Quality of Life in Intermittent Exotropia

Child and Parent Concerns

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Objective: To identify specific health-related quality-of-life (HRQOL) concerns for children with intermittent exotropia (IXT) and their parents.

Methods: Twenty-four children aged 5 to 17 years with IXT and 1 parent for each child were recruited. Individual interviews with the child and then the parent were audiotaped and transcribed. Transcripts were reviewed, phrases regarding effects of IXT on HRQOL recorded, and specific topic areas identified. Topic frequency was analyzed to determine children’s perceptions of their own HRQOL, parents’ perceptions of their child’s HRQOL, and parents’ own HRQOL.

Results: Child interviews generated 18 topics. Worry (10 of 24 patients [42%]) was most frequently mentioned. Parent interviews generated 22 topics regarding their children’s HRQOL. The most frequently mentioned topic was comments from others (15 of 24 patients [63%]). Regarding the parents’ own HRQOL, 14 topics were identified; the most frequently mentioned was worry regarding possible surgery (15 of 24 patients [63%]).

Conclusions: Multiple individual interviews revealed specific HRQOL concerns, such as worry, in children with IXT and their parents. We will use the concerns identified to develop condition-specific HRQOL instruments for IXT.

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INTERVENTION IN CHILDHOOD intermittent exotropia (IXT) aims to preserve binocular function and to address any social stigma associated with manifest strabismus. Nevertheless, the natural history of IXT and indications for intervention are poorly defined, partly because of a lack of robust outcome measures. Formal assessment of health-related quality of life (HRQOL) may therefore be particularly helpful in clarifying indications for intervention and potential benefits of treatment. Others have compared HRQOL between patients with IXT and control subjects using generic questionnaires, but not with condition-specific instruments. A condition-specific instrument is designed to better capture HRQOL effects specific to a given condition and therefore is more likely to be sensitive to changes over time and with treatment.

A condition-specific HRQOL instrument may be warranted for IXT because it is a common condition and differs from other forms of childhood strabismus in its intermittency and often normal binocular function. As a first step toward developing condition-specific HRQOL questionnaires, we interviewed children with IXT and their parents to identify specific HRQOL concerns. We also aimed to compare HRQOL concerns perceived by the parent and child because such concerns may influence management of this condition.

METHODS

Institutional review board approval was obtained and the parent of each child gave informed consent before participating. Children aged 8 years or older provided informed assent. All procedures and data collection were conducted in a manner compliant with the Health Insurance Portability and Accountability Act.

PATIENTS

Twenty-four children aged 5 to 17 years (median age, 10.5 years) with IXT who had not undergone previous surgery were recruited. Subjects were identified in outpatient clinics and by searching clinical databases. Seventeen subjects were girls and, for 21 subjects, race was self-reported as white. All children had diver-
Present. For the parent, a series of 12 open-ended questions given the choice to be interviewed with or without the parent views ranged from approximately 2 to 5 minutes. The child was asked each question. The duration of child inter-
vence excess or basic-type exotropia; no child had a near angle of 10 or more prism diopters (PD) greater than distance (convergence insufficiency-type exotropia). Patients with coexisting ocular pathology were excluded. The median angle of deviation by alternating prism cover test at distance was 20 PD (range, 10-40 PD). Control and stereoaucity are not reported because we have found both to vary throughout the day in children with IXT9,16 and we did not make multiple measurements in this particular cohort. For each child, 1 accompanying parent or legal guardian was also recruited. Patients and parents were required to be comfortably conversant in English to be included.

PROCEDURES

Individual interviews, first with the child and then with the parent, were conducted and audiotaped. All interviews were conducted by a research orthoptist (S.R.H.) and were performed separately from the clinical examination, some before the examination and some after. For the child, a series of 10 open-ended questions (Table 1) were used as the basis for the interview and discussion was encouraged for each question. Each child was asked each question. The duration of child interviews ranged from approximately 2 to 5 minutes. The child was given the choice to be interviewed with or without the parent present. For the parent, a series of 12 open-ended questions (Table 2) were used as the basis for the interview and discussion was encouraged for each question. Each parent was asked each question. Both parent and child questions were selected after informal discussion within our research group, but were chosen to be general and open-ended to capture as many concerns as possible. The duration of the parent interviews ranged from approximately 3 to 15 minutes. The parent was given the choice of being interviewed alone (if possible) or with the child present.

