Pediatric Enucleation

Analysis of Volume Replacement

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Objectives: To determine the percent volume replacement by an implant and prosthesis, the long-term sequelae of poor volume replacement, and specific guidelines for volume replacement in pediatric patients.

Methods: A retrospective observational study of 16 pediatric patients who underwent enucleation (group 1) and 31 adult patients enucleated in childhood (group 2) was performed. The total volume replacement and the volume replacement by the implant were determined. The incidence of secondary surgical procedures for superior sulcus deformity, enophthalmos, ptosis, ectropion, and socket contraction was determined.

Results: The average percent volume replaced by the implants in group 1 patients was 68%. The average percent volume replaced by the implants in group 2 patients was 21%. Seventy-one percent of patients in group 2 underwent secondary implant surgery to augment volume and eliminate superior sulcus deformity and enophthalmos.

Conclusions: The placement of an adequately sized implant in pediatric patients may obviate the need for secondary augmentation of socket volume in adulthood. The authors suggest an implant 2 mm less in diameter than the axial length of the eye in pediatric patients.

by the age of 2 years, and continues to grow 1% per year until its final size is reached. An issue that arises in pediatric patients is the potential need for implant exchange with secondary insertion of larger implants into the muscle cone as growth occurs. This becomes problematic if one uses porous implants in the muscle cone because the dissection required for implant removal could damage the extraocular muscles and their innervation, or create restrictive fibrous tissue planes that inhibit motility. We hypothesize that larger implants may be used at the time of primary enucleation, thus making larger secondary implants unnecessary in many instances.

Fundamental questions are (1) Is the volume removed by enucleation in a pediatric patient being adequately replaced by the implant and prosthesis? To answer this question, one must compare the volume removed with the total volume used to replace the globe, including the implant and the prosthesis. (2) What are the sequelae of implants that do not replace enough volume in the posterior socket in pediatric patients? (3) What

PATIENTS, MATERIALS, AND METHODS

PATIENTS

Forty-seven consecutive anophthalmic patients who had undergone enucleation in childhood were studied retrospectively, including pediatric patients (group 1, n = 16; age range, 3.5-16 years), and adult patients who underwent enucleation in childhood (group 2, n = 31; age range, 17-71 years). None of the patients had a diagnosis of congenital microphthalmos. Only 1 patient (patient 26, group 2) had a history of radiation treatment to the eye socket. Table 1 presents individual patient data.

Group 1 patients underwent enucleation between 1988 and 1998 by 6 different surgeons using similar techniques. After informed consent, the patients underwent enucleation under general anesthesia. Conjunctiva was preserved in each case. Fifteen of 16 patients had a hydroxyapatite sphere implant and 1 patient had a silicone implant. Sizers were used to estimate appropriate implant size for 4 patients. One patient's operative report showed documentation of the axial length of the globe. No specific means of determining implant size was noted in the remaining cases. Extraocular muscles were attached to wrapping around the spherical implant, except in 1 case in which the muscles were "imbricated." Tenon's capsule and conjunctiva were closed in separate layers.

Patients in group 2 underwent enucleation in childhood and most of these patients (74%) came for evaluation of eyelid or socket problems during 7 years. Other patients (26%) returned for continuing follow-up after enucleation. Clinical problems following enucleation were documented, including implant exposure and infection, ptosis, enophthalmos, ptosis, superior sulcus deformity, ec- tropion, implant migration, and socket contraction. Operative reports were reviewed to obtain the diameter of the implant used (Table 1). In 6 of 8 patients (group 2) who underwent enucleation between 1978 and 1982, operative reports were available but none documented method(s) of implant size determination.

Pathology reports containing the axial length of the enucleation specimen were obtained in 12 of 16 patients in group 1, and 8 of 31 patients in group 2. A-scan ultrasonography of the remaining eye was available in 5 of 16 patients in group 1, and in 21 of 31 patients in group 2.

