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Language Profile of a Vietnamese Child with Prader-Willi Syndrome

Yen Nguyen [1], Ben Phạm [2*], Hang Phạm [3], Van Phạm [4], Thu Dinh [2], Phuong Nguyen [2], Thanh Nguyen [1]

[1] Psychology and Education Department, Vietnam National Academy of Educational Management, Hanoi, Vietnam

[2] Faculty of Special Education, Hanoi National University of Education, Hanoi, Vietnam

[3] Research Center for General Education Development, Vietnam National Institute of Educational Sciences, Hanoi, Vietnam

[4] VietSpeech EDU Early Intervention Center, Hanoi, Vietnam

ABSTRACT

Prader-Willi Syndrome (PWS), a low incidence genetic syndrome, exhibits complex difficulties characterized by infantile hypotonia and feeding, motor, cognition, behavioral phenotype, and hypoplasia. Children with PWS also may demonstrate delay in language development. However, this has not been well-investigated in research. To date, there is no research reporting the speech and language abilities of Vietnamese speaking population with PWS. This case report is the first attempt to describe the speech and language profile of a Vietnamese-speaking child with PWS. A 5-year-old boy with PWS participated in the study by completing speech and language tasks available in the Vietnamese language, including oral motor, speech accuracy, intelligibility, speech participation, receptive and expressive vocabulary, sentence repetition, rapid naming, phonological awareness skills, nonword repetition and telling and retelling stories. The performances of the majority of the tasks were digitally audio-recorded, then transcribed for speech and language analysis. The tasks' scores were calculated then compared to the corresponding results available for typically developing same-aged peers and those with speech and language disorders. The child's scores demonstrated poor oral motor skills, severe impairment of speech accuracy, and low speech intelligibility and participation. Language skills were low in vocabulary, grammar and narrative skills. Despite some facial abnormalities, the child demonstrated normal vocal characteristics. This case report supports the fact that Vietnamese individuals with PWS may experience a delay in language development compared to typically developing peers, even though the individuals receive early intervention services; therefore, continually speech

*CORRESPONDING AUTHOR:

Ben Phạm, Faculty of Special Education, Hanoi National University of Education, Hanoi, Vietnam; Email: ben.phamthi@hnue.edu.vn

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and language support the individuals in mainstream school is essential.

Keywords: Case Report; Comprehensive Assessment; Assessment Protocol; Speech and Language; Vietnamese; Prader-Willi

1. Introduction

Prader-Willi Syndrome (PWS) is a complex multisystem genetic disorder[1, 2]. Prevalence of this disorder is rare, affecting only two out of 10,000–15,000 individuals, with an equal ratio between boys and girls[3]. PWS is caused by the malfunction of imprinted genes on chromosome 15 that is identified by a molecular genetic test[1]. Based on the causes, PWS includes three subtypes: (1) the absence of the paternally active genes in the chromosome 15q11-13 region (DEL), causing about 75% of all PWS cases; (2) the inheritance of two copies of the maternal chromosome 15 (mUPD), causing about 20%; and abnormalities in the structure of chromosome 15, causing 5% of PWS cases[1].

PWS may be clinically diagnosed by a number of phenotype characteristics[4]. Individuals with PWS exhibit universal physical characteristics as "short stature, small narrow hands, short broad feet"[5], "neonatal and infantile hypotonia, feeding problems in infancy, obesity with onset between 12 months and 6 years and distinct facial features including a narrow bifrontal (forehead), diameter, almond-shaped eyes, a narrow nasal bridge, and a down-turned mouth with a thin upper lip, hypogonadism"[6] and hyperphagia. Hypopigmentation and distinctive facial features are experienced more by individuals with the DEL subtype of PWS compared to those with the mUPD and the structural abnormalities[1, 7].

