Health-related quality-of-life of children with speech and language difficulties: A review of the literature

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Abstract

A review of the literature has been undertaken to examine health-related quality-of-life (HRQoL) of children and adolescents with speech and language difficulties (SaLD), with a particular focus on evidence regarding the domains of HRQoL most affected by SaLD. Twelve electronic databases were searched for articles on this topic published from 1966 to January 2011. Seven studies suitable for inclusion were identified. These papers were reviewed in relation to their participants, sample size, study design, and outcome measures, which differed considerably across studies. From the studies reviewed, there is emerging evidence that HRQoL can be compromised for children and adolescents with SaLD relative to their peers, and some consensus that the social domain of HRQoL is most impacted. Overall, the review highlights a paucity of research in this area and recommendations are made as to how research may advance. Most importantly, studies examining the effect of child and family factors in mediating or moderating the relationship between SaLD and HRQoL are required. This knowledge will support the identification of children with SaLD at risk of poorer HRQoL outcomes and inform intervention strategies through the identification of relevant risk and protective factors.

Keywords: Systematic review, quality-of-life, children.

Introduction

Outcomes for children with speech and language difficulties

Speech and/or language difficulties (SaLD) are among the most frequently observed developmental problems affecting children (Prelock, Hutchings, & Glascoe, 2008). Speech difficulties include problems with the production (articulation) of speech sounds, the flow or rhythm (fluency) with which speech is produced, or the pitch, volume, or intonation (quality) of the voice (Biddle, Watson, Hooper, Lohr, & Sutton, 2002). Language difficulties relate to problems with understanding (receptive) and/or use (expressive) of spoken, written, and/or other communication systems (signs, symbols, etc.) (Biddle et al., 2002). Language difficulties may involve any combination of (1) the form of language (phonology, morphology, and syntax), (2) the function of language (pragmatics), and/or (3) the content of language (semantics) in communication (American Speech-Language-Hearing Association, 1993).

Prevalence rates of SaLD vary widely; however, in one recent population-based Australian study of 4–5-year-old children, a quarter of parents surveyed expressed concerns regarding their child’s ability to talk and produce sounds, and 9.5% of parents reported concerns about their child’s receptive language skills (McLeod & Harrison, 2009). Children with more apparent speech and expressive language deficits and those with more severe SaLD are generally identified before school age, while those with less severe difficulties may only be recognized following school entry (Laing, Law, Levin, & Logan, 2002). It is well established that, for many children, SaLD are associated with a range of adverse social, emotional, and educational outcomes (Cohen, 2006). Children with receptive language difficulties tend to exhibit poorer long-term outcomes than those with expressive language difficulties, especially in relation to behaviour, learning, and academic attainment (Laing et al., 2002).

The extent to which SaLD tend to persist or resolve depends on the nature of the child’s difficulty (i.e. speech difficulties only, language difficulties only, or both speech and language difficulties), and their access to speech-language pathology intervention. Law, Boyle, Harris, Harkness, and Nye...
(2000) reviewed the literature on the natural history of childhood SaLD to identify the factors associated with the persistence or resolution of these difficulties in children who were not receiving speech-language pathology services. Significant variability was reported across studies, even within specific types of SaLD. However, there was evidence that more persistent deficits were exhibited by children with language problems (particularly those with both receptive and expressive difficulties) compared to children with speech difficulties. Where SaLD persist into adulthood, individuals are at greater risk of having co-morbid mental health conditions and also tend to have poorer vocational outcomes (Rutter & Mawhood, 1991), although outcomes tend to differ depending on the nature of the individual's SaLD (Johnson, Beitchman, & Brownlie, 2010).

Health-related quality-of-life

While the impact of childhood SaLD on specific outcomes such as literacy/educational achievement, behaviour, and self-esteem is widely recognized, there has been limited attention to the impact of SaLD on health-related quality-of-life (HRQoL). HRQoL is generally considered to be part of the broader construct of Quality-of-Life (QoL), which the World Health Organization defines as “individuals’ perception of their position in life in the context of culture and value systems in which they live and in relation to their goals, expectations, standards and concerns” (WHOQOL Group, 1998, p. 551).

HRQoL is defined as the impact that a particular health condition (or intervention) has on an individual (Petersen, 2003). It is seen as encompassing those elements of QoL specifically related to health (Spieth & Harris, 1996). HRQoL is a multidimensional construct, consisting of numerous distinct dimensions or “domains” which commonly include physical, psychological, and social functioning (Spieth & Harris, 1996). The physical domain encompasses areas such as mobility, self-care, exercise, and energy/fatigue; the psychological domain encompasses cognitive functioning, behaviours, and attitudes; and the social domain encompasses social role participation, community engagement, interactions with others, and relationships (Walker & Rosser, 1993).

The relationship between HRQoL and concepts such as overall QoL, life satisfaction, and well-being is debated. Some researchers consider HRQoL to consist solely of a subjective assessment of well-being or life satisfaction (e.g., Tennant & McKenna, 1995). Others argue that it also includes an objective description of the individual’s health status, and that measurement of HRQoL should therefore include both objective and subjective assessments (e.g., Spilker, 1990; Titman, Smith, & Graham, 1997). According to Eiser and Morse (2001, p. 10), “The objective assessment of [HRQoL] focuses on what the individual can do, and is important in defining the degree of health. The subjective assessment of [HRQoL] includes the meaning to the individual”. Items related to objective (health status) aspects of HRQoL tend to focus on symptomatology, functioning, and activity limitations, whereas items related to subjective (well-being) aspects of HRQoL focus on feelings, concerns, and worries about the condition (Eiser & Morse, 2001).

In health research the term quality-of-life is sometimes used instead of health-related quality-of-life for the purposes of brevity. Where QoL has been used in this manner within the studies reviewed here, we have replaced this with HRQoL to aid conceptual clarity.

