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**CHILD AND PARENT
REPORTED OUTCOME
MEASURES: A SCOPING
REPORT FOCUSING ON
FEASIBILITY FOR
ROUTINE USE IN THE
NHS**

A Report to the Department
of Health, 2009



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**CHILD AND PARENT REPORTED OUTCOME MEASURES: A SCOPING
REPORT FOCUSING ON FEASIBILITY FOR ROUTINE USE IN THE NHS**

A Report to the Department of Health, 2009

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EXECUTIVE SUMMARY

The purpose of this report was to assess the feasibility of developing child and parent-reported outcome measures alongside other quality service indicators, particularly for children with long-term conditions.

A scoping and synthesis of pertinent policy and academic literature was undertaken; reviews of generic and condition-specific instruments were identified and further references collected from citation searches and by hand-searching reference lists.

Attention was paid particularly to the constructs assessed by instruments, the age at which children can self report their health and complete questionnaires, administration and processes, and also legal issues.

The findings suggest that it would be feasible to implement routine collection of child and parent reported outcome measures, but that a number of key conceptual and methodological complexities must be carefully considered:

1. Given the variability of constructs assessed by generic PROMs for children, and the range of important outcomes for ‘healthy’ children advocated in Healthy Lives, Brighter Futures, it is crucial that the purpose of measurement be clearly defined to distinguish ‘wellbeing’ from ‘health status’ and physical and social ‘functioning’.
2. Variability in children’s development and abilities means chronological age is not a hard and fast criterion for judging when children can self report their health and complete a questionnaire. Nonetheless, children aged eight years and above are widely believed to be competent to complete such questionnaires; children aged five to eight years may be able to self report but some are likely to require assistance to complete a questionnaire.
3. A generic child and family-reported outcome measure could be selected across health conditions. One of several candidate instruments candidates could be used, with the selection process principally dependent on the construct of interest. In addition, structured reviews of child and family-reported outcome measures for selected specific conditions affecting children could also be undertaken.
4. Established health utility measures for children are limited to the parent-reported HUI, which assesses health states without reference to child development or social roles. The HUI is accepted by NICE in their technology appraisal guidance, however we suggest that further evidence is required before any utility instrument is implemented for routine use. Nevertheless, this situation may change in the near future as two child-appropriate health utility measures are being evaluated.
5. Informed consent from both the child and their parent or legal guardian must be documented prior to administration of questionnaires. Preferentially, the consent procedure would be conducted face to face in a clinical setting. Questionnaires can then be administered in the clinic or hospital, by mail or telephone interview, or using the Internet or email.

6. Children's own self reports should be gathered whenever possible; parent reports can be gathered for children unable to self-report but these must not be aggregated with self reports. Ideally both child and parent reports would be collected to enhance the interpretability of results when children's self reports are unavailable, but this must be considered from the perspective of cost and burden.

7. Given the heavy emphasis on the provision of family-centred services (FCS) in Health Lives, Brighter Futures, and the availability of a well developed parent-reported instrument for assessing FCS (the Measure of Process of Care, or MPOC), consideration should be given to implementing a measure of this important aspect of health care.

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1. INTRODUCTION

1.1 Purpose of the report

The policy document ‘Healthy Lives, Brighter Futures, the strategy for children and young people’s health’ contains the following commitment:

- Exploring the feasibility of developing child and parent reported outcome measures to sit alongside quality measures and accreditation schemes. This will particularly be the case for children with long-term conditions.

The aim of this report is to take this commitment forward, explore pertinent issues, and scope what further work is required to move towards implementing the use of child and parent reported outcome measures in the NHS. First, the policy context is reviewed and patient reported outcome measures are introduced. Second, an overview of generic, condition-specific and utility child and parent reported outcome measures is described. Third, several key feasibility issues are assessed including the constructs assessed, children’s age, administration and processing, and legal considerations. The findings are discussed and key issues meriting further consideration are highlighted in the executive summary.

1.2 Methodology

This report presents the findings of a scoping and synthesis of pertinent policy and academic literature relating to the feasibility of routine implementation of child and parent reported outcome measures. Targeted searching on *Pubmed* and knowledge of the topics identified structured reviews of generic and condition-specific instruments and these were appraised. Further references were collected through citation searches and by hand-searching reference lists of retrieved papers.

1.3 Children’s health and wellbeing

The Children’s Act 2004 provided legislation to support the Every Child Matters programme aiming to support children to be healthy, stay safe, enjoy and achieve, make a positive contribution, and achieve economic well-being. These policies were further consolidated by The Children’s Plan (Department for Children, Schools and Families, 2007) which set out ways in which Government will achieve the stated objectives through modifying public services and children’s environments. An emphasis was placed on supporting parents in bringing up their children through providing services that are family-centred.