In addition to physical features, individuals with PWS also experience psychological and mental health issues including behavior, cognition, and speech and language problems. Complex and multifaceted behavioral and emotional problems are one of the universal characteristics associated with PWS and are well described in many studies over the decades in many countries[5, 8–11] developed a Prader-Willi Syndrome Profile to measure behavioral and emotional problems in PWS, and reported eight distinctive characteristics: rigidity, insistence, aggression, repetitive questioning, compulsivity, depression and anxiety, hoarding, negative distorted thinking and magical distorted thinking, in addition to two lower internal consistency factors such as skin-picking and nail biting. According to results from the Prader-Willi Syndrome Profile[10], there are generally no significant differences between behavior problems and demographics, including IQ scores, gender, age, BMI, parental socio-economic status, and region. Some behavior problems of PWS differ between the genetic subtypes. For example, individuals with the DEL subtype scored strongly in the repetitive questioning and speech behaviors but less strongly in the skin-picking behavior compared to other subtypes.

With respect to cognition, the majority of individuals with PWS experience mild to moderate intellectual disabilities, with their overall IQ score ranging from 55 to 70[5]. It has been reported that individuals with PWS present better performance IQ than verbal IQ, show strengths in long-term memory, visual-spatial and jigsaw puzzle skills, and weaknesses in auditory verbal processing, auditory short-term memory and linear or temporal processing[1, 9, 12]. Individuals with the DEL subtype show relative strength in block design, object assembly and picture completion, whereas the mUPD subtype shows strengths in information, arithmetic, vocabulary and receptive language skills. Therefore, those with the mUPD subtype have higher verbal IQ scores than those with the DEL[13, 14].

Language delays have been reported as a common characteristic associated with PWS; however, the analysis of the language deficits of PWS has received relatively less attention compared to the clinical and behavioral idiosyncrasies[5, 6, 15]. Language deficits in PWS may be presented in a wide range of types and severity throughout one's lifespan. Some of these deficits may be the result of abnormalities associated with PWS[6]. For instance, due to the distinctive mouth and dentition abnormalities, PWS present difficulties in articulatory abilities and vocal pitch[6]. Similarly, individuals with PWS present low verbal IQ scores that may be the reason for their poor receptive and expressive language skills. The behavioral disturbance, in turn, affects social interaction and pragmatic skills[6]. It has also been indicated that overall language abilities in individuals with PWS are below chronological age, with expressive language

more impaired than receptive language[6, 16]. This population presents limitations with respect to all language aspects and language development stages. At the prelinguistic stage, individuals with PWS show limited babblings[6]. At the emergence language stage, this group begins speaking the first words late, ranging from 18 months to 6 years old[6]. At the developing language stage, individuals with PWS show limitations in lexical comprehension and expression, average mean length utterance (MLU), morphosyntactic abilities, narrative skills, conversational skills, and pragmatic skills[6, 16]. Language deficits were also reported in individuals with PWS who were bilingual speakers[15]. Within the PWS population, different genotypes and ages present phenotypic language discrepancies[5, 6, 15]. To our knowledge, research on language abilities is far behind research on medicine, genetics and behavior problems in the PWS population. Furthermore, there is no research on the language abilities of the Vietnamese-language speaking population as of yet. Although there are universal linguistic characteristics in the PWS population across different languages, there are specific linguistic features, particularly for a syllabic and tonal language like Vietnamese.

