· Clinical Research ·

Superior rectus/levator complex in acquired anophthalmic socket repaired with spheric implant—a computed tomography scan and topographic study

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Abstract
● **AIM:** To determine whether the levator palpebrae superioris (LPS)/superior rectus (SR) muscle complex, can influence the position of the upper lid and fornix in acquired anophthalmic sockets.
● **METHODS:** This comparative non-randomized and non-interventional study included retrospective data of 21 patients with unilateral acquired anophthalmic sockets repaired with spheric implants. High-resolution computed tomography (CT) measurements of the LPM/SR muscle complex and clinical topographic position of the upper lid, superior and inferior fornix depth in primary gaze position were evaluated. Demographic data were presented as frequency and percentage proportions and quantitative variables comparing the socket measurements with the normal contralateral orbit was statistically analyzed using non-parametric tests considering $P<0.05$.
● **RESULTS:** The anophthalmic orbits had a significantly shorter LPS length ($P=0.01$) and significantly thicker SR ($P=0.02$) than the normal orbit. Lagophthalmos was present in anophthalmic sockets but not in normal orbits ($P=0.002$), while levator function was normal in both ($P>0.05$, all comparisons). The superior fornix depth was similar in the anophthalmic socket and the contralateral normal orbit ($P=0.192$) as well the inferior fornix depth ($P=0.351$).
● **CONCLUSION:** Acquired anophthalmic sockets repaired with spheric implants have shorter LPS, thicker SR, and more lagophthalmos than normal orbits. The relationship of the LPS and SR with other orbital structures, associated with passive or active forces acting in the final position of the lids and external ocular prosthesis should be further investigated.
● **KEYWORDS:** anophthalmic socket; spheric implant; levator palpebrae superioris; superior rectus; lagophthalmos; fornix depth
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INTRODUCTION

The absence of the globe, or its contents, can lead to anophthalmic socket syndrome, concurring with several anatomic alterations and lid deformities differing greatly from the normal orbit, strongly impacting the patient[1-4]. Postoperative socket changes initiate a complex sequence of interrelationships that affect the appearance, and the function, of the socket and lids[5-8].

Progressive intraorbital volume loss following an enucleation or evisceration procedure due to reduced blood supply and fat atrophy[9-10] results in the rotatory displacement of the orbital contents from superior to posterior, and from the posterior to the inferior orbit[5,11]. There is retraction of the levator palpebrae superioris (LPS)/superior rectus (SR) muscle complex, a downward and forward redistribution of the orbital fat, and an upward movement of the distal end of the inferior rectus muscle, resulting in a shallow inferior fornix and potential tilting of the external prosthesis. The superior palpebrae sulcus may deepen, and the position of the upper lid may be displaced, probably because of the reduced tendon support after the loss of the eye[12-13]. Progressive lid laxity of senescence associated to a heavy external prosthesis can aggravate the inferior lid position even more. Loss of lower lid support and reduced inferior fornix depth make it more difficult to fit and wear an external prosthesis. Insufficient volume replacement, rotational socket content changes, and tendon laxity associated with aging results in the "post enucleation socket syndrome" or "anophthalmic socket syndrome" characterized by a deep superior sulcus, upper lid ptosis, enophthalmic appearance, and lower lid malposition, requiring an even larger than desirable external prosthesis[5,11,14-17].

All the described alterations occurring in anophthalmic sockets post-enucleation without an implant are well documented by computed tomography (CT)[5,11,18]. However, the relationship between the orbital structures as well as changes in the lid and fornix due to the rearrangement of the tissues in the anophthalmic socket after replacing the lost orbital volume using an orbital implant warrants further investigation. The purpose of this study was quantitative analyze the LPS/SR muscle complex and its influence on the position of the lids and fornix in patients with unilateral acquired anophthalmic sockets repaired with spheric implants.

SUBJECTS AND METHODS

Ethical Approval This study was approved by the King Khaled Eye Specialist Hospital ethical board (IRB-1849R) and conducted in accordance with the principles of the Declaration of Helsinki. Informed consent was waived because of the retrospective and anonymous nature of this study.

This comparative non-randomized and non-interventional study included retrospective data of 21 patients with unilateral acquired anophthalmic sockets all repaired with spheric implants, managed at King Khaled Eye Specialist Hospital, Riyadh, Saudi Arabia between 1999 and 2018. The anophthalmic socket was the anophthalmia group and the contralateral normal orbit was considered the comparison (control) group. Analysis was performed using high-resolution orbital CT and clinical topographic analysis.

