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Review

Health-related quality of life, service utilization and costs of low language: A systematic review

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Abstract

Background: Low language (LL) is a common childhood condition affecting 7–17% of children. It is associated with life-long adverse outcomes and can affect various aspects of a child's life. However, the literature on its impact on health-related quality of life (HRQoL), service use and costs are limited. To date, there has been no systematic review of the overall economic burden of LL. A systematic review regarding the economic burden of LL is important for clinical, educational, policy decision-making and theoretical aspects. We adopted the term 'low language' to refer to children whose language performance falls below well-recognized cut-points regardless of known or unknown aetiology.
Aims: To review the literature systematically on how LL is associated with HRQoL, service utilization and costs.
Methods & Procedures: A systematic search was conducted across various databases, including MEDLINE, Embase, PsycINFO, CINAHL, up to July 2017. Data on study design, population and outcomes were extracted and screened by two pairs of reviewers with the revision of other experts in the panel on any discrepancies. The Effective Public Health Practice Project tool was used to assess the risk of bias of the included studies. The findings of the included studies were summarized in a narrative synthesis.
Outcomes & Results: We identified 22 relevant articles, of which 12 reported HRQoL and 11 reported service utilization and costs associated with LL. Preference-based instruments, which include the relative importance attached to different aspects of HRQoL, were less employed in the literature. Most studies found poorer HRQoL in children with LL compared with their peers. About half the families having children with LL did not actively seek professional help, and many families felt they did not receive sufficient services when needed. Healthcare costs associated with LL were substantial. Non-healthcare costs were largely unexplored.
Conclusions & Implications: LL was associated with reduced children's HRQoL, higher service use and costs. Under-servicing was evident in children with LL. LL also imposed large costs on the healthcare system. Further research is required to examine (1) the overall HRQoL of children with LL, in particular studies using and testing the performance of preference-based instruments; and (2) the service use and costs specific to LL, especially non-healthcare costs.

Keywords: health-related quality of life (HRQoL), children, low language, service use and cost.

What this paper adds
What is already known on the subject
LL is common and has significant adverse long-term outcomes. Little is known about how it impacts HRQoL, service utilization and costs.

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What this paper adds to existing knowledge
The paper provides a systematic review of the literature on the economic burdens of LL, including HRQoL, service utilization and costs.

What are the potential or actual clinical implications of this work?
LL has substantial healthcare costs. Interventions to improve the condition at an early stage may help mitigate its economic burden. There are still gaps in the literature for further research on the association between LL and the child's overall HRQoL, service utilization and costs, especially research using preference-based instruments to capture HRQoL and research that examines costs of LL to the education and employment sectors.

Introduction

The ability to communicate verbally is fundamental to an individual's development and well-being (McCormack *et al.* 2018). Most children acquire language skills quite easily, but many do not have a typical language developmental pathway. Low language (LL) is a common childhood condition, affecting 7–17% of children and represents one of the largest disability groups in preschool children (Law *et al.* 2000, Bishop *et al.* 2017). LL, which is characterized by late achievement of developmental communication milestones (such as vocabulary, sentence formulation and comprehension) (Law *et al.* 2000), can be detected in children as young as 2 years of age and may persist into late childhood (Rice *et al.* 2008) and adolescence (Law *et al.* 1998). In this paper we consistently used the term 'low language', which refers to children whose language performance falls below well-recognized cut-points (1.25 SD from the mean) regardless of known or unknown aetiology as not all the children in the studies from which our data are drawn will have had a diagnosis of language disorder (e.g., when there is a known biomedical aetiology likely to impact on language development) or developmental language disorder (e.g., language disorder not associated with a known biomedical aetiology) (Bishop *et al.* 2017).

LL imposes a heavy burden on individuals, families, communities and countries (Ruben 2000, Law *et al.* 2009). There are negative associations between LL and various outcomes including social skills and social cognition, problem-solving, literacy, education achievement, emotional problems, behaviour and self-esteem (Charman *et al.* 2015) as well as mental health and socioeconomic outcome in adulthood (Law *et al.* 2009). LL involves substantial treatment costs and impacts on individual's productivity due to limited employment opportunities (Law *et al.* 2009). However, literature regarding the costs and service utilization of LL is scarce. No published systematic review on the costs and service use of LL is available. Of the few studies that have explored this topic, substantial costs were associated with LL, and these varied by child age (Sciberras *et al.* 2015).

Research on health-related quality of life (HRQoL) of children with LL is also limited. HRQoL, a multidimensional construct encompassing physical, mental and social facets of life (Bullinger 2002), is measured by generic and disease-specific HRQoL instruments. Generic instruments have the advantage of being applicable to a wide range of populations and conditions, and therefore may provide opportunities for comparison across populations (Brazier *et al.* 1999). Specific instruments, which focus on one particular disease or health condition, may be more sensitive to the specific condition and more suitable for use within particular patient groups or populations (Brazier *et al.* 1999, Sung *et al.* 2010). Within the generic instruments, preference-based instruments (e.g., Health Utility Index 3 (HUI3)), which incorporate the relative importance attached to different aspects (domains) of HRQoL, are preferred by policy-makers because they allow for the calculation of quality adjusted life years (QALYs), which is needed in the common technique to assess resource allocation in healthcare (cost-utility analysis) (Sung *et al.* 2010). Conversely, non-preference-based instruments do not incorporate individual's relative importance attached to different aspects of HRQoL.

A previous systematic review in 2012 found that the burden of LL on children's overall HRQoL was variable, with half the studies not finding any association between LL and HRQoL (Feeney *et al.* 2012). More recently, in examining the existing approaches of measuring HRQoL in children with speech and language problems, Gomersall *et al.* (2015) found that generic instruments are more widely used but the association between HRQoL and LL had not been explored. This systematic review identified 19 studies, which is almost three times the number found in the 2012 systematic review. The review indicated that there has been more emerging research regarding HRQoL in children with LL over the past several years. Therefore, an update on the literature about HRQoL in children with LL is necessary.

A systematic review regarding HRQoL, service utilization and costs (we refer to all three areas as economic burden hereafter) associated with LL is important for clinical, educational, policy decision-making and theoretical aspects. This knowledge could improve

decision-making on efficient resource allocation and, in turn, enhance HRQoL of children with LL and improve societal productivity. We aimed to review systematically the available literature to assess the economic burden of LL.

Methods

This review adhered to the guidelines in the PRISMA statement 2009 (Moher *et al.* 2009). The original protocol of the review was registered on PROSPERO (Le *et al.* 2017a).

Identification of studies

Extensive searches of the literature were conducted to identify studies examining the economic burden of LL. We were interested in exploring literature over the past 15 years so that the review reflected more recent healthcare experiences. Therefore, the searches performed in this systematic review applied to literature published from January 2002 to July 2017. Owing to limited capacity for language translation, searches were restricted to English publications. The searches were conducted in two stages. Initial searches for existing reviews of economic burden associated with LL were conducted in the Cochrane Controlled Trial Register, DARE, MEDLINE and PsychInfo. The search strategy was developed by the primary reviewer (H.L.) in consultation with the review team and an expert librarian (see appendix A). Searches from this first stage identified two recent published reviews relating to HRQoL of children with LL (Gomersall *et al.* 2015, Feeney *et al.* 2012) in which all included publications were published after 2005. We did not identify any systematic review on costs or service use associated with LL.

