

Inverse Knapp Procedure for Bilateral Hypoplastic Inferior Rectus Muscles

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ABSTRACT

The case of a 39-year-old woman with vertical strabismus with no concordance between intraoperative findings and the preoperative magnetic resonance imaging (MRI) has been presented here. Orbital MRI revealed apparent agenesis of the inferior rectus muscles. After undergoing surgery a satisfactory postoperative result with reduction of lateral incomitance has been observed.

Keywords: Oculomotor muscles; Inferior rectus muscles; Inferior rectus hypoplasia; Magnetic resonance imaging; Inverse Knapp procedure

INTRODUCTION

A 39-year-old woman presented with incomitant right eye hypertropia (RHT), with bilateral limitation of infraduction and over-depression on adduction of the left eye. Orbital MRI revealed apparent agenesis of the inferior rectus muscles. Surgical exploration of extraocular muscles was performed, where hypoplasia of both inferior rectus muscles was found.

Transposition of right eye horizontal rectus muscles to right eye inferior rectus muscle and fasciotomy of the left superior oblique was performed, obtaining a satisfactory postoperative result with reduction of lateral incomitance.

CASE PRESENTATION

A 39-year-old woman presented to the Strabismus Clinic with RHT and left eye esotropia (ET) since childhood. She used optical correction for ET in childhood and amblyopia of the left eye. On ophthalmological examination, best-corrected visual acuity was 20/20 in the right eye and 20/40 in the left eye.

The cover test revealed incomitant RHT of 25 PD in primary gaze position (PGP) in both far and near vision, 55 prism diopters (PD) in supraversion, and limitation of infraversion. Her ocular motility had under action of the right inferior rectus muscle and right inferior oblique muscle, over action of both

superior recti, and over action of both superior oblique muscles mainly in the left eye producing over-depression on adduction.

Conjugated horizontal nystagmus in extreme horizontal gaze (Figure 1). Slit-lamp examination and fundoscopy were normal. Brain MRI was normal, orbits MRI evidenced the absence of inferior rectus muscles (Figure 2). She had a Neurology consult that ruled-out associated neurological deficit. She was taken to surgery for extraocular muscle exploration.



Figure 1: Pre-operative ocular motility exploration. Incomitant RHT of 25 PD in primary gaze position, with under action of the right inferior rectus muscle and right inferior oblique muscle, over action of both superior recti, and over action of both superior oblique muscles mainly in the left eye producing over-depression on adduction.

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Received date: October 19, 2019; **Accepted date:** November 01, 2019; **Published date:** November 08, 2019

Citation: Grice J, Lonngi M (2019) Inverse Knapp Procedure for Bilateral Hypoplastic Inferior Rectus Muscles. J Eye Dis Disord 4: 130 Doi: 10.35248/2684-1622.19.4.130

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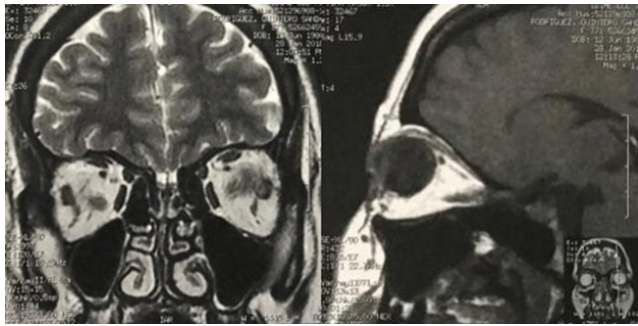


Figure 2: Orbital MRI with the apparent absence of the inferior recti of both eyes. Right, coronal plane and Left, sagittal plane.

Significant atrophy of both inferior rectus muscles was found, with a thickness of 3.5 mm in the right eye and 2.5 mm in the left eye (Figure 3). Transposition of horizontal rectus muscles to the right inferior rectus muscle was performed following the Tillaux spiral (Figure 4), along with fasciotomy of the left superior oblique muscle to reduce incomitance. At one month follow up, the patient had a clear improvement, with an RHT of 5 PD at a distance in PGP and 4 PD in near vision. There was also an improvement in the motility of the inferior rectus muscle in the right eye (Figure 5).

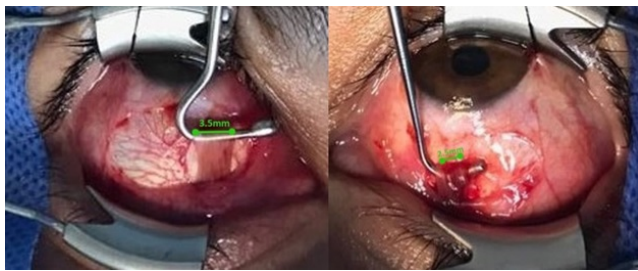


Figure 3: Intraoperative picture showing hypoplastic inferior rectus muscles. Right, right inferior rectus muscle with 3.5 mm thickness. Left, left inferior rectus muscle with 2.5 mm thickness.



