Orbital adherence syndrome: clinical characterization and risk factor tracing (retrospective clinical research)

Original Article

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Background: Titanium mesh is one of the most used reconstruction material in the management of orbital floor fracture, with successful reported outcomes. Few reports describe unfavorable tissue reaction to titanium mesh with subsequent diplopia and lid retraction secondary to periorbita adhesions, or as mentioned in literature, orbital adhesion syndrome (OAS). However, there is no accurate description of such condition and how to avoid it.
Aim: This manuscript was made in an attempt to review characterization of this syndrome, associated risk factors, and recommendations to avoid it.
Patients and methods: A thorough revision of orbital floor fracture cases treated with titanium mesh in oral and maxillofacial surgery units in both Cairo University and Ain Shams University between 2015 and 2017 was done according to criteria described by Lee (2009).
Results: Only six cases of 100 cases treated with titanium mesh in the orbital floor and/or medial wall were diagnosed clinically and radiographically as OAS. All cases underwent removal of implant (3–6 months) after primary repair with replacement by polydaxon sheet. Four cases showed clinical improvement in ocular motility within 2 weeks, whereas two cases did not improve.
Conclusion: OAS is an unfavorable tissue reaction to rough surface of titanium mesh, where delayed primary repair and use of large mesh with wide pores in an un-intact periorbita extending to inferior orbital rim are main risk factors. Early detection of this rare complication with removal of titanium mesh is essential for successful management.

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Key Words: orbital adhesion, orbital fracture, postoperative diplopia, titanium mesh.

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INTRODUCTION

Management of orbital floor injuries either alone or in combination with other facial fractures is a challenging task. The aim of treatment is to relieve herniated tissues, regain normal ocular motility, reconstruct floor defect, and restore orbital volume with minimal trauma [1]. In spite of advances in biomaterial available in the market, the ideal reconstruction material is not present yet. Titanium mesh preformed, customized, or manually adapted is one of the most widely used material for orbital floor reconstruction, with reports of high success and minimal complications [2]. Titanium is preferred, despite its known tendency for provoking fibrosis, as it possesses enough rigidity for the support of orbital contents, is easily shaped to each patient's anatomy, is biocompatible, has a low extrusion rate, and displays good osseointegration. However, there are a few reports about unfavorable tissue reaction to titanium mesh (adhesions), with subsequent limitation of inferior rectus muscle and diplopia [3,4]. This was nominated as orbital adhesion syndrome (OAS), which was first described by Lee et al. [5] where fibrinogenic response to titanium has been reported. Titanium forms thin oxide film when exposed to air; this film allows attachment and subsequent proliferation of fibroblasts around titanium. Adhesions of inflammatory cells to titanium surface were proportional to surface irregularities. Even smoothest titanium surface has greater tissue adhesion response than nonporous plastic implant material [6,7].

Clinical reports of this complication were not clear about the diagnostic criteria of such condition perhaps owing to rare nature or being managed by other specialty (occuloplastic). The present manuscript uses the Lee

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description of OAS to retrospectively analyze cases treated with titanium mesh in orbital floor reconstruction in an effort to define its diagnostic criteria and trace possible risk factors.

PATIENTS AND METHODS

A retrospective review was conducted of cases that showed complications after open reduction and rigid fixation of orbital fracture repair using titanium mesh in Department of Oral and Maxillofacial Surgery, Ain Shams University, and Plastic Surgery, Cairo University, using described criteria of OAS (progressive ocular mobility with or without lower lid retraction) over 2 years (2015–2016). A total of 25 cases showed postoperative complication after use of titanium mesh for orbital floor reconstruction. After revision of ophthalmic findings, only 6 of 25 cases met the criteria of OAS. Intraoperative findings and postoperative follow-up of positive cases were analyzed. Thin cuts computerized tomography with three-dimensional reconstructions of both bone and soft tissue windows was achieved. The study protocol was approved by Ethical Committee of Ain Shams University, Faculty of Dentistry, and an informed consent of the details of procedure and use of data for publication was obtained.

RESULTS

Six patients (five males and one female) aged between 20 and 38 years old presented with significant postoperative progressive limitation of ocular movement and diplopia with or without retraction of lower eyelid; coronal computed tomography thin sections were used to confirm diagnosis (Figs 1 and 2). The mean time between trauma and primary surgery was 2 weeks (1–4 weeks) where titanium mesh was used for floor reconstruction. The mean time between primary intervention and presentation of symptoms was 2 weeks. Clinical findings of cases are shown in Table 1. All patients had undergone surgical removal of titanium mesh with or without replacement. Intraoperative findings in each case revealed that there was a dense meshwork of fibrotic tissues that was adherent to and growing through the holes of titanium mesh. Blunt and sharp dissection was done to separate the tissues from the implant. The forced duction test before and after implant removal in each case showed improvement in the movement of the globe. Lower lid correction was performed in three cases (Fig. 3).

