Cost of illness and health-related quality of life for stuttering: Two systematic reviews

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Abstract

Background: For those who stutter, verbal communication is typically compromised in social situations. This may attract negative responses from listeners and stigmatization by society. These have the potential to impair health-related quality of life across a range of domains, including qualitative and quantitative impacts on speech output, mental health issues, and failure to attain educational and occupational potential. These systematic reviews were designed to explore this matter using traditional health economics perspectives of utility measures and cost of illness.

*Methods***:** Studies were included if they involved children, adolescents, or adults with stuttering as a primary diagnosis. The quality of life search strategy identified 2,607 reports, of which three were included in the quality of life analysis. The cost of illness search strategy identified 3,778 reports, of which 39 were included in the cost of illness analysis.

Results: Two of the three studies included in the quality of life analysis had a high risk of bias. When measured using utility scores, quality of life for people who stutter was in the range of those reported for chronic health conditions such as diabetes mellitus, cardiovascular disease, and cancer. However, there is little such evidence of quality of life impairment during the preschool years. Studies included in the cost of illness analysis carried considerable risk of bias overall.

Conclusions: For people who stutter, there are substantive direct and indirect costs of illness. These include impairment, challenges, and distress across many domains throughout life, including income, education, employment, and social functioning. Evidence of quality of life impairment using utility measures is extremely limited. If this situation is not remedied, the lifetime impairment, challenges, and distress experienced by those who stutter cannot be documented in a form that can be used to influence health policy and healthcare spending.

Introduction

Health-related quality of life and health economic evaluation

Health economics relates decisions about the use of health care to the costs and outcomes of that health care. A key consideration within health economics is health-related quality of life, which is a multidimensional concept that extends beyond clinical measures of health, such as diagnosis and physiological function, to include physical and social functionality, energy level, and mental and physical wellbeing (Wilson & Cleary, 1995). Health-related quality of life includes interaction between those variables, individual characteristics, and the environment in which the individual functions.

All health issues impact not just the individuals and families who experience them, but also health care systems and the societies they serve. Health economic evaluation informs what is therefore a “range of very different but unavoidable decisions in health care” (Drummond et al., 2015, p. 3). Government and private health care funders, and heath care policy makers, need to compare different interventions for different diseases and make decisions about where to direct health care spending to maximize benefits. This can be done with overarching measures that assess health-related quality of life across many diseases and support health economic evaluations.

Utility values are fundamental data for conducting health economic evaluations, being a means to measure health-related quality of life across different diseases. Utility values represent an individual’s preference for different health states, traditionally measured on a scale between 0 and 1, spanning the extremes of death and perfect health (Drummond et al., 2015). The other fundamental datum for health economics is cost of illness, which is a monetary measure of the burden of an illness to society. The costs of such a burden involve the fundamental expense to the individual in terms of receiving treatment and lost work hours, as well as wider societal costs.

Economic evaluations of healthcare interventions can be conducted through cost-utility analysis. The summary outcome of a cost-utility analysis is cost per quality-adjusted life year. A quality-adjusted life year is obtained by multiplying a utility value by the number of years lived in a certain health state. This provides an index that combines quality of life and length of life obtained after treatment. One quality-adjusted life year is equivalent to one year lived in perfect health. Cost-utility analyses allow health care funders and policy makers to compare different interventions for the same disease, and for different diseases, using one summary outcome. This facilitates maximum health benefit from healthcare spending.

Health-related quality of life and stuttering

Stuttering is a prevalent and potentially lifelong disorder, with a lifetime incidence in the range of 8–10% (Bloodstein et al., 2021; Yairi & Ambrose, 2013). While around 10% of preschool children start to stutter, most recover naturally within a few years of onset, leaving around 1% to stutter through childhood, adolescence, and adulthood. Verbal communication is compromised for many people who stutter, and this has the potential to impact health-related quality of life. This is reflected in how commonly treatment is sought. A study by Boyce et al. (2022) reported that 73% of adults who stuttered received treatment from a speech-language-pathologist. In that report, 92% of parents indicated that their children (mean age 11 years) had received treatment. The following sections give an overview of the many features of the disorder that are potential sources of such impact.

Qualitative and quantitative aspects of speech output

Fundamentally, the speaker experiences disruptive speech behaviors. These have been categorized as various types of repeated movements, fixed postures (“blocks”) with or without audible airflow, and verbal or nonverbal superfluous behaviors (Teesson et al., 2003). Those disruptive speech behaviors, more often than not, occur together during stuttering moments (O’Brian et al., 2022). Based on a 12-hour speaking day, a report found a mean number of 33,617 syllables spoken for adults with a mean 7.8 percent syllables stuttered (Karimi et al., 2013a). Those data suggest that participants stuttered around 2–3 thousand times per day. Because stuttering moments occur so frequently, and each of them is time consuming, adults who stutter take, on average, three times as long as others to convey their message (Johnson, 1961; Spencer et al., 2009).

It seems that the complexity of stuttering moments increases during life, with repeated movements being predominant at onset during the preschool years (Ambrose & Yairi, 1999; Reilly et al., 2009; Yairi & Lewis, 1984) and with evidence of their advancing behavioral complexity from adolescence to adulthood (O'Brian et al., 2022). Part of stuttering causality appears to involve anomalies of brain structure and function in areas subserving spoken language (Chang et al., 2018; Packman, 2012). Perhaps for that reason, the broad notion "loss of control" has been considered as fundamental to the disorder (Perkins, 1983), and around half of adults who stutter report a loss of control while speaking, "either often or always" (Tichenor & Yaruss, 2019, p. 4339).

The experience of anticipating stuttering during speech is common (Jackson et al., 2015; Johnson & Solomon, 1937; Martin & Haroldson, 1967; Milisen, 1938). Word avoidance and circumlocution are common responses to anticipation (Crichton-Smith, 2002; Jackson et al., 2015; Martens & Engel, 1986; Vanryckeghem et al., 2004). According to analyses using Systemic Functional Linguistics, adults who stutter have been shown to consciously change their use of language in order to limit conversational interaction. (Lee et al., 2015; Spencer et al., 2005, 2009). Systemic Functional Linguistics reflects how people modulate language in different contexts and situations. There is also some evidence of that effect during childhood (Weiss & Zebrowski, 1994), but there is no evidence that children who stutter have reduced language ability per se (Nippold, 2019). Constant effort to conceal stuttering with word avoidance and circumlocution, and also situation avoidance, occurs commonly (Boyle &

Gabel, 2020; Douglass et al., 2018) and has been labelled “covert stuttering” (Murphy et al., 2007) or “interiorized stuttering” (Sønsterud et al., 2022).

Stuttering variability

Stuttering severity varies across individuals, from mild to severe, and also within individuals. Survey responses of 204 adults (Tichenor & Yaruss, 2021) indicated that 97% of them experienced variability of their stuttering across time and situations. That result is consistent with a control chart study of stuttering variability during a day for 10 adults (Karimi et al., 2013b), which showed that half of them had stuttering that varied more than three standard deviations from their mean daily score. The majority of the Tichenor and Yaruss (2021) participants attributed frustration with the disorder to stuttering variability across times and situations. Stuttering severity will vary according to the nature of an audience, particularly its size (Porter, 1939; Siegel & Haugen, 1964; Steer & Johnson, 1936). Stuttering severity can vary from situation to situation (Ulliana & Ingham, 1984) and can vary when the speaker is in the same situation (Constantino et al., 2016).

Social anxiety

Stuttering moments may invoke social penalty by drawing attention to speech and interfering with communication. Accordingly, the disorder is commonly associated with mental health issues associated with social anxiety. A large body of evidence confirms this to be the case, with a meta-analysis showing those who stutter to be a standard deviation above controls for social anxiety scores (Craig & Tran, 2014). In particular, those who stutter are at high risk of social anxiety disorder diagnosis (Blumgart et al., 2010b; Iverach & Rapee, 2014; Iverach et al., 2016; Stein et al., 1996). It appears that the origins of these issues can be detected during early childhood (Briley et al., 2019; Langevin et al, 2010; McAllister, 2016). Briley et al. (2019) and McAllister (2016) used the Strengths and Difficulties Questionnaire in two large population studies; the questionnaire measures behavioral, emotional, and social well-being, with high scores indicating anxiety. Both studies reported significantly higher scores for preschool-age children who were stuttering up to age 5 years, compared with non-stuttering children. Langevin et al. (2010) reported the findings of a questionnaire sent to 77 parents of preschoolers who stutter, in which 90% of respondents reported clear signs of anxiety in their child, such as withdrawal and avoidance. One report of older children (Iverach et al., 2016) found that 24% of 7–12-year-old children who stutter received a social anxiety disorder diagnosis, compared with 4.6% of control children. For social anxiety in general, a systematic review and meta-analysis (Bernard et al., 2022) concluded that children and adolescents who stutter present with increased anxiety symptoms in comparison to their non-stuttering peers. The connection between stuttering and social anxiety is probably due to marginalisation caused by negative social stereotypes, microaggressions, bullying, stigma, and social exclusion (Boyle, 2018; Coalson et al. 2022; Doody et al., 1993; Erickson & Block, 2013; Ham, 1990; White & Collins, 1984). Negative social stereotypes pertain to all ages (Horsley & Fitzgibbon, 1987; Woods & Williams, 1976; Yairi & Williams, 1970). There is evidence that those stereotypes extend to perceptions of physical attractiveness (Van Borsel et al., 2011) and that stuttering can affect personal relationships (Connery et al., 2020).

Educational and occupational attainment

Stuttering may lead to impaired educational and occupational attainment. For adults, an inverse relationship has been reported for stuttering severity and educational attainment (Boyce et al., 2022; O'Brian et al., 2011). A large cohort report by Boyle et al. (1994) found significantly more chance of repeating a grade for children who stutter compared with control children. That effect was replicated by Berchiatti et al. (2020). There is evidence that children experience social isolation in school from fear of speaking in the classroom, and that they habitually avoid it (Daniels et al., 2012; Klompas & Ross, 2004). College professors in the US have been shown to have more negative perceptions of students who stutter than controls, and students who stutter were less comfortable than controls when approaching professors (Werle & Byrd, 2022). Not surprisingly, then, adults who stutter tend to have restricted occupational opportunities and outcomes (Blumgart et al., 2010a; Gerlach et al., 2018; Klein & Hood, 2004; McAllister et al., 2012).

The present reviews

In summary, stuttering is a prevalent and potentially lifelong disorder, which is capable of impairing health-related quality of life across a range of domains, including qualitative and quantitative impacts on speech output, mental health issues, and failure to attain educational and occupational potential. Although some individuals find strength, emotional growth and relationship benefits through their stuttering (Boyle et al, 2019; Constantino, 2016), a review of qualitative studies of the matter (Connery et al., 2020) concluded that stuttering has a “profound and predominantly negative impact” (p. 2232) on the experiences of those affected.

The present systematic review was designed to explore this matter by seeking information from existing literature to understand how stuttering affects quality of life and how interventions have potential to change quality of life. For the review, we did not explore measures that pertain specifically to the disorder of stuttering. Instead, we sought overarching quality of life utility measures from traditional health economics that are pertinent to health policy and funding. Such measures place health care for stuttering into a broader perspective. The review was also designed to explore economic impact of stuttering: the cost of illness or the economic costs of stuttering interventions. Costs involved are to the individual in terms of treatment expenses and lost work hours, along with carer burden, health and education system costs, and wider societal costs. Those costs are an essential comparison against quality of life and how it can be changed by intervention, and how it can guide health care funders and heath care policy makers to compare different interventions for different diseases.

Methods

Inclusion criteria

Studies were included if they involved children, adolescents, or adults with stuttering as a primary diagnosis. Participants with comorbid disorders were excluded because the outcomes of treating them would not be generalisable to participants with stuttering only. The review included quality of life instruments that directly measured health-related quality of life with utility scores. For the cost analysis all studies that detailed direct, indirect, and societal costs of stuttering were included. All interventions and all study types were included.