Prosthetic volume was determined by the volume displacement of an alginate cast of the prosthesis in 7 patients in group 1 and 15 patients in group 2 (>10 months after enucleation). A clay cast of the patient's prosthesis was made. Alginate was injected into the cast and allowed to set. The alginate cast was removed from the clay mold, and the volume was determined by volume displacement to the nearest 0.05 mL. Plain x-ray skull films (anterior to posterior views) were available in 5 patients (group 2) for comparison of the anophthalmic and normal orbital entry at the level of the orbital rim. Horizontal and vertical measurements of both sockets were obtained.

Surgical procedures performed on the eyelids or sockets on the anophthalmic side after the date of the primary enucleation were documented. The average length of follow-up for group 1 was 3 years, and for group 2 was 29 years.

VOLUME CALCULATIONS

All of the volume calculations were based on the primary implant and the corresponding optimal prosthesis 10 months or more following enucleation. The volume removed was calculated using the axial length of the enucleated specimen (to the nearest 0.5 mm), and/or estimated by an A-scan measurement of the contralateral eye. The formula $4/3\pi r^3$ was used to determine the volume of the eye, with $r = \text{axial length (from the pathology report)}$ or $r = \frac{1}{2} \text{(A-scan measurement + 1 mm)}$. Although the eye is not a perfect sphere, the axial length corresponds to the A-scan, a clinically accessible and useful parameter for estimating pre-morbid ocular volume.

The volume replaced by the implant was calculated from the radius of the implant using the formula $4/3\pi r^3$, or by volume displacement if the implant was not spherical (to the nearest 0.05 mL). The volume of the prosthesis plus the volume of the implant was the total volume replaced. The percent volume replaced by the implant was the volume of the implant divided by the volume removed times 100. The percent total volume replacement was the volume of the implant plus the volume of the prosthesis divided by the volume removed times 100.

Calculations of volume replacement at the time of enucleation (volume removed based on axial length of the enucleation specimen) and at a distant follow-up examination (volume removed based on the A-scan of the remaining eye) were available in 6 patients. These measurements allowed observation of the change in percent volume replacement by the implant as growth occurred.

In group 2, the mean percent volume replacement in subgroups with and without superior sulcus deformity, enophthalmos, ptosis, ectropion/laxity, and socket contraction, were compared for statistical significance using the 2-tailed $t$ test. Confidence intervals were stated in terms of $P$ values.
implant sizes (percent volume replacements) are associated with the need for secondary procedures to augment soft tissue volume? (4) What critical volume replacement should be achieved to prevent asymmetry of bony orbital development? (5) Should replacement of a larger percentage of the volume be considered in pediatric patients to compensate for growth of the eye and orbit to adult size? (6) Can we anticipate a certain percent change in the relative percent volume that the implant provides, and then replace a larger percentage of volume to compensate for this growth? (7) What are specific guidelines for determining appropriate implant size in pediatric patients?

**RESULTS**

A trend toward greater volume replacement was noted in the more recent enucleations (group 1) (Figure). Two patients in group 1 required secondary implants because of implant exposure and infection, not for volume supplementation. The exposure in these 2 patients correlated clinically with anteriorly malpositioned implants.

In group 2, 77% of patients had superior sulcus deformity and 71% of patients had secondary implant surgery to increase volume replacement because of insufficient volume replacement by the primary intraconal implant. Two patients with 13% and 30% volume replacement by the implant had superior sulcus deformity but chose not to have surgery to augment the volume replacement.

Four patients in group 1 (25%) had ptosis, and only 1 of them underwent ptosis repair. In group 2, 20 patients (64%) had ptosis, and 13 of those patients (63%) had ptosis repair. The mean percent volume replaced by the implant in group 2 patients with ptosis was 30%, whereas the mean percent volume replaced by the implant in group 2 patients without ptosis was 84%. The comparison of the means (group 2) using the 2-tailed t test was statistically significant ($P<.01$).

