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Authors	Lee, Alice S.;Hayes, Orla;Ní Mhurchú, Damhnait;Gibbon, Fiona E.
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A preliminary study of prosody skills in children with spina bifida

Alice Lee[1], Orla Hayes[1], Damhnait Ní Mhurchú[2], and Fiona E. Gibbon[1]
[1]University College Cork
[2]Central Remedial Clinic, Waterford

Introduction

Spina bifida (SB) is a common congenital structural defect of the central nervous system that affects approximately 3–5 per 10,000 births depending on the population (Fletcher & Brei, 2010). The defect occurs when the caudal end of the neural tube – the precursor to the brain and spinal cord – fails to close properly during the third and fourth week of gestation (Schoenwolf, Bleyl, Brauer, & Francis-West, 2015), resulting in abnormalities in the central nervous system and its supportive tissues including the spine, muscle, and connective tissue (Murdoch, 2011). SB can be further classified into different types depending on the extent of the structural anomalies (see e.g. Copp et al., 2015). Myelomeningocele is the most common and severe form of SB, which is characterised by an open lesion in the lumbar and lumbosacral regions of the back, with the meninges and spinal cord protruding through the defect and adhering to the inner surface of a swelling meningeal cyst (Murdoch, 2011). In addition, myelomeningocele is associated with malformation of the brain (particularly, the cerebellum, midbrain, corpus callosum, and the posterior cerebral cortex) and hydrocephalus (see e.g. Fletcher, Barnes, & Dennis, 2002; Murdoch, 2011). Hence, aside from the sensory and motor impairments of the parts of body below the lesion as well as bowel and bladder dysfunction, individuals with SB myelomeningocele often show difficulties in the cognitive and language domains (Fletcher et al., 2002; Murdoch, 2011).

There has been an extensive body of work on language and related skills in children with SB (e.g. Dennis & Barnes, 2010; Dennis, Hendrick, Hoffman, & Humphreys, 1987; Fletcher et al., 2002; Huber-Okrainec, Dennis, Brettschneider, & Spiegler, 2002). Recent reviews show that most of the children with SB have difficulties with specific aspects of language, but show relative strengths in other areas of language (Dennis & Barnes, 2010; Fletcher et al., 2002; Murdoch, 2011). In general, language abilities on the surface appear strong in most of the children with SB in terms of single word vocabulary, morphology, syntax and phonology (Brookshire et al., 1995; Dennis & Barnes, 2010; Fletcher et al., 2002; Tew, 1979). Discourse can appear as a strength in that children often engage readily in conversations. However, on closer examination, children with SB experience notable difficulties with pragmatic aspects of communication, that is, the appropriate use of language in conversations and social interaction (Schwartz, 1974). Difficulties in conversations include problems shifting attention and topic, deriving and expressing meaning in conversations and integrating information from social and written context. In social situations, children are described as being overly "chatty" with conversational partners (Fletcher et al., 2002; Murdoch, 2011; Tew, 1979). Hence, although children produce a lot of language, it often lacks content, cohesion and it can be inappropriate in terms of interaction style. Much is known about the language profile of children with SB; however, prosody has not been investigated systematically. This is a surprising gap because receptive or expressive difficulties with prosody could account for at least some of the difficulties – pragmatic, interactional, semantic – experienced by children with SB.

