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Review Article

Centurion Syndrome- Rarity Revisited

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Abstract *Centurion syndrome is a rare cause of unexplained epiphora which often goes unrecognized due to a lack of awareness among ophthalmologists. This entity should be considered in the differential diagnosis of causes of epiphora which can be managed surgically. This article is a short review of the available literature on etiopathogenesis, clinical features, diagnosis, and current management of Centurion syndrome.*

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Introduction

Centurion syndrome is a rare idiopathic disorder characterised by abnormal anterior insertion of medial canthal tendon along with a prominent nasal bridge and malposition of the medial part of lid away from the globe displacing the lacrimal puncta out of lacus lacrimalis, resulting in epiphora. It was first described by Sullivan, Welham and Collins in 1993 in an article based on a series of thirteen patients with unexplained epiphora in whom a peculiar anterior insertion of medial canthal tendon (MCT) along with a high nasal bridge was observed. They described these patients as having 'Centurion syndrome' owing to the nasal structural similarity between their patients to those of Roman centurions.[1] Few studies have been published regarding the diagnosis and management of this clinical entity.[2-6] The exact insertion of anterior limb of MCT to the anterior lacrimal crest could be anterior, normal or maldeveloped and can be visualized on magnetic resonance imaging [7] The techniques described in literature for surgical correction are disinsertion/release of anterior limb of medial canthal tendon followed by suturing of the MCT stump to the anterior lacrimal crest. This can be accompanied by adjunctive procedures like canthoplasty, retractor plication and dacryocystorhinostomy. The purpose of this review is to summarize the clinical features which clinch to the diagnosis of this rare condition and further analyse the various surgical techniques utilized to treat this condition.

Material and Methods

An online search was made using the key words "centurion syndrome eyelid" on Google scholar, PubMed and Medline.

Results

A total of 11 articles were retrieved which comprised of 5 case series and 6 case reports. Two articles were excluded considring linguistic barriers of other than English language. A total of 53 cases were analysed on the basis of clinical features, clinical tests performed, surgical procedure and their outcomes and summarized in (Table 1).

Discussion

Various ocular abnormalities may lead to epiphora including hyper lacrimation, eyelid abnormalities, lacrimal outflow obstruction. In this literature review we have described Centurion Syndrome which is a rare cause of epiphora and often goes undetected. It was first published as a series of 13 case reports in 1993 by Timothy J. Sullivan et al. Based on review of medical records of those 13 patients, they reached to the conclusion that epiphora in them was caused due to abnormal anterior insertion of medial canthal tendon which causes inferior punctum to displace anteriorly out of lacus lacrimalis and named the disorder as Centurion Syndrome due to similarity in nasal structure of their patients and the description of Roman Centurions. Two theories have been proposed for pathogenesis- one theory as proposed by Sullivan et al is the abnormal anterior insertion of medial canthal tendon, which has been confirmed based on pre and postoperative (i.e., after performing medial canthal tendon release) findings of lacrimal scintillography and the other theory is based on a more complex anatomic disparity between orbit and its contents.[1]

It is important to look for medial canthal tendon abnormality in patients presenting with epiphora. This disorder is more common in males (89.8%). Age at presentation of Centurion syndrome cases may vary from 1st decade to 4thdecade of life mean age being around 21 years of age. Age of onset varied in different case reports as Sullivan et al noted it to be between 3-10 years of age whereas majority of other case reports mentioned epiphora becoming apparent in 2nd

Figure 1: *Twenty years young male presenting with epiphora with typical ''beak sign''' diagnosed as Centurion syndrome*

decade of life. However, Sullivan et al mentioned that the symptom aggravated in their patients during puberty [1]. The reason being growth of mid facial structures especially frontal process of maxilla and nasal bone during puberty which draws medial canthal tendon further forwards. Epiphora is mostly bilateral, in this review we found that 95% of cases whose laterality is mentioned are reported to be bilateral. Sullivan et al however reported that 15% cases may be unilateral. Prominent nasal bridge is an important factor aiding in diagnosis but its absence doesn't exclude

