Orbital Growth After Unilateral Enucleation in Infancy Without an Orbital Implant

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Purpose: To measure the volume of the anophthalmic orbit in adults who had undergone enucleation during infancy and to determine its growth.

Methods: The orbital volume in 5 adults who had undergone unilateral enucleation during infancy without an orbital implant was measured on x-ray computed tomography images. Comparisons were made between the anophthalmic and normal sides. In addition, we evaluated the morphology of the orbits showing growth retardation and the association between the prosthesis, if present, and orbital growth.

Results: In adults who underwent unilateral enucleation as infants, without an orbital implant, orbital growth was more retarded on the anophthalmic side than on the normal side. The difference was most marked in the area corresponding to the equator of the eyeball. This growth retardation was more severe in patients whose prosthesis was not replaced during childhood than in those who had regular replacement of their prosthesis.

Conclusion: For orbital growth in anophthalmic orbits, an intraorbital volume that replaces the eyeball is necessary. An orbital implant at the time of enucleation and the replacement of the prosthesis with growth are important.

Key Words: Anophthalmia, enucleation, orbital implant, orbital volume, prosthesis.

Introduction

The ocular orbital volume increases rapidly until 3 years after birth and expands gradually thereafter, reaching nearly adult orbital volume at the age of 12 years. It has long been recognized that enucleation during the orbital growth period causes orbital growth retardation in the anophthalmic orbit. This growth retardation has been evaluated in animal models by direct measurement of the orbital volume of skulls. It has been suggested that the absence of an eyeball and a defect in the orbital tissue can cause orbital growth retardation, and that greater orbital volume can be provided by gradual enlargement with tissue expanders. In clinical cases, the growth of anophthalmic orbits has been evaluated by measuring the transverse and longitudinal axes of the orbit on x-ray films or by morphometry. However, there have not been any clinical studies that evaluated orbital growth by measuring the orbital volume.

With the recent improvements in x-ray computed tomography (CT), measurement of orbital volume can be made more easily and accurately. We measured the orbital volume on CT images in 5 adults who underwent enucleation without an orbital implant during infancy, and evaluated growth of their anophthalmic orbits.

Materials and Methods

The subjects were 5 adults who underwent enucleation for retinoblastoma or injury during infancy without orbital implants and who had some prob-
lems in their sockets (Table 1). None of the subjects had undergone radiotherapy after enucleation. The prostheses worn by patients 1, 2, and 3 were occasionally replaced by larger devices with growth (replacement group), but in patients 4 and 5 prosthetics were replaced infrequently (nonreplacement group).

The orbital volume of these 5 subjects was measured on serial coronal images reconstructed from x-ray CT data. Imaging was performed using a Toshiba X-Vigor CT whole body scanner that uses helical scanning. CT image data were reconstructed to serial coronal images with a slice thickness of 2 mm. The bony orbit was traced using the NIH Image (National Institutes of Health, Bethesda, MD, USA) measurement software on a personal computer. Measurements were made from the CT slices preceding the slices containing the posterior lacrimal crest, in the anterior portion, to the optic canal opening in the posterior portion. Areas without bony borders were traced using straight lines. Cross-sectional areas were summed in each slice to calculate the orbital volume.

The volume of the anophthalmic orbit was compared with that on the normal side in the same subjects, and the volume on the normal side in test subjects was compared with normal controls. The cross-sectional area in each slice of the anophthalmic orbit was compared with the corresponding area on the normal side to determine whether the degree of growth retardation differed at different locations in the orbit.

**Results**

**Results of Measurements**

The orbital volumes in the 5 subjects are shown in Table 2. The differences in the orbital volumes between the anophthalmic and normal sides ranged from 1.3 cm$^3$ (anophthalmic/normal side ratio = 94%) in patient 3 to 7.1 cm$^3$ (63%) in patient 5. The volume difference was relatively small (< 10%) in the replacement group (patients 1, 2, 3) but was more marked (30% to 40%) in the nonreplacement group (patients 4 and 5, Figure 1).