Search strategies

An initial search of Embase and MEDLINE Medical Subject Headings was conducted to identify articles relating to the costs and outcomes of stuttering therapy. Key words from the titles, abstracts, subject headings, and other index terms were used to develop a full Boolean search strategy. The search strategy was refined at a multidisciplinary meeting that included speech-language pathologists, health economists, and a clinical psychologist. The details of the search strategy are presented in Supplemental Material S1.

The databases searched were: Embase; MEDLINE; PsycINFO; Cumulative Index to Nursing and Allied Health Literature (CINAHL); the Cochrane Library, including the Cochrane Database of Systematic Reviews and Cochrane Central Register of Controlled Trials; ProQuest; Web of Science; Scopus; Paediatric Economic Database Evaluation (PEDE); and the National Health Service Economic Evaluation Database (NHS EED). To identify unpublished literature The Australian New Zealand clinical trials registry (ANZCTR), United States National Institutes of Health trial register, and Open Grey were searched. Searches were limited to English language. No limits on publication year were applied.

Search dates

The cost of illness and quality of life searches were run between 19th January 2021 and 20th May 2021. After the initial results were retrieved, searches of Embase, MEDLINE, PsycINFO, and Scopus were automatically re-run at weekly intervals and searches of CINAHL, ProQuest and Web of Science were re-run at monthly intervals until 13th August 2021. Database searches were re-run between the 14th and 15th February, 2022, to identify records indexed after 13th August, 2021. The Cochrane Library, ANZCTR, and the United States National Institutes of Health trial register were manually re-searched on 13th August 2021 and 14–15th February 2022. PEDE, NHS EED, and Open Grey ceased to be updated on 31st Dec 2020, 31st March 2018, and 27th July 2018, respectively so were not re-searched in 2022. Additional records identified were added to the search results. Citations included in the reference lists of included studies were manually searched to identify additional papers of interest.

Search procedures

Following the searches, citations were collated in Excel, and duplicates removed. Titles and abstracts were screened against the inclusion criteria and potentially relevant studies retrieved in full. Retrieved full text articles were screened in detail against the inclusion criteria, and relevant articles were included in the review. Eligibility decisions at the title/abstract and full text stage were recorded in a spreadsheet. Two investigators independently conducted title/abstract and full text reviews for relevance and any disagreement was resolved through consensus. The search strategies are presented in PRISMA flow diagrams (Moher et al., 2009): quality of life in Figure 1 and cost of illness in Figure 2.

INSERT FIGURES 1 and 2 ABOUT HERE

Data extraction form

A data extraction form for included studies was developed, based on the Cochrane checklist of items to consider in data collection (Li et al., 2021), and piloted prior to use. The form is presented in Supplemental Material S2. Relevant data were extracted from each study and recorded in Microsoft Access. The data extracted included details about the study (design, methods, year, location), participants (age, gender, comorbidity, socioeconomic status, ethnicity), intervention (setting, duration), and outcomes (costs and health outcomes). No summary measures were pre-specified.

Risk of bias assessment

The risk of bias was assessed at the study level using the RoB 2 tool for randomized trials (Sterne et al., 2019), and the ROBINS-I tool (Sterne et al., 2016) for non-randomized studies. Qualitative research was assessed using the CASP qualitative studies checklist (Critical Appraisal Skills Programme, 2018). The CASP checklist is cited by The Cochrane Qualitative and Implementation Methods Group as “the most commonly used tool in qualitative evidence synthesis in Cochrane and World Health organisation guideline processes” (Noyes et al., 2018, p. 50). Risk of bias details for included studies are presented in Supplemental Material S3.

Cost standardisation

Costs were inflated to 2020/21 values using an inflation calculator relevant to the reported currency, then converted into US dollars where necessary using the exchange rate listed at www.xe.com on 20 August, 2021.

Results

Quality of life

The quality of life search strategy identified 2,067 reports, of which three were included in the quality of life analysis. From these reports, 30 studies were excluded during the review because they did not generate a utility measure. Of these 30 studies, 19 used eleven different instruments directly pertinent to quality of life. Details are presented in Figure 1 and Table 1.

Three reports measured quality of life using utility measures (de Sonneville-Koedoot et al., 2015; McAllister et al., 2017; Omori et al., 2021). These involved 242 participants: 199 children, 31 adults, 12 in a mixed cohort. The proportion of participants who were male ranged by treatment group from 60% to 94%. The three studies are summarized in Table 2.

INSERT TABLE 1 and 2 ABOUT HERE

The utility measures used were the Health Utilities Index 3 (HUI3), the 3-level version of EuroQol-5 Dimension (EQ-5D-3L), and the 5-level EQ-5D questionnaire. HUI3 considers eight attributes: vision, hearing, speech, ambulation, dexterity, emotion, cognition, and pain. EQ-5D considers five dimensions: mobility, self-care, usual activities, pain/discomfort, and anxiety/depression.

In the de Sonneville-Koedoot et al. (2015) study, using the HUI3 instrument with preschool children, the mean baseline utility score was 0.88 for both treatment groups (RESTART-DCM and Lidcombe Program). At 18-months follow-up, the mean utility score improved for both groups by 0.07 in the Lidcombe Program group and 0.06 for the RESTART-DCM group. In the Omori et al. (2021) study of adults, the mean baseline utility score was 0.80 for the intervention group (Cognitive Behavior Therapy (CBT) + speech treatment) and 0.71 for the control group (speech treatment), using the 5-level EQ-5D instrument. After 4 months of treatment, the mean intervention group (CBT + speech treatment) utility score increased by 0.10 utility score values, and with the control group (speech treatment), it increased in a favourable direction by 0.06 utility score values. In the McAllister et al. (2017) study of adults, the mean baseline utility score was 0.82 for the intervention group and 0.83 for the control group, using the EQ-5D-3L instrument. At 4 months post intervention, the mean intervention group utility score worsened slightly in both groups by 0.02 utility score values. No study reported a statistically significant difference in utility scores between interventions.

The McAllister et al. (2017) randomized trial was considered to have a low risk of bias, and the de Sonneville-Koedoot et al. (2015) trial was considered to have a high risk of bias due to the potential for loss of allocation concealment. The Omori et al. (2021) non-randomized trial had a high risk of bias for several reasons (see Supplemental Material S3).

Cost of illness

Interventions and Comparators

The cost of illness search strategy identified 3,778 reports. From these reports, 39 studies were included in the cost of illness analysis. Of these studies, 36 did not include interventions or comparators. Details are presented in Figure 2. One report compared two stuttering interventions (the Lidcombe Program and RESTART-DCM) (de Sonneville-Koedoot et al., 2015), and one report compared a web-based cognitive bias modification program with placebo (McAllister et al., 2017). One report presented long-term follow-up data for two single-arm trials of smooth speech treatment for stuttering (Craig & Calver, 1991).

Outcomes

The direct costs of treatment, indirect costs connected with treatment, and societal costs of stuttering were of interest for the cost analysis. Societal costs were grouped into three themes: education, employment, and social outcomes. In total, 155 cost outcomes were extracted. Of the 39 included studies, six provided evidence on healthcare utilisation, six provided evidence on direct and indirect costs of stuttering, 20 provided evidence for the impact of stuttering on educational outcomes, 22 provided evidence on employment outcomes, and 11 provided evidence on social outcomes, and one presented information on society's willingness to pay for stuttering treatment. A summary of studies included in the cost of illness analysis is presented in Supplemental Material S4, along with overall risk of bias for the studies (see Supplemental Material S3 for further risk of bias details).

Healthcare utilisation

There is evidence of trends in health care utilisation for those who stutter, though most findings were not statistically significant. Children who stutter were (a) more likely to be hospitalized and spend more days in hospital than children with no developmental disabilities (Boyle et al., 1994); (b) more likely to attend hospital emergency departments ($p < .01$) and undergo surgical or medical procedures ($p < .05$) than children with no developmental disabilities (Boulet et al., 2009); (c) more likely to make frequent doctor visits than children with no developmental disabilities (Boyle et al., 1994); (d) more likely to visit specialists ($p < .05$), allied health professionals ($p < .01$), and mental health professionals (difference not significant); and (e) more likely to take medication for longer than 3 months and require special equipment than children with no developmental disabilities (Boulet et al., 2009).

A retrospective analysis of health insurance data by Sommer et al. (2021) reported that approximately 45% of individuals of any age diagnosed with stuttering seek speech treatment within a year of diagnosis and receive an average of 13.5 speech therapy sessions in the first year of treatment. A report by de Sonneville-Koedoot (2015) indicated that children treated with the Lidcombe Program received 20 hours of stuttering therapy over an 18-month period, compared with 18 hours with RESTART-DCM. McAllister et. al. (2017) collected data at baseline and 4 months follow-up for adults on hospitalisations, day hospital, primary care visits, specialists, other therapy, medication and special equipment. In this report, individuals in the intervention group, at baseline, had received 3.1 primary care visits and 2.4 prescriptions in the previous 4 months. Individuals in the placebo group received 2.0 primary care visits and 4.5 prescriptions at baseline. Mean specialist visits, other therapy, hospitalisations, and special equipment use were all below 1.0 at baseline in the intervention and control groups.

Direct and indirect costs of stuttering

A study by Blumgart et al. (2010a) reported the mean cost of stuttering treatment over five years to be the equivalent of US$2,528. The overall direct cost of stuttering over five years, including speech treatment, other treatment, technology, self-help, and other costs was US$5,101 per adult client (Blumgart et al., 2010b). The mean cost of stuttering treatment in one randomized controlled trial of early intervention was US$1,976 for the Lidcombe Program and US$1,788 for RESTART-DCM (de Sonneville-Koedoot et al., 2015). Total direct costs were US$3,681 for the Lidcombe Program and US$3,541 for RESTART-DCM: including additional allied health, home therapy, travel costs, and parent time associated with home therapy. Total costs, including absenteeism and productivity losses over the 18-month study period were US$4,444 for the Lidcombe Program and US$4,212 for RESTART-DCM.

One qualitative research report conducted by Georges (2017) considered the financial impact of stuttering. Seven of 10 participants reported no financial strain associated with stuttering. However, the same number noted that their health insurance did not cover stuttering treatment. These individuals described finding alternative methods to fund therapy or seeking alternatives to treatment. A report of the impact of stuttering on adolescents and their families by Erickson and Block (2013) indicated that stuttering placed at least some financial strain on 61% of families. Of the parents interviewed, 19% missed work and 10% had made time and financial sacrifices due to their child’s stuttering.

In a cross-sectional study investigating society's willingness to pay for stuttering treatment, Franic et al. (2012) reported that the mean amount respondents were willing to pay ranged from US$24,927 for an intervention that improved stuttering from severe to mild to US$61,810 for an intervention that stopped severe stuttering; this was equivalent to between two and four times their annual income.

Education

Stuttering impacted education for a range of measures, including performance, attainment, attendance, and the requirement of special support services. In two reports, McClure and Yaruss (2003) reported 80% of respondents stating that stuttering interfered with their schoolwork, and Hayhow et al. (2002) reported 95%. Two reports indicated academic difficulties associated with stuttering that were indirectly linked to the occurrence of bullying (Erickson & Block, 2013; Hugh-Jones & Smith, 1999). Another two reports indicated that children who stutter were significantly more likely to require special education services or early intervention compared with children who have no developmental disabilities (Boyle et al., 1994; Boulet et al., 2009). Two further reports noted a link between stuttering and avoiding school, mediated by the presence of bullying (Erickson & Block, 2013; Hugh-Jones & Smith, 1999); and another reported the mean number of school days lost each year by children who stutter to be 7.7 compared with 3.0 days for children with no developmental disabilities ($p < .01$) (Boyle et al., 1994).

