Congenital anophthalmia treated with a dermis fat graft combined with a skin graft in the upper lid in early childhood

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Abstract:

Three infants with unilateral congenital anophthalmia and contracted sockets received early dermis fat and eyelid skin grafts, plus progressive conformer therapy. This approach expanded socket volume, improved periorbital and eyelid dimensions, and enhanced facial esthetics. All patients successfully adapted to ocular prostheses, showing the benefits of timely intervention and reconstruction.

Keywords:

Congenital anophthalmia, dermis fat graft, eyelid skin graft

Introduction

Congenital anophthalmia (CA) is a rare malformation, with an estimated incidence of 0.18–0.4 cases/10,000 births, caused by failure in the development of the primary optic vesicle during the embryonic period (around 6–10 weeks of gestation) or by nonclosure of the fetal fissure.[1] CA may be unilateral or bilateral, isolated or syndromic and can present as either total absence of the eye (true anophthalmia) or with a rudimentary eye (clinical anophthalmia), distinguished by orbital imaging.

Deficient orbital volume leads to abnormal development of the periorbital region and progressive eyelid/facial asymmetry, causing functional, esthetic, and psychological challenges.[2]

Classical treatment involves progressively larger conformers. However, in true CA, palpebral fissure dimensions may be so reduced that even the smallest conformer is difficult to adapt. Expanding orbital volume is essential. Several techniques have been proposed for this, including dermis fat grafts (DFG), spherical implants, hydrophilic/hydrogel expanders, inflatable expanders, mucosal, bone, or muscle grafts.[1,3-5]

DFG is widely used for orbital volume augmentation, stimulating orbital development.[6] Nevertheless, the optimal age for surgery remains debatable, generally recommended for children over the age of three. However, by this time, approximately 80% of adult orbital volume has already developed.[7]

Effectiveness of DFG at an early age remains uncertain, and there is no consensus on the ideal timing for surgery in CA. We report three cases using a novel technique combining DFG with upper lid skin graft and simultaneous horizontal fissure enlargement. This report was approved by our ethics committee and follows the Declaration of Helsinki. Parental consent was obtained.

Case Reports

Three children with CA and severely contracted sockets, which prevented conformer adaptation, presented for treatment. Cases 1 and 2 were < 1-month-old, and Case 3 was 7-month-old. Case 1 had a contralateral microphthalmic eye with light perception, and Case 2 and 3 had normal contralateral eye. All children underwent DFG and lid surgery upon presentation. Surgeries were under general anesthesia. DFG were harvested in oval shape from the upper outer quadrant of the gluteal region. The overlying skin was carefully removed for use as graft. Donor

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site deep layers were sutured with polyamide 4-0; skin with polyamide 5-0 (Johnson and Johnson, SP, Brazil). To enlarge the palpebral fissure, a horizontal incision from the lateral canthi was made, matching the length of the contralateral lid fissure, and the skin was sutured to the underlying tissue. The upper eyelid was incised at a height corresponding to the contralateral normal lid crease, and the harvested skin graft was positioned and sutured with a continuous 6-0 polyamide suture (Johnson and Johnson, SP, Brazil). The DFG was inserted into the socket and sutured to Tenon's capsule with interrupted sutures; the conjunctiva was closed with continuous 6-0 polyamide (Johnson and Johnson, SP, Brazil) suture. A customized conformer was placed, followed by tarsorrhaphy for 1 month, then removed [Figure 1a-f]. The conformer or external prosthesis was then progressively enlarged monthly postoperatively.

Standardized periocular photographs were taken preoperatively and 6 months postoperative using an iPhone 10 (Apple, USA). Images were analyzed using ImageJ software from the National Institute of Health (USA). Comparisons between CA-affected and contralateral normal sides included upper eyelid height (from lid margin to upper eyebrow in internal, medial, and lateral segments), horizontal fissure length, and palpebral fissure area. Pre- and postoperative values for each eyelid segment were compared as percentages: (100 × [CA − normal side]/normal side) [Figure 2].

