Ophthalmology Research: An International Journal
3(3): 59-66, 2015, Article no.OR.2015.011
ISSN: 2321-7227

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Child-rated and Parent-rated Quality of Life in Childhood Intermittent Exotropia: Findings from an Observational Cohort Study

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Authors' contributions

Author DB contributed to study design, data analysis and interpretation, drafting, revising and approving the manuscript; authors NA, PT and RHT contributed to data interpretation, manuscript revision and approval; author CJP contributed to data acquisition and interpretation, manuscript revision and approval; author MPC contributed to funding acquisition, study design, data interpretation, manuscript revision and approval.

Article Information

DOI: 10.9734/OR/2015/15246
Editor(s):
(1) Tatsuya Mimura, Department of Ophthalmology, Tokyo Women's Medical University Medical Center East, Japan.
Reviewers:
(1) Anonymous, Israel.
(2) Anonymous, Turkey.
Complete Peer review History: http://www.sciencedomain.org/review-history.php?iid=889&id=23&aid=7360

Original Research Article

Received 15th November 2014
Accepted 3rd December 2014
Published 16th December 2014

ABSTRACT

Purpose: To use the Pediatric Quality of Life Inventory (PedsQL™) to describe generic quality of life (QOL) in children with intermittent exotropia [X(T)], to examine changes in scores, and to compare scores in children with X(T) to those of age-matched healthy cohorts.
Methods: PedsQL™ was administered to children and parents as part of the UK Improving Outcomes in Intermittent Exotropia (IOXT) study. Excluding 27 children with co-morbidity, PedsQL data was available from 365 parents and 152 children. Paired-samples t-tests examined change in PedsQL™ scores over time. One-sample t-tests and mean differences compared scores between

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children with X(T) and healthy UK samples.
Results: Mean parent-rated PedsQL™ scores from the X(T) cohort at baseline were: 90.6 (Physical Health), 78.2 (Emotional Functioning), 88.8 (Social Functioning), 83.4 (School/Nursery Functioning), 83.6 (Psychosocial Summary), 86.2 (Total). Mean baseline child-rated scores were: 78.1 (Physical Health), 76.5 (Emotional Functioning), 73.6 (Social Functioning), 72.2 (School/Nursery Functioning), 74.2 (Psychosocial Summary), 75.5 (Total). X(T) parents rated their child's QOL similar to healthy children's parents, except for poorer School/Nursery Functioning in 2-4 year olds. X(T) children rated their QOL significantly better than age-matched healthy children. There were no significant changes over time.
Conclusion: Using the PedsQL™ we were unable to detect significant effects of X(T) on generic QOL. However, evidence for PedsQL's utility in this condition remains limited without further investigation in larger samples and concurrent control groups. Further qualitative work and consideration of condition-specific measures in UK cohorts are needed before practitioners can better inform parents about psychosocial impacts of X(T).

Keywords: Intermittent exotropia; divergent strabismus; quality of life; surgery; child; parent; proxy.

1. INTRODUCTION

Intermittent exotropia [X(T)] is one of the commonest forms of childhood strabismus. In X(T), one eye intermittently drifts outwards when the child is looking at distant objects, or is tired or inattentive. Potential functional consequences include loss of stereovision and amblyopia. Parents often have concerns relating to social exclusion and bullying because of the appearance of the strabismus [1].

Childhood X(T) has been associated with subsequent poor psychological well-being into young adulthood [2,3]. There may also be more immediate implications for the child's quality of life (QOL) yet relatively little exists in the literature about this [4-8]. One group of researchers, from the Mayo Clinic in North America, has studied parental and children's concerns regarding the effect of X(T) on QOL, leading to the recent development of their Intermittent Exotropia Questionnaire (IXTQ) [6-10]. As far as we are aware, there have been no other reports of paediatric QOL in this specific condition and the relevance of this group's work to a different population and healthcare system in the UK is unestablished.

QOL estimates of patients, including those with ophthalmologic conditions, often differ significantly from those of physicians [11]. Capturing child and parental perspectives is important since parental insight may not correspond with that of their children [12] and there is growing recognition that each needs to be taken into consideration. Parent-child agreement on QOL ratings in X(T) has been found to be poor [13].