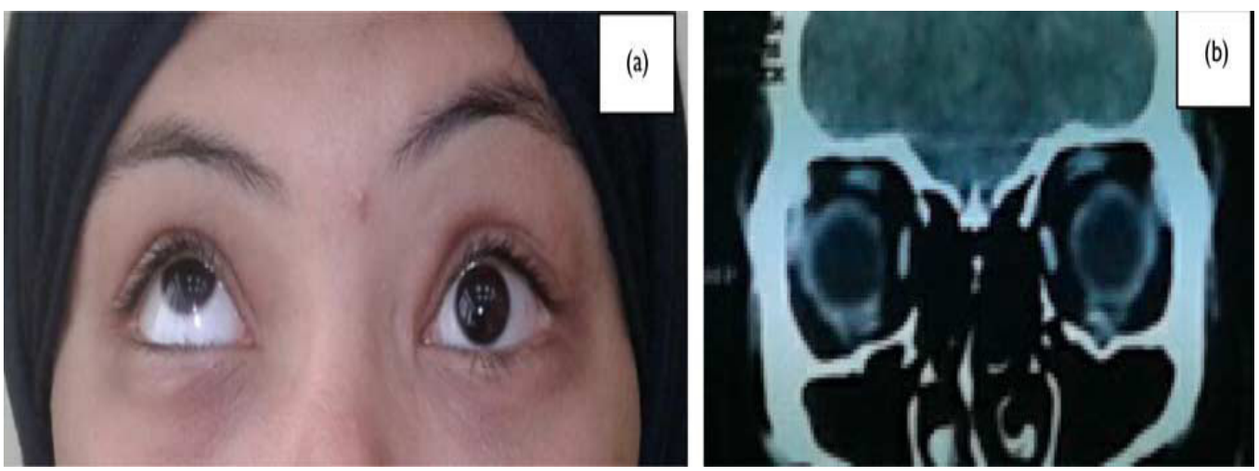

Fig 1: (a) Clinical photograph showing normal lid in spite of severe limitation of ocular motility in upward gaze (left side) in patient no. 4 two months after placement of titanium mesh. **(b)** Coronal computed tomographic view showing adhesions at the orbital floor 2 months after removal of titanium mesh in an attempt to regain ocular motility.

The patients were followed-up for residual diplopia for a minimum of 2 months. Patients reported subjectively regarding an improvement in their gaze restrictions. There was persistent diplopia in extreme up and down gaze. There was no loss or deterioration of visual acuity in any of the patients. One case (no. 4) did not improve after mesh removal and strabismus surgery in the unaffected side.

DISCUSSION

CLINICAL PROBLEM

Patients presented in this manuscript had two chief complaints: lid retraction, which has an issue with esthetics more than function, and diplopia, which is functional rather than esthetic. Persistent or developing diplopia after orbital floor fracture repair has three possible scenarios: irreparable neuromuscular trauma to orbital tissues, incomplete reduction of entrapped orbital tissues, and orbital adherence of implant material used. The first scenario is not surgically correctable; however, incomplete repair of entrapped muscles or induction of OAS through the use of fibrogenic materials is avoidable [8].

Incomplete reduction of herniated tissues is usually excluded by intraoperative forced duction test after finishing reduction and fixation of damaged floor [9].