Next, searches for the economic burden of LL were conducted in the following electronic bibliographic databases: MEDLINE (via EBSCOHOST database); The Cochrane Library (Wiley); HTA and NHSEED (CRD York); Web of Science Core Collection; Econlit (via EBSCOHOST database); Embase; PsycINFO (via EBSCOHOST database); and CINAHL (via EBSCOhost database). Studies included in the previous reviews that did not appear in the search results were added if they met inclusion criteria. The search terms used for these searches included a broad variety of terms for children, language problems, and HRQoL or service use or costs (see Tables A1 and A2 in Appendix).

Inclusion criteria

Quantitative research studies published in English in peer-reviewed journals and meeting the criteria set out below were included.

Population

Children and adolescents (age ≤ 18 years) with LL (no restriction on definition). LL needed to be the primary health condition of participants, with no co-occurring health diagnoses such as Asperger spectrum disorder, attention deficit hyperactivity disorder (ADHD) or cerebral palsy.

Study types

There was no restriction on the type of study included in the review as long as relevant data were related to our research aims. Therefore, randomized control trial (RCT), economic evaluation and observational studies were eligible for this review.

Comparison group

There was no specific restriction on whether or not the included study had a comparison group of children without LL.

Outcomes

Studies that reported HRQoL/domains of HRQoL or use of services (health or education services) or costs associated with LL from primary data were eligible for inclusion. Where multiple studies reported outcomes (either HRQoL or service use/costs) from the same data, only one (the most recently published) was presented in the final synthesis.

Study selection and data extraction

Data from the searches were extracted into Rayyan QCRI website (Ouzzani *et al.* 2016) by the primary author (H.L.). Extracted research studies were then screened through a two-stage process taking into account the above inclusion criteria. Pilot screening was conducted to test agreement on inclusion/exclusion among four reviewers (H.L., L.L., P.N., S.B.). A random sample of 50 extracted titles and abstracts were screened until 90% agreement on inclusion/exclusion was reached. After the pilot screening, titles and abstracts of articles were screened independently by two authors. Full-text articles included after title and abstract screening were then retrieved and reviewed in full by the same two authors. Consistent rules for pilot screening were applied to title and abstract and full text screening. Any discrepancies were discussed extensively within the review team and decisions were made by consensus.

Quality assessment

The Effective Public Health Practice Project (EPHPP) tool (http://www.ephpp.ca/tools) was used for quality

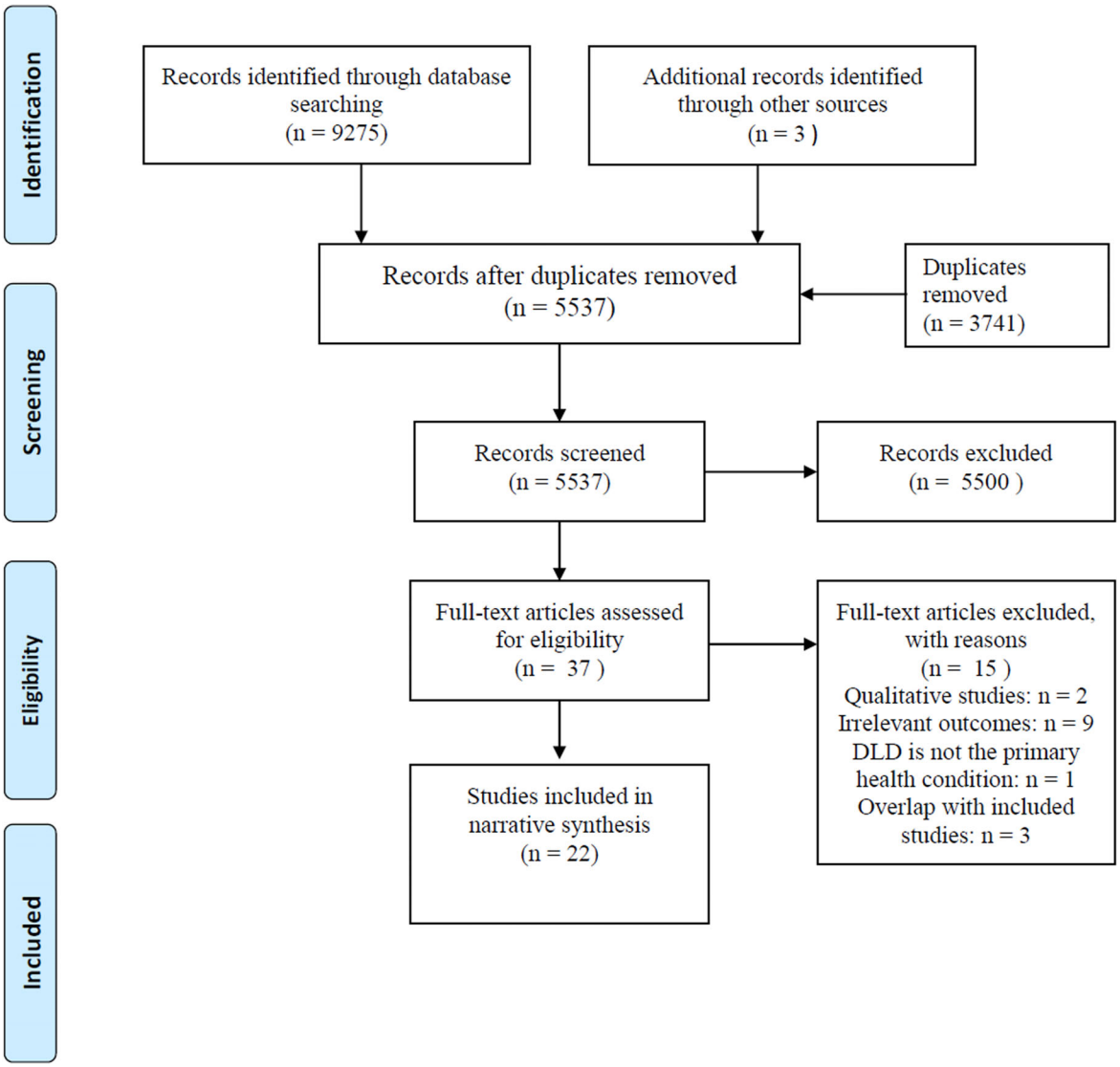

Figure 1. PRISMA flow diagram. [Colour figure can be viewed at wileyonlinelibrary.com]

assessment of the included studies because it covers a broad range of quantitative study types (Armijo-Olivo *et al.* 2012). The tool has been reported to have content and construct validity (e.g., kappa statistic ranged from 0.61 to 0.74) (Thomas *et al.* 2004). It comprises six components including selection bias, study design, confounders, blinding, data collection, and withdrawals and drop-outs. Each component scored as strong, moderate or weak. A global rating score was produced by combining these six components.

Reporting cost associated with low language (LL)

For the convenience of comparing service costs of LL across studies, costs reported were converted to 2017 Australian dollars (A$) using the Australian consumer price inflator (Australian Institute of Health and Welfare (AIHW) 2017). Costs of international studies, reported in currency other than A$, were converted to 2017 A$ using an online cost-converter tool based on the purchasing power parity adapted from the International Monetary Fund (IMF) (http://eppi.ioe.ac.uk/costconversion).

