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#### Review

# Bilingual children who stutter: Convergence, gaps and directions for research



Ai Leen Choo<sup>a,\*</sup>, Sara Ashley Smith<sup>b</sup>

- <sup>a</sup> Department of Communication Sciences and Disorders, Georgia State University, 30 Pryor St SW, Atlanta, GA 30303, United States
- <sup>b</sup> Department of Teaching and Learning, University of South Florida, Tampa, FL 33620, United States

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#### ABSTRACT

*Purpose*: The aim of this systematic review is to examine the early interactions between bilingualism and stuttering to synthesize knowledge that could inform diagnosis and treatment for bilingual children who stutter.

Method: Scopus, Science Direct, PubMed, ERIC Ebsco, and Google Scholar were searched with no limits placed on the year of publication. Search terms consisted of: ("stuttering" [MeSH] OR "stutter") AND ("child" [MeSH] OR "children") AND ("multilingualism" [MeSH] OR "bilingualism"). Inclusion criteria were children who stutter, bilinguals who stutter, empirical research articles, and published in peer review journals. Exclusion criteria were studies that reported on only adults, only monolinguals, or were not published in English.

Results: A total of 50 articles met the criteria. There was convergence with monolingual studies reporting sexually dimorphic and familial trends in the prevalence of stuttering and rates of recovery. Findings surrounding language proficiency, cross-linguistic stuttering severity, and development were ambivalent. Results point to the difficulty in identifying stuttering in bilingual children, and the need for culturally competent research and interpretations.

Conclusion: Current findings offer a fragmented view of bilingual development and echoes a recurring theme, i.e., the current understanding of bilingualism and stuttering is limited and more research is warranted.

# 1. Introduction

One definition of bilingual is the use of more than one language in everyday life (Grosjean, 2010a). Although bilingualism has generally been treated as uncommon in the fluency literature, according to this definition more than half of the world's population fit into this category, and this number is expected to increase (Grosjean, 2010b). The presence of English as a *lingua franca* and growing international mobility will likely result in an even greater global population of bilinguals. Research in fields such as communication disorders and language acquisition must increasingly approach childhood development in more than one language as a norm, rather than an exception. In the U.S. alone, the number of bilinguals has doubled in the past 40 years, coinciding with the growth of Hispanic/Latino and other immigrant populations (Grosjean, 2018; U.S. Census Bureau, 2018). As many as one third of U.S. children between the ages of 0–8 speak a language other than English at home with one or more parents (Park, Zong, & Batalova, 2018). Linguistic diversity in U.S. is predicted to further increase with shifting migration trends, i.e., decreasing immigration from Mexico, increasing immigration from Africa and Asia, especially China and India (Park et al., 2018). Growing national and international

E-mail address: achoo1@gsu.edu (A.L. Choo).

<sup>\*</sup> Corresponding author.

within-population variation would increase the numbers of children who will likely speak multiple languages and have varied background characteristics that influence development.

Increasingly heterogeneous combinations of language and cultural backgrounds among bilingual populations demand a fine-grained understanding of bilingualism and communication disorders during childhood. Changing demographics due to immigration, resulting in a greater number of children who speak another language in the home, will likely increase the number of bilingual children accessing education and intervention services. However, treatment may be challenging for this population without a clear understanding of how bilingualism interacts with specific disorders in shaping prognosis and development (Arias & Friberg, 2017; Centeno & Ansaldo, 2016; Kohnert & Medina, 2009; Stow & Dodd, 2003). Bilingual children are at higher risk for misdiagnosis and disproportionally referred for special education and speech-language interventions (Artiles, Rueda, Salazar, & Higareda, 2005; Muñoz, White, & Horton-Ikard, 2014). Misdiagnosis of bilinguals may be a function of failure to take into account language proficiency, developmental variability, cultural differences, or limitations of traditional assessments that are generally not normed for bilinguals (Fletcher & Navarrete, 2003; Muñoz et al., 2014). A review of empirical literature conducted a decade ago on the relationship between bilingualism and communication disorders reported a "complete dearth of studies in some areas, including autism, stuttering and acquired traumatic brain injury" (Kohnert & Medina, 2009, p. 230). Although some advances have been made, significant gaps remain.

In the topic of stuttering, the default population is monolingual. However, in a disorder such as stuttering where language development and abilities are hypothesized to play crucial roles in symptomology and prognosis, understanding cross-linguistic influence is essential to elucidate the nature of the disorder (Bloodstein, 2006; Ntourou, Conture, & Lipsey, 2011; Ratner & Benitez, 1985; Yairi, Ambrose, Paden, & Throneburg, 1996). Such knowledge would guide clinical practice, for example, by informing whether treatment should be conducted in one or both languages for bilinguals, and addressing previous suggestions that acquisition of a second language should be delayed or eliminated in managing stuttering. Treatment guidelines and practices that optimize outcomes are critical as 81% of children who stutter experience peer victimization, including physical aggression and isolation, that occurs at least once a week (Langevin, Bortnick, Hammer, & Wiebe, 1998). Children who stutter also face difficulty in establishing friendships; negative self-perceptions, shame, lower self-confidence, lower academic achievements, higher rates of anxiety; and are less likely to graduate high school or attend college (Blood & Blood, 2007; Blood, Blood, Tellis, & Gabel, 2003, 2011; Davis, Howell, & Cooke, 2002; Iverach et al., 2016; Langevin, Packman, & Onslow, 2009; Rees & Sabia, 2014). Outcomes do not improve with age. Chronic stuttering is associated with emotional and mental health challenges, lower quality of life, lower employability, and limited career advancement (Blumgart, Tran, & Craig, 2010; Craig, Blumgart, & Tran, 2009; Klein & Hood, 2004; Messenger, Onslow, Packman, & Menzies, 2004). Optimal, differentiated remediation informed by targeted research among bilingual children who stutter is needed to effectively serve this population.

Two reviews examining stuttering and bilingualism across all ages by Van Borsel, Maes, and Foulon (2001), and Van Borsel (2011) highlight significant gaps related to prevalence, risk factors, assessment and treatment in this population. First, only a handful of studies have investigated the prevalence of stuttering in bilinguals, and findings were ambivalent. Comparably, studies that examine risk factors in bilinguals have also been scant. Second, although there were more studies which evaluated stuttering severity across languages, the underlying determinants which impact cross-linguistic severity remain unclear. Third, diagnosing stuttering may be particularly challenging for clinicians who are not familiar with the languages spoken by the person who stutters. Fourth, there are no clear guidelines for treating bilinguals who stutter including whether treatment should be conducted in all languages spoken by the person who stutters. Further, the efficacy of treatments for bilinguals, which are typically based on the monolingual profile, is not known. The most recent review (see Van Borsel, 2011) was performed nearly a decade ago, a reappraisal and update of this topic is needed. A more comprehensive understanding of the link between bilingualism and stuttering could facilitate treatment strategies specific for bilinguals leading to improved outcomes, and inform models which implicate language development or deficits in the manifestation and maintenance of the disorder. We use the term bilingual to refer to the knowledge and use of two or more languages, encompassing multilingual. We conducted a systematic review to analyze the interaction between bilingualism and stuttering, and narrowed the focus to examine the early stages of this interaction, i.e., in children, with the aim of increasing knowledge that could increase specificity of diagnosis and treatment guidelines for this group. We will outline areas where the knowledge remains weak and inconclusive, and attempt to further the theoretical knowledge of the intersection of bilingualism and stuttering that could inform practice and guide future research.

#### 2. Method

# 2.1. Search strategy and information sources

A systematic review was conducted using Scopus, Science Direct, PubMed, and ERIC Ebsco, up to May 2019, according to the Preferred Reporting Items for Systematic Review and Meta-Analyses (PRISMA) guidelines (Moher, Liberati, Tetzlaff, & Altman, 2009). The search terms used consist of the following: ("stuttering" [MeSH] OR "stutter") AND ("child" [MeSH] OR "children") AND ("multilingualism" [MeSH] OR "bilingualism"). No limits were placed on the years of publication. A Google Scholar Search was also performed, and the first 500 hits were crossed-checked manually to identify relevant articles that were not detected by the database search. Each author individually scrutinized citations from the database and Google Scholar searches to identify articles that met the criteria, a 100% reliability was achieved. A hand search of the bibliography of articles that met the criteria was also conducted to identify additional empirical articles that were relevant.

#### 2.2. Study selection

#### 2.2.1. Review process

First, duplicate records were removed, and initial title and abstract screening were conducted for relevance. Records that were clearly not related to stuttering (e.g., Narrative Medicines: Challenge and Resistance) were removed. Second, the abstract or method section of records with titles that were ambiguously related to stuttering (e.g., Morphological inflections of children with normal and impaired articulation) were reviewed and the record removed if it did not mention stuttering. Third, the remaining records were reviewed, and selected based on a predefined criteria (see section on Inclusion and exclusion criteria). Each article was reviewed independently by the authors and relevant studies which did not meet the criteria were placed in a supplementary list (see supplementary material). The review process was conducted individually by each author, and the results of the selection were compared. There were no discrepancies in article selection between authors.

#### 2.2.2. Inclusion and exclusion criteria

Studies were included if they met the following criteria: (a) included children who stutter, (b) included bilinguals who stutter, (c) empirical research articles, and (d) published in peer-reviewed journals. Studies were ineligible if they included: (a) only adults, (b) only monolinguals, or (c) not published in English.

#### 3. Results

The database search yielded a total of 466 titles (Scopus = 24, ScienceDirect = 331, PubMed = 14, PubMed Central = 81, Medline = 4, and ERIC Ebsco = 12), of which 445 were unique, and 28 met the criteria. The Google Scholar search yielded a total of 7,510 titles and the first 500 were crossed checked manually; this yielded 399 unique titles, of which nine met the criteria. The reference list of articles that met the criteria yielded a total of 1,823 titles, of which 1,042 were unique, and 13 met the criteria. Overall, 50 empirical studies met the criteria and were categorized into four main topic areas: epidemiology (n = 14), profile of the bilingual child (n = 15), treatment/assessment (n = 19) and culture (n = 2), although most studies had multiple foci (Fig. 1). Thirteen studies which met the criteria also included adults who stutter ( $\geq 18$  years old). Our search identified a total of 39 articles (bolded in Table 1) which were previously not included in the Van Borsel et al. (2001) or Van Borsel (2011) reviews. A list of selected studies which were relevant to the topic but did not meet the criteria (e.g., not published in peer-reviewed journals, unable to obtain full access to the article to determine if the inclusion criteria was met) are listed in Appendix A in Supplementary material.