There currently exists an extraordinarily large number of theoretical models and frameworks of HRQoL. In their systematic review, Taillefer, Dupuis, Roberge, and LeMay (2003) examined 68 HRQoL frameworks and models, and highlighted that this number represented only 60% of those published since 1965. Many of these lacked explicit details about their conceptualization of HRQoL and have had limited use (Taillefer et al., 2003). Nonetheless, the importance of this construct in understanding the impact of health conditions and interventions is clear. HRQoL measures are increasingly being employed in research and service evaluation contexts, and for assisting intervention planning and outcome assessment in clinical settings (Varni, Burwinke, & Lane, 2005a). Use of self-reported HRQoL data ensures that clients’ perspectives are factored into clinical decision-making (Sullivan, 2003). Identification of poor HRQoL can highlight the need for targeted intervention strategies or referral of clients to appropriate professionals (Varni et al., 2005a). Further, longitudinal collection of HRQoL data can be used to inform clinicians of the effectiveness of interventions in improving the lives of their clients (Varni et al., 2005a). Historically, most of the HRQoL research conducted to date has been with adults; however, a growing number of paediatric studies have been undertaken in recent years (Petersen, 2003).

Paediatric studies and measures of HRQoL

There are numerous difficulties inherent in conducting research on the HRQoL of children and adolescents. A first key challenge relates to the poor definition and conceptualization of HRQoL. Understanding of HRQoL in children is still developing (Titman et al., 1997), and conceptualization of this construct for children and young people has been even more variable than for adults (Davis, Waters, Mackinnon, Reddihough, Graham, Mehmet-Radji, et al., 2006). In children, HRQoL should account for things such as the unique experiences and attitudes of children, and changes that occur as part of children’s development (Eiser & Morse 2001).

A second challenge relates to the measurement and particularly the issue of appropriate informants
Quality-of-life of children with speech and language difficulties has child self-report (age 5 years and older) and parent proxy report forms, with age-appropriate versions that differ in content and language complexity. School functioning is only measured in children aged 5 and above. Reliability, validity, and clinical utility have been well established (Varni, Sied, & Kurtin, 2001), and the PedsQL is considered to be the gold standard measure of paediatric HRQoL (Schmitt, Paterno, & Huang, 2010).

The KINDL (Ravens-Sieberer & Bullinger, 1998) is a self-report measure designed for use with children and adolescents of 8–16 years of age. It contains 40 items in four scales: mental; physical; everyday life; and social life. Reliability and validity have been established; however, the KINDL's sensitivity to clinical change has not yet been examined.

The TAPQOL (Fekkes, 2000) is a parent proxy measure designed for use with infants and young children aged 1–5 years. It has 56 items across five scales: motor; autonomy; cognitive; emotional; and social. Reliability and validity have been assessed to be adequate (Davis et al., 2006).

Paediatric studies of HRQoL have mostly concerned children with chronic illnesses and infants in intensive care (Petersen, 2003). These have generally shown that HRQoL is adversely affected, regardless of the condition type. The chronic conditions studied to date have included such things as cancer, asthma, and epilepsy, which tend to be characterized primarily by physical signs and symptoms. In contrast, Sald are characterized by difficulties with social interactions, school achievement, and emotional/behavioural difficulties (Cohen, 2006). Nevertheless, the broader paediatric studies offer insight into issues relevant to SaLD.

Variations in measurement of HRQoL by child self-report compared to parent proxy report has been examined in a comprehensive study of children with chronic illnesses (Varni, Limbers, & Burwinkle, 2007). The sample comprised 2500 children with a wide range of conditions (gastrointestinal conditions, diabetes, cancer, cardiac conditions, cerebral palsy, asthma, obesity, end stage renal disease, psychiatric disorders, and rheumatologic conditions) and 9500 control children without health conditions. For each health condition, children with chronic illness and their parents reported significantly lower HRQoL than controls. However, children's overall HRQoL scores varied according to health condition. For example, children with cerebral palsy were rated as having the lowest overall HRQoL, and children with diabetes and cardiac conditions the highest overall ratings. Differences were also noted at the domain level, with some domains being reported by both children and parents as being unaffected in particular conditions (e.g., physical and social domains for children with diabetes, school functioning for obese children, and the social domain for children with cardiac conditions). While child self and parent proxy reports generally showed reasonable agreement,
there was poor agreement for children with some conditions. For example, parents’ ratings were lower than children’s for children with cerebral palsy, and parent ratings were higher than children’s for those with cardiac conditions and obesity. Across domains, ratings of emotional functioning showed the least agreement, with many parents reporting significantly lower HRQoL in this domain than children.

More recently, Ingerski, Modi, Hood, Pai, Zeller, Piazza-Waggoner, et al. (2010) conducted a study of HRQoL outcomes for children with eight chronic health conditions. Their study utilized secondary data for 589 children and adolescents with various illnesses (inflammatory bowel disease, epilepsy, gastrointestinal disorders, sickle cell disease, cystic fibrosis, type 1 diabetes, obesity, and post-renal transplantation) as well as healthy controls (using population level data). HRQoL was measured using child self and parent proxy report versions of the PedsQL. Children with all condition types had significantly lower HRQoL scores than healthy controls for at least some HRQoL domains. HRQoL ratings varied depending on their illness type, with children in the obesity and gastrointestinal disorder groups having the lowest scores. Overall, scores for social functioning differed significantly across condition type, as did the psychosocial summary score (mean scores from the emotional, social, and school functioning scales). Agreement between child self and parent proxy report was poor: parents reported significantly lower HRQoL in this domain than children.