Prosody is an umbrella term to cover aspects of speech that centres on how variations in loudness, relative syllable-length and vocal pitch combine to enhance or change the meaning of what is said (e.g. Cruttenden, 1997; Peppé, 2009b). Prosody is central to language and conveys meaning at lexical, grammatical and pragmatic levels (Peppé, 2009a). At the lexical level, prosody can indicate word-class (e.g. "INsult" the noun versus "inSULT" the verb; the syllable in capitals carries the main stress). Prosody also plays a grammatical role, and can be thought of as operating as the verbal equivalent of written punctuation; that is, to indicate delimitation of utterances including major and minor syntactic divisions. For example, "FRUIT-salad and MILK" indicates two food items, whereas "FRUIT, SAlad and MILK" indicates three food items. Prosody also operates in various interactional categorisations, and so has an important role to play in pragmatics. For example, prosody indicates whether a conversational turn has ended and distinguishes between utterance types (e.g. question, statement, or request). Prosody also shows where new information (emphasis or focus) is in an utterance, for example, in sentential stress or accent (e.g. "It's a blue CAR" versus "It's a BLUE car"; Peppé & McCann, 2003). As far as the communication of affect/mood is concerned, prosody ('tone of voice') conveys the personal attitude and emotion of the speaker (e.g. emotional/neutral, humorous/serious).

Despite the important role of prosody in language and communication, there have been no published research studies on prosody in children with SB. A few previous studies investigating neuromotor speech or language in children with SB have made reference to abnormal expressive prosody, however. Tew (1979) mentioned that children with SB used an inappropriately overfamiliar style of talking in terms of expressive prosody. The study by Henderson, Murdoch and Ozanne (1989, cited by Murdoch, 2011) found that five of nine children with SB showed abnormal expressive prosody, characterised by inappropriate prosody, vocal intensity, or vocal quality. Huber-Okrainec and colleagues (2002: 598) reported that children with SB "were more dysfluent, had more ataxic dysarthria features, and had a slower speech rate" than typically developing (TD) children. Although these studies highlighted expressive prosody can be atypical in children with SB, none have reported the strengths and weaknesses of expressive and receptive prosodic skills in this clinical group in a systematic way. Therefore, a preliminary study was conducted to address this gap by using *Profiling Elements of Prosody in Speech-Communication* (PEPS-C) (Peppé & McCann, 2003).

PEPS-C is a computerised test of prosodic skills which adopts a psycholinguistic approach (McCann, Peppé, Gibbon, O'Hare, & Rutherford, 2007; Peppé, Cleland, Gibbon, O'Hare, & Martínez-Castilla, 2011; Peppé & McCann, 2003). A psycholinguistic approach allows for the user to identify the area of deficits in prosody processing in terms of input (e.g. perception or comprehension), mental representations (e.g. knowledge relating phonetic form to meaning) or output (e.g. lower level phonetic production). Hence, unlike other prosody assessments (see Peppé, 2011), PEPS-C assesses both receptive and expressive prosodic skills. The test produces numeric results which can be used to build a comprehensive profile of strengths and weaknesses in prosodic skills. Different versions of PEPS-C have been developed for assessing speakers of a number of European languages and different varieties of English, such as Irish-English (Foley, Gibbon, & Peppé, 2011; Gibbon & Smyth, 2013).

With reference to Stackhouse and Wells' psycholinguistic model of speech processing (1997), PEPS-C assesses the following components involved in

prosody processing: (1) the Input Form tasks assess a child's ability to discriminate prosodic patterns (i.e. variations in pitch, loudness, and duration) without reference to lexical representations; (2) the Input Function tasks assess whether the child has an accurate mental representations of prosodic patterns; (3) the Output Function tasks assess whether the child is able to access accurate motor programmes; and (4) the Output Form tasks assess the integrity of motor planning and execution. As the results of the 12 PEPS-C subtests indicate whether there is a breakdown in receptive and expressive prosody processing in children with prosodic disturbances, the test has been used to evaluate prosodic skills in children with high-functioning autism (see Peppé, 2011) and those with Williams syndrome or Down's syndrome (see Stojanovik & Setter, 2011). This preliminary study investigated (1) whether PEPS-C is an appropriate tool for assessing children with SB; and (2) whether prosodic skills are affected in children with SB by comparing the prosodic skills profile of children with SB to that of age-matched typically developing children.