Data synthesis

We narratively synthesized HRQoL, as well as service use and costs associated with LL because the data were heterogeneous, thus precluding a meta-analysis.

Results

The search strategy identified 9278 articles from which 22 articles were included in the final synthesis (figure 1). Among the included articles, 11 reported HRQoL in

children with LL (McKean *et al.* 2017, Nicola and Watter 2015, Thomas-Stonell *et al.* 2010, Van Agt *et al.* 2005, 2010, Wake *et al.* 2013, Arkkila *et al.* 2009, Feeney *et al.* 2017, Flapper and Schoemaker 2013, Hubert-Dibon *et al.* 2016); 10 reported service utilization and/or costs associated with LL (Mazer *et al.* 2017, Nasuuna *et al.* 2016, Skeat *et al.* 2011, 2014, Sciberras *et al.* 2015, Boyle *et al.* 2009, Gibbard *et al.* 2004, Law *et al.* 2006, Le *et al.* 2017b, Cronin *et al.* 2017); and one reported both costs and HRQoL (Wake *et al.* 2015).

Study characteristics

Study participants included children and adolescents aged 0–18 years and their parents. Most studies ($n = 19$) were cross-sectional, and six studies were RCTs (Wake *et al.* 2013, 2015, Van Agt *et al.* 2005, 2010, Boyle *et al.* 2009, Gibbard *et al.* 2004). Sample size ranged from 22 (Gibbard *et al.* 2004) to 24,678 (Nasuuna *et al.* 2016). There was large variation in LL definitions in the included studies: seven studies used the Clinical Evaluation of Language Fundamentals (CELF) (McKean *et al.* 2017, Wake *et al.* 2013, 2015, Skeat *et al.* 2011, 2014, Boyle *et al.* 2009, Le *et al.* 2017b), four studies used the International Classification of Diseases (Thomas-Stonell *et al.* 2010, Arkkila *et al.* 2009, 2011, Hubert-Dibon *et al.* 2016), and three studies used parent report of concerns of LL and/or clinically diagnosed by expert panel (Van Agt *et al.* 2005, 2010, Nasuuna *et al.* 2016) (table 1). Most study outcomes (HRQoL, service use and costs) were parent-proxy reported. Studies were based in and collected data from Australia ($n = 11$), Europe ($n = 9$) and Canada ($n = 2$). Further information is presented in tables 1 and 3.

Health-related quality of life (HRQoL) in children with low language (LL)

The 12 articles reporting HRQoL in children with LL varied in terms of the HRQoL instrument used and whether HRQoL ratings were provided by children themselves or by parent or teacher as proxy (table 1).

Measures

Among the studies reporting HRQoL in children with LL, eight employed generic non-preference-based instruments (with PedsQL being the most commonly used) (McKean *et al.* 2017, Nicola and Watter 2015, Thomas-Stonell *et al.* 2010, Feeney *et al.* 2017, Van Agt *et al.* 2005, 2010, Flapper and Schoemaker 2013, Hubert-Dibon *et al.* 2016); two used preference-based instruments (Arkkila *et al.* 2009, 2011); and two (Wake *et al.* 2013, 2015) used both preference-based (HUI3) and non-preference based instruments (PedsQL). All these instruments are validated for use with children and adolescents. Further details of HRQoL instruments are presented in table 2.

Raters

The majority of studies included parent or teacher-proxy-reported data (McKean *et al.* 2017, Thomas-Stonell *et al.* 2010, Van Agt *et al.* 2005, 2010, Wake *et al.* 2013, 2015, Feeney *et al.* 2017, Hubert-Dibon *et al.* 2016). Two studies (Arkkila *et al.* 2009, 2011) included child self-reported data and one study (Nicola and Watter 2015) included both child self-reported and parent-reported data. The latter found consistencies between child self-report and parent report on physical, emotional and school functioning but children rated themselves significantly better than their parents in social functioning (Nicola and Watter 2015).

Association between LL and children's overall HRQoL

Seven studies (six non-matched control and one matched control) found poorer overall HRQoL/most HRQoL domains in children (aged 4–18 years) with LL compared with their peers with typical language (McKean *et al.* 2017, Van Agt *et al.* 2005, 2010, Hubert-Dibon *et al.* 2016, Feeney *et al.* 2017, Thomas-Stonell *et al.* 2010, Flapper and Schoemaker 2013). Of these seven studies, only two reported medium (Cohen's $d > 0.5$) (Flapper and Schoemaker 2013) to large ($d > 0.8$) (Van Agt *et al.* 2010) effect size. Two matched-control studies (Arkkila *et al.* 2009, 2011) did not find an association between LL and overall HRQoL in adolescents although there was association between LL and impairment in some of HRQoL domains (e.g., mental function or sleep). Among three studies that did not include a control group (children with typical language), two (Wake *et al.* 2013, 2015) reported overall HRQoL of children with LL that were similar to the healthy population mean (Varni *et al.* 2003) and one (Nicola and Watter 2015) reported HRQoL of children with severe LL that fell < 1 SD below the population mean.

Although most studies found an impact of LL on children's social functioning, there was variation in this and other HRQoL domains across studies due to the use of different HRQoL instruments, the severity of the condition, and the different ages of children in the studies. For example, compared with children with typical language, children with LL had impaired communication, liveliness and social functioning (Van Agt *et al.* 2005), mental function and vitality (Arkkila *et al.* 2009)

Table 1. Included studies of HRQoL in children with low language

Study	Population	Study design	Study setting	Low language definition	HRQoL instrument used	Rater	Main findings
Arkkila *et al.* (2009)	$n = 302$ (67 with childhood LL) Age: 12–16 years	Quantitative matched case control	Helsinki, Finland	Clinically diagnosed using the International Classification of Diseases—10th Revision	16D	Self-report	No significant difference in overall HRQoL between adolescents with LL and adolescents with typical language Compared with adolescents with typical language, adolescents with LL experienced lower mental function ($p = 0.001$) and vitality ($p = 0.003$) Among children with LL, low vitality was associated with low verbal IQ in childhood ($r = -0.43$, $p = 0.004$)
Arkkila *et al.* (2011)	$n = 308$ (55 with childhood LL) Aged 8–11 years	Quantitative matched case control	Helsinki, Finland	Clinically diagnosed using the International Classification of Diseases—10th Revision	17D	Self-report	No significant difference in overall HRQoL between adolescents with and without LL Compared with adolescents with typical language, adolescent with LL experienced lower HRQoL in sleep ($p = 0.001$) and speech ($p < 0.001$) domains Feelings of distress domain was correlated with low verbal IQ ($p = 0.008$, $r = 0.4$)