**HRQoL within speech-language pathology research**

Relatively little research on HRQoL has been conducted within the field of speech-language pathology, possibly due to a traditional focus on impairment-based approaches to assessment, intervention, and outcome measurement (Hesketh & Sage, 1999). In recent years, speech-language pathologists have adopted a more socially-based view of communication. This approach places a greater emphasis on the individual with the communication difficulty (and their families), their personal goals and experiences, ability to communicate functionally within their everyday situations, and the environmental factors that facilitate or impede optimal communication (Duchan, 2001). Within this social-contextual approach speech-language pathologists have become interested in a broader range of intervention outcomes, including social participation and HRQoL (Duchan, 2001).

**Adult studies.** Most of the research undertaken on HRQoL within the field of speech-language pathology has concerned adults rather than children. Adult studies have included individuals with aphasia (e.g., Cruice, Hirsch, Worrall, Holland, & Hickson, 2000; Cruice, Worrall, & Hickson, 2010; Hilaris, Wiggins, Roy, Byng, & Smith, 2003), voice disorders (Baylor, Yorkston, & Edie, 2005; Jones, Carding, & Drinnan, 2006; Krischke, Weigel, Hoppe, Kollner, Klotz, Eysholdt, et al., 2005), laryngectomies (Edie & Doyle, 2004; Gritz, Carmack, de Moor, Coscarelli, Schacherer, Meyers, et al., 1999; Hammerlid, Bjordal, Ahihner-Elmqvist, Boyse, Evensen, Björklund, Aet al., 2001), and fluency disorders (stuttering) (Bramlett, Bothe, & Franic, 2006; Craig, Blungart, & Tran, 2009; Yaruss, 2010). Outcome measures have spanned both generic measures, which are more likely to be standardized, and non-standardized, often condition-specific measures. The Short Form Health Survey 36 (SF-36) (Ware, Snow, Kosinski, & Gandek, 1993), a well-validated, generic measure of HRQoL assessing physical health, mental health, and role functioning, has been used in numerous studies and with a range of clinical populations. The SF-36 has been used with adults with a range of communication disorders, although its use as a self-administered measure with adults with aphasia is problematic as a result of the complex language and inconsistent response format used (Cruice et al., 2000).

Adults with communication disorders have generally been found to have reduced HRQoL across multiple domains. For example, adults who stutter report lower HRQoL scores on the social, emotional, mental health, and vitality scales of the SF-36 than controls who do not stutter (Craig et al. 2009). Similarly, adults with voice disorders show evidence of adverse effects on voice quality, levels of effort/fatigue, social interactions, and psychological functioning (Baylor et al., 2005). Adults with voice problems also report limitations in voice-related activities and participation restrictions relative to adults without these problems (Ma & Yiu, 2001), and activities and participation are closely related to HRQoL in adults (Lawton, Winter, Kleban, & Ruckdeschel, 1999).

For adults with aphasia, there is also evidence of adverse effects on HRQoL, but the evidence is mixed regarding which domains are affected, possibly due to variability in the comparison groups examined. Compared to adults without aphasia who have also experienced stroke or brain injury, adults with aphasia have reported lower HRQoL in the emotional and mental functioning scales on the SF-36 (Cruice et al., 2010) and communication, psychological, social, and total scores on the Stroke and Aphasia Quality of Life Scale (SAQoL) (Manders, Dammeke, Leemans, & Michiels, 2010). However, compared to healthy controls, these adults with aphasia...
showed additional impairments in the SF-36 scales measuring general health, pain, physical functioning, and physical impact on role functioning (Cruice et al., 2010) and the physical and energy scales on the SAQoL (Manders et al., 2010).

Finally, research on young adults with childhood histories of specific language impairment (SLI) has produced mixed findings. One study has reported significant differences compared to peers, in some aspects of HRQoL particularly for speech, mental functioning, usual activities, and distress (Arkilla, Råsänen, Roine, Sintonen, & Vilkman, 2008a). In contrast, Johnson et al. (2010) and Records, Tomblin, and Freese (1992) reported no differences in HRQoL between adults with and without histories of SLI, despite evidence in both studies of continued language impairment in adulthood.

There has also been considerable research examining the HRQoL of people who have had laryngectomies. However, this research has largely focused on comparisons between people receiving different vocal restoration methods and hence findings will not be discussed in this review.

**Paediatric studies.** To date research on HRQoL in paediatric SLP populations has mainly examined children who have voice disorders and hearing impairment/cochlear implant (CI). Measures used with children with voice disorders have tended to be standardized, condition-specific tools. The most widely used is the *Paediatric Voice Outcomes Survey* (PVOS) (Hartnick, 2002), adapted from the adult *Voice Outcomes Survey* (Gliklich, Glovsky, & Montgomery, 1999). These studies show the PVOS has good reliability and validity (Hartnick, 2002; Hartnick, Volk, & Cunningham, 2003), but limited sensitivity for detecting changes over time (Boseley, Cunningham, Volk, & Hartnick, 2006). Studies of children with hearing impairment/cochlear implants have utilized a wide range of generic measures of HRQoL, with some studies only using the communication sub-scales (Lin & Niparko, 2006).

Results for children with voice disorders and those with hearing impairment are similar to those for children with chronic health conditions; both groups show poorer HRQoL than non-affected children. Children with dysphonia had lower HRQoL ratings across all scales (functional, physical, and emotional) of the *Paediatric Voice Handicap Index* (Zur, Cotton, Kelchner, Baker, Weinrich, & Lee, 2007). Similarly, hearing impairment in children is associated with lower HRQoL, particularly in subscales related to behaviour, mental health, family activities, and social/physical impact on role functioning (Wake, Hughes, Collins, & Poulakis, 2004) and social and psychological domains of HRQoL (e.g., assertiveness, making friends, sadness/anxiety; Keilmann, Limberger, & Mann, 2007). However, there was no evidence of reduced HRQoL in the physical domain for these children.