A widely-used and accepted generic QOL instrument is the Pediatric Quality of Life Inventory (PedsQL™), which has strong evidence of reliability and validity in healthy children and those with a range of acute and chronic illnesses [14]. However, evidence of its usefulness in assessing generic QOL in children with X(T) is very limited. Here we describe parent- and child-rated PedsQL™ scores from a UK observational study of X(T) (the Improving Outcomes in Intermittent Exotropia (IOXT) study) [15-17]. The primary aim of this paper is to examine change over time in self- and parent-rated PedsQL™ scores. A secondary aim is to compare baseline PedsQL™ scores to those of unaffected age-matched cohorts.

2. METHODS

2.1 Participants

The IOXT study involved 26 children's eye clinics /orthoptic departments [15]. Between May 2005 and December 2006, collaborating departments approached parents/carers of children under 12 years, diagnosed with X(T) within the previous 12 months, without significant coexisting ocular pathology (e.g. cataract). In addition to clinical assessment, demographic details were recorded at enrolment including age, gender, and general health. Treatment status was documented (observation only, conservative treatment for squint such as glasses/patching, eye muscle surgery, or treatment for visual acuity only). Clinical and QOL follow-up ended in December 2008 (December 2009 for surgery patients). As this was an observational study, clinicians managed children according to their usual clinical

criteria. Informed consent was obtained from parents/guardians. The study received a favourable opinion from UK North West Multi-Centre Research Ethics Committee.

2.2 Outcome Measures

Generic QOL data were collected from parents and children using the PedsQL™ 4.0 Generic Core Scales (http://www.pedsql.org/index.html). We used the age-appropriate 'proxy' (parent/carer) version for those with children aged 2-4 years (toddler version), 5-7 years and 8 years or older, and the child's self-rated version for those aged 5-7 and 8 years or older. The PedsQL assesses QOL on four individual scales: Physical Health (8 items), Emotional Functioning (5 items), Social Functioning (5 items) and School Functioning (5 items, or 'Nursery/Day Care Functioning' for the toddler version which has 3 items). These four scales can be amalgamated to form a Total score (23 or 21 items) and 2 summary scores: Physical Health summary and Psychosocial Health summary. We followed developers' scoring instructions by reversing and linearly transforming scores to a 0-100 scale (higher scores indicating better QOL), and, for missing data, imputation of the mean of completed items in a scale when 50% or more are completed.

PedsQL™ questionnaires were administered in the clinic setting as part of the IOXT study assessment in all except 2 cases. In accordance with PedsQL™ administration guidelines, child versions were self-completed by those aged 8 or older. If the child was 5-7 years, instructions and questions were read to the child word for word by an orthoptist who then circled his/her response. Parent versions were self-completed. A written protocol and training video were produced for use by study orthoptists to ensure PedsQLs were administered in a standardised way. This generic instrument has been widely validated in numerous conditions and in healthy groups, and we found that its psychometric properties specifically in children with X(T) were generally good [13].

Baseline PedsQL™ data were scheduled to be collected within 12 weeks of enrolment. Follow-up appointments were arranged on a 3-monthly basis during the first year of the study and 6-monthly thereafter for an average period of 2 years. These visits involved both clinical and QOL review. Outcome QOL is the latest PedsQL™ completed, but at least 6 months after the initial QOL assessment.

2.3 Data Analysis

Data were analysed using SPSS version 19.0. Summary statistics (mean scores, standard deviations (SD)) are reported for each PedsQL™ scale. Analyses are performed across the whole cohort of children with X(T), i.e. for all age groups combined, and (see Supplemental Material) repeated for age-specific versions of the PedsQL™. Paired-samples t-tests were performed to examine change over time.

Baseline scores are compared with age-matched published scores [18,19] from healthy UK samples: mean differences and one-sample t-tests were used. Comparisons with healthy UK samples are possible for those aged between 2 years and 4 years 11 months [18] and for those aged between 5.5 and 8.5 years [19].All scales/sub-scales are available for use in the 2-4 year old comparison. Only the Total, Physical Health and Psychosocial Summary scores can be used in the older age-group comparison, as individual scores for Emotional, Social and School/Nursery Functioning were not published for the 5.5 to 8.5 year-old healthy group.