Continued

Table 1. Continued

Study	Population	Study design	Study setting	Low language definition	HRQoL instrument used	Rater	Main findings
Hubert-Dibon *et al.* (2016)	$n = 134$ (67 with LL) Age 8–18 years	Quantitative prospective controlled study	Nantes University Hospital, France	Clinically diagnosed using the International Classification of Diseases—10th Revision	KIDSCREEN-27	Parent proxy report	Overall HRQoL scores were not reported Compared with children with typical language, mean HRQoL scores in children with LL were significantly lower for physical ($p = 0.03$), psychological well-being ($p = 0.04$), autonomy and parent relation ($p = 0.005$), social support ($p = 0.004$), and school environment ($p = 0.01$)
Feeney *et al.* (2017)	Wave 3: $n = 4384$ (1381 with LL) (PEDS receptive and expressive language) or 617 (PPVT-III) Wave 4: $n = 4163$ (1143 with LL) (PEDS receptive and expressive language) or 674 (PPVT-III) Wave 5: $n =$ 4014 (556 with LL on PPVT-III) Age 4–9 years	Quantitative longitudinal study	Longitudinal Study of Australian Children, Australia	The Peabody Picture Vocabulary Test—III and parent-reported 10-item PEDS	PedsQL 4.0	Parent proxy report	Parent concern about child's language problem was negatively associated with all HRQoL domains ($p < 0.05$) Child's general health, maternal mental health, parental warmth and primary caregiver's engagement in the labour force were positively associated with all HRQoL domains ($p < 0.05$)
Flapper and Schoemaker (2013)	$n = 65$ (21 with LL) Age: 5–8 years	Quantitative matched-control study	The Northern part of the Netherlands	Language test scores for auditory processing, grammar or lexical–semantic skills < 1.25 (SD) below the norm	TNO-AZL Child Quality of life (TACQoL)	Both parent and teacher report	Overall HRQoL scores were not reported Compared with children with typical language, children with LL had significantly lower mean TACQOL scores in the autonomy, cognitive, social and positive moods domains ($p < 0.05$).

Continued

Table 1. Continued

Study	Population	Study design	Study setting	Low language definition	HRQoL instrument used	Rater	Main findings
McKean *et al.* (2017)	$n = 1204$ (227 with LL) Age: 7 years	Quantitative cohort study	Victoria, Australia	Receptive or expressive standard score on the CELF-4 ≤ 1.25 SD below the population means	PedsQL	Parent proxy report	Overall HRQoL scores were not reported Children with LL were experiencing limitations (scores ≥ 1 SD below the sample mean) at more than twice the frequency reported in their peers with typical language Compared with children with typical language, children with LL were more likely to have limitations in HRQoL across all domains including physical health, emotional, social and school functioning ($p < 0.05$)
Nicola and Watter (2015)	$n = 43$ with LL Age: 5–18 years	Quantitative cohort study	Queensland, Victoria, Australia	Scores of general language on a psychometric test < 1.5 SD; (2) overall standard score IQ > 70 performing better on the non-verbal test of IQ compared with language ability; and (3) have a confirmed failure of normal language development in the absence of other possibilities	PedsQL4.0	Both child self-report and parent proxy	HRQoL of children with severe LL that fell < 1 SD below the population mean No significant differences between child self-report and parent proxy report on all scales except social functioning, where children scored themselves significantly better than parents did ($p = 0.014$)

Continued

Table 1. Continued

Study	Population	Study design	Study setting	Low language definition	HRQoL instrument used	Rater	Main findings
Thomas-Stonell (2010)	$n = 165$ with LL Age: 5 years 6 months	Quantitative case series study	Ontario, Newfoundland and Labrador, and Nova Scotia, Canada	The International Classification of Diseases—9th Revision	PedsQL	Parent proxy report	Children with better communication skills had higher PedsQL total scores ($r = 0.466$, $p = 0.029$) PedsQL psychosocial domain was highly correlated with communication ability ($r = 0.518$, $p = 0.013$)
Van Agt (2005)	$n = 8877$ (LL: parent report: 252; specialist's judgement: 131; panel: 316) Age: 3 years	RCT	6 regions in the Netherland	Both parent-reported and clinically diagnosed by expert panel	6 domains TNO-AZL Pre-school Quality of life questionnaire (TAPQoL)	Parent proxy report	Overall HRQoL scores were not reported For both parent-reported and expert panel assessed LL groups, children with LL had significantly lower HRQoL on the communication and social functioning ($p < 0.01$) than children with typical language
Van Agt (2010)	$n = 6051$ (377 with LL) Aged: 8 years	RCT	6 regions in the Netherland	Parent reported	CHQ-PF28	Parent proxy report	Compared with children with typical language, children with LL had significantly lower scores in general behaviour ($p < 0.001$), self-esteem ($p < 0.001$), parental impact—emotional ($p < 0.001$), parental impact—time ($p < 0.001$), mental health ($p = 0.003$) and psychosocial summary score ($p < 0.001$) There were significant differences in the psychosocial summary score by gender and parental education but not by socioeconomic neighbourhood, region and foreign language in the family

Continued

Table 1. Continued

Study	Population	Study design	Study setting	Low language definition	HRQoL instrument used	Rater	Main findings
Wake *et al.* (2013)	$n = 200$ with LL Age: 4–5 years	RCT	Melbourne, Australia	Expressive and/or receptive language scores ≤ 1.25 SD below the normative mean of 100 on the CELF-P2	HUI3 PedsQL4.0	Parent proxy report	Association between LL and HRQoL was not explored HRQoL scores in children with LL were similar to the healthy population mean, measured by either the HUI3 or the PedsQL at child ages 4 or 5 year.
Wake *et al.* (2015)	$n = 200$ with LL Age: 5–6 years	RCT	Melbourne, Australia	Expressive and/or receptive language scores ≤ 1.25 SD below the normative mean of 100 on the CELF-P2	HUI3 PedsQL4.0	Parent proxy report	Association between LL and HRQoL was not explored HRQoL scores in children with LL were similar to the healthy population mean, measured by either the HUI3 or the PedsQL at child aged 6 years

Notes: We have consistently used the term 'low language', which is not necessarily the same as the original terms the authors of these included studies used in their studies.
CELF-4, Clinical Evaluation of Language Fundamentals—Fourth Edition; CELF-P2, Clinical Evaluation of Language Fundamentals—Preschool Edition; CHQ-PF28, Child Health Questionnaire—Parent Form; LL, low language; HRQoL, health-related quality of life; HUI3, Health Utility Index Mark 3; IQ, Intelligence Quotient; PEDS, parents' evaluation of developmental status; PedsQL, Pediatric Quality of Life Inventory; PPVT, Peabody Picture Vocabulary Test; RCT, randomized controlled trial; SD, standard deviation; TACQOL, TNO-AZL Children Quality of Life; TAPQOL, TNO-AZL Pre-school Children Quality of Life Questionnaire.