Three studies reported that children who stutter perform worse academically than children who do not stutter (Berchiatti et al., 2020; Calnan & Richardson, 1977; Williams et al. 1969). Boyle et al. (1994) reported that 29% of children who stutter had repeated a grade, compared with 13% of children with no developmental disabilities ($p < .01$). Conversely, a study of college instructors evaluating oral presentations reported that the performances of individuals who stutter were evaluated significantly higher than fluent controls ($p < .01$), equating to a "full letter" grade difference in total score (Werle & Byrd, 2022). Another study reported a significant inverse relationship between stuttering severity and highest educational achievement (O'Brian et al., 2022); and another reported that stuttering predicted lower educational attainment ($p < .01$) (Rosenbaum, 2018). However, two additional follow-up studies reported that after controlling for confounding variables such as comorbidities and family background, stuttering was not a significant contributor to educational attainment (McAllister et al. 2012; Rees & Sabia, 2014).

Similar findings were observed in qualitative research. Participants in one study reported that stuttering had a negative effect on academic performance (Silverman & Zimmer, 1982). Another study contained reports of being marked down for not attending university seminars, and even choosing university courses with less interaction (Butler, 2013). The impact on educational attainment was mixed, with some studies reporting high levels of tertiary completion for adults who stutter (Leko Krhen et al., 2021; Silverman & Zimmer, 1982). Participants in other studies reported low educational achievement and leaving school prematurely (Butler, 2014; Crichton-Smith, 2002; Johnson, 1934).

Employment

Stuttering impacted employment for a range of measures, including job performance, occupational choice, unemployment, underemployment, workplace discrimination, promotion, and income. From three reports, a range of 69–86% of respondents stated that stuttering interfered with their job performance (Klein & Hood, 2004; McClure & Yaruss, 2003; Rice and Kroll, 2006). Another report stated that stuttering severity had a medium to high correlation with self-rated job difficulties ($p < .01$) (Iimura & Miyamoto, 2022). Two reports indicated that approximately 50% of respondents stated that stuttering had affected their occupational choice (Hayhow et al., 2002; Rice & Kroll, 2006), with a similar proportion in two other reports stating that finding employment was difficult (Blumgart et al., 2010a; Boyle, 2018). A workforce analysis comparing adults who stutter with controls reported significant differences between the groups in terms of choice of industry (Plexico et al., 2019); it was also reported that there were more adults who stutter than controls in the information industry ($p = .008$) and fewer in sales or service roles ($p = .02$).

An assessment of United States labor market outcomes found no significant difference between adults who stutter and controls in terms of the number of people who were employed in the labor force, underemployed, or receiving public assistance after controlling for confounding (Gerlach et al., 2018). However, a similar study conducted in the United Kingdom found that, although stuttering at age 16 was not a significant predictor of unemployment or socioeconomic class of occupation at 23, it was a significant predictor of socioeconomic class of occupation at 50 ($p = .047$) (McAllister et al., 2012).

In a report by Boyle (2018), 70% of respondents indicated discrimination in the workplace (Boyle, 2018). The workforce analysis by Plexico et al. (2019) demonstrated that adults who stutter reported higher levels of discrimination than controls ($p < .01$). In another report, 8% of respondents said their employment was terminated due to their stuttering (Blumgart et al., 2010a). A further two reports described discrimination in employment (Hayhow et al., 2002; Rice & Kroll, 1994). Reports indicate that 20–27% of respondents turned down a job or promotion, and 28–38% were denied a promotion due to their stuttering (Blumgart et al., 2010a; Klein & Hood, 2004; Rice & Kroll, 2006). The workforce analysis by Plexico et al. reported significantly fewer adults who stutter than controls in management roles ($p = .04$) and supervision ($p = .01$) roles. Craig and Calver (1991) found that within 10 months of completing a speech treatment program, 44% of participants who stutter who were eligible for promotion. Evidence for the impact of stuttering on income is mixed, Gerlach et al. (2018) reporting that stuttering is associated with an annual income deficit of at least US$9,054 ($p < .05$) and McAllister et al. (2012) reporting that stuttering at 16 years old is not a significant predictor of income at 23 or 50 years. The workforce analysis by Plexico et al. reported that adults who stutter had less income than controls ($p < .05$).

Many participants in qualitative studies reported that stuttering had affected their working lives (Crichton-Smith, 2002; Georges, 2017; Silverman & Zimmer, 1982). In a report by Nang et al. (2018), all the women interviewed indicated that stuttering limited their ability to secure work and perform work-related tasks. There were several other reports of stuttering influencing occupational choice (Butler, 2014; Crichton-Smith, 2002; Georges, 2017; Johnson, 1934). There were also reports of discrimination (Nang et al., 2018), challenges in recruitment (Butler, 2013; Crichton-Smith, 2002), and challenges in promotion (Bricker-Katz et al., 2013). However, in one report by Leko Krhen et al. (2021), none of the women indicated that stuttering had affected their occupation choice or presented significant professional challenges.

Social

Stuttering also has a social cost, affecting individuals through bullying, stigma, and social exclusion, with the potential to negatively impact friendships and life relationships. There are reports indicating that 53–77% of respondents were teased or bullied and 20–55% were avoided by others or excluded from social situations (Boyle, 2018; Erickson & Block, 2013). Boyle (2018) reported that 58–81% of respondents experienced other types of enacted stigma, including discrimination: treated unfairly, considered as inferior, considered less seriously, or being patronized. A reported personal effect of bullying can be a difficulty in forming relationships (Hayhow et al., 2002; Hugh-Jones & Smith, 1999). In the Hayhow et al. (2002) report, 61% of respondents reported that stuttering had impacted their life relationships, and 64% of respondents reported that stuttering had impacted their friendships. An analysis of school children by Berchiatti et al. (2020) reported that children who stutter were less popular (*p* < .01) and more rejected (*p* < .001) in the peer group than control children.

Thematic results of qualitative research support these findings. Reponses suggest that stuttering can lead to anxiety, avoidance, embarrassment, and frustration in those who stutter and their life partners (Beilby et al., 2013). Some respondents reported problems in life relationships (Georges, 2017). One report noted the emotional impact on life partners (Boberg & Boberg, 1990). However, a key theme was the importance of supportive partners, many of whom were not bothered by stuttering (Beilby et al., 2013; Leko Krhen et al., 2021; Nang et al., 2018).

Discussion

General study limitations

Interpretation of the present results requires consideration of their limitations. Results cannot be generalized to non-English literature, because only English language publications were searched. As with all systematic reviews, results were limited by the comprehensiveness of the databases searched. For the present review, 14 databases were searched, and Open Grey was searched for unpublished data. Additionally, manual searches of the reference lists of all identified studies served as a cross-check. However, it is conceivable that some data published in textbooks and presented in unpublished conference proceedings may have been overlooked. The sparse finding of only three studies identified in the quality of life search needs to be considered in light of two of them being classified as having a high risk of bias. This limits the extent to which their results can be considered admissible. The exclusion of participants with diagnosed comorbid disorders could also be seen as a limitation. This exclusion criterion was intended to clarify the findings for stuttering alone. However, stuttering and comorbidity is, of course, of great interest for health economics research and requires further exploration.

Quality of life

Summary of results

This systematic review involved a full-text review of 135 articles, and only three provided standard utility values that could be incorporated into health economics analyses. Baseline utility scores for stuttering adult groups in two studies ranged from 0.71–0.83, and for preschool children in one study, they were 0.88. Two of the three studies had a high risk of bias. Post-intervention improvements in utility values were reported in the range of 0.06–0.10 for two studies, and a slight worsening was reported in one study. No study reported a statistically significant difference in utility scores between treatment groups.

Instrument limitations

Age restrictions for the quality of life test instruments limit the usefulness of results identified in this review. The cost utility analysis based on HUI3 utilities in de Sonneville-Koedoot et al. (2015) is limited by the number of participants 5 years of age or older for whom the instrument was applicable, which was 25% of total participants. The cost utility analysis based on transformed EQ-VAS scores—a visual analogue scale from 0 to 100—used by de Sonneville-Koedoot et al. is not supported by a published mapping algorithm. The Child Health Utility 9 dimensions (CHU-9D) is an alternative quality of life instrument appropriate for children 4–17 years of age (Rowen et al., 2020). Using the CHU-9D instead of HUI3 by de Sonneville-Koedoot et al. would have expanded the cohort with applicable utility scores to children older than 4 years of age, increasing the sample size with utility data to 120 participants (60% of the study total).

There is currently no generic quality of life instrument appropriate for children younger than 4 years of age (Rowen et al., 2020). However, the Pediatric Quality of Life Inventory measures quality of life in children 2 years of age and older, and it could be used in stuttering research to maximize the amount of quality of life data available for economic evaluation.

Test instrument sensitivity is also a limitation of the quality of life assessments identified in this review. The EQ-5D and HUI3 consider psychological distress; however, both were found to be less sensitive to mental health quality of life than other instruments, such as Assessment of Quality of Life–Eight Dimension Scale (Mihalopoulos et al., 2014).

Conversion of instruments to utility measures

It is possible that two of the quality of life instruments excluded during this review could be converted into utility measures: EQ-VAS and SF-36 (the 36-Item Short Form Health Survey). Although, a recent review of mapping algorithms that link results from quality of life instruments to EQ-5D utility values did not identify a method to convert EQ-VAS to EQ-5D (Dakin et al., 2018). However, a scoring system linking the visual analogue scale to the utility measure of the EQ-5D youth version (EQ-5D-Y) exists (Wu, 2014), which suggests that conversion of EQ-VAS scores to utility measures is possible. The results of SF-36 can be mapped to multiple generic utility measures, including HUI3 and EQ-5D (Brazier et al., 2010). Given that 21 studies evaluated the impact of stuttering interventions on quality of life using the Quality of Life scale of the Overall Assessment of the Speaker's Experience of Stuttering (OASES), mapping OASES to EQ-5D or another generic quality of life measure would facilitate the inclusion of valuable existing research in future economic evaluations.

Interpretation of reported stuttering utility values

The baseline utility scores for the two studies with adults (McAllister et al., 2017; Omori et al., 2021) were limited by low participant numbers. McAllister et al. (2017) reported utility scores for 22 participants, and Omori et al. (2021) reported utility scores for 12 participants. With the caveat of limited sampling, those data did substantiate an earlier suggestion (Craig et al., 2009) that the quality of life impact of stuttering is similar to other conditions that are of prime concern to health care provision. Stuttering utility scores for adults in the range of 0.71–0.83 are broadly consistent with values associated with utility scores for some conditions reported in another systematic review (Zhou et al., 2021). For example, Zhou et al. (2021) reported a meta-analytic utility estimate of 0.83 for diabetes mellitus, and pooled values of 0.77 for cardiovascular disease, 0.75 for cancer, 0.84 for HIV, and 0.70 for chronic kidney disease.

Sampling was more substantive in the de Sonneville-Koedoot et al. (2015) report for preschoolers, although, as noted above, it was limited to participants 5 years of age or older for whom standard utility scores were available. Baseline utility scores for children (mean = 0.88) were higher than for adults, reflecting less quality of life impact. It is intuitively correct that quality of life impact is lower for preschoolers than adults. This is because shortly after stuttering potentially debilitating factors of mental health and educational and occupational disadvantage would have had limited influence.

Interpretation of treatment results

The Omori et al. (2021) study involved basic stuttering control training for both groups, with the intervention group receiving a 7-week CBT package. Results were encouraging after 4 months, with the intervention group increasing its utility score by 0.10 and the standard care group by 0.06. Arguably, that was a conservative estimate, considering that the standard stuttering control training was limited to “20-minute speech therapy sessions once every three or four weeks for four months” (p. 59). With preschool participants in the de Sonneville-Koedoot et al. (2015) report for whom standard utility scores were available, results are a little more convincing, with similar utility score improvements in both intervention groups at 18 months follow-up: 0.07 for Lidcombe Program and 0.06 for RESTART-DCM. However, arguably again, those results were conservative estimates because at follow-up, 28% of childen in the Lidcombe Program arm and 35% in the RESTART-DCM arm had not completed their treatments (Franken, 2016). The de Sonneville-Koedoot et al. report did not involve a no-treatment control group, which makes it unclear to what extent follow-up improvements were due to natural recovery (Bergþórsdóttir & Ingham, 2017).

