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Brown Syndrome from Local Anesthesia for Inferior Orbital Fat Decompression

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Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Conflict of interest: None declared

Patient: Female, 36-year-old
Final Diagnosis: Acquired Brown syndrome
Symptoms: Diplopia • vertical diplopia
Medication: —
Clinical Procedure: Local anesthesia • orbital fat decompression
Specialty: Ophthalmology

Objective: Unusual or unexpected effect of treatment

Background: Vertical diplopia that follows local anesthesia is usually due to inferior rectus muscle fibrosis. Here, we report a rare case of acquired Brown syndrome following local anesthesia.

Case Report: A 36-year-old woman underwent right inferior orbital fat decompression under local anesthesia. On the first postoperative day, she developed vertical diplopia. She had left hypertropia, which increased on left gaze, with limitation of elevation of the right eye on attempted adduction. Forced duction test of the right eye revealed resistance on elevation in adduction. Magnetic resonance imaging showed signal alteration, thickening, and irregularity involving the right superior oblique tendon and trochlea region. The diagnosis of iatrogenic Brown syndrome was made. Then, a single dose of 10 mg triamcinolone injection was given near the intratrochlear region. On follow-up, complete resolution of diplopia on primary gaze occurred 12 weeks after the incident.

Conclusions: The reported case highlights that local anesthesia carries a risk of Brown syndrome. We believe bupivacaine-induced superior oblique hypertrophy is the underlying mechanism. The patient showed excellent outcome after medical management, with no surgical intervention required after 3 months of follow-up.

MeSH Keywords: Brown Syndrome • Diplopia • Ocular Motility Disorders • Strabismus

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Background

Acquired Brown syndrome may be inflammatory, traumatic, or iatrogenic. Iatrogenic causes include scleral buckle, or glaucoma valve implants. However, Brown syndrome is not expected after local anesthesia. Vertical diplopia that follows local anesthesia is usually due to inferior rectus muscle fibrosis [1]. Here, we report a rare case of acquired Brown syndrome following local anesthesia.

Case Report

A 36-year-old woman, otherwise healthy, presented to the oculoplastics clinic with mild unilateral 2-mm proptosis of the right eye. Systemic work-up was negative, including thyroid function test and computerized tomography of the orbit. She underwent right inferior orbital fat decompression afterwards under local anesthesia and masked sedation. Local anesthesia was injected inferiorly and superonasally using a 25-gauge needle with 2% lidocaine, 1: 100 000 epinephrine, 0.5% bupivacaine, and 1500 IU of hyaluronidase. The aim of the superior-nasal injection was to block the infratrochlear nerve responsible for innervating the medial aspect of the lower lid. The inferior oblique muscle was clearly identified and was avoided during fat decompression. About 2.0 mL of inferior orbital fat was removed. Surprisingly, the patient developed vertical diplopia on the first postoperative day. Examination revealed a corrected distance visual acuity of 20/20 in both eyes. On inspection, she was using right head tilt position to relieve her diplopia. On primary gaze, she had left hypertropia of 6 and 12 prism diopters (PD) at near and distance, respectively. The hypertropia increased to 25–30 PD with limited elevation of the right eye on attempted adduction (Figure 1). Moreover, she had a mild degree of 8 PD of exotropia at primary position. In the outpatient clinic, under topical anesthesia, forced duction test of the right eye revealed resistance on elevation in adduction. The rest of the eye examination was unremarkable. She had not had strabismus surgery nor diplopia prior to the surgery.

Magnetic resonance imaging (MRI) of the orbit without contrast showed signal alteration and thickening and irregularity involving the right superior oblique tendon and trochlea region (Figures 2, 3). The diagnosis of acquired Brown syndrome was made based on clinical presentation and MRI finding. Then, in the minor operation room under topical anesthesia, forced duction was repeated several times, and a single dose of 10-mg triamcinolone injection was given near the intratrochlear region. Furthermore, a topical nonsteroidal anti-inflammatory drug and Fresnel prisms were prescribed. Additionally, she was asked to perform elevation exercises at home, which were frequent adduction and elevation of the right eye 3 times per day. On serial follow-up, clinical improvement was noticed 2 weeks after the injection (Figure 4). Complete resolution of head tilt and diplopia on primary gaze occurred 12 weeks after the local anesthesia injection. However, there was residual left hypertropia of 18 PD on left gaze. The patient did not require any further surgical intervention.

