Silicone oil chiasmal syndrome: an atypical vitreoretinal complication

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Abstract

Purpose: Present a case-based rare post-vitrectomy complication secondary to silicone oil (SO) migration through the visual pathway, to the central nervous system (CNS).
Case report: A 75-year-old woman consulted for acute decreased visual acuity (VA) in her left eye (OS), with history of stable glaucoma but IOP peaks in her right eye and no light perception after vitrectomy with SO 14 months prior. She had bilateral disc cupping and visual field loss compatible with a chiasmal syndrome. Magnetic resonance images showed SO in the visual pathway with progression to the intracranial subarachnoid space and into the ventricles, recovering VA and visual field (VF) in contralateral eye (OS) after the combined antiinflamatory corticotherapy and the ocular SO extraction in the right eye in order to discontinue the leakage inside the CNS.
Conclusions: Silicone oil optic neuropathy may be more frequent than diagnosed. It is therefore advisable to perform urgent neuroimaging studies in

patients with optic disk risk factors (cupping, congenital anomalies) associated to otherwise unexplained visual incoveniences in the fellow eye after a successful vitrectomy, since a neurosurgery could be avoided.
Keywords: chiasmal syndrome; silicone oil; silicone oil migration; vitrectomy; vitreoretinal surgical complications

Síndrome quiasmático por aceite de silicón: complicación atípica de cirugía vitreorretinal

Resumen

Objetivos: Presentar una rara complicación pos-vitrectomía a propósito de un caso con migración de aceite de silicón a través de la vía óptica hacia el sistema nervioso central (SNC).
Reporte de caso: Paciente de 75 años, femenina, consulta por disminución de agudeza visual (AV) en su ojo izquierdo (OI), con antecedentes de glaucoma estable, pero picos de hipertensión ocular y visión no luz pos-vitrectomía con colocación de aceite de silicón (AS) 14 meses previos. Presentaba excavación papilar y alteración del campo visual (CV) bilateral, compatible con un síndrome quiasmático. Las imágenes de resonancia magnética mostraron aceite de silicón en la vía óptica que progresó al espacio subaracnoideo intracraneal y luego hacia los ventrículos cerebrales. Recuperó su AV y CV en el ojo contralateral (OI) luego del tratamiento combinado de corticoterapia antiinflamatoria y extracción del AS intraocular del ojo derecho (OD), con el objeto de discontinuar la fuga al SNC.
Conclusión: La neuropatía óptica por aceite de silicón puede ser más frecuente de lo que se piensa, por lo que se recomienda realizar estudios de neuroimágenes urgentes en pacientes con discos ópticos de riesgo (excavados, anomalías congénitas) y molestias visuales en el ojo contralateral al operado, a pesar de una vitrectomía exitosa, ya que se puede evitar una intervención neuroquirúrgica.
Palabras clave: síndrome quiasmático; aceite de silicón; migración de aceite de silicón; complicaciones de cirugía vitreorretinal; vitrectomías.

Síndrome quiasmática por aceite de silicone: complicação atípica de cirurgia vitreorretinal

Resumo

Objetivos: Apresentar uma rara complicação pós-vitrectomia a propósito de um caso com migração de aceite de silicone através da via óptica para o sistema nervoso central (SNC).
Reporte de caso: Paciente de 75 anos, feminina, consulta por diminuição de acuidade visual (AV) em seu olho esquerdo (OI), com antecedentes de glaucoma estável, mas picos de hipertensão ocular e visão não luz pós-vitrectomia com colocação de aceite de silicone (AS) 14 meses prévios. Apresentava escavação papilar e alteração do campo visual (CV) bilateral, compatível com uma síndrome quiasmática. As imagens de ressonância magnética mostraram aceite de silicone na via óptica que progrediu ao espaço subaracnóidea intracraniana e logo para os ventrículos cerebrais. Recuperou sua AV e CV no olho contralateral (OI) logo do tratamento combinado de corticoterapia antiinflamatória e extração do AS intraocular do olho direito (OD), com o objetivo de descontinuar a fuga para o SNC.
Conclusão: a neuropatia óptica por aceite de silicone pode ser mais frequente do que se pensa, pelo que é recomendado realizar estudos de neuroimagens urgentes em pacientes com discos ópticos de risco (escavados, anomalias congênitas) e desconforto visual no olho contralateral ao operado, apesar de uma vitrectomia com sucesso, já que é possível evitar uma intervenção neurocirúrgica.
Palavras chave: síndrome quiasmática; aceite de silicone; migração de aceite de silicone; complicações de cirurgia vitreorretinal; vitrectomias.