Child and parent interviews were transcribed and the transcripts were reviewed independently by 1 clinician (W.E.A.) and 2 nonclinicians (D.A.L. and P.A.K.). Each investigator independently recorded any phrases pertaining to the effects of IXT on quality of life, resulting in 3 lists of phrases for each child and 3 lists of phrases for each parent. For each child and parent the 3 lists of phrases were combined, duplicate phrases were removed, and responses were reviewed to identify themes or topic areas. Any phrases describing pure symptoms but not any effect of symptoms on HRQOL were removed. If the phrase indicated that the child or parent was troubled by the symp-
tom, then the phrase was retained. For example, the phrase “my eye wanders” was removed, whereas “I’m bothered by my eyes going in and out” was retained.

Once data collection was completed, 2 investigators (D.A.L. and S.R.H.) reviewed the final list of phrases and topics for all children and parents to agree on the assignment of each phrase to a topic. If a response could be assigned to more than 1 topic area, the allocation was agreed on by discussion. We consid-
ered a topic to have been mentioned whether it occurred once or multiple times during an interview, therefore topic frequency was determined by analyzing whether a topic was mentioned at least once by individual children and parents. Overall topic frequency was calculated for the children’s responses. For the parents, phrases and topics were analyzed according to whether they reflected the parent’s perception of their child’s quality of life or whether they reflected the parent’s own quality-of-life concerns. Topics relating to the child’s HRQOL that were common to both the parent and child groups were analyzed to determine whether they were mentioned by parent-child pairs within the same family.

RESULTS

CHILD INTERVIEWS

For the 24 children with IXT, 106 phrases were extracted and 18 topic areas were apparent. The number of topics mentioned per child ranged from 0 (3 patients) to 11 (median, 3 topics). The overall topic frequency is shown in Table 3. The most frequently mentioned topic was worry (10 of 24 patients [42%]), including phrases such as “I worry a lot about my eyes,” “I worry about it getting worse,” and “I worry that people will think I’m weird.” “Troubled by blurriness” was mentioned by 8 of 24 patients (33%), eg, “I don’t like it wandering because it goes all blurry.” Comments from others, eg, “kids ask what is wrong with my eye” and “people say your eye is drifting” was also mentioned by 8 of 24 patients (33%). The next most frequently mentioned topics (7 of 24 children each [29%]) were ocular discomfort, eg, “My eyes hurt when I’m tired,” self-consciousness including phrases such as “My eyes are different from other kids” and “it kind of feels a little bit embarrassing,” and wish didn’t have it, eg, “I wish my eyes would stay straight” and “wish I could stop them from going in and out.”
Table 3. Topic Frequency of Health-Related Quality-of-Life Concerns Related to Children With Intermittent Exotropia, as Expressed by Children Themselves and by Parents