Improving the health and wellbeing of children, and services to achieve that aim, is also underpinned by the National Service Framework for Children, Young People and Maternity Services (Department of Health, 2004). Standards 6-10 pertain to children receiving health and social services; Standard 10 specifically concerns children with long term conditions. Standard 10.1 states “For many children with long term health conditions, the aim of treatment and care is to manage their illness in such a way that they are able to enjoy and achieve fully in their lives and make a positive

contribution.” Therefore, when considering what outcomes to measure, one should be clear about the object of measurement, specifically whether it is health status per se, or their enjoyment of life, or their ability to achieve and contribute.

There have been further reports and policies to help particular groups of children, including disabled children (HM Treasury and the Department for Education and Skills, 2007) and those with mental health problems (Department of Health, 2008). The most recent report, *Healthy Lives, Brighter Futures*, recommends promoting a stronger emphasis on the voices of children and young people, explicitly in reporting their health and in the organisation of services. Better measures of service quality are advocated, notably assessing the extent to which services are family-centred.

The Office for National Statistics (ONS) is also becoming interested in measuring children’s wellbeing using routinely collected statistics and information from social surveys (Thomas, 2009).

1.3 Long-term conditions in childhood

There are numerous health problems that constitute long term conditions in childhood. These include respiratory problems (e.g. asthma), endocrine problems (e.g. diabetes), neurodisability (e.g. epilepsy, cerebral palsy, hearing or vision loss), mental health problems (e.g. depression, behavioural disorders), congenital anomalies and genetic disorders (e.g. cleft palate, clubfoot, cystic fibrosis, and neuromuscular conditions), incontinence, and also some cancers. There are also children who suffer major trauma causing brain, spinal or limb injury causing long term consequences.

Estimates of the prevalence of childhood disability in the UK have varied from 5-18% depending on the population and definition or indicator employed. A social gradient has been consistently observed across a number of studies; disabled children are much more likely to live in relatively poorer households and in families with lower income (Read et al., 2007).

Children with long term conditions are unlikely to be ‘cured’ and typically require ongoing support from health and other public services. Nevertheless, the health of children with organ level problems, such as renal or liver failure, can be improved dramatically through transplants. Similarly, congenital anomalies such as clubfoot and cleft palate can often be significantly improved or resolved through surgery. Epilepsy seizures or diabetes can be controlled to some extent through medication, and children with fluctuating conditions such as arthritis and asthma may have cycles of remission and relapse. Hence the health trajectories for children with long term conditions are extremely variable.

Although a cure may currently be beyond reach for most, much can be done to improve the health and wellbeing of children and young people with long term conditions. In fact, it is recognised (in *Healthy Lives, Brighter Futures*) that the outcomes for children with long term conditions have not improved to the same extent that has occurred for adults with long term conditions (Department of Health & Department for Children, Schools and Families, 2009).

Children with long term conditions frequently require support from a variety of health services, including various medical specialists, and also hospital and community-based allied health professionals. It is known that families sometimes have difficulties accessing appropriate services or acquiring necessary equipment such as wheelchairs. There is recognition that the way services are delivered and providing adequate information about services are crucial to improving children's health and wellbeing, in part through reducing burden on families. Outcome indicators of service quality must, wherever possible, include reporting from children and parents (Department of Health & Department for Children, Schools and Families, 2009).

1.4 Patient-reported outcome measures

Patient-reported outcome measures (PROMs) offer enormous potential to improve the quality and results of health services. They provide validated evidence of health from the point of view of the user or patient. They may be used to assess levels of health and need in populations, and in users of services, and over time they can provide evidence of the outcomes of services for the purposes of audit, quality assurance and comparative performance evaluation. They may also improve the quality of interactions between health professionals and individual service users.

Lord Darzi's Interim Report on the future of the NHS recommends that PROMs should have a greater role in the NHS (Darzi, 2007). The new Standard NHS Contract for Acute Services, introduced in April 2008, includes a requirement to report from April 2009 on patient-reported outcome measures (PROMs) for patients undergoing Primary Unilateral Hip or Knee replacements, Groin Hernia or Varicose Vein surgery. Furthermore, Lord Darzi's report 'High Quality Care for All' (2008b) outlines policy regarding payments to hospitals based on quality measures as well as volume. These measures include PROMs as a reflection of patients' experiences and views. Routine collection of PROMs is now in place for the selected elective procedures. Building upon this policy, a group at the University of Oxford were commissioned to undertake structured reviews and have recommended PROMs for several adult long-term conditions. The feasibility of implementing the adult measures on a widespread basis across the NHS is currently being evaluated. To date, there has been less attention paid to child and parent reported measures for assessing child health.

1.5 Child and parent reported outcome measures

The DH has committed to exploring the feasibility of developing child and parent-reported outcome measures for use in health and social care. There is a considerable body of evidence that children can self report their health using questionnaires (Eiser & Morse, 2001a; Riley, 2004). Furthermore, a plethora of instruments have been developed to assess various aspects of children's health and wellbeing; these have been catalogued and reviewed by various groups (Harding 2001; Eiser & Morse 2001a; Schmidt et al., 2002; Ravens-Sieberer et al., 2006; De Civita et al., 2005; Davis et al., 2006; and most recently by Solans et al., 2008).