Method

Participants

Two children with SB and eight TD children took part in this preliminary study. The two children with SB were recruited from a hospital in the south of Ireland. Both children were female – the first participant (SB1) was 12;4 of age and had a diagnosis of spina bifida myelomeningocele with a thoracolumbar lesion; and the second participant (SB2) was 13;9 of age and had a diagnosis of spina bifida myelomeningocele with a sacral lesion. Both children had hydrocephalus which was treated with a ventriculoperitoneal (VP) shunt. SB1 was born at 31 weeks and had a VP shunt inserted at two months of age; the course of which has been unremarkable. SB2 was born full term. A VP shunt was inserted at six months of age for SB2 and it remained uneventful prior to participating in this study. SB2 also takes sodium valproate due to a previous history of epilepsy (no episodes for the past ten years).

According to the speech and language therapy report, both children with SB had concomitant language difficulties – their language abilities fell anywhere from being within normal limits to 2 or more standard deviations (SDs) below the mean of their age-matched peers depending on the area of language being tested. Abstract language was the most challenging area for them. SB1 achieved a core language score of 82 on the *Clinical Evaluation of Language Fundamentals* (CELF-4) (Semel, Wiig, & Secord, 2006) at age 11 years, with standard scores falling between 74 and 86 (norm = 100; SD = 15) on other domains. SB2 achieved a total standard score of 69 (norm = 100; SD = 15) on the Test of Language Competence (Wiig & Secord, 1989) at the age of around 12 years, with domain standard scores falling between 3 and 9 (norm = 10; range = 3).

For cognitive ability, psychological assessment for SB1 administered prior to this study using *Wechsler Intelligence Scale for Children* (WISC-IV) (Wechsler, 2003) identified clinically significant variability among index scores; hence, a full scale IQ score could not be meaningfully interpreted. However, her Verbal Comprehension Index and Perceptual Reasoning Index were combined to obtain a General Ability Index, which fell within the borderline range of learning difficulty. SB2 was assessed using the same test at age 12 years. Similar to SB1, SB2's results could not be globally interpreted due to variability among index scores, however, a General Ability Index was calculated and the score fell within the low average range of ability.

Both children also had visual perception difficulties in line with their diagnosis of spina bifida and hydrocephalus. SB1 was assessed using the *Beery-Buktenica Developmental Test of Visual-Motor Integration* (Beery & Buktenica, 2010) at age 11 years. She attained a standard score of 77 (norm = 100; range = 10) on the Visual Perception section. SB2 was assessed using the *Test of Visual-Perceptual Skills* (Martin, 2006) at age 12 years. She scored below the normal range for two subtests – Form Constancy and Figure Ground. There was no indication that either participant had hearing difficulties at the time of data collection.

The eight TD children (four females aged between 12;2 and 13;9, and four males aged between 12;2 and 13;3) were recruited from a primary school and a secondary school in the Cork region. All the TD children had no history of speech, language, hearing, or learning difficulties, nor structural anomalies according to their parents' report. The absence of any structural anomalies of the oral cavity was confirmed by an oral peripheral examination (see below). All ten participants spoke Irish-English as their first language.

Materials

The materials included an oral peripheral examination adapted from the one by Robbins and Klee (1987) and the Irish-English version of PEPS-C. The oral peripheral examination included evaluation of the appearance and function of the lips, mandible, and tongue; as well as respiratory function and the ability to produce sound sequences. PEPS-C was administered using a laptop and a headset with noise cancelling microphone was used to record the

participants' responses for the Output tasks of the test. The test consists of 12 subtests (but see PEPS-C (n.d.) for the latest 2015 version of the test) incorporating the following two dimensions: (1) "Input" tasks that assess perception and comprehension versus "Output" tasks that assess speech generation and production; and (2) "Form" tasks that test lower level phonetic processing devoid of meaning versus "Function" tasks that test higher level processing accessing meaning. The "Forms" tasks assess intonation patterns in single words ("Intonation") and short phrases ("Prosody"). Whereas the "Function" tasks assess the following communicative functions: (a) "Turnend", where prosody indicates whether it is a question or statement; (b) "Affect", where prosody communicates emotion through vocal pitch movement; (c) "Chunking", where prosody delimits an utterance into grammatical, lexical or pragmatic units; and (d) "Focus", where phonetic prominence or emphasis is measured. Each subtest contains 16 test items and a score of 12 or higher indicates competence in the Function or Form assessed. The 12 subtests are also detailed in a number of publications (e.g. McCann et al., 2007; Peppé et al., 2011; Peppé & McCann, 2003).