Two patients in group 1 had mild socket contraction but did not require mucous membrane grafting for reconstruction of the fornixes. Eight of 9 patients in group 2 who had socket contraction required mucous membrane grafting. Comparison of the mean percent volume supplied by the implant in group 2 patients withocket contraction (21%) to the mean percent volume supplied by the implant in group 2 patients without socket contraction (45%) was statistically significant ($P<.05$).

Eight patients in group 2 underwent horizontal shortening of the lower eyelid. Comparison of the mean volume replaced by the implant in group 2 patients (28%)
to the mean volume replaced by the implant in group 2 patients without ectropion or lower eyelid laxity (42%) was statistically significant (P < .01).

X-ray films were available for 5 patients from group 2. Two patients (aged 4 and 5 years) had no implant, 1 patient (aged 5 years) had a 15-mm implant, 1 (aged 7 years) had a 1.8-mL nonspherical implant, and 1 (aged 5 years) had an implant of unknown size. The anophthalmic orbits were smaller in 4 of 5 patients. Enlargement of the ethmoid air cells on the side ipsilateral to enucleation was evident on 1 x-ray film. The area of the orbital entry, determined by multiplying the horizontal and vertical dimensions measured on the anterior to posterior skull x-ray film, was 68% to 100% of the value on the normal side.

**COMMENT**

Adult patients undergoing enucleation in childhood provide an opportunity to assess the effects of volume replacement in the intraconal implant. This study demonstrates that group 2 patients, undergoing enucleation from 1932 to 1985, with 21% volume replacement by the implant (range, 0%-44%), had grossly inadequate posterior volume replacement. Seventy-seven percent of patients had superior sulcus deformity, and 92% of those patients had secondary orbital implant surgery to supplement volume replacement in the posterior compartment. Poor volume replacement by the implant had a statistically significant association with superior sulcus deformity, enophthalmos, ectropion/lower eyelid laxity, and ptosis. Clearly, the insertion of an implant that replaces only 21% of the volume removed is insufficient, and predestines the patient to the pain and morbidity of additional orbital surgery to correct posterior volume deficiency. This observation underscores the need for meticulous planning of volume replacement, including a preoperative A-scan of the opposite eye, intraoperative measurement of the axial length of the enucleated specimen, and appropriate selection of implant size.

The mean follow-up time for patients (group 2) with superior sulcus deformity was 37 years, and for patients without superior sulcus deformity, 19 years. A trend of placing larger implants is noted. Progressive, undefined physiologic and anatomic changes may also occur. Fat atrophy has been generally accepted as a cause of superior sulcus deformity but is not substantiated by scientific evidence. Several studies have shed doubt on this mechanism. Kronish et al found no evidence of fat atrophy following enucleation in a primate animal model. Smit et al studied computed tomographic scans of anophthalmic sockets and documented a rotation of the orbital contents posteriorly and inferiorly, accounting for deepening of the superior sulcus. More complete volume replacement in the anophthalmic socket has been shown to improve superior sulcus deformity. A prospective study is underway to determine if placing an implant 2 mm less in diameter than the axial length will eliminate superior sulcus deformity in uncomplicated enucleation.

Based on the following observations, implants larger than those traditionally placed in pediatric eye sockets may be used in pediatric patients: (1) The eye is more than 85% of its adult size by age 2 years. (2) The axial lengths and A-scans of infants and children suggest placement of implants 18.5 to 22 mm in diameter. (3) Clinical experience indicates that pediatric patients do well with implants 16 to 20 mm in diameter. (4) In this study, 22-mm implants were used in 2 patients, both aged 7 years, without complications. Two instances of implant exposure occurred in group 1 patients (aged 21 months and 24 months) who had 16- and 18-mm implants, respectively. The exposure correlated with anterior positioning of the implant in both cases. Clinical evidence supports that exposure is related more consistently with technique and implant positioning than with implant size.