Nevertheless, prior to selecting one or more instruments, several fundamental issues must be considered before their use can be implemented. Among the most important

issues are: (i) deciding which aspects of children's health and wellbeing should be assessed? (ii) at what age do children become competent to report their own health? (iii) can the reports of parents or carers be used as a proxy for their children?

In addition, there are legal aspects to consider regarding children's right to consent and report their own health, and when it is appropriate for parents to answer on their behalf without their consent. For instance, some children are unable to consent or give their own responses due to age or cognitive limitations. Furthermore, from a feasibility perspective, some children who have the cognitive ability will only be able to answer with assistance to read the question and communicate a response.

1.6 Types of outcome measures

Patient reported outcome measures (PROMs) can be used to assess a variety of constructs with different populations to answer specific questions. Broadly speaking, PROMs can be categorised into one of the following types:

- Generic multidimensional measures of subjective quality of life or wellbeing assess how people feel about aspects of life that are commonly believed to be important.
- Generic multidimensional measures of health status assess the level of objective functioning in various domains, such as physical, psychological and social.
- Generic measures of activities and participation assess whether and to what extent people take part in discretionary (recreational) and non-discretionary (activities of daily living) life situations in the context of the environment in which they live.
- Generic dimension-specific measures assess functioning or wellbeing in a single domain, such as physical or psychological or social, but also pain or self-esteem. As a single domain is assessed it may be explored in greater detail.
- Condition-specific measures assess functioning and/or wellbeing in aspects of life that are thought to be affected by a particular condition. The issues assessed are unlikely to be examined, in sufficient detail, in generic instruments.
- Health utility measures assess the value of health states, usually from a societal perspective. Initially there were only generic instruments; however, as the generic instruments could not be expected to be sensitive to changes in health for certain conditions, condition-specific health utility instruments are being developed.
- Service evaluation and satisfaction with service measures assess whether services are delivered in a certain way are adequate for needs, and/or whether respondents are satisfied and would recommend the service to others. Examples are measures of family-centred services or the process of transition to adult services.
- Patient-reported measures of their living environment seek to identify enabling or disabling aspects of home and community life. Overcoming environmental barriers can significantly improve the lives of people with long term conditions.

Although the categories described above appear discrete, a cursory view of available PROMs shows that they often combine objective and subjective elements, and may also include elements of service evaluation. The terms health status, quality of life, health-related quality of life, and functional status are frequently used interchangeably and there is no consistent agreement about what these titles really mean (Fitzpatrick et al. 1998). However, the implications of this inconsistency are not insignificant. Colver & Jessen (2000) emphasise the conceptual difference between measuring children's 'objective' health status as distinct from their 'subjective' perception of their quality of life. As McDowell and Newell emphasise, the distinction between objectivity and subjectivity is determined by 'what' is being assessed and not by 'who' makes the assessment (McDowell & Newell 1996).

The purpose of measurement is inextricably linked to the constructs that should be assessed as the results can otherwise be confusing. Using cerebral palsy as an example (N.B. cerebral palsy is an exemplar in the NSF for Children), studies have shown that whilst children's objective functioning is reduced by the severity of their impairments, their self-reported subjective quality of life was similar to that of general population norms and not mediated by severity (Dickinson et al., 2007). Similarly, whilst the health utility of children with cerebral palsy was in many cases considered poor, and in some cases extremely poor, their level of self-reported subjective wellbeing was fairly good and not associated with the severity of their disability (Rosenbaum et al. 2007). This is known as the 'disability paradox' (Albrecht & Devlieger, 1999), but is particularly salient for children with congenital or developmental conditions who may not have experienced life in any other health state. The implication of this issue is that, when selecting PROMs, both the purpose of measurement and the reference population to which the findings are being compared need to be clearly defined.

2. OVERVIEW OF PROMs FOR CHILDREN

Several substantive reviews were identified that focus on measures of children's health status, health-related quality of life and/or wellbeing. Eiser and Morse, (2001) reported a comprehensive review of measures of 'quality of life' for children with chronic conditions, and Schmidt et al., (2002) reviewed generic measures of health-related quality of life for children. A number of other reviews have been conducted to assess various conceptual, methodological and psychometric issues in the use of generic PROMs with children (notably: Harding, 2001; Ravens-Sieberer et al., 2006; De Civita et al., 2005; Davis et al., 2006; and Solans et al., 2008). A large number of PROMs that can be used with children are available, especially for children with a variety of long term conditions.

2.1 Generic PROMs

Solans et al. (2008) identified 30 generic PROMs for children of which 17 were developed in English speaking countries. These instruments cover various age groups but are predominantly intended for children at least five or eight years old.