Procedure

The testing took place in a quiet room in the hospital or the school from which the participants were recruited. Written consent was obtained from each participant before data collection. The tester administered the oral peripheral examination first, followed by PEPS-C. None of the participants showed any structural anomalies of the oral cavity, nor problems in producing sound sequences. One TD child was noted to have a frontal lisp; however, this did not seem to be of clinical relevance to this study.

As instructed in the PEPS-C user manual, a pre-test vocabulary check and a 'same/different' concept check were completed before administering the actual test, to ensure that any incorrect responses were not due to problems understanding the vocabularies or the concept of 'same/different' (Peppé & McCann, 2003). Each of the 12 subtests includes two demonstration items and two practice items to familiarise the participants with the task format. Each task was introduced separately and instructions were presented as per the script that comes with the test.

For the four Function Input tasks, the participants were required to listen to an audio stimulus each time and select the picture from two options that matches the audio stimulus. In the two Form Input tasks, the participants were played two audio stimuli and they had to indicate whether the stimuli are the same or different. For the four Function Output tasks, the participants were

prompted by a picture each time to produce a response using an appropriate prosody pattern (e.g. asking "apple?" when the picture showed a person offering an apple in "Turnend"). The tester (second author, a native Irish-English speaker and Speech and Language Therapist-in-training) then made an online judgement of the participants' intended meaning (e.g. "Apple." versus "Apple?"). In the two Form Output tasks, the participants were played a target word (or short phrase) each time and they had to produce the utterance in exactly the same way. The tester judged online whether the imitation was 'good', 'fair' or 'poor'.

All responses for the Output tasks were audio-recorded and the tester repeated the perceptual judgements within one week after the date of testing to measure intra-rater agreement. The intra-rater agreement was 91.3 % (876 out of a total of 960 Output task responses). For the 84 responses that showed an intra-rater disagreement, a second rater (a native speaker of Irish-English and Speech and Language Therapist-in-training) was asked to listen and score the responses. These judgements were then discussed and the scores of the participants were revised according to the consensus judgements.

The entire testing for each participant took approximately one hour. Ethics approval for conducting this study was obtained from the Clinical Research Ethics Committee of the Cork Teaching Hospital.

Data analysis

Once all the subtests were completed, the auto-scoring suite which accompanies the PEPS-C assessment was used to convert the participants' responses saved on the laptop into scores. For the six Input tasks and the four Function Output tasks, a score of 1 was given to each correct response and a score of 0 to each incorrect response. For the two Form Output tasks, a score of 1 was assigned to a good imitation of the intonation or prosody pattern, 0.5 to a fair imitation, and 0 to a poor imitation. Mann-Whitney U tests were conducted to examine whether there was any difference in the scores between the children with SB and the typically developing children in each of the 12 PEPS-C subtests.

Results

The raw scores for the six Input tasks and six Output tasks of the two children with SB and the mean scores of the eight TD children are displayed in Figure 1 and 2 respectively. Results of the Mann-Whitney U tests showed that

children with SB had significantly fewer correct responses than their TD peers in two of the six Input tasks – "Focus" (mean score = 14 for SB; mean score = 16 for TD; $p < 0.05$) and "Prosody" (mean = 13 for SB; mean = 15.6 for TD; $p < 0.05$); as well as five of the six Output tasks – "Turnend" (mean = 9 for SB; mean = 15.1 for TD; $p < 0.05$), "Affect" (mean = 7 for SB; mean = 15.9 for TD; $p < 0.05$), "Focus" (mean = 8.5 for SB; mean = 11.9 for TD; $p < 0.05$), "Intonation" (mean = 7 for SB; mean = 15.6 for TD; $p < 0.05$), and "Prosody" (mean = 10.3 for SD; mean = 15.8 for TD; $p < 0.05$). The difference in the mean scores between the two groups of children was not significant for the other four Input tasks – "Turnend", "Affect", "Chunking", and "Intonation"; and the Chunking Output task ($p > 0.05$).

