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Case Report

Surgical Drainage *Plus* Optic Nerve Decompression in Acute Optic Neuropathy by an Onodi Cell Mucocele

Tiago Fuzeta Eça[1*], Pedro Correia Rodrigues[1] and Paulo Borges Dinis[1]

[1]Otolaryngology Department, Centro Hospitalar Lisboa Norte, Hospital Santa Maria, Portugal

Abstract

Unilateral acute optic neuropathy, with rapid visual loss, due to an isolated mucocele in a sphenoethmoidal cell, also known as Onodi cell, is an exceedingly rare condition of which no more than a very few cases are reported in the English literature. We describe a new case, in a patient who became blind in just 48 hours but successfully recovered his eyesight after prompt endoscopic endonasal sinus surgery, aimed at both draining the mucocele and decompressing the optic nerve, the later performed as a novel procedure for this specific indication.

Keywords: Acute optic neuropathy; Onodi Cell Mucocele; Optic Nerve Decompression

Introduction

The sphenoethmoidal cell, or Onodi cell (OC), is a posterior-most ethmoid air cell which develops a lateral and/or superior pneumatization that engulfs the optic canal, producing the anatomic variant of an optic nerve bulge in the posterior ethmoid [1-5]. Its prevalence is unknown, but reportedly ranges from 8 to 60% (from data employing various methods of assessment – imaging, intra-operative finding or cadaver dissection), some claiming that is perhaps more frequent in the Asian population [5]. The OC renders the optic nerve vulnerable to posterior ethmoid pathology and to surgical dissection, particularly if the nerve is dehiscent.[2,5] Even if the optic canal is not originally exposed, certain posterior ethmoid pathology, such as a mucocele, which erodes surrounding bone, can eventually create an OC through disease progression, and, by exposing the optic canal, later risking direct damage to the nerve itself [5]. This occurrence is an exceptionally rare cause of rapid visual loss, [1-3]. And, to the best of our knowledge, only a very few cases (we found less than a two digit number of cases in the English literature) have been reported [1-3].

Case Report

A 53-year-old African male attended the emergency department of a tertiary-care hospital due to the rapidly progressive loss of vision in his left eye over the last 48 hours. No nasal symptoms were elicited. Ophthalmologic evaluation at admission revealed a virtually complete loss of vision on the left eye (0/10), with no perception of light, and with anisocoria due to left mydriasis, and an afferent left pupillary defect. Orbit CT-scans disclosed an isolated left posterior ethmoid opacification of soft tissue density, with expansion and erosion of the orbital apex region. An MRI showed that the lesion, hyper-intense on T1 and T2-weighted images, was causing the remodeling of the left lamina papyracea and the optic canal, and was consistent with the diagnosis of a mucocele/pyocele in an OC (fig.1) [4]. The patient was immediately submitted to surgery through an endonasal endoscopic approach, with opening and drainage of the mucocele and optic nerve decompression. Once the mucocele was opened, pus came out and was collected for microbiological analysis, which yielded Morganella morgani and Staphylococcus aureus. The optic nerve was indeed found dehiscent in the posterior ethmoid and nerve decompression was subsequently carried out by gently incising, longitudinally, its myelin sheath, with care not to damage any underlining nerve fiber (fig.2). The patient was immediately started on IV antibiotics and steroids (ceftriaxone 2g, 80mg prednisolone id). On day 4 post-op, pupillary reflexes became normal, whilst left eye visual acuity improved up to 4/10. After 10 days of IV treatment the left eye visual acuity reached 8/10. At 6 months post-op the visual loss reverted completely (10/10), with the only anomaly still found being a reduced amplitude of the visual evoked potentials waves.

***Corresponding Author:** Corresponding Author: Tiago Fuzeta Eça, Otolaryngology Department, Centro Hospitalar Lisboa Norte, Hospital Santa Maria, Portugal, E-mail: tiago.eca@gmail.com

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Discussion

All unilateral rapid visual loss cases due to acute optic neuropathy need to address the possibility that sinus pathology may be at stake, even in the absence of nasal symptoms [1-5]. Sphenoid sinus disease is the most common sinonasal pathology to cause optic neuropathy [4], but it should be reminded that posterior ethmoid disease is also able to damage the optic nerve, particularly if an OC is present [1-3]. An isolated OC mucocele is an exceeding rare condition, and no more than a very few cases are in the literature as cause of acute optic neuropathy.[1-3] In our case, we successfully managed to revert the visual loss by the endonasal endoscopic drainage of the pathology at an early stage of neural suffering. The additional inclusion in the surgical protocol of an optic nerve decompression (fig.2), who's most common indication is traumatic optic neuropathy [5], was not performed on any other of the reported cases, [1-3] and is, admittedly, open to debate. Some may argue that the opening of the myelin nerve sheath not only risks further damage to the nerve itself but also renders it more exposed to local pathogens. Nevertheless, we feel that the complete loss of optic nerve function fully justifies all therapeutic actions aimed at improvement, and the additional potential benefit of having a suffering, inflamed, nerve decompressed out of its constricting myelin sheath makes sense, and is probably worth the extra risk.

This report is exempted of approval by the institutional ethics committee (Comissão de Ética do Centro Hospitalar Lisboa Norte).

Acknowledgement

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Figure 1: Sinus MRI scan, T1-SPIR, showing the hyper-intense left posterior ethmoid lesion, which causes remodeling of the lamina papyracea and the optic canal and compresses the optic nerve.

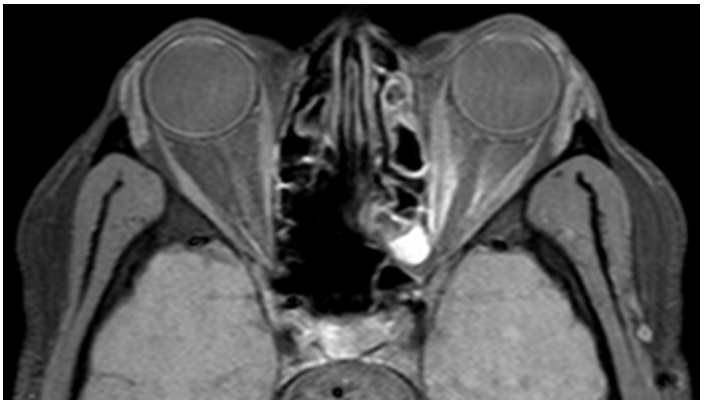
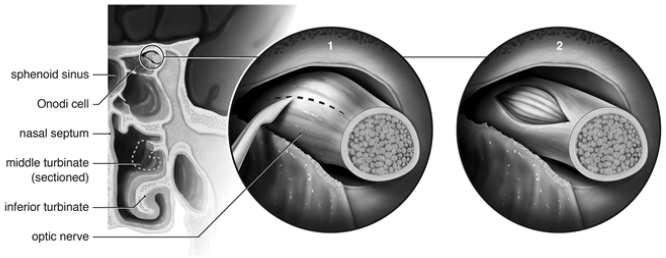


Figure 2: Artist's depicturing of the optic nerve decompression surgery.



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