One-way analysis of variance was used to examine whether severity of X(T) at baseline was associated with QOL at initial assessment. Severity was determined by scores on the Newcastle Control Score (NCS) [20] with a score of 1 to 3 denoting mild, 4 to 6 moderate, and 7 to 9 severe X(T). One-way analysis of variance was used to explore any association between treatment and PedsQL™ scores (see Supplemental Material).Given the scarcity of reports of QOL in X(T), and to place this study in context, further descriptive comparison was made of PedsQL™ scores in this sample and those reported in the American study [8].

Co-morbidity was present in 27 children, the most common co-existing health issues being asthma and developmental delay. We have excluded those with co-morbidity from our analyses as they had significantly poorer QOL compared to those without other health conditions (see Supplemental Material).

2.4 Statement of Ethics

We certify that all applicable institutional and governmental regulations concerning the ethical use of human volunteers were followed during this research.

3. RESULTS

3.1 Participant Characteristics

Baseline PedsQL™ data were available from 365 parents and 152 children (excluding 27 children with comorbidity). Mean (SD) age at time of initial parent PedsQL™ was 52 (22) months. Mean (SD) age at time of initial child PedsQL™ was 74 (16) months. 57% of the children were female.

3.2 Baseline PedsQL™ Scores

Mean (SD) baseline parent-rated scores for all 365 parents were 86.2 (12.8) on Total score, 90.6 (12.8) on Physical Health, 78.2 (18.8) on Emotional Functioning, 88.8 (15.5) on Social, 83.4 (17.6) on School and 83.6 (14.7) on Psychosocial Summary. Mean (SD) baseline scores for all 152 children with QOL data were 75.5 (14.7) on Total score, 78.1 (17.4) on Physical Health, 76.5 (19.5) on Emotional Functioning, 73.6 (23.2) on Social, 72.2 (19.6) on School and 74.2 (16.2) on Psychosocial Summary. Age-specific PedsQL™ scores are provided in the Supplemental Material.

3.3 Paired Data

Mean interval between baseline and outcome PedsQL™ was 24 months (range 6 to 48) for parents and 20 months (6 to 48) for children. Paired data (baseline and outcome scores) were available from 301 parents and 128 children: Fig. 1 shows their baseline and corresponding outcome scores. For parent-ratings there was a small, statistically significant deterioration over time on Total (mean difference 1.5, 95% CIs 0.1 to 2.9, p=0.036), School/Nursery (mean difference 2.5, 95% CIs 0.3 to 4.8, p=0.025), and Psychosocial Summary (mean difference 1.9, 95% CIs 0.3 to 3.5, p=0.023) scales. Paired data within age-specific versions are provided in the Supplemental Material. For child-ratings (Fig. 1) there were no significant changes in self-rated scores.

Treatment received was not significantly associated with PedsQL™ scores in any of the parent- or child-rated PedsQL™ scales at baseline or outcome (see Supplemental Material). Mean parental PedsQL™ scores were higher at outcome on each scale (except Physical Health) for those with a better surgical outcome, defined as a NCS of between 0 and 2. However, this was not statistically significant (see Supplemental Material).

