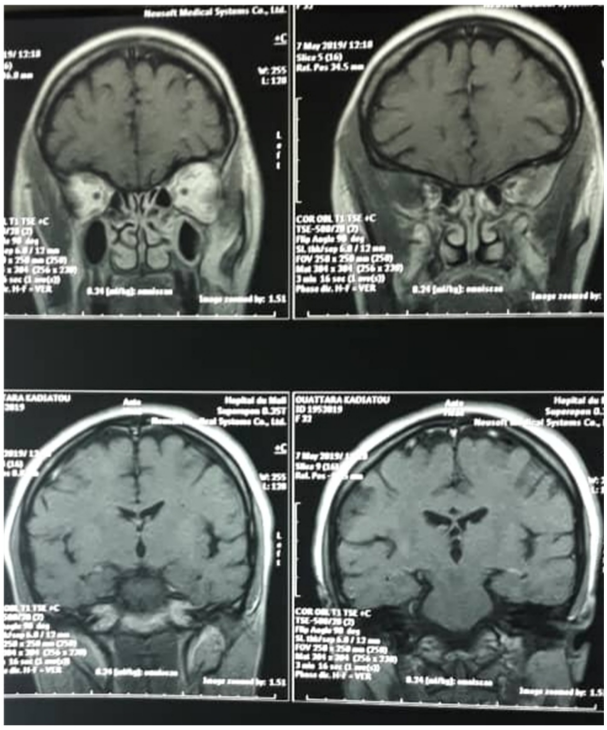

Figure 2. MRI (Stage II bilateral exophthalmos, no orbital abnormalities or sellar and supra sellar tumors).

Jenouiz, Z. [4], in 2018, reported a case similar to ours in the context of a severe Means orbitopathy syndrome, in a 68-year-old woman who consulted for bilateral asymmetric exophthalmos of progressive worsening, evolving for 2 years with diplopia and visual blur. She was clinically and biologically euthyroid. However, she had a history of right loboisthmectomy.

The female predominance of OB has been demonstrated in several studies; we report one case in a female subject. The sex ratio was 86.4% in favor of women in a study in Mali on a series of 815 patients [5], N. Diagne in Senegal found a female predominance of 88% for 108 patients in a hospital study [6], while S. Brah found 94% in the internal medicine department in Niamey, Niger, for 60 patients [7]. N. Daldoul found a male predominance of 54.5%, in a study involving 22 subjects [8]. Despite the existence of several risk factors favoring the onset of OB, we found no evidence of smoking, hormonal contraception or other medications. The occurrence of OB in young subjects has been reported by many authors [5] [6] [8]. Our patient was 35 years old, which corresponds to the average age found by A.K. Koumaré [5], close to the average age found by Nafissatou Diagne [6], *i.e.* 34.6 years, and lower than that of N. Daldoul [8], who had an average age of 40 years in Sousse, Tunisia.

The reason for consultation was exophthalmos and moderate pain. Exophthalmos is known to be the most frequent sign in the diagnosis of OB. Exophthalmos accounted for 93% of clinical signs in S. MBarek's study of a series of 82 patients [3]. The pain was due to exposure keratitis, which accounted for only 1% of clinical signs in Tunisia [3].

A thyroid ultrasound and Doppler ultrasound of the supra-aortic trunks performed on our patient revealed no particularities, namely a thyroid of normal size and echostructure. N. Diagne in Senegal, in a series of 44 patients who underwent thyroid ultrasound, found thyroid hypervascularization in 86.4% of cases [6].

Thyroid ultrasound was performed in 11 patients and revealed 7 cases of multi-nodular goiter (14.29%) [7].

MRI plays an important role in assessing oculomotor muscles, as well as inflammation, fatty infiltration and staging of exophthalmos. MRI revealed stage II bilateral exophthalmos, with no orbital abnormalities and no sellar or supra sellar tumors. Retout [9] found a T2-mode hyper signal in the center of the muscles, indicating muscle edema and an active period of ophthalmopathy. Morax *et al.* [10] found large muscles at the orbital apex, which were responsible for limited eyeball movement, which was not the case in our patient.

Subclinical forms of thyroid orbitopathy are common. MRI or CT scans reveal oculomotor muscle enlargement in 70% of young patients with basedow, whereas clinical manifestations are evident in only 25% - 50% of patients with basedow hyperthyroidism [11].

Basedowian orbitopathy may appear after the onset of basedow's disease, or it may precede it. According to MBarek, basedow's disease may be revealed by ocular involvement in 21% of cases, concomitantly in 45%, or appear after the diagnosis of the disease in 34% [3].

Management was medical and consisted in administering wetting eye drops and boluses of low-dose corticosteroids (solumedrol 500 mg) intravenously for 3 days. This was followed by oral corticosteroid relay therapy at a dose of 1 mg/kg/day for 1 month, then tapering off until the 2nd month, when ophthalmological functional signs improved.

Most teams opt for medical treatment with corticosteroids and external radiotherapy at the inflammatory stage [10].

OB treatment depends on the severity of the disease. It can range from artificial tears in cases of minimal severity to radiotherapy and surgical decompression in more severe cases [12].

Our patient, classified in the minimal severity category, received exclusive medical treatment and endocrinological monitoring due to the clinical and paraclinical signs, with an MRI that revealed stage II exophthalmos with no signs of neurological involvement. Given that basedow's disease can evolve in a wide variety of ways, we continued monitoring with thyroid hormone assays, which showed normal HRT at 2 months.

Functional signs completely disappeared after 1 month of treatment, and continuous ophthalmological and endocrinological monitoring was introduced first every two months, then every six months.

According to MBarek [3], OB is a chronic pathology, which evolves in 2 phases. An active phase lasted up to 1 year or more, and manifested by relapses and spontaneous remissions. A second, inactive and sequellar phase, with fibrosis

leaded to a progressive improvement of the disease, lasting from 1 to 1.5 years.

Basedowian orthopathy without thyrotoxicosis appears to be a rare clinical entity, with a better evolution and prognosis than hyperthyroid patients. This seems to be confirmed by this study, carried out in Korea [13], which compared the clinical characteristics and thyroid-stimulating hormone receptor antibodies in thyroid-associated ophthalmopathy in euthyroid patients with those in hyperthyroid patients; or the authors concluded, euthyroid patients showed less active and severe clinical course, greater unilateral involvement and lower levels of thyroid-stimulating hormone receptor antibodies than hyperthyroid patients.

4. Conclusion

The circumstances in which OB is discovered are dominated by exophthalmos. Despite the predominance of dysthyroid orbitopathy in the etiology of this pathology, we must always bear in mind that the diagnosis must not be obscured by a normal thyroid workup. Diagnosis and early, appropriate medical treatment of Graves' orbitopathy require a multidisciplinary approach.

It is therefore important to inform these patients of the need for good clinical and paraclinical monitoring to avoid complications.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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