Four generic instruments are described briefly to illustrate the different constructs that are assessed by each measure. The Child Health Questionnaire (CHQ) was developed in the USA for assessing 'health status' (Landgraf et al., 1998). The CHQ is most often reported using a parent reported version with 50 items, 14 domain scores and/or physical and psychosocial summary scores; self-reported versions are available for older children, though these are of different length and include different items than the parent versions. The KIDSCREEN instrument was developed simultaneously across the European Union as a self and/or parent report of health-related quality of life for children aged between 8 and 18 years (Ravens-Sieberer et al., 2008). KIDSCREEN takes into account children's concepts of health in terms of their physical, mental and social 'wellbeing'. In contrast, the Pediatric Quality of Life Inventory (PedsQL) (Varni et al., 2001), developed in the USA, is a child and/or parent reported measure of 'ill-being' by the frequency that a child is affected by a range of problems with their health and functioning. The Assessment of Life Habits (LIFE-H) is an interview-administered instrument for assessing the emerging concept of children's 'social participation' in daily and discretionary activities (Noreau et al., 2007). Participation in this context is synonymous with social inclusion, most definitions of which emphasise 'participation in social activities as the core characteristic' (Morgan et al., 2007).

Most generic measures have undergone tests of construct validity; nevertheless, as noted by Davis et al. (2006) and illustrated above, the constructs targeted by each instrument varies considerably. Most generic instruments have undergone evaluation of test-retest reliability and internal consistency, but relatively few have been assessed using both criteria (Solans et al., 2008). Some instruments have been shown to have discriminative validity by distinguishing extreme or known groups; however very few instruments have been evaluated for their responsiveness to change (longitudinal validity). Limited evidence is available to inform issues of acceptability or feasibility.

2.2 Condition-specific PROMs

Solans et al. (2008) identified 60 condition-specific instruments for children covering 27 conditions, of which 42 were developed in English-speaking countries.

There are several candidate instruments specific to asthma, diabetes and epilepsy. The PedsQL and DISABKIDS instruments both have condition-specific modules for assessing relevant problems. Structured reviews of condition-specific PROMs appraise the quality of instruments for a number of conditions, for example for children with respiratory conditions including asthma and cystic fibrosis (Quittner et al. 2008) and diabetes (de Wit et al., 2007). Mokkink et al. (2009) evaluated the quality of such papers, and provide a useful list of 21 reviews of condition-specific PROMs for children. However, Mokkink and colleagues are critical of inconsistent methodology and appraisal criteria used in these reviews.

The majority of the condition-specific instruments are reported to have acceptable internal consistency and validity, though less evidence is available to support test-retest reliability (Solans et al., 2008). Disappointingly, the property of responsiveness to change has been evaluated for relatively few condition-specific instruments. Limited evidence is available to inform issues of acceptability or feasibility.

There are several more condition-specific PROMs for children than those identified in the review by Solans et al; at least one instrument was omitted (for children with epilepsy) and we are aware of other PROMs that have become available since their search. It is beyond the scope of this report to appraise available condition-specific PROMs; however, targeted structured reviews would be likely to be successful in identifying instruments appropriate to many common long term conditions.

2.3 Health utility measures for children

Measuring health utility in children is problematic for a variety of reasons: pertinent issues include incorporating content that is developmentally appropriate, the extent to which children understand health states, and the appropriate source of valuation for preference scaling (Petrou 2003). Hence, perhaps unsurprisingly, a review of cost utility studies in children reported by Griebisch et al. (2005) concluded that the quality of research in this area was poor. A principal problem is that almost all health utility measures were developed for adults and lack validity for use with children.

The measure of health utility generally preferred by NICE is the EQ-5D. However, as the EQ-5D was not designed for use by children, NICE considers alternative preference-based measures such as the Health Utilities Index (HUI). The Health Utility Index Mark 2 (HUI2) was developed specifically for valuing children's health states (Feeny et al., 1995). The HUI2 health state profile is typically parent-reported and a sample of parents in Canada provides the reference population (Torrance et al. 1996); preference valuation from a UK population sample is also available (McCabe et al., 2005). The HUI measures focus on children's 'capacity' for generic aspects of physical function. In contrast, the EQ-5D assesses 'performance' of physical functions including social roles. As with all PROMs, the varying constructs will affect the results elicited and the interpretation. Thus, the purpose of measurement must be carefully considered. A licence fee must be paid to the developers in Canada to enable use of the HUI. The 15D, 16D and 17D family of health utility instruments were designed with children and adolescents in mind.¹ However these appear to be used infrequently in the UK.

Child focused utility measures recently developed include a child version of the EQ-5D² and the Child Health Utility 9D (CHU-9D).³ The dimensions of the CHU-9D were developed from qualitative research with children aged 7-11 years, and the valuation of children's health states has been conducted in the UK. The child version of the EQ-5D assesses the same concepts as the adult version; however the wording has been modified to be understood by children to enable self-report. The same preference weighting for EQ-5D health states is applied as for adult populations. We are not aware of any reports in peer reviewed literature detailing the performance of these two new instruments in English speaking countries.