Table 1: Detailed analysis of various studies related to centurion syndrome

Name of study	Type of study	Total no. of patients	Gender (M/F)	Average age at presentation (years)	Typical clinical presentation (Epiphora, prominent nasal bridge, deep set eyes, displaced lower puncta)	Clinical tests supporting Centurion syndrome (Increased tear film height, delayed FDDT, negative primary Jones dye test, hard stop on probing, patent syringing)	No. of patients who underwent surgical correction (eyes)	Surgical procedure	Outcome (No /persistent epiphora
Sullivan et al (1993)	Case series	13	-	At birth-4 cases 3-10 years-9 cases	Present	Present Special tests- Lacrimal scintillography-10 functional block ROPLAS positive-3 Hertel's Exoph-normal in all	8(12 eyes)	Anterior MCT release with posterior plication of tendon stump	No epiphora (7/8), one required DCR for associated outflow obstruction
							1	Bilateral lateral tarsal strip procedure for concomitant anterior displacement of lateral lid margin	No epiphora (1/1)
							3	Anterior MCT release with DCR due to associated outflow obstruction	No epiphora (3/3)
Sujatha et al (1999)	Retrospective case series	40	38/40	20.5	Present	Present Special tests-none	22	Disinsertion of anterior limb of MCT without posterior plication	No epiphora (22/22)
Ma'luf et al (2003)	Case series	4	4/0	20-33	Present	Present	4(8 eyes)	Disinsertion of anterior limb of MCT without posterior plication	Persistent epiphora (4/4)
Chang (2006)	Case report	1	1/0	21	Present	Present Special test-none	1(2 eyes)	Anterior MCT release with medial lower eyelid retractor plication	No epiphora (2/2)
Huerva et al (2007)	Case report	1	1/0	33	Present	Not mentioned	1	Anterior MCT release and posterior plication of tendon stump followed by retractor plication	No epiphora
Murthy et al (2009)	Retrospective case series	13	10/3	12-35	Present	Present	4	Anterior MCT release alone	No epiphora
							5	Anterior MCT release with conjunctivoplasty	No epiphora
							4	Anterior MCT release alone with punctoplasty	Persistent epiphora (3/4)

the diagnosis. Beak sign is the inferior angulation of medial canthus which can be looked for in suspected cases. Other causes of epiphora have to be ruled out by excluding history of discharge, pain over lacrimal sac area, any ocular trauma or surgery and by performing tests like ROPLAS, syringing or lacrimal passage irrigation, probing of puncta, Jones Primary Dye test. Deep set eyes or enophthalmos has been reported to be associated in 25.7% of cases in this review which contributes to the pathogenesis of disease by pulling the globe further inwards away from the lid by causing relative retro displacement of globe. Sullivan et al however didn't find enophthalmos in their cases and concluded that it only has a minor role to play in the pathogenesis, if at all any.

Tests like Fluorescein dye disappearance test which shows delayed emptying of dye (i.e., persistence of dye beyond 3 minutes), tear film height which is raised, and lacrimal scintillography which shows functional block at medial canthus with hold up of activity before entry into the sac, are performed to help in finalising the diagnosis.

Treatment options include disinsertion of anterior limb of medial canthal tendon with or without posterior plication of the tendon stump with periosteum of lacrimal fossa just posterior to anterior lacrimal crest. Decision to do posterior plication can be made on table during surgery depending on whether adequate retro displacement of lid to globe has been achieved by anterior MCT release alone or not. Similarly, anterior MCT release can be supplemented with conjunctivoplasty to achieve adequate lid globe apposition and align the punctum with lacus lacrimalis. It can also be combined with punctoplasty. This surgery has been proven to be effective in almost all cases without enophthalmos except for one case reported by Huerva et al[6] in which status of enophthalmos was not mentioned. However, it has been reported to be ineffective in patients having enophthalmos as seen in 4 case reports by Ma'luf et al[4] where epiphora persisted after anterior MCT release and in 4 cases mentioned by Murthy et al who also reported 4 of their cases having enophthalmos.

Anterior MCT release along with medial eyelid retractor plication has been effective in both the cases it was performed, one of them having enophthalmos suggesting it as the preferred surgery for Centurion syndrome cases with enophthalmos. Bilateral lateral tarsal strip procedure done in a case having concomitant anterior displacement of lateral lid margin by Sullivan showed successful result.[1] Post op follow up should be done up to a minimum period of 6 months to assess the outcome of surgery subjectively by grading epiphora and objectively by Fluorescein dye disappearance test. Post op lacrimal scintillography can also be done and compared with the pre op findings.

Conclusion

Diagnosis of Centurion Syndrome should be kept in mind while dealing with patients complaining of epiphora. A typical patient is a male in 2nd to 4th decade of life complaining of bilateral epiphora which affects his daily studies or work with the symptom becoming apparent during puberty having a prominent nasal bridge, deep set eyes, forward displacement of punctum out of tear lake and loss of lid globe apposition mostly involving medial side is likely to have Centurion syndrome. General ophthalmologists should be aware of this condition, recognizing and referring them to specialized personnel at appropriate time for surgical correction.

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