Introduction

The silicone oil (SO) was introduced in 1962 for the treatment of retinal detachment by Cibis *et al*. and is selected according to the vitreoretinal pathology and the technique[1]. It has been described that is well tolerated for up to 6 months.

Later, complications were reported, such as emulsification, keratopathy, cataract formation, high IOP, closure of inferior iridectomies, migration to the subconjunctival space[2], and to the upper eyelid causing ptosis and rarely, retinal toxicity[3-5].

We report a case herein, where not only we describe silicone optic neuropathy as an atypical complication of vitreoretinal surgery, but also how the diagnosis was arisen after recognizing a chiasmal syndrome, and neuroimages were opportunely done and analyzed in order to treat promptly avoiding devastating visual consequences, and also neurosurgical intervention.

Case report

A 75-year-old woman consulted for acute decreased visual acuity (VA) (20/40) in her left eye (OS). She had history of glaucoma diagnosed 20 years ago, trabeculoplasty and cataract surgery in both eyes (10 years ago) and vitreoretinal surgery with SO 1000 centistokes (cs), followed by ocular hypertension in right eye (OD) (40 mmHg) with very poor visual recovery (no light perception), 14 months prior to consultation.

Her IOP was normal (OD 15 mmHg, OS 9 mmHg); with bilateral relative afferent pupillary defect (RAPD), most accentuated in OD, and color vision was impaired OS with the Ishihara color plates. The fundoscopy showed bilateral glaucomatous atrophy, (megalopapillae with 0.9 cup-disk excavation) and generalized pallor, more in the temporal area of the disk, with applicated retina in OD (Fig. 1).

An Octopus G1X visual field (VF) showed amaurosis in OD and inferior-nasal remanent in OS (Fig. 1).

Differential diagnoses of optic neuropathies were considered rapidly: giant cell arteritis was ruled out (normal Erythrocyte Sedimentation Rate and C-Reactive Protein test, normal temporal arteries ultrasonography); and also rheumatology lab routine was done arousing negative results.

A brain magnetic resonance image (MRI) study ruled out a sellar tumor, but the findings were the most unusual: Fluid-Attenuated Inversion Recovery (FLAIR), T1 and T2 scans showed hyperintense material in the right optic nerve as chiasmal region, and this material showed to be hypointense in T1 images with fat suppression and gadolinium (both characteristics of the silicone oil); in the right eye and optic chiasm (Fig. 2a-b).

After 1 month of observation and corticotherapy (4 intramuscular dexamethasone injections separated for 7 days), a surgery was performed to extract the intraocular SO from OD. After that, the MRI findings showed migration of this substance to the cerebral subarachnoid space (Fig. 2c), and a notorious increase in OS VA to 20/20 as well as her VF recovery, from a week after the initial treatment (only superior arcuate scotomatous defect, related with her previous glaucomatous disease) and still stable after a year-follow-up.

Discussion

After an extensive literature review, we can say that intracranial (subarachnoid and intraventricular) migration of SO from the vitreous cavity of the eye is a rare phenomenon, with approximately 25 cases reported in the literature[6], firstly reported in 1983 by Ni *et al.*, intravitreal SO has been demonstrated in pathological specimens in the retrolaminar optic nerve causing visual loss due to an optic neuropathy[7].

In this article we report a new case with a chiasmal syndrome presentation (worsening in VA, temporal hemianopia and infero-nasal remanent, pupillary reflex and color vision impairment, as well as optic disk pallor).

