analysis was essential for management and definitive diagnosis. However, collection of the specimen was limited by the patient's clinical status and the delicate anatomical location.

In short, invasive aspergillosis of the cavernous sinus is a rare and difficult-to-diagnose condition that should be considered in immunocompromised patients. Imaging and laboratory studies are frequently inconclusive; therefore, we should obtain biopsy specimens of the lesion where possible. Management of these patients requires a multidisciplinary approach involving neurologists, internists, neurosurgeons, radiologists, and pathologists.

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Liquorice-associated bilateral non-arteritic anterior ischaemic optic neuropathy☆

Neuropatía óptica isquémica anterior no arterítica bilateral asociada a consumo de regaliz

Dear Editor:

Atypical non-arteritic anterior ischaemic optic neuropathy (NAION) is defined as those cases that manifest in young patients (<50 years) or those lacking classical risk factors for typical NAION.[1,2]

Atypical NAION may have a more insidious clinical presentation, usually with a milder loss of visual acuity, and with greater frequency of bilateral involvement than typical cases. Cases have been described of atypical NAION related to such drugs as amiodarone,[1] 5-alpha-reductase inhibitors,[3] nasal decongestants,[4] and epinephrine[5]; as well as cases associated with tumours, prothrombotic states, and perioperative bleeding.[1]

We present the case of a young, healthy patient with no history of cardiovascular risk factors, who presented sequential bilateral atypical NAION during a hypertensive crisis secondary to massive intake of liquorice root extract over a period of 5 years.

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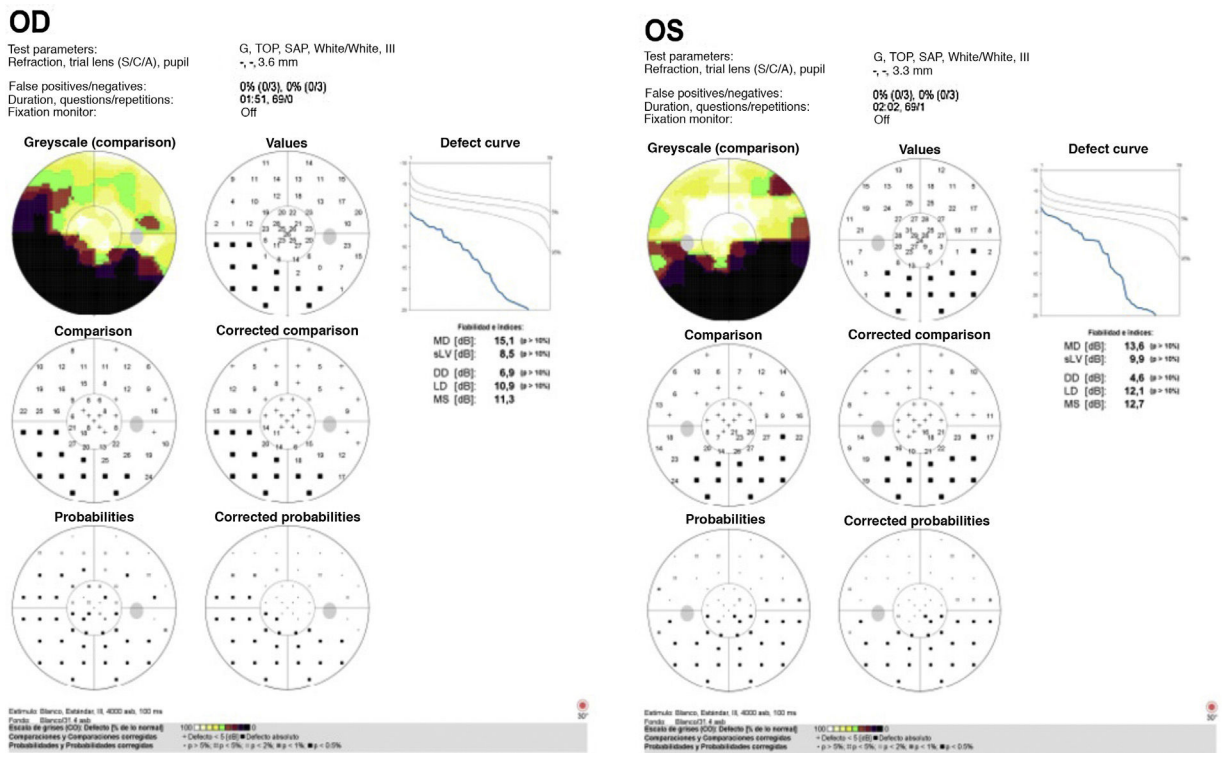

Figure 1 Visual field test showing inferior altitudinal defects in both eyes.

