Long-term quality-of-life outcomes in children undergoing adenotonsillectomy for obstructive sleep apnoea: a longitudinal study

Randhawa, P.S.,* Cetto, R.,* Chilvers, G.,* Georgalas, C.† & Narula, A.A.*

*Department of Otolaryngology, St Mary's Hospital, Imperial Healthcare NHS Trust, London, UK, and
†Endoscopic Skull Base Center, Academic Medical Centre, University of Amsterdam, Amsterdam, The Netherlands

Accepted for publication 9 August 2011
Clin. Otolaryngol. 2011, **36**, 475–481

Objectives: To assess a cohort of patients who underwent adenotonsillectomy for obstructive sleep apnoea, 4 years after surgery for evidence of continued and long-term improvement in quality of life. We also sought to compare our results to Child Health Questionnaire scores obtained from our previous study. We also compared our data with a healthy UK children population from normative data available.
Design: Longitudinal study.
Settings: University Hospital Tertiary Referral Centre.
Participants: A 4-year follow-up study of 37 children who underwent adenotonsillectomy for obstructive sleep apnoea confirmed on polysomnography. There were 19 boys and 18 girls from our initial cohort. The primary caregiver completed the validated Child Health Questionnaire Parental Form version-28, 4 years after initial surgery. Our control group consist of 221 healthy children aged 6–18 that were included as 'normal' controls in a study looking at children with juvenile arthritis. The children were defined as healthy by a physician and/or after declaration by the parent.
Main outcome measure: Child Health Questionnaire Parental Form version-28 scores.
Results: A total of 33 patients (89%) from our initial cohort were contacted. The mean age was 10.6 (median, 11; range, 5–16). When compared with results obtained 3 months postoperatively, the mean scores were higher in five domains and were statistically significant in three subscales (Role Limitations $P < 0.00001$; Bodily Pain $P < 0.002$; and Global Health $P < 0.02$). There was a significant deterioration in Behaviour subscale ($P < 0.0007$) in spite of surgery. Compared with controls, 4-year follow-up scores were higher in five domains with the Global Health domain ($P < 0.0004$) being statistically significant. When the 4-year follow-up scores were compared with preoperative values, these were higher in all 13 domains with statistically significant improvements in nine domains, indicating that improvements had persisted 4 years after surgery. At 4 years, however, the means scores in many domains remain lower when compared with controls.
Conclusion: Quality-of-life data are an important measure when deciding on a specific clinical intervention. In the short term, quality-of-life measures have been shown to improve after adenotonsillectomy for obstructive sleep apnoea. Our study demonstrates that the benefits of surgery are still persistent and the children continue to improve in the long term.

Sleep-related disordered breathing encompasses a range of conditions from simple snoring, upper airway obstruction to obstructive sleep apnoea (OSA). It is relatively common in the paediatric population, and between 3.7% and 12.1% of children have a sleep-related breathing disorder and up to 4% may have OSA.[1,2] The peak incidence of OSA is in the 2–8 year age group because of prominent lymphoid hyperplasia. OSA can lead to significant and well-documented cardiovascular, neurocognitive, developmental and behavioural problems.[3]

Children with OSA have a poorer health status, and the impact on quality of life (QoL) has been poorly investigated. There is evidence to suggest that morbidity associated with OSA is comparable to juvenile arthritis or moderate asthma.[4–6] Adenotonsillectomy has been shown to be effective in this group of patients, and it helps prevent long-term complications.[7,8]

It is important for clinicians and parents to know the impact on QoL obtained from surgery in this group of

Correspondence: P. Randhawa, Department of Otolaryngology, St Mary's Hospital, Imperial Healthcare NHS Trust, Praed Street, London W2 1NY, UK. Tel.: 0207 8867566; Fax: 02078861394; e-mail: prandhawa13@hotmail.com

patients, both short and long term, as this will not only help in the decision-making process regarding surgery but also help third-party commissioners of health care make decisions on the effectiveness of an intervention. A number of studies have shown short-term improvement in quality-of-life measures post-adenotonsillectomy for OSA.[9–13] Data for long-term benefit on the other hand are not so readily available.[10–12,14]

Although more than 100 quality-of-life questionnaires exist for the adult population, only a handful exists for children.[15–18] These questionnaires form the basis of assessing the need for intervention; validate efficacy and measure relative and absolute cost-effectiveness. These questionnaires are necessary as the paediatric population are unable to adequately express suffering verbally or satisfaction with an intervention.