Research on children and adolescents with CI has produced mixed findings. Warner-Czyz, Loy, Roland, Tong, and Tobey (2009) found no difference in HRQoL scores for children with CI compared to controls. Conversely, Huber (2005) reported that, while HRQoL was lower for children with CI, scores were similar to controls for adolescents. In both studies age at implantation and duration of CI use were not related to HRQoL, however Huber (2005) reported that audiological outcomes (ability to hear and comprehend spoken language) were related to HRQoL.

To date, there have been no reviews of the HRQoL of children with SaLD. This paper addresses this gap by systematically reviewing the literature reporting the HRQoL of children with SaLD. Specifically, it aims to examine whether there is any evidence that children with SaLD have poor HRQoL overall and in the physical, psychological, and social domains. Given the findings of previous child and adult studies within speech-language pathology, it is hypothesized that children with SaLD and their parents will report reduced HRQoL for this group, particularly in the social and psychological domains. Findings will directly inform speech-language pathology practice by aiding with the identification of children in need of interventions targeting improved HRQoL.

**Method**


1) child OR adolescent
2) AND (well-being OR personal satisfaction OR “quality of life” OR “health related quality of life” OR “communication related quality of life” OR perception OR QOL OR HRQOL)
3) AND (communication OR speech OR language)
4) AND (problem OR difficulty OR disorder OR delay OR impairment).

First, the strategy used search terms to limit the findings to children and adolescents from birth to 18 years. The medical subject heading (MeSH) terms
and text words/phrases associated with HRQoL were combined with words/phrases used to identify children with SaLD (steps three and four). The phrases “well-being” and “personal satisfaction” were included in the search strategy to avoid missing papers that may have been published before terms such as QoL and HRQoL were widely used (step 2). The authors then examined the reference lists of the included studies to check for any other articles that may have been missed.

A total of 1064 papers were retrieved during electronic database searching. After duplicates were removed 739 unique studies remained. Titles and abstracts were screened by one author (RF) for suitability, according to the following inclusion criterion: the study sample was children or adolescents up to the age of 18 years with identified speech and/or language difficulties, or the parents of these children. This process resulted in 735 papers being excluded, mostly because they did not satisfy criteria number 3 (were not of children with SaLD). Scanning of reference lists yielded an additional three studies, resulting in identification of seven studies suitable for inclusion in the review.

Results

Table I summarizes the participant details, study design, outcome measures, and findings of the seven identified studies of HRQoL in children with SaLD.

<table>
<thead>
<tr>
<th>Study Details</th>
<th>Findings</th>
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<tr>
<td>Arkkila et al. (2009, 2011)</td>
<td>Used four generic, standardized measures of HRQoL (TAPQOL, 16D, 17D, and the PedsQL). The 16D and 17D are generic, multidimensional HRQoL instruments designed for use with adolescents aged 12–15 and children aged 8–11 years, respectively (Arkkila et al., 2009, 2011), adapted from the 15D, a HRQoL instrument used with adults (Sintonen, 2001). They examine HRQoL across 16 (16D) and 17 (17D) “dimensions” of HRQoL, including vitality, mobility, hearing, speech, vision, sleeping, breathing, eating, elimination, appearance, discomfort and symptoms, school and hobbies, friends and relations, mental function, and depression, with the 17D containing an additional dimension on learning (Arkkila et al., 2009; 2011). Both instruments can be completed via interview.</td>
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<tr>
<td>Barr et al. (2007)</td>
<td>Utilized age- and gender-matched controls recruited through a national survey, while Barr et al. (2007) utilized age-matched controls randomly selected in a medical centre clinical practice. In the study by Van Agt et al. (2005), children with language delay were compared to an unmatched population-based cohort without language delay. Studies were conducted in the UK (Markham &amp; Dean, 2006; Markham et al., 2009), US (Barr et al., 2007), the Netherlands (van Agt et al., 2005), Canada (Thomas-Stonell et al., 2010), and Finland (Arkkila et al., 2009; 2011).</td>
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<tr>
<td>van Agt et al. (2005)</td>
<td>Used focus group interviews to discuss the impacts of SaLD on the HRQoL of children and young people. Participants were asked a single broad question and asked to discuss their responses as fully as possible. Interviewers also prompted participants to discuss particular topics (e.g., emotional function, behaviour, hobbies, autonomy, social skills, school, etc.) if these issues had not already been raised by participants. Children and young people were provided with communication support to facilitate their participation in the interview. Interviews were audio-taped with participant consent and transcribed verbatim. In addition to audio recordings, interview content was noted by researchers during the interviews. Thematic charting was then undertaken to code and then group related participant responses. Data analysis was performed in accordance with grounded theory principles and framework analysis. Markham and colleagues did not relate emergent themes to HRQoL domains. Rather, the authors of the current review reported these themes within individual HRQoL domains to enhance comparison of findings across studies.</td>
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Participants