Discussion

Strabismus following peribulbar anesthesia is typically vertical and is usually a result of vertical rectus muscles dysfunction. In one report, vertical diplopia was seen in 0.64% of phacoemulsification under peribulbar anesthesia [2]. The reported cases were due to inferior rectus restriction caused by peribulbar anesthesia. In this case, we describe an unexpected case of Brown syndrome following local anesthesia. The reported case highlights that local superonasal anesthesia carries a risk of Brown syndrome.

Vertical diplopia following local anesthesia may be explained by different mechanisms. First, this could be attributed to the myotoxic effects of anesthesia. Carleson et al. reported that injection of local anesthesia into the extraocular muscles of monkeys caused muscle fibrosis [3]. Second, another reason could be scarring induced by the needle within the superior oblique tendon-trochlea complex. Another mechanism is hematoma from the ciliary artery causing muscle ischemia [4]. However,

Figure 1. Nine-gaze image showing left hypertropia which increases on left gaze. There is limitation of elevation of the right eye on attempted adduction.

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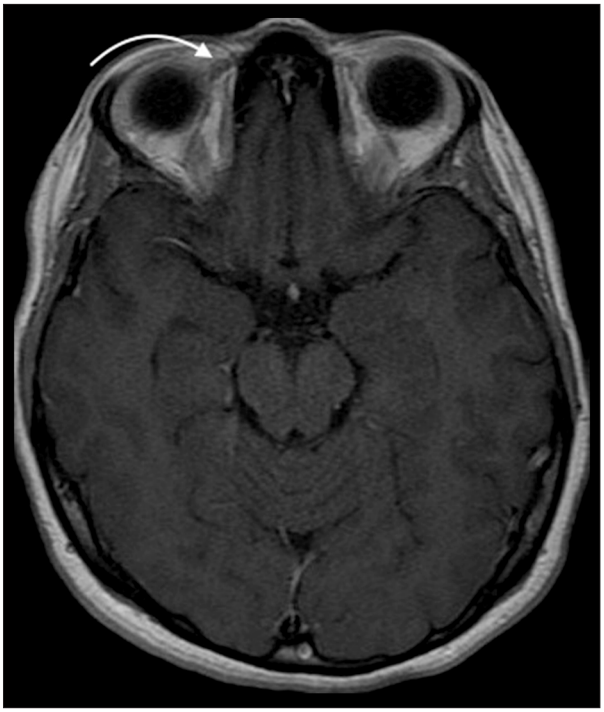

Figure 2. MRI T1 showing thickening and irregularity involving the right superior oblique tendon-trochlea complex (arrow).

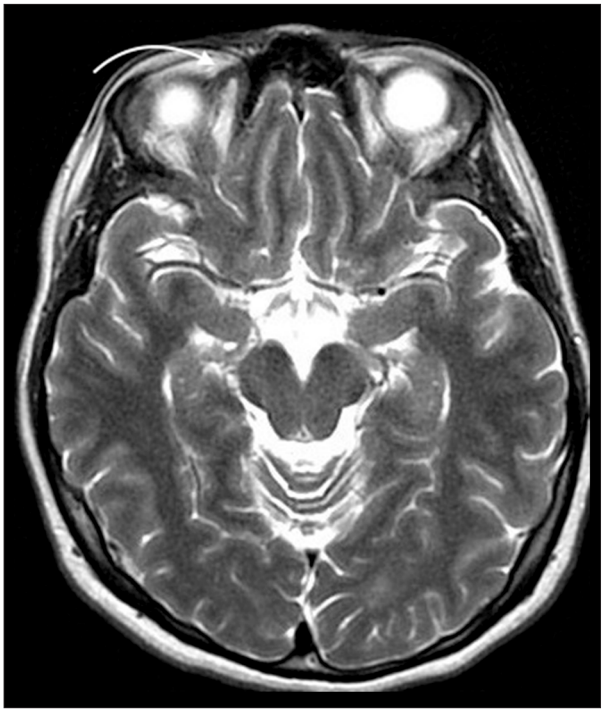

Figure 3. MRI T2 showing thickening and irregularity involving of right superior oblique tendon-trochlea complex (arrow).