An earlier study from our centre demonstrated significant short-term benefits in QoL in a cohort of children who underwent adenotonsillectomy for OSA using the Child Health Questionnaire Parental Form version 28 (CHQ-PF28),[13] which is a generic parental questionnaire that is sensitive and reliable for measuring QoL in young children through their parents' responses.[19] The 28-item questionnaire is subgrouped into 13 domains evaluating the family's and child's well-being. This questionnaire has been extensively used in assessing QoL of children with asthma,[20] juvenile arthritis[21] and epilepsy[22] and has also been previously validated for use in adenotonsillar disease in the UK.[23]

The aim of our current study was to assess the same cohort of patients who underwent adenotonsillectomy for OSA, 4 years after surgery to assess any continued and long-term improvements in QoL.

Materials and methods

Design

Longitudinal study.

Setting

A tertiary, academic paediatric unit serving an inner city population of multi-ethnic background.

Patients

Of 37 patients who underwent adenotonsillectomy for OSA at our centre 4 years earlier, 33 were contacted. The data used as our control group of healthy children were obtained from a study on children with juvenile arthritis.[21] This control group consist of 221 healthy children who were recruited from local schools in the UK (age, 6–18 years) and among the healthy brother(s) and sister(s) of the juvenile arthritis children. The children were from a multi-ethnic background and socio-economic status recruited from centres participating in the study on juvenile arthritis around the UK. A child was defined as healthy after examination by a physician and/or based on the parent's declaration.

Main outcome measure

The main outcome measure was to determine whether health-related quality-of-life benefits post-adenotonsillectomy as shown with the CHQ-PF28 persisted in the long term. We compared our results with data obtained from our previous study[13] and with existing normative data, as derived from a recent UK study.[21]

Methods

We conducted a telephone questionnaire administering the CHQ-PF28 to the primary caregiver of the same cohort of patients who underwent adenotonsillectomy for OSA 4 years after initial surgery. All patients had preoperative polysomnography which confirmed their OSA. An apnoea/hypopnea of ≥1/h was used to diagnose OSA.

Statistical analysis

Data were collected using Microsoft Excel, and subsequent statistical analysis was carried out using SPSS release 17.0 for Windows (SPSS Inc., Chicago, IL, USA) and $P < 0.05$ was considered statistically significant. A two-tailed paired and unpaired, as required, *t*-test with Bonferroni adjustments for multiple comparisons was used for comparison between groups. No financial incentives were offered for participating in the study.

Ethical consideration

Ethical approval was obtained and granted from the Imperial College Healthcare Ethics board. Verbal consent was given by the primary caregiver to participate in our study.

Results

The study was carried out between February and June 2010. The CHQ-PF28 was administered to the primary caregiver in all cases and the mother in 88%. A total of 33 of the 37 (89%) patients of the initial cohort who underwent surgery were contacted. The mean age was 10.6 (median, 11; range, 5–16) *versus* 6.1 (median, 5.7;

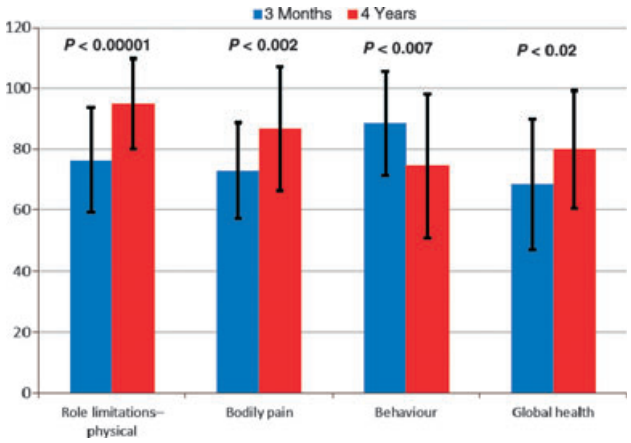

Fig. 1. Significant domain(s) of Child Health Questionnaire: 3 months postoperative and 4-year follow-up.