Inclusion Criteria Patients over 18 years old, with unilateral acquired anophthalmia after evisceration or enucleation, managed with a spheric polymethylmethacrylate (PMMA) implant, with anophthalmic sockets classified as grade 0 (no fornix contraction) or grade 1 (contraction of the lower fornix)[19], having a well-documented CT scan exam.

Exclusion Criteria Congenital anophthalmic socket, previous upper lid or orbital trauma, concurrent myopathies, previous lid ptosis or lid retraction surgeries, or other types of contracted anophthalmic sockets[19].

Parameters of Study 1) CT scan orbit image evaluation: length and thickness of the complex composed of the LPS and the SR and the space between the LPS and the SR were obtained for all patients with the external ocular prosthesis in place. 2) Topographic lid parameters: degree of lagophthalmos, margin reflex distance of the upper lid to the center of the pupil (MRD1), and levator function were manually measured in millimeters (mm) using a plastic malleable ruler (Figure 1A) by one of the authors. The measurements were taken with the external ocular prosthesis in place, in primary gaze position. The absence of lagophthalmos was defined as a lid fissure of 0 with the lids closed; a normal MRD1 was considered 3 or 4, and 10 mm or higher was considered a normal levator function. The contralateral normal side was measured as the control group. 3) Superior and inferior fornix depth: the measurement of the superior and inferior fornix depth in primary gaze position was performed in the affected socket and the contralateral normal eye, in millimeters, using a plastic malleable ruler, after instilling a single drop of topical anesthetic (Minims, oxybuprocaine hydrochloride 0.4%, Bausch and Lomb Inc., Rochester, NY, USA) and removal of the external ocular prosthesis (Figure 1B, 1C).

CT Scan Methodology for Evaluation The anophthalmic and the contralateral normal orbits were evaluated in the sagittal plane by a senior neuro-radiologist (SE), measuring the length and thickness of the LPS and the SR as well the space between the LPS and the SR, representing the amount of fat between the complex of the LPS and SR. CT scan images were acquired with the Discovery 750 HD 64-slice scan (GE Healthcare, Milwaukee, WI, USA). The scanning protocol included 0.2 mm axial, non-overlapping contiguous sections for the orbits, achieved by tilting the patient's head parallel to the Frankfurt plane. Bone and soft-tissue algorithm reconstructions were available for review. For image processing and analysis, all non-contrast CT images were exported in DICOM format into an Enterprise Imaging/Agfa Advantage Window, version 4.6 (Barco, Belgium)[20]. The rules for measurements and anatomical landmarks were standardized. The LPS and SR curved path were measured in the sagittal plane, selecting a standard method to trace the muscle along its course from the origin to the insertion. The SR is identified as a parallel line to the roof of the orbit beneath the LPS from its insertion in the limbus following the curvature of the implant or the globe to its origin in the orbital apex, close to the optic nerve sheath (Figure 1D–1F). Measurements were compared between groups.

Surgical Techniques Unilateral evisceration was performed removing the ocular content, with the placement of a spherical PMMA implant inside the patient's sclera for volume replacement. Unilateral eye enucleation was performed

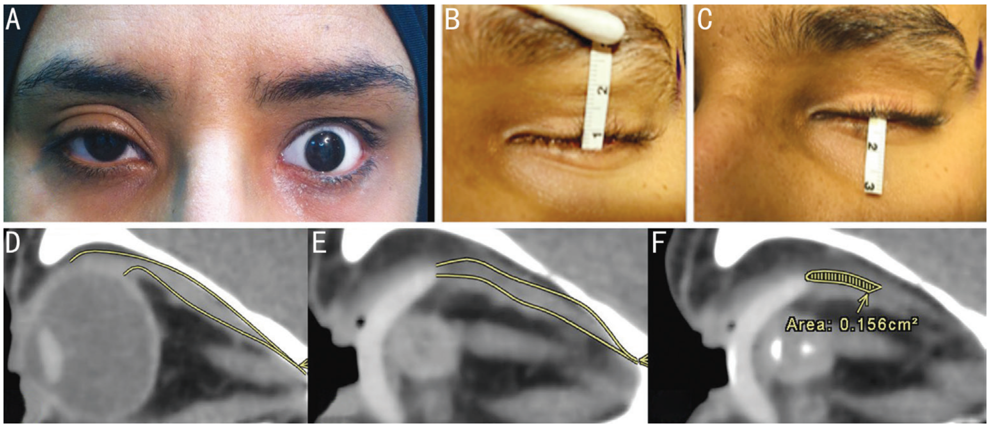

Figure 1 Levator/superior rectus complex in anophthalmia A: Clinical photo of a patient with left side anophthalmic socket and wearing external ocular prothesis presenting upper lid retraction; B: Measurement of the lower fornixes using rulers; C: Measurements of the upper fornixes using rulers; D: Sagittal plane computed tomography (CT) scan of the normal orbit and the levator/superior rectus (LPS/SR) complex along its course from the origin to the insertion; E: Anophthalmic socket with implant and the schematic representation of the curve distance measurement tracing the SR from its insertion in the sclera and following the curvature of the globe; F: The SR parallel to the roof of the orbit beneath the levator palpebrae superior (LPS) muscle in a schematic representation of measurement area of the LPS/SR complex.

