



Pulled in Two Syndrome (PITS), a Serious Strabismus Surgery Complication

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Abstract

Aim of the Study: To consider an important and rare complication that can happen during strabismus surgery, to identify the causes, the warning signs, the risk factors so as to prevent it from happening.

Subjects and Methods: This is a retrospective study, studying the files of operated cases of strabismus, and available photos, during the period from January 2010 till January 2021, searching cases of Pulled In Two Syndrome, reporting what happened and cases where this complication could be avoided.

The study included the files of 1610 patients operated by the authors from strabismus in one of the centers of Alexandria, where the details of surgery and complications are monitored, and where most of them had photos for documentation.

Results: Cases of pulled in two syndrome were six cases. This complication was shown in long standing strabismus, specially in adults, where the muscle to be operated was fibrotic, and the minimal pull of the muscle during hooking caused the muscle to be cut spontaneously at the junction of the belly and tendon of insertion. In the earlier cases, the muscle was torn, starting at one edge, and rapidly completely cut in two when pulled with the hook.

Conclusion: This complication should be considered when operating long standing strabismus, restrictive strabismus, Congenital fibrosis of extraocular muscles, and strabismus operated in older age.

Keywords: Pulled in Two Syndrome (PITS); Restrictive Strabismus; Congenital Fibrosis of Extraocular Muscles (CFEOM)

Introduction

One of the serious complications that happened rarely during strabismus surgery, is the spontaneous rupture of the operated muscle under tension during hooking and even (with the gentle pull of the muscle by the strabismus hook, at the junction between muscle and tendon of insertion). In the beginning, in the earlier cases, we accused the assistant of applying extra pulling force over the muscle while using the strabismus hook, but rapidly we could realize that the real cause was unknown, which was probably due to the muscle changes and loss of elasticity in long standing

neglected strabismus cases, in congenital fibrosis of extraocular muscle, in restrictive strabismus. The muscle was spontaneously torn in all cases by the same manner; it started the tear at one edge of the muscle, over the hook, at the junction muscle - tendon, and within seconds continues to the whole junction and the muscle recedes back and lost within seconds. This important complication although rare, but has to be reported, because it happened spontaneously, within seconds, and the surgeon is facing a problem that has to react immediately to prevent its happening, or to change his plan immediately if it happened already.

Aim of the Work

The aim of this work is to focus on this complication that may happen in strabismus surgery, to know the predisposing and risk factors, before surgery, the warning signs during surgery, the ways to react to prevent its happening, and also how to modify the surgical procedures if it already happened and the muscle is partially cut or if is totally cut and lost.

Subjects and Methods

This is a retrospective study, undergone in Alexandria Hospitals, studying the files and records of patients undergoing strabismus surgery, during the period from January 2010 till the end of January 2021. The details of muscle surgery was studied from the records, preoperative data, operative data and available videos, searching for accidental, unplanned muscle cutting during surgery, the common predisposing cause in these patients, and the methods followed to cure them.

Results

This study included the records of 1610 patients, where the details of preoperative presentation, type of squint were documented, also the preoperative surgical plan, the surgical steps, intraoperative complications and management were documented. Many of these patients were having photos preoperative and postoperative, and many had detailed surgical steps.

One important rare complication was searched in these records, it happened that in some rare cases, where the muscle was tight, as in long-standing strabismus, and muscle fibrosis as in Congenital Fibrosis of Extraocular Muscles, that just after exposing and holding the muscle over the strabismus hook, the muscle was torn rapidly at the junction between the tendon and muscle, recedes back and lost.

This was noticed in six cases, one of them was a lady over 50 years, having longstanding esotropia since childhood in left eye, with big angle of esotropia, the plan of surgery was to fully recess the medial rectus by hang-back technique, maximum recession, and to resect the lateral rectus maximum resection. The medial rectus was too tight, dissected and once hooked, is torn immediately and lost. Surgery is stopped at this step, to wait for the clinical picture after this mishappening, but the second day postoperative, the patient came with esotropia again.

The second case was an old man, aged 56 years, with long standing esotropia in one eye, in this case it was written in his preoperative notes : take care when recessing Medial Rectus, hooking gently, no pull. In this case, when hooking the Medial Rectus, even gently, it started to be torn at one edge, the hook was immediately removed and surgery was stopped, with the muscle still attached, postponed for muscle recovery for later surgery. Six months later, this patient was subjected to uneventful Medial Rectus recession with hang-back technique.

One patient, a lady 30 years old, was having long standing left comitant esotropia, operated in another country, presenting with left consecutive exotropia and having a medical report that during surgery, planned left medial rectus recession, the medial rectus was accidentally torn and lost. In this patient, hummelsheim operation was performed with half transposition of medial half of superior and inferior recti muscles and suturing at the insertion of the lost medial rectus (Figure 1,2).



Figure 1: Left MR, PITS.

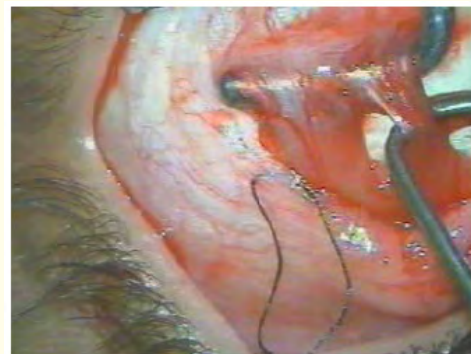


Figure 2: Hummelsheim operation.