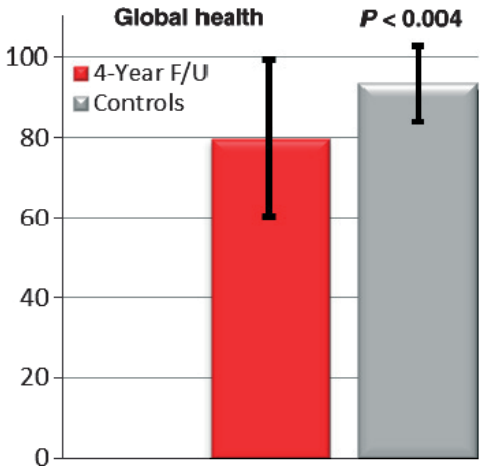

Fig. 2. Significant domain(s) of Child Health Questionnaire: 4-year follow-up and controls.

range, 1–11) at the time of surgery. There were 15 boys and 18 girls in our study population.

When our data was compared with data obtained 3 months postoperatively, there was improvement in the mean scores in five domains (Role Limitations Physical/Bodily Pain/Global Health/Family Cohesion/Parental Impact–Emotional). This was statistically significant in three domains (Role Limitations $P < 0.00001$; Bodily Pain $P < 0.002$; and Global Health $P < 0.02$) (Fig. 1).

The mean scores were lower in eight domains (Physical Functioning/Role Emotional Behaviour/Behaviour/Mental Health/Self-Esteem/General Health Perceptions/Parental Impact–Time/Family Activities) with the deterioration only in the Behaviour domain being statistically significant ($P < 0.007$).

When we compared our data with a healthy UK population, which acted as our controls, the mean scores were higher in domains related to behaviour, mental health, self esteem, family cohesion, parental impact–emotional and family activities, although these were not statistically significant. The mean scores of our follow-up study were once again lower in many domains (Physical Functioning/Role Emotional Behaviour/Role Limitation–Physical/Bodily Pain/Global Health/General Health Perception/Family Cohesion). This however was only statistically significant in the Global Health domain ($P < 0.0004$) (Fig. 2).

When our long-term data were compared with preoperative values, there were statistically significant improvements in nine of the 13 domains. Essentially, the beneficial effects of surgery were still evident 4 years after surgery and were statistically significant in Physical Functioning ($P < 0.01$), Role Limitations–Physical ($P < 0.002$), Bodily Pain ($P < 0.03$), Mental Health ($P < 0.007$), Global Health ($P < 0.0001$), General Health Perceptions ($P < 0.006$), Parental Impact–Emotional ($P < 0.0001$), Parental Impact–Time ($P < 0.003$) and Family Activities ($P < 0.0002$) (Fig. 3). A snapshot of the CHQ scores are provided in Fig 4. A summary of the mean CHQ scores are provided in Table 1.

Discussion

Synopsis of key/new findings

In our study, we measured quality-of-life improvements in children 4 years after initial surgery. Our results show an improvement in the mean scores and hence, QoL, in all individual domains of the CHQ-PF28 when compared with preoperative results.

Five domains continue to improve in the long term when compared results obtained from our previous study at 3 months postoperatively.[13] There were significant improvements in the domain for Bodily Pain ($P < 0.002$), Role Limitation–Physical ($P < 0.00001$) and Global Health ($P < 0.02$). This could be explained by the generalised improvement in general health that is obtained by surgery causing the child to be more active and hence less physical limitation. There was a significant deterioration in the Behaviour domain (88.5 ± 17.1 *versus* 74.7 ± 23.6; $P < 0.007$) when compared with results obtained 3 months postoperatively, which once again could well be related to the improvement in the child's general well-being and activeness. This long-term mean score however was comparable to the score in our controls (74.7 ± 23.6 *versus* 74.3 ± 17.5; $P < 0.93$).