Table 1 Comparison of the computed tomographic scan measurements and clinical parameters obtained in anophthalmic socket and contralateral normal eyes Median, IQR

Parameters	Anophthalmic socket (*n*=21)	Contralateral normal eye (*n*=21)	Validation
Distance between levator and superior rectus	0.33, 0.01; 15.0	0.22, 0.12; 12.3	*P*=0.5
Levator palpebral muscle thickness	2.7, 1.95; 3.3	2.5, 2.0; 2.6	*P*=0.09
Levator palpebral muscle length	36.7, 14.6; 41.3	42.1, 38.1; 44.0	*P*=0.01[a]
Superior rectus muscle thickness	2.6, 2.05; 3.1	2.1, 1.8; 2.6	*P*=0.02[a]
Superior rectus length	29.9, 27.3; 37.1	35, 29.8; 38.7	*P*=0.5
Levator function (mm)	14.0, 10.0; 15.0	15, 11; 15	*P*=0.6
Lagophthalmos with prosthesis in place (mm)	3.0, 0.0; 4.0	0	*P*=0.002[a]
MRD1 (mm), mean±SD	4.05±1.2	3.6±2.1	*P*=0.3
Superior fornix depth (mm)	13, 10.5; 14.0	14, 12; 15	*P*=0.192
Inferior fornix depth (mm)	7.76, 7.0; 8.0	8, 7; 9.75	*P*=0.351

IQR: Interquartile range; CI: Confidence interval; SD: Standard deviation; N: Number of participants; MRD1: Distance between the superior lid margin and the pupil. [a]$P<0.05$ is statistically significant.

by sectioning the four rectus muscles and the optic nerve, followed by the placement of a spherical PMMA implant wrapped in a donor sclera attached to the rectus muscles. The implant size was chosen intraoperatively using a sizer, varying from 18 to 22 mm.

Statistical Analysis All data were collected on a pretested data collection form and then transferred to an Excel spreadsheet (Microsoft Corp., Redmond, WA, USA). Statistical analysis was performed with Statistical Package for the Social Sciences (SPSS 25; IBM Corp., Armonk, NY, USA). Demographic data are presented as frequency and percentage proportions. Quantitative data were analyzed using the non-parametric method due to non-normal distribution, and all values are reported as median with interquartile range (IQR). A two-sided P value <0.05 was considered statistically significant.

RESULTS

Twenty-one patients with unilateral acquired anophthalmia comprised the study group and the contralateral normal eye was the control group. The mean age of the participants was 41.3±11.2y and there were 13 (61.9%) males. Fifteen (71.4%) patients had undergone evisceration and 6 (28.6%) had undergone enucleation. The median interval between evisceration or enucleation and the current measurement was 9.0 (IQR 2.5; 15.5)y.

Table 1 presents the CT scan measurements, the clinical topographic lid measurements, and the fornix depth of both groups.

CT in the sagittal plane showed a not measured hypodense space (“sagging space”) between the roof and the LPS/SR complex in the entire anophthalmia group compared to the control group. LPS thickness was similar in both groups. However, the LPS length was statistically significantly shorter in the anophthalmia group compared to the control group (P=0.01). The SR was statistically significantly thicker in the anophthalmia group compared to the control group (*P*=0.02), but both did not differ in length. The space between the LPS and the SR was similar in both groups.

Topographic evaluations of the lids indicated lagophthalmos only in the anophthalmia group. MRD1 was mildly higher in the anophthalmia group but not significantly different than the control group. LPS function was similar in both groups (*P*>0.05).

Superior fornix depth in the anophthalmic socket was not significantly different compared to the normal orbit (*P*=0.192). The inferior fornix depth was similar between groups (*P*=0.351).

DISCUSSION

In the present study, a shorter LPS and thicker SR occurred in conjunction with lagophthalmos in unilateral acquired anophthalmia managed with a spheric implant for replacing orbital volume. These changes can be responsible for the altered upper lid position observed in patients with acquired anophthalmia.