Cost of illness

Summary of results

Overall, this body of research carries considerable risk of bias. Regardless, it seems that those who stutter—children and adults— consume more health care resources than their non-stuttering peers, including those relating to hospitalisations, outpatient visits, consultations with mental health professionals, and medications. Many of the results in the reviewed reports were not statistically significant. However, that could be attributed to study designs, in particular, limited controls for confounding variables. The direct, overall costs of treatments obtained for the adult population are in the vicinity of US$1,000 per year, and the total costs of well-known early interventions are around US$4,000-4,500. There is some evidence of financial strain on families with adolescents who are receiving treatment. One report considered willingness to pay for stuttering interventions. Respondent willingness to pay for treatment to improve or cure stuttering was quantified as two to four times their annual income.

Stuttering appears to be associated with lower academic performance in childhood and may contribute to reduced attainment of tertiary qualifications. Children who stutter are more likely than their peers to require special education services. They are twice as likely to not attend school days, apparently because of their stuttering, and this has been found to be related to bullying. It is possible that there is an inverse relationship between stuttering severity and educational attainment, and stuttering has been linked to leaving school prematurely. However, those findings are potentially confounded by uncontrolled variables. One report suggested that tertiary educators may overcorrect performance assessments to account for stuttering. The authors of that report note that such overcorrection may ultimately have a negative impact and limit long-term academic achievement.

There is clearly a relationship between stuttering and employment, where those who stutter are limited in terms of income, promotion, and socioeconomic status of occupation. Those limitations may affect women particularly. Stuttering is reported to affect occupational choice, and it is associated with workplace discrimination. There is some evidence associating stuttering with reduced income.

There are social costs associated with stuttering, involving bullying, stigma, and social exclusion. Stuttering has the potential to negatively impact friendships and life relationships. Children who stutter are less popular and rejected more often than their non-stuttering peers. Stuttering can lead to anxiety, avoidance, embarrassment, and frustration in those who stutter and their life partners. Supportive life partners assist those who stutter to deal with the condition.

Conclusions

In health economics terms, stuttering has substantive direct and indirect costs of illness. It is likely to cause impairment, challenges, and distress across many domains throughout life, including income, education, employment, and social functioning. There is some evidence that, in health economics terms, stuttering causes impairment of health-related quality of life and that it can be improved by treatment. However, that evidence is extremely limited and is derived from only 85 participants across three studies. This evidence gap limits economic evaluation and cost-utility analyses of stuttering interventions.

There is a growing body of clinical trials evidence to support treatments for stuttering. For adult and adolescents, variations of speech restructuring have been evaluated in more than 30 clinical trials (Brignell et al., 2020). There are eight randomized controlled trials of the Lidcombe Program for children younger than 6 years (Sjøstrand et al., 2021). Additionally, there are five successful clinical trials of CBT specifically for the social anxiety of those who stutter (Gunn et al., 2019; Helgadóttir et al., 2014; Menzies et al., 2008, 2009, 2019). Yet, to date, that evidence has been overlooked for its capacity to improve quality of life for children and adults who stutter.

Eleven instruments pertinent to quality of life were excluded from the analysis because they did not generate a utility measure. Most strikingly, the Overall Assessment of the Speakers Experience of Stuttering, with its Quality of Life subscale, was presented in 21 reports but was excluded from this review. Because of this, the literature to date contains much potentially useful information that cannot be used to inform cost-utility analyses of stuttering interventions. Without utility values, treatment for the impairment, challenges, and distress experienced by those who stutter cannot be presented in a cost-utility analysis, conveying cost per quality-adjusted life year. This means that the health-related quality of life impairment of those who stutter is not documented in a form that can be used to influence health policy and healthcare spending. That problem could be remedied if future clinical trials of stuttering treatment routinely incorporate a health-related quality of life measure that can be converted to a utility score. That course of action is our recommendation from this systematic review.

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Figure Captions

Figure 1. Results of the quality of life search strategy.

Figure 2. Results of the cost of illness search strategy.

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Figure 2. Results of the quality of life search strategy.

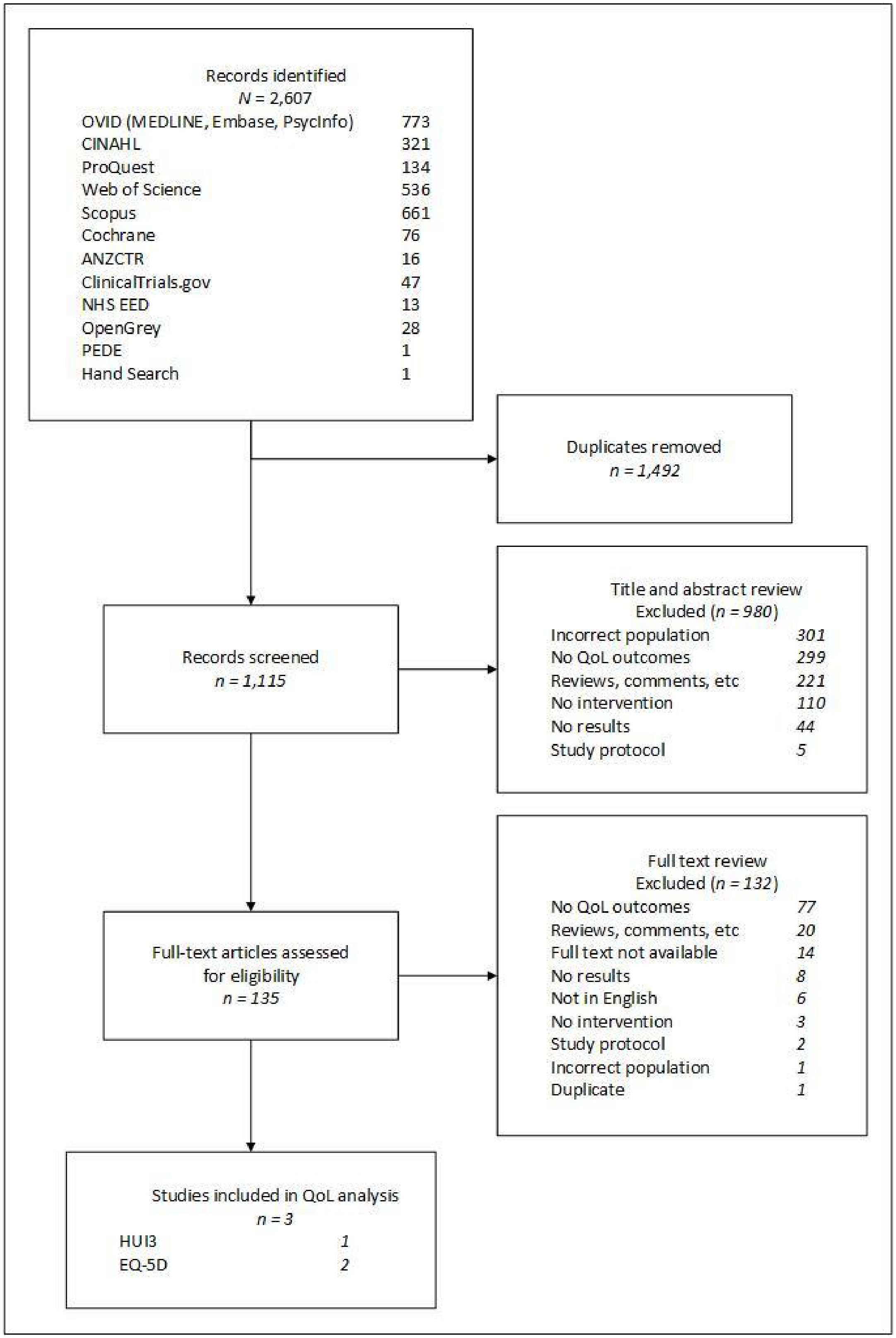
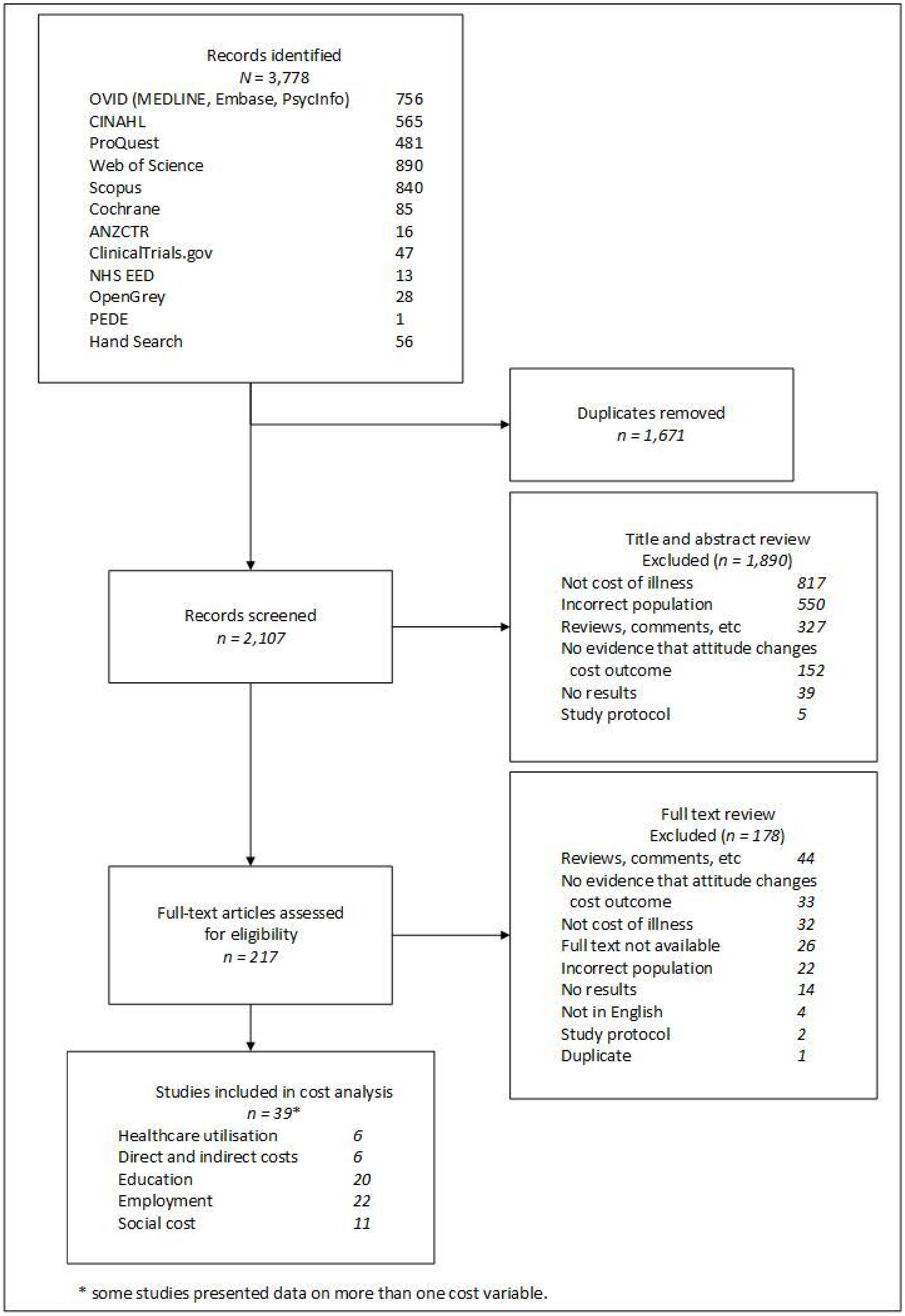


Figure 2. Results of the cost of illness search strategy.



* some studies presented data on more than one cost variable.