When long-term results were compared with controls, they were not significantly different, with the exception of the Global Health domain (79.8 ± 19.4 *versus* 93.3 ± 9.5;

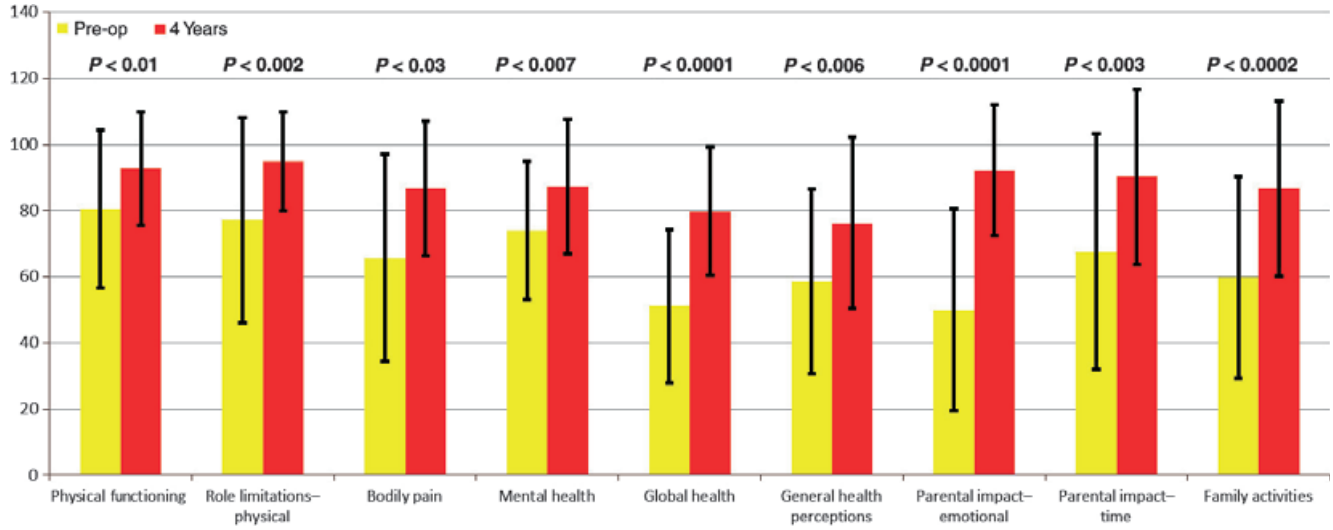

Fig. 3. Significant domain(s) of Child Health Questionnaire: preoperatively and at 4 years.