Clinical case

Our patient was a 39-year-old man who in the context of a hypertensive crisis presented altered visual field in the right eye (OD), describing difficulty perceiving images in the inferior field, leading to his referral to the neuro-ophthalmology unit. The patient had no relevant personal or family history. He reported episodes of holocranial headache progressing for months, which improved with analgesics. The ophthalmological examination revealed visual acuity of 0.8 in OD, normal anterior pole, papilloedema with poorly-defined edges in the fundus examination, and 2 flame-shaped haemorrhages in OD; in the left eye (OS), the papilla was apparently normal, with optic disc cupping of 0.2. A visual field test revealed a lower altitudinal defect (Fig. 1) and a blood analysis including complete blood count, C reactive protein, and glomerular sedimentation rate yielded normal results. The absence of any other systemic or neurological symptoms, as well as the normal results obtained in the complementary tests, ruled out other causes of unilateral papilloedema in young patients, including infiltrative, compressive, or post-traumatic neuropathy. The patient was diagnosed with atypical NAION in the context of uncontrolled essential arterial hypertension (AHT). After 5 months of follow-up, the patient visited the emergency department due to similar visual symptoms, but this time involving the left eye. After the examination, he was diagnosed with atypical NAION. During a more detailed history-taking, the patient reported daily consumption of liquorice root extract capsules for 5 years. After examination at the internal medicine department, he was diagnosed with arterial hypertension secondary to primary pseudohyperaldosteronism (pseudo-HAP) due to excessive liquorice intake.

Discussion and conclusions

The isoenzymes 11β-hydroxysteroid dehydrogenase type 1 (HSD11B1) and type 2 (HSD11B2) play an important role in ion regulation and arterial pressure, catalysing conversion between cortisol and its inactive metabolite, cortisone. HSD11B2 is predominantly expressed in tissues sensitive to mineralocorticoids; when locally catalysing the conversion of cortisol into cortisone, it prevents the activation of mineralocorticoid receptors through physiological concentrations of cortisol in the body.[6]

Liquorice contains glycyrrhizin, which is absorbed in the intestine and converted into glycyrrhizinic acid (GA), which competitively inhibits HSD11B2. High concentrations of GA prevent the catabolism of cortisol into cortisone, causing activation of mineralocorticoid receptors, which results in a state of hypermineralocorticoidism that may clinically manifest as AHT secondary to hypokalaemia.[7] The neurological alterations described to date in association with liquorice consumption, including paresis, paralysis, and encephalopathies, are secondary to AHT and electrolyte imbalance.[8]

One gramme of liquorice contains an average of 2 mg of GA; however, this concentration is variable, particularly in extracts such as those consumed by our patient, in which concentrations of up to 98 mg/g have been reported.[6,7]

We suspect that excessive consumption of this extract caused secondary AHT, which predisposed our patient to sequential bilateral atypical NAION.

Our case is particularly interesting due to the unusual association between pseudo-HAP and atypical NAION. We should underscore the importance of detailed history-taking in cases of atypical NAION, given its association with a broad range of drugs, conditions, and clinical situations.

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Myasthenia gravis and systemic lupus erythematosus: presentation of 5 cases and PubMed review☆

Asociación de miastenia gravis y lupus eritematoso sistémico: aportación de 5 casos y revisión de PubMed

Dear Editor:

The contribution of Alba Isasi et al.,[1] who present 3 patients with myasthenia gravis (MG) who developed systemic lupus erythematosus (SLE) after thymectomy, is of great importance given the rareness of this association and the scarcity of research into the causes and triggers of lupus disorder.[2]

We searched on the PubMed database using the keywords SLE and MG, and obtained the following interesting results: the search returned 586 articles, although 483 do not report cases. The first article dates from 1961, although the first case of association of both conditions was published in 1954.[3] Of the 103 articles describing patients, 76 report a single case. We found a total of 180 cases from 32 different countries, mainly in Europe (54 articles/126 cases) and Asia (22 articles/24 cases). Prevalence of SLE in patients with MG was 1.12%-8.4% and prevalence of MG in patients with SLE was 1.3%. MG manifested first in more than twice as many cases; both conditions rarely presented simultaneously. Both adults and children were affected, with the great majority being women. Patients with MG and SLE were younger than those with MG only; they also showed higher prevalence of anti-acetylcholine receptor antibodies, received more immunosuppressants, more frequently underwent thymectomy, and presented a higher rate of remission.

The possible association between SLE and thymectomy in patients with MG is controversial.[4,5] We identified 41 reported cases of SLE onset after undergoing thymectomy to treat MG, with SLE manifesting between 3 months and 40 years after surgery (mean, 10 years). Some studies suggest that thymectomy may play a role in the subsequent development of SLE, due to a defect in lymphocyte suppressive activity caused by decreased thymic hormonal activity, although other authors have been unable to confirm this in experimental studies. The thymectomies performed in some patients with MG and SLE caused no significant changes in disease activity. We should highlight 2 epidemiological studies from Norway and Sweden, which identified 8 patients with SLE among 48 with MG (8.4%) and 23 with SLE among 2045 with MG (1.12%), respectively; no association could be demonstrated with thymectomy.[6,7] Furthermore, Fang et al.[7] indicate that the 3 diseases most frequently associated with MG (polymyositis/dermatomyositis, SLE, and Addison disease) are all regulated by the HLA-B8-DR3 haplotype. In a recent meta-analysis, Chen et al.[8] mention the association of the HLA-DRB1*1602 allele with different autoimmune diseases, including SLE and MG. In our review, most cases of SLE associated with MG were not related to thymectomy.

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