Study participants were children and adolescents, aged birth–18 years, as well as their parents, speech language therapists, and professional carers. Of the seven studies reviewed, one comprised children with language delays (van Agt, Essink-Bot, van der Stege, de Ridder-Sluieter, & de Koning, 2005), two were children and adolescents with specific language impairment (SLI) (Arkkila, Räisänen, Roine, Sintonen, Saar, & Vilkman, 2009; 2011), a further three were children with a range of SaLD sub-types (Markham & Dean, 2006; Markham, van Laar, Gibbard, & Dean, 2009; Thomas-Stonell, Oddson, Robertson, & Rosenbaum, 2010), and one was children with speech disorders secondary to velopharyngeal insufficiency (VPI, inadequate closure of the soft palate during speech) (Barr, Thibeault, Muntz, & De Serres, 2007). Sample sizes ranged from 29 (Markham et al., 2009) to 8877 (van Agt et al., 2005). Most quantitative studies (Arkkila et al., 2009; 2011; Barr et al., 2007; van Agt et al., 2005) utilized a control group. Two studies (Arkkila et al., 2009; 2011) employed age- and gender-matched controls recruited through a national survey, while Barr et al. (2007) utilized age-matched controls randomly selected in a medical centre clinical practice. In the study by Van Agt et al. (2005), children with language delay were compared to an unmatched population-based cohort without language delay. Studies were conducted in the UK (Markham & Dean, 2006; Markham et al., 2009), US (Barr et al., 2007), the Netherlands (van Agt et al., 2005), Canada (Thomas-Stonell et al., 2010), and Finland (Arkkila et al., 2009; 2011).
<table>
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<tr>
<th>Reference</th>
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<th>Design</th>
<th>HRQoL measures</th>
<th>Main findings</th>
<th>Domains of HRQoL measured</th>
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<tr>
<td>van Agt et al. (2005)</td>
<td>Age: 3 years</td>
<td>Quantitative cross-sectional study.</td>
<td>TNO-AZL Pre-school children Quality of Life questionnaire (TAPQOL) completed by parents.</td>
<td>Communication scale of the TAPQQL moderately correlated with language production scores on the Language Screening Instrument (Parent Form) and the Van Wiechen (language assessment). The following scales of the TAPQQL discriminated between children with and without language problems: 1. Communication (control mean = 88.9, SD = 13; language delay mean = 67.2, SD = 19; p &lt; .01); and 2. Social Functioning (control mean = 92.2, SD = 15; language delay mean = 82.3, SD = 24, p &lt; .01)</td>
<td>Psychological/Social</td>
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<tr>
<td>Arkkila et al. (2009)</td>
<td>Age: 12–16 years</td>
<td>Quantitative cross-sectional study.</td>
<td>16D questionnaire completed by adolescents.</td>
<td>Overall, no significant reduction in scores on the 16D for adolescents with SLI compared with controls. However, differences in scores were statistically significant for the mental functioning (lower for adolescents with SLI) and vitality dimensions (lower for controls).</td>
<td>Physical/Psychological/Social</td>
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<tr>
<td>Arkkila et al. (2011)</td>
<td>Age: 8–11 years</td>
<td>Quantitative cross-sectional study.</td>
<td>17D questionnaire completed by children.</td>
<td>Overall, no significant reduction in scores on the 17D for children with SLI compared with controls. However, differences in scores were statistically significant for the speech (lower for children with SLI) and sleep dimensions (lower for controls).</td>
<td>Physical/Psychological/Social</td>
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<tr>
<td>Barr et al. (2007)</td>
<td>Age: 5–17 years</td>
<td>Quantitative cross-sectional study.</td>
<td>Velopharyngeal Insufficiency Quality of Life (VPIQL) and Pediatric Quality of Life Inventory, version 4.0 (PedsQL®) completed by children and parents.</td>
<td>Reduction in scores on the VPIQL and PedsQL® for children with VPI compared with controls. For both measures, differences in scores were statistically significant for all sub-scales (representing various HRQoL domains).</td>
<td>Physical/Psychological/Social</td>
</tr>
<tr>
<td>Markham &amp; Dean (2006)</td>
<td>Age: 11 parents, 12 speech language therapists and 12 professional carers of children aged 0–18 years</td>
<td>Qualitative study utilizing focus group interviews.</td>
<td>Single broad, open ended question asked initially.</td>
<td>Key themes: inclusion, behaviour and reactions of others, education and awareness, friendships and family relations, schooling, the child’s needs, dependence–independence, quality of care, choice, and potential, variability.</td>
<td>Physical/Psychological/Social</td>
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<tr>
<td>Markham et al. (2009)</td>
<td>Age: 6–18 years</td>
<td>Qualitative study utilizing focus group interviews.</td>
<td>Interviews consisted of open-ended questions, prompts and probes.</td>
<td>Key themes: achievement, emotions, independence, individual needs, relationships, relaxation, school, support.</td>
<td>Physical/Psychological/Social</td>
</tr>
<tr>
<td>Thomas-Stonell et al. (2010)</td>
<td>Age: 1–5 years</td>
<td>Quantitative case series study.</td>
<td>PedsQL completed by parents.</td>
<td>Reduction in total scores on the PedsQL for children with SaLD compared with controls. PedsQL psychosocial domain (the social, emotional, and school functioning sub-scales) was most highly correlated with communication ability.</td>
<td>Physical/Psychological/Social</td>
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</table>

*Domains of HRQoL linked with SaLD in italics.*
or self-administered for either self or proxy report (Arkkila et al., 2009; 2011). In the two studies by Arkkila et al. (2009; 2011), questionnaires were self-completed by children and adolescents with SLI where possible, however participants also had assistance from parents to complete the questionnaire if required.

In addition to the generic measures of HRQoL employed, for the study of children with VPI, a condition-specific measure was developed: the Velopharyngeal Insufficiency Quality of Life (VPIQL) (Barr et al., 2007). This assesses children's and parents' perceptions of HRQoL in 43 items across six subscales: speech limitations, situational difficulty, emotional impact, perception by others, activity limitations, and swallowing problems (Barr et al., 2007). Reliability and validity of the VPIQL is yet to be established (Barr et al., 2007). In this study, parents also provided additional information on the impact that their child’s VPI has on them (Barr et al., 2007) by completing a parent version of the VPIQL.

Overall, included studies utilized a mixture of self and proxy report measures. However, only the study by Barr et al. (2007) used reports from both informant types, with both children and parents completing the same two HRQoL measures. This approach allowed for direct comparison of child and parent perspectives.