With respect to research on the PWS Vietnamese-speaking population, there are three studies that mainly focused on clinical and genetic features[17–19]. By using retrospective design[19], a cohort of 80 patients diagnosed with PWS by fluorescence in *situ* hybridization (FISH) was conducted at the National Hospital of Pediatrics from 2007 to 2015. Clinical features showed that there were six times more males with PWS compared to females. Half of the cohort that was diagnosed was under 5 years old, and the majority of the cohort had hypotonia (85.7%), hyperphagia (86.4%), cryptorchidism (91.7%). Similarly, a cohort of 118 patients diagnosed with PWS by FISH at the same hospital over 10 years starting from 2007[17]. 74.6% (88/118) of this cohort originated from the deletion of the paternal chromosome 15 q11-q13 region (DEL) and 8.47% (10/118) resulted from the mUPD subtype. The majority of the participants were diagnosed during the neonatal period (8.5%) and were under 2 years old (58.3%). Hypogonadism and hypotonia were common phenotypic characteristics of this Vietnamese cohort with PWS. Recently, the molecular genetic analysis of a case study of a one-month-old boy diagnosed with PWS caused by a rare abnormal uniparental disomy of maternal chromosome 15q11.2-q.13 region (mUPD15) was reported[18]. To the best of our knowledge, only the three abovementioned studies which focused on the clinical and genetic features of Vietnamese children with PWS have been published. Psychological characteristics including behaviors, cognition and language skills within the Vietnamese PWS population have not been studied as yet. This present study is the first attempt to contribute to previous research with data from a case study describing the linguistic characteristics of a Vietnamese-speaking boy with PWS through a comprehensive speech and language assessment that was conducted by an interdisciplinary assessment team. To obtain the overarching aim, the research questions are as follows:

1) What are the speech and language characteristics of a Vietnamese-speaking child with PWS?

2) What are the differences in the speech and language characteristics of this boy with PWS compared to the speech and language abilities of those with typical development and language disorder?

2. Materials and Methods

2.1. Ethical Considerations

This case report was part of our research project managed by the Institute of Clinical Psychology Vietnam under the grant number NCUD.05-2019.67. Ethical approval for the research project was obtained from the Institute board (number: 2021/05). Written consent forms were obtained from the child's parents for volunteering involvement to provide information about their child. They also permitted their child to attend all assessment sessions. The child's parents allowed the assessment team to use their child's assessment information for any forms of publication including this case report. In addition to the parents' consent, we also asked permission from the child participant by obtaining his assent to participate after hearing a child-friendly description of the research.

2.2. Participant with Prader-Willi Syndrome

A boy aged 5 years and 11 months old visited our educational early intervention center for a comprehensive speech and language assessment by a request from a primary school where he was going to enrol in the next three months. He met

our assessment team who were the author team of this paper, including three speech and language pathologists, three special educators and a psychologist. The referral letter from the primary school reported that he was diagnosed with Prader-Willi Syndrome after birth. He received different self-funded intervention services from birth and he progressed in many development areas to the point that he could attend a mainstream preschool. Currently, he has daily intervention services after school. However, the primary school's biggest concern was on his speech and language skills, which it raised by requesting a comprehensive assessment when he was in a transition program from the preschool to this primary school.

All information in this section on the participant's development history was provided by the participant's mother and data analysis is content analysis for qualitative description. The participant is the first child in the family, and born through caesarean section in the 36th week of gestation due to the mother's depleted amniotic fluid. His weight at birth was 2.8 kg. After birth, he had to be hospitalized because of respiratory failure. Being a dentist with basic clinical knowledge, the participant's mother was suspicious about his distinctive physical appearance and obvious decreased muscle tone. Clinical examination performed at the hospital showed obvious symptoms of decreased muscle tone, inability to suckle, bilateral undescended testicles, overlapping skull joints, abnormal face with almond-shaped eyes that were far apart, flat nose base, thin lips and pale skin. He was diagnosed with PWS in the DEL subtype by a FISH genetic test in the first month of age. He stayed at the hospital for a month. He started taking growth hormone treatment when he was 4 years old that was reported to support early psychomotor development among the PWS population[20] and is provided at hospitals in Vietnam to patients in the first 6 months of their life[17].

He was screened for hearing ability at the hospital after birth through testing and was found to have normal hearing. He had problems with feeding during infancy. Eating was a big concern as he ate uncontrolled but only preferred soft to crunchy or hard solid food. He had hypotonia that needed to be checked every day. Upper respiratory infections occurred frequently. For motor skills, he was able to manage independent walking by 3 years old; however, going up and down-stairs still required leaning on the stair banisters. His mother complained that he was clumsy in using both hands. His facial appearance was said to influence articulation, such as having a small lower jaw and limited saliva output that resulted in dental decay and xerostomia. He did not have excessive drooling but the saliva output was always seen as white on both sides of the mouth when he talked. He said his first words when he turned 3. He had a number of behavior problems such as temper tantrums, stubbornness, compulsive behavior, skin picking, being argumentative, emotional lability, depression, and verbal and physical aggression. All of the above was reported to seriously interrupt the frequency and density of the speech and language intervention sessions.