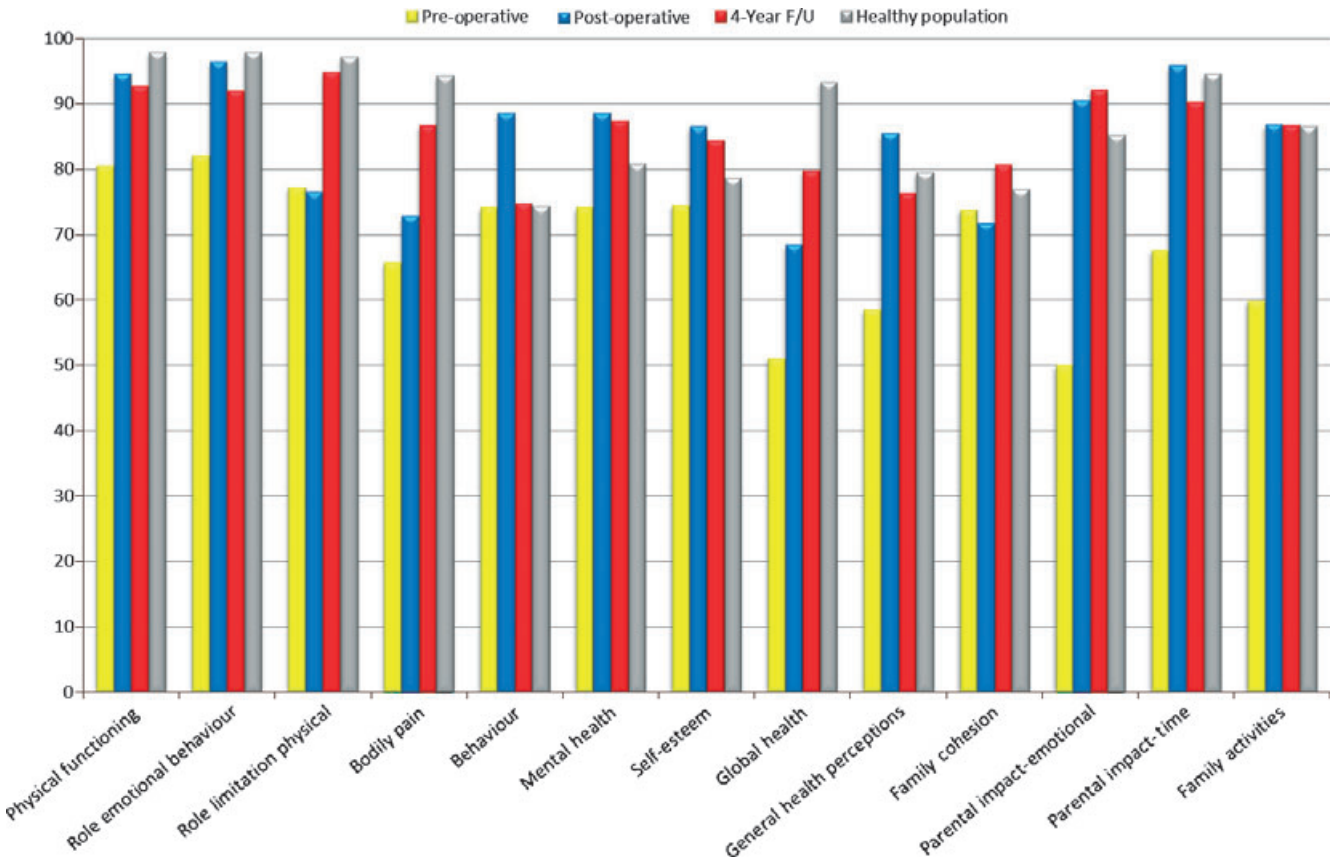

Fig. 4. Summary of Child Health Questionnaire scores.

$P < 0.0004$). This score however did show continued improvement when compared with values preoperatively, 3 months postoperatively and in the long term (51.1 ± 23.1 *versus* 68.4 ± 21.4 *versus* 79.8 ± 19.4). This finding supports the evidence that children with OSA have a poorer general health as a consequence of the disease (4, 5, 6) and that surgery confers improvements in Global Health which continues to improve in the long term.

Table 1. Comparison of Child Health Questionnaire scores 3 months postoperative, 4 years and controls

Scale	Preoperative (n = 42)		Postoperative (n = 37)		4-year follow-up (n = 33)		Controls (n = 221)	
	Mean	SD	Mean	SD	Mean	SD	Mean	SD
Physical Functioning	80.5	23.8	94.6	19.2	97.9*	9.6	92.7	17.1
Role Limitations–Emotional Behavioural	82.1	30.8	96.4	10.5	97.8	10.0	91.9	23.7
Role Limitations–Physical	77.2	31.1	76.4	17.2	97.2*†	11.9	94.9	14.9
Bodily Pain	65.7	31.3	72.9	15.8	94.3*†	13.8	86.7	20.4
Behaviour	74.1	22.6	88.5	17.1	74.3†	17.5	74.7	23.6
Mental Health	74.1	20.9	88.5	19.6	80.8*	10.9	87.3	20.4
Self-Esteem	74.5	24.3	86.5	26.3	78.6	14.9	84.3	22.3
Global Health	51.1	23.1	68.4	21.4	93.3*†	9.5	79.8‡	19.4
General Health Perceptions	58.5	27.9	85.5	23.1	79.5*	13.7	76.3	25.9
Family Cohesion	73.8	23.9	71.8	20.9	76.9	22.0	80.6	24.4
Parental Impact–Emotional	50.0	30.6	90.5	20	85.2*	15.9	92.1	19.8
Parental impact–time	67.6	35.7	95.8	13.4	94.5*	10.4	90.2	26.4
Family Activities	59.8	30.6	86.8	16.1	86.5*	16.9	86.7	26.5

Statistically significant domains.
†Three months *versus* 4 years.
*Preoperative *versus* 4 years.
‡Four years *versus* controls.