### 3.1. Definition of bilingual

The definition of bilingual varied across studies. For example, some studies defined bilingualism according the age of second language acquisition (Bakhtiar & Packman, 2009; Carias & Ingram, 2006; Druce, Debney, & Byrt, 1997; Howell, Davis, & Williams, 2009; Koushik, Shenker, & Onslow, 2009; Lee, Robb, Ormond, & Blomgren, 2014; Lim, Lincoln, Onslow, & Chan, 2015; Osipovskaya, Sharifzyanova, & Zamaletdinova, 2016; Stern & Log, 1948; Vong, Wilson, & Lincoln, 2011, 2016), years of exposure to another language (Lim, Lincoln, Chan, & Onslow, 2008, 2015; Maruthy, Raj, Geetha, & Priya, 2015; Schäfer & Robb, 2012), whether a language other than English is spoken in the home (Bebout & Arthur, 1997; Howell & Davis, 2011; Reilly et al., 2013; Rousseau, Packman, Onslow, Harrison, & Jones, 2007; Trajkovski et al., 2011), exposure to formal instruction in another language (Mamdoh & Gomaa, 2015; Mohammadi, Nilipour, & Yadegari, 2008; Mohammadi, Bakhtiar, Rezaei, & Sadeghi, 2012; Mohammadi, Khazaie, Rezaei, & Joghataei, 2016; Nwokah, 1988; Ralston, 1981), or number of languages spoken (McLeod, Verdon, & Bennetts Kneebone, 2014). Notably, a majority of the studies did not operationally define or quantify bilingualism (Andrews et al., 2012, 2016; Aron, 1962; Baker & Cantwell, 1982; Blanton, 1916; Dale, 1977; Firozjaei, 2013; Gkalitsiou, Byrd, Bedore, & Taliancich-Klinger, 2017; Harrison, Onslow, & Menzies, 2004; Lattermann, Shenker, & Thordardottir, 2005; Lincoln, Onslow, Lewis, & Wilson, 1996; Malek, Amiri, Hekmati, Pirzadeh, & Gholizadeh, 2013; Mirawdeli, 2015; Ralston, 1981; Raza et al., 2013; Tellis, 2008; Tetnowski, Richels, Shenker, Sisskin, & Wolk, 2012; Travis, Johnson, & Shover, 1937; Van Zaalen-op't Hof, Wijnen, & De Jonckere, 2009; Vong, Wilson, McAllister, & Lincoln, 2010; Wright & Sherrard, 1994a, 1994b). Only a handful of studies utilized standardized assessments or selfrating scale to measure bilingual proficiency (Byrd, Watson, Bedore, & Mullis, 2015; Lim, Lincoln et al., 2008, 2015; Taliancich-Klinger, Byrd, & Bedore, 2013).

#### 3.2. Epidemiology

#### 3.2.1. Prevalence

Findings on the prevalence of stuttering in bilinguals were ambivalent with the exception of those related to differences between sexes. *Higher rates of stuttering in bilinguals*. Travis et al. (1937); Stern and Log (1948), and Firozjaei (2013) reported that the prevalence of stuttering were 1.3 to 3.1 times higher in bilingual compared to monolingual children. The prevalence of stuttering has also been reported to increase up to around 11 years old in bidialectal children (Ralston, 1981). *Similar rates compared to monolinguals*. The prevalence of stuttering in bilingual children were reported to be around 4.9% by McLeod et al. (2014), and 4.7% by Ralston (1976), similar to previous reports in monolingual children who stutter (Yairi & Ambrose, 1999). For studies which included young adults who stutter, the prevalence was reported to be 1.26% in bilingual Bantu children between 6 to 21 years by Aron (1962), and 1.13% in Kurdish-Farsi speaking children between 6 to 19 years by Mohammadi et al. (2008). Further, Howell, Davis, Williams et al.

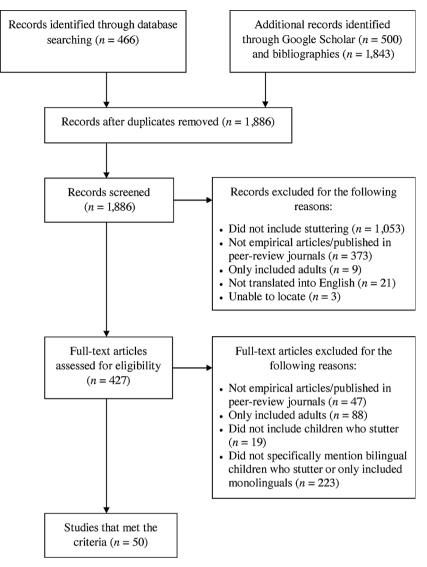


Fig. 1. Article selection process.

(2009) reported that only 21.8% of children in their clinical cohort who stuttered were bilingual, which is lower than the prevalence of bilingual children in the general population (28.4%). *Higher ratio of stuttering in bilingual males compared to females*. Studies which compared rates of stuttering across sexes consistently found higher rates in males compared to females (Aron, 1962; Howell, Davis, Williams et al., 2009; Ralston, 1976; Stern & Log, 1948; Travis et al., 1937). The highest ratio of bilingual males to females who stutter was 4:75 for children between 8 to 12 years (Howell, Davis, Williams et al., 2009) while the lowest ratio was 2:1 for bilingual children between 4 to 17 years (Travis et al., 1937). For monolingual children, the ratio of male to female who stutter ranged between 3:4:1 to 8:1 (Howell, Davis, Williams et al., 2009; Stern & Log, 1948; Travis et al., 1937).

#### 3.2.2. Age of onset

There were no clear trends for the age of stuttering onset, with the exception of differences between sexes. Stuttering has been reported to occur between 1 to 11 years old in bilingual children who stutter (Bakhtiar & Packman, 2009; Harrison et al., 2004; Howell, Davis, Williams et al., 2009; Karniol, 1992; Schäfer & Robb, 2012; Taliancich-Klinger et al., 2013; Vong et al., 2016). Bilinguals versus monolinguals. Howell, Davis, Williams et al. (2009) reported earlier stuttering onset for monolinguals (4;3) compared to early (4;10) and late (4;9) bilinguals. However, Lincoln et al. (1996) reported similar age of onset, around 4;3, for bilingual and monolingual children who stutter in a clinical cohort. Bilingual males versus females. Schäfer and Robb (2012) found earlier stuttering onset for German-English speaking females (mean age = 5 years old) compared to males (mean age = 6 years old). Similarly, Vong et al. (2016) reported earlier stuttering onset (between 1 to 2 years old) for the majority of females (50%, n = 2) compared to the only male (around 3 years old) in the study. Cross-cultural/race comparisons. Aron (1962) found earlier stuttering onset for children of African ancestry (3;4) relative to children of European ancestry (4;10) based on parent reports in South Africa.

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Study (year)	Age (yrs)	N	Languages/Bilingual criteria or age of L2 acauisition	Topic area	Results
Andrews et al. (2012)	6-11	bCWS = $5 (F = 1)$ mCWS = $5 (F = 1)$	Languages spoken and bilingual criteria were not reported.	Treatment	Syllable-timed speech <sup>®</sup> along with parental verbal feedback were observed to reduce the frequency of stuttering by more than 50% in about half of the participants. However, one bCWS did not show a
Andrews et al. (2016)	6;0-11;11	bCWS = 17 (F = 3) mCWS = 5 (F = 3)	Arabic/Croatian/Gujarati/Hindi/ Mandarin/Spanish/Tamil/Turkish/ English Not defined.	Treatment	reduction in stuttering. Syllable-timed speech <sup>b</sup> along with parental verbal feedback were observed to reduce the frequency of stuttering and self-reported stuttering severity during treatment and 12 months post-treatment. Reduction in avoidance behavior, perceived impact of stuttering, and
Aron (1962)	6-21	bCWS = $6.581$ (F = $3.476$ )	Afrikaans/Bantu languages/English Not defined <sup>c</sup> .	Epidemiology	increased speech naturalness were also reported.  The prevalence of stuttering was 1.26% ( $n = 83$ , $F = 21$ ). The ratio of males to females who stutter was 3.28.1. For children who spoke Bantu languages, the mean age of onset based on 16 parent interviews was 3.4, while the mean age of onset based on self-report was 9.4. For children of European descent, age of onset based on parent interview was 4;10 while the self-reported mean age of onset was 8;5. The prevalence of sinistrality was higher for females who stutter compared
Baker and Cantwell (1982)	Children with SD (including stuttering) = 2.3–15 Children with LD = 3.0–15.8 Children S.D. = 1.9–13.9	SD = 108 (F = 33; CWS = 16) LD = 19 (F = 8) S-LD = 164 (F = 44)	About 4% of children with SD, and 5% of children with S-LD were from bilingual homes.  Languages spoken and age of bilingual exposure were not reported.	Profile	to remain with speech disorders (SD, 29%) were least likely to be claidren with speech disorders (SD, 29%) were least likely to be diagnosed with a concomitant psychiatric disorder, based on the DSM-III, compared to children with language disorders (LD, 95%), or speech and language disorders (S-LD, 45%). The most frequently reported concomitant disorders were behavioral (e.g., attention deficit) and
Bakhtiar and Packman (2009)	8;11	bCWS = $1$	Baluchi/Persian Learned Persian in school at age 7 years.	Treatment	emotional (separation anxiety).  The Lidcombe Program <sup>d</sup> was reported to decrease rates of stuttering in the school-age child across both languages spoken. Parent verbal feedback was conducted in L1 (Baluchi) and L2 (Persian), while parent
Bebout and Arthur (1997)	17-71 °	Bilingual immigrant respondents = $60 \text{ (F} = 40)$ Monolingual second-generation American respondents = $46 \text{ (F} = 29)$	Cantonese/English Bilinguals were either an immigrant or a first generation Chinese-American who used Cantonese in the home. Monolingual groups were U.S-born.	Culture	training by SLP was conducted in L.2.  Monolingual respondents agreed more strongly than bilingual respondents with these statements: "Severe child stutterers have trouble making friends" and "It is normal for other children to make fun of a badly stuttering child". In contrast, bilingual respondents were more likely to agree with the statement that persons with disorders
Blanton (1916)	6	$bCWS = 1^{f}$	Italian/English	Epidemiology	could reduce the severity of their disorder by "trying hard".  Onset of stuttering in a 9-year old boy who spoke only Italian at home many and to not old their braits in calculations.
Byrd, Watson et al. (2015)	bCWS = 6;1 $bTDC = 5;11$	bCWS = 1 (F = 1) bTDC = 1 (F = 1)	Not urgined. Spanish/English Assessed by BESA, bCWS was 66%	Assessment	was observed to controle with realning English in School.  Twenty-nine percent $(n = 4)$ of Spanish-English SLPs incorrectly identified the bCWS as a child who does not stutter. About 86%
Carias and Ingram (2006)	4-10	SLES = 14 (F = 13) bCWS = 2 (F = 1)	ragusa donntan. Spanish/English Age of bilingual exposure ranged between 4-6 year old for males. Age of exposure was not reported for the female	Profile	(n = 12) of SLL's incorrectly Identified the DLD, as a child who stutiers. Repetition was the most common type of disfluency in the more proficient language, while insertions and prolongations were more common in the less proficient language. Children produced higher rates of disfluencies in their more proficient language.
Dale (1977)	Mean = 13 years	bCWS = 4	oom. Spanish/English Not defined.	Profile	Disfluencies were reported in Spanish (1.1) but not English (1.2). bCWS reported being forced to speak Spanish and seeing parental distress when stuttering in Spanish. One bCWS showed physical concomitant during moments of stuttering.