Table 2. Details of HRQoL instruments used in the included studies

HRQoL instruments used	Studies using the HRQoL instruments	HRQoL domains measured
Non preference-based instruments		
Child Health Questionnaire—Parent Form (CHQ-PF28)	Van Agt (2010)	Change in health, physical functioning, role functioning—behaviour, role functioning—physical, bodily pain, general behaviour, mental health, self-esteem, general health perception, parental impact emotional, parental impact time, family activities, family cohesion
KIDSCREEN-27	Hubert-Dibon *et al.* (2016)	Physical well-being, psychological well-being, autonomy and parent relation, peers and social support, and school environment
Pediatric Quality of Life Inventory (PedsQL)	McKean *et al.* (2017), Nicola and Watter (2015), Thomas-Stonell (2010), Wake *et al.* (2013, 2015), Feeney *et al.* (2017)	Physical, emotional, social and school functioning
TNO-AZL Children Quality of Life (TACQOL)	Flapper and Schoemaker (2013)	Physical-functioning, motor-functioning, autonomic-functioning, cognitive-functioning, social-functioning, positive moods, negative moods
TNO-AZL Pre-school Children Quality of Life Questionnaire (TAPQOL)	Van Agt (2005)	Communication, social functioning, anxiety, positive mood, problem behaviour and liveliness
Preference-based instruments		
Health Utility Index 3 (HUI3)	Wake *et al.* (2013, 2015)	Vision, hearing, speech, emotion, cognition, ambulation, dexterity and pain
16D	Arkkila *et al.* (2009)	Vitality, breathing, vision, distress, hearing, sleeping, eating, discomfort/symptoms, speech, appearance, school and hobbies, mobility, friends and relations, mental function, elimination, depression
17D	Arkkila *et al.* (2011)	Vitality, breathing, vision, distress, hearing, sleeping, eating, discomfort/symptoms, speech, appearance, school and hobbies, mobility, friends and relations, learning and memory, depression, concentration, excretion

as well as sleep and speech domains (Arkkila *et al.* 2011). Among six studies using the Pediatric Quality of Life Index (PedsQL), one (McKean *et al.* 2017) reported that LL was associated with all HRQoL domains including physical, social, emotional and school functioning in children younger than 9 years. Another study (Nicola and Watter 2015) reported that children with severe LL perceived to be at risk of impaired social and physical functioning, experienced overall HRQoL scores below the healthy population mean.

Variation was also found regarding factors affecting the association between LL and HRQoL. For instance, gender and parental education were found to impact psychosocial function, but socioeconomic neighbourhood, region and foreign language in the family did not affect this relationship (Van Agt *et al.* 2010). Children's general health, maternal mental health, parental warmth and caregiver's engagement in the labour force were also positively associated with physical, social, emotional and school function (Feeney *et al.* 2017).

Service utilization and costs of low language (LL)

Six studies (Boyle *et al.* 2009, Cronin *et al.* 2017, Gibbard *et al.* 2004, Law *et al.* 2006, Wake *et al.* 2015, Sciberras *et al.* 2015) reported costs, two studies (Mazer *et al.* 2017, Nasuuna *et al.* 2016) reported service utilization, and two studies reported both service utilization and costs associated with LL (Skeat *et al.* 2011, Le *et al.* 2017b) (table 3).

Service utilization

Patterns of service utilization varied by child age. For example, in Australia, up to 21% of families of children with and without LL accessed any health services for speech and language concerns for 4–9-year-old children, with the use of services decreasing with age (Le *et al.* 2017b). Among children with LL, up to 63% were assessed by a speech–language therapist between 4 and 9 years (Le *et al.* 2017b). Compared with children with

typical language, children with LL were more likely to access health services in general as well as speech–language therapy (SLT) services, in particular at a later age (8–9 years) (Le *et al.* 2017b).

In terms of over- or under-servicing for children with LL, a Canadian study reported about 48% (29/60) of preschool children with LL were not receiving services at school (kindergarten) despite needing them and 25% (15/60) of children with LL were not receiving health services outside of school as needed (Mazer *et al.* 2017). Similarly, in an Australian community cohort, only 45% of parents of preschool children with LL reported receiving help from a professional for their child's communication problem while 7% of parents of preschool children without LL reported that their child received help from a health professional (Skeat *et al.* 2014). Parent concern was the strongest predictor of service use, while child and family factors including socioeconomic status and maternal education did not drive the utilization of health services (Skeat *et al.* 2014).

Costs associated with LL

Most studies reported costs to the healthcare system and/or out-of-pocket (OOP) costs (co-payment) to family (table 3). Overall 2-year healthcare costs associated with LL (ranging from 2017 A$430 to A$2560) varied by child age (from birth to 13 years). Large variation in healthcare costs was observed between studies. For example, two Australian studies reported 2-year overall healthcare costs (i.e., not just language-related service use) in the same population of children aged 4–9 years up to A$687 (Sciberras *et al.* 2015) and up to A$1878 (Cronin *et al.* 2017), with differences in LL definitions, analysis methods (e.g., cross-sectional versus longitudinal) and in the cost components included in these two studies (e.g., exclusion versus inclusion of OOP costs and hospital costs). Children with LL aged 4–11 years had higher overall healthcare costs (higher 2-year medical costs at 4–5 and 8–9 years but also 2-year pharmaceutical costs at 10–11 years) than children with typical language (Cronin *et al.* 2017).

OOP costs associated with LL also varied by child age. For instance, 2-year OOP costs associated with LL at 4–5 years were almost doubled at 6–7 years (Le *et al.* 2017b). Significantly higher OOP costs were also noted in children with LL compared with children with typical language at 4–7 (Le *et al.* 2017b) and 10–11 years (Cronin *et al.* 2017).

Quality assessment of studies

The quality ratings of the included studies are presented in figure 2. Among 22 studies, three (14%), 16 (73%) and three (14%) studies were rated as strong, moderate and weak, respectively, according to the EPHPP global rating scales. Most studies used validated data-collection tools and comprehensively considered confounders in the analyses so they were rated as strong for these components (figure 2; see also Table B1 in the Appendix). Included studies did not blind both the outcome assessor and participants, therefore all studies performed relatively poorly on the blinding component.

Discussion

To our knowledge, this is the first review on the broader economic burden of LL, including service use and costs as well as HRQoL. Findings from this review indicated that there was an association between LL and impairment in different aspects of children's HRQoL but it is unclear whether or not there is a burden of LL on overall HRQoL. This is consistent with the previous review published in 2012 (Feeney *et al.* 2012). A combination of the variety of HRQoL instruments used in the studies and the heterogeneity in study population, study designs and methodologies may explain the variability in findings among the studies included in our review. For example, two thirds of studies using non-preference-based instruments (e.g., PedsQL) found an association between LL and impairment in either various aspects of children's HRQoL or overall HRQoL while studies using preference-based instruments (e.g., HUI3, 16D or 17D) did not find this association. Thus, it is possible that non-preference-based instruments may be more sensitive to detect HRQoL impairment associated with LL than preference-based instruments. Future research is needed to disentangle whether or not the choice of HRQoL instruments impacts study results and the performance of these HRQoL instruments in children with LL.

On the other hand, there was variation in findings within studies with a control group or without a control group. For example, among three studies without a control group of children with typical language, two studies found similar HRQoL in children with LL to the healthy population mean while one study found lower HRQoL in children with severe LL than the healthy population mean. In this instance, the severity of the condition may explain the difference in findings among these studies. It is also noteworthy that finding from the studies without a control group can only be descriptive and thus limits the extent to which a conclusion about an association between LL and children's HRQoL could be drawn. Among studies with a control group (both matched and non-matched control studies), variation was found in the various HRQoL aspects that were associated with LL, probably due to the differences in HRQoL instruments used and study populations.