Clinical applicability of the study

The impact of adenotonsillar disease on the paediatric population has been shown to be significant and largely underestimated. It has been estimated that 27 000 tonsillectomies are carried out annually in the UK.[24] Although the majority of tonsillectomies are for recurrent tonsillitis, there has been a gradual increase in the number of children having surgery for OSA over the last few years. During this period, the criteria for tonsillectomy have become more stringent and have been classed as 'non-essential' procedures by certain health authorities. Although monetary aspects are always considered when these decisions are made, quality-of-life data are rarely taken into account because they are scanty.

In our study, we used the CHQ-PF28 parental questionnaire, which is a validated generic instrument for measuring QoL in young children through their parents' responses. By using a generic questionnaire, we have the advantage of measuring overall health improvement in the child, unlike disease-specific instruments such as OSD-6 and OSA-18 which focus on areas of function that pertain to a particular disease. The CHQ-PF28, being generic, also allows health status in children with different disease processes to be compared. It also allows for production of generic health status data that may be of value when health policies and management decisions are made based on quality-adjusted life years.

The Glasgow Children's Benefit Inventory is another recently developed and validated generic post-interventional questionnaire. It has been used and validated in a variety of paediatric surgical procedures. At the time our initial study was carried out, this questionnaire was not in wide usage and as such we opted for the CHQ-PF28 which we continued in our follow-up study so that the long-term outcomes could be compared.

A number of studies have shown short-term improvement in quality-of-life measures. Georgalas *et al.*[13] showed an improvement in quality-of-life 3 months post-adenotonsillectomy. Flanary *et al.*,[11] using the same CHQ-PF28 questionnaire and also the OSA-18 questionnaire, also showed an improvement in QoL at between 3 and 6 weeks postoperatively. Using the OSA-18 disease-specific questionnaire, Goldstein *et al.* and Mitchell *et al.* showed improvement in QoL at 3, 4 and 7 months, respectively.[5,9,10] de Serres *et al.*[12] used the CHQ-6 questionnaire to show an improvement in QoL at 3 weeks postoperatively.

In the long term, Flanary's study also showed an improvement at between 6 and 12 months; however, the mean scores on the CHQ-PF28 showed a downward trend in the long-term responses which led the authors to suggest that patients sustained a long-term benefit immediately after surgery. The OSA-18 score in their study, on the other hand, showed a statistically significant improvement in the long term. Stewart *et al.*[14] using the CHQ-PF28 also showed long-term improvement in quality-of-life parameters at 12 months post-adenotonsillectomy. de Serres's study also showed an improvement in the long term although this was only 12 weeks postoperatively.

Mitchell *et al.*[10] to date has the longest follow-up study at 24 months using the non-generic OSA-18 tool. Their study revealed some important differences in short- and long-term follow-up. Scores for domains measuring sleep disturbances and physical suffering, i.e., loud snoring, breath holding spells, mouth breathing and nasal discharge, were lower in the long-term group suggesting that some symptoms recur in time. Scores in the domains measuring emotional distress, daytime problems and caregiver concerns did not differ in the long term. They inferred that although symptoms do recur in time, they were not severe enough for caregivers to be sufficiently concerned. Their study was not able to infer that QoL improvements were a result of surgical therapy as there was no control group. They felt the improvements in the underlying disorder may have occurred as part of its natural history, but they did not specifically measure general health with this instrument.

Using the GCBI, Schwenter *et al.* showed improvement in the four domains (vitality, emotional, physical health and learning) of the questionnaire. Their retrospective study had a mean follow-up duration of 3.4 ± 2.2 years. However, their response rate of only 43% could have led to significant bias as only those that had maximal benefit or vice versa would have responded.[25] A more recent study conducted by Wood *et al.* with a follow-up period of between 3 months and 2 years had a response rate of 59%. The study was essentially to compare a reduction tonsillectomy and conventional tonsillectomy and this showed an improvement in all domains of the GCBI but no differences in the subgroups.[26] It is important to mention that neither of these studies had objective confirmation of OSA with polysomnography. Both of these being post-interventional studies may be confounded by bias in that parents were feel their children were worse beforehand although the GCBI is constructed to eliminate this. In our patient population, we had obtained a preoperative CHQ-PF28 score that was used as a baseline.