**Domains of HRQoL measured**

Outcome measures used in the quantitative studies were variable with respect to the domains of HRQoL measured. The study by van Agt et al. (2005) utilized a shortened version of the TAPQOL which included only sub-scales related to communication, social interaction, and emotion/behaviour, and hence measured the effects of language delay on social and psychological rather than physical domains of HRQoL. In the two studies by Arkkila et al. (2009; 2011), all items of the 16D and 17D questionnaires, respectively, were completed by participants, thereby exploring physical, psychological, and social domains of HRQoL. Barr et al. (2007) and Thomas-Stonell et al. (2010) also examined all of these HRQoL domains in their studies (of children with VPI and speech and/or language disorders, respectively), with the all sub-scales of the PedsQL being completed by parents. However, the second measure employed by Barr et al., the VPIQL, related to psychological and social domains of HRQoL only. This focus is not surprising given that the VPIQL is a condition-specific rather than generic HRQoL measure. For both qualitative studies (Markham & Dean, 2006; Markham et al., 2009), participants were asked a single broad question and, hence, themes related to any domains of HRQoL could potentially be identified.

**Relationship between HRQoL and SaLD**

The seven studies set out to examine whether or not there was a relationship between SaLD and HRQoL and to determine what domains of HRQoL were affected. Overall, findings supported a relationship between HRQoL and childhood SaLD. Results of the quantitative studies generally suggested that children with SaLD have lower HRQoL than controls; however, there were mixed findings regarding which domains of HRQoL were affected (as shown in Table I). Barr et al. (2007) reported HRQoL was significantly lower for children with VPI compared to a control group, as measured by the PedsQL (all domains) and VPIQL (social and psychological domains). Conversely, van Agt et al. (2005) reported that HRQoL in children with language delays was only significantly lower than that of controls for sub-scales of the TAPQOL related to social functioning. Arkkila et al. (2011) similarly found the social domain to be the only one in which children with SLI had lower scores than controls, reporting statistically significant reductions for the speech dimension of the 17D. Thomas-Stonell et al. (2010) reported that overall scores were better for children with higher scores on the FOCUS (Focus on the Outcome of Communication Under Six), a measure of communication ability. While they did not detail results for each sub-scale of the PedsQL, the authors also reported that the social, emotional, and school functioning sub-scales were most highly correlated with communication ability (Thomas-Stonell et al., 2010). Last, Arkkila et al. (2009) found the psychological domain to be the only one in which children with SLI scored lower than controls, reporting statistically significant reductions in scores for the mental functioning dimension of the 16D.

In the qualitative studies (Markham & Dean, 2006; Markham et al., 2009) all participant groups (children/adolescents, parents, speech language therapists, and professional carers) discussed the negative impact they perceived SaLD to have on the HRQoL of children and adolescents with SaLD. In these two studies the emergent themes related to all three domains of HRQoL, as well as school functioning, and participants consistently reported that SaLD adversely affected children’s functioning and experiences in these areas.

**Discussion**

**Measurement approaches**

Studies were variable with respect to the type of outcome measures employed, with both standardized/informal and generic/condition-specific measures being utilized. This variation in approaches to HRQoL measurement also occurs in the broader HRQoL literature within speech-language pathology. For
included studies, outcome measures and overall study designs generally reflected the fact that HRQoL issues change over time. Markham et al. (2009) grouped participants by educational level, thus focus groups consisted of children of similar ages, and participants’ ages were taken into consideration during data collection and discussion of emergent themes. The studies by Barr et al. (2007) and Thomas-Stonell et al. (2010) measured HRQoL using the PedsQL, for which different versions are used with different age ranges (Varni et al., 1999). The studies by van Agt et al. (2005) and Arkkila et al. (2009; 2011) all utilized measures designed for use with specific age groups (pre-school aged children, pre-adolescents, and adolescents, respectively). Conversely, Markham and Dean (2006) did not appear to consider the effects of children’s age either in the development of interview questions or during data analysis, despite the fact that participants were proxy respondents for children of widely differing ages (birth to 18 years).

Included studies utilized a mixture of self and proxy report measures; thereby enabling some comparison of these two approaches for the study of HRQoL and SaLD in children. Barr et al. (2007) required children and parents to complete the same two HRQoL measures. Overall, there was a high level of agreement between children and their parents on both questionnaires, except on the emotional functioning scale of the PedsQL, where parents tended to report more difficulties than children. Further conclusions regarding commonalities between reports from child and parent/professional perspectives can be drawn from the studies by Markham and Dean (2006) and Markham et al. (2009), which utilized similar methodologies but different study samples (children with SaLD and parents/professionals in the two studies, respectively). Both were qualitative studies employing grounded theory in which focus group interviews were used to gain insights into participants’ perceptions of the HRQoL of children with SaLD. While there were some differences in the themes which emerged in the two groups in each of the studies, there were also many commonalities. These occurred around communication and social issues such as the importance of awareness of SaLD and supportive communication partners and environments, and problems and rewards associated with peer and family relationships (including difficulties establishing and maintaining these relationships as well as the potentially positive impact of relationships on HRQoL). Commonalities were also observed around emotional issues for children with SaLD, especially feelings of frustration, anger, and anxiety. Finally, both children/adolescents and their parents/professionals highlighted the importance of school-related issues (such as inclusion, learning/achievement) and independence with activities and decision-making on children’s HRQoL.

The dearth of paediatric speech-language pathology studies of HRQoL may in part reflect the difficulties inherent in assessing this construct in children. As previously discussed, there are limitations in using proxy report alone for measuring HRQoL (White-Koning et al., 2007). However, given the difficulties children with SaLD experience with communication, obtaining self-reports on HRQoL, even for older children, is not common. There is a need for measures to be developed that are suitable for use with young children and those with cognitive/language difficulties that enable self-report. Strategies that could be employed for this purpose include the use of symbols, pictures, closed choice yes/no questions, and simplification of content (Sturgess, Rodger, & Ozanne, 2002).