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Study (year)	Age (yrs)	N	Languages/Bilingual criteria or age of L2 acquisition	Topic area	Results
Druce et al. (1997)	6;9–8;1	mCWS = 9 bCWS = 6 (F = 2 but unclear if they were bCWS)	German/Greek/Hindi/Italian/ Slovenian/English Simultaneous acquisition of languages during the first 5 years life.	Treatment	Both bCWS and mCWS showed reduced frequency of stuttering immediately after treatment. Fluency was maintained for 18 months after the intensive treatment program <sup>8</sup> . Speech naturalness also increased during this period.
Firozjaei (2013)	Primary school children (age range was not provided).	Bilingual = $550 (F = 275)$ Monolingual = $550$ (F = $275$ )	Languages spoken and bilingual criteria were not reported.	Epidemiology	There was a higher prevalence of stuttering for bilinguals (2.37%) compared to monolinguals (0.76%) and overall higher ratio of male to female who stutter (1.1.59).
Gkalitsiou et al. (2017)	3;10-6;8	bcws = 4 (F = 2)	Spanish/English Age of language acquisition was not reported.	Profile	Overall, bCWS showed higher frequency of stuttering for function compared to content words for the Spanish narrative and conversation tasks, and for the English narrative speech task. However, similar rates of stuttering on function and content words were found for the English conversation sample for Spanish dominant bCWS $(n = 3)$ . The English-dominant bCWS showed higher frequency of stuttering on function compared to content words for the English-conversation sample
Harrison et al. (2004)	2;0-5;11	mCWS = 33 bCWS = 5 (F = not specified for either group)	Arab/Greek/English Not defined.	Treatment	Confidence and ministered the Lidcombe Program with parent contingency reduced or maintained their rate of stuttering during a 4-week period. However, rates of stuttering increased for children who did not receive parental eleback. Seventy one percent of participants reported a family history of entirely.
Howell, Davis, Bartrip et al. (2009), Howell, Davis, Williams et al. (2009)	8-12	mCWS = 248 bCWS = 69 (F = not specified for either group)	English; other languages spoken were not reported.  Early bilinguals = exposed to a second language from birth, late bilinguals = learned a second language in preschool.	Epidemiology	for early painty of statements of stuttering was 60.5% and about a quarter recovered. For late bilinguals, the prevalence of stuttering was 39.5%. The recovery rates for late bilinguals and monolinguals (as a group) was 55%. Onset age was closer to 5 years for bilinguals but closer to 4 years for monolinguals. More males than females stutter (> 4:1) in all groups. There were no differences in academic performance (English, Mathematics, and Science) between early bcWS, late bcWS, and tvoically fluent bilinguals.
Howell and Davis (2011) Karniol (1992)	Age range was not provided but all children were assessed during their teenage years.	mCWS = 103 bCWS = 29 (F = 26 for both mCWS and bCWS) bCWS = 1	Languages spoken were not reported. bCWS predominantly spoke another language (not English) in the home. Hebrew/Hungarian/English Exposure to languages prior to age 1 year.	Epidemiology Epidemiology	Billingual status was not predictive of persistent stuttering or recovery for children who were confirmed to stutter at age 8 years. Only stuttering severity, measured by the SSI-3, at initial assessment was found to be a significant predictor of stuttering outcome. Child started stuttering about a month after parents reported signs of billingualism, stuttering became more severe 4 weeks later (at 2;0). Over a period of 4 months, reduction in the use of L2 (English)
Koushik et al. (2009)	6;8–10;8	bCWS = 12 (F = 2)	French/Greek/Hebrew/Italian/ Portuguese/English Children were simultaneous (two languages since birth) or sequential hilmonuk (17 or noe 4 worve or older)	Treatment	coincided with a decrease in stuttering.  Overall, treatment using the Lidcombe Program reduced rates of stuttering (from 9.2% SS to 1.9% SS) and increased the rate of syllables produced (from 145.8 spm to 179.3 spm). However, follow-up parent interviews reported slight increases in stuttering rates in two bCWS.
Lattermann et al. (2005)	4;1-5;11	bCWS = 4	French/Italian/English Not defined.	Treatment	Treatment using the Lidcombe Program increased fluency, MLU, use of complex sentence and NDW in children over the 12-week treatment period and at the 6-month follow-up.
Lee et al. (2014) <sup>-</sup>				Assessment	(continued on next page)

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Study (year)	Age (yrs)	N	Languages/Bilingual criteria or age of 12 acquisition	Topic area	Results
	bCWS = 16 bAWS = 22 (F = 1) SLP = not provided	Bilinguals who stutter = 2 (F = 1) SLP = 19 (F = 18)	bCWS and bAWS = Spanish/English SLP = Afrikaans/Cantonese/Dutch/ Kannada/Malay/Mandarin/English bCWS (age 16 years) acquired L2 around age 5.		SLPs identified more instances of stuttering in the L1 (Spanish) compared to L2 (English) for bCWS, but rates of stuttering in the L1 and L2 were similar for bAWS. English-speaking SLPs showed higher accuracy in identifying instances of stuttering in English compared to Spanish. Overall, bilingual SLPs identified more instances of stuttering than monolineual SLPs recardless of the laneuage.
Lim, Lincoln et al. (2008), Lim, Liow et al. (2008)	12-44	Bilinguals who stutter $= 30 \text{ (F} = 2)$	Mandarin/English Bilngualism was based on self-report of language proficiency and use (see Lim,	Profile	Balanced bilinguals showed similar rates of stuttering in both languages, while dominant bilinguals showed higher rates of stuttering in their less proficient language.
Lim et al. (2015)	12–47	Bilinguals who stutter $= 19 \; (F = 1)$	Mandarin/English Mandarin/English Was age of exposure to both languages was around 3.5 years for Mandarin and 3.4 wars for English	Treatment	The effects of the speech restructuring intensive program conducted in English generalized to Mandarin. Reduction in rates of stuttering were observed in both languages.
Lincoln et al. (1996)	mCWS = 7;0-12;4 bCWS = 6;10-9;10	mCWS = 8 $bCWS = 3 (F = 1)$	Languages spoken by bilinguals were not reported.  Not defined	Treatment	Two out of the three bCWS showed the smallest increases in fluency in the study, and both experienced relapse with the Operant Program $\!^{\rm I}$
Malek et al. (2013)	6-11	bCWS = 40 bilinguals with dyslexia = 40 bilingual controls = 40 (F = not specified for (F = not specified for either errorm)	Languages spoken were not specified <sup>k</sup> Not defined.	Profile	bCWS showed lower speech motor skills as reflected in lower number of syllables uttered, slower reaction and completion time, and higher error rates during diadochokinetic tasks compared to bilingual children with dyslexia and controls.
Mamdoh and Gomaa (2015)	10;1–11;8	bCWS = 31 (F = 8)	Arabic/English Learned L2 (English) at around 4 years old	Profile	Higher rates of stuttering in L2 (English) compared to L1 (Arabic).
Maruthy et al. (2015)	16-28	Bilinguals who stutter = $25 (F = 2)$	ou. Kannada/English Mean age of exposure to L2 (English) ranead between 4–15 voars old	Profile	Higher rates of stuttering in L2 (English) compared to L1 (Kannada). More stuttering on content compared to function words in L1 while the onnosite was observed for 1.2.
McLeod et al. (2014)	3-5	Bidialectal children = $692$ (F = $338$ )	English of languages spoken and Number of languages spoken and	Epidemiology	About 4.9% of parents in the survey expressed concern about their child's stuttering.
Mirawdeli (2015)	4-6	Total = 730 (F = 361) Native English speakers = 55.6% For = $44.40$	projecting were provinces or provinces. Akan African/Bengali/Polish/ Portuguese/Romanian/ Tamil/Urdu/ Other/English	Assessment	The SSI-3 was found to have high sensitivity in identifying those who stutter. The probability of being diagnosed with stuttering, as measured with the SSI-3 in English, was not higher for ESL versus native English
Mohammadi et al. (2008)	6–19	DTDC and bCWS = 11,425 (F = 5,137)	no againea. Kardish-Farsi Acquisiton of L2 from age 5 or 6 years.	Epidemiology	Packeds.  The prevalence of stuttering was lowest for ages 14–19 years (0.5%), followed by ages 11–14 years (0.87%), and ages 6–11 years (2.06%). Across all age groups, the prevalence was 1.3%. The highest male to female ratio of stuttering was for ages 11–14 years (3.25:1), followed by ages 14–19 (1.76:1), and ages 6–11 years (1.28:1). Across all age promus the ratio of male to female who stuttering was 1.8:1.
Mohammadi et al. (2012)	9-13	bCWS = 31 (F = 15)	Kurdish/Persian Acquisition of L2 from age 6 years.	Profile	Stuttering was more severe in the L2 (Persian). Severity in the L1 (Kurdish) predicted severity in the L2 and vice versa. MUU of the L2 predicted severity in the L2 but this pattern was not found for the L1. (continued on next page)

Study (year)	Age (yrs)	N	Languages/Bilingual criteria or age of L2 acquisition	Topic area	Results
Mohammadi et al. (2016)	7–14	bCWS = 22 (F = 11)	Kurdish/Persian The L2 (Persian) was learned in school although the specific ages were not reported.	Epidemiology	Recovery rate was 22.7%, and correlated with right hand dominance on the Purdue Pegboard test. Children with persistent stuttering did not show a difference in performance between the right and left hand. Recovery was not found to be correlated with age or sex.
Nwokah (1988)	16-40	Bilinguals who stutter = $16$ (F = 4)	Igbo/English Balanced bilinguals, the L2 was learned in primary school at around 6 years old.	Profile	Stuttering occurred in both languages. The majority of bilinguals (94%, $n = 15$ ) showed higher frequency of stuttering in one language, while only 6% ( $n = 1$ ) showed similar frequency across languages for spontaneous speech. For reading, 88% ( $n = 14$ ) of bilinguals showed higher rates of stuttering in one language, while only 12% ( $n = 2$ ) showed similar rates across languages.
Osipovskaya et al. (2016)	9-10	bCWS = 19 Controls = 46 (F = not specified for either group)	Tatar/English Early bilinguals = acquisition of languages prior to age 6 years, late bilinguals = acquisition of L2 after or at age 7 years.	Profile	Early bilinguals with high proficiency in their 1.2 showed similar rates of stuttering in their L1 (Tatar) and L2 (Russian). However, early bilinguals with lower proficiency in their L2, and late bilinguals showed higher rates of stuttering in their L2.
Ralston (1981)	5-11	Bidialectal children = 1,999 (F = not specified).	Nevisian dialect/Standard Nevisian English Nevisian Dialect (1.1) and Standard Nevisian English (1.2) were learned in schools.	Epidemiology	The prevalence of stuttering was 4.7%. The ratio of male to female who stutter was 2.48.1. About 88% of children who stutter were reported to have above average intelligence by their teachers.
Raza et al. (2013)	Not reported <sup>m</sup>	Total number of family members = 71, of which 33 were reported to stutter (F = not specified).	Cameroonian dialect/English	Epidemiology	Evidence for linkage loci to stuttering on chromosomes 2p, 3p, 3q, and 15q.
Reilly et al. (2013)	4.2	CWS = 181 (bCWS = 4; F = 75) TDC = 1438 (bTDC = 98; F = 718)	Languages spoken were not provided. Non-English speaking background	Epidemiology	Across all language status (including bilinguals), stuttering onset was correlated with higher maternal education, being male, and being a twin. Recovery rates within 12 months of onset were found to be higher for children who did not repeat whole words and for boys. Stuttering onset and recovery were not correlated with bilingual status.
Rousseau et al. (2007)	3-6	mCWS = 21 bCWS = 8 (Overall, F = 8, however, the numbers in each group were not reported).	Languages spoken were not provided. Exposed to a second language at home.	Treatment	As a group (mCWS and bCWS together), a median of 16 clinic visits were required to enter the maintenance stage of the Lidcombe Program. The mean stuttering severity prior to treatment was 3% SS and the mean severity was below 1% SS after treatment. Higher MLU prior to treatment was correlated with shorter treatment time while higher receptive language scores was correlated with longer treatment time.
Schäfer and Robb (2012)	13-59	bCWS = 6 bAWS = 9 (F = 4, however, it is unclear if there were bCWS in this groun)	German/English Exposure to English (L2) prior to age 5 years.	Profile	Overall, higher rates of stuttering were found in the L2 (English). In the L1 (German), higher rates of stuttering were found on content words, while higher rates of stuttering were found on function words in L2 (English).
Stern and Log (1948)	6–16	Total = 1,861 including CWS (F = not specified).	Afrikaans/Arabic"/French/Hebrew"/ English Acquisition of languages prior to age 6 years	Epidemiology	The prevalence of stuttering was higher in bilinguals (2.16%) compared to monolinguals (1.66%). The ratio of males to females who stutter was 3:1 for bilinguals, and 8:1 for monolinguals.