¹ <http://www.15d-instrument.net>

² www.euroqol.org/eq-5d/eq-5d-versions.html

³ www.shef.ac.uk/scharr/sections/heds/mvh/paediatric

Finally, any move towards the use of age-specific health utility measures will limit our ability to inform decisions across individuals of different ages, and consequently, potentially affect the equity and efficiency of resource allocation decisions.

3. FEASIBILITY ISSUES

3.1 Age at which children can self report health

It is proposed that any children of school age can self report their health (Riley 2004). By this age, children typically have an awareness of the concept of health and illness, and, providing questions are administered using an appropriate means, they can respond to questions relating to these concepts. Children who have experienced health problems may have an increased awareness of such issues compared to their peers who have not needed health services (Kozinetz et al., 1999; Colver & Jessen 2000).

It is generally accepted that children eight years and older can reliably self report their health (Raven-Seiberer et al., 2006); but what about children seven years old and younger? Analyses of children's self reports using the PedsQL instrument stratifying by one year age bands demonstrated that indices of reliability and validity remained acceptable for children as young as five years old (Varni et al., 2007). In the UK children take the National Curriculum Tests (SATs) for Key Stage 1 in Year 2, when they are aged six to seven years old. The test involves completing a 15-20 minute exercise involving independently reading and answering questions.

Younger children and those with cognitive, sensory or communication difficulties may require assistance to complete questionnaires, such as having the questions read to them or someone to mark their responses. A small proportion of children will not be able to self report even with assistance when their cognitive and intellectual ability is such that they cannot understand the meaning of questions or formulate appropriate responses. Other children simply may not want to complete a questionnaire. However, these issues are not confined to children and have also to be considered with adults.

From the review by De Civita et al. (2005), before the age of 6 years children's thinking is largely dichotomous and their self concept and emotional response is predominantly based upon physical attributes and actions. Children progressively develop awareness of their psychological self but it is not until adolescence that children integrate physical and psychological aspects of self and contemplate the future systematically (De Civita et al., 2005). Harter and Whitesell (1989) provide further details of how children progressively develop an awareness of their sense of self and their emotional wellbeing.

Variability in children's development and ability means chronological age is not a hard and fast criterion for judging when children can self report their health and complete a questionnaire. However, the evidence is substantial that children aged eight years and older can self report; children between five and eight years old may be able to self report but some will require assistance to complete a questionnaire.

3.2 Content & constructs

Broadly speaking, qualitative analysis of the dimensions assessed by child and adult instruments would suggest that they address similar constructs, namely physiological, psychological, and social wellbeing and functional ability (Rajmil et al., 2004). However, the social context of children's lives differs from that of adults because of their dependence on their family, the relative importance of friends, experiences of school and play, and their aspirations for the future (Matza et al., 2004). Thus it is broadly accepted that adult questionnaires are not entirely appropriate for children because of deficiencies in their content and face validity (Levi & Drotar 1998; Ravens-Sieberer et al., 2006).

Empirical evidence of how adult instruments perform with children comes from the field of paediatric orthopaedics where the SF-36 and Euroqol EQ-5D were compared with child-focused instruments. Neither of the adult measures was able to discriminate between known differences in health states and both showed severe ceiling effects (Vitale et al., 2001a). Conversely, the Child Health Questionnaire and a condition-specific measure were shown to detect the known differences in some domains and demonstrated superior performance overall (Vitale et al., 2001b).

Furthermore, the social context of children's lives varies at different stages of development. Therefore, depending on the purpose of measurement, the content may need to be adjusted for different age groups. This is particularly the case when aiming to assess children's activities and social participation.

The conceptual basis behind the content of instruments for children under five years of age (parent-reported questionnaires) has been noted to be particularly weak, and no instrument appears to capture all relevant domains (Grange et al. 2007). In addition, as we have emphasised, the conceptual basis underlying the constructs assessed by generic measures for children up to 12 years old is inconsistent, with various domains and constructs assessed by each instrument (Davis et al., 2006; Waters et al., 2009).

Although a large number of instruments appropriate for children are available, the item content, constructs and domains assessed by any candidate instruments should be carefully examined with reference to the purpose of use.

3.3 Questions / items

In addition to broad content issues, the way specific questions are phrased must be age-appropriate for children. Conceptually, there may be differences when questions assess well-being (feelings) and ill-being (problems/symptoms) (Davis et al., 2006). Negatively worded items that focus on difficulties and what children cannot do or achieve, for instance as is assessed in the PedsQL and DISAKIDS instruments, may undermine children's self-esteem (Waters et al., 2009).

The terms used in questions must be easy for children to understand, and this can be assessed through cognitive interviewing. The Discrepancy QoL (formerly ExQoL) uses pictures to frame the questions (Eiser et al., 2000), while other instruments use storytelling or vignettes. Child-friendly fonts are commonly used in questionnaires.