<table>
<thead>
<tr>
<th>Concerns Expressed by Child</th>
<th>Concerns Expressed by Parent About Child</th>
<th>Concerns Expressed by Parents</th>
</tr>
</thead>
<tbody>
<tr>
<td>Worry</td>
<td>10 (42)</td>
<td>15 (63)</td>
</tr>
<tr>
<td>Troubled by blurriness</td>
<td>8 (33)</td>
<td>9 (38)</td>
</tr>
<tr>
<td>Comments from others</td>
<td>8 (33)</td>
<td>6 (25)</td>
</tr>
<tr>
<td>Ocular discomfort</td>
<td>7 (29)</td>
<td>Reading 5 (21)</td>
</tr>
<tr>
<td>Self-consciousness</td>
<td>7 (29)</td>
<td>Self-consciousness 5 (21)</td>
</tr>
<tr>
<td>Wishes he or she didn’t have it</td>
<td>7 (29)</td>
<td>Relationships 4 (17)</td>
</tr>
<tr>
<td>Appearance to others</td>
<td>4 (17)</td>
<td>Self-confidence 4 (17)</td>
</tr>
<tr>
<td>Self esteem</td>
<td>4 (17)</td>
<td>Concentration 3 (13)</td>
</tr>
<tr>
<td>Bothered by eyes</td>
<td>3 (13)</td>
<td>Self esteem 3 (13)</td>
</tr>
<tr>
<td>Reading</td>
<td>3 (13)</td>
<td>Clinic visits 2 (8)</td>
</tr>
<tr>
<td>Appearance to self</td>
<td>2 (8)</td>
<td>Computer 2 (8)</td>
</tr>
<tr>
<td>Depth perception</td>
<td>2 (8)</td>
<td>Eye contact 2 (8)</td>
</tr>
<tr>
<td>Monocular eye closure</td>
<td>2 (8)</td>
<td>Ocular discomfort 2 (8)</td>
</tr>
<tr>
<td>Ocular sensation</td>
<td>2 (8)</td>
<td>School 2 (8)</td>
</tr>
<tr>
<td>Troubled by need to correct exotropia</td>
<td>2 (8)</td>
<td>Troubled by blurriness 2 (8)</td>
</tr>
<tr>
<td>Exotropia triggered by staring</td>
<td>1 (4)</td>
<td>Worry 2 (8)</td>
</tr>
</tbody>
</table>
| Parent’s need to correct    | 1 (4)                                  | Exotropia triggered by tele 
 vision b | 1 (4) | Depth perception 1 (4) |
| Troubled by double vision   | 1 (4)                                  | Exotropia triggered by te
vision b | 1 (4) | Eye fatigue 1 (4) |
|                              |                                        |                              |

Table 4. Topic Frequency of Health-Related Quality-of-Life Concerns of Parents Whose Children Have Intermittent Exotropia

<table>
<thead>
<tr>
<th>Concerns Expressed by Parents</th>
<th>Concerns Expressed by Parents, No. (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Worry regarding possible surgery</td>
<td>15 (63)</td>
</tr>
<tr>
<td>Worry regarding possible deterioration</td>
<td>14 (58)</td>
</tr>
<tr>
<td>Worry regarding other potential effects of IXT</td>
<td>11 (46)</td>
</tr>
<tr>
<td>Worry regarding others noticing</td>
<td>8 (33)</td>
</tr>
<tr>
<td>Comments from others</td>
<td>7 (29)</td>
</tr>
<tr>
<td>Parent’s need to correct</td>
<td>6 (25)</td>
</tr>
<tr>
<td>Bothered by eyes</td>
<td>5 (21)</td>
</tr>
<tr>
<td>Clinic visits</td>
<td>5 (21)</td>
</tr>
<tr>
<td>Wish child didn’t have it</td>
<td>5 (21)</td>
</tr>
<tr>
<td>Forced to accept it</td>
<td>4 (17)</td>
</tr>
<tr>
<td>Guilt</td>
<td>3 (13)</td>
</tr>
<tr>
<td>Appearance to others</td>
<td>1 (4)</td>
</tr>
<tr>
<td>Lack of definitive medical plan</td>
<td>1 (4)</td>
</tr>
<tr>
<td>Parent-child relationship</td>
<td>1 (4)</td>
</tr>
</tbody>
</table>