Table 3. Included studies of costs and service utilization associated with child low language

Study	Population	Study design	Study setting	Study perspective	LL definition	Cost instrument used	Cost outcomes	Reference year	Main findings (cost estimates as conversion to 2017 A$)
Boyle *et al.* (2009)	$n = 161$ with LL Age: 6–11 years	RCT	Scotland	Healthcare provider	Expressive and/or receptive language scores ≤ 1.25 SD below the normative mean of 100 on the CELF-3 UK	Program record, NHS annual salary rate, taxi tariffs	Average cost per child over 30 weeks	2004	Average cost per child with LL: A$480
Cronin *et al.* (2017)	$n = 4983$ (LL: 4–5 years: 340 6–7 years: 278 8–9 years: 408 10–11 years: 451 12–13 years: 474) Age: 4–13 years	Longitudinal study	Australia	Healthcare system and family	Standardized language scores ≤ 1.25 SD below the normative mean on the corresponding language measure at each age groups (PPVT-III, academic rating scale-language and literacy, parents' evaluation of developmental status (PEDS) receptive language)	Medicare (PBS/MBS), private hospital data	Two-year cost to healthcare system and family per child	2014	*Two-year total healthcare costs associated with LL* 4–5 years: A$2112 6–7 years: A$2062 8–9 years: A$1548 10–11 years: A$2560 12–13 years: A$2472 *Two-year family costs associated with LL* 4–5 years: A$326 6–7 years: A$324 8–9 years: A$240 10–11 years: A$361 12–13 years: A$431 Compared with children with typical language, children with LL had higher total 2-year healthcare costs (including family OOP costs) at child ages 4–5, 6–7 and 10–11 years
Gibbard *et al.* (2004)	$n = 22$ with LL Age: 22–36 months	RCT	UK	Healthcare provider and family	The Reynell Developmental Language Scales, Preschool Language Scale—3 UK (comprehension and expression)	Program record and parent report	Cost to healthcare system and family per child over 6 months	1999	Healthcare cost, mean (SD): A$215 (5) Family cost: A$17 (travel) plus 178 min (travel and time spent at clinic)

Continued

Table 3. Continued

Study	Population	Study design	Study setting	Study perspective	LL definition	Cost instrument used	Cost outcomes	Reference year	Main findings (cost estimates as conversion to 2017 A$)
Law *et al.* (2006)	$n = 91$ with LL Age: 2.9 years	Case-controlled study	UK	Healthcare provider	Preschool Language Scale—3 UK	Program record and parent report	Annual health service cost per child	2004	Costs of speech language therapy (SLT) services per year, mean (SD): A$461 (665) to A$480 (620)
Le *et al.* (2017b)	$n = 778$ (180 with LL) Age 4–9 years	Prospective cohort study	Victoria, Australia	Government and family	Expressive and/or receptive language scores ≤ 1.25 SD below the normative mean of 100 on the CELF-P2	Study designed questionnaire	Two-year health service cost to government and family per child	2014	*Cost to government* (mean (SD)) 4–5 years: A$287 (503) 6–7 years: A$357 (489) *Cost to family* (mean (SD)) 4–5 years: A$401 (831) 6–7 years: A$759 (1304) *Two-year overall cost per child* Normal Language: A$134 (576) Transient LL: A$610 (1695) Persistent LL: A$920 (1603) At 5, 7 and 9 years, respectively, 21%, 11% and 8% of families reported using services for speech and/or language concerns Between 4 and 9 years, children with persistent LL had significantly higher service use and costs than those with typical language

Continued

Table 3. Continued

Study	Population	Study design	Study setting	Study perspective	LL definition	Cost instrument used	Cost outcomes	Reference year	Main findings (cost estimates as conversion to 2017 A$)
Mazer *et al.* (2017)	$n = 60$ with LL Age: School age, not specified	Case-controlled study	Quebec, Canada	n.a.	Not specified	n.a.	n.a.	n.a.	Children with LL received more health services in the rehabilitation centre than in the school system. Children with LL were more likely to continue to receive both OT and SLT services in their first year of school The change in receipt of services from the rehabilitation centre to the school setting did not vary by any demographic factors except for those with a family history of developmental delay 48% parents reported their child should be receiving services at school that they were not getting and 25% parents reported their child was not receiving health services outside of school as they should be
Nasuuna *et al.* (2016)	$n = 24678$ (3561 with LL) Age: 4–7 years	Quasi-longitudinal study	Victoria, Australia	n.a.	Parent report on the School Entry Health Questionnaire	n.a.	n.a.	n.a.	1836 (55%) used a speech–language therapy service, and 164 (9%) continued to attend at school entry

Continued

Table 3. Continued

Study	Population	Study design	Study setting	Study perspective	LL definition	Cost instrument used	Cost outcomes	Reference year	Main findings (cost estimates as conversion to 2017 A$)
Skeat *et al.* (2014)	$n = 983$ (196 with LL) Age: 4–5 years	Prospective cohort study	Victoria, Australia	n.a.	Receptive or expressive standard score on the CELF-P2 ≤ 1.25 SD below the population means	n.a.	n.a.	n.a.	About 45% children with communication problem and 7% children without communication problem received help from a professional Parent concern was the strongest predictor of service use
Skeat *et al.* (2011)	$n = 983$ (196 with LL) Age: 4–5 years	Prospective cohort study	Victoria, Australia	Healthcare system and family	Expressive and/or receptive language scores ≤ 1.25 SD below the normative mean of 100 on the CELF-P2	Study designed questionnaire	Total cost to healthcare system and family per child	2010	16% had used services (one-third of whom used more than one service); 11% had accessed SLT treatment, and 6% had accessed other professionals Total healthcare and family costs of all services varied from A$45 to A$21,655 (median of A$1396)

Continued

Table 3. Continued

Study	Population	Study design	Study setting	Study perspective	LL definition	Cost instrument used	Cost outcomes	Reference year	Main findings (cost estimates as conversion to 2017 A$)
Sciberras *et al.* (2015)	Birth cohort: $n = 4191$ (LL: 0–1 years: 422 2–3 years: 385 4–5 years: 369); Kindergarten cohort: $n = 4406$ (LL: 4–5 years: 422 6–7 years: 385 8–9 years: 369)	Prospective cohort study	Australia	Healthcare system	Standardized language scores ≤ 1.25 SD below the normative mean on the corresponding language measure at each age (CSBS, CDI-Words and Sentences, PPVT-III, CDI-Grammatical markers, CCC-2)	MBS, PBS	Two-year total Medicare cost and pharmaceutical costs per child	2012	*Birth cohort:* A$1483, A$839, A$945 at 0–1, 2–3, 4–5 years, respectively *Kindergarten cohort:* A$687, A$577 and A$757 at 4–5, 6–7 and 8–9 years, respectively. Compared with children with typical language, children with LL had higher 2-year total Medicare costs at child ages 0–1, 2–3, 4–5 years (B cohort) and 4–5 and 8–9 years (K cohort)
Wake *et al.* (2015)	$n = 200$ with LL Age: 4–6 years	RCT	Victoria, Australia	Government and family	Expressive and/or receptive language scores ≤ 1.25 SD below the normative mean of 100 on the CELF-P2	Study designed questionnaire	Two-year health service cost to government and family per child	2012	*Service use costs to government:* mean (SD) 4–5 years: A$324 (A$842) 5–6 years: A$165 (A$516) *Two-year service use costs to family*: mean (SD) 4–5 years: A$343 (A$1169) 5–6 years: A$306 (A$1159)

Note: B, birth; CCC, Child's Communication Checklist; CDBS, Communication and Symbolic Behavior Scales; CDI, MacArthur–Bates Communicative Development Inventory; LL, low language; MBS, Medicare Benefit Schedule; n.a., not applicable for non-cost-related studies; NHS, National Health Service; K, kindergarten; OOP, out-of-pocket; OT, occupational therapy; PBS, Pharmaceutical Benefit Schedule; PPVT, Peabody Picture Vocabulary Test; RCT, randomized controlled trial; SD, standard deviation; SLT, speech–language therapy.