As Boren *et al.* described, the retrolaminar migration of intraocular SO can be presumed to be related to the dimension of the optic disk (big disks), high IOP, and the determining fact of a vitreoretinal surgery with SO, which stimulates the leakage through the lamina cribosa[6].

Another case described the same pattern that also affects the retrochiasmal optic tract[8] and Eckle *et al.* also report a similar case: a 66 year-old-man that had a vitreoretinal surgery secondary to retinal detachment in OS with the introduction of SO with atrophic and glaucomatous optic disk[9]. The patient suffered a VF defect in the fellow eye (although VA was 20/20) in association with chi-

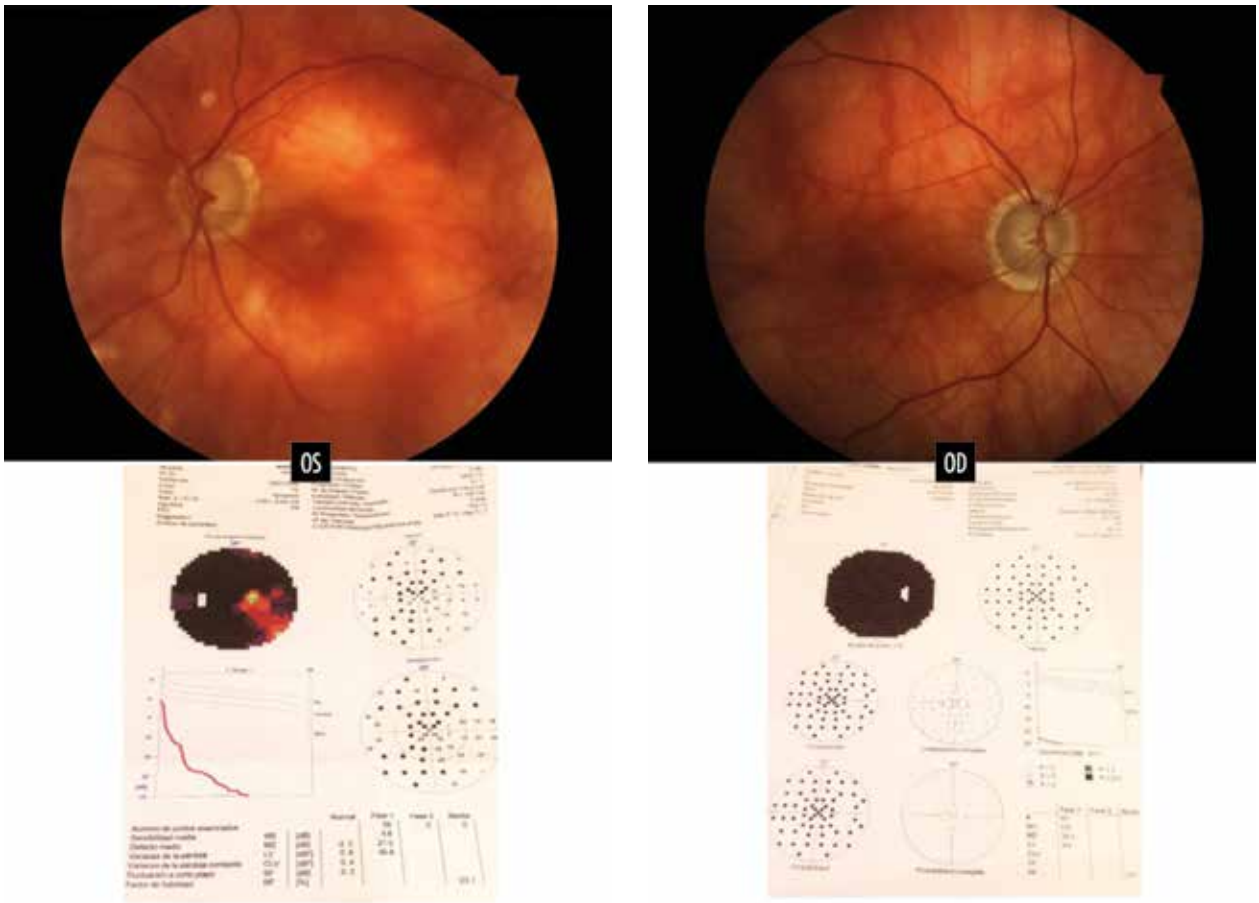

Figure 1. Retinography and VF OS and OD.

asmal migration of intraocular SO, and resulted in neurosurgery (which was the *principal difference* with the present case who recovered with systemic corticosteroids): left optic nerve sheath fenestration, with posterior suction of the SO and irrigation. And VF was recovered after surgery.