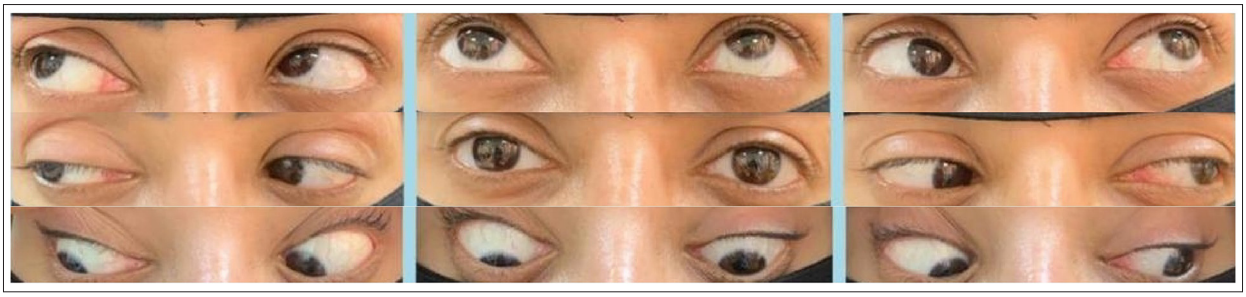

Figure 4. Nine-gaze image showing improvement in vertical deviation in primary gaze 1 month after the incident.

the latter 2 mechanisms are typically seen with deep local anesthesia, as in retrobulbar anesthesia. Since our patient's diplopia started on the first postoperative day and then resolved without any surgical intervention, we think scarring of the tendon-trochlea complex is less likely to be the underlying mechanism. We believe the most likely mechanism in our case is muscle hypertrophy. This is supported by the MRI finding, which showed irregular thickening of the superior oblique tendon. A study by Miller et al. showed bupivacaine causes significant increase in extraocular muscle volume. Maximum muscle volume was noted 10 minutes after the injection and then decreased gradually over many months until it reached pre-injection muscle volume [5].

On review of the literature, we found only 4 cases of acquired superior oblique dysfunction following peribulbar anesthesia. Erie reported a case of acquired Brown syndrome following superonasal peribulbar anesthesia [6]. The anesthesia used was lidocaine 4%, bupivacaine hydrochloride 0.75%, and 1 mL of hyaluronidase. Erie stated the Brown syndrome was a result of scarring within the tendon or trochlea. Phillips et al. reported a case of superior oblique overaction following peribulbar anesthesia [7]. They described a case of Brown syndrome 7 weeks after phacoemulsification surgery under peribulbar anesthesia. The anesthesia agent used was a 50: 50 mixture of 2% lidocaine and 0.75% bupivacaine, with hyaluronidase injected inferotemporally and superonasally. Their patient eventually required superior oblique tenotomy and inferior rectus muscle recession. Hyaluronidase was part of the anesthesia mixture given in our case as well as in the cases reported by Erie and Phillips et al. [6,7]. There is controversy over the role of hyaluronidase in reducing the risk of postoperative diplopia.

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Hamada et al. showed hyaluronidase reduced the risk of postoperative diplopia [8]. It is hypothesized that hyaluronidase causes diffusion of the anesthesia and reduces the adverse effects on the muscles. However, in a larger study on 17 531 eyes, Johnson reported no significant increase in postoperative diplopia during periods when hyaluronidase was not available [9].

Superior oblique hypoplasia is seen in congenital Brown syndrome but not in acquired causes [10]. MRI in our case showed asymmetric thickening of the superior oblique tendon. Lang et al. reported a similar MRI finding in 2 cases [11]. In our case, contrast could have shown better characterization of the superior oblique tendon, similar to that reported by Lang et al. [11]. Although spontaneous recovery has been previously reported in the literature [12], less invasive methods may be initially attempted to treat acquired Brown syndrome, such as local steroid injection or anti-inflammatory agents, as in our case [13,14]. Single intramuscular injection of betamethasone was shown to be effective in a case of inflammatory Brown syndrome [15]. Ravilla et al. reported a case series of 5 children with acquired Brown syndrome of idiopathic and presumed inflammatory etiology, which showed significant improvement following intratrochlear injection of 4 mg betamethasone [16]. In a recent study, intratrochlear injection of steroids was beneficial in the treatment of 11 cases of acquired Brown syndrome secondary to trochleitis [17]. However, there have been no reports on the role of local steroid injection in reversing the adverse effects of local anesthesia on extraocular muscles. We are not sure if peritrochlear steroid injection in our case had any impact, or if the resolution of the patient's diplopia was part of the natural course of the disease. A larger study on the role of local steroid injection in such cases is needed to evaluate whether steroids are capable of accelerating recovery, but performing larger studies may be difficult due to the rarity of this complication.

Conclusions

Brown syndrome is a rare but possible adverse effect of local anesthesia. We believe bupivacaine-induced superior oblique hypertrophy is the underlying mechanism. The patient showed excellent outcome after medical management, with no surgical intervention required after 3 months of follow-up.

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