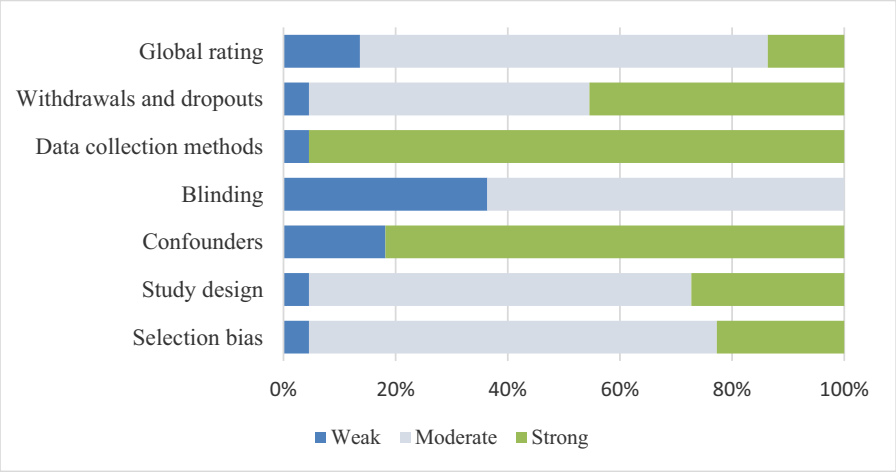

Figure 2. Quality assessment using the EPHPP generic quantitative tool of the 22 included studies. [Colour figure can be viewed at wileyonlinelibrary.com]

The variation in HRQoL domains affected by LL reflects the various aspects of life measured by different HRQoL instruments as well as the nature of the disorder (e.g., LL is likely to affect some domains more than others). Children with LL had limitations on social, emotional and school functioning (Rice *et al.* 2008), thus these specific HRQoL domains were more likely to be affected by LL. This finding was similar to those from qualitative studies on HRQoL of children with LL (Markham and Dean 2009, Markham *et al.* 2009).

The diversity of HRQoL instruments employed in the included studies also contributed to the variation in the factors affecting the association between LL and different aspects of life. Given the link between the child's relationship with family and HRQoL ratings (Markham and Dean 2009), factors such as parental warmth and parental mental health are expected to affect this association.

Service utilization and child LL

We found that just over half the children with LL had accessed SLT services before school entry (4–5 years). The OOP costs of SLT services borne by the family are high (Le *et al.* 2017b), thus, patient access to these services may be largely impacted by families' capacity to incur the cost. Long waiting times may also discourage families to access SLT services, for example, in Australia, 25% of parents of children with LL reported a waiting time of more than 6 months and 15% of parents reported a waiting time of more than 1 year (Senate Community Affairs References Committee 2014). Location and availability of speech-pathology services may also influence the use of these services (McCormack and Verdon 2015). For example, McCormack and Verdon (2015) found a lack of SLT services in the vast majority of non-metropolitan areas, and that there were people living in non-metropolitan areas located > 50 km from an SLT service in New South Wales and Victoria, Australia.

We also found a small proportion of parents reported receiving professional help when their child did not actually meet criteria for LL (e.g., child language assessment scores did not fall below the cut-point of 1.25 SD below the mean). In these instances, parental concern was a significant predictor of service utilization.

Costs associated with child LL

There is large variation in the costs associated with LL reported in this review because of the heterogeneity in the study settings, participant's characteristics, LL definition, and analysis approaches employed in the included studies. For example, while some studies did not include costs of health services associated with LL other than SLT services, other studies considered all costs of health services (e.g., general practitioner and paediatrician), some of which may not be specific to the LL condition per se but rather for other non-LL health conditions (e.g., common cold). Variation in cost data sources (self-reported versus medical record) were also noted. Although self-reported cost data may generate bias due to recall issues, it could be a valuable supplementary data source or alternative to consider when cost or medical record data are not available (e.g., services are not covered by public health service or additional expenses are incurred by families). Furthermore, different studies adapted different methodologies (e.g., inclusion of various cost components), resulting in the reporting of slightly different findings by studies using the same dataset.

It is unsurprising that differences are not evident in pharmaceuticals costs between children with and

without LL who were younger than 10 years of age as children require SLT services for LL rather than medications. The higher pharmaceutical costs in children with LL than children with typical language at 10–11 years could, therefore, be due to associated health conditions such as ADHD.

No studies included in this review explored the associated costs incurred by the education system, which includes costs related to special education, grade retention and school support services. Indirect costs associated with LL such as productivity loss or absenteeism from the workforce of parents of children with LL, or productivity loss (school absenteeism) of children with LL were largely unexplored. The economic burden of LL reported in the included studies may underestimate the true overall societal costs of LL.

Strengths and limitations

While the strengths of our review include the extensive search of the research literature and that this is the first systematic review on costs and service utilization associated with LL, there are a few limitations to note. First, given a lack of consensus about quality assessment of studies and which approach is preferable (Dixon-Woods *et al.* 2004), especially the best approach to assess HRQoL or service use/cost studies, we used the generic EPHPP quantitative study quality assessment tool. The EPHPP is used to evaluate quality across a variety of study types including RCTs, observational and cohort studies. However, some domains/questions of the assessment tool seem to be more appropriate for RCTs than for non-RCT designs, leading to non-RCT studies being more frequently rated as moderate or weak. Second, although we employed a comprehensive search strategy, we did not have capacity to include non-English literature. Third, our review focused on children with LL, and this may limit the generalization of our findings to children with other communication problems (e.g., speech or stuttering). Fourth, the inclusion of studies without a control group and reliance on observational data may limit interpretation of the causal impact of LL. Finally, as cost studies included in this review used validated language measures (e.g., CELF, PLS) to define child LL, it is not possible to compare costs associated with LL based on clinical diagnosis versus approximation methods (screening tools such as PPVT and parental concern).

Policy relevance and economic implications

This review shows a wide utilization of non-preference-based HRQoL instruments in the language literature. Being preferred by policy-makers (Sung *et al.* 2010), further research using preference-based instruments to capture HRQoL in paediatric cohorts is encouraged. Although LL negatively affects various aspects of HRQoL, research to date cannot confirm the adverse effect of LL on children's overall HRQoL. Further research is needed to explore the impact of LL on children's HRQoL, especially longitudinal studies in which the sequences between LL and associated HRQoL could be explored. It is also important to understand that LL impacts different aspects of children's HRQoL (more commonly social functioning) and these impacts vary by child age. Therefore, interventions to improve language for children experiencing LL could benefit other aspects of life e.g., school and social functioning, as well as speech and language, with effects potentially varying according to child age.