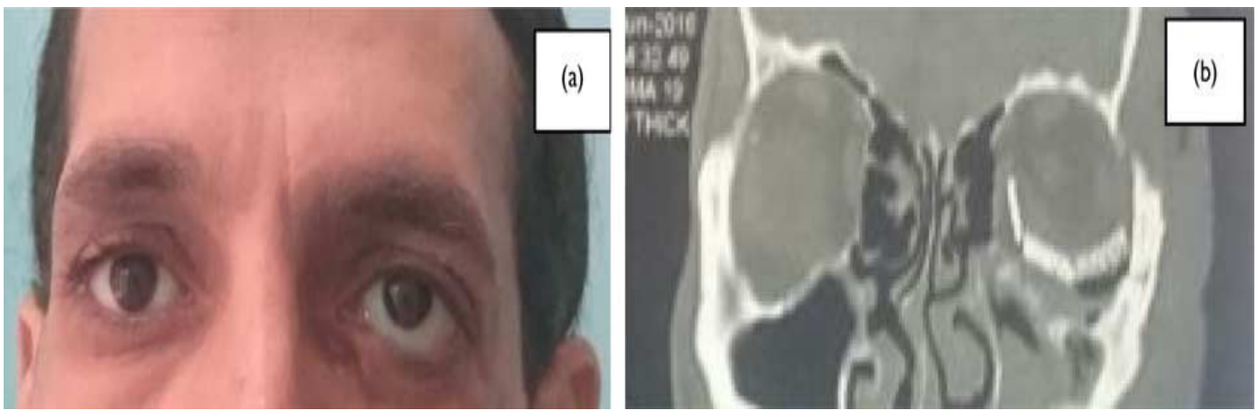

Fig. 2: (a) Clinical photograph showing lower lid retraction in with limitation of ocular motility in upward gaze (left side) in patient no. 6 four weeks after placement of titanium mesh in the orbital floor. **(b)** Coronal computed tomographic view showing soft tissues intervening between inadequately adapted titanium mesh and orbital floor in patient no. 6.

Table 1: Clinical findings of cases that were diagnosed as orbital adhesion syndrome

Patient no.	Site and type of implant	Diplopia after primary repair	Eyelid retraction	Time between injury and primary repair	Secondary surgery
1	Floor	Yes, upward gaze	No	1 week	Mesh removal
2	Floor	Yes, upward and bilateral gazes	Yes	28 days	Mesh replacement and lid repair
3	Floor and medial wall	Yes, upward gaze	Yes	14 days	Mesh replacement and lid repair
4	Floor and inferior rim	Yes, all fields of gaze	No	20 days	Mesh replacement
5	Floor and inferior rim	Yes, upward and down ward	Yes	18 days	Mesh replacement and lid repair
6	Floor and med wall	Yes, upward gaze	Yes	15 days	Mesh removal

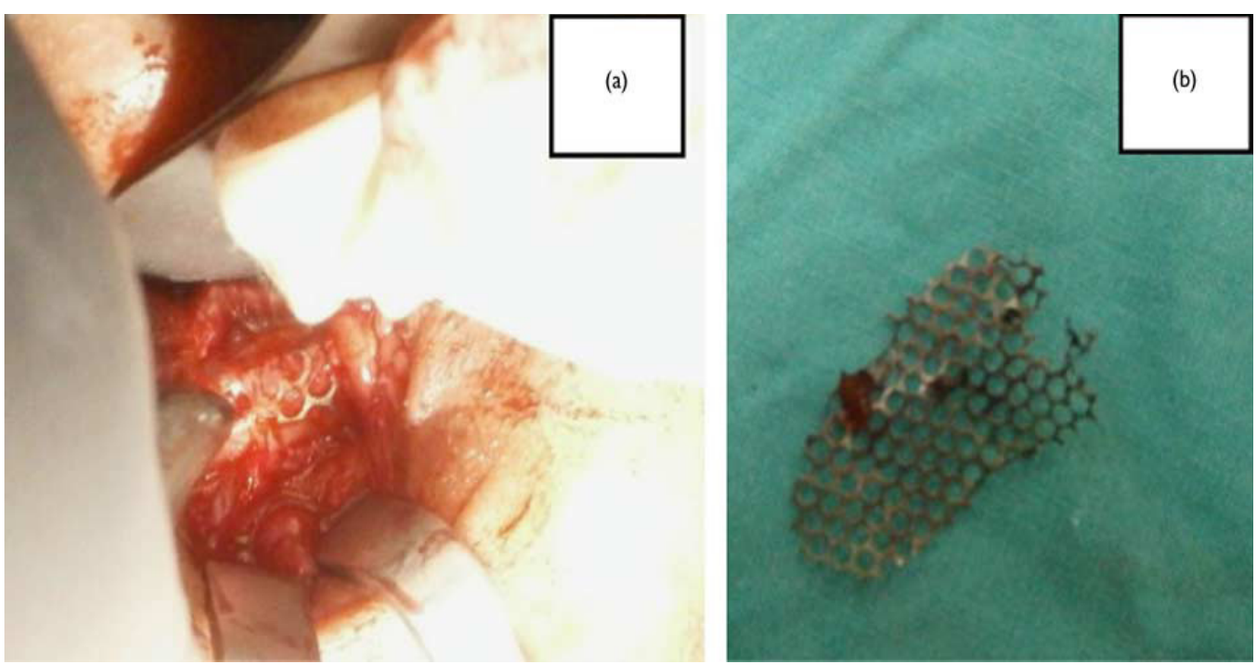

Fig. 3: (a) Clinical photograph of titanium mesh removal with adhesions along floor and inferior rim. **(b)** Tissues adhere to mesh after removal.