One issue to be considered is the appropriate recall period for assigning health states. Younger children's ability to distinguish between periods such as 'past day', 'past week' and 'past month' or 'four weeks' is unknown, though children 8 years and older are believed to be able to reasonably recall and consider a four-week period (Ravens-Sieberer et al., 2006).

3.4 Response scaling

Response options must be interpretable and salient for children. The most common scaling in children's instruments uses frequency of problems or difficulties, intensity or frequency of feelings, and comparison with perceived 'ideal' or other children (Davis et al., 2006). Five response options appear to be valid for children 8 years and older, however there is some evidence that younger children polarise their responses and tend to select extreme options compared to parents (Davis et al., 2007). This means that they effectively use a three point scale regardless of the number of increments, consequently with less precision but not necessarily less validity or reliability. Elsewhere, there is evidence that children generally tended to avoid the extreme options (Theunissen et al., 1998).

The use of pictures (faces with different expressions) is common in instruments used to assess pain as these are thought to be readily understood by children (Stanford et al. 2006). Images of facial expressions are used to scale response options in an asthma-specific instrument (Christie et al., 1993) and other questionnaires. In the Discrepancy QoL children are asked to rate whether each of a series of pictures is 'like me' and then as 'I would like to be' (Eiser et al., 2000).

One problem in designing scales, especially probing sensitive subjects, is enabling children to endorse negative statements without being too self-critical, or undermining their self-esteem (Waters et al., 2009). One way around this issue is to offer alternative paired options of forced responses. Based on the work of Harter and Whitesell (1989), such an approach was used by Ronen et al. (2003) when designing their epilepsy-specific instrument; children are asked to compare two examples and indicate which children they are more like, and thereby legitimising either choice. Then in a second stage children rate to what extent they are like the example, creating a four-point scale that avoids neutral responses. Although perhaps this is seemingly more complex than standard response options, children above 8 years old appear able to understand and follow the instructions.

3.5 Use of proxy reporters

Although it is now accepted that children should self-report their health whenever possible, there are occasions when children are unable to self report, for example pre-school children or those with severe cognitive impairment. For pre-school age children self reports cannot be expected. A review of parent-reported instruments for children three to eight years old identified several instruments but was critical of the extent to which psychometric criteria were met (Cremeens et al., 2006). When a child is unable to self report due to cognitive limitations parents and/or other people who know the child well (such as peers, carers and teachers) may be asked to respond as a

proxy reporter. As there will be no child report for comparison in these cases, proxy reports are typically gathered even for children who are self-reporting so that the presence of any systematic biases can be assessed.

When parallel child and proxy report versions are available, it is imperative that these include the same items using the same words. However, this is not always the case; one of the most established instruments, the Child Health Questionnaire, uses different wording in the child and parent versions, and the length of the children's and parent's versions vary, making direct comparisons impossible.

The reliability of proxy reports has been examined using a number of instruments, and is now expected as an integral part of PROMs instrument development (U.S. Food & Drug Administration, 2006). Reliability is assessed by comparing child and proxy group means and standard deviations; and by computing the Intraclass Correlation Coefficient (ICC). An ICC exceeding 0.7 is proposed as a criterion for acceptable reliability. Absolute or chance-corrected agreement between dyads (calculated using the kappa statistic) is also informative, and is often only moderate (Upton et al., 2008).

Systematic biases occur when either children or proxies consistently report better or worse health than the comparator. Typically, children who have experienced health problems report better health than their parents do about them; conversely, children in non-clinical samples (predominantly without health conditions) tend to rate their health slightly worse than their parents (Eiser & Morse 2001b). The level of child-proxy reliability has often been presumed to be dependent on the construct assessed. Typically, higher reliability is found when assessing objective observable phenomena such as physical functioning. Conversely, lower reliability is reported for instruments or domains assessing more subjective states, such as social and emotional functioning or wellbeing (Pantell & Lewis 1987; Eiser & Morse 2001b).

However, such findings are not universal and higher reliability has also been shown in psychosocial domains in some instruments (Upton et al., 2008). Hence, the construct may not be the only complicating factor, and the salience of the construct to the health condition may also play a role (Upton et al., 2008). Disagreement has been suggested to relate to children's and parents' different reasoning and different response styles, rather than interpretation of items (Davis et al., 2007). Parent-related factors that have been shown to affect child-proxy reliability are aspects of parental wellbeing, such as stress (Upton et al., 2008). Other influences such as child's age, sex and culture have also been examined to a limited extent but have not been found to explain discordance between children and proxies (Upton et al., 2008).

It is important to recognise that disagreement and poor reliability between children's and proxies' ratings may be due to genuine difference in perspectives and opinion. Thus it is incorrect to conclude that either reporter is wrong (Eiser & Morse, 2001b). Rather, both perspectives should be gathered whenever possible, and valued equally despite any differences (Upton et al., 2008). Collusion between parents and children completing questionnaires creates a potential confounding factor when separate child and proxy reports are being sought. However, the noted differences between child and parent reports, described above, suggest that this presents little risk.