**HRQoL domains**

Studies included in this review provided evidence of poorer HRQoL for children with SaLD compared with children without these difficulties, and this finding was consistent irrespective of the nature of the type of SaLD investigated. The negative relationship between SaLD and HRQoL found in the included studies is consistent with the broader literature on HRQoL in chronic health conditions, which has generally found poorer outcomes compared to healthy children (Varni et al., 2007). Further, findings of reviewed studies suggest that children with SaLD share many similarities with previous studies of both adults and children within other areas of speech-language pathology. As with other conditions, it appears that SaLD affect some HRQoL domains more than others. Most consistently, the included studies reported reduced HRQoL in the social domain, and often also found some problems with psychological functioning. This finding is consistent with the literature on adults with aphasia, dysphonia, and some studies of SaLD in adulthood, as well as research on children with dysphonia and hearing impairment. Given that children with SaLD experience difficulties with understanding others and making themselves understood by communication partners, it is not surprising that they tend to experience lower HRQoL in the social domain. In addition to difficulties with specific communication tasks, problems with social relationships are well-documented in this population (Conti-Ramsden & Botting, 2004).

**Social domain**

The study by Arkkila et al. (2009) was the only paper which did not report significant differences between adolescents with SLI and controls in the social domain. This may be due to the relatively older age of participants (12–16 years). Studies have demonstrated the increasing importance of life
circumstances, aside from communication ability, in determining perceptions of well-being for adults with histories of SaLD (Johnson et al., 2010; Records et al., 1992). Other studies revealed diverse ways in which SaLD may be related to social domains of HRQoL. Both qualitative studies described key themes related to social functioning such as behaviour/reactions of others and friendships/family relations (Markham & Dean, 2006), and relationships (Markham et al., 2009). Peer relationships were found to be particularly central to children’s HRQoL, especially their ability to play and feel comfortable and form friendships (Markham & Dean, 2006; Markham et al., 2009; van Agt et al., 2005). Negative aspects of peer relationships were described in two studies (Barr et al., 2007; Markham et al., 2009), in which children and their parents expressed concerns regarding teasing and bullying which occurred as a result of children’s SaLD. While peer relations were emphasized, children’s relationships with family was also strongly linked with HRQoL (Markham et al., 2009). Some children stressed the importance of the relationship with their parents and identified this relationship as a source of happiness and something which improved their lives.

Psychological domain

Findings were mixed regarding the links between SaLD and the existence and nature of difficulties within the domain of psychological HRQoL. Barr et al. (2007) found that children with VPI reported frequent feelings of anger, and these reports were confirmed by their parents. The qualitative studies by Markham and Dean (2006) and Markham et al. (2009) highlighted the fact that, although some children responded to teasing and communication breakdowns with frustration and anger, others experienced sadness or anxiety. These have been found to be common reactions of children with SaLD to communication breakdowns (McCormack, McLeod, McAllister, & Harrison, 2010). In contrast, Arkkila et al. (2011) did not find any differences between children with SLI and controls for aspects of psychological functioning (feeling scared/tense or sad/unhappy/depressed). van Agt et al. (2005) reported no differences between children with language delays and controls on the Problem Behaviour or Anxiety scales of the Pre-school Children Quality of Life Questionnaire (Fekkes, 2000), and Arkkila et al. (2011) also did not find any differences between children with SLI and controls in terms of psychological functioning. Based on the included studies, findings regarding the psychological domain of HRQoL do not appear to be explained by the type of SaLD, age of participants, or use of self or proxy report.

The mixed findings in terms of psychological functioning in children with SaLD is in contrast with HRQoL studies of children with dysphonia (Zur et al., 2007) and hearing impairment (Keilmann et al., 2007; Wake et al., 2004), which have consistently reported poorer outcomes in this domain of HRQoL, including anxiety and depression. Considering the broader literature examining emotional, behavioural, and psychological outcomes in children with SaLD, there is evidence to suggest higher prevalence anxiety disorders in children and adolescents with SaLD (Beitchman & Brownlie, 2010), although there has been little investigation of the relationship between childhood SaLD and depression. Behaviour problems were also consistently found to be more common in children/adolescents with SaLD, which has been widely reported in the literature (Law et al., 2000). Further research is needed to clarify HRQoL within the psychological domain for children with SaLD, particularly in terms of anxiety/depression.

Physical domain

On the basis of the studies reviewed, it is unclear whether or not physical aspects of HRQoL are typically affected in children and adolescents with SaLD. In the study by van Agt et al. (2005), subscales of the TAPQoL related to physical functioning were excluded. Barr et al. (2007) demonstrated significant reductions across all three domains of HRQoL, including physical functioning. Some of the key emerging themes in the interviews with parents, health/education professionals, and children with SaLD conducted by Markham and Dean (2006) and Markham et al. (2009) could be seen to relate to the domain of physical HRQoL, e.g., life skills, hobbies and play, relaxation. However, Arkkila et al. (2009; 2011) did not find any reduction in the physical domain, and Thomas-Stonell (2010) reported that the physical functioning sub-scale of the PedsQL was less highly correlated with communication competence than the other sub-scales. While these findings suggest that physical aspects of HRQoL may be affected in children with SaLD, the evidence is not compelling or supported by the broader speech-language pathology literature. For example, in studies of adults with aphasia, (e.g., Cruice et al., 2010; Manders et al., 2010), participants only report lower physical functioning when compared to healthy controls rather than non-aphasic adults who are also post-stroke, demonstrating that physical difficulties relate to co-morbid conditions rather than language problems. There would be no reason to expect that SaLD per se would result in lower scores for physical functioning and it is likely that any observed effects are due to co-morbid conditions, as children with SaLD often also have motor difficulties (Visscher, Houwen, Scherder, Moolenaar, & Hartman, 2010). For further clarification of this issue, future studies should include both the measurement of physical functioning as part of their overall assessment of HRQoL, and detail on co-morbid conditions.
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School functioning