However OAS could not be excluded as it is not fully understood complication.

DIAGNOSTIC CRITERIA

Findings from this retrospective study and previous reports suggest the following diagnostic criteria of OAS: worsening of pre-existing ocular limitation with negative intraoperative forced duction test and delayed onset limitation after 2–3 weeks of improved ocular mobility.

Another diagnostic criterion is progressive worsening by time. Moreover, an intense fibrotic adherence is noted between the titanium implant and periorbital tissues during mesh removal. Imaging could be used to confirm suspected cases, including thin-section (1.5 mm) coronal computed tomography, which discloses presence of tissue adhesion; however, characterization of such tissues could not be confirmed without MRI [10,11].

PATHOGENESIS AND RISK FACTORS

Muscle trauma (either original or surgical trauma), incomplete reduction of herniated tissues, and fibrous adhesions of titanium implant are the main scenarios that could be attributed to postoperative ocular motility restriction [5,10]. Cases presented in this article showed definitive criteria of OAS, where restriction of ocular mobility occurs after 1 week or more postoperatively and worsens by time. This clinical finding excludes the first two mechanisms and correlates well with fibrous response to implant. Either muscle trauma or entrapment would be presented as immediate postoperative restriction. Lee and Ho [10] suggested that large pore size of titanium mesh and extension from the floor to rim are risk factors for OAS. The intact periorbita provide smooth gliding surface between ocular structures and bony orbital wall. Accordingly, Lee and Nunery [5] treats one of two reported cases by placement of smooth suprafoil on the top of titaniummesh instead of mesh removal [12]. Another risk factor postulated is time between trauma and primary intervention [7].

PROPHYLAXIS

It has been concluded from the review of previous reports as well as presented cases that several actions could be adopted to minimize the risk of OAS, including minimize time lapse between trauma and repair, use smallest possible available titanium mesh (1.0-mm lowprofile plate) placed as far as possible posterior to orbital rim, minimize dissection of orbital rim, and place smooth interface medium in initial surgery [5,10].

Another option to use titanium mesh without risk of soft tissue adhesion is by combining titanium mesh with porous polyethylene, as the composite becomes radiopaque and more rigid than Medpor of similar thickness, with the added advantages of stability, ease of contouring, and tissue incorporation, which are common to both materials (Medpor Titan; Stryker, Kalamazoo, Michigan, USA) [13]. Placement of smooth suprafoil sheet on top of titanium implant in high-risk cases is another proposal for prevention of OAS [14].

MANAGEMENT

Fortunately, OAS is not a common complication of orbital floor reconstruction, as Lee and Ho [10] reported only two cases from 30 cases treated over 2 years [11]. The low incidence of OAS makes its management unclear without certain guidelines. Previous reports include surgical and nonsurgical management of OAS, without mentioning about indications for each treatment. Nonsurgical management includes steroid therapy, physiotherapy of ocular muscles, lower lid massage, and hyaluronic acid injection. The hyaluronic acid filler acts to restore the volume of the posterior lamella and results in relief of lower eyelid retraction, but it has nothing to do with ocular limitation [15]; accordingly, it is preserved for cases with lower lid problem. Surgical treatment, which was greatly adopted especially in cases with positive forced duction test result, includes removal of implant with or without replacement according to the integrity of floor at time of correction surgery. Early detection of the condition with immediate removal of implant is crucial for restoration of ocular mobility [10,11].

Inherent limitations of this study include its retrospective nature and different primary interventions adopted, which would inevitably lead to a degree of bias. Unluckily, owing to the sporadic presentation of orbital adherence syndrome, and lack of definitive criteria, all published studies on this topic are retrospective, with poorly understood pathogenesis.

Titanium mesh either readymade or customized is used regularly to treat orbital fractures and has shown good clinical outcome when used appropriately [16,17]. However, in highrisk cases of periorbita disruption and orbital fat herniation, it is better to shift to another type of synthetic implant or placement of smooth surface interface on top of the titanium mesh. Early detection of condition and removal of implant especially in cases with positive forced duction test result are very crucial for successful management of such uncommon complication.

CONFLICT OF INTEREST

There are no conflicts of interest.

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