One key area where the child's parent or carer might be the appropriate respondent, even more so than the child, is in measures of service quality. In practice, the parent seeks health care on their child's behalf; thus the extent to which services meet their needs can best be judged by the parent or carer. The strategy Health Lives, Brighter Futures strongly advocates that child health services should be family-centred. The Measure of Processes of Care (MPOC) is a parent reported measure of the extent to which the health services are perceived to be family-centred (King et al., 1996; McConachie & Logan, 2003).

To conclude, proxy-reporting by a parent or carer will be the only way of assessing the health status of children who are not able or willing to self report. Nevertheless, the child and parent reports should not be aggregated as they represent different perspectives. Ideally, both child and parent reports should be gathered for all children, as this makes it possible to interpret the results when children do not self report. However, pragmatically, this must be weighed against the additional burden and cost of gathering and analysing these data.

3.6 Administration & Engagement

There are several modes of administering questionnaires; principally these are in the clinical setting when the child is attends an outpatient appointment (as is being done for the elective procedures); on the ward as an inpatient; by mail or telephone; or electronically using email, Internet or mobile phone/personal data assistant. Each has advantages and disadvantages (Streiner & Norman, 2008). A critical consideration is response rate; non-participation leads to sampling biases, and particularly risks excluding people with less education and lower socio-economic status. There are two stages to be considered: first the invitation, and second, administration of the questionnaire.

Inviting children and/or parents to complete questionnaires in a clinical setting is likely to achieve maximum participation, as staff will be on hand to seek informed consent and also to assist any children who need help in answering questions. Mailed surveys do not always achieve the 70-80% participation benchmark preferred by statisticians for increasing confidence in the absence of response bias. For example, in a survey by the Healthcare Commission in 2004 with families of children (0-17 years old) who had been in hospital, even with two reminder mailings the response varied from as low as 32% to a high of 64%, with a average of 50% (Ramm et al., 2004). In addition, as noted in the introduction, children with long term conditions are more often living in relatively more deprived households which are less likely to take part and respond to mailed surveys.

When invitations are to be sent by mail, either accompanied by questionnaires or directing families to a website, some consideration needs to be given on whether the addressee is the child or their parent/carer. Hospitals usually address appointment letters to the parents as the legal guardian; however in the context of PROMs, the invitation might be more appropriately addressed to the child directly.

Increasing interest has been paid to electronic systems for PROMs data collection, with such instruments sometimes referred to as an ePRO. Children are likely to be familiar with using computers and may enjoy and be more willing to use innovative methods when completing questionnaires; there is some evidence that children prefer the electronic method, for instance in children with asthma (Bushnell et al. 2003). An additional advantage may be completeness in the proportions of items answered. The electronic system for administering varies, for instance using computers, handheld devices and mobile phones, and Interactive Voice Response (IVR) programmes. Advances in such technology and resources, and their availability, are progressing rapidly year on year.

Gwaltney et al. (2008) reviewed empirical comparisons of responses provided by electronic and paper-and-pencil methods and report general equivalence using different methods. Such equivalence has been reported using a condition-specific for children with diabetes (Varni et al., 2008). Coons et al. (2008) recommend criteria for the evidence required to support such equivalence. They recommend that cognitive debriefing interviews and usability testing should be undertaken to ensure respondents understand questions and response options in the same way. Full psychometric testing is necessary only when modifications have been made in the migration to electronic format that are likely to introduce the potential for response bias.

Another issue is the costs inherent with different methods. If children and/or parents complete questionnaires in clinic using pen and paper then the main costs are in data entry; mailed questionnaires also require data entry and additionally incur costs for assembling questionnaire packages, reminder letters and postage. From a financial perspective, perhaps the most efficient method would be for questionnaires to be completed electronically using the Internet. This circumvents the need for data entry, thus avoiding the cost and risk of errors during this stage. There is a set up cost for creating the website and this must be compared with the costs of setting up a database for manual data entry of questionnaires completed by pen and paper. Invitations could be made in clinic and/or by mail; questionnaires could be completed online in clinic or at home. There is little evidence available to predict likely participation using this method of administration.

In summary, depending on the target population, children can be invited to complete questionnaires in clinic or by mail, with the face-to-face invitation likely to increase response. There are likely to be significant advantages in administering questionnaires electronically in terms of cost, completeness and minimising data entry errors.

3.7 Ethics & legal issues

The UN Conventions on the Rights of the Child and the Rights of Persons with Disabilities advocate that children themselves should report their own health outcomes whenever possible. Therefore children should be shown the same respect for personal autonomy accorded to adults when invited to complete questionnaires about them. Children should be approached directly and their explicit informed consent should be sought prior to administering a questionnaire. The Conventions have not been directly implemented into statute within the UK but we see these principles underlying the Children Act. The legal position in the UK regarding

decision-making by children in a medical context is largely dependent upon the age of the child. This law has predominantly developed in regard to physical intervention rather than the giving of information by children as is the case with PROMS.