The child received a report on nonverbal intelligence measured by the Primary Test of Nonverbal Intelligence (PTONI)[21] with a score of 82 (the standard score is > 70) when he was 5 years old. When the child was two years old, he also completed the Vietnamese version of the K-test. This test was validated according to Kyoto's Developmental Scale[22] to assess three areas: Postural-Motor Area (PM), Cognitive-Adaption Area (CA) and Language-Social (LS). Unfortunately, the mother did not provide this assessment report; however, she remembered that his scores for all sessions were one year behind compared to those of his peers. Additionally, he only started verbalising his first words when he turned three years old.

2.3. Speech and Language Assessment Protocol and Instruments

The speech and language protocol used for the child comprised both indirect and direct assessment. Indirect assessment refers to collecting information about the speech and language skills of the children from the child's parents whereas direct assessment refers to what the child performed on specific speech and language tasks.

The instruments used for indirect assessment included the Intelligibility in Context Scale Vietnamese (ICS-VN)[23, 24] and the Vietnamese version of the FOCUS©-34: Focus on the Outcomes of Communication Under Six[25]. The ICS-VN and FOCUS-34 were completed by the child's mother.

ICS-VN[23] was originally created in English and translated into over 60 languages across the world. This scale was translated and validated with Vietnamese-speaking children. This scale is available for free at: https://cdn.csu.edu.au/__data/assets/pdf_file/0004/

2175988/ICS-Vietnamese-English-Final-Nov-2015.pdf.

The ICS-VN is a parent's reporting form comprising 7 questions to examine how the child's speech intelligibility was understood by different communicative partners. Communicative partners include parents, family members, relatives, friends, teachers, acquaintances and strangers. Each question was rated by a 5-Likert scale with the items Always, Usually, Sometimes, Rarely and Never. The total raw score of the ICS-VN was 35 and the average score was 5.0. Normative data of the ICS-VN was 4.4 and 4.5 out of 5.0 for typically developing Vietnamese children who spoke the Vietnamese northern dialect [24, 26] and southern dialect [27] respectively.

FOCUS-34 [25] was originally created in English and translated into many different languages, including Vietnamese. The Vietnamese version of the FOCUS-34 for parents and clinicians can be found at: . The parent version of the FOCUS-34 includes 34 questions related to children's communication skills. Each question is rated by a 7-Likert scale. The total raw score of the FOCUS-34 is 238 points.

For direct assessment activities, instruments were selected to assess oral motor skills, speech accuracy, receptive and expressive vocabulary, rapid automize naming task, nonword repetition, sentence repetition and narrative skills.

The Oral Motor Assessment (OMA) was conducted based on the Oral Speech Motor Protocol for children from 2 years old and 6 months to 7 years old [28]. The checklist of OMA included 24 items for oral structure, where each item was rated as "normal" = 1 point and "abnormal" = 0; and 7 items for oral function, including speaking aloud the sounds /p/, /t/, /k/ separately, the sequences /p-t-k/, /u-i/ and repeated two sentences for hypo- and hyper-nasality checking. The 7 items had a total score of 14 as they were rated at three levels, 0 point = "do not perform", 1 point = "initially perform", and 2 point = "perform correctly". Due to limited normative data on Vietnamese-speaking children for the OMA, the results of this task were described qualitatively.