Patients requiring correction of ptosis, socket contraction, and lower eyelid laxity had statistically less volume replacement at the time of enucleation than patients not exhibiting these clinical findings. Some secondary eyelid procedures may be attributed to augmentation of the prosthesis to compensate for poor volume in the implant, chronic manipulation of the eyelids to insert and remove a large prosthesis, duration of prosthetic wear, and other factors.

Multiple complex mechanisms contribute to ptosis in the anophthalmic socket, including levator dehiscence, scarring, and small implant with collapse of the superior orbital structures. Results of this study concur that the latter mechanism may be a contributing factor, as well as the long-term use of a bulky prosthesis. In group 1, 25% of patients had ptosis. One patient had ptosis related to mechanical tethering of the levator in the horizontal closure site, which was confirmed intraoperatively. In group 2, 64% of patients had ptosis and the comparison of the means of implant volumes in patients with and without ptosis was statistically significant. Mean follow-up of patients with (34 years) and without (31 years) ptosis was comparable. Additional studies of anophthalmic patients with ptosis, first-hand intraoperative observations of the levator, and perhaps sagittal magnetic resonance imaging studies will further clarify the multiple mechanisms contributing to ptosis in anophthalmic patients.

Although socket contraction is an anterior socket phenomenon, lack of volume in the posterior compartment, forcing maximal replacement in the anterior compartment, could conceivably contribute to socket contraction. Bulky prostheses accumulate dried mucus on the anterior surface, which causes inflammation and discomfort. Chronic inflammation could instigate fibrous contraction in the subconjunctival and Tenon’s space. The comparison of the means of volume replacement by the implant in group 2 patients with and without socket contraction was statistically significant. Difference in mean follow-up time of patients (group 2) with and without (31 years) ptosis was comparable. Additional studies of anophthalmic patients with ptosis, first-hand intraoperative observations of the levator, and perhaps sagittal magnetic resonance imaging studies will further clarify the multiple mechanisms contributing to ptosis in anophthalmic patients.

Anophthalmic ectropion, or eyelid laxity, may be a consequence of aging tissues, chronic prosthetic wear and manipulation of the eyelids on insertion and removal of the prosthesis, excessively bulky prostheses, cicatrical...
anterior lamellar changes in the lower eyelid, and other factors. Inadequate volume of the implant with a compensatory large ocular prosthesis has an effect on the development of lower eyelid laxity in these patients. The comparison of the mean percent implant volume between patients with and without ectropion was statistically significant in group 2 patients. Mean follow-up was similar in the 2 groups (35 years in those with laxity and 30 years in those without laxity).

Poor bony development was observed in 3 patients with poor volume replacement. A relationship has been established between the lack of an implant, or a small implant, and abnormal orbital bone development in animals and humans.13 A recent study suggests that the bony orbital entry is minimally affected in pediatric patients who have had 15- to 19-mm implants.6 Follow-up of group 1 patients was short, not extending past puberty. We suspect that superior sulcus deformity and enophthalmos may not develop as readily in pediatric patients because poor bone development, a consequence of poor volume replacement by the implant, means less bony volume to be filled by the soft tissues. The soft tissue deficit may not be evident until the implant migrates forward or out of the muscle cone. Larger radiographic studies of adults who received implants of various sizes as children will establish meaningful parameters of the critical volume replacement and implant diameter that will support normal, symmetric bony development.

The status of orbital bony development becomes a key clinical issue when evaluating patients for secondary intraconal or subperiosteal implants for volume augmentation. A screening anterior-posterior or Water skull x-ray film is recommended prior to secondary orbital implants in patients who undergo enucleation in childhood. The difference in vertical dimension should be subtracted from the implant diameter. Computed tomographic scans or magnetic resonance imaging are helpful in some cases to visualize the extraocular muscles, the existing implant, and their interrelationships.