Owing to the exclusion of non-English literature and the uneven SLT research activities across English-speaking countries, most cost studies included in this review are from Australia, the UK and Canada, which have relatively similar healthcare systems. The differences in the healthcare systems and practices internationally may hinder the generalization of service utilization and costs from this review to other countries such as the United States or low- and middle-income countries. Further research from international contexts is encouraged.

This review also highlights a need for stronger financial support from government for families of children with LL to be supported to access services, especially disadvantaged families. Strategies to raise parental awareness of LL may encourage families to seek professional help for their child as parental perceptions of their child's health and well-being are the foremost driver of health care service utilization (Nicola and Watter 2015).

The cost burden associated with LL was comparable to that for childhood underweight, overweight or obesity (Clifford *et al.* 2015), thus, LL should be identified as a priority for population health resource allocation. Although this review did not find any studies that reported on the cost incurred to education and employment sectors, Ruben (2000) estimated substantial costs to the education and employment sectors for people with communication problems. Further research to examine non-healthcare costs associated with LL (e.g., costs to the education system) is, thus, warranted.

Conclusions

In summary, this comprehensive review showed an association between LL and reduced children's HRQoL in some aspects such as social functioning. However, whether or not the condition negatively influences children's overall HRQoL is inconclusive. A large proportion of families of children with LL did not access speech–language therapy services. We also found substantial

costs associated with LL to the healthcare system and to families, but costs to education and employment sectors are largely unexplored. Given the limited literature on either HRQoL or service use/costs associated with LL and the heterogeneity in the study samples and methodologies, further research is required, especially to examine the burden of LL on education and employment sectors.

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Appendix A

Table A1. Search term in the first stage

S1	(review OR 'meta-analysis' OR 'meta-analytic')
S2	(speech OR language OR communicati*)
S3	(problem* OR difficult* OR disorder* OR delay OR impair*)
S4	S2 NEAR S3
S5	('quality of life' OR QoL OR HRQoL).
S6	cost* OR service* OR resource* OR 'healthcare' OR healthcare
S7	use OR utili* OR cost*
S8	S5 NEAR S7
S9	S1 AND S4 AND S5
S10	S1 AND S4 AND S8
S11	Limiters: human
S12	S10 AND S11

Table A2. Search term in the second stage

S1	Toddler* OR child* OR p#ediatri*OR boy* OR girl* OR adolescen* OR 'young people' OR teen* OR youth*	Population
S2	(communicati* OR speech OR language) N3 (problem* OR difficult* OR disorder* OR delay OR impair* OR deficit* OR disab*)	Condition
S3	('well-being' OR wellbeing OR 'well being' OR 'personal satisfaction' OR 'quality of life' OR perception OR QoL OR HRQoL OR utilit*)	Outcomes: quality of life outcome
S4	Econom* OR cost* OR burden	Outcomes: economic burden (service use and costs)
S5	(service* OR resource* OR 'healthcare' OR healthcare) N3 (use OR utili* OR cost* OR expen*)	
S6	(GP OR clinician OR 'general practitioner' OR doctor OR 'speech-pathology*' OR 'speech therap*' OR 'specialist' OR ENT OR p#ediatrician OR psycholog*) N1 visit*	
S7	S4 OR S5 OR S6	
S8	S1 AND S2 AND S7	Combined search
S9	Limiters: English, human	
S10	Limiters: 2002–2017	
S11	S8 AND S9 AND S10	Combined search with limiters

Appendix B

Table B1. Quality assessment of the included studies

Study	Selection bias	Study design	Confounders	Blinding	Data collection methods	Withdrawals and dropouts	Global rating
Mazer *et al.* (2017)	Moderate	Moderate	Strong	Moderate	Strong	Moderate	Moderate
McKean *et al.* (2017)	Strong	Moderate	Strong	Moderate	Strong	Moderate	Moderate
Nasuuna *et al.* (2016)	Moderate	Moderate	Strong	Moderate	Strong	Moderate	Moderate
Nicola and Watter (2015)	Moderate	Moderate	Strong	Moderate	Strong	Moderate	Moderate
Sciberras *et al.* (2015)	Strong	Moderate	Strong	Moderate	Strong	Moderate	Moderate
Skeat *et al.* (2011)	Strong	Moderate	Weak	Moderate	Strong	Weak	Weak
Thomas-Stonell *et al.* (2010)	Moderate	Weak	Weak	Moderate	Strong	Strong	Weak
Van Agt *et al.* (2010)	Weak	Strong	Strong	Moderate	Strong	Strong	Moderate

Continued

Table B1. Continued

Study	Selection bias	Study design	Confounders	Blinding	Data collection methods	Withdrawals and dropouts	Global rating
Van Agt *et al.* (2005)	Moderate	Strong	Strong	Moderate	Strong	Moderate	Moderate
Wake *et al.* (2015)	Moderate	Strong	Strong	Moderate	Strong	Strong	Strong
Wake *et al.* (2013)	Moderate	Strong	Strong	Moderate	Strong	Strong	Strong
Arkkila *et al.* (2009)	Moderate	Moderate	Strong	Weak	Strong	Moderate	Moderate
Arkkila *et al.* (2011)	Moderate	Moderate	Strong	Weak	Strong	Strong	Moderate
Feeney *et al.* (2017)	Moderate	Moderate	Weak	Moderate	Strong	Strong	Moderate
Boyle *et al.* (2009)	Strong	Strong	Strong	Moderate	Strong	Strong	Strong
Law *et al.* (2006)	Moderate	Moderate	Strong	Moderate	Weak	Moderate	Moderate
Flapper and Schoemaker *et al.* (2013)	Moderate	Moderate	Weak	Weak	Strong	Moderate	Weak
Cronin *et al.* (2017)	Moderate	Moderate	Strong	Weak	Strong	Moderate	Moderate
Hubert-Dibon *et al.* (2016)	Moderate	Moderate	Strong	Weak	Strong	Moderate	Moderate
Le *et al.* (2017)	Moderate	Moderate	Strong	Weak	Strong	Strong	Moderate
Gibbard *et al.* (2004)	Strong	Strong	Strong	Weak	Strong	Strong	Moderate
Skeat *et al.* (2014)	Moderate	Moderate	Strong	Weak	Strong	Strong	Moderate

Notes: Selection bias: assesses the representativeness of the target population (e.g., whether they are randomly selected from a comprehensive list of individuals in the target population) and the percentage of subjects in the control and intervention groups who agreed to participate in the study before they were assigned to intervention or control groups.
Study design: assesses the likelihood of bias due to the allocation process in an experimental study or the independence of the assessments of exposure and outcome for observational studies.
Confounders: assesses if confounders were controlled in the design (by stratification or matching) or in the analysis.
Blinding: assesses if study assessors were blinded to participants or study participants were blinded to the research questions.
Data collection methods: assess the reliability and validity of tools for primary outcome measures.
Withdrawals and drop-outs: assess the percentage of participants completing the study and refer to the percentage of subjects remaining in the study at the final data-collection period in all groups.