The pathophysiology is also uncertain. Possibly, the SO penetrates the subarachnoid space through the laminar holes and dehiscences (demonstrated on behalf optical coherence tomography [OCT] in glaucomatous eyes), or through the space where the vessels penetrate the optic nerve (both central retinal artery and central retinal vein). This route was also proposed as an explanation for cases of Terson's syndrome, as well as the pseudo-Schnabel's degeneration, where the lamina cribosa reflects the infiltration of silicone vacuoles. The Schnabel's degeneration itself is the ischemic necrosis of the retrolaminar optic nerve in acute glaucoma creating cystic spaces; that filled with vitreous humor create a reflection in this location. Carol L. Shields *et al*. in 1989 explains why many retinal detachments are successfully operated, but have poor vision, remarking the idea that "in glaucomatous eyes, intraocular SO appears to have a worrisome potential for posterior migration into the central nervous system (CNS)"[10].

OCT images can actually help us to recognize a possible infiltration of this substance in the retina, since it's been described that there could be seen different findings such as thinning of the ganglion cell layer and microcystic changes in the inner nuclear layer of the retina, even thought not specific for SO, since it could also be seen in other eti-

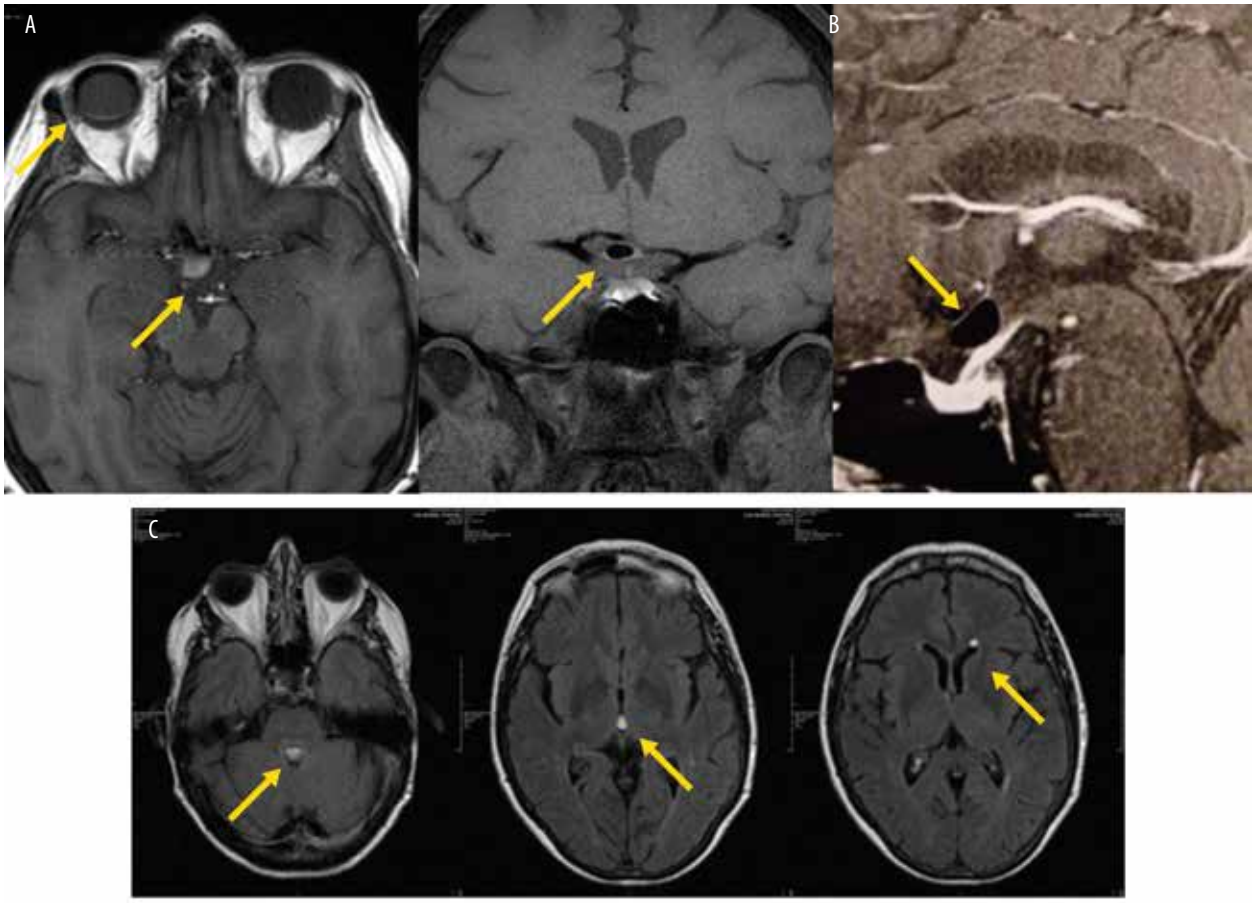

Figure 2. Brain MRI. (A) Axial T1-weighted image showing SO in right vitreous cavity, optic nerve and chiasm. (B) Coronal and sagittal T1-weighted, fat suppressed and gadolinium images showing SO inside the chiasm. (C) Axial T1-weighted images with the SO in fourth, third and lateral ventricles. Arrows indicate the location of silicone oil.

ologies such as Leber's hereditary optic neuropathy and multiple sclerosis-associated optic neuritis[11-12]. However, neuroimaging would be crucial in this case for the specific detection of the infiltration of SO in the CNS. Pathognomonic imaging findings should be taken into consideration, mostly when differentiating from blood, and in cases where there is the precedent of a retinal detachment surgery. Blood can be determined as 30-60 Hounsfield units (HU) and SO on average of 82 HU in computed tomography (CT) images. The