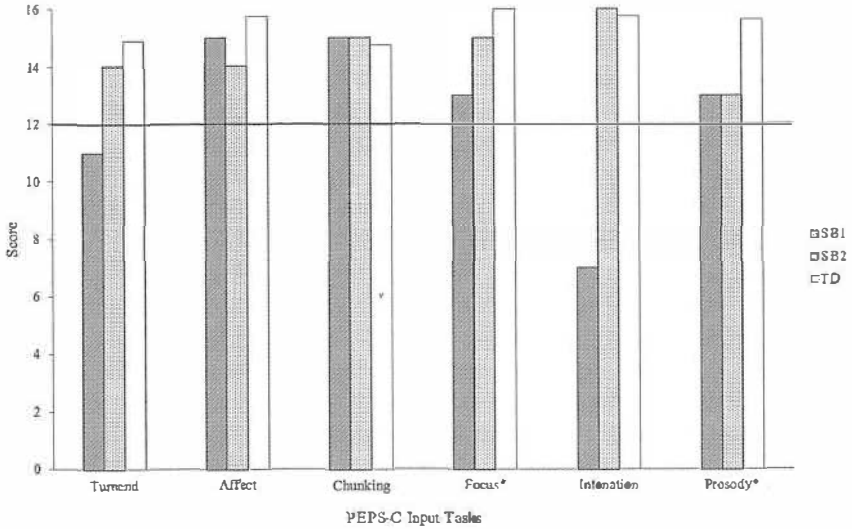


Figure 1. Scores for the six PEPS-C Input tasks of the two children with spina bifida (SB1 and SB2) and mean scores of the eight typically developing (TD) children. The horizontal line (at score 12) indicates minimum level of competence and the asterisks show significant difference between the two groups of children.

Applying the cutoff score of 12, SB2 and all TD children (except TD8 who scored 9 in Chunking Input) reached competence level for all Input tasks (see Figure 1 and Table 1 p. 140). SB1 reached competence level in four of the Input tasks – "Affect", "Chunking", "Focus", and "Prosody". For the Output tasks, all TD children scored 12 or above in all the tasks, except TD1 and TD8