Table 1. Instruments excluded from the review because they do not generate a utility measure

Instrument	Pertinence to quality of life
Rosenberg Self-Esteem Scale	N
Modified Erickson Scale of Communication Attitude	N
Self-Efficacy Scaling for Adult Stutterers	N
Revised Communication Attitude Inventory (S24)	N
Stuttering Severity Instrument (SSI-3 & SSI-4)	N
Perception of Stuttering Inventory	N
Beck Anxiety Inventory	N
Subjective Screening of Stuttering	N
Clinician-based Global Impression	N
Overall Assessment of the Speakers Experience of Stuttering–Quality of Life subscale	Y
Assessment of the Child's Experience of Stuttering	Y
Self-Assessment Protocol–version for adults	Y
Satisfaction with Life Scale	Y
EuroQol-visual analogue scale (EQ-VAS)	Y
Therapy Outcome Measure	Y
Wright and Ayre Stuttering Self-Rating Profile	Y
Strength and Difficulties Questionnaire	Y
36-Item Short Form Survey (SF-36)	Y
Quality of Life Assessment Schedule	Y
Visual Analog Rating Scale of Health-Related Quality of Life (VAS)	Y

Table 2. Summary of studies included in the quality of life analysis

Author (Year)	Region	Treatment Setting	Study design	Size (*N*)	Cohort	Male (%)	Population characteristics (stuttering diagnosis, severity, treatment)	Intervention & Comparator	Outcomes (utility measure)	Risk of bias
de Sonneville-Koedoot et al. (2015)	The Netherlands	Outpatient and home-based	Multi-centre parallel group RCT	199	Children	Lidcombe Program: 69.4 RESTART-DCM: 70.0	Stuttering severity rating ≥ 2 (‘mild’) and stuttered at least 3% of syllables.	Two stuttering interventions: Lidcombe Program and RESTART-DCM	HUI3	High
McAllister et al. (2017)	Britain	Online	Two-group parallel design (treatment vs placebo), double-blinded feasibility study.	31	Adults	Treatment: 94 Placebo: 67	Individuals who stutter and have social anxiety disorder.	Cognitive bias modification using neutral/disgusted faces and letter prompts. Intervention: letter replaces neutral face. Comparator: letter replaces neutral or disgusted face	EQ-5D-3L	Low
Omori et al. (2021)	Japan	Outpatient and home-based	Single-centre parallel design (CBT + speech therapy vs speech therapy), non-randomised pilot study.	12	Mixed (age 12 to 65 years)	Treatment: 71.4 Control: 60.0	Meet criteria of childhood-onset fluency disorder (DSM-5) after being diagnosed by an otolaryngologist	Intervention: 7 guided self-hep CBT sessions + 4 speech therapy sessions (fluency shaping method) Control: 4 speech therapy sessions (fluency shaping method)	EQ-5D-5L	High

CBT = Cognitive Behavior Therapy; DCM = Demands and Capacities Model; DSM-5 = Diagnostic and Statistical Manual of Mental Disorders, 5th Edition; EQ-5D-3L = EuroQol-5 Dimension, 3 level; HUI3 = Health Utilities Index 3; RCT = Randomised Controlled Trial.

Supplemental Material S1 – Search Terms

Search term mapping

Embase subject headings were mapped to subject headings or topics in CINAHL, ProQuest, and Web of Science (Tables S1 and S2).

Title/abstract/keyword searches were used in place of subject heading searches in Scopus, PEDE, NHS EED, and the Cochrane Library, which do not use subject headings or topics to index records. Word variations were searched on all lines of the Cochrane Library search. PEDE, NHS EED, ANZCTR, ClinicalTrials.gov, and OpenGrey had limited search functionality and a small number of results. Searches of these databases were truncated after combining the population search terms and results exported for title/abstract review.

Table S1: Quality of life search term mapping from Embase to CINAHL, ProQuest and Web of Science

Embase ‘subject heading’	CINAHL ‘subject heading’	ProQuest ‘mainsubject’	Web of Science ‘topic’
Fluency disorder	Fluency Disorders	No subject heading	Fluency disorder*
Stuttering	No subject heading	Stuttering	Stuttering
Therapy	Therapeutics+	Therapy	Therapy
Clinical outcome	No subject heading	Clinical outcomes	Clinical outcome*
Outcome variable	Dependent Variable	Clinical outcomes	Outcome variable*
Outcome assessment	Outcome Assessment	Clinical outcomes	Outcome assessment*
Treatment outcome	Treatment Outcome+	Clinical outcomes	Treatment outcome*
Patient-reported outcome	Patient-Reported Outcomes+	No subject heading	Patient-reported outcome* OR Patient reported outcome*
Quality of life	Quality of Life+	Quality of life	Quality of life
Quality adjusted life year	Quality-Adjusted Life Years	No subject heading	Quality-adjusted life year* or Quality adjusted life year*

Table S2: Quality of life search term mapping from Embase to CINAHL, ProQuest and Web of Science

Embase ‘subject heading’	CINAHL ‘subject heading’	ProQuest ‘mainsubject’	Web of Science ‘topic’
Fluency disorder	Fluency Disorders	No subject heading	Fluency disorder*
Stuttering	No subject heading	Stuttering	Stuttering
Disease burden	No subject heading	No subject heading	Disease burden
Caregiver burden	Caregiver Burden	No subject heading	Caregiver burden
Health care cost	Health Resource Utilization OR Health Care Costs+	Health care expenditures	Health care cost* OR Healthcare cost*
Cost	No subject heading	Cost	Not used [c]
Cost of illness	Economic Aspects of Illness	Cost of illness	Cost of illness
Education	Outcomes of Education [a]	Academic achievement OR Academic underachievement OR Education attainment [b]	Outcomes of education OR Education outcomes OR Academic achievement OR Academic underachievement OR Educational attainment [d]
Occupation	Occupations and Professions+	Occupations	Occupation
Employment	Employment+	Employment	Employment
Attitude	Attitude+	Attitudes	Not used [c]
Personnel management	Personnel Management+	Personnel management	Not used [c]
Mental Health	Mental Health	Mental health	Mental health
Human relation	No subject heading	Human relations	Human relation*
Productivity	Productivity	Productivity	Productivity
Social aspect	No subject heading	No subject heading	Social aspect*

[a] Education+ subject heading expanded; outcomes of education selected from list of sub-headings

[b] Education subject heading expanded; academic achievement, academic underachievement, and education attainment selected from list of sub-headings

[c] Topic returned many irrelevant results. Title/abstract used instead.

[d] Education topic expanded; outcomes of education, education outcomes, academic achievement, academic underachievement, and educational attainment selected as topics, consistent with CINAHL and ProQuest subject headings.

Quality of Life search terms

Tables S3 to S8 detail the quality of life search terms used for Embase, CINAHL, ProQuest, Web of Science, Scopus, and the Cochrane Library.

Table S3: Quality of Life search terms used in Embase

Databases: Embase (1974 to 2021 January 15), Ovid MEDLINE ® ALL (1946 to January 15, 2021) & APA PsycInfo 1806 to January Week 2 2021		
Search date: 20-Jan-21		
#	**Search terms**	**Results**
1	fluency disorder/ or stuttering/	13,680
2	(stuttering or stammering or disfluency).kw. or (stuttering or stammering or disfluency).tw.	14,504
3	1 or 2	17,350
4	intervention.kw. or intervention.tw.	1,844,627
5	treat*.kw. or treat*.tw.	14,205,344
6	therapy/	1,328,181
7	4 or 5 or 6	16,037,827
8	clinical outcome/ or outcome variable/ or outcome assessment/ or treatment outcome/ or patient-reported outcome/	2,602,676
9	quality of life/	737,668
10	quality adjusted life year/	40,878
11	health related outcome*.tw. or health related outcome*.kw.	5,267
12	8 or 9 or 10 or 11	3,235,601
13	3 and 7 and 12	751
14	remove duplicates from 13	516

Explanatory notes: / = subject heading; * = unlimited truncation (i.e., unlimited suffix variations); (…) = limiter; kw = keyword heading; tw = text word (includes title, abstract, and drug trade name).

Table S4: Quality of Life search terms used in CINAHL

Databases: CINAHL Complete		
Search date: 22-Feb-21		
#	**Search terms**	**Results**

1	(MH "Fluency Disorders")	3,166
2	TI stuttering or fluency disorder or stammering or dysfluency	1,486
3	AB stuttering or fluency disorder or stammering or dysfluency	1,487
4	1 or 2 or 3	3,424
5	TI intervention or AB intervention	424,759
6	TI treat* or AB treat*	1,080,074
7	(MH "Therapeutics+")	1,585,326
8	5 or 6 or 7	2,541,569
9	TI clinical outcome or AB clinical outcome	75,289
10	(MH "Dependent Variable")	2,957
11	(MH "Outcome Assessment") or (MH "Treatment Outcomes+") or (MH "Patient-Reported Outcomes+")	428,083
12	(MH "Quality of Life+")	125,305
13	(MH "Quality-Adjusted Life Years")	5,025
14	TI health related outcome* or AB health related outcome*	5,087
15	9 or 10 or 11 or 12 or 13 or 14	587,035
16	4 and 8 and 15	314

Explanatory notes: * = truncation wildcard (i.e., unlimited suffix variations); + = explode subject heading; (…) = limiter; "…" = exact phrase; AB = abstract; MH = exact subject heading; TI = title.

Table S5: Quality of Life search terms used in ProQuest

Databases: ProQuest		
Search date: 26-Feb-21		
#	**Search terms**	**Results**
1	MAINSUBJECT.EXACT("stuttering")	6,150
2	AB(stuttering or stammering or disfluency) or TI(stuttering or stammering or disfluency)	13,878
3	1 or 2	15,128
4	AB(intervention) or TI(intervention)	2,054,834
5	AB(treat*) or ti(treat*)	12,202,146
6	MAINSUBJECT.EXACT("therapy")	77,618
7	4 or 5 or 6	13,696,514
8	MAINSUBJECT.EXACT("Clinical outcomes")	47,463
9	AB(patient reported outcome) or TI(patient reported outcome)	240,347

10	MAINSUBJECT.EXACT("Quality of life")	354,124
11	AB(quality adjusted life year) or TI(quality adjusted life year)	28,738
12	AB(health related outcome) or TI(health related outcome)	152,245
13	8 or 9 or 10 or 11 or 12	744,537
14	3 and 7 and 13	123

Explanatory notes: * = truncation (up to 500 word variations); (…) = limiter; "…" = exact phrase; AB = abstract; MAINSUBJECT.EXACT = exact subject heading; TI = title.

Table S6: Quality of Life search terms used in Web of Science

Database: Web of Science (All years, 1864-2021)		
Search date: 27-Feb-21		
#	**Search terms**	**Results**
1	TS=("fluency disorder*") or TS=(stuttering)	9,146
2	TI=(stuttering or stammering or disfluency) or AB=(stuttering or stammering or disfluency)	9,293
3	1 or 2	10,434
4	TI=(intervention) or AB=(intervention)	1,441,805
5	TI=(treat*) or AB=(treat*)	9,534,163
6	TS=(therapy)	11,596,190
7	4 or 5 or 6	17,618,885
8	TS=("clinical outcome*" or "outcome variable*" or "outcome* assessment*" or "treatment outcome*" or "patient-reported outcome*" or "patient reported outcome*")	1,382,075
9	TS=("quality of life")	604,326
10	TS=("quality adjusted life year*" or "quality-adjusted life year*")	21,227
11	TI=("health related outcome*") or AB=("health related outcome*")	2,350
12	8 or 9 or 10 or 11	1,895,176
13	3 and 7 and 12	496

Explanatory notes: * = wildcard for right hand truncation (any group of suffix characters, including no character); (…) = limiter to override operator precedence; "…" = exact phrase; AB = abstract; TI = title; TS = topic.