Strengths of the study

The strengths of our study are that we have the longest follow-up period to date at 4 years, and our long-term results are comparable to other published studies. The CHQ-PF28 questionnaire we used in our study has been widely used to assess varies disease processes and has been previously validated for adenotonsillar disease in the UK.[23] We feel that by using the CHQ-PF28 data of a healthy population obtained by Nugent *et al.*[21] in their study of patients comparing QoL of patients with juvenile rheumatoid arthritis and an age-matched control group, our patient population age was well matched [mean age, 10.6 years (range, 5–16) *versus* 10.1 years (range, 1–14)]. All the patients in our study group had objective confirmation of OSA with polysomnography, which many studies have not had. We also used the now accepted Apnoea/Hypopnoea index (AHI) of one to make a diagnosis of OSA.[27] Many studies in the past have used an AHI of between 1 and 5, and this inevitably would not only have affected the number of children diagnosed with OSA but also those that were 'cured'.[28]

Limitations of study

The main weakness in our study was there was no formal randomised control group; however, this is difficult to achieve as the gold standard treatment for paediatric OSA is adenotonsillectomy, and it would be un-ethical not to treat documented OSA in view of the well-known short-term benefits and also the long-term complications. We acknowledge that the natural history of the disease may have played a part in the resolution of some of the symptoms and hence the improvements in some of the scores of the CHQ-PF28 domains.

Conclusion

Quality-of-life data are an important measure when deciding on a particular intervention. Adenotonsillectomy is curative for paediatric OSA, and we have evidence that quality-of-life measures improve in the short term. Our study demonstrates that quality-of-life measures continue to improve in the long term.

Keypoints

- OSA affects up to 4% of the paediatric population and has significant long-term consequences if left untreated.
- Children with OSA have a poorer health status and this is comparable to children with juvenile arthritis or moderate asthma.
- Whilst adenotonsilectomy is curative for the majority of children, the impact of QoL has been poorly investigated.
- QoL is an important measure to guide clinicians, parents and health care commissioners into decision making and also the effectiveness of an intervention.
- In the short-term, QoL has been shown to improve after surgery.
- Our longitudinal study, which is he longest to date, shows that this improvement is persistent in the long-term.

Conflict of interest

None.

References

1 Lumeng J.C. & Chervin R.D. (2008) Epidemiology of pediatric obstructive sleep apnea. *Proc. Am. Thorac. Soc.* **5,** 242–252

2 Powell S., Kubba H., O'Brien C. *et al.* (2010) Paediatric obstructive sleep apnoea. *BMJ* **340,** c1918

3 Kohler M.J., Lushington K., van den Heuvel C.J. *et al.* (2009) Adenotonsillectomy and neurocognitive deficits in children with Sleep Disordered Breathing. *PLoS ONE* **4,** e7343

4 Franco R.A. Jr, Rosenfeld R.M. & Rao M. (2000) First place – resident clinical science award 1999. Quality of life for children with obstructive sleep apnea. *Otolaryngol. Head Neck Surg.* **123** (1 Pt 1), 9–16

5 Mitchell R.B., Kelly J., Call E. *et al.* (2004) Quality of life after adenotonsillectomy for obstructive sleep apnea in children. *Arch. Otolaryngol. Head Neck Surg.* **130,** 190–194

6 Baldassari C.M., Mitchell R.B., Schubert C. *et al.* (2008) Pediatric obstructive sleep apnea and quality of life: a meta-analysis. *Otolaryngol. Head Neck Surg.* **138,** 265–273

7 Friedman B.C., Hendeles-Amitai A., Kozminsky E. *et al.* (2003) Adenotonsillectomy improves neurocognitive function in children with obstructive sleep apnea syndrome. *Sleep* **26,** 999–1005

8 Burg C.J. & Friedman N.R. (2010) Diagnosis and treatment of sleep apnea in adolescents. *Adolesc. Med. State Art Rev.* **21,** 457–479, viii