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Table 1 (continued)

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Study (year)	Age (yrs)	N	Languages/Bilingual criteria or age of 12 acquisition	Topic area	Results
Taliancich-Klinger et al. (2013)	6;1	bCWS = 1	Spanish/English Exposed to Spanish in the home and English in kindergarten. Better Spanish language skills and lower English language skills and sassesed by BESA.	Profile	Higher rates of stuttering were found for the less proficient language (English). In terms of disfluency types, there were more SLDs in the Spanish than English narrative samples, and more ODs in the English than Spanish narrative samples.
Tellis (2008)	Mean age = 22.89°	Hispanic American college students = 258 (F = 154)	Languages spoken by respondents were not specified. Not defined.	Culture	Based on the survey, 2.2.4% of Hispanic American college students were found to agree with the statement that stuttering was the result of pressure placed by parents on a child to speak two languages, English and Spanish. Further, 39.4% of the respondents believed that switching from the more proficient (Spanish) to less proficient (English) language
Tetnowski et al. (2012)	4	bCWS = 1	French/Tamil/English Not defined.	Treatment	The SLP advisor are the speak English in the home to expose the child to English although the child's parents spoke mainly Tamil. The father spoke a little English and French while mother does not.
Trajkovski et al. (2011)	3-5;9	mCWS = 11 bCWS = 6	Arabic/Punjabi/Russian/Spanish/ Telegu/English Language spoken at home.	Treatment	Five out of the six bCWS withdrew from the syllable-timed speech treatment for various reasons. The one remaining bCWS achieved < 1% SS, i.e., criterion for entrance to the maintenance stage, in 11 visits. Overall, mCWS achieved < 1% SS between 4 to 10 visits, with a mean of 12.5 visits.
Travis et al. (1937)	4-17	Total: 4,827 (F = 2,422) bCWS = 2.8% mCWS = 1.8%	Afro-Asiatic/Indo-European/Sino- Tibetian language groups <sup>p</sup> Not defined.	Epidemiology	There was a higher percentage of children who spoke two (2.8%) or three (2.38%) languages who were identified with stuttering compared to monolingual children (1.8%). The study also identified a higher prevalence of stuttering in non-Caucasian children (3.76%) compared to Caucasian children (3.76%) compared to Caucasian children (3.76%) of siblings were reported to stutter, while 45% of siblings in monolingual families stutter. The prevalence of stuttering in children who did not speak English was 7.41%. Ratio of male to female who stutter was 2:1 for bilinguals, and 3.4:1 for monolinguals. The study also reported lower intelligence, and poorer physical development in bilinguals compared to monolinguals.
Van Zaalen-op't Hof et al. (2009) Vong et al. (2010)	Stutter = 6-39.4 Control = 12.6-47.3 2;7-4;9	Stutter = 54 (F = 13) Control = 25 (F = 8) bCWS (F = 3) and mothers = 6 dyads	Dutch <sup>q</sup> Not defined. Malay/Mandarin/English Not defined.	Assessment	Low agreement between clinicians for the diagnoses of stuttering, cluttering or stuttering-cluttering.  Malaysian parents code-switched to English when praising their children although their L1 was Malay or Mandarin. The frequency and expressions of praise were reported to be lower and more limited than other transes of reservoirs and another interest than expersions of praise were accordance and more limited than expressions of praise were accordance and more limited than
Vong et al. (2011)	3;9-4;9	bCWS = $3 (F = 2)$	Malay/Mandarin/English Acquisition of languages prior to age 4 years.	Profile	The two female bCWS showed higher rates of stuttering in their L2 (English) compared to their L1 (Mandarin), while the male bCWS showed similar rates of stuttering in the L1 (Mandarin) and L2 (English). Treatment conducted in one language, whether L1 or L2, generalized to the other language. For the male bCWS who received treatment in his L2 (English), he was reported to refuse to speak
Vong et al. (2016)	3,3-4,9	bCWS = 3 (F = 2) mCWS = 1 (F = 1)	Mandarin/Malay/Chinese dialects/ English Aquisition of languages prior to or during preschool.	Treatment	Manual III. In the case of the disciplination of the second second second in fluency was observed across L1 and L2 languages, although treatment was delivered in only one language, either the L1 or L2.
	Not provided <sup>r</sup>			Treatment	

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Study (year)	Age (yrs)	N	Languages/Bilingual criteria or age of Topic area L2 acquisition	Topic area	Results
Wright and Sherrard (1994a)		SLP = 116 contacted, however, only 87 completed the survey on their caseloads of bCWS (British Asian children and adolescents).	SLPs identified as Asian (6%), British (81%) and other (13% - including South African, Dutch, West Indian, European).  Languages spoken were not provided and bilingualism was not defined.		Respondents reported discussing their caseloads with interpreters or co-workers (68%, $n = 55$ ) or bilingual SLPs (27%, $n = 22$ ), and attending training courses (54%, $n = 44$ ) and acquiring materials (42%, $n = 34$ ) for working with Asian clients. Respondents also reported that typically or sometimes, the adult relative of the child who stutters (96%, $n = 52$ ), a friend of the child's family (54%, $n = 29$ ), and child relatives of the child who stutters (74%, $n = 40$ ) served as the interpretable.
Wright and Sherrard (1994b)	Not provided <sup>8</sup>	Similar to Wright and Sherrard (1994a)	Similar to Wright and Sherrard (1994a)	Treatment	Mean years. About 75% ( $n = 54$ ) of non-Asian SLPs reported lower perceived success with their Asian clients than their non-Asian clients, and SLPs who were dissatisfied with the quality of interpreter services were more

(ren) who stutter(s), NDW = number of different words, SD = speech disorder, SLP = speech-language pathologist, S-LD = speech and language disorder, spm = syllable per minute, %SS = percent bAWS = bilingual adult(s) who stutter(s), bCWS = bilingual child(ren) who stutter(s), BESA = Bilingual English-Spanish Assessment, bTDC = typically developing bilingual child(ren), CWS = children who stutter; ESL = English as a second language, F = female, L1 = native/first language, L2 = second language, LD = language disorder, MLU = mean length utterance, mCWS = monolingual child syllable stuttered, SSI-3 = Stuttering Severity Instrument 3<sup>rd</sup> edition. Bolded = not previously included in the Van Borsel, Maes, and Foulon (2001) or Van Borsel (2011) reviews.

a The treatment included speaking each syllable in time to a rhythmic beat. Parents were taught to use verbal contingencies and to praise their child's use of syllable-timed speech.

likely to perceive lower success with their Asian clients.

c The study examined children in the Orlando Township, Johannesburg. Children were reported to be bilingual, spoke two to three Bantu languages in addition to the official South African languages, English or Afrikaans. The language combinations and number of children who spoke these combinations were not reported.

The program uses parent-administered response-contingent feedback.

e Study examined attitudes toward children with communication disorders. Respondents were not individuals who stutter but bilingual immigrants and monolingual second-generation Americans who The study looked at the prevalence of speech disorders including lisping, aphasia, mutism in schools around Madison, Wisconsin. spoke English. Respondents were asked to rate statements to evaluate attitudes toward stuttering, deaf speech and cleft palate.

g. The intensive program was conducted over 5 days for 6.5 hours a day. Children learned to identify moments of stuttering in their own speech and a peer's, and clinicians modeled slowed speech rate. Fluent speech was reinforced with rewards consisting of stickers, games or social praise. h The child's L1 and L2 speech was recorded by the mother during this period.

i The study examined the assessment of stuttering by monolingual and bilingual SLPs in two Spanish-English bilinguals who stutter. SLPs reported between 1–22 years of clinical experience.

k Children were matched for bilingual proficiency but the authors did not specify the languages spoken. The study was conducted in Tabriz where Azerbaijani is widely spoken in addition to Farsi (see <sup>j</sup> Uses parent-administered response-contingent operant feedback. Mirvahedi, 2016).

1 Some of the indigenous Australian languages spoken include Anindilyakwa, Arrernte, Djambarrpuyngu, Galpu, Gurindji, Kalaw KawawYa/Kalaw Lagaw Ya, Luritja, and Murrinh Patha. m The study participants were part of a family which included children. However, the specific ages of adults and children were not reported.

n In the article, the languages were listed as Syrian and Jewish. However, the language is most likely Arabic, which is the most widely spoken language by Syrians, and Hebrew which is the most widely spoken Jewish language.

o. The study was a survey of perceptions of stuttering based on the Stuttering Inventory for Hispanic Americans. Respondents were Hispanic American college students from Pennsylvania, New York and

P The reported languages were listed as Armenian, Austrian, Bohemian, Bulgarian, Chinese, Croatian, Czechoslovakian, Danish, Finnish, French, German, Greek, Hungarian, Italian, Lithuanian,

<sup>q</sup> Participants included monolingual and bilingual children who spoke Dutch. The number of bilinguals and languages spoken were not reported. Macedonian, Mexican, Polish, Romanian, Russian, Serbian, Swedish, Syrian, Turkish, Ukranian, Yiddish, and Yugoslavian.

<sup>r</sup> The study surveyed SLPs who work with British Asian children in the United Kingdom.

#### 3.2.3. Familial risk

Findings suggest the majority of bilingual children who stutter have a familial history of stuttering that may be related to stuttering outcomes. *Positive family history*. Most studies report a positive family history of stuttering in their cohort of bilingual children who stutter, ranging between 75% to 90% (Andrews et al., 2012, 2016; Mohammadi et al., 2016; Vong et al., 2016). However, Aron (1962) found that only about a third of bilingual children had a family history of stuttering, and parents were more likely to report a positive family history compared to the child. It is unclear if the prevalence of family history in the Aron (1962) study was based on the parent or child report. *Family history and recovery*. Only 20% of bilingual children who recovered reported a positive family history of stuttering compared to 80% of bilingual children with chronic stuttering who did so (Mohammadi et al., 2016). *Genetic linkage*. Although no single gene has been found to cause stuttering, Raza et al. (2013) reported evidence for linkage on chromosomes 2, 3, 14, and 15 from a bilingual Cameroonian family with a high number of individuals who stutter.

#### 3.2.4. Bilingualism as a risk factor

The onset of stuttering was reported to coincide with the acquisition of a second language in three studies. *Onset of stuttering*. These studies reported a link between the onset of stuttering and learning a second language (Blanton, 1916; Karniol, 1992; Travis et al., 1937). Travis et al. (1937) reported that for 26% of bilingual children (13 boys and 4 girls) in their study, the onset of stuttering coincided with learning a second language. However, it is important to note that in each of the Blanton (1916) and Karniol (1995) studies, findings were based on a single child. Dale (1977) observations of four Spanish-English bilingual Cuban-American children prompted support for the Diagnosogenic Theory of Stuttering (Johnson, 1942), that is, stuttering resulted from the pressure to speak their first language (L1; Spanish) fluently for fear of parental displeasure, and the labeling of typical disfluencies as stuttering. Additionally, the study by Reilly et al. (2013) which included eight bilingual children who stutter reported that being male, being a twin, and higher maternal education but not bilingual status were correlated with an increased risk of stuttering. *Age of language acquisition*. Howell, Davis, Williams et al. (2009) pointed to a higher proportion of early bilinguals (exposed to two languages since birth) than late bilinguals (acquired a second language in school) who stutter in a clinical cohort of children between 8 to 12 years old.