A sagittal plane CT scan is essential for assessing anophthalmic orbits anatomy[5,11]. We used this imaging study to quantitatively measure and evaluate the position of the LPS/SR complex. Despite improved imaging techniques, it is difficult to differentiate LPS and SR due to the strict relation of these structures with surrounding tissue and other structures such as the superior ophthalmic vein, the transverse superior intermuscular fascia sheath between both muscles and the stronger attachments at the medial and lateral portion of the muscles with Whitnall's ligament[21]. In addition, a loose connective tissue connects the transverse ligament with the overlying LPS and the underlying SR, with delicate connective tissue fibers extending from the ligament into the superior fornix, previously described as the "suspensory ligament of the superior fornix" indicating the close interaction of the structures in this quadrant of the orbit[22].

Normally the LPS/SR complex can be seen in the sagittal plane as contiguous with the roof of the orbit. However, "sagging" space has been reported after the removal of the eye without volume replacement with orbital implants. "Sagging" suggests that the globe provides additional support to the orbital structures, preventing the collapse of the upper orbital compartment and the rotatory displacement of the orbital contents in anophthalmic sockets[22-23]. However, in the present study even using a spherical implant selected by a sizer during the surgical procedure to replace orbital volume efficiently, "sagging" was observed. This indicates that despite volume replacement with orbital implants and a more physiologic position of the muscles in the orbit, the LPS/SR complex is still displaced downwards, influencing the position of the upper lid.

In our study, the LPS length was statistically shorter, and the SR was statistically thicker in anophthalmic sockets compared to the normal orbit. These alterations can have functional implications. The sustained LPS/SR complex contraction can be related to reduced mobility of the orbital contents, contributing to the displacement of the eyelids, and reducing the amplitude of movements of the external ocular prosthesis in the anophthalmic socket syndrome[17,23-28].

The sample of this study was composed of grade 0 or 1 anophthalmic sockets and we detected mild lagophthalmos and a non-significant upper lid retraction in the anophthalmic sockets. Perhaps higher grades of socket contraction can be associated with even greater lagophthalmos and upper lid retraction. Although ptosis is part of the definition and is generally included as a major sign of anophthalmic socket syndrome, ptosis occurs only in 11% to 18% of anophthalmic cases[7,10,15,23,29]. Upper lid retraction has already been reported in anophthalmia[5,17] and should be included as a component of anophthalmic socket syndrome.

The origin of lagophthalmos and mild lid retraction can be explained by the contraction of the LPS, levator damage during eye removal, changes in the position of the levator muscle, or inadequate replacement of orbital volume associated with a large external ocular prosthesis[5,10,30]. Upper lid retraction can be associated with a deep upper lid sulcus and lower lid laxity, worsened by progressive laxity due to aging and a possible lack of volume, leaving an appearance of enophthalmos[11].

In our study, the upper fornix depth had no statistical difference comparing the anophthalmic sockets to the normal orbit. The upper fornix has a close anatomical relationship with the SR muscle, and the physiological rectus pulley movement might be expected to shift the soft tissues of the fornix[25]. Perhaps the more contracted sockets that were not evaluated in the present study can have shallow upper fornix and this must be ruled out in future research.

The inferior fornix was similar to the normal control side, but after many years of wearing an external prosthesis, the inferior fornix can become shallow, probably because of repetitive trauma induced by the external prosthesis to the delicate conjunctival tissue, provoking conjunctival adhesions in the fornix[7].

The alterations observed in the current study such as lagophthalmos and mild upper lid retraction can result in the inability to complete closure of the upper lid, resulting in exposure of the external ocular prosthesis, causing a mucin crust on the prosthesis surface, foreign body sensation, irritation of the anterior surface of the socket and the lids, and worsening adaption of an external prosthesis.

This is a preliminary study and as such, has several limitations including the small sample size and the lack of comparison between eviscerated and enucleated sockets. However, we have demonstrated alterations in the LPS/SR complex in the upper quadrant of the orbit, explaining lid position changes in anophthalmic socket. Further studies using high-resolution magnetic resonance images can better clarify the role and interactions of the LPS and SR with other adjacent structures such as Müller's muscle, orbicularis oculi, as well the "sagging" in the roof of the orbit despite the presence of an orbital implant in anophthalmic socket. Another critical point is the influence of the external prosthesis in the final position of the lids and fornix; this is another essential variant to be studied because even with customized prostheses, asymmetry is pursued[6].

In conclusion, there is evidence of the interaction between the shorter LPS and thicker SR with mild lid retraction resulting in lagophthalmos in acquired anophthalmic socket orbits repaired with spherical implants. Post-enucleation or evisceration anatomic changes and the relationship of the LPS and SR with other orbital structures associated with passive or active forces acting in the final position of the lids and external ocular prosthesis should be further investigated.

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