One patient, a boy aged eleven years, having fibrosis of Extraocular muscles, CFEOM, with severe hypotropia in both eyes, ptosis and chin elevation, was planned for a primary surgery to correct the eye position, before correcting the lid position. Inferior rectus muscle recession was planned in both eyes to correct the hypotropia. During surgery, once the right inferior rectus was hooked with difficulty, it was spontaneously cut and the muscle escaped. So, in the left eye, the plan of surgery was to disinsertion the left inferior rectus, to have a symmetrical surgery. The inferior rectus was hooked with difficulty, and disinserted slowly by cutting with a super blade over the hook, and living the muscle to spontaneously recede back. Postoperative, the hypotropia was corrected and the chin elevation was corrected, then planned for lid surgery later on (Figure 3,4,5,6).



Figure 3: CFEOM hypotropia.

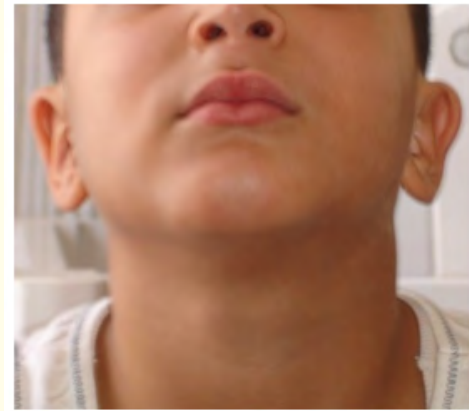


Figure 5: Neck extension.

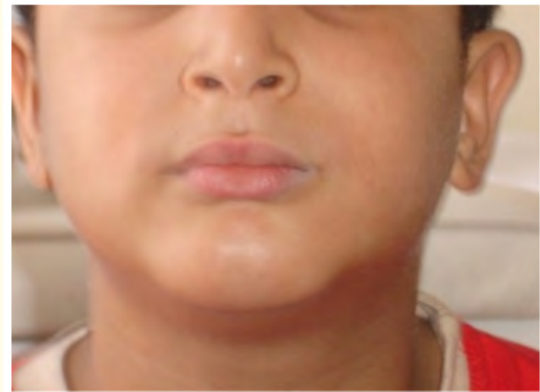


Figure 6: Postoperative.



Figure 4: Postoperative.

Another patient, a lady aged 28 years, presenting with Congenital Fibrosis of Extraocular muscles CFEOM, with bilateral hypotropia and exotropia, forced duction test was positive for both lateral recti and both inferior recti muscles, and bilateral jaw winking (Figure 7,8). In this case, the two inferior recti muscles were hooked without pulling the muscles, were fully recessed by hang back technique due to difficulty in placing of lower posterior scleral sutures in the hypotropic eyes, and performed successfully, also for correction of exotropia in these fibrotic muscles, left lateral rectus recession was slowly hooked, sutures taken by hang back was planned in both eyes, left lateral rectus was recessed by hang back technique, but right lateral rectus, while hooked started to be spontaneously cut, so the hook was immediately removed, and the procedure was stopped to prevent this PITS complication from happening, and the

surgery stopped at this step, and the right lateral rectus recession was postponed to a later time about 6 months (Figure 9) to have the muscle recovered, then recessed successfully by hang back technique (Figure 10).



Figure 7: Rt hypotropia Lt exotropia.



Figure 8: Rt exotropia, Lt hypotropia (alternating).

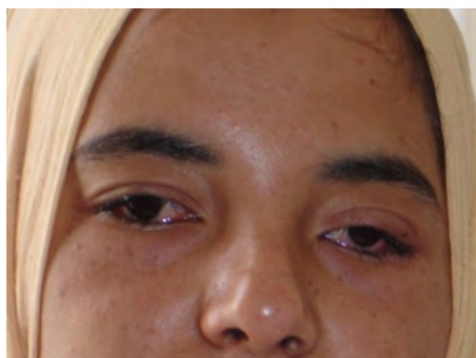


Figure 9: Rt PITS left to recover.



Figure 10: Final postoperative.

The sixth case, was a boy, having long standing left oculomotor nerve palsy, with large angle exotropia, very tight lateral rectus, positive forced duction test, and just hooking the lateral rectus muscle, PITS occurred immediately, no follow up data for this patient, for he didn't show again.

Discussion

Pull in two syndrome PITS, is a serious complication encountered in strabismus surgery when the muscle ruptures under tension. It occurs in tight, fibrotic muscles, when the strabismus hook is inserted underneath the muscle in order to perform the muscle surgery, it was noticed that even a gentle pull on the tight fibrotic muscle, can lead to cut in two at the junction muscle tendon, and so complicating the surgery.

In the current study, this complications occurred in two cases of CFEOM, three long standing strabismus operated in old age, and one case of long-standing oculomotor palsy. The commonly involved muscles were the inferior rectus, then medial and lateral rectus. When comparing with other studies, it was reported by Pujari et al [1] in a case of congenital fibrosis of extraocular muscles. In a multicentric study, a total of 40 cases of PITS from 29 physicians in 6 countries were collected. The most commonly involved muscles were the medial rectus and the inferior rectus. The most commonly identified risk factors were previous ocular surgery and advanced age in patients aged 50 years and older [2,3].

In other studies PITS was encountered in a case of breast carcinoma with metastasis to the medial rectus muscle [4], and in chronic external ophthalmoplegia [5].

The management of PITS in this current study was waiting for muscle recovery and then operating upon. If still partial, the muscle was left to recover and operating later on. If total cut and lost muscle, muscle transposition Was performed. Comparing with other studies, the preferred management for PITS is surgical recovery and reattachment of the muscle; however, if the muscle is lost, transposition surgery or observation are common forms of management [5].

Conclusion

Our data suggests that PITS could be encountered every few years, the risk factors for PITS could be attributed to changes of extraocular muscles in longstanding cases of strabismus, in fibrotic muscles, in previously operated muscles, and surgeons have to be cautious and manage accordingly.

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