For speech accuracy, the Vietnamese Speech Assessment [29, 30] was used to assess the production of initial consonants, semi-vowels (medial and final), vowels, final consonants, and tones. VSA is a single picture naming task including 77 words and it takes about 10 to 15 minutes to complete. Measures were calculated, including the percentage of initial consonants correct (PICC), percentage of semi-vowels correct (PSVC), percentage of vowels correct (PVC), percentage of final consonants correct (PFCC), and percentage of tones correct (PTC). The child's results were compared to the normative data provided for speech accuracy of typically developing Vietnamese-speaking children aged 5 years old [31]. Additionally, phonological processes spoken by the child were described and were compared to phonological processes spoken by typically developing peers [31].

The child completed a form on Speech Participation and Activity Assessment of Children Vietnamese (SPAAC) [32] to measure how he feels when he talks with different people and in different situations by colouring the corresponding symbols that match to his emotions. The symbols used in SPAAC are five types of face symbols corresponding to emotions of "happy", "in the middle", "sad", "another feeling" and "don't know". The Vietnamese version of SPAAC is available for free at: https://cdn.csu.edu.au/__data/assets/pdf_file/0011/2755406/SPAA-C-Child-Vietnamese-English.pdf.

To assess language skills, six instruments were selected to administer to the child, which are available to use for free with Vietnamese-speaking children by registering for an account from the VietSLP webpage at: . The six instruments included: (1) Picture Identification, (2) Picture Naming, (3) Rapid Naming (RAN), (4) Sentence Repetition, (5) Nonword Repetition (NWR), and (6) Phonological Awareness Measure in Vietnamese (PhAM-V). All these assessment tasks were conducted online by using a computer. The examiner rated the corresponding printed record forms. At the word level, Picture Identification and Picture Naming were used to assess receptive and expressive vocabulary skills respectively [33]. In the Picture Identification task, the child was asked to hear a pre-recorded word while looking at four black and white pictures in two arrays on the computer screen at a time, then point to the corresponding picture. In the Picture Naming task, the child was asked to name a picture depicting the object at the same time as seeing it on the computer screen. Each task had a total of 60 items and the items between both tasks were not overlapped. The number of correct items of each task was calculated.

Another task used for the child at the word level was Rapid Naming (RAN). This test was used to assess the processing speed of naming familiar objects and digits [34]. The child was asked to complete two sections of objects and two sections of digits, each section comprising 36 items pre-

sented in random order in four rows per array. The time for completing the task was recorded and the result of the RAN was calculated by the total number of correctly named items of four sections divided by the total number of seconds [34].

For sentence level, the Sentence Repetition task was used to assess grammatical knowledge [35]. The task included two items for practice and 28 testing items. Each one consisted of 8 syllables and was placed in order by increasing the complexity of grammatical forms. The child was asked to complete the task by listening to each pre-recorded sentence at a time, then repeating the whole sentence. Precluding error and grammatical scoring systems, the results of the child's performance for sentence repetition used a binary scoring system by calculation of the total number of correctly repeated sentences divided by the total 28 sentences because under the binary scoring, the sensitivity value for the task was good in identifying language difficulties of the child. Errors made while the child repeated each sentence included substitution, omission, insertion or transposition, excluding dialectal variants and consistent articulation errors reported in the speech test.

To assess short-term phonological processing, Nonword Repetition [35] was used for the child as this measure has a significant difference between typically developing children and those with language difficulties. NWR has 20 items divided into 4 groups: 5 items of monosyllable nonwords, 5 items of disyllable nonwords, 5 items of trisyllable nonwords and 5 four-syllable nonwords, 50 syllables and 180 phonemes measures in total. The internal consistency of this task was high (Cronbach's alpha=0.76). The result calculated for the NWR task for the child was the percentage of correct phonemes out of 180 phonemes. The incorrect phonemes for this task excluded articulation errors made by the child reported in the speech test.

Together with NWR accessing phonological processing, the Phonological Awareness Measure in Vietnamese (PhAMV) was used [35]. This task included 36 items, 12 for blending, 6 for tone detection, 12 for segmentation and 6 for rime detection. The internal consistency of this task was high (Cronbach's alpha=0.84). The results for PhAMV were calculated by the total number of correct items out of 36.