9 Goldstein N., Fatima M., Campbell T. *et al.* (2002) Child behaviour and quality of life before and after tonsillectomy and adenoidectomy. *Arch. Otolaryngol. Head Neck Surg.* **128,** 770–775

10 Mitchell R.B., Kelly J., Call E. *et al.* (2004) Long-term changes in quality of life after surgery for pediatric obstructive sleep apnea. *Arch. Otolaryngol. Head Neck Surg.* **130,** 409–412

11 Flanary A. (2003) Long-term effects of adenotonsillectomy on quality of life in paediatric patients. *Laryngoscope* **113,** 1639–1644

12 de Serres L.M., Derkay C., Astley S. *et al.* (2000) Measuring quality of life in children with obstructive sleep apnea. *Arch. Otolaryngol. Head Neck Surg.* **126,** 1423–1429

13 Georgalas C., Babar-Craig H., Arora A. *et al.* (2007) Health outcome measurements in children with sleep disordered breathing. *Clin. Otolaryngol*. **32,** 268–274

14 Stewart M.G., Friedman E.F., Sulek M. *et al.* (2001) Validation of an outcomes instrument for tonsil and adenoid disease. *Arch. Otolaryngol. Head Neck Surg.* **127,** 29–35

15 Kubba H., Swan I.R. & Gatehouse S. (2004) The Glasgow Children's Benefit Inventory: a new instrument for assessing health-related benefit after an intervention. *Ann. Otol. Rhinol. Laryngol.* **113,** 980–986

16 Berzon R.A., Simeon G.P., Simpson R.L. Jr *et al.* (1995) Quality of life bibliography and indexes: 1993 update. *Qual. Life Res.* **4,** 53–74

17 Erling A. (1999) Methodological considerations in the assessment of health related quality of life in children. *Acta Paediatr. Suppl.* **88,** 106–107

18 Pal D.K. (1996) Quality of life assessment in children: a review of the conceptual and methodological issues in multidimensional health status measures. *J. Epidemiol. Community Health* **50,** 391–396

19 Landgraf J.M., Abetz L. & Ware J.E. (1999) *Child Health Questionnaire (CHQ): A User's Manual*. Second Printing. Health Act, Boston, MA

20 Rutishauser C., Sawyer S.M. & Bowes G. (1998) Quality-of-life assessment in children and adolescents with asthma. *Eur. Respir. J.* **12,** 486–494

21 Nugent J., Ruperto N., Grainger J. *et al.* (2001) The British version of the Childhood Health Assessment Questionnaire (CHAQ) and the Child Health Questionnaire (CHQ). *Clin. Exp. Rheumatol.* **19** (Suppl. 23), S163–S167

22 Gilliam F., Wyllie E., Kashden J. *et al.* (1997) Epilepsy surgery outcome: comprehensive assessment in children. *Neurology* **48,** 1368–1374

23 Georgalas C., Tolley N. & Kanagalingam J. (2004) Measuring quality of life in children with adenotonsillar disease with the Child Health Questionnaire: a first UK study. *Laryngoscope* **114,** 1849–1855

24 Indications for Tonsillectomy: Position Paper. ENT UK, 2009. http://www.entuk.org/position_papers/documents/tonsillectomy

25 Schwentner I., Schmutzhard J., Schwentner C. *et al.* (2008) The impact of adenotonsillecotomy on children's quality of life. *Clin. Otolaryngol.* **33,** 56–66

26 Wood J.M., Harris P.K., Woods C.M. *et al.* (2011) Quality of life following surgery for sleep disordered breathing: subtotal reduction adenotonsillectomy versus adenotonsillectomy in Australian children. *ANZ J. Surg.* **81,** 340–344

27 Marcus C.L., Omlin K.J., Basinki D.J. *et al.* (1992) Normal polysomnographic values for children and adolescents. *Am. Rev. Respir. Dis.* **146** (5 Pt 1), 1235–1239

28 Hoban T. & Chervin R. (2007) Sleep-related breathing disorders in childhood: description and clinical picture, diagnosis and treatment approaches. *Sleep Med. Clin.* **2,** 445–462