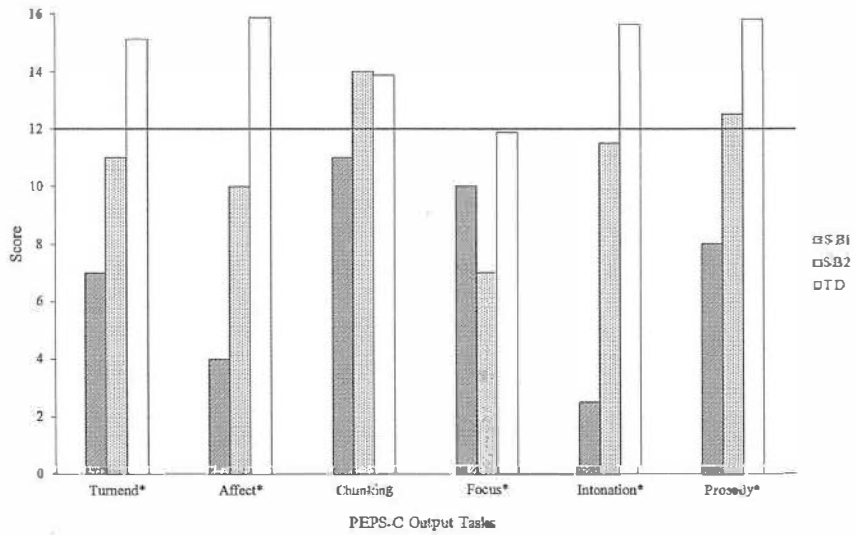


Figure 2. Scores for the six PEPS-C Output tasks of the two children with spina bifida (SB1 and SB2) and mean scores of the eight typically developing (TD) children. The horizontal line (at score 12) indicates minimum level of competence and the asterisks show significant difference between the two groups of children.

who scored 11 in "Focus Output", resulting in a mean of 11.9 for the TD group. SB1 did not score 12 or above in any of the Output tasks; while SB2 scored above 12 in two tasks – "Chunking" and "Prosody". The total score of the six Input tasks was 74 for SB1 and 87 for SB2; whereas the total score ranged between 85 and 95 with a mean of 92.8 for the TD children. For the Output tasks, the total score was 42.5 for SB1 and 66 for SB2; whereas the TD children scored between 85 and 91, with a mean of 88.2.

Table 1. Mean, standard deviation (SD), and range of scores for the PEPS-C Input and Output tasks of the eight typically developing (TD) children

Task	Input			Output		
	Mean	SD	Range	Mean	SD	Range
Function						
Turnend	14.9	1.0	13-16	15.1	1.1	13-16
Affect	15.8	0.5	15-16	15.9	0.4	15-16
Chunking	14.8	2.4	9-16	13.9	0.4	13-14
Focus	16.0	N/A	N/A	11.9	0.6	11-13
Form						
Intonation	15.8	0.5	15-16	15.6	0.4	15-16
Prosody	15.6	0.5	15-16	15.8	0.3	15-16
Total score	92.8	3.3	85-95	88.2	1.9	85-91

Note. N/A – all children scored 16 for Focus Input

Discussion

This is a preliminary study which explored whether PEPS-C is an appropriate tool for evaluating children with SB; and whether there was any difference in terms of expressive and receptive prosodic skills between children with SB and TD children of comparable chronological age. The results showed that the children with SB had significantly lower scores than the TD children in two of the six subtests that assess receptive prosodic skills and five of the six subtests that assess expressive prosodic skills. "Chunking" is the only prosodic function where there was no significant between-group difference in both Input and Output tasks. Both children with SB demonstrated better receptive prosodic skills compared to their expressive prosodic skills – their total score of the Input tasks were relatively higher than that of the Output tasks; and there were more Output tasks than Input tasks where they scored significantly lower than the TD children. Furthermore, SB1 had lower score than SB2 for both Input and Output tasks, which might be related to the age of the child (SB1 was younger than SB2), the level of spinal cord lesion and the different levels of language abilities of the two children (see Method section above). It is possible that the visual perception difficulties of the two children with SB might have impacted on their performace on PEPS-C (and other psychometric tests) as visual stimuli were used. However, both children were able to complete the pre-test vocabulary check and 'same/different' concept check of PEPS-C, which indicated sufficient visual perception abilities for undertaking the test.

The present results revealed a useful profile of each child's prosodic strengths and weaknesses. Applying the psycholinguistic model (Stackhouse & Wells, 1997), the results of PEPS-C showed that SB1 was probably able to discriminate the intonation patterns of one- to two-syllable words but had difficulties in discriminating the prosodic patterns of longer utterances, such as four- to five-words phrases; whereas SB2 was competent in both. Based on the results from PEPS-C, the two children with SB had fairly accurate mental representations of prosodic patterns used for signalling a question or a statement (assessed in "Turnend"); like or dislike ("Affect"); phrase boundary ("Chunking"); and contrastive stress ("Focus"). Although there was a significant difference between the children with SB and the TD children in the "Focus Input" tasks, both children with SB scored above 12 in the task and were therefore considered as competent for this function. The significant between-group difference in "Focus Input" was probably due to the fact that all the TD children attained the maximum score of 16 in this task.