At the discourse level, the Multilingual Assessment Instrument for Narratives (MAIN) [36] and translated into Vietnamese [37] was used to assess the narrative skills of the child. The MAIN manual to collect and analyze narrative language samples for the Vietnamese language is available at https://www.leibniz-zas.de/fileadmin/media/Dokumente/ZASPiL/ZASPiL_63/MAIN_Vietnamese_2020_Revised.pdf. Cat and Baby Goat Stories were selected to collect language samples. The Cat Story was read to the child and the child retold it while looking at the 6-sequencing pictures; then, the child was asked to tell the Baby Goat Story while viewing the 6-sequencing pictures. All responses were audio-recorded. Two examiners, who were speech and language pathologists and special educators, transcribed the language samples from two stories and got an inter-judge agreement at 94.9%. Language samples were analysed for Story Structure (with a maximum of 17 scores) and Story Comprehension (with a maximum of 10 scores). At the sentence level, the mean length of utterances (MLU) and the number of grammatically correct utterances were calculated. At the word level, the ratio of type token (TTR) was calculated. These measures were compared to the results of typically developing same-age peers [33].

2.4. Data Collection Procedure and Data Analysis

The procedure of this case report comprised five steps (illustrated in **Figure 1** below). Step one was referral when the primary school sent a referral letter and requested the child's parents to meet professional(s) for comprehensive speech and language assessment. The main purpose of the request was to have information about the child's speech and language skills to determine whether the child needs speech and language support once he was enrolled in primary school. The child's parents contacted an early intervention center to register for a comprehensive speech and language assessment for their child. A co-author of this paper, a speech and language pathologist, was working at this early intervention center. Given a background that Prader-Willi was a rare genetic syndrome and there was a lack of information on speech and language characteristics of children speaking Vietnamese, we decided to obtain the consent agreement from the child's parents to describe their child's speech and language characteristics in a paper after conducting the comprehensive assessment.

The next step was to collect the child's medical information and developmental history. We asked the parents to

provide any documents they kept for their child's medical profile. The child's mother was a dentist and she had a good manner of keeping medical records from her son's birth to the time of visiting our team. She also gave us information about his development history and answered the assessment team's questions related to her son's speech and language skills. It took us some time to go through the medical, health and development records and we only extracted information associated with the child's speech and language performance.

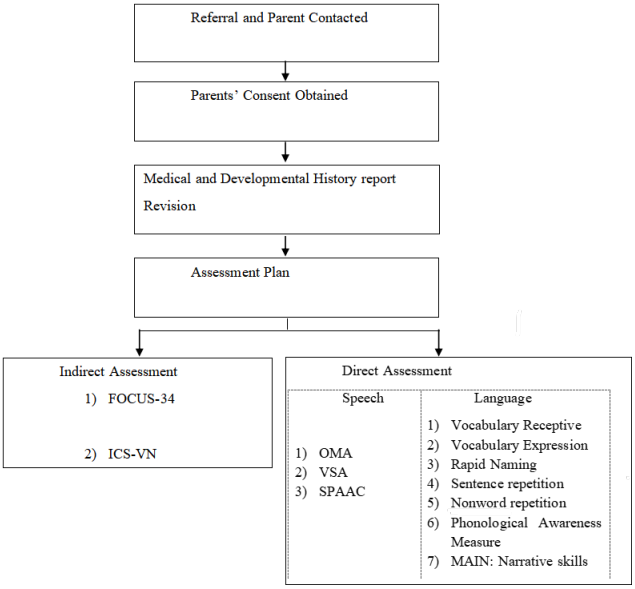

Figure 1. Procedure of collecting data.

Based on the background information provided by the child's parents, the assessment team made an assessment plan. We discussed the protocol of speech and language assessment used for this case and identified areas in speech and language skills that the child should be comprehensively assessed. We also discussed which assessment tools were available in the Vietnamese language that could be used to capture his speech and language skills. A number of available tests were collated to use with the child.