While this literature review has focused on three broad domains of HRQoL (social, psychological, and physical), an additional construct which was explored in the included studies was ‘school functioning’. School functioning is often included in pediatric HRQoL measures (Davis et al., 2006) and in the current review, school participation was often affected for school-aged children with SaLD in studies where role functioning was examined. Both children and their parents reported significantly lower HRQoL in relation to school functioning on the PedsQL (Barr et al., 2007). Further, within the qualitative studies, a number of key themes related to school functioning, e.g., inclusion (Markham & Dean, 2006) and achievement (Markham et al., 2009). Both parents and health/education professionals highlighted issues associated with children’s academic difficulties and parents’ decision-making around educational placements for their children (i.e. special or mainstream settings) (Markham & Dean, 2006). Children with SaLD further stressed the importance of being in an accepting and encouraging educational setting which provided a structured and quiet learning environment (Markham et al., 2009). Both at school and more generally, children, parents, and professionals all highlighted the difficulties encountered by children with SaLD in being able to complete tasks independently and make their own decisions (Markham & Dean, 2006). Where children were able to achieve these things, they reported great satisfaction in doing so (Markham et al., 2009). Neither of the studies by Arkkila et al. (2009; 2011) found a reduction in scores on the 16D and 17D, respectively, (2009; 2011) found a reduction in scores on the 16D and 17D, respectively, in mediating or moderating the relationship between these difficulties and HRQoL. Studies conducted with children with voice problems, and adults with childhood diagnosis of SLI, highlight some possible mediating factors worthy of further investigation, e.g., severity of the communication difficulty (Zur et al., 2007) and verbal IQ (Arkilla, Räsänen, Roine, & Vilkman, 2008b). Studies of children’s experiences of having SaLD also highlight child, family, and environmental factors which may relate to HRQoL in this population. When examining the experiences of pre-school aged children with speech impairments, McCormack et al. (2010) reported that a persistent temperament and support from parents and siblings may help minimize the negative impact of SaLD on children. In addition, examination of the effects of co-morbid difficulties (e.g., motor, literacy problems) on the HRQoL of children with SaLD would also be valuable, as children with co-morbidities have also generally been excluded from relevant studies. Further, no research has examined the effects of SaLD sub-type (e.g., speech or expressive and/or receptive difficulties only, combined speech/language difficulties). This area warrants investigation given that existing research suggests that children from these various sub-groups have different experiences. For example, children with language difficulties tend to experience more problems related to schooling than those with speech difficulties (Law et al., 2000). There is also a need for longitudinal studies investigating HRQoL, as these may be most informative when considering causal pathways and understanding how HRQoL changes with age. Further research is required to

Limitations of this review

The major limitation of this literature review was that the specific focus on SaLD meant that the number of studies identified was very limited. Nevertheless, given the increasing interest in the experiences and HRQoL of children with SaLD it was felt that a review of the existing literature was warranted. A further limitation was the diversity of study samples, methodologies, and outcome measures utilized, making direct comparisons difficult. However, broad similarities and differences were able to be identified, providing a basis for recommendations regarding further research in the area.

Suggestions for further research

Given existing gaps in the literature, there are numerous issues related to the HRQoL of children with SaLD which warrant exploration. While studies have examined child and family characteristics related to the existence of SaLD (e.g., Fox, Dodd, & Howard, 2002; Harel, Greenstein, Kramer, Yifat, Samuel, Nevo, et al., 1996; Reilly, Wake, Bavin, Prior, Williams, Bretherton, et al., 2007), there has been no investigation of the role these factors may play in mediating or moderating the relationship between these difficulties and HRQoL. Studies conducted with children with voice problems, and adults with childhood diagnosis of SLI, highlight some possible mediating factors worthy of further investigation, e.g., severity of the communication difficulty (Zur et al., 2007) and verbal IQ (Arkilla, Räsänen, Roine, & Vilkman, 2008b). Studies of children’s experiences of having SaLD also highlight child, family, and environmental factors which may relate to HRQoL in this population. When examining the experiences of pre-school aged children with speech impairments, McCormack et al. (2010) reported that a persistent temperament and support from parents and siblings may help minimize the negative impact of SaLD on children. In addition, examination of the effects of co-morbid difficulties (e.g., motor, literacy problems) on the HRQoL of children with SaLD would also be valuable, as children with co-morbidities have also generally been excluded from relevant studies. Further, no research has examined the effects of SaLD sub-type (e.g., speech or expressive and/or receptive difficulties only, combined speech/language difficulties). This area warrants investigation given that existing research suggests that children from these various sub-groups have different experiences. For example, children with language difficulties tend to experience more problems related to schooling than those with speech difficulties (Law et al., 2000). There is also a need for longitudinal studies investigating HRQoL, as these may be most informative when considering causal pathways and understanding how HRQoL changes with age. Further research is required to
build further consensus around the specific domains of HRQoL affected in children with SaLD, as studies provide inconclusive findings on this issue to date. Research in these areas will aid the identification of children at high risk of reduced HRQoL secondary to their SaLD and guide the development of appropriate interventions for these children.

Conclusion
SaLD are a high prevalence condition in childhood and it is well established that a range of adverse outcomes are associated with these difficulties. The HRQoL outcomes for children with SaLD are less well understood, and hence the current review provides a summary of findings from existing studies on this topic. This review revealed only seven studies that have investigated HRQoL in children with SaLD. All found that children with SaLD have significantly reduced HRQoL compared to children without HRQoL. In particular, children with SaLD appear to have difficulties with social and emotional domains of HRQoL as well as school functioning. Because so few studies have been conducted in this area and there has been considerable variation in study samples and methodologies, further research is needed. In order to direct intervention and promote positive outcomes for children with SaLD it is necessary to improve understanding of the HRQoL domains affected in this population, as well as the effects of SaLD sub-type/severity and co-morbid difficulties on HRQoL outcomes. In addition, longitudinal studies and an examination of the effect of child and family factors in mediating or moderating the relationship between SaLD and HRQoL are also required.

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