Six patients having volume determinations in childhood and adulthood demonstrated a 6% relative negative change in the percent volume replacement during an average of 14 years. One might consider replacing 6% of the volume of the implant for pediatric patients. That is, if 70% to 80% volume replacement in the implant is desirable in adulthood, then 76% to 86% of the volume of the globe should be replaced by the implant in pediatric patients. For example, the eye of a 2-year-old is 85% the size of an adult’s eye (average, 7.9 mL), and the volume is 6.7 mL. Replacement of 76% to 86% of 6.7 mL, 5.1 to 5.8 mL, would require a 21- to 22-mm sphere. This leaves 6.7 mL minus 5.1 to 5.8 mL for the prosthesis in childhood (0.9-1.6 mL), and 7.9 mL minus 5.1 to 5.8 mL for the prosthesis in adulthood (2.1-2.8 mL). The pediatric prosthesis might resemble a scleral shell initially but would be gradually augmented into adulthood. A recent study demonstrated a range of prosthetic volumes in adults of 0.75 mL to 4.2 mL.16 A volume of 1.8 mL produces a thickness conducive to integration and avoids complications of larger prostheses (3.0-4.0 mL).16

In this study, as in a previous study, none of the pediatric patients had an A-scan that was less than 20.0 mm.3 Two patients in group 1 had too much volume replacement by the implant (100% and 114%), leaving very little flexibility for volume replacement by the prosthesis. Preoperative planning or an intraoperative observation of the axial length of the specimen would have avoided over-replacement of implant volume in these cases.

Infants (2 aged 5 months and 1 aged 8 months) undergoing enucleation in this study had ocular volumes of 4.3 mL, 5.6 mL, and 7.2 mL, respectively. Implant sizes of 18 mm, 20 mm, and 22 mm, respectively, could be accommodated in these infants, leaving adequate space for the prosthesis in each case (Table 2). Measurements of the enucleated specimen would have provided valuable information in these cases. Unless phthisis or microphthalmos is present, the diameter of the enucleated specimen will be greater than the diameter of the selected implant. If one is not treating a microphthalmic patient with poor bone development, no logical argument exists that an implant, sized correctly (axial length minus 2 mm), would not easily fit into an infant socket.

The authors suggest placing an implant 2 mm less in diameter than the axial length of the enucleated specimen or 1 mm less than the A-scan measurement of the other eye. Estimation of implant diameter based on the A-scan of the opposite eye allows a preoperative decision, requires less intraoperative calculation than other methods, establishes a guideline for patients with phthisis and buphthalmos, and is useful for patients undergoing evisceration. Table 2 presents the distribution of the volume replacement in the implant and the prosthesis if this algorithm is followed. This approach, combined with deliberate placement of the implant posteriorly, would account for additional axial growth and optimal augmentation of the prosthesis.

As the eyes and bony sockets of group 1 patients grow, comparison of their radiographic results with those of the patients in group 2 will determine if the increase from 21% to greater than 68% of volume supplied by the implant dramatically improves their outcomes as adults and eliminates the morbidity of additional orbital surgery.

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<th>Prosthetic Volume</th>
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Axial length – 2 mm = implant diameter.
REFERENCES


Ophthalmological Numismatics

A look at the past . . .

Julius Hirschberg (1843-1925), was an ophthalmologist who practiced in Berlin, Germany, and was an assistant to von Graefe. He became professor at the University of Berlin and introduced the use of the electromagnet for metallic foreign body removal in eye surgery. Today he is best known for his 11-volume History of Ophthalmology (1899-1918), which was later translated into English by Frederick C. Blodi.

This bronze plaquette (Figure 1) was struck in honor of Hirschberg’s 70th birthday by his friends, colleagues, and students in 1913. The reverse (Figure 2) depicts “Science” removing the bandages from the eyes of an old man who is gazing out in amazement. On the left are some of Hirschberg’s publications, and in the background, buildings of the University of Berlin. The medallion plaquette was engraved by Ernest Herter and struck by B. H. Mayer.

Courtesy of: Jay M. Galst, MD, 30 E 60th St, New York, NY 10022.