The final step was face-to-face assessment. We made the appointment with the parents to bring him to visit our assessment team at the early intervention center for a face-to-face speech and language assessment. At this step, we conducted an indirect assessment which the parents filled in with further specific forms or scales. We conducted a direct assessment with the child in which he completed speech and language tasks with the assessment team. We planned to meet him for about three hours in a morning based on an estimate of the time taken to complete all the collated speech and language tests available.

However, due to limited cooperation from the child, we had to conduct the direct speech and language assessment with him within five visits in two consecutive weeks, the first visit lasted 45 minutes, the second was an hour, the third was two hours, the fourth was 45 minutes and the last visit was an hour. In total, it took five and a half hours to conduct face-to-face assessment sessions in speech and language skills for the child.

With respect to data analysis, scoring all the tasks conducted for the child was independently rated by two or three members of the research team who had training in speech and language testing with children. The inter-rater reliability of task scores was high, at over 90% agreement for all tasks.

The language samples collected from the narrative skills were also used to assess voice characteristics that were rated based on the Voice Rating Severity Scale [38]. The three-level ratings were high-normal-low for pitch and hypernasal-normal-hyponasal for resonance and vocal quality was rated as normal, soft, harsh, hoarse or strained. Prosody was also noted. Voice characteristics were rated by two speech and language pathologists for a consensus on these scores.

The scores the child gained for each individual speech and language task was calculated and then compared to the scores of typically developing children at the same age if these data were available.

3. Results

3.1. Indirect Assessment: Intelligibility in the Context Scale and Communication Scores

The intelligibility score measured by the ICS-VN [23] was a total of 23 out of 35 points with a mean score of 3.3, compared to a mean score of 4.4 for typically developing children speaking the northern Vietnamese dialect.

The child's score from FOCUS-34 [25] rated by his mother was 137 out of 238 points. This score was similar to those of Vietnamese-speaking children with developmental language disorder (DLD) at141.90 points, and lower than those of the Vietnamese-speaking children who were developing normally at 202.27 points [33].

3.2. Oral Motor Assessment Results

Based on the OMA scale developed by Robbins and Klee[28], the participant received 19 out of 24 items for oral structures by rating "normal" = 1, "abnormal" = 0. "Abnormal" was considered by observable characteristics with 5 items of teeth decay, micrognathia (small lower jaw), narrow palatal arch, limited tongue movement, and swollen tonsils. Teeth decay and swollen tonsils were two characteristics seen in normal preschool children in Vietnam. Swollen tonsils were seen in many children, particularly in winter time in the North because children would get bronchitis or respiratory track inflammation. However, three other characteristics of micrognathia, narrow palatal arch and limited tongue movement were typically seen in PWS and these were cited as factors influencing articulation[6]. For oral function items, the participant achieved 10 out of 14 points. The participant could produce correctly the /p/ and /t/ sounds, the /u-i/ sequence, two sentences imitated but no hyper- or hypo-nasality was presented. The participant was not able to produce the /k/ sound and the sequence sounds of /p-t-k/ continuously.

3.3. Speech Accuracy Scores

The results of the percentage of correct phonemes were calculated from the VSA. The results are presented in **Table 1**.

Table 1. Speech accuracy scores of a Vietnamese child with Prader-Willi Syndrome in comparison to scores from typically developing children with speech sound disorder.

Measures	The Prader-Willi Syndrome participant (5;11)	SSD (*n*=15, age 5;5–5;11)[39]	TD (*n*=31, age 5;5–5;11)[31]
PICC	36.7%	76.9% (*SD*=19.3, 34.2–96.2)	92.11% (SD=7.02)
PFCC	40.5%	63.2% (*SD*=19.2, 4.8–88.1)	92.55% (SD=10.06)
PSVC	87.5%	89.2% (*SD*=27.1, 0–100)	99.60% (SD=1.60)
PVC	92.4%	92.8% (*SD*=8.36,65.8–98.7)	98.11% (SD=2.79)
PTC	91.4%	89.2% (*SD*=27.1, 0–100)	96.55% (SD=3.42)

The participant had lower scores in all speech accuracy measures than those of children with typical development and children with SSD. The participant's PVC, PTC, and PSVC scores were higher than his PICC and PFCC scores. These trends were similar to those of same-aged peers with typical development and SSD[31, 39, 40]. Based on the child's PICC and PFCC scores and diagnosis criteria of speech sound disorder[40], the child's speech accuracy skills met the criteria of a speech sound disorder diagnosis.