Figure 1: Demonstration of the surgical technique performed in Case 1. (a) Child before the surgical procedure; (b) Horizontal fissure enlarged to a measurement similar to the contralateral normal side; (c) Skin removed from the surface of the dermis fat graft harvested from the bottom region; (d) Placement of the skin graft on the upper lid; (e) Immediate postoperative appearance after dermis fat and skin graft surgery; (f) Child 3 months postsurgery, showing an increase in orbital volume, improved lid fissure, and height of the upper lid

Preoperative measures showed shorter eyelid heights on the CA side, especially in Case 1's internal segment. Postoperative improvements were noted in all cases. Horizontal fissure length increased postoperatively in all three children, with Case 2 showing the greatest gain. Cases 1 and 2, operated in the 1st month of life, had better outcomes than Case 3 [Figure 2]. The palpebral fissure area increased in all cases, allowing for conformer or prosthesis retention. Children remain under follow-up at our hospital. Case 1 is now 1 year old; Case 2 is 3; Case 3 is 9 years old and received an additional DFG and skin graft to improve esthetics. All retain external prostheses [Figures 1 and 3].

Discussion

We described three children with severe unilateral true CA who underwent DFG combined with upper lid skin graft, enlargement of lid fissure length, associated with progressive conformers at an early age. CA often causes marked periorbital deformity and facial asymmetry. Our technique helped stimulate simultaneously periorbital and orbital growth.

DFG with skin graft in the upper lid had not been previously reported. Skin typically discarded from the DFG surface was reused to improve eyelid dimensions. This is important since true CA presents poorly developed upper eyelids, and grafting substantially improves measurements. We also addressed short palpebral fissures length combining horizontal enlargement with the other procedures. Although classic treatment recommends conformer use within the 1st month of life, conformers alone cannot improve lid measurements as effectively as surgery. Alterations in the levator muscle, including ptosis

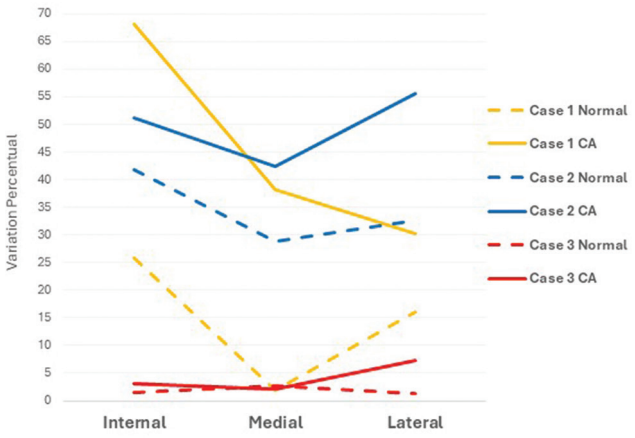

Figure 2: Percentage of variation between the preoperative and 6 months after dermis fat graft combined with skin graft in the upper eyelid. The measurements include the internal, medial, lateral, and horizontal fissures in the children reported – UNESP, 2025. CA: Congenital anophthalmia

Figure 3: Photodocumentation of patients 2(a,b) and 3(c,d), showing the preoperative and the postoperative images of the children – UNESP, 2025

and malformed creases, were observed in Cases 1 and 3, requiring future correction.

Orbital development depends on volume and progresses most rapidly from birth to 12 months.[7,8] Growth occurs in two phases: the first growth phase occurs before the age of 3 years, and the second phase between 7 and 12 years of age, with greater cranial changes between 12 and 24 or 48 months of life.[7,8] The growth speed in the first phase is approximately three times greater than in the second phase.[9] Orbital volume increases by 1%–2% annually through adolescence.[8]

Our cases had early DFG to address volume deficits. Although age guidelines for surgery are lacking, known orbital development patterns support early intervention for better outcomes in CA, promoting growth and enabling external prosthesis adaptation. Based on our data, we recommend early volume correction.

Disadvantages of DFG include requiring two surgical sites with scarring, unpredictable graft volume, potential for necrosis, and excessive growth requiring future debulking.[10] Yet in children, graft growth may be advantageous, contributing to orbital expansion. Even if enlargement occurs, revision of the external prosthesis can be enough.

Despite this study involved only three children and limited follow-up, its strengths lie in the quantitative comparison of periocular development, emphasizing the urgency of early surgical intervention combining DFG, skin grafts, lid fissure enlargement, and progressive conformers for optimal results in the CA treatment.

In conclusion, DFG in the socket, combined with upper eyelid skin graft, lid fissure enlargement, and progressive conformers, performed early in life, is recommended for true CA effective treatment. This technique improves orbital volume, eyelid and palpebral fissure dimensions, facilitates external prosthesis adaptation, enhancing cosmetic outcomes in CA-affected children.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients have given their consent for their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published, and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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