Expressive prosody is the area in which the children with SB in this study showed major difficulties. SB1 did not achieve competence level for all of the Output Function and Form tasks; and SB2 was competent in only one Output Function task ("Chunking") and one Output Form task ("Prosody"). As a group, both children with SB showed significantly lower scores than the TD children in all of the tasks except "Output Chunking". Moreover, investigators' informal observation of the children's speech revealed low vocal loudness, high pitch, and limited variation in intonation (i.e. monopitch). The results of PEPS-C in this study indicated that the children with SB might have difficulties in accessing the motor programme as well as problems with motor planning and execution. Although further analysis of the speech errors would be needed in order to find out whether the expressive prosody difficulties were due to deficits with motor programming (i.e. apraxic type of errors) or motor execution (i.e. dysarthric type of errors), the current literature suggests that it is dysarthria that was observed in this clinical group (see Huber-Okrainec et al., 2002). Huber-Okrainec and colleagues (2002) hypothesised that children with SB would have ataxic dysarthria based on the fact that bilateral cerebellar dysmorphology is often associated with SB. The authors assessed for the presence of ataxic dysarthria speech features and they reported that children and adults with SB showed significantly more articulatory inaccuracies, phonatory-prosodic insufficiency (defined as "harshness, monopitch, and monoloudness" (Huber-Okrainec et al.: 596)), and prosodic excess (defined

as "excess and equal stress, prolonged phonemes, slow rate, and prolonged intervals" and "short phrasing" (ibid.)) than the TD individuals of comparable age.

For the TD children in this study, as their age was between 12;2 and 13;9, it was expected that their test scores should be similar (if there was a ceiling effect) or higher than those of the 10–11 year olds reported in Foley and colleagues' study (2011). Similar to the study by Foley and colleagues, there were some TD children in this study who attained the maximum score of 16 in each of the subtests, except "Chunking Output" and "Focus Output". However, none of the TD children achieved the maximum score in all Input tasks or all Output tasks. Overall, the 12–13 year olds in this study showed a slightly higher mean score than the 10–11 year olds in Foley and colleagues' study in five Input tasks and five Output tasks. The exceptions were (1) "Chunking Input", where the mean score of the two groups was the same and the 12–13 year olds in this study had a bigger SD and range of scores; and (2) "Focus Output", where the mean score of the 12–13 year olds in this study was lower than that of the 10–11 year olds in Foley and colleagues' study but the SD and range of scores of the 12–13 year olds were smaller. We agree with Foley and colleagues (2011: 38) that the "development of these [prosodic] skills continues, leading to acquisition of all prosodic skills by the age of 10 or 11 years". Moreover, we suggest that, due to the variations in prosodic skills among TD children, some of these children may still show further progress in their prosodic skills by the age of 12 or 13 years. However, further investigation employing a larger sample size is warranted as the sample size for each age group in Foley and colleagues' study and the present study was small.

Conclusion

This preliminary study showed that the computerised prosody test, PEPS-C, is adequate for evaluating prosodic skills in children with SB. As revealed by the test, the two children with SB had relative strengths and weaknesses in different areas of prosody comprehension and production. Specifically, they showed major difficulties in expressive prosody, which is in agreement with the findings reported in the literature (e.g. Huber-Okrainec et al., 2002; Murdoch, 2011). Further investigation using perceptual judgements and acoustic analysis of speech are recommended in order to gain insights into the nature of the expressive prosody difficulties in this group of children. Similar to the prosody profile, the two children with SB also showed relative strengths and

weaknesses in the language domain and cognitive domain. Although SB1 seemed to have relatively better language skills than SB2, her PEPS-C scores were lower than those of SB2. Hence, future research is needed to investigate how prosodic skills relate to language comprehension and production in children with SB.

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