Concerns Expressed by the Parents Relating to Their Child’s HRQOL

The overall frequency of the 14 topics mentioned by parents regarding their own HRQOL is shown in Table 4. Because of the large number of phrases describing specific worries, topics relating to worry were divided into individual areas of concern rather than grouping all types of worry together. The number of topics mentioned per parent ranged from 1 to 8 (median, 3 topics). The most frequently mentioned topics were worry-related, specifically worry regarding possible surgery (15 of 24 parents [63%]), including phrases such as “worry about needing surgery,” “worry about surgery making it worse,” and “worry about risks of surgery” and worry regarding possible deterioration (14 of 24 parents [58%]), eg, “worried it will get worse,” “worried about losing depth perception,” and “I don’t want 1 eye to give up.” The topic of worry regarding other potential effects of IXT (11 of 24 parents [46%]) included worry about individual concerns such as ability to drive in the future, potential affects on schooling, and potential affects on personality. Worry regarding others noticing was mentioned by 8 of 24 parents (33%), including phrases such as “worry that people will think he is mentally handicapped” and “worried about the social thing.” Seven of 24 parents (29%) mentioned phrases regarding comments from others, eg, “people say there is something wrong with my child’s eyes” and “other people ask if we notice.”

Comparison of Child and Parent Responses Regarding Child’s HRQOL

Regarding the child’s HRQOL, 7 topics were mentioned only by the children with IXT and not by the parents (Table 3). Three of the 24 children (13%) mentioned that they were bothered by their eyes, eg, “bothered that my eyes go out.” Other topics mentioned only by children were appearance to self, monocular eye closure, ocular sensation, troubled by double vision, parent’s need to correct, and exotropia triggered by staring (Table 3). Eleven topics were mentioned by the parents that were not mentioned by the children (Table 3). Four parents

PARENT INTERVIEWS

For the 24 parents, 217 phrases were extracted. Ninety-six of 217 phrases (44%) related to the child’s quality of life, covering 22 topic areas; 121 of 217 phrases (56%) related to the parent’s quality of life, covering 14 topic areas.

Concerns Expressed by the Parents Relating to Their Own HRQOL

The number of topics mentioned per parent ranged from 1 to 8 (median, 3 topics). The most frequently mentioned topics were worry-related, specifically worry regarding possible surgery (15 of 24 parents [63%]), including phrases such as “worry about needing surgery,” “worry about surgery making it worse,” and “worry about risks of surgery” and worry regarding possible deterioration (14 of 24 parents [58%]), eg, “worried it will get worse,” “worried about losing depth perception,” and “I don’t want 1 eye to give up.” The topic of worry regarding other potential effects of IXT (11 of 24 parents [46%]) included worry about individual concerns such as ability to drive in the future, potential affects on schooling, and potential affects on personality. Worry regarding others noticing was mentioned by 8 of 24 parents (33%), including phrases such as “worry that people will think he is mentally handicapped” and “worried about the social thing.” Seven of 24 parents (29%) mentioned phrases regarding comments from others, eg, “people say there is something wrong with my child’s eyes” and “other people ask if we notice.”
(17%) thought that IXT affected their child’s self-confidence, using phrases such as “sometimes he holds himself back because of his eyes” and “it affects self-confidence” and relationships, eg, “affects his ability to interact with others.” Three of the 24 parents (13%) perceived IXT to cause problems with their child’s concentration, eg, “he lacks some concentration because of it.” Other topics mentioned by parents only were clinic visits, computer, eye contact, school, eye fatigue, general awareness of deviation, headaches, and exotropia triggered by television (Table 3).

There were 11 topics relating to the children’s HRQOL that were mentioned in both child and parent interviews (Table 3). Analysis of these 11 topics by parent-child pairs (ie, a parent and child within the same family) showed poor agreement between parents and their children (Table 5). Of the topics mentioned by both parents and children, the topics with the best agreement were reading (2 of 6 families [33%]), appearance to others (3 of 10 families [30%]), and comments from others (4 of 19 families [21%]) (Table 5).

We have identified a range of specific HRQOL concerns affecting children with IXT and their parents. The topics most frequently expressed by children with IXT were worry concerning their eyes, comments from others, and that they were troubled by bluriness. Parents identified the main effects of IXT on their children to be dealing with comments from others. Interviews also identified affects of the children’s IXT on the HRQOL of the parents themselves, primarily worry relating to the possibilities of surgery, deterioration, and other effects of IXT.