Guidance is provided by the General Medical Council (2007) for doctors and others to assess children's competency to consent. The Gillick case established that children under sixteen years can give consent to advice, treatment or physical examination, if they have 'sufficient understanding and intelligence to enable [them] to understand fully what is proposed'. The ruling established that children under sixteen years may have capacity to consent to medical treatment, and that doctors may treat such a child without parental consent or knowledge. However the ruling stressed that it would be very unusual circumstances that would justify a doctor withholding information from a parent. Therefore, unless there are compelling reasons to exclude parents from the consent procedure (which seems unlikely in the context of PROMs), parents should also be asked to consent to the use of the questionnaires.

Thinking about and responding to questions about their health is not without some risk of harm for the child. The process could be puzzling and bring into focus issues that may make them unhappy. Therefore procedures are required for supporting children who become confused or upset by the process (McIntosh et al., 2000). Parents should be fully informed of these potential concerns. In addition, when answering an apparently anonymous questionnaire, a child could potentially report information that they are reluctant to communicate in other ways. This places a dutiful responsibility on those gathering such information to have explicit procedures in place to screen questionnaires for any signs that raise suspicion of, for example, child abuse or neglect, or suicidal ideation or criminal behaviour.

For infants and children with intellectual deficits who are unable to provide informed consent, or complete questionnaires themselves, a parent/carer can be approached as a proxy informant. Nevertheless, children who are competent can decline to consent and not complete questionnaires. In these instances, guidelines recommend that children be asked to assent or consent for their parent/carer to complete a questionnaire on their behalf (General Medical Council, 2007).

Notwithstanding our recommendation that questionnaires should be screened for any evidence suggestive of child abuse or neglect, suicidal ideation or criminal behaviour, potential respondents should be assured at the invitation stage that their responses will be kept confidential and reported anonymously once data are aggregated for analysis.

In summary, the invitation and consent stages should preferably be conducted face to face in a clinical setting; the informed consent of both children and parents should be documented. Children should be offered the opportunity to self-report their health whenever possible or assent for a parent to proxy report on their behalf. Confidential screening of questionnaires should occur before questionnaires are made anonymous to check for any issues requiring clinical or legal advice or action.

4. DISCUSSION

The purpose of this report was to assess the feasibility of developing child and parent-reported outcome measures alongside other quality service indicators, particularly for children with long-term conditions. On the face of the findings presented, it would certainly appear to be feasible; however, a number of conceptual and methodological complexities must be considered and/or monitored and evaluated.

First, although a large number of generic and condition-specific PROMs for children already exist, they are believed to assess a broad and inconsistent range of constructs. Hence, it is essential to define clearly the purpose of measurement before selecting any specific instruments. The main distinction is between instruments that assess functioning, or what children are able or choose to do, versus instruments that assess how children feel about their health state, i.e. their wellbeing. If the latter were the purpose then KIDSCREEN, a generic instrument that has been thoroughly developed and tested across the European Union in recent years, would be a strong candidate.

Second, if specific conditions are to be targeted, from a pragmatic point of view, it would be advisable to start with conditions where cognitive difficulties are relatively rare. Nevertheless, one should not exclude important areas of child health and long-term conditions that are heavily dependent on health services. Intellectual impairment is associated with some neurodisabilities such as epilepsy and cerebral palsy; however there is ample evidence that many children with these conditions are competent and willing to complete questionnaires.

Third, proxy reporting can, to some extent, overcome barriers to assessing the health of children who cannot self-report; but, as they represent quite different perspectives, these should never be aggregated. Ideally both child and parent reports would be collected to enhance the interpretability of results when children's self reports are unavailable, but this must be considered from the perspective of cost and burden.

Fourth, the age at which children should be invited will depend on the age group targeted by the developers, and the extent to which PROMs have been shown to be valid and reliable for different age groups. Where appropriate children aged 5 years and older could be considered competent and included, though children under 8 years of age are likely to need parent or other assistance to complete questionnaires.

Fifth, attention should be paid to methods of administration likely to engage children and to ensure that they are supported during and after completing questionnaires. Availability of resources (people to assist children and/or computers) may depend on whether data are collected in primary, hospital, or community health care settings. Providing a small token reward (e.g. stickers, etc.) merits consideration.

Finally, the content and psychometric performance of potential PROMs should be appraised. The criteria proposed by Fitzpatrick et al. (1998) are pertinent, namely appropriateness, reliability, validity, responsiveness, precision, interpretability, acceptability, and feasibility. A Task Force of the International Society for Pharmacoeconomics and Outcomes Research (ISPOR) will shortly report

recommendations for good practice in the development and use of PROMs with children.⁴ The document will help identify other important criteria for consideration.

⁴ www.ispor.org/TaskForces/PROCA_background.asp

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