Table S7: Quality of Life search terms used in Scopus

Database: Scopus (All years)

Search date: 02-Mar-21		
#	**Search terms**	**Results**
1	TITLE-ABS-KEY ("fluency disorder*" or stuttering)	8,230
2	TITLE-ABS (stuttering OR stammering or disfluency)	7,421
3	1 or 2	9,469
4	TITLE-ABS (intervention)	1,360,735
5	TITLE-ABS (treat*)	8,598,971
6	TITLE-ABS-KEY (therapy)	4,659,707
7	4 or 5 or 6	11,885,688
8	TITLE-ABS-KEY ("clinical outcome*" or "outcome variable*" or "outcome* assessment*" or "treatment outcome*" or "patient-reported outcome*" or "patient reported outcome*")	2,015,330
9	TITLE-ABS-KEY ("quality of life")	539,489
10	TITLE-ABS-KEY ("quality adjusted life year" or "quality-adjusted life year")	25,642
11	TITLE-ABS ("health related outcome*")	2,207
12	8 or 9 or 10 or 11	2,245,159
13	3 and 7 and 12	616

Explanatory notes: * = wildcard; (…) = limiter; "…" = loose phrase (exact words, ignores punctuation); TITLE-ABS = article title or abstract; TITLE-ABS-KEY = article title or abstract or keywords.

Table S8: Quality of Life search terms used in the Cochrane Library

Database: Cochrane Library (including Cochrane Database of Systematic Reviews and Cochrane Central Register of Controlled Trials)		
Search date: 20-May-21		
#	**Search terms**	**Results**
1	("fluency disorder" or stuttering):ti,ab,kw	276
2	(stuttering or stammering or disfluency):ti or (stuttering or stammering or disfluency):ab	271
3	1 or 2	291
4	intervention:ti,ab	422,259
5	treat*:ti,ab	806,531
6	therapy:ti,ab,kw	710,888
7	4 or 5 or 6	1,212,754

8	("clinical outcome*" or "outcome variable*" or "outcome* assessment*" or "treatment outcome*" or "patient-reported outcome*" or "patient reported outcome*"):ti,ab,kw	267,756
9	"quality of life":ti,ab,kw	115,018
10	("quality adjusted life year" or "quality-adjusted life year"):ti,ab,kw	0
11	("health related outcome*"):ti,ab	429
12	8 or 9 or 10 or 11	0
13	8 or 9 or 11	348,994
14	3 and 7 and 12	0
15	3 and 7 and 13	69

Explanatory notes: * = wildcard to search zero or more characters; (…) = nesting to change search precedence; "…" = phrase search; ab = abstract; kw = keyword; ti = title.

Cost of illness search terms

Tables S9 to S14 detail the cost search terms used for Embase, CINAHL, ProQuest, Web of Science, Scopus, and the Cochrane Library.

Table S9: Cost search terms used in Embase

Databases: Embase (1974 to 2021 January 15), Ovid MEDLINE ® ALL (1946 to January 15, 2021) & APA PsycInfo 1806 to January Week 2 2021		
Search date: 19-Jan-21		
#	**Search terms**	**Results**
1	fluency disorder/ or stuttering/	13,680
2	(stuttering or stammering or disfluency).kw. or (stuttering or stammering or disfluency).tw.	14,504
3	1 or 2	17,350
4	economic.kw. or economic.tw.	614,353
5	health resource*.kw. or health resource*.tw.	18,082
6	disease burden/ or caregiver burden/	36,056
7	economic burden.kw. or economic burden.tw.	32,765
8	emotional burden.kw. or emotional burden.tw.	2,482
9	"health care cost"/	245,531
10	"cost"/	108,319

11	cost.kw. or cost.tw.	1,126,791
12	"cost of illness"/	47,767
13	education/	480,763
14	occupation/ or employment/	187,403
15	attitude/ or personnel management/	199,338
16	mental health/	256,431
17	human relation/	89,318
18	productivity/	57,123
19	social aspect/	78,028
20	4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19	3,045,646
21	3 and 20	662
22	Remove duplicates from 21	518

Explanatory notes: / = subject heading; * = unlimited truncation (i.e., unlimited suffix variations); (…) = limiter; "…" = literal string (i.e., an exact phrase); kw = keyword heading; tw = text word (includes title, abstract, and drug trade name).

Table S10: Cost search terms used in CINAHL

Database: CINAHL		
Search date: 22-Feb-21		
#	**Search terms**	**Results**
1	(MH "Fluency Disorders")	3,166
2	TI stuttering or fluency disorder or stammering or dysfluency	1,486
3	AB stuttering or fluency disorder or stammering or dysfluency	1,487
4	1 or 2 or 3	3,424
5	TI economic or AB economic	70,268
6	(MH "Health Resource Utilization")	19,592
7	TI health resource* or AB health resource*	18,217
8	TI disease burden or AB disease burden	14,159
9	(MH "Caregiver Burden")	10,172
10	TI caregiver burden or AB caregiver burden	4,247
11	TI economic burden or AB economic burden	4,958
12	TI emotional burden or AB emotional burden	987
13	(MH "Health Care Costs+")	61,717
14	TI cost or AB cost	163,546

15	(MH "Economic Aspects of Illness")	10,072
16	(MH "Education+")	948,231
17	(MH "Outcomes of Education")	14,151
18	(MH "Occupations and Professions+") or (MH "Employment+")	143,243
19	(MH "Attitude+") or (MH "Personnel Management+")	715,705
20	(MH "Mental Health")	41,151
21	TI human relation or AB human relation	969
22	(MH "Productivity")	11,248
23	TI social aspect OR AB social aspect	4,499
24	5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 18 or 19 or 20 or 21 or 22 or 23	1,741,435
25	4 and 24	1,392
26	5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 17 or 18 or 19 or 20 or 21 or 22 or 23	1,103,452
27	4 and 26	534

Explanatory notes: * = truncation wildcard (i.e., unlimited suffix variations); + = explode subject heading; (…) = limiter; "…" = exact phrase; AB = abstract; MH = exact subject heading; TI = title.

Table S11: Cost search terms used in ProQuest

Database: ProQuest		
Search date: 26-Feb-21		
#	**Search terms**	**Results**
1	MAINSUBJECT.EXACT("stuttering")	6,150
2	AB(stuttering or stammering or disfluency) or TI(stuttering or stammering or disfluency)	13,878
3	1 or 2	15,128
4	AB(economics) or TI(economics)	368,891
5	AB(health resource) or TI(health resource)	307,910
6	AB(disease burden) or TI(disease burden) or AB(caregiver burden) or TI(caregiver burden)	185,051
7	AB(economic burden) or TI(economic burden)	56,629
8	AB(emotional burden) or TI(emotional burden)	9,914
9	MAINSUBJECT.EXACT("health care expenditures")	23,951
10	MAINSUBJECT.EXACT("cost")	17,285
11	AB(cost) or TI(cost)	4,449,850

12	MAINSUBJECT.EXACT("cost of illness")	30,643
13	MAINSUBJECT(education)	3,101,730
14	MAINSUBJECT.EXACT("Academic achievement") or MAINSUBJECT.EXACT("Academic underachievement") or MAINSUBJECT.EXACT("Educational attainment")	156,203
15	MAINSUBJECT(Occupations) or MAINSUBJECT(Employment)	617,811
16	MAINSUBJECT.EXACT("attitudes") or MAINSUBJECT.EXACT("personnel management")	163,449
17	MAINSUBJECT.EXACT("mental health")	246,652
18	MAINSUBJECT.EXACT("human relations")	15,322
19	MAINSUBJECT.EXACT("productivity")	234,137
20	AB(social aspect) or TI(social aspect)	72,028
21	4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 15 or 16 or 17 or 18 or 19 or 20	10,258,536
22	3 and 21	818
23	4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 16 or 17 or 18 or 19 or 20	7,603,501
24	3 and 23	449

Explanatory notes: * = truncation (up to 500 word variations); (…) = limiter; "…" = exact phrase; AB = abstract; MAINSUBJECT.EXACT = exact subject heading; TI = title.

Table S12: Cost search terms used in Web of Science

Database: Web of Science (All years, 1864-2021)		
Search date: 27-Feb-21		
#	**Search terms**	**Results**
1	TS=("fluency disorder*") or TS=(stuttering)	9,146
2	TI=(stuttering or stammering or disfluency) or AB=(stuttering or stammering or disfluency)	9,293
3	1 or 2	10,434
4	TI=(economic) or AB=(economic)	1,314,049
5	TI=("health resource*") or AB=("health resource*")	8,302
6	TS=("disease burden" or "caregiver burden")	29,506
7	TI=("economic burden") or AB=("economic burden")	16,123
8	TI=("emotional burden") or AB=("emotional burden")	1,046
9	TS=("health care cost*" or "healthcare cost*")	80,388
10	TI=("cost") or AB=("cost")	1,921,726

11	TS=("cost of illness")	30,415
12	TS=("outcomes of education" or "education outcomes" or "academic achievement" or "academic underachievement" or "educational attainment")	48,068
13	TS=(occupation or employment)	460,611
14	TI=(attitude or "personnel management") or AB=(attitude or "personnel management")	493,853
15	TS=(mental health)	580,975
16	TS=("human relation*")	9,504
17	TS=(productivity)	548,342
18	TS=("social aspect*")	60,479
19	4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18	5,046,284
20	3 and 19	807

Explanatory notes: * = wildcard for right hand truncation (any group of suffix characters, including no character); (…) = limiter to override operator precedence; "…" = exact phrase; AB = abstract; TI = title; TS = topic.

Table S13: Cost search terms used in Scopus

Database: Scopus (All years)		
Search date: 02-Mar-21		
#	**Search terms**	**Results**
1	TITLE-ABS-KEY ("fluency disorder*" or stuttering)	8,230
2	TITLE-ABS (stuttering OR stammering or disfluency)	7,421
3	1 or 2	9,469
4	TITLE-ABS (economic)	1,335,483
5	TITLE-ABS ("health resource*")	7,926
6	TITLE-ABS-KEY ("disease burden" or "caregiver burden")	43,876
7	TITLE-ABS ("economic burden")	14,870
8	TITLE-ABS ("emotional burden")	1,168
9	TITLE-ABS-KEY ("health care cost*" or "healthcare cost*")	209,210
10	TITLE-ABS ("cost")	2,482,855
11	TITLE-ABS-KEY ("cost of illness")	34,851
12	TITLE-ABS-KEY ("outcomes of education" or "education outcomes" or "academic achievement" or "academic underachievement" or "educational attainment")	73,056

13	TITLE-ABS-KEY (occupation or employment)	443,479
14	TITLE-ABS (attitude or "personnel management")	460,999
15	TITLE-ABS-KEY (mental and health)	484,748
16	TITLE-ABS ("human relation*")	7,505
17	TITLE-ABS-KEY (productivity)	412,547
18	TITLE-ABS-KEY ("social aspect*")	115,313
19	4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18	5,364,509
20	3 and 19	761

Explanatory notes: * = wildcard; (…) = limiter; “…” = loose phrase (exact words, ignores punctuation); TITLE-ABS = abstract title or abstract; TITLE-ABS-KEY = article title or abstract or keywords.

Table S14: Cost search terms used in the Cochrane Library

Database: Cochrane Library (including Cochrane Database of Systematic Reviews and Cochrane Central Register of Controlled Trials)		
Search date: 12-Mar-21		
#	**Search terms**	**Results**
1	("fluency disorder" or stuttering):ti,ab,kw	272
2	(stuttering or stammering or disfluency):ti or (stuttering or stammering or disfluency):ab	268
3	1 or 2	287
4	(economic):ti or (economic):ab	18,767
5	("health resource"):ti or ("health resource"):ab	762
6	("disease burden" or “caregiver burden”):ti,ab,kw	3,110
7	("economic burden"):ti or ("economic burden"):ab	1,050
8	("emotional burden"):ti or ("emotional burden"):ab	100
9	("health care cost" or “healthcare cost”):ti,ab,kw	9,037
10	("cost"):ti,ab	69,665
11	("cost of illness"):ti,ab,kw	1,115
12	("outcomes of education" or "education outcomes" or "academic achievement" or "academic underachievement" or "educational attainment"):ti,ab,kw	2,231
13	(occupation or employment):ti,ab,kw	34,867
14	(attitude or "personnel management"):ti,ab	12,689
15	("mental health"):ti,ab,kw	20,867

16	("human relation"):ti,ab	30
17	(productivity):ti,ab,kw	82,265
18	("social aspect"):ti,ab,kw	762
19	4 or 5 or 6 or 7 or 8 or 9 or 10 11 or 12 or 13 or 14 or 15 or 16 or 17 18	212,907
20	3 and 19	77

Explanatory notes: (…) = nesting to change search precedence; "…" = phrase search; ab = abstract; kw = keyword; ti = title.