#### 3.2.5. Recovery

There were no distinct patterns for recovery, with the exception of sexually dimorphic trajectories, which mirrored those in monolinguals. Age of second language (L2) acquisition. The correlation between age of language acquisition and rates of recovery have been ambivalent. Howell, Davis, Williams et al. (2009) reported that only a fourth of early bilingual children who were exposed to a second language from birth recovered from stuttering while more than half (55%) of monolinguals and late bilinguals (together as a group) recovered. In contrast, Mohammadi et al. (2016) found slightly lower recovery rates for late bilingual children (22.7%; learned a second language after age 6 years) compared to early bilingual children (25%; exposed to a second language since birth). L2 and recovery. Karniol (1992) reported recovery that coincided with cessation of L2 use, and when the L2 was reintroduced about a year after the observed recovery, symptoms of stuttering did not return. Sex. More bilingual girls have been reported to recover compared to bilingual boys (37.5% vs. 14.3%, Mohammadi et al., 2016). This sexually dimorphic trend is similar to unassisted recovery in monolinguals where girls are twice as likely to recover compared to boys (Ambrose, Cox, & Yairi, 1997; Yairi & Ambrose, 1999). Developmental factors. Bilingual children who recovered showed better performance with their right hand compared to the left during the Purdue Pegboard test, however, bilinguals with persistent stuttering did not show a difference between the left and right hand performance (Mohammadi et al., 2016). In contrast to bilinguals, Seider, Kidd, and Gladstien (1983) found higher rates of recovery for monolingual females who were right-handed compared to left-handed females although there were no differences between monolingual right- and left-handed males. No differences were found for inhibitory control, as evaluated by Stroop tests, between bilinguals with chronic stuttering and who recovered (Mohammadi et al., 2016). Handedness has also been examined as a factor in recovery for monolinguals (Bloodstein & Ratner, 2008). Similar to bilinguals, there were no differences between monolingual children who did and did not recover in temperament, including inhibitory control (Ambrose, Yairi, Loucks, Seery, & Throneburg, 2015; Singer, Walden, & Jones, 2019).

# 3.3. Profile of bilingual children who stutter

# 3.3.1. Stuttering behaviors and speech characteristics

There were consistent trends across studies including the cross-linguistic occurrence of disfluencies, and production of both stuttering-like disfluences (SLD) and typical/other disfluencies (OD) across languages. Nonetheless, findings related to the interaction between stuttering severity, language proficiency and speech tasks were less conclusive. *Stuttering in one or multiple languages*. The majority of studies reported cross-linguistic occurrence of stuttering in bilinguals. In a study that included 69 bilingual children who stutter, about 95% were found to exhibit disfluencies in all languages spoken (Howell, Davis, Williams et al., 2009). This finding was supported by three smaller studies ( $12 \le n \ge 31$ ) where all bilingual children stuttered in all languages spoken (Koushik et al., 2009; Mamdoh & Gomaa, 2015; Mohammadi et al., 2012). *Types of stuttering*. Bilinguals produced both SLD and OD in all languages spoken (2016, Ambrose & Yairi, 1999; Carias & Ingram, 2006; Gkalitsiou et al., 2017; Lattermann et al., 2005; Lim, Lincoln et al., 2008; Taliancich-Klinger et al., 2013; Vong et al., 2011). Carias and Ingram (2006) reported that insertions and prolongations were the most common types of disfluency in the less proficient language while repetitions were the most common in the more proficient language. *Language proficiency and stuttering severity*. Most studies reported higher frequency of stuttering in the less proficient language (Dale, 1977; Lim, Lincoln et al., 2008; Mamdoh & Gomaa, 2015; Maruthy et al., 2015; Mohammadi et al., 2012; Osipovskaya et al., 2016;

Schäfer & Robb, 2012; Vong et al., 2011). However, two studies found higher frequency of stuttering in the more proficient language (Carias & Ingram, 2006; Lee et al., 2014), and one study reported similar rates of stuttering across languages in a child with unequal language proficiency (Vong et al., 2011). Findings for balanced bilinguals were similarly ambivalent. Lim, Liow, Lincoln, Chan, and Onslow (2008) reported similar frequency of stuttering across languages during spontaneous speech for balanced bilinguals. However, Nwokah (1988) found higher frequency of stuttering in one language for the majority of balanced bilinguals (94%), and only one bilingual (6%) showed similar severity across languages during spontaneous speech. Both the Lim, Liow et al. (2008) and Nwokah (1988) studies included adults. Speech tasks and stuttering severity. Although the frequency of stuttering has been reported to vary across speaking situations and tasks (Bloodstein & Ratner, 2008), current results were not consistent with previous findings. Lim, Lincoln et al. (2008) and Nwokah (1988) did not find differences in stuttering severity across different speaking situations (e.g., clinic, work, home, telephone, reading). Similarly, Druce et al. (1997) failed to find differences in the frequency of stuttering during high- (speaking with a stranger) and low-stress (speaking with a family member) speaking situations in bilingual or monolingual children who stutter. Language status and stuttering severity. While 55.6% of bilingual children were diagnosed with severe stuttering, only 16.67% of monolingual children were classified as severe (Stern & Log, 1948). Sex differences. Bilingual boys have been reported to show more severe stuttering compared to bilingual girls (Mohammadi et al., 2012). Physical concomitant. Similar to monolinguals, physical concomitants have also been observed in bilingual children who stutter during moments of stuttering (Dale, 1977; Howell & Davis, 2011; Karniol, 1992; Lee et al., 2014). Word class (function vs. content) and stuttering severity. Frequency of stuttering across word types maybe impacted by the age of the speaker. In the Maruthy et al. (2015) and Schäfer and Robb (2012) studies which included bilingual adults, higher rates of stuttering were found on content words in the L1 (Kannada or German), and function words in the L2 (English). In contrast, findings from Gkalitsiou et al. (2017) suggests an interaction between age, word type, and speech task. In their study of four Spanish-English bilinguals between 3;10 to 6;8, higher rates of stuttering were found on function compared to content words regardless of language proficiency for conversation and narrative tasks in Spanish, and narrative tasks in English. However, the frequency of stuttering was similar for function and contents words for the English conversation tasks.

#### 3.3.2. Development

Few studies have reported on the development of bilingual children who stutter, accordingly, is it unclear if current findings are robust. Motor. Bilingual children who stutter showed slower reaction time, produced less syllables and more errors, and took longer to complete mono- and long-syllabic diadochokinetic tasks compared to typically developing bilingual children and bilinguals with dyslexia (Malek et al., 2013). Language. In a treatment study of four preschool bilingual children who stutter, Lattermann et al. (2005) reported normal speech articulation, and receptive and expressive language development in their L1 (English) prior to treatment. Cognitive. Findings related to cognitive development have been ambivalent. While some studies report academic delays between 1 to 2 years and lower IQ in bilingual children who stutter compared to bilinguals children who do not stutter (Aron, 1962; Travis et al., 1937), other studies have not supported these findings (Howell, Davis, Williams et al., 2009; Ralston, 1981). A recent study by Howell, Davis, Williams et al. (2009) found similar performance in English, Mathematics and Science for bilingual children who stutter, monolingual children who stutter, and typically developing bilingual children. Interestingly, Ralston (1981) reported that 88% of bidialectal child who stutter have been noted to show above-average intelligence by their teachers. Physical development/ health. Travis et al. (1937) reported a higher percentage of bilingual children who stutter than monolingual children who stutter who have poor dental development (40% vs. 25.8%) and tonsillar disorders (13.8% vs. 6.5%). However, a lower percentage of bilinguals who stutter compared to monolinguals who stutter were found to have poor skeletal development (10.8% vs/ 12.9%) and vision (9.2% vs. 22.6%; Travis et al., 1937). Handedness/laterality. Aron (1962) reported a higher prevalence of sinistrality in bilingual females who stutter (23.8%) compared to bilingual females who do not stutter (2.3%), however, no differences were found between bilingual males who do and do not stutter.

#### 3.3.3. Comorbidity

Stuttering has been found to be comorbid with other disorders in bilingual children although rates of comorbidity were ambivalent (Andrews et al., 2016; Koushik et al., 2009; Lincoln et al., 1996). In clinical studies, concomitant language, speech, and behavioral disorders (e.g., expressive language, receptive language, articulation, phonology, and attention deficit hyperactivity disorder [ADHD]) have been reported in about a third of bilingual children who stutter (Andrews et al., 2016; Koushik et al., 2009). Further, in the Baker and Cantwell (1982) study, 29% of children with speech disorders, including bilinguals who stutter (n = 4), showed concomitant psychiatric disorders (based on DSM-III) compared to children with language (95%), and mixed speech-language (45%) disorders. In contrast to these studies, Andrews et al. (2012) did not find comorbidities in bilingual children who stutter (n = 5), while 80% (n = 4) of monolingual children who stutter presented comorbid disorders.

#### 3.4. Assessment and treatment

# 3.4.1. Identification

Findings suggest that identifying stuttering in bilingual children may be challenging. *Non-clinicians*. Typically developing children have been mislabeled as children who stutter by their teachers (Stern & Log, 1948). Nonetheless, a more recent study by Mirawdeli (2015) reported consensus between teachers and the Stuttering Severity Instrument (Riley & Bakker, 1994) in identifying children who stutter in bilingual English language learners. *Bilingual clinicians*. Misidentification of stuttering has also been reported for clinicians. Byrd, Watson et al. (2015) reported that 86% (n = 12) of Spanish-English bilingual speech-language pathologists (SLP) in their study incorrectly diagnosed a typically fluent Spanish-English bilingual child with stuttering, and 29% (n = 4) erroneously

identified the Spanish-English child who stutters as normally fluent based on Spanish and English audio speech samples. Van Zaalenop't Hof et al. (2009) found low agreement between two SLPs with experience with fluency disorders in diagnosing stuttering and cluttering in both bilingual and monolingual children and adults. Agreement was achieved in only 50% of the cases, 13% of the cases were identified as stuttering by one SLP but cluttering by the other, and 37% were diagnosed with only stuttering or cluttering by one SLP but with comorbid stuttering-cluttering by the other SLP (Van Zaalen-op't Hof et al., 2009). The Van Zaalen-op't Hof et al. (2009) study did not report diagnostic agreement separately for bilinguals and monolinguals. Bilingual versus monolingual clinicians. English-speaking SLPs who were bilinguals (e.g., English-Dutch, English-Mandarin, English-Indonesian) who were not exposed to Spanish, reported higher frequency of stuttering in reading samples and judged physical concomitants of bilingual Spanish-English children who stutter to be more severe relative to monolingual English-speaking SLPs (Lee et al., 2014). Despite this, there were high levels of agreement between the monolingual and bilingual SLPs in judging the overall stuttering severity of bilingual children in both the Spanish and English speech samples, and both groups reported higher rates of stuttering in Spanish (L1) compared to English (L2; Lee et al., 2014).

# 3.4.2. Treatment latency, relapse, practices, and perceptions

Treatments for bilingual children who stutter varied widely in approach. While the primary component of some treatments was operant training, others used speech modification techniques, or a hybrid of approaches to achieve fluency. The latency of response to treatment also varied widely across and within treatment types, ranging from a few clinic sessions to more extended visits. Markedly, signs of relapse were found across all treatment types.

3.4.2.1. Operant treatments. Lidcombe Program. The preponderance of published research on treatment in bilingual children were focused on the Lidcombe Program (53%, n = 8; Bakhtiar & Packman, 2009; Harrison et al., 2004; Koushik et al., 2009; Lattermann et al., 2005; Lincoln et al., 1996; Rousseau et al., 2007; Vong et al., 2011, 2016). The program could be delivered in one or more languages and does not use any speech restructuring techniques (Bakhtiar & Packman, 2009). The mainstay of the program is parental verbal contingency (i.e., praising stutter-free utterances and correcting stuttered utterances), in addition to weekly sessions with a clinician to increase and maintain fluent speech. During Stage 1 of the program, 15 min parental feedback sessions administered three times per day, and weekly clinic visits lasting about 45 min were required to achieve around 1% SS (syllable stuttered; Kingston, Huber, Onslow, Jones, & Packman, 2003; Onslow & Packman, 1999). Once children achieve around 1% SS, the maintenance portion of the treatment is initiated. Clinic visits are gradually decreased during the maintenance stage, however, if a child fails to meet the criteria for %SS during any of the maintenance sessions, clinical visits are increased (Lincoln et al., 1996). The total number of visits required to achieve less than 1% SS ranged between 4 to 57 visits (Bakhtiar & Packman, 2009; Koushik et al., 2009; Lincoln et al., 1996). However, some children were unable to achieve this criteria (Koushik et al., 2009; Vong et al., 2016). For both bilingual and monolingual children who stutter, higher mean length utterance (MLU) before treatment was correlated with shorter treatment time while higher receptive language ability was correlated with longer treatment time (Rousseau et al., 2007). Up to 92% reduction in %SS across languages spoken have been reported post-treatment (Bakhtiar & Packman, 2009; Koushik et al., 2009; Lincoln et al., 1996; Vong et al., 2016). Lattermann et al. (2005) found a correlation between reduction in stuttering severity and increases in MLU, syntactic complexity, and vocabulary diversity (NDW) in bilingual children between 4 to 6 years old.

Relapses were commonly reported. Several studies found increased rates of stuttering, above the maintenance criteria after treatment in some children (Koushik et al., 2009; Lincoln et al., 1996; Vong et al., 2016). Parental feedback have been found to impact fluency after treatment (Harrison et al., 2004). In a study of 38 preschool monolingual and bilingual children, those who received parental feedback were observed to maintain or increase their fluency gains 4 weeks after therapy while children who did not showed a slight increase in their stuttering severity (Harrison et al., 2004). Koushik et al. (2009) reported that 80% of parents continue to provide feedback after the end of treatment although 60% found it challenging to participate in the program due to busy home schedules. The Lincoln et al. (1996) and Vong et al. (2016) studies included monolingual children.

Slow speech model. The slow speech model reported by Druce et al. (1997) also includes verbal feedback administered by clinicians. Clinicians modeled slow speech although the technique was not explicitly taught to children. During treatment children engaged in various speech tasks (e.g., repetition of single words, naming pictures, producing single word utterances when cued, conversation), and were praised or rewarded for fluent speech (Druce et al., 1997). Children received 4.5 h of therapy per day over 5 days while parents attended about 6 h of workshops and observation sessions over the same period (Druce et al., 1997). The treatment also included a 3-month maintenance phase where clinical visits were gradually reduced to weekly visits in the first month, fortnightly in the second month, and a final visit in the third month. By the third clinical visit, Druce et al. (1997) reported a 77% reduction in %SS (from 7.6% SS to 1.75% SS). The study included monolingual children who stutter, but there were no significant group differences (bilinguals vs. monolinguals) in reduction rates. Data collected up to 21 months following the end of treatment indicate relapse in 80% (n = 12) of children, with increases in %SS ranging between 2.48% to 3.83%. The language used in treatment was not reported but all children including bilinguals spoke English.

3.4.2.2. Speech restructuring. Syllable-timed speech. The treatment was a hybrid of speech restructuring and parental feedback (Andrews et al., 2012, 2016; Trajkovski et al., 2011). Children were taught to break up their utterances into syllables to achieve fluent speech and parents were instructed to provide verbal feedback on their child's use of the technique (Andrews et al., 2012). Each parent and child dyad attended 45 min weekly sessions in the clinic, and techniques were practiced 4 to 6 times a day, up to 5 to 10 min each time outside the clinic in everyday situations (Andrews et al., 2016; Trajkovski et al., 2011). When stuttering was reduced to 1.5% SS, children entered into the maintenance phase (Andrews et al., 2016). Across studies, a mean of 11 or 12 visits,

ranging between 4 to 22 clinic sessions, were required to attain < 1.5% SS or a 50% reduction in stuttering frequency (Andrews et al., 2012, 2016; Trajkovski et al., 2011). During maintenance, follow-up visits were gradually reduced (Andrews et al., 2012). Languages used in the syllable-timed treatment were not reported, although adequate proficiency in English was an inclusion criteria for all studies. Across studies, bilingual children showed a mean reduction of 77.58% in stuttering severity (from 7.54% SS to 1.68% SS) while monolinguals showed a mean reduction of 86% (from 7.85% SS to 1.1% SS) from pretreatment to 12-months post-treatment (Andrews et al., 2016; Trajkovski et al., 2011). Increased fluency was correlated with lower self-reported avoidance behaviors, more satisfaction with their speech and improved self-reported quality of life (Andrews 2012, 2016). In all three studies, some children showed increased stuttering severity after treatment or failed to meet the fluency criteria. All studies included monolingual children who stutter.

Smooth Speech. Bilingual Mandarin-English speaking children learned to use target speech pattern modeled by clinicians, initially at a slower speech rate but increasing with mastery of the technique (Lim et al., 2015). The treatment was conducted in English over 3 days, 8 h a days, with six 2 h follow-up sessions once a week. Across languages spoken, an 84% reduction in stuttering frequency in English (from 8.5% to 1.4% SS) and 80% reduction in Mandarin (from 9.17% to 1.81% SS) were observed immediately after the 3-day treatment (Lim et al., 2015). Further reduction in stuttering severity was reported four weeks after the end of treatment for English (1.4% SS) and Mandarin (2.78% SS). However, increases in stuttering severity in both English (45% increase in %SS) and Mandarin (9.6% increase in %SS) were found 12 weeks post-treatment. This study also included adult bilinguals who stutter.

#### 3.4.3. Generalization

Treatments delivered in one language, typically English, have been found to reduce the frequency of stuttering in all languages spoken (Bakhtiar & Packman, 2009; Lim et al., 2015; Vong et al., 2011). *Delivery in one language.* Vong et al. (2011) and Lim et al. (2015) reported that treatment effects generalized to the non-treatment language. However, a non-significant but greater reduction in stuttering severity was found for the language used in treatment (English, 70% decrease in %SS) compared to the untreated language (Mandarin, 67% decrease in %SS) in bilingual adolescents and adults (Lim et al., 2015). *Delivery in multiple languages.* Bakhtiar and Packman (2009) reported reductions in the frequency of stuttering (near or at 0% SS) for both Baluchi (L1) and Persian (L2) in a school-age (8;11) boy where the parent treatment components were conducted in both languages, and the clinical component in Persian (Bakhtiar & Packman, 2009).

#### 3.4.4. Practices and perceptions

Two studies which surveyed treatment practices in the United Kingdom found varying approaches when working with bilingual Asian children who stutter (Wright & Sherrard, 1994a, 1994b). Some respondents discussed their bilingual caseloads with interpreters or co-workers (68%, n = 55), or bilingual SLPs (27%, n = 22); and attended training courses (54%, n = 44) and acquired materials (42%, n = 34) for working with Asian clients (Wright & Sherrard, 1994a). About 75% (n = 54) of non-Asian SLPs reported lower perceived success with their Asian clients than their non-Asian clients (Wright & Sherrard, 1994b). Further, SLPs who were dissatisfied with the quality of interpreter services were more likely to perceive lower success with their Asian clients (Wright & Sherrard, 1994b). Respondents also reported that the adult relative of the child who stutters (96%, n = 52), a friend of the child's family (54%, n = 29), relatives of the child who stutters (74%, n = 40) typically or sometimes served as the interpreter (Wright & Sherrard, 1994a). Additionally, only 2% of SLPs reported delivering therapy across languages for bilingual children who stutter (Wright & Sherrard, 1994a).

#### 3.5. Culture

# 3.5.1. Attitudes toward children who stutter and perceptions of stuttering

Attitudes toward children who stutter and perceptions of stuttering may be influenced by cultural values and norms. Dale (1977) reported that parents of their Cuban-American children who stutter "become upset and try to embarrass them when they 'stutter' in Spanish or forget a word" and "immediately label their sons as stutterers and chastised them for speaking 'imperfect' or dysfluent Spanish" (p. 312). In a more recent study, 22.4% of Hispanic American college students believed that the pressure placed by parents on a child to speak two languages caused stuttering, and 39.4% agreed with the statement that switching from the L1 (Spanish) to L2 (English) was the source of stuttering (Tellis, 2008). Bilingual Cantonese/English speaking immigrants and monolingual second generation Asian-Americans have been found to differ in their perceptions of children with communication disorders including children who stutter (Bebout & Arthur, 1997). Second generation monolingual respondents were more likely to agree with the statements that "Severe child stutterers have trouble making friends" and "It is normal for other children to make fun of a badly stuttering child" compared to bilingual immigrant respondents (Bebout & Arthur, 1997). In contrast, the bilingual respondents were more likely than the monolingual respondents to agree with the statement that those with speech disorders could reduce the severity of their disorder if they "tried hard" (Bebout & Arthur, 1997).

#### 3.5.2. Cultural expectations and experience

Culture has been hypothesized to play a role in the development of stuttering. *Expectations*. Dale (1977) proposed that parental displeasure with their child for speaking "imperfect" Spanish and labeling of typical disfluencies in four Cuban-American children was the root of stuttering (p. 312). Similarly, Nwokah (1988) suggested that the emphasis on eloquence and fluency, particularly for first-born males, in Igbo culture may increase the risk of stuttering. Differences in societal expectations and values for males and females are also thought to impact the distribution of stuttering across sexes (Ralston, 1981). First, Ralston (1981) posits that the

smaller disparity between the male and female prevalence of stuttering in Nevis is an outcome of a highly matriarchal society, although no further details are given. Second, the higher prevalence of females who stutter in urban compared to rural areas but the opposite pattern for males was hypothesized to be the result of higher societal pressures for girls in urban areas and boys in rural areas (Ralston, 1981). Aron (1962) reported similar prevalence of stuttering in urban and rural areas in Johannesburg, however, the distribution across sex was not reported. *Experience*. Nwokah (1988) proposed that experience may interact with stuttering severity. Igbo children who had more negative experiences in school stuttered more in English (language of instruction) while those who had difficulty at home stuttered more in Igbo, their home language (Nwokah, 1988).