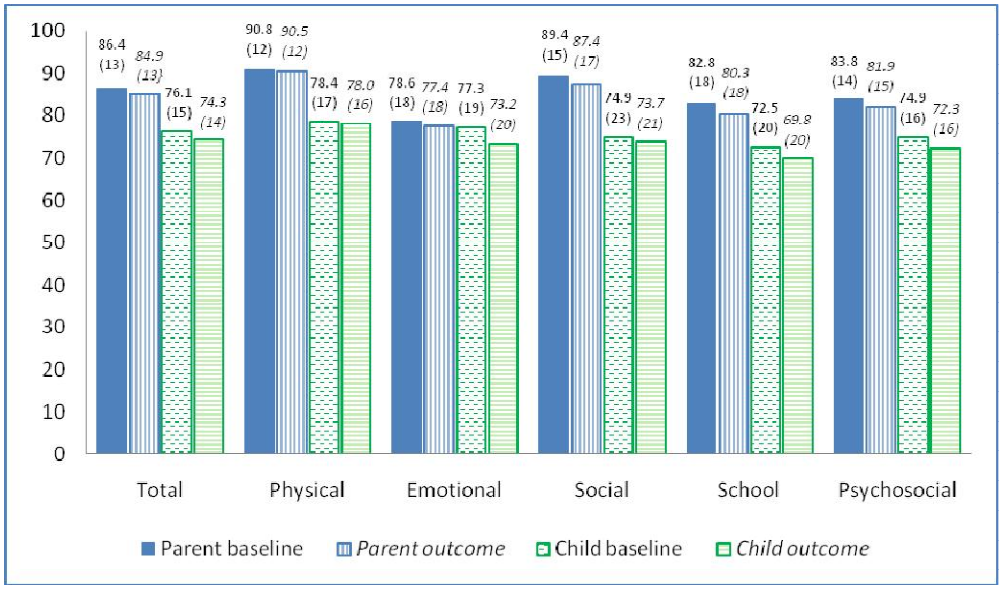


Fig. 1. Mean (SD) parent and child PedsQL scores at baseline and outcome

3.4 Comparisons with Unaffected Children (UK Sample)

3.4.1 2-4 year olds

Parents of 2 to 4 year-old children with X(T) rated their child's QOL similarly to those of age-matched healthy children in terms of Total and summary (Physical Health and Psychosocial) scores (Table 1). Further details are provided in the Supplemental Material.

3.4.2 5.5 to 8.5 year olds

In the 5.5 to 8.5 year-old group, there were no significant differences between X(T) parents' and healthy parents' ratings on Total or summary scores (Table 1). Children with X(T) rated their own QOL as significantly better than the age-matched healthy sample on the Total, Physical and Psychosocial Summary scales (Table 1).

3.5 Cross-cultural Comparisons

Fig. 2 shows that baseline parental PedsQL™ scores were very similar to those in a recent North American study of QOL in X(T).[7] In contrast, child-rated scores were significantly lower amongst our sample compared to their American counterparts with X(T) (one-sample t tests, p=0.005 to p<0.001).

3.6 Squint Severity

Although children with severe X(T) generally had poorer mean parent- and self-rated PedsQL™ scores than those with mild or moderate X(T), this was not statistically significant. Details are reported in the Supplemental Material.

4. DISCUSSION

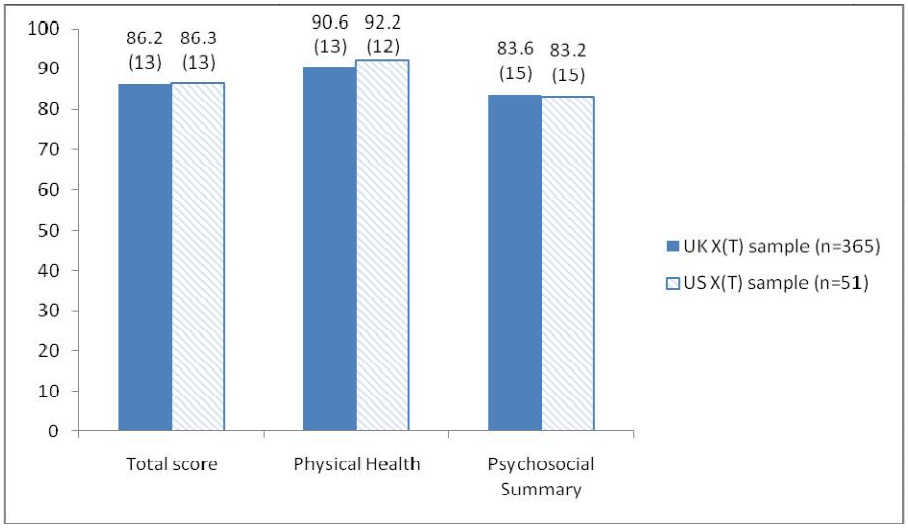
Drawing on data from a large UK multicentre observational study of children with X(T), we have described child-rated and parent-rated generic QOL using the widely-used and validated PedsQL™. We found that parent-rated PedsQL™ scores for children with X(T) were similar to those of unaffected cohorts in the UK, while the self-rated scores of the children themselves were better than those of unaffected children. While this finding is of interest, it is important to keep in mind that, as a generic measure, the PedsQL™ may not be detecting QOL issues that are especially pertinent to X(T). Our findings differ from those of a research group from the Mayo Clinic in North America (the only other group to have used the PedsQL™ specifically in this condition) [8]. In their study of 51 children with X(T) and 47 controls, they found that parent-rated scores were poorer in children with X(T) than in controls, while child-rated scores were similar between affected and unaffected children [8]. The differences between the UK and North American studies are most likely due to variations in the way the healthy samples were recruited given that the latter benefitted from a concurrent control group. Age is also a likely factor, since the age of the healthy sample in the American study ranged from 5 to 16 years whereas those in the present study ranged from 2 to 8.5 years.