Search terms using population only

Tables S15 to S19 detail the truncated search terms used for the Paediatric Economic Database Evaluation (PEDE), the National Health Service Economic Evaluation Database (NHS EED), the Australian New Zealand clinical trials registry (ANZCTR), United States National Institutes of Health trial register, and Open Grey.

Table S15: Search terms used in the Paediatric Economic Database Evaluation (PEDE) database

Database: PEDE (All years, 1980 - 2019)		
Search date: 02-Mar-21		
#	**Search terms**	**Results**
1	TITLE_ABSTRACT_KEYWORDS (stuttering)	1
2	TITLE_ABSTRACT_KEYWORDS (stammering)	0
3	TITLE_ABSTRACT_KEYWORDS (disfluency)	0
4	TITLE_ABSTRACT_KEYWORDS ("fluency disorder")	0
5	1 or 2 or 3 or 4	1

Explanatory notes: (…) = limiter; "…" = phrase search.

Table S16: Search terms used in the National Health Service Economic Evaluation Database (NHS EED)

Database: NHS EED (All years, 1994 - 2015)		
Search date: 02-Mar-21		

#	Search terms	Results
1	Any field (stuttering or stammering or disfluency)	13
2	Any field ("fluency disorder")	0
3	1 or 2	13

Explanatory notes: (…) = limiter; “…” = phrase search.

Table S17: Search terms used in the Australian New Zealand clinical trials registry (ANZCTR)

Database: ANZCTR		
Search date: 20-May-21		
#	**Search terms**	**Results**
1	Health condition(s) or problem(s) studied: (stuttering)	10
2	Health condition(s) or problem(s) studied: (stammering)	0
3	Health condition(s) or problem(s) studied: (disfluency)	1
4	Health condition(s) or problem(s) studied: ("fluency disorder")	0
5	1 or 2 or 3 or 4	10

Explanatory notes: (…) = limiter; “…” = phrase search.

Table S18: Search terms used in the United States National Institutes of Health trial register (ClinicalTrials.gov)

Database: United States National Institutes of Health trial register (ClinicalTrials.gov)		
Search date: 20-May-21		
#	**Search terms**	**Results**
1	Condition or disease: (stuttering or stutters or stammering)	34
2	Condition or disease: (disfluency)	3
3	Condition or disease: ("fluency disorder")	14
4	1 or 2 or 3	45

Explanatory notes: (…) = limiter; “…” = phrase search.

Table S19: Search terms used in OpenGrey

Database: OpenGrey

Search date: 20-May-21		
#	**Search terms**	**Results**
1	stuttering	6
2	stammering	6
3	disfluency	12
4	fluency disorder	5
5	1 or 2 or 3 or 4	28

Supplemental Material S2.

Data extraction form

The data extraction form was developed, based on the Cochrane checklist of items to consider in data collection (Li et al., 2021). Relevant data from each of the included studies was extracted and recorded in Microsoft Access.

1. Study design
Study aim
Study design (e.g., cluster, cross-over)
Recruitment and sampling
Was there randomization? If yes, how was it achieved? If no, how were groups allocated?
Was there blinding?
Sequence generation and concealment
Incomplete outcome data/selective outcome reporting?
Enrolment start/end
Duration of follow-up
2. Population
Inclusion criteria
Exclusion criteria
Total number (N)
Population description (including diagnostic characteristics & disease severity)
Setting
Region/countries involved
Baseline characteristics Age Sex Comorbidities Socio-economic status Ethnicity
3. Intervention and comparator
Number of intervention groups
Intervention/comparator description Components Delivery method Timing/intensity/frequency Duration of treatment
Implementation description Format and content Staffing and equipment
Intervention/comparator integrity/fidelity/compliance
Co-interventions (for intervention and comparator arm)
4. Utility measures outcomes
Overview
Measurement instrument (e.g., CHU9D) Name of instrument Unit of measurement Upper/lower limits

Whether high or low is favourable Clinical thresholds Relevance to study aim
Metric (e.g., change pre/post intervention)
Timing of outcome measurements
Method of collection
Method of aggregation (e.g., mean & SD; proportion with condition)
Between group estimate (e.g., RR, OR, mean difference)
At each time point
Description (e.g., baseline, 6mo, 12mo)
Number of participants: In each intervention Censored (withdrawn/lost to follow-up)
Result - summary data (e.g., mean & SD)
Estimate of effect (effect size, 95%CI, *p* value)
Summary/conclusion
Is there any variation in measurement or reporting of the outcome?
Content available/appropriate for meta-analysis?
Were subgroup analyses conducted?
5. Outcomes - cost
Analytic perspective (e.g., health system)
Time horizon
Setting (i.e., study country)
Context and relevance
Cost items included, grouped as: Health sector Other sector (e.g., education) Patient and family Productivity impacts
Resource use (e.g., number of speech path visits)
Unit costs (currency, price year) (e.g., cost per speech path visit)
Discount rate
6. Miscellaneous
Key conclusions & limitations noted by the study authors
Miscellaneous comments from study authors
References to relevant studies
Generalisability of the results
Funding source
Conflicts of interest
Correspondence with study authors required?
Study quality notes (ethical approval, sample size calculation)

Supplemental Material S3.

Details of the risk of bias for the quality of life and cost studies using (i) the Rob 2 tool (Sterne et al., 2019) for randomized trials, (ii) the ROBINS-I tool (Sterne et al., 2016) for non-randomized studies, and (iii) the CASP qualitative studies checklist (Critical Appraisal Skills Programme, 2018) for qualitative research.

(i) Randomized trials

	Randomization	Deviations from intended interventions	Missing outcome data	Measurement of outcomes	Selection of reported result	Overall score
de Sonneville-Koedoot et al. (2015)	High	Low	Low	Some concerns	Low	High
McAllister et al. (2017)	Low	Low	Low	Low	Low	Low

(ii) Non-randomized trials

	Confounding	Participant selection	Classification of interventions	Deviations from intended intervention	Missing data	Measurement of outcomes	Selection of reported result	Overall score
Berchiatti et al. (2020)	High	Low	Not Applicable	Not Applicable	Low	Low	Low	High
Blumgart et al. (2010a)	Moderate	Low	Not Applicable	Not Applicable	Low	Moderate	Low	Moderate
Boulet et al. (2009)	Moderate	Low	Not Applicable	Not Applicable	Moderate	Low	Low	Moderate
Boyle (2018)	High	Low	Not Applicable	Not Applicable	High	Low	Low	High

Boyle et al. (1994)	High	Low	Not Applicable	Not Applicable	Moderate	Low	Low	High
Calnan and Richardson (1977)	High	Low	Not Applicable	Not Applicable	Low	Low	Low	High
Craig and Calver (1991)	High	Low	Low	Low	High	Low	Low	High
Erickson and Block (2013)	High	Low	Not Applicable	Not Applicable	Low	Low	Low	High
Franic et al. (2012)	High	Low	Not Applicable	Not Applicable	Moderate	Low	Low	High
Gerlach et al. (2018)	Moderate	Low	Not Applicable	Not Applicable	No Information	Low	Low	Moderate
Hayhow et al. (2002)	High	Low	Not Applicable	Not Applicable	Low	Low	Low	High
Hugh-Jones and Smith (1999)	High	Low	Not Applicable	Not Applicable	Low	Low	Low	High
Iimura and Miyamoto (2022)	High	Low	Not Applicable	Not Applicable	Moderate	Low	Low	High
Klein and Hood (2004)	High	Low	Not Applicable	Not Applicable	No Information	Low	Low	High
McAllister et al. (2012)	Moderate	Low	Not Applicable	Not Applicable	Moderate	Low	Low	Moderate
McClure and Yaruss (2003)	High	Low	Not Applicable	Not Applicable	No Information	Low	High	Critical
O’Brian et al. (2011)	High	Low	Not Applicable	Not Applicable	Low	Low	Low	High

Omori et al. (2021)	High	Low	Low	Moderate	High	High	Low	High
Palasik (2012)	High	Low	Not Applicable	Not Applicable	No Information	Low	Low	High
Plexico et al. (2019)	High	Low	Not Applicable	Not Applicable	Low	Low	Low	High
Rees and Sabia (2014)	Moderate	Low	Not Applicable	Not Applicable	Moderate	Low	Low	Moderate
Rice (1994)	High	No Information	Not Applicable	Not Applicable	No Information	Low	High	Critical
Rice and Kroll (2006)	High	Low	Not Applicable	Not Applicable	No Information	Low	Low	High
Rosenbaum (2018)	Moderate	Low	Not Applicable	Not Applicable	Moderate	Low	Low	Moderate
Sommer et al. (2021)	High	Low	Not Applicable	Not Applicable	Low	Low	Low	High
Werle and Byrd (2022)	Moderate	Low	Not Applicable	Not Applicable	Moderate	Low	Low	Moderate
Williams et al. (1969)	Moderate	Low	Not Applicable	Not Applicable	No Information	Low	Low	Moderate

(iii) Qualitative research

	Clear research aim	Appropriate methodology	Appropriate design	Appropriate recruitment	Appropriate data selection	Researcher-participant relationship considered	Ethical issues considered	Rigorous data analysis	Statement of findings	Valuable research	Overall
Beilby et al. (2013)	Yes	Yes	Yes	Unclear	Yes	Unclear	Yes	Yes	Yes	Yes	Lower

Boberg and Boberg (1990)	Yes	Yes	Unclear	Unclear	Yes	Unclear	No	Unclear	Yes	Yes	Higher
Bricker-Katz et al. (2013)	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Lower
Butler (2013)	Yes	Yes	Yes	Yes	Yes	Unclear	Yes	Yes	Yes	Yes	Lower
Butler (2014)	Yes	Yes	Yes	Yes	Yes	Unclear	Unclear	Yes	Yes	Yes	Lower
Crichton-Smith (2002)	Yes	Yes	Yes	Yes	Yes	Unclear	Yes	Yes	Yes	Yes	Lower
Georges (2017)	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Lower
Johnson (1934)	Yes	Yes	Yes	Unclear	Unclear	No	No	Unclear	Yes	Yes	Higher
Leko Krhen et al. (2021)	Yes	Yes	Unclear	No	Yes	No	Unclear	Yes	Yes	Yes	Higher
Nang et al. (2018)	Yes	Yes	Unclear	Yes	Yes	Unclear	Yes	Yes	Yes	Yes	Lower
Silverman and Zimmer (1982)	Yes	Yes	Unclear	No	Unclear	No	No	No	Yes	Yes	Higher

Supplemental Material S4.

Summary of studies included in the cost of illness analysis.