The cross-sectional area in each coronal slice was compared between the anophthalmic and normal sides in both the replacement and nonreplacement groups (Figure 2). In both groups, the differences representing growth retardation were most apparent in the anterior portion of the orbit, particularly near the equator of the eyeball.

**Case Reports**

**Two Typical Patients Are Presented**

**Patient 2 (replacement group).** A 31-year-old woman underwent enucleation of the right eye for retinoblastoma at the age of 2 years. As she matured, her prostheses were replaced with larger ones and were well maintained. At age 31, she visited our hospital to have slippage of her prosthesis corrected. The orbital volume was 21.9 cm$^3$ on the right side and 24.4 cm$^3$ on the left side without any substantial difference on CT images (Figure 3). Because the lower fornix was observed to be loose, an orbital implant was inserted and the fornix was reconstructed. Her postoperative course was good.

**Patient 4 (nonreplacement group).** A 21-year-old woman had undergone enucleation of the right eye for retinoblastoma at the age of 9 months. The prosthesis was seldom replaced, and she stopped seeing her ophthalmologist. Recently, she visited our hospital because manipulation of the prosthesis became difficult. Her socket was very small with marked enophthalmos. Her orbital volume was 17.6 cm$^3$ on the right side and 24.3 cm$^3$ on the left side, a marked difference, and definite growth retardation on the right side was observed on CT images (Figure 4). An orbital implant was inserted and her socket was reconstructed by skin grafting, but the enophthalmos remained.

**Discussion**

The presence of an eyeball is important for normal orbital growth, and orbital growth retardation is observed in patients with congenital anophthalmos.

Table 1. Subjects

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Sex</th>
<th>Age</th>
<th>Age at Enucleation</th>
<th>Reason for Enucleation</th>
<th>Replacement of Prosthesis</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>M</td>
<td>23</td>
<td>2 mo</td>
<td>Retinoblastoma</td>
<td>+</td>
</tr>
<tr>
<td>2</td>
<td>F</td>
<td>31</td>
<td>2 y</td>
<td>Retinoblastoma</td>
<td>+</td>
</tr>
<tr>
<td>3</td>
<td>F</td>
<td>57</td>
<td>4 y</td>
<td>Injury</td>
<td>+</td>
</tr>
<tr>
<td>4</td>
<td>F</td>
<td>21</td>
<td>9 mo</td>
<td>Retinoblastoma</td>
<td>-</td>
</tr>
<tr>
<td>5</td>
<td>F</td>
<td>47</td>
<td>2 y</td>
<td>Retinitis ?</td>
<td>-</td>
</tr>
</tbody>
</table>

† mo: month, y: year.
or those undergoing enucleation during early childhood. Immediately after birth, the orbits of patients with congenital anophthalmos and those with acquired anophthalmos cannot be compared on the same level because they differ to such a high degree. However, in both congenital and acquired anophthalmos, orbital growth can be promoted by the use of conformers or the insertion of orbital implants. We previously confirmed that the orbit grows rapidly during childhood, reaching the adult level between the ages of 12 to 14 years. In particular, orbital growth is most rapid up to 3 years of age, and an adequate intraorbital volume during this period appears to be associated with subsequent growth. Therefore, it is probable that orbital growth is impaired after enucleation without an orbital implant.

The 5 patients in this study underwent enucleation for retinoblastoma, injury, or retinitis, but had no facial abnormalities or other orbital diseases, and they did not undergo postoperative radiotherapy. Comparison of the orbital volume of the normal eye in these 5 patients with normal adult controls showed a slightly smaller volume in case 1, a male patient, than in the controls (22.5 mL), partly because he was slightly shorter in height than normal male adults. A larger volume in the 4 female patients compared to normal controls (19.0 mL), suggested that there were no growth problems in the orbit on the patients’ normal side. Growth retardation due to the absence of the eyeball caused the volume of the anophthalmic orbit to be less than the orbit on the normal side in the 5 patients.