The participant had a wider range of phonological processes compared to those of typically developing children at the same age[31] The phonological processes range from common (more than 10% of occurence) to rare (less than 5% of occurences). The common systematic phonological processes were: fronting (e.g., /k/ → [t]), deaspiration (e.g., /t͡ʰ/ → [t]), glottal replacement (e.g., /x/ → [h]), stopping (e.g., /s/ → [t]) and common structural phonological processes were medial semivowel deletion (e.g., *hoa* (flower) /hwɑ1/ → [hɑ1]) and initial consonant deletion (*đũa* (chopstick) /duo3/ → [o5]). Phonological processes that were rare or atypical patterns included nasalization (e.g., *lược* (comb) /lɯɤk6/ → [mɯɤt6]), and gliding (e.g, *gà* (chicken) /ɣɑ2/→ [jɑ2]).

The child chose "sad" faces in the SPAAC form[32] for questions: "how do you feel when you talk to the whole class", "when you play with the children at preschool", and "when people do not understand what you say".

Regarding vocal characteristics rated by the voice scale[38], it was described that he had normal pitch, normal resonance and voice quality.

3.4. Language Ability Scores

Language skills were measured in three areas: vocabulary, grammar, and narration.

4. Discussion

The main aim of this case report is to describe the speech and language characteristics of a Vietnamese-speaking boy with Prader-Willi Syndrome. Since Prader-Willi Syndrome is a low incidence syndrome[1], the paper

presents a single case through a comprehensive assessment. This is the first paper describing the speech and language skills of Vietnamese-speaking individuals with this rare syndrome. In addition, although the profession of speech and language therapy in Vietnam is emerging, assessment protocols and tools for the Vietnamese language are sparse [41]. The paper demonstrates the assessment procedure that was implemented and the available tests that were used.

We conducted a number of assessment tasks that were available in Vietnamese to measure different areas of speech and language skills. Most tests were available for free so that professionals who are working with Vietnamese-speaking children in Vietnam and across the world may access these tools for use with this population.

The child was reported to have a history of speech and language difficulties when he started to talk at the age of three. The scores that the child gained in all speech and language tasks demonstrated that this child with Prader-Willi Syndrome had difficulties in both speech and language skills. His scores for speech accuracy met the diagnostic criteria for a speech sound disorder. Similarly, his scores in the language domain met the diagnostic criteria for language disorders. These findings align with studies on the speech and language skills of children with Prader-Willi Syndrome across different languages [6, 16].

Regarding oral motor skills, he presented facial abnormalities and poor coordination of oral movements which affected his articulation skills, particularly the velar sounds. This finding is similar to findings reported in previous studies [16, 42].

Regarding speech accuracy, the child's scores of PICC and PFCC from the VSA test and connected speech samples were below the mean scores of typically developing children, exhibiting severe speech disorder. Children with Prader-Willi Syndrome were reported to have speech sound disorders [43, 44] and this was true for the case described in this paper. Speech errors produced by the child included both substitutions and omissions exhibiting both systematic and structural phonological processes: fronting, backing, deaspiration, initial consonant deletion, and medial semivowel deletion. The child's speech errors showed the phonological contrast loss, meaning that he pronounced an error sound to substitute for a number of sounds in Vietnamese phonology. Phonological contrast loss is a clinical marker in identifying children with speech sound disorder [45]. These characteristics were similarly found in studies [43, 44] as speech accuracy of children with Prader-Willi Syndrome characterized by both typical and atypical phonological processes.

The child also obