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Prevalence and features of comorbid stuttering and speech sound disorder at age 4 years

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ABSTRACT

Background: Stuttering and speech sound disorder may co-occur during early childhood, although the exact rate of comorbidity in a community-cohort sample remains unknown. In isolation, both disorders have the potential for long-term negative effects. Comorbidity rates of 16%–46% reported in previous studies were based on parent report, speech-language therapist surveys, case file audits or direct observation studies from clinical samples. Rigorous methodology utilising a prospective, longitudinal community-cohort design is required to support these previous findings.

Aims: First, to identify the proportion of children with comorbid stuttering and speech sound disorder at 4 years of age drawn from a community-cohort study. Second, to compare demographic and clinical features of this comorbid diagnosis group compared to children with no diagnosis of either disorder, or those with either disorder in isolation.

Methods & procedures: Participants were drawn from a prospective, longitudinal community cohort study (the Early Language in Victoria Study) at 4 years of age (n = 1607). Demographic and clinical features for comparison were theoretically driven and included: gender, birth history, feeding status, speech and language status, family history of communication difficulties, maternal education, maternal vocabulary, maternal mental health and socioeconomic status.

Outcomes & results: Of the 160 children diagnosed with stuttering between 2 and 4 years of age, 6.88 % (n = 11) also had a speech sound disorder. Given the small sample size and number of comparisons performed, there was insufficient evidence to rule out that group differences observed were not simply due to chance.

Conclusions & implications: The prevalence of comorbid stuttering and speech sound disorder was lower in a community cohort compared to that reported in clinical studies. Higher rates reported in clinical samples may be due to increased parental help-seeking behaviour when the two disorders co-occur. Subsequently, these children may present to clinics more frequently. Accurate representation of prevalence allows for population specific research on best practice assessment and intervention. Currently little is known about how best to manage this caseload, therefore more research is required in this area, including the determination of prognostic variables to provide efficient and effective management.

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1. Introduction

Communication impairment in childhood is relatively common and may involve a number of domains including speech, language, fluency (e.g., stuttering), voice, and/or literacy (Speech Pathology Australia, 2016). Additionally, disorders involving these domains can co-occur. One commonly reported example is when stuttering co-occurs with speech sound disorder (SSD). Stuttering is a neurobiological disorder of communication (Drayna & Kang, 2011) which disturbs the flow of speech (Onslow et al., 2003; Sowman, Crain, Harrison, & Johnson, 2014), and typically appears between the ages of 2 and 4 years (Reilly et al., 2013). Speech sound disorder is a broad term, but here we focus on atypical phonetic or phonological speech errors, or delayed speech errors, which occur beyond age-expected norms (Morgan et al., 2017).

The potential negative impact of stuttering is well documented, and can be long reaching across the lifespan (Blood & Blood, 2007; Iverach et al., 2009; Langevin, Packman, & Onslow, 2009; O'Brian, Jones, Packman, Menzies, & Onslow, 2011). For example, stuttering in the preschool years can elicit negative peer interactions (Langevin et al., 2009). In the school years, children who stutter have a heightened risk of being teased and bullied compared to their non-stuttering peers (Blood & Blood, 2007). Adults who stutter are at greater risk of developing mental health disorders (Iverach et al., 2009), and stuttering can effect educational (O'Brian et al., 2011) and vocational attainment (Klein & Hood, 2004).

Similar to stuttering, having a SSD may also have negative consequences on an individual across a lifetime (Law, Boyle, Harris, Harkness, & Nye, 1998; Mccormack, Mcleod, Mcallister, & Harrison, 2009). For example, if SSDs persist into the school years, children may be at-risk for poorer literacy outcomes compared to their typical peers (Nathan, Stackhouse, Goulandris, & Snowling, 2004). Additionally, other academic skills may be impacted, as well as poorer psychosocial outcomes and vocational attainment (Mccormack et al., 2009).

Given the potential negative impact across stuttering and SSD, it is important to be able to identify and better understand which children are at risk for these conditions when they co-occur to offset potential deleterious and possibly additive long-term effects. Yet the exact rate of co-occurrence of these disorders remains unknown and little is understood of potential prognostic variables in this field. It has been reported that between 16–46 % of children who stutter will also have co-occurring SSDs (Conture, Louko, & Edwards, 1993; Louko, 1995; Melnick & Conture, 2000; Ratner, 1995; Wolk, Blomgren, & Smith, 2000; Wolk, 1998). Yet the methodologies used to determine these rates in the surrounding literature have varied, and may explain the inconsistent findings (Unicomb, 2015). Three main methodological approaches employed in this field have been survey studies, retrospective file (chart) audits, and observation studies of clinical samples, discussed here in turn.

A number of survey studies have been conducted in this area over the past few decades (Arndt & Healey, 2001; Blood & Seider, 1981; Blood, Ridenour, Qualls, & Scheffner Hammer, 2003). Arndt and Healey (2001) and Blood and Seider (1981) both conducted surveys of school-based speech-language therapists (SLTs) in the US. Blood and Seider reported that 16 % (n = 170/1,060) of children who stuttered had a co-occurring articulation disorder whereas Arndt and Healey reported a co-occurring SSD rate of 28 % (n = 133/467). Diagnostic/eligibility criteria for both disorders differed across studies, as did the measures (e.g., standardized vs. non-standardized) used to determine the presence of both disorders. Data obtained from both studies were based on uncontrolled, subjective evaluations of participants. In an attempt to address this caseload subjectivity, Blood et al. (2003) conducted a nationwide survey of randomly selected school-based SLTs to determine the percentage of children who stutter with a co-occurring communication disorder. In contrast to the aforementioned studies, SLTs were required to provide a separate data sheet for each child on their caseload identified as stuttering. Speech-language therapists were advised that children could only be included based on verified disorders documented through case histories, client files and other professional/caregiver information. The results identified 2628 children who stuttered and were currently receiving therapy. Of these, 46.2 % exhibited a co-occurring speech (articulatory and/or phonological) disorder; SSDs were the most frequently reported comorbidity with stuttering. Almost 90 % of the children with co-occurring disorders were receiving treatment for both disorders. This was in line with previous findings where 91 % of children with co-occurring stuttering and SSD were receiving therapy for both conditions (Blood & Seider, 1981). These figures indicate that the rate of co-occurrence may be over-estimated, as children with co-occurring disorders are more likely to receive referrals for treatment (Nippold, 2004). As this was a nationwide survey, there were possibly broad variations in state-wide diagnostic criteria for both disorders.

The other main methodological approaches to studying rates of co-occurrence in this field are retrospective file audits (Yaruss, Lasalle, & Conture, 1998), and direct observation studies (Louko, Edwards, & Conture, 1990; Ratner, 1998; Riley & Riley, 1979; Ryan, 1992; St Louis & Hinzman, 1988; St. Louis, Murray, & Ashworth, 1991; Thompson, 1983). Yaruss et al. (1998) conducted a retrospective file audit on children who were referred to a speech language therapy clinic following caregiver concerns relating to fluency. The participants had a mean age of 54.7 months, and were administered an extensive battery of speech and language assessments. The authors reported that 37 % of participants presented with a concomitant phonological disorder. Others (e.g., Nippold, 2001) have cautioned these findings as the children were recruited based on concerns they were at risk for stuttering, and a firm diagnosis was not reported in this study. There is query around how many children were actually stuttering, given only approximately half the children were subsequently referred for fluency intervention.

Of the four direct observation studies conducted since 1990, two have reported clinical prevalence for co-occurring stuttering and SSD (Louko et al., 1990; St. Louis et al., 1991). Louko et al. (1990) conducted direct assessment of preschool-aged children who stutter (mean age 4;6 years, range 2;5–6;11 years), matched with fluent controls, and found that 40 % of the children who stuttered exhibited disordered phonology compared to 7% of the matched controls. Disordered phonology was defined by the presence of at least one atypical error and/or at least one age inappropriate error. St Louis, Murray, and Ashworth (1991) observed school-aged children (mean age 12;6 years, range 6;8–17;5 years) who stuttered and reported that 42 % of participants had a co-occurring

phonological disorder. Phonological disorder was determined to be present if a child exhibited > 1 speech error. Both of these studies reported similar rates of comorbid stuttering and SSDs despite using different assessment protocols. Louko et al. (1990) analysed data from spontaneous speech samples while St Louis et al. (1991) administered the Goldman Fristoe Test of Articulation (Goldman & Fristoe, 1968), a single-word naming test.

Varying results between previous studies may reflect methodological differences. These include (but are not limited to): the absence of matched controls; use of surveys and informal observations to document speech development; differing definitions of stuttering and SSD; small sample sizes; and approaches to elicit speech production (i.e. using word level assessments vs. spontaneous connected speech samples). Further, there may be reason to question the results of the findings of some of the direct observation studies because only a portion of children went on to receive referrals for therapy (Nippold, 2001). Future research examining co-occurrence should therefore include control groups of non-stuttering children to obtain more rigorous estimates of the co-occurrence of stuttering and SSD. In doing so, studies may reduce potential risk for bias and allow for optimal comparison and interpretation, as participants are recruited from the same cohort with similar backgrounds (e.g., socio-economic status (SES) etc.) and the same methods for diagnosis can be applied across the whole cohort. Further, there has been much research focusing on variables associated with stuttering and SSD in isolation, yet little is known about these variables when the disorders co-occur. For example, variables such as gender, twinning status and language status are reportedly associated with stuttering. There are a higher proportion of males who stutter compared to females as reported in Bloodstein and Ratner (2008). Twin status and more advanced vocabulary development have also been reported as being positively associated with stuttering onset (Reilly et al., 2009). Being a boy and having a reactive temperament have been reported as risk factors for SSD (Harrison & Mcleod, 2010), as have maternal level of education and socioeconomic factors such as household income (To, Cheung, & Mcleod, 2013). These data help generate hypotheses regarding which variables are associated with and potentially predictive for children with dual diagnoses. Identifying predictors of dual diagnosis cases may function as potential preventative measures and may support earlier identification and intervention for these children.

In summary, stuttering and SSD have been reported to co-occur at rates as high as 46 %; however these findings are largely based on selected clinical samples, an approach more likely to introduce study bias. Co-occurrence may be a key factor driving referral and influence parental help-seeking behaviour. The use of prospective, longitudinal community cohort data to examine the co-occurrence rate of stuttering and SSD is needed. Screening children using this methodology will enable more precise estimations of population prevalence and resolve some of the variance previously reported in the literature. Further, participants must be compared to controls to make findings generalizable to the broader population.

Here we use data from a prospective community cohort. Our study aims to:

1 Identify the proportion of children diagnosed with co-occurring stuttering and SSDs at age 4 years; and
2 Describe and compare demographic and clinical features of this comorbid diagnosis group compared to children with no diagnosis of either disorder, or those with either disorder in isolation. These features are known to be associated with stuttering or SSD in isolation and include examples such as: language development, family history, maternal education, SES.

2. Method

Ethical approval for this research was obtained from the Royal Children's Hospital Human Research Ethics Committee (27078/33195) and the La Trobe University Human Ethics Committee (03–32).

2.1. Participant recruitment and procedures

Participant recruitment procedures relevant to this study are described below in three stages (see Fig. 1).

The Early Language in Victoria Study (ELVS) baseline community sample was recruited when children were aged 8–10 months (wave 1) from six socio-economically diverse local government areas in Melbourne, Australia between September 2003 and April 2004 (see Reilly et al., 2006, Reilly et al., 2017 for more detail). Maternal and Child Health (MCH) nurses approached parents of all infants attending an 8-month 'universal well-child' appointment to participate in the study. These appointments have an uptake of 82 % (i.e., 82 % of families attend the free consultations to monitor their child's health, growth, and development). In addition, participants were also recruited at 8-month hearing screening sessions and through advertisements in local newspapers. As indicated in Fig. 1, of the 2335 families approached for the study, 1910 families were eligible and consented to participate and formed the baseline sample. All consenting families completed a parent-report questionnaire when participants were 8–10 months old and then on an annual basis from 1 to 4 years of age. Questionnaires contained measures related to children's communication development, and a range of other child and family-related variables (Reilly et al., 2006). Direct assessments were completed on a subsample at 12 months and all participating children 4 years of age.

At age 2 years, parents who completed and returned the annual questionnaire were invited to participate in the 'nested' ELVS Stuttering Study (Reilly et al., 2009, 2013). Parents were sent an opt-out letter and provided with a refrigerator magnet that described stuttering behaviours and encouraged to contact the research team by telephone if they observed their child exhibiting these stuttering behaviours. Reminder letters were sent out to participating families every 4 months for a year.

At age 4 years, participants were invited to attend an individualised face-to-face assessment for SSD, language delay and pre-literacy ability (per procedures cited by Eadie et al., 2015); 1623 parents returned their completed questionnaires and 1607 children underwent direct face-to-face assessment.

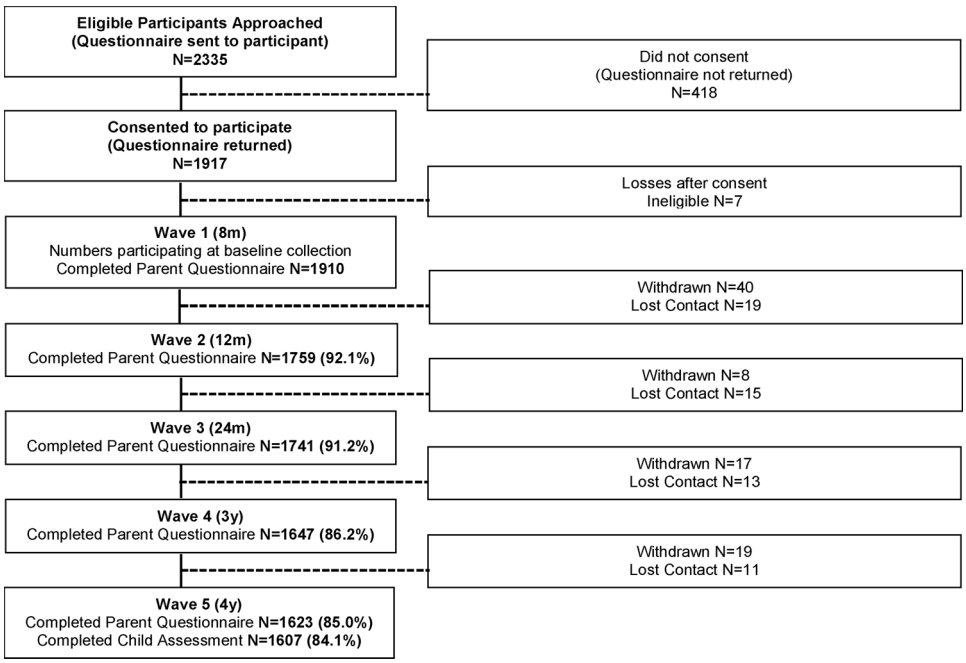

Fig. 1. Participant flowchart from baseline (8 months) to 4 years (denominator for percentages is number participating at baseline – 1910).

2.2. Measures

2.2.1. Ascertainment of stuttering and SSD

Members of the research team contacted any parent who reported that their child was exhibiting any of the stuttering behaviours described on the fridge magnet, or if they indicated in any of the questionnaires that their child had a stutter between the ages of 2 and 4 years. The research assistant first confirmed that the descriptions were consistent with characteristics of stuttering and if so, a 45-minute home visit was scheduled by an experienced SLT. Where possible, these visits were conducted within a two-week period of the parents' initial report. A thorough case history was obtained, followed by a 25-minute videotaped play session between parent and child using a standard set of stimuli (e.g., toys, pictures and prompts for conversation). At the conclusion of the play session, the research assistant and parent jointly confirmed the presence of stuttering using a 10-point severity rating scale (where 1 = no stuttering, 2= extremely mild stuttering and 10 = extremely severe stuttering). Stuttering was confirmed when both the parent and research assistant (RA) assigned a severity score ≥ 2 on the scale. Ambiguous cases were reviewed on a case-by-case basis by an expert panel who independently reviewed the samples to ascertain if the child was stuttering. This procedure was used to identify children who started to stutter up to 4 years of age (Reilly et al., 2013).

The Goldman Fristoe Test of Articulation (GFTA-2, Sounds-in-Words subtest) (Goldman & Fristoe, 2000) was administered to all participating children. Procedures for administration and scoring of this assessment were completed in line with the GFTA-2 manual. Research assistants (with a background in speech-language therapy or psychology) administered this assessment (Sounds-in-Words subtest) and were formally trained in the protocolised procedures by an experienced SLT to do so. Only the trained SLTs scored the assessments. Children's responses to each stimulus were recorded. Speech sound disorder was defined on the basis of standardized scores that were ≤79 on the GFTA -2 and this also encompassed scores that were in the bottom 10 % as compared to the entire ELVS cohort[1] (Eadie et al., 2014). Similar to previous research examining comorbid stuttering and SSD, all children in this study who received a diagnosis of speech sound disorder had > 1 speech error (St Louis et al., 1991), as analysed by the GFTA-2.

Video and audio recordings of these direct assessments were transcribed using narrow phonetic transcription conventions. Reliability of the data was evaluated by two experienced SLPs (per procedures cited by Dodd et al., 2017) who were presented with 10 % of randomly selected assessment data. Each SLP therefore transcribed these chosen 10 % of data twice and obtained a point-to-point reliability of 96.8 %.

In this study, a history of stuttering was defined as years any child verified to have stuttering onset between 2 and 4 years. Comorbidity was determined by identifying groups of children at 4 years of age as follows: children with a history of stuttering (n = 149); children with SSD only (n = 129; and children with a history of stuttering and SSD (n = 11). Children without a history of stuttering between 2 and 4 years and with no SSD at 4 years were included as control or comparison group (n = 1318).

[1] For a more complete summary on the rationale and procedures of the use of this tool to diagnose SSD with the ELVS cohort, readers are advised to refer to Eadie et al., 2014 and Eadie et al., 2015.

2.2.2. Demographic features

Child variables were collected within the parent-report questionnaires completed from baseline to 4 years of age or direct assessments completed at 12-months or 4 years of age. Factors such as child demographic details, birth history, feeding problems, language/s spoken at home and family history of communication difficulties were provided in the questionnaires.

Family and environmental factors included socio-economic status, which was indicated by scores derived from the continuous Socio-Economic Indexes for Areas (SEIFA) Index of Relative Disadvantage (by postcode) at 8-months of age. On this index, lower scores based on an Australian normative mean of 1000 (SD = 100), were indicative of more disadvantage (Australian Bureau Of Statistics, 2001). At 1 year of age, maternal mental health and vocabulary scores were measured by the Kessler screen for Psychological Stress (Kessler & Mroczek, 1994) and the Mill Hill vocabulary scale respectively (Raven, 1997).

2.2.3. Speech, language and communication features

Parents also completed scales/checklists of features specifically related to speech and/or language. At ages 2- and 3-years, parents completed the MacArthur Bates Communicative Development Inventory (CDI) (Words and Sentences version) which was modified (with author permission) to include some Australian specific item substitutions. In total, 24 vocabulary items were substituted on the Words and Sentences inventory, and it is unlikely that these changes impacted the validity of the results. Children were classified as late talkers if they scored below the 10th percentile based on the norms provided by this assessment (Fenson, Dale, & Reznick, 1993). At age 3 years, completion of the Communication and Symbolic Behavior Scales Infant-Toddler checklist (CSBS) allowed collection of standardized scores (Wetherby & Prizant, 2002). Parents also completed the Short Temperament Scale for Children (Prior, Sanson, & Oberklaid, 1989) at age 3 years which includes 6 dimensions of temperament including approach, distractibility, rhythmicity, co-operation, persistence and reactivity. At age 4 years, children were directly assessed for language ability using the Clinical Evaluation of Language Fundamentals Preschool (2nd Edition, Australia and New Zealand Standardized Edition; CELF-P2) (Wiig, Secord, & Semel, 2006). They also underwent a test of pre-literacy ability using a purpose-designed letter knowledge task (for more detail, see Eadie et al., 2015). Nonverbal cognitive skills were directly assessed using the matrices subtest of the Kaufman Brief Intelligence Test (KBIT; 2nd edition) (Kaufman & Kaufman, 1990).

The abovementioned child, family and environmental variables were used to examine associations with a dual diagnosis of stuttering and SSD. Table 1 provides a summary of all of variables and the time-points from which they were gathered.

2.3. Statistical analysis

Chi-square tests compared groups on categorical socio-demographic variables. T-tests were used to compare groups on continuous variables examining family and maternal characteristics, child temperament, non-verbal IQ and language performance. Effect sizes were also calculated for continuous variables using Cohen's d (interpreted as small (d = 0.2), moderate (d = 0.5), and large (d = 0.8) (Cohen, 1988)), and for categorical variables using Cramer's V (φ) (interpreted as weak (φ ≤ .2), moderate (φ.21–.3), and strong (φ ≥ .3)). Due to the small group size of the Stutter + SSD group (n = 11), further analysis of potential predictor variables could not be carried out. Instead we present some of the first statistics describing those with comorbid stuttering and SSD.

Table 1
Summary of clinical and demographic variables and corresponding time-points of collection.

	8 months	12 months	2 years	3 years	4 years
Family/environmental and maternal measures					
Gender	*				
Birth type (single/twin)	*				
Prematurity	*				
Late talker			*		
Feeding problems	*				
Family language	*				
Family history of speech and/or language problems	*				
Maternal education	*				
SEIFA index of disad.	*				
Kessler total score		*			
Maternal Mill Hill score		*			
Child measures					
CDI			*	*	
Temperament				*	
CSBS				*	
CELF					*
KBIT					*
Goldman Fristoe					*
Letter awareness					*

Table 2
Chi-square analysis comparing the Stutter + SSD group to all other groups on categorical variables, with n (%) indicated.

		Stutter + SSD versus Control group				versus Stutter only			versus SSD only		
		Stutter + SSD	Controls	φ	p	Stutter only	φ	p	SSD only	φ	p
Infant											
Gender											
	Male	7 (63.64)	661 (50.15)	.02	.373	87 (58.39)	.03	.733	71 (55.04)	−.05	.582
	Female	4 (36.36)	657 (49.85)			62 (41.61)			58 (44.96)		
Birth type											
	Single	11 (100.00)	1287 (97.65)	.01	.607	138 (92.62)	.07	.350	126 (97.67)	−.04	.609
	Twin	0 (0.00)	31 (2.35)			11 (7.38)			3 (2.33)		
Premature											
	No	10 (90.91)	1274 (96.66)	−.03	.293	146 (97.99)	−.11	.147	124 (96.12)	.07	.412
	Yes	1 (9.09)	44 (3.34)			3 (2.01)			5 (3.88)		
Late talker at 2yrs											
	Yes	4 (36.36)	232 (18.74)	.04	.137	13 (8.84)	.23	.004	51 (43.22)	.04	.660
	No	7 (63.64)	1006 (81.26)			134 (91.16)			67 (56.78)		
Feed problems (8 m)											
	Smooth food, yes	0 (0.00)	24 (1.82)	.01	.652	2 (1.34)	.03	.699	2 (1.55)	−.04	.677
	Smooth food, no	11 (100.00)	1294 (98.18)			147 (98.66)			127 (98.45)		
	Mashed food, yes	1 (9.09)	134 (10.17)	.00	.906	13 (8.72)	−.00	.967	10 (7.75)	.01	.874
	Mashed food, no	10 (90.91)	1184 (89.83)			136 (91.28			119 (92.25)		
	Lumpy food, yes	3 (27.27)	549 (41.65)	.03	.335	58 (38.93)	.06	.443	61 (47.29)	−.11	.201
	Lumpy food, no	8 (72.73)	769 (58.35)			91 (61.07)			68 (52.71)		
Family											
Language											
	English	11 (100.00)	1246 (94.61)	.02	.731	148 (99.33)	.02	.785	126 (97.67)	−.04	.609
	English + Other	0 (0.00)	13 (0.99)			0 (0.0)			0 (0.0)		
	Other	0 (0.00)	58 (4.40)			1 (0.67)			3 (2.33)		
Family history											
	8 m										
	Yes	6 (54.55)	317 (24.05)	−.06	.019	32 (21.48)	−.20	.013	47 (36.43)	.10	.235
	No	5 (45.45)	1001 (75.95)			117 (78.52)			82 (63.57)		
	6 years										
	Yes	5 (62.50)	223 (27.50)	.08	.028	46 (41.07)	.11	.236	32 (52.46)	−.06	.592
	No	3 (37.50)	588 (72.50)			66 (58.93)			29 (47.54)		
Maternal education											
	Year 12 or greater	11 (100.0)	1020 (78.34)	.05	.082	121 (82.31)	.05	.127	93 (73.23)	−.17	.048
	< Year 12	0 (0.00)	282 (21.66)			26 (17.69)			34 (26.77)		

Note: φ = Cramer's V; Participants included in each group for each measure ranged from n = 811–1318 for the control group, n = 8–11 for the 'SSD + Stutter' group, n = 112–149 for the 'Stutter only' group and n = 61–129 for the 'SSD only' group.

3. Results

3.1. Rate of comorbidity: Stuttering and SSD outcomes by 4 years

Of the 1607 children who underwent assessment at 4 years of age, 11 obtained a dual diagnosis of a stutter and SSD, equating to 0.68 % of the population assessed. This indicates that around 1 in every 146 four year old children will meet criteria for dual diagnosis. Overall, 6.88 % (11/160) of those with a stutter also had a SSD. The following mean scores were obtained from the GFTA-2: Stutter + SSD group ($M = 82.18$, $SD = 5.76$); SSD only ($M = 81.76$, $SD = 8.24$); Stutter only ($M = 107.91$, $SD = 7.85$).

3.2. Clinical and demographic features

Table 2 summarises the characteristics of participants assigned to the Stutter + SSD group relative to the three subgroups: Stutter only, SSD only and control group. Chi-square analysis indicated the Stutter + SSD group had a higher percentage of participants (54.55 %) who reported a positive family history of communication difficulties at 8 months of age compared to the Control group (24.05 %, $p = 0.019$) and to the Stutter only group (21.48 %, $p = 0.013$). The Stutter + SSD group also had a higher percentage of participants (36.36 %) classified as late talkers at age 2 years compared to the Stutter only group (8.84 %, $p = 0.004$). Finally, a larger percentage of participants in the Stutter + SSD group (100 %) had mothers who had completed a tertiary qualification and/or Year 12[2] compared to the SSD *only* group (73.23 %, $p = 0.048$).

Table 3 summarises the *t*-test results comparing the Stutter + SSD group against all three subgroups on continuous variables

[2] A tertiary qualification is also known as third stage/third level education. In Australia, it refers to education that a student undertakes following their compulsory secondary schooling. In Australia, Year 12 is considered the final year (end-point) of a student's secondary education.

Table 3
T-test results comparing the stutter + SSD group to all other groups on continuous variables with Mean (SD) indicated.

	Stutter + SSD versus Control group						versus Stutter only				versus SSD only			
	n	Stutter + SSD	n	Controls	*d*	p	n	Stutter	*d*	p	n	SSD	*d*	p
Family and maternal characteristics														
SEIFA index of disadvantage (8 m)	11	1036.31 (46.04)	1318	1037.93 (59.81)	−.03	.929	149	1050.14 (45.07)	−.31	.328	129	1031.45 (59.16)	−.08	.791
Kessler total score (12 m)	11	4.18 (3.28)	1249	2.98 (2.91)	.63	.037	148	2.88 (2.76)	.69	.028	121	3.27 (3.06)	−.50	.113
Maternal Mill Hill score (12 m)	11	28.18 (3.82)	1254	27.58 (4.92)	.12	.687	148	29.41 (3.88)	−.32	.311	121	27.28 (5.56)	−.17	.600
Child at 2 years														
CDI														
Vocabulary production %rank	11	28.00 (28.31)	1238	42.19 (28.30)	−.50	.099	147	47.26 (27.83)	−.69	.029	118	23.22 (23.38)	−.20	.525
Irregular nouns/verbs % rank	11	29.73 (26.19)	1260	45.12 (26.16)	−.59	.052	147	47.11 (25.56)	−.68	.031	122	31.40 (25.30)	.07	.834
Sentence complexity %rank	11	61.09 (35.87)	1260	54.12 (30.45)	.23	.450	147	55.51 (28.80)	.19	.543	122	58.16 (34.66)	−.08	.789
Child at 3 years														
Temperament														
Approach score	11	17.66 (4.55)	1212	16.58 (4.40)	.24	.421	141	16.88 (4.03)	.19	.545	113	16.67 (4.41)	−.22	.480
Reactivity score	11	29.00 (3.52)	1213	28.38 (5.11)	.12	.687	141	26.81 (4.71)	.47	.133	113	28.48 (5.04)	−.10	.741
CSBS														
CSBS Standard score	11	102.18 (5.28)	1226	104.63 (14.58)	−.17	.580	143	107.70 (15.26)	−.36	.255	117	98.65 (14.50)	−.24	.449
CDI														
Vocabulary production %rank	11	23.09 (24.94)	1208	28.45 (26.22)	−.20	.499	141	31.33 (24.74)	−.33	.289	115	17.50 (20.74)	−.27	.403
Grammatical complexity %rank	11	24.64 (38.02)	1208	35.77 (35.92)	−.31	.307	141	44.66 (37.71)	−.53	.092	115	18.79 (24.32)	−.23	.472
Language use %rank	11	29.73 (21.90)	1208	33.27 (29.12)	−.12	.687	141	39.50 (28.49)	−.35	.269	115	21.77 (24.16)	−.33	.295
Child at 4 years														
CELF														
Core language	11	102.55 (15.18)	1311	99.31 (14.63)	.22	.466	149	105.42 (12.55)	−.23	.471	128	94.63 (15.41)	−.51	.104
Receptive language	11	96.00 (13.73)	1307	96.32 (14.88)	−.02	.942	149	103.62 (13.29)	−.57	.069	128	92.24 (14.87)	−.25	.420
Expressive language	11	100.82 (18.47)	1275	99.68 (14.97)	.08	.803	149	104.69 (13.63)	−.28	.377	126	92.85 (15.84)	−.50	.117
Other														
KBIT matrices SS	11	104.00 (9.17)	1307	104.18 (13.50)	−.01	.966	149	106.72 (11.56)	−.24	.447	128	101.07 (13.88)	−.22	.494
Goldman Fristoe SS	11	82.18 (5.76)	1290	108.06 (8.77)	−2.96	< .001	149	107.91 (7.85)	−3.33	< .001	129	81.76 (8.24)	−.05	.871
Letter awareness raw score	10	9.10 (9.36)	1263	8.34 (8.96)	.08	.791	144	9.28 (9.02)	−.02	.952	122	6.87 (8.71)	−.25	.440

Note: d = Cohen's d, SS = standardised score.

measured. At age 12 months, mothers of participants in the Stutter + SSD group had a tendency to report greater mental distress compared to the Control and Stutter only groups ($p = 0.037$ and $p = 0.028$). Vocabulary production percentile rank at age 2 years was lower in the Stutter + SSD group compared to the Stutter only group ($p = 0.029$). Similarly, irregular noun/verb percentile rank at age 2 years was lower in the Stutter + SSD group compared to the Control group ($p = 0.052$) and the Stutter only group ($p = 0.031$). Children in the Stutter + SSD group scored significantly lower in the GFTA-2 compared to the Control group ($p < .001$ $d = 2.96$; mean difference 25.88) and the Stutter only group ($p < .001$, $d = 3.33$; mean difference 25.73). Both comparisons were also supported by very large effect sizes, which is also indicative of what would be expected clinically for these groups as well.

There was no evidence of any other significant differences between the Stutter + SSD group and other subgroups; however there were almost twice as many boys as there were girls in the Stutter + SSD group (63.64 % vs. 36.36 %). There were also no differences observed between groups in relation to SES status and maternal vocabulary. In relation to child temperament, no differences were found between groups. Across the various language measures recorded at 3 and 4 years, there were no differences observed between groups. The SSD only group achieved a lower mean score (M = 6.87, SD = 8.71) on pre-literacy ability, though this was not significantly different across all groups.

4. Discussion

This study identified the proportion of children diagnosed with comorbid stuttering and SSD in a prospective, longitudinal community cohort of children growing up in Melbourne, Australia. Of the 1607 children assessed at age 4 years, 11 children received a dual diagnosis indicating a community cohort prevalence of 0.68 %. In total, 6.88 % (11/160) children who stuttered also had a SSD. This clinical prevalence rate is significantly lower than prevalence rates reported in previous studies (16–46 %). Methodological differences between the current study and previous studies may account for these conflicting findings. Previous research in this area has mostly reported on clinical prevalence (i.e., children who presented to clinics with a diagnosis of stuttering who were subsequently diagnosed with a SSD as well). Of the 160 children who stuttered, 11 had a diagnosis of SSD (6.88 %). Again, this finding is much lower than any figure previously reported, likely due to the fact that findings were based on clinical cohorts. Comorbid diagnoses may be a key factor driving referral and attendance at clinics, and influence parental help-seeking behaviour. Additionally, some of the previously reported results were based on data collected from surveys and therefore may be based on uncontrolled and subjective participant evaluation/diagnosis (Arndt & Healey, 2001; Blood & Seider, 1981; Blood et al., 2003). In contrast, the present study used community-cohort controls to enable comparisons and all participants were rigorously diagnosed with stuttering and SSD using the same protocol and criteria.

Other studies based on direct observation have also reported higher prevalence rates than the present study, that is, rates between 40–42 % (Louko et al., 1990; St Louis et al., 1991). One methodological difference separating these two studies from the current is the definition used to determine the presence of SSD. The current study determined a speech sound disorder if a child scored ≤ 79 on the GFTA -2 (Goldman & Fristoe, 2000), which encompassed scores in the bottom 10 %, whereas both St Louis and Louko used lower cut-off points. Another methodological difference lies in the sampling context. Similar to St Louis, the current study also based the diagnosis of SSD on standardized scores obtained from the GFTA, although the current study reports a much lower prevalence rate. It is suspected that only using a single word naming test context may have limited the prevalence findings in the current study, and that further assessment for SSD in spontaneous connected speech would yield a higher rate of clinical prevalence as demonstrated in Wren, Mcleod, White, Miller, and Roulstone, (2013). For SSD, it is optimal to confirm diagnosis based on a combination of a single-word naming test, which samples relevant sounds systematically, and an appropriate connected speech sample. The single-word naming test should include all consonants and clusters in appropriate word positions for a given language (Morrison & Shriberg, 1992). Future research may consider incorporating both spontaneous connected speech and single word naming test contexts to verify the presence of SSD. The tool used to assess SSD was considered valid and reliable for the purposes of the ELVS; however, it is acknowledged that the norms used in this assessment are not Australian. This has been countered by statistical modification as discussed in previous research (Eadie et al., 2015). When using a spontaneous speech sample alone, children are free to talk about whatever they choose, and may therefore fail to produce certain consonants and consonant clusters (Nippold, 2002).

A further aim of the current study was to describe and compare theoretically driven variables associated with either stuttering or SSD in isolation in relation to the Stutter + SSD group. Whilst statistically significant findings between groups were revealed, we cannot rule out that these occurred due to chance. That is, due to the large number of comparisons conducted here, it is anticipated that around 5% of findings would have p-values below 0.05 even if there were no true differences between groups (as they are likely occurring due to chance). Hence there is insufficient evidence to suggest real differences between groups in the present study, although any group differences observed will be summarised below

A higher percentage of participants in the Stutter + SSD group reported a positive family history of communication difficulties compared to the control and stutter only groups. This is consistent with previous reports which have reported family history of communication difficulties as a predictor of both stuttering and SSD in isolation (Campbell et al., 2003; Bloodstein & Ratner, 2008). Similarly, previous studies have indicated that being a boy is associated with a heightened risk for both disorders (Harrison & Mcleod, 2010; Reilly et al., 2009). A larger percentage of mothers of participants in the Stutter + SSD group had completed a higher level of education compared to the SSD only group. These findings are consistent with previous research reporting that maternal higher education level may predict stuttering onset (Reilly et al., 2009) and may also be a risk factor associated with SSDs (To et al., 2013; Campbell et al., 2003).

In relation to language variables, the results of the current study revealed that at age 2 years, a larger percentage of participants in the Stutter + SSD group were classified as late talkers. Other language measures indicated that this group had an overall lower

overall vocabulary production score compared to participants in the Stutter only group, and scored lower on their use of irregular nouns/verbs than both the Stutter only group and the Control group at 2 years of age. This is an interesting finding since previous literature has reported that stuttering onset often coincides with rapid language development and that having a more advanced vocabulary may predict stuttering onset by age 3 years (Reilly et al., 2009). While Reilly et al. (2013) reported that advanced vocabulary was no longer a predictor of stuttering onset by age 4 years, other researchers have reported that children who stutter have similar or more advanced language skills compared to non-stuttering controls (Nippold, 2018). The current study found no significant differences in language measures between groups at 3- and 4-years of age. In contrast, there is a reported correlation between language ability and SSDs. Children whose language is less advanced may have weaker phonological skills and display more speech sound errors than their typically developing peers (Roulstone et al., 2002; Smith et al., 2006).

In relation to pre-literacy skills, there is much literature reporting a strong positive correlation between literacy skills and SSDs. Children with SSDs are at significant risk for delays in development of phonological awareness skills and, subsequently, literacy acquisition (Anthony et al., 2011; Rvachew & Grawburg, 2006). The results of this study indicated no evidence of significant differences between the groups' pre-literacy skills. However, the SSD-only group did achieve a lower overall mean score compared to all other groups.

The association between maternal mental health and communication disorders has generated inconsistent findings in the literature. Some studies have reported no association between maternal mental health and speech/language disorders (Reilly et al., 2007; Reilly et al., 2006), while others have found a positive correlation (Prior et al., 2008; Weindrich et al., 2000). Harrison and Mcleod (2010) reported that better maternal mental health was a protective factor against speech and language disorders in 4- to 5-year-old children. In relation to stuttering, maternal mental health has not been found to be associated with stuttering onset at 3- or 4-years of age (Reilly et al., 2009, 2013). In the current study, the mothers of participants in the Stutter + SSD group reported poorer maternal mental health compared to the Control group when participants were aged 12-months.

Across all subgroups, there was no evidence of significant differences on measures of SES status, maternal vocabulary or child temperament. This is consistent with previous research that has indicated SES is not associated with stuttering onset by age 4-years (Reilly et al., 2009, 2013). In contrast, the association between SES and SSDs is less clear, with some research noting no relationship between SES and speech acquisition (Dodd, Holm, Hua, & Crosbie, 2003) and others reporting that low SES may be a risk factor for development of SSD (To et al., 2013). Particular temperament traits in early childhood are a widely accepted risk factor for the development of anxiety later in life (Fox & Pine, 2012). A previous study based on the same cohort used in the present study reported that temperament precursors of anxiety are not indicated in children prior to or shortly after stuttering onset (Kefalianos, Onslow, Ukoumunne, Block, & Reilly, 2014). Instead, temperament differences may emerge as children get older and experience the negative consequences of stuttering over time. Other studies examining the association between temperament precursors of anxiety and stuttering have yielded similar results finding no differences between children who stutter and their non-stuttering peers (Eggers et al., 2010; Anderson et al., 2003). In relation to SSDs, our findings conflict with previous research. Hauner, Shriberg, Kwiatkowski, and Allen, (2005) reported that the temperament precursors of anxiety (including approach/withdrawal dimension) were associated with greater severity of SSDs. Similarly, Harrison and Mcleod (2010) reported that a reactive temperament was a risk factor for speech and language impairment, while a more persistent and sociable temperament was considered a protective factor. The findings in relation to temperament from this study may be explained by the fact that data were collected from a community cohort (where severity of stuttering and/or SSDs may be milder than those presenting clinically).

Finally, and unsurprisingly, children in the Stutter + SSD group had overall lower scores on speech sound production as assessed by the GFTA-2 compared to the Control group and to those in the Stutter only group, further confirming the diagnosis of SSD in this group of children. What is interesting to note is that the children in the Stutter only group scored very similarly on this measure compared to those in the Control group (mean difference 0.15). This finding perhaps further supports the viewpoint that there may be limited interaction between stuttering and phonology (Nippold, 2002).

While the abovementioned findings are interesting to note, they cannot be used to reliably predict the onset of comorbid stuttering and SSDs. The small sample size ($n = 11$) reported in this study prohibited the use of more rigorous statistical analysis (e.g., logistic regression) to infer predictability. The large number of comparisons made in the current study make it difficult to interpret the findings as statistically significant, as there is a possibility the differences noted occurred due to chance. Additionally, as previously mentioned, it is more ideal to assess the production of children's speech in both spontaneous connected speech and single-word naming test contexts. Whilst all assessments (including speech assessment) used in the current study were considered valid and reliable, it is acknowledged that they were not exhaustive and may only have provided a snap shot of the participants' communicative abilities. More thorough analysis of speech and language abilities may have yielded different results. Finally, it is worthwhile noting that the results found in this study were based on children confirmed as stuttering between 2 and 4 years of age, and confirmed with a SSD at 4 years of age. It is therefore a consideration that during this period, some children may have naturally recovered from stuttering. Similarly, stuttering onset may continue after 4 years of age for some children (Reilly et al., 2013). Future studies should consider all of the abovementioned factors and potentially conduct later follow-up studies taking these into account.

5. Conclusion

This is the first study to report the prevalence of comorbid stuttering and SSD in a cohort of children recruited prospectively from the community. We found that 6.88 % of children diagnosed with stuttering also had a diagnosis of SSD. Further, only 0.68 % of participants received this comorbid diagnosis. Both of these findings are much lower than those previously reported and likely reflect methodological differences between studies. Further, general population prevalence has not been previously reported. Therefore,

consideration of methodological differences must be taken into account when interpreting the surrounding literature in this area. Further, it is likely that more children are presenting to clinic because of the dual diagnoses and parents seeking help more so than if the disorders were presenting in isolation. These children may also be presenting with more severe forms of both stuttering and/or SSDs than what is represented in a broad community cohort sample.

Our secondary aim was to examine the association between theoretically driven variables, previously associated with either stuttering or SSD in isolation, and a dual diagnosis of these conditions. At 4-years of age, there was evidence of differences for a range of child and family variables between children with a comorbid diagnosis of stuttering and SSD and other subgroups. Such differences related to family history of communication difficulties, late talking status, speech sound production, and maternal education levels. Other group differences were reported on some variables, including: maternal mental health, vocabulary production, and use of irregular nouns/verbs. However, as previously reported, these results should be interpreted with caution due to the large number of comparisons made in this study. In order to determine whether these differences are real, this study would need to be replicated on a larger sample.

As a research exercise, future studies should use more rigorous assessment in relation to diagnosis of SSDs by including analysis of both spontaneous connected speech samples and single word naming tests, as a first step to inform practice. This may impact on the number of children diagnosed with SSDs, and therefore the prevalence of comorbid stuttering and SSDs. Having a valid indication of the prevalence of co-occurring stuttering and SSDs is an important step that paves the way for a better understanding of this caseload, which in turn may lead to specific research for the management of this population. Similarly, identifying predictors of dual diagnoses may function as potential preventative measures and may support earlier identification and intervention for these children.

Overall, more research in the area of co-occurring stuttering and SSDs is required in order to further understand this caseload and subsequently, to efficiently and effectively manage this population.

CRediT authorship contribution statement

Rachael Unicomb: Conceptualization, Resources, Writing - original draft, Writing - review & editing, Visualization, Supervision. **Elaina Kefalianos:** Conceptualization, Resources, Writing - review & editing. **Sheena Reilly:** Methodology, Validation, Investigation, Resources, Writing - review & editing, Project administration, Funding acquisition. **Fallon Cook:** Software, Visualization, Formal analysis, Writing - review & editing. **Angela Morgan:** Conceptualization, Methodology, Validation, Formal analysis, Investigation, Resources, Writing - review & editing, Supervision, Project administration, Funding acquisition.

Declaration of Competing Interest

The authors of this paper report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

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Appendix A. Supplementary data

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