appearance of intraocular SO has been described to be hyper-intense relative to contralateral humoral vitreous on T1 weighted and hyper-, hypo- or iso-intense on T2 -weighted MRI sequences, these variations probably due to the different viscosities of the oil (more viscosity will be more hypo-intense on T2 scans), and therefore since our patient received 1000 cs SO, it appeared more hyperintense in T2 scans, with respect to 5000 cs SO tamponades[13]. Several MRI protocols are used to determine SO detection and include short T1 inversion recovery (STIR: single tau inversion recovery sequence) or FAT-SAT which suppress the signal originating from fat leaving the water signal unaffected, and T1 and T2-turbo spin echo (TSE) sequences. Silicone has a different relaxation time than the one of tissue-fat, which at 1.5 Tesla has a radiofrequency approximately 440 Hz lower than the frequency of water[14]. In our case, neuroimaging interpretation

was the key to determining the etiology, fat-supression generated hypointense signal (T1 FAT-SAT). *We suggest the best protocol for the detection of silicone oil is fat-suppression fast T1 and T2 weighted MRI images with gadolinium.*

How long should the SO remain in the operated eye is still unclear, especially in patients with high postoperative IOP as well as pre-existing glaucoma, and with optic disk risk factors such as: megalopapillae, optic pit, morning glory. These should be considered for the management of these patients before and after vitrectomy. Nevertheless, there is much more to be studied to understand the pathophysiology of our patient, as well as the characteristics of the SO used in the surgical procedure. Although Hruby Paul *et al.* described headaches secondary to intraventricular SO successfully managed with ventriculoperitoneal shunt[15-16], amongst other authors, it's almost innocuous to the CNS[17], with no significant complications described to the actual moment.

In conclusion, silicone oil optic neuropathy may be more frequent than diagnosed. It is therefore advisable not only to perform a detailed ophthalmologic exam, but also neuroimaging studies in patients with otherwise *unexplained VA or VF loss after successful vitrectomy in the contralateral eye.*

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