#### 3.5.3. Implications for treatment and research findings

Cultural practices and historical perspectives may influence treatment practices and interpretation of findings. Consequences of cultural and treatment practices. First, Tetnowski et al. (2012) reported that parents of a 4-year old bilingual boy who speaks Tamil at home were asked to speak English with the child despite one parent not being able to speak English and the other parent had only minimal proficiency. Second, a bilingual boy who stutters (4;9) was reported to refuse to speak in Mandarin (L1) at the end of treatment which was conducted in English (L2; Vong et al., 2011). Third, Vong et al. (2010) found that Malaysian parents frequently code-switched to English (typically the language of treatment) when praising their child although they spoke Malay or Mandarin as their first language. Historical research. Contemporary understanding and perspectives may influence how findings are interpreted. Travis et al. (1937) proposed that the higher prevalence of stuttering in bilinguals in their study was related to the bilingual home environment. The authors stated, "We cannot be certain, however, that this difference is due solely to the factor of bilingualism. It may be due to the economic insecurity and emotional instability found in many foreign homes as a result of the recent economic depression, or it may be due merely to a confusion which arises from being placed in a totally strange and new environment." (Travis et al., 1937, p. 189). In a clear example of a prejudiced interpretation, Aron (1962) stated that "Whereas the European stutterer usually attempts less bizarre bodily movements for fear of severe reaction from his audience, the less sophisticated African audience might conceivably accept these bodily movements of the stutterer more readily since the Africans themselves take pleasure in using gesticulations" (p. 123).

#### 4. Discussion

The need for more systematic research on prevalence, risk factors, cross-linguistic symptomology, assessment and treatment in bilinguals who stutter is a recurring theme (Finn & Cordes, 1997; Packman, Onslow, Reilly, Attanasio, & Shenker, 2009; Ratner & Benitez, 1985; Shenker, 2011; Van Borsel et al., 2001; Van Borsel, 2011). Our findings echo the need to close gaps in our understanding of how stuttering interacts with bilingualism, particularly during the early stages of development, and to translate knowledge into clinical practice. Some lines of research including those focused on uncovering the extent to which stuttering modulates the development of bilingual children, and the impact of culture on the disorder have received little attention. Although our review identified points of convergence with monolingual research, findings do not provide a unified view of stuttering, with potentially negative consequences for how we manage and treat the disorder in bilinguals.

# 4.1. Convergent findings

Findings from this review confirm the sexually dimorphic and familial nature of stuttering. First, similar to monolingual children who stutter, there is a *higher male to female ratio of stuttering* in bilingual children (Yairi & Ambrose, 1999). In monolinguals, this ratio is driven by higher rates of recovery in females (Ambrose et al., 1997; Yairi & Ambrose, 1999). Second, *rates of recovery are also higher for bilingual girls* compared to bilingual boys. Third, the *onset of stuttering is earlier for bilingual girls* compared to boys which mirrors observations in monolingual children (Yairi & Ambrose, 1992; Yairi, 1983). Fourth, the majority of bilingual children who stutter report a *positive family history of stuttering*, similar to monolingual children who stutter (Ambrose, Yairi, & Cox, 1993; Buck, Lees, & Cook, 2002). Other sexually dimorphic and familial patterns in epidemiology found in monolingual children who stutter have yet to be substantiated in bilingual children who stutter. For example, monolingual girls experience earlier recovery and are less likely to have a familial history of stuttering compared to monolingual boys (Ambrose et al., 1993, 1997; Yairi & Ambrose, 1999). Monolingual children with a family history of recovery are more likely to recover themselves (Ambrose et al., 1993, 1997). In monolinguals, the earlier onset of stuttering for girls may be related to earlier language development, and across sexes, stronger language skills are associated with a higher likelihood of recovery (Watkins & Yairi, 1997; Yairi et al., 1996).

In terms of intervention, post-treatment relapse was also commonly reported in bilinguals. The majority of treatment studies in the current review reported relapse (i.e., increased rates of stuttering compared to levels immediately after treatment) in some children within a year of treatment completion. In general, higher pre-treatment stuttering severity, slower speech rates, and low trait anxiety (general response anxiety) have been identified as predictors of relapse (Craig, 1998; Hancock & Craig, 1998). It is likely that these same predictors apply to bilingual children, although the extent to which bilingualism interacts with these factors is unclear. Our review identified points of convergence with monolingual research, confirming specific aspects of stuttering across different populations. Nonetheless, the scope of these findings is narrow and based on a limited number of studies. More importantly, integration of these findings into conventional frameworks that could elucidate the interaction between bilingualism and stuttering, or the nature of the disorder is generally absent.

#### 4.2. Gaps in the literature

Knowledge gaps identified in the current review reflect the paucity of systematic cross-linguistic approaches to stuttering research. Definition of bilingual. In general, research related to bilingualism across disparate fields (e.g., linguistics, cognition, education) have been inconsistent in their definition of bilingual, and as a whole have struggled to reach a consensus on the operational definition (Surrain & Luk, 2017). One of the most significant challenges in exploring the link between bilingualism and stuttering is the lack of a common and measurable definition for bilingual. A majority of the studies identified in this systematic review did not operationally define or quantify bilingualism which limits the interpretation and application of existing knowledge. Further, when bilingualism was defined, different criteria were adopted. These disparities make it difficult to compare findings across studies and may contribute to contradictory findings. However, a common definition of bilingual across studies and individuals may be difficult to achieve as language exposure, use, and proficiency lie on a continuum (Luk & Bialystok, 2013). A multidimensional approach using self-reports, interviews, and testing across languages spoken could resolve some of these challenges (J. Anderson, Mak, Chahi, & Bialystok, 2018; Sheng, Lu, & Gollan, 2014). Epidemiology. Epidemiological findings do not offer a comprehensive understanding of how bilingualism interacts with stuttering. Inconsistent reports of prevalence rates, age of stuttering onset, and rates of recovery in bilingual children offer conflicting perspectives on whether the acquisition of a second language increases the risk of developing stuttering and lowers the probability of recovery. Bilingual profile and development. Current findings offer a fragmented view of bilingual development. Studies examining language proficiency, cross-linguistic stuttering severity, and motor and linguistic development have produced ambivalent reports. Further, only one study has investigated the link between developmental factors and recovery in bilingual children. Although comorbid speech, language and behavioral disorders has been observed in bilingual children who stutter, it is unclear if bilingual children show similar susceptibility to comorbidity, compared to their monolingual peers. To answer these questions, studies examining bilingual children who stutter must operationally define and quantify bilingualism. Assessment and treatment. Results indicate that identifying stuttering in bilingual children may be challenging, which is consistent with findings of Byrd, Bedore, and Ramos (2015) in typically developing bilingual children. Relatedly, studies by Dockrell, Howell and colleagues identified several areas which pose challenges to identifying and treating bilingual children (Dockrell & Howell, 2015; Dockrell, Howell, Leung, & Fugard, 2017; Howell, 2013). First, SLPs and educators cite difficulty in differentiating between speechlanguage impairments from language proficiency in bilingual children (Dockrell & Howell, 2015; Dockrell et al., 2017). Accordingly, utilizing assessments that are able to distinguish between behaviors related to language proficiency and symptoms of stuttering such as the Universal Non-Word Repetition (UNWR) test are crucial (Howell et al., 2017). Second, a majority of SLPs and educators associate speech-language difficulties with being bilingual (Dockrell & Howell, 2015; Dockrell et al., 2017). Third, SLPs and educators, including those working with bilingual children, cite the lack of training as a barrier to working with children with speechlanguage impairments (Dockrell et al., 2017). Collectively, these findings suggest that bilingual children may be disproportionately referred for services, and treatment remains a challenge. However, establishing the efficacy of treatments, including latency to response, and causes of relapse may prove difficult due to the heterogeneity of bilinguals (e.g., in language combination, crosslinguistic proficiency), and without a clear understanding of bilingual development. An effective treatment strategy for bilinguals will be difficult to achieve without the ability to identify bilingual children who are at risk, and without a comprehensive understanding of bilingual development and factors that influence recovery or relapse in this population. Culture. Findings suggest cultural beliefs and practices influence treatment delivery and strategies with consequences beyond fluency for the child who stutters. Results also point to the need for culturally competent research and interpretations. Investigations to determine how multicultural factors interact with stuttering, from the perspective of the child who stutters and clinician, will be an important area for future research, and findings could impact treatment recommendations, practices and efficacy (e.g., whether to limit or eliminate the use of another language, particularly, a non-English language; Finn & Cordes, 1997).

Closing these gaps in knowledge will require large scale, systematic studies to determine within-group differences in bilingual children who stutter and between-group differences relative to monolingual children who stutter. Longitudinal epidemiological studies will be necessary to establish factors related to recovery and chronicity. One challenge to conducting these types of large-scale studies is recruitment. However, open data repositories such as the UCL Archive of Stuttered Speech (UCLASS), TalkBank, and FluencyBank – specific to the study of stuttering – could support such efforts (Brundage, Corcoran, Wu, & Sturgill, 2016; Howell, Davis, & Bartrip, 2009; Ratner & MacWhinney, 2018). Our understanding of cross-linguistic manifestations of stuttering are based on relatively few languages. There are over 6,000 languages spoken, accordingly, it is unclear if findings generalize to different linguistic systems (S. R. Anderson, 2010). Thus, defining and quantifying proficiency across different linguistic systems will be crucial to establishing comprehensive descriptions of the bilingual profile and allow comparisons across studies and linguistic systems to gain meaningful interpretations of findings (Coalson, Peña, & Byrd, 2013). Such knowledge would scaffold the development of differentiated assessment tools and evidence-based treatments grounded in the specific linguistic typology and cultural background of the child, and support a critical evaluation of findings in bilinguals (Shenker, 2011).

# 4.3. Insights from typically developing bilinguals

Growing rates of bilingualism have supported increased interest in the development of bilinguals, mostly in typically developing children (Bialystok, 2011; De Houwer, 2017). Results from this review also point to a surge in research on bilingual children who stutter. Nearly half of the articles identified in this study were published after the last review by Van Borsel (2011), despite this, important questions remain. Findings in typically developing bilinguals and models of bilingualism may provide insights into the nature of stuttering in bilingual children. A study by Byrd, Bedore et al. (2015) which reported higher rates of SLD in Spanish

compared to English in typically developing bilingual children point to the possibility of bilingual children being over-identified with stuttering. Also, research in typical bilinguals suggests that language-switching, which requires the ability to select and maintain representations of the target language while inhibiting the non-target, enhances executive functions (EF; encompassing inhibitory control, working memory and attention control skills) in this population (Adesope, Lavin, Thompson, & Ungerleider, 2010; Bialystok & Martin, 2004; Bialystok, 2015; Thorn & Gathercole, 1999). A recent investigation by Hartanto, Toh, and Yang (2018) which surveyed 18,200 children between 5 to 7 years old reported that bilingualism attenuates the effect of socioeconomic status on EF. Executive function components, including working memory and attention, are proposed to play key roles in speech processes, thus, bilingualism may have direct consequences for stuttering (Vasic & Wijnen, 2001; Baddeley, 2002; Hickok, Buchsbaum, Humphries, & Muftuler, 2003). Nonetheless, it is unknown if EF components are impacted by stuttering in bilingual children.