Table 1. Mean difference in PedsQL scores: X(T) sample compared to age-matched healthy samples

	Mean (SD):			
Parental scores 2 to 4 years	**Healthy sample** (n=256)	**X(T) sample** (n=234)	**Mean difference; 95% CIs**	**p value**
Total PedsQL	87.8 (8.7)	89.0 (10.2)	1.2; -0.2 to 2.5	0.083
Physical Health	92.6 (9.1)	92.3 (10.3)	-0.3; -1.7 to 0.99	0.62
Psychosocial Summary	84.6 (10.5)	86.8 (12.2)	2.2; 0.63 to 3.8	0.006
Parental scores 5.5 to 8.5 years	**Healthy sample** (n=149)	**X(T) sample** (n=82)	**Mean difference; 95% CIs**	**p value**
Total PedsQL	79.9 (11.7)	80.8 (15.2)	0.9; -2.4 to 4.3	0.58
Physical Health	86.1 (11.4)	88.0 (15.9)	1.9; -1.6 to 5.4	0.28
Psychosocial Summary	76.7 (13.0)	76.9 (17.0)	0.2; -3.5 to 3.9	0.89
Child scores 5.5 to 8.5 years	**Healthy sample** (n=149)	**X(T) sample** (n=92)	**Mean difference; 95% CIs**	**p value**
Total PedsQL	71.8 (14.4)	77.8 (13.9)	6.0; 3.1 to 8.9	<0.001
Physical Health	76.4 (14.0)	80.8 (15.7)	4.4; 1.1 to 7.6	<0.001
Psychosocial Summary	68.9 (16.2)	76.3 (15.2)	7.4; 4.2 to 10.5	<0.001