Figure 4: Horizontal rectus muscles to inferior rectus muscle transposition following the Tillaux spiral.



Figure 5: Ocular motility exploration one month after the surgery. RHT of 5 PD in PGP at distance and 4 PD at near, with a reduction of lateral incomitance and of the right eye depression.

DISCUSSION

With this report, we introduce an exceptional case due to clinical and imaging features suggesting a bilateral inferior rectus muscle aplasia, with surgical confirmation of bilateral inferior rectus muscle hypoplasia. There are previous reports about patients with aplasia of the inferior rectus reported with higher prevalence than hypoplasia, being frequently associated with craniofacial malformations or Apert syndrome [1].

In general, aplasias of extraocular muscles are rare, being the superior oblique muscle aplasia, the most commonly reported, followed by the inferior rectus muscle [2]. Other authors, have established hypotheses about the origin of these congenital alterations, suggesting that they could result from changes in the development of the mesodermal complex [1,2] with a variable spectrum between aplasia and hypoplasia of extraocular muscles, and being associated to syndromes like Axenfeld-Rieger syndrome or Chitty syndrome, whether they have extraocular manifestations or not [3,4].

Matsuo et al., in 2009, in a case series of 16 patients with aplasia of the inferior rectus muscle found a right eye prevalence of 62.5%, often related to microphthalmia, microcornea, and coloboma. This last one, had a prevalence of 31, 25%, suggesting a strong association between the early closure of the optic fissure and aplasia of the inferior rectus muscle. Also, in that same series, a third of the patients presented heterotropia of the medial rectus muscle, which manifested as esotropias, in addition to alterations given by the aplasia of the inferior rectus muscles, being this the more extensive case series reported [5].

In 2002, Demer reported 261 cases with strabismus that underwent orbital MRI or CT scan, finding inferior rectus muscle atrophy in 4 (1.5%) of those patients. Given these results, the author insisted on the importance of MRI in patients with atypical clinical findings [6]. In 1997 Taylor reported for the first time, the absence of the inferior rectus muscle in an orbital MRI before a surgical procedure [1]. In 2009, Matsuo described the same findings in 2 of 16 patients with similar presentations [5]. However, the number of patients reported in the literature with clinical features suggest hypoplasia of inferior rectus muscle and requiring medical or surgical management have been scarce, and a normative for the care of these group of patients does not exist [1,5].

Our patient had decreased muscle thickness and alteration in vertical movements with downshoots of the left eye that could correspond to an abnormal innervation not previously described in patients with hypoplasia of the inferior rectus muscle. Initially, a complete aplasia of both inferior rectus muscles was

suspected due to MRI. However, the intraoperative findings were compatible with hypoplasia. These surgical findings reporting greater hypoplasia of the left eye, which was in hypotropia, are paradoxical, representing a diagnostic challenge and an uncertain outcome.

Regarding the management of these rare cases, in 2007, Ozkan reported the presence of hypoplasia of both inferior rectus muscles associated with the presence of a retrobulbar accessory muscle, exotropia, upshoots and A-pattern in a 12-year-old adolescent. However, surgical treatment was not described [2]. In 2012, Bathe et al. reported a case of a child with inferior rectus muscle hypoplasia linked to Axenfeld-Rieger syndrome, the patient who had a hypertropia of 40 PD and exotropia of 45 PD.

This patient was surgically treated with a recession of both superior and lateral rectus, achieving a residual hypertropia of 15 PD at six months' follow-up without receiving a second intervention [3]. In addition, anterior transposition of inferior oblique muscle has been performed in patients with inferior rectus hypoplasia, also elongation of the superior oblique muscle associated with recession and infratransposition of the medial rectus muscle for a patient with hypoplasia of the inferior rectus muscle and esotropia, achieving favorable results with both procedures in that kind of patients [7,8].

Our case is exotic and presents in an uncommon fashion being effectively diagnosed intraoperatively rather than by images. This discrepancy suggests that despite its undeniable advantages, MRI is not infallible and can result in misdiagnosis. Second, the present case was successfully treated by performing a transposition of the medial and lateral rectus muscles to the insertion of the inferior rectus muscle in the right eye, and fasciotenectomy of the superior oblique muscle in the left eye. The patient had a favorable result on the primary gaze position with an improvement of lateral incomitance given by downshoot in adduction. This approach is like the one used by Burke, which described an average correction of hypertropia of 19.5 PD with a similar type of transposition [9-15].

CONCLUSION

The incomitant horizontal strabismus with limitation of vertical movements suggests an alteration in the development of extraocular muscles; given an inconsistent result in the ocular motility examination, an orbital MRI should be performed. The procedure performed in our case presents favorable results in patients with hypoplasia of the inferior rectus muscle.

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