Nearly half of the children interviewed expressed worry related to their eye condition. There are few data on the presence or nature of worry in children with strabismus, but worry regarding health has been identified even in children without health concerns, affecting 69% in a study by Silverman et al.17 Whether children with IXT worry more about their eyes than children without will be analyzed in the next phase of our study. It is possible, especially in younger children, that the word worry encompasses other more specific concerns, such as self-consciousness, that children were not able to articulate.

Concern regarding comments from others was expressed by one-third of children, with most phrases indicating that comments were from other children. Also, more than half of the parents identified comments from others as affecting their child’s HRQOL. The age at which children may become aware of strabismus in others was studied by Paysse et al5 who found that at approximately 6 years of age children began to express an overall negative response to playing with dolls with strabismus. It is unclear whether comments from others should influence management decisions, but assessing such comments appears important in evaluating HRQOL in patients with IXT.

Children expressed concern regarding bluriness of vision more frequently than we expected. Expressions related to being troubled by bluriness were associated with awareness of the eye wandering. It is possible that children used the word blurry to describe diplopia, although patients with IXT are typically thought to suppress one image. Alternatively, it may be that comments relate to experiences of children when not wearing their habitual refractive correction (glasses were prescribed in 10 of 24 children [42%]). Nevertheless, in the context of the interview it was clear that the experience of bluriness was associated with an episode of exotropia. We speculate that in IXT a child’s observations of blurry vision may indicate the effect of accommodative or motor convergence mechanisms used to control the exodeviation, but this remains controversial.18,19 Alternatively, it may be that children describe as blur the change in visual experience noticed when suppression occurs.

Analysis of parent-child pairs showed poor agreement, with most parents raising concerns that were different than those raised by their child and vice versa. Our finding of poor parent-child concordance is consistent with the abstract by Powell et al12 who found poor correlation between parent and child scores on all Pediatric Quality of Life Inventory20 questionnaire subscales. Lack of agreement between self- and proxy-reporting, especially in social or emotional domains, is well recognized20,21 and creates challenges for management decisions. For children with
IXT, the differences between child and parent concerns raise important questions regarding the indications for and aims of treatment, ie, whether we are treating the child or the parent. Self-reporting is considered the standard measure of quality of life, but in children this cannot always be achieved. Where possible, a combination of parent- and self-reporting has been recommended. Using data from this study, we aim to develop questionnaires for self-reporting and parental reporting of HRQOL in IXT.

Regarding the effect of IXT on the HRQOL of parents, the most frequently occurring topics were worry-related. Without exception, worry was caused by potential, not actual, effects of IXT, ie, the possibility of surgery, deterioration, or others noticing the strabismus. It is possible that transference of parental concerns may lead to worry in children. Some parental comments in our study suggest that medical advice conveys a sense of uncertainty as to the likely course of disease and the optimal treatment strategy. Improved understanding of the natural history of IXT and indications for surgery may lead to improved parental education and result in reduced parental worry and possibly reduced child worry.

There are several potential weaknesses to our study. Most of the children recruited were girls, although this is consistent with the reported prevalence of IXT. We did not find a noticeable difference between concerns reported by girls and boys, but this may be worthy of further study. To create a less threatening atmosphere, we did not insist on parents leaving the room when the child was interviewed. As a result, most of the younger children (<10 years) were interviewed in the presence of their parents. This may have limited the openness of the child, although there was no indication that the parent's presence restricted what the child said. When interviewing parents of younger children, it was not always possible to ask children to leave. Again, this may have limited how much the parent revealed regarding particular concerns. An additional potential weakness is that we did not standardize the timing of interviews relative to the clinical examination. It is possible that parent and child responses might have differed before vs after the examination.

We have identified a range of HRQOL concerns affecting children with IXT. Parental perceptions of the effects of IXT on their children's HRQOL differed from concerns identified by the child. We also identified concerns affecting the HRQOL of the parents. The specific concerns identified in this study will be used to develop patient-derived condition-specific HRQOL questionnaires both for children with IXT and their parents. Such questionnaires should prove valuable in assessing the needs of an individual patient, determining criteria for intervention, and improving our understanding of the nature of IXT.

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