Author (Year)	Country	Study design	Size (*N*)	Cohort	Male (%)	Population characteristics (stuttering diagnosis, severity, treatment)	Cost outcomes	Risk of bias
Randomized controlled trials								
de Sonneville-Koedoot et al. (2015)	The Netherlands	Multi-centre parallel group RCT	199	Children	Lidcombe Program: 69.4 RESTART-DCM: 70.0	Stuttering severity rating ≥ 2 (‘mild’) and stuttered at least 3% of syllables.	Health care utilisation, direct and indirect costs.	High
McAllister et al. (2017)	Britain	Two-group parallel design (treatment vs placebo), double-blinded feasibility study.	31	Adults	Treatment: 94 Placebo: 67	Individuals who stutter and have social anxiety disorder.	Health care utilisation, direct costs	Low
Non-randomized studies								
Berchiatti et al. (2020)	Italy	Case-control	572 (CWS $n = 62$, CWNS $n = 474$, Teachers $n = 36$)	Children	CWS: 58.1 CWNS: 49.2	Stuttering diagnosis made by speech therapist in medical centres. CWS had prior/current formal therapy.	Education (teacher relationships; academic performance), social (friendships)	High
Blumgart et al. (2010a)	Australia	Cross-sectional [a]	200	Adults	75.5	Mean (*SD*) SS = 3.7 (2.8). Mean (*SD*) perceived stuttering severity = 4.2 (2). 94% had sought prior treatment.	Direct and indirect costs, employment	Moderate

Author (Year)	**Country**	**Study design**	**Size (*N*)**	**Cohort**	**Male (%)**	**Population characteristics (stuttering diagnosis, severity, treatment)**	**Cost outcomes**	**Risk of bias**
Boulet et al. (2009)	USA	Cross-sectional	95,132 (CWS *n* = 1,530)	Children	71.4	Parent/ guardian report based on clinical diagnosis	Health care utilisation, education (special services/early intervention)	Moderate
Boyle (2018)	USA	Cross-sectional	324	Adults	67	Self-diagnosed PWS. 95% had prior therapy	Social (discrimination), employment (recruitment)	High
Boyle et al. (1994)	USA	Cross-sectional	2,779 (CWS *n* = 297)	Children with disabilities	Not reported	Parent/ guardian report	Health care utilisation, education (school attendance and performance)	High
Calnan and Richardson (1977)	England, Scotland, and Wales	Cross-sectional	11,455 (CWS *n* = 65)	Children	Not reported	Stuttering ‘diagnosed’ in three ways: clinician assessment, teacher assessment, speech test. Results based on different diagnosis methods	Education (performance)	High
Craig and Calver (1991)	Australia	Study II: 10-month follow-up of 2 non-randomized experimental trials	62	Adults	Not reported	Craig 1984: AWS Craig 1985: AWS, mean %SS = 12.9 All participants were successfully treated with smooth speech prior to 10-month follow-up.	Employment (promotion, career improvement)	High
Erickson and Block (2013)	Australia	Cross-sectional	36	Adolescents	77.8	Mean (*SD*) stuttering frequency 6.9% (5.7%) SS, range = 2.1–26.8% Mean (*SD*) onset: 8.5 (3.0). Previously received treatment: 30 (84%). Major treatment type: speech restructuring (63% of those treated).	Education (avoid school, affected schoolwork), social (teasing/bullying, exclusion), indirect costs	High

Author (Year)	**Country**	**Study design**	**Size (*N*)**	**Cohort**	**Male (%)**	**Population characteristics (stuttering diagnosis, severity, treatment)**	**Cost outcomes**	**Risk of bias**
Franic et al. (2012)	USA	Cross-sectional	80 (included *n* = 78, history of stuttering *n* = 4)	Adults	17.9%	Not discussed	Direct and indirect costs (willingness to pay)	High
Gerlach et al. (2018)	USA	Cohort	20,745 (included *n* = 13,564, PWS *n* = 261)	Stuttered in adolescence, outcomes as adults	PWS: 64.8 PWNS: 47.5	PWS: 84% of respondents described their stuttering as “mild”; 15%, as “moderate”; and less than 1%, as “severe.”.	Employment (earnings, employment status)	Modera te
Hayhow et al. (2002)	UK	Cross-sectional	332	Adolescents and adults (age range 16–86 years)	71	Self-reported severity (10-point scale, 1=mild and 10=severe). 264 (80%) rating their stammering as “mild” (1–3) on a good day. Bad day 8% (1–3), 15% (4–5), 32% (6–7), 45% (8–10)	Education (performance), employment (occupation choice, promotion), social (friendships, romantic partners)	High
Hugh-Jones and Smith (1999)	UK	Cross-sectional	276	Adolescents or adults	75.7	Not discussed	Education (attendance, performance), employment (performance), social (relationships)	High
Iimura and Miyamoto (2022)	Japan	Cross-sectional	112 (included *n* = 110, AWS without comorbidity *n* = 52*)*	Adults	82.7	Participates in self-help group: 5% no, 18% rarely, 30% sometimes, 35% often, 12% almost always.	Employment (self-rated job difficulties)	High
Klein and Hood (2004)	USA	Cross-sectional	232	Adults	71.1	Self-rated severity: 33 (14%) very mind, 72 (31%) mild, 103 (44%) moderate, 20 (9%) severe, 4 (<2%) very severe. 91% had been enrolled in speech therapy at one time.	Employment (performance, promotion)	High

Author (Year)	Country	Study design	Size (*N*)	Cohort	Male (%)	Population characteristics (stuttering diagnosis, severity, treatment)	Cost outcomes	Risk of bias
McAllister et al. (2012)	UK	Cohort	18,558 (no stutter at age 16 and no history of speech problems *n* = 15,694, stutter at age 16 *n* = 217)	Stuttered in adolescence, outcomes as adults	PWS: 78.7 to 86.6 [b] PWNS: 47.1 to 67.6 [b]	CWS identified by parent report	Educational (attainment), employment (status and income)	Modera te
McClure and Yaruss (2003)	USA	Cross-sectional	642 (AWS *n* = 544, parents of CWS *n* = 98)	Children, adolescents, and adults	Not reported	Prior treatment: of those who had received treatment, 85% had ≥ 2 experiences. All survey respondents were NSA members, <50% had attended convention, workshop, or meeting	Education (performance), employment (performance, promotion, recruitment)	Critical
O'Brian et al. (2011)	Australia and New Zealand	Cross-sectional	147	Adults	78.9	Stuttering confirmed by SLP. 123 (81.6%) had previously received treatment for stuttering.	Education (achievement)	High
Palasik (2012)	USA	Cross-sectional	184	Adults	72.8	Self-rated stuttering severity: 51% mild, 41% moderate, 8% severe. 21% currently in therapy, 90% have had therapy at any stage	Employment (performance, career development)	High
Plexico et al. (2019)	USA (Geographic locations not equally distributed between PWS and PWNS groups *p* = .000)	Cross-sectional	164 (PWS *n* = 72, PWNS *n* = 92)	Adults	PWS: 63.4 PWNS: 34.1	On a 1-10 (worst) scale, mean (SD) severity: 5.17 (2.37) 86% had some past treatment for stuttering. 40% had received treatment ≥4 times. 74% of those receiving prior treatment experienced relapse. 60% were members of NSA.	Employment (roles, career progression, income, discrimination)	High

Author (Year)	**Country**	**Study design**	**Size (N)**	**Cohort**	**Male (%)**	**Population characteristics (stuttering diagnosis, severity, treatment)**	**Cost outcomes**	**Risk of bias**
Rees and Sabia (2014)	USA	Cohort	15,170 (analysed n = 13,549)	Stuttered in adolescence, outcomes as adults	PWS: 57.8 PWNS: 45.5	Self-reported stutter. 7% answered affirmative in Wave III ($n \approx 948$).	Education (achievement, attainment)	Moderate
Rice (1994)	Canada	Cross-sectional	>250	Not reported	Not reported	Not discussed	Employment	Critical
Rice and Kroll (2006)	USA (n = 239), Britain (n = 64), Canada (n = 32), Australia (n = 24), India (n = 13), other (n = 40). 32 countries total	Cross-sectional	412	Adults	71.6	Stuttering severity: mild (39%), moderate (54%) severe (7%).	Employment	High
Rosenbaum (2018)	USA	Case-control	9,909 enrolled in community college n = 1494, enrolled in 4-year college n = 2,721)	Adults	39.6	Self-reported stutter: 7.2% overall (n = 714), 8.1% not attending college, 7.0% attending community college, 5.5% attending 4-yr college	Education (attainment)	Moderate
Sommer et al. (2021)	Germany	Cross-sectional	27,977	Children, adolescents, and adults	75	Confirmed outpatient diagnosis (ICD-10 code recorded in insurer database)	Healthcare utilisation	High
Werle and Byrd (2022)	USA	Cross-sectional	158	College instructors	Not reported	Instructors who teach or evaluate oral presentations in university foundational oral communications courses within last 5 yrs.	Education (performance)	Moderate

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Williams et al. (1969)	USA	Case-control	1. 400 (CWS *n* = 100, CWNS *n* = 300). 2. 200 (male CWS *n* = 50, male CWNS *n* = 150)	Children	87	Names of CWS submitted by SLP	Education (performance)	Moderate
Qualitative research								
Beilby et al. (2013)	Unclear	Mixed method (questionnaires, interviews).	20	Adults	PWS: 90 Partner: 10	Clinical diagnosis of stuttering confirmed by a SLP with ≥10 years of experience in assessment and treatment of fluency disorders;	Social (romantic partner)	Lower
Boberg and Boberg (1990)	Canada	Interviews	15	Adults	0	Non-stuttering wives of PWS. 7/15 husbands completed 3wk intensive stuttering program, 3 completed precision fluency program, 3 avoidance reduction, 2 no therapy	Social (family)	Higher
Bricker-Katz et al. (2013)	Australia	Interviews	9	Adults	66.6	Stuttering severity: 2 severe, 3 moderate, 1 mod/mild, 3 mild. Treatment history: 2 current, 3 intermittent/not current, 3 never, 1 early childhood	Employment (occupational progression)	Lower
Butler (2013)	UK	Interviews, focus groups	38	Adults	Just over 80	Typically experiencing dysfluency from age 5yrs	Education (academic achievement, progression)	Lower

Author (Year)	**Country**	**Study design**	**Size (*N*)**	**Cohort**	**Male (%)**	**Population characteristics (stuttering diagnosis, severity, treatment)**	**Cost outcomes**	**Risk of bias**
Butler (2014)	UK	Interviews, focus groups, conversations	36	Adults	100	Diagnosis/ severity not discussed.	Education (achievement), employment (aspirations, recruitment)	Lower
Crichton-Smith (2002)	UK	Interviews	14	Adults	78.6	13 persistent developmental stuttering, 1 acquired stammer. Previous therapy: 2/14 never, 3/14 as child, 4/14 as adult, 5/14 as child + adult.	Education (attainment), employment	Lower
Georges (2017)	USA (*n* = 9) NZ (*n* = 1)	Interviews	10	Adults	0	Not described	Employment, social, direct and indirect costs	Lower
Johnson (1934)	USA	Mixed method (interviews, questionnaires, clinical examination, autobiographies).	80	Children, adolescents, and adults (age range 7–42 years)	76.3	Greater proportion of severe cases among boys than girls.	Education (attainment), employment (occupation choice)	Higher
Leko Krhen et al. (2021)	Croatia	Interviews	6	Adults	0	Self-report. PWS >5 yrs. Half perceived stuttering moderate at onset, remainder severe. Stutter onset: half <6rs, remainder 6–10 yrs.	Education, employment, social	Higher
Nang et al. (2018)	Australia	Interviews	9	Adults	0	Stuttering severity: 5 mild, 2 mild-moderate, 2 moderate. All started stuttering in childhood and had received some SLP intervention. Support group membership: 3 < 10yrs, 4 10-20 years, 2 > 30 years.	Social (romantic relationships), employment (recruitment)	Lower

Author (Year)	Country	Study design	Size (*N*)	Cohort	Male (%)	Population characteristics (stuttering diagnosis, severity, treatment)	Cost outcomes	Risk of bias
Silverman and Zimmer (1982)	US	Interviews	20 (Group 1 *n* = 10, Group 2 *n* = 10)	Adults	Group 1: 0 Group 2: 100	Some parent, self, teachers, relative, or friend-diagnosed (no mention of clinical diagnosis). Stutter onset: 4yrs women, 6.2yrs men. Treatment onset: 11.4yrs w, 9.8yr m.	Education, employment, social, healthcare utilisation	Higher

AWS = adults who stutter; BSA = British Stuttering Association; CWNS = children who do not stutter; CWS = children who stutter; DCM = Demands and Capacities Model; LP = Lidcombe Program; NSA = National Stuttering Association; NZ = New Zealand; PWNS = people who do not stutter; PWS = people who stutter; RCT = Randomized Controlled Trial; *SD* = standard deviation; SLP = speech-language pathologist; SS = sample size; UK = United Kingdom; USA = United States of America; yrs. = years.

[a] This study is described by the authors as a population group cohort study. However, it specifically recruited PWS so has been classified as a cross-sectional study in this review.

[b] Proportion of males was presented by outcome variable (e.g., highest qualification at 50 years, unemployment at 23 years, pay at age 23)