**one-sample t-test*

a) Parental scores



b) Child-rated scores

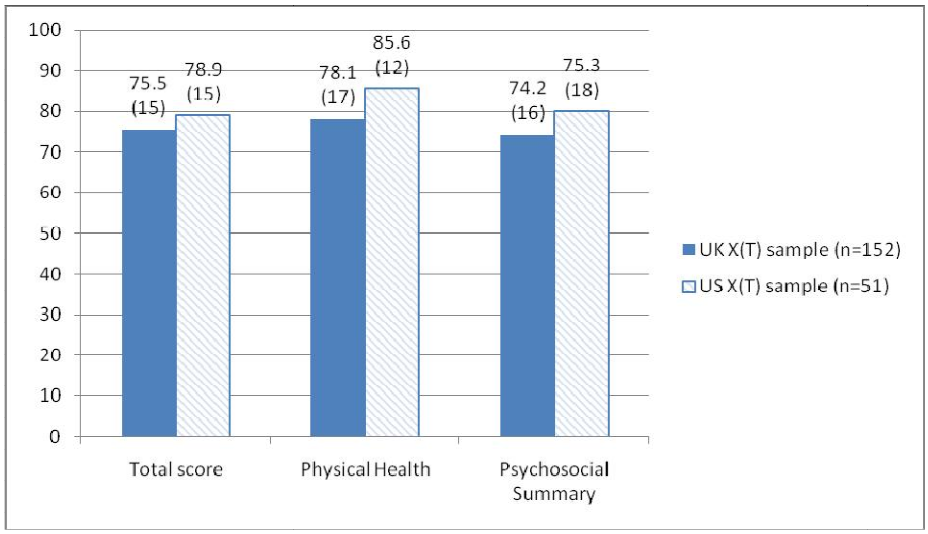


Fig. 2. Comparison of IOXT study PedsQL™ scores with those from a recent study of X(T) in North America

We found no effects of treatment on PedsQL™ scores, and no significant deterioration in self-rated PedsQL™ scores over an average follow-up period two years. While some parent-rated PedsQL™ scores did worsen slightly over time within the youngest age group, the size of these changes is unlikely to be meaningful [21].

Generic measures such as the PedsQL™ are important because of their applicability to a diverse range of conditions, enabling comparisons between various groups including healthy populations and thereby placing scores into context. However, they may not be sensitive enough to demonstrate effects associated with strabismus generally or X(T) in particular, or its treatment. A particularly informative way to determine any effect of X(T) on QOL is through qualitative methods. Indeed qualitative research, also by the Mayo Group in North America, reveals the most frequently mentioned QOL 'concerns' of children with X(T) was worry [6]. From the parents' perspective, "comments from other people" was the most frequently mentioned concern.

Although no paediatric strabismus-specific measures were available at the start of our study, the qualitative findings in America have recently informed the development of the Intermittent Exotropia Questionnaire (IXTQ) [7]. The advantage of such condition-specific measures is that they should be responsive to any change over time, for example in those who undergo treatment for their condition, while the main disadvantage is that they can not be used across different conditions or to compare with unaffected populations. Thus there will remain a need for both condition-specific and generic patient-reported outcome measures, depending on the context of the study. A popular solution is the use of a 'battery' of outcome measures incorporating both specific and generic components, although this can be more time-consuming and may result in respondent burden.

An important limitation of this paper is that we did not have a concurrent control group with which to compare the PedsQL^{TM} scores of children with X(T) and we therefore used published, age-matched scores from unaffected UK children. The published data for the older control group were collected from one region of the UK. Consequently, there may have been geographical and socio-economic differences between this group and our national IOXT cohort. The data for unaffected toddlers were collected from a wider geographical area and from a variety of sources. Another important limitation is that, due to the young age of the children in the IOXT study, there were less baseline child-rated PedsQL^{TM} data with which to evaluate change over time from their own perspective.

The findings from the IOXT study show that UK children with X(T) did not appear to fare any worse than unaffected peers in terms of PedsQL^{TM} scores, and that there was no detrimental effect on their PedsQL^{TM} scores over time. These findings do not necessarily imply a lack of effect of X(T) on QOL. However, they do add to the sparse literature on the condition itself and on the utility of the universally-accepted PedsQL^{TM} as an outcome measure specifically in ophthalmology. While the PedsQL^{TM} has demonstrated reasonable psychometric properties in X(T),[13] the evidence regarding its direct relevance to children with eye conditions remains limited. The implications for the management of this condition in the UK are difficult to establish, although the lack of any significant difference between children with X(T) and those without it should be reassuring to worried parents. Further investigation of the comparison of generic and condition-specific instruments in larger cohorts will further help clinicians.

5. CONCLUSION

In conclusion, parent- and child-rated PedsQL^{TM} scores were similar to or better than those of unaffected cohorts in the UK. Scores were not altered by treatment or time. Local and international evidence about the impact of intermittent exotropia on a child's quality of life remains limited. Further qualitative work should be pursued together with further investigation of established QOL measures in larger samples, and with concurrent control groups, in order to inform researchers, practitioners and parents about the specific effects of childhood X(T) and in turn how to evaluate its treatment.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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Peer-review history:
The peer review history for this paper can be accessed here:
http://www.sciencedomain.org/review-history.php?iid=889&id=23&aid=7360