Table 2. Orbital Volume

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Anophthalmic Side (A)</th>
<th>Normal Side (N)</th>
<th>Between Sides A – N</th>
<th>A/N %</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>18.2</td>
<td>20.3</td>
<td>−2.1</td>
<td>90</td>
</tr>
<tr>
<td>2</td>
<td>21.9</td>
<td>24.4</td>
<td>−2.5</td>
<td>90</td>
</tr>
<tr>
<td>3</td>
<td>18.7</td>
<td>20.0</td>
<td>−1.3</td>
<td>94</td>
</tr>
<tr>
<td>4</td>
<td>17.6</td>
<td>24.3</td>
<td>−6.7</td>
<td>72</td>
</tr>
<tr>
<td>5</td>
<td>12.2</td>
<td>19.3</td>
<td>−7.1</td>
<td>63</td>
</tr>
</tbody>
</table>

Figure 1. Orbital volume in nonreplacement group was smaller than in replacement group. In both groups, orbital volume on anophthalmic side was less than on normal side. ○: nonreplacement group (n = 2), △: replacement group (n = 3); □: normal orbit (n = 5).

Figure 2. Comparison of cross-sectional areas in coronal slices. Rear computed tomography image represents depth of orbit. Sectional areas on anophthalmic side were markedly small near equator of eyeball, especially in nonreplacement group. ○: nonreplacement group; △: replacement group; □: normal orbit.
Cepela et al.8 inserted tissue expanders that allowed gradual, continuous expansion of the anophthalmic orbits of cats that underwent enucleation soon after birth, and adequate orbital growth was observed. In addition, they compared the orbital volume in animals with a normal-size orbital implant and animals without an orbital implant. They observed that orbital growth without an implant was less than 50% on the normal side, and that the orbital growth with an implant was 50% of that on the normal side, suggesting that the use of adjustable tissue expanders or regular replacement of orbital implants with larger ones is necessary for adequate orbital growth.

Our 5 patients also showed smaller orbital volumes on the anophthalmic side than on the normal side. However, the differences in the orbital volume between the sides were much larger in patients 4 and 5 (28% and 37%, respectively) than in patients 1, 2, and 3 (about 10%). The state of the socket at the time of consultation also differed. Patients 1, 2, and 3 had sufficiently wide cul-de-sac but complained that their prostheses slid down due to a loosening or contracture of the inferior fornix; while patients 4 and 5 had severe contracture of the entire socket and difficulty in applying their prostheses. We speculated that the differences in orbital volume and the state of the socket were associated with regular replacement of the prosthesis with growth in patients 1, 2, and 3, and infrequent replacement in patients 4 and 5.

Comparisons of the areas in each coronal CT image slice between the anophthalmic and normal sides revealed marked differences on the site where the eyeball was originally present, particularly near its equator. The differences were most pronounced in the patients without regular replacement of their prostheses. These results suggested that an intraorbital volume, in place of the eyeball, is necessary for orbital growth after enucleation. Orbital implant insertion is the optimal means to provide a proper intraorbital volume. However, even in patients who did not undergo orbital implant insertion, if the pros-
thesis was repeatedly replaced by larger ones with growth, it served the role of the eyeball by providing intraorbital volume, and marked growth retardation could be avoided.

Fountain et al.\(^\text{11}\) performed morphometric measurements of the orbit in 9 patients who were followed for 5.5 years or more after enucleation and evaluated the effects of the size of the orbital implant on orbital growth. When an implant of adequate size was inserted at the time of enucleation, orbital growth was maintained even if the volume of that implant was less than 50% of the eyeball as an adult. Based on these results, they suggested that changing the size of the orbital implant or the use of tissue expanders was unnecessary. Although they did not describe the prosthetics in their study, the prosthesis is usually provided soon after enucleation in most clinical cases, and the role of the prosthesis in orbital growth cannot be ignored.

One of the methods of evaluating the orbital volume is direct measurement of the orbital volume using animal skull models. In clinical cases, measurements are performed on x-ray film or x-ray CT images, along with direct morphometric measurements. The accuracy and reproducibility of these methods has become a topic of discussion, but the measurement method using serial coronal x-ray reconstruction images performed in this study may be the most satisfactory one at present.\(^\text{12}\)

In the future, we intend to measure the orbital volume in children who undergo enucleation and receive an orbital implant, in order to evaluate the growth in anophthalmic orbits.

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### References