#### 4.4. Could models of bilingual memory shed light on current findings?

Many models of bilingual memory organization characterize bilingual memory as separate lexical systems that map onto shared conceptual representations (Chen & Leung, 1989; Kroll & Curley, 1988; Kroll & Sholl, 1992; Potter, So, Von Eckardt, & Feldman, 1984). Among so-called "balanced" bilinguals, the representation for two lexical items in the L1 and L2 (i.e., cat and kucing for an English-Malay bilingual) are represented separately and both map onto the same conceptual representation. The Bilingual Interactive Activation Model of word recognition (BIA; Dijkstra & Van Heuven, 2002) proposes that bilinguals have hierarchically organized word representations that are activated and competing for attention. It is hypothesized that bilinguals manage their attention between two (or more) jointly activated languages at all times, diverting attention away from the language not in use (Luk, Anderson, Craik, Grady, & Bialystok, 2010; Bialystok, Craik, & Luk, 2012). The Inhibitory Control Model (Green, 1998) posits that bilinguals experience competition from both continuously activated languages at all times, including competition between lexical representations during speaking, and must use cognitive control resources to resolve the competition from the activated non-target language (Hermans, Bongaerts, De Bot, & Schreuder, 1998). Neuroimaging research generally supports a common neural network for processing the L1 and L2 among bilinguals, including shared structures for language control and lexical selection (for a review, see Abutalebi & Green, 2007; Hernandez, 2009). Both languages are continuously active when a bilingual listens, reads, and plans speech in any of their multiple languages (for a review see Kroll, Dussias, Bice, & Perrotti, 2015). Language interactions are bidirectional, each language impacts processing of the other (see review, Kroll et al., 2015). Regular bilingual activity is posited to necessitate: selectively attending to a broader range of linguistic and social cues (Friesen, Jared, & Haigh, 2014), inhibiting lexical and semantic representations in a non-target language while using another (Bialystok & Viswanathan, 2009; Gandolfi, Viterbori, Traverso, & Usai, 2014; Martin-Rhee & Bialystok, 2008; Poarch & Van Hell, 2012), and constraining language production to the target language (Kovács, 2007). In these bilingual models, allocation of resources to resolve attentional competition would reduce or exceed available resources for linguistic processes, increasing the likelihood of linguistic and selection errors that manifests as disfluencies (Bergmann, Sprenger, & Schmid, 2015). These models do not make a distinction between the production of stuttering-like and typical disfluencies. It is plausible that reduced or inadequate resources is a necessary condition for normal fluency breakdown but insufficient for stuttering. The manifestation of stuttering in bilinguals may arise from the interaction between multiple factors including competition for attention resources, accordingly, a multifactorial, developmental approach will be an important strategy for future investigations in bilingual children who stutter.

These bilingual models (BIA, Inhibitory Control Model, and neuroanatomical) are consistent with frameworks for stuttering. Consistency with the Suprasegmental Sentence Plan Align (SPA; Karniol, 1995) and Neuroscience model of stuttering (Nudelman, Herbrich, Hoyt, & Rosenfield, 1989) will be discussed. In the SPA, linguistic deficit (e.g., lexical, syntactic) is a necessary condition for stuttering (Karniol, 1995). When speakers revise their utterance, corresponding suprasegmental features, such as rhythm and memory, would also need to be modified and realigned with the new utterance. Stuttering is proposed to occur at this juncture, as more time is required to revise the linguistic features of the utterance, readjust the fundamental voice frequency, and realign the suprasegmental plan, as a consequence of language deficits. For bilinguals, more disfluencies are hypothesized to occur in the less proficient language, as more revisions and realignment are required to resolve competition with the predominant plan (i.e., the dominant language). However, our review findings point to variability in cross-linguistic stuttering, while most studies report higher frequency of stuttering in the less proficient language, others found the opposite trend. It is plausible other factors, such as increased self-repairs, linguistic complexity, and social context may impact cross-linguistic severity in addition to the factors proposed in this model, thus, more research is warranted to uncover how various factors interact in bilinguals. The Neuroscience model of stuttering (Nudelman et al., 1989) proposes that disfluencies result from moments of "instability" among speech motor control loops; bilingualism requires additional processing and thus, increases loop instability. Van Borsel et al. (2001, p. 186) similarly posit that if stuttering is due to "overload" of brain structures associated with language learning, early bilinguals using these same structures for dual language learning would be even more "overloaded" and likely to stutter. Research indicates that bilinguals access word meaning just as quickly as monolinguals, but perform more slowly and make more errors during picture naming tasks in the dominant language (Gollan, Montoya, Fennema-Notestine, & Morris, 2005). Typically developing bilinguals also report more tip-of-the-tongue disfluencies (i.e., retrieval failures where the speaker feels they know the word but cannot immediately recall it, sometimes recalling characteristics such as initial sound; Gollan et al., 2005). Prior researchers have also noted increased disfluencies among bilinguals who stutter associated with switching between languages (Aguis, 1994; Cabrera & Ratner, 2000; Mussafia, 1967), including codeswitching events, however, it is unclear if a switch causes or is the result of disfluencies (i.e., the speaker switches languages as a coping strategy; Karniol, 1992). The Adaptive Control Hypothesis (Green & Abutalebi, 2013) posits that context is also relevant for cognitive control among bilinguals, such that different switches (i.e., switching languages within a single conversation as compared

to switching languages from one interlocutor to another) require different control processes.

Alternatively, bilingualism may be advantageous to fluency by enhancing speech-related EF components (see section on Insights from typically developing bilinguals). The Conditional Routing Model (CRM) proposes that language-switching enhances EF in dual language speakers, and this interaction lies in the basal ganglia (Stocco, Yamasaki, Natalenko, & Prat, 2014). The basal ganglia selects the appropriate rules for language by strengthening or rerouting specific signals, and the ability to filter and reduce storage of irrelevant information or distractors, which improves with practice and is mediated by the basal ganglia increases EF capacity (McNab & Klingberg, 2007; Stocco et al., 2014). A study which found an inverse correlation between bilingual proficiency and attention deficit in 5 to 16 year old bilingual Spanish-English children with psychopathology suggest this enhancement may also be present in atypical developing systems (Toppelberg, Medrano, Morgens, & Nieto-Castañon, 2002). Neuroimaging research suggest that atypical white and gray matter development in the basal ganglia is characteristic of stuttering (Alm, 2004; Beal, Gracco, Brettschneider, Kroll, & Luc, 2013; Etchell, Johnson, & Sowman, 2014; Lu et al., 2010). If bilingualism also augments basal ganglia function in children who stutter, it is plausible that with age, as bilingual proficiency increases with practice, bilingual children may experience greater fluency. If so, bilingual children who stutter may experience recovery at an older age, and the likelihood of recovery will be greater in more proficient compared to less proficient bilingual children who stutter. While not the focus of the current paper, it is relevant to note that there are no reports of increased disfluency associated with onset of an L2 among adult learners, possibly indicating that simultaneous or sequential bilingualism in early life is distinct, from a neurological perspective and, correspondingly, in relation to stuttering. Nonetheless, there is a paucity of research on recovery in bilingual children who stutter, thus, how recovery is distributed across age and proficiency in bilingual children is unclear.

#### 4.5. Limitations

Current findings should be understood in the context of some limitations. First, studies not published in peer-reviewed journals, such as conference abstracts and posters, and articles not published in English were excluded due to inability to critically evaluate the methodology or overall study, however, this may have biased our findings. Restricting the language of the search and inclusion of English only publications may affect some topic areas more than others (see Hartling et al., 2017; Morrison et al., 2012), nonetheless, it is unclear which topic areas were more impacted in this review. Further, the exclusion of non-empirical sources may have introduced a sampling bias, however, these sources generally did not contain enough information (e.g., age of participants) to ascertain whether the inclusion or exclusion were met. Second, we did not specify a time range for the search, accordingly, inclusion of early investigations with different criteria of bilingualism and stuttering may have added to this bias. A third source of bias may be the inclusion of epidemiological findings based on clinical cohorts. It is plausible that the odds of finding positive results may be higher in clinical cohorts. Further, the review included case studies or studies with single-subject research design. In general, although case studies and single-subject designs could provide a means to explore complex interactions, their generalizability is constrained compared to group designs. In stuttering, single-subject design could be an important tool to determine the efficacy of stuttering treatments, however, their utility in exploring other aspects of the disorder such as epidemiology is limited. Nonetheless, singlesubject and group studies may fulfill complementary functions in answering fundamental questions about stuttering. Fourth, we included studies that did not operationally define or quantify bilingualism, constraining the ability to compare results across studies or children with varying levels of proficiency and age of L2 exposure. The heterogeneity of bilinguals would therefore invite a cautious interpretation of these studies. Overall, the number of published studies on bilingual children who stutter is relatively small, more research is needed to determine if findings from this review are robust.

#### 5. Conclusion and New Directions

This systemic review echoes a recurring theme, current understanding of bilingualism and stuttering is limited, and more research is warranted. Epidemiological and developmental findings do not present a consistent view of the effects of stuttering on bilingual children, and the underlying nature of disorder. The long-term efficacy of treatment for bilinguals and factors that impact treatment outcomes in bilinguals are unclear. Further, despite the significance of culture to bilingualism, this area of research has not received much attention. Results from this review also underscore clinicians' challenge to identify and treat bilingual children who stutter. Without a comprehensive, unified framework that accounts for differentiated findings in bilinguals, clinical practice must rely on our limited knowledge of this population and insights from monolinguals, with potentially negative consequences. Nonetheless, there are points of convergence with findings in monolinguals across a limited subset of investigations, mainly related to sexual dimorphism and familial risk. However, more coherent cross-linguistic research and novel approaches are needed beyond validation to elucidate the relationship between bilingualism and stuttering, and for that matter, the nature of stuttering itself. Findings from this systematic review point to the applicability of bilinguals as an approach to test theories and assumptions about the disorder. Some recommendations for future research include: (a) large-scale, longitudinal studies to uncover the risk and prognosis of stuttering in bilinguals, including those related to language acquisition, (b) longitudinal studies to determine the effects of stuttering on speechrelated cognitive components impacted by bilingualism, (c) large-scale and international collaborations to examine cultural and language-specific factors in the development and presentation of stuttering, and (d) bilingualism must be operationally defined and quantified to enable comparisons across studies. As the number of research focused on bilingual children who stutter continues to increase, what we learn from them will bring us into closer proximity to a comprehensive, unified framework for stuttering.

If future research indicates that simultaneous early bilingualism or the addition of an L2 among sequential bilingual children does increase disfluencies, we must be cautious and mindful when drawing implications. Delaying addition of an L2 or eliminating

bilingual education for children at risk for stuttering is not a viable "therapeutic solution" in many cases (i.e., children from minority language backgrounds in which the majority language cannot be used in the home, children from settings in which L1 schooling is not available, children growing up in bilingual countries in which use of multiple languages is needed for daily life). For minority language children, bilingual schooling is often preferable over L2 only "sink-or-swim" programs for language development and academic outcomes (Bali, 2001). Suggesting minority language parents of children who stutter instead use only majority language in the home may be even worse, potentially resulting in less language input overall, lower quality input, less robust models (Hoff, 2018), not to mention possible negative impacts on socio-cultural identity and emotional wellbeing. Instead, the field should focus on research that would lead to treatments tailored to the reality of children who are bilingual by nature of birth and who stutter.

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#### Appendix A. Supplementary data

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Ai Leen Choo is an Assistant Professor in the Department of Communication Sciences and Disorders at Georgia State University. Her research aims to understand the effects of stuttering on cognitive development in monolinguals and bilinguals, and how specific cognitive factors are involved in the trajectory of stuttering. She received her Ph.D. in Speech and Hearing Science from the University of Illinois at Urbana-Champaign.

Sara A. Smith is an Assistant Professor in the College of Education at the University of South Florida. She received her Doctorate in Applied Linguistics and Second Language Acquisition from the Department of Education at the University of Oxford. Her research interests include within-population variation among bilinguals, typical and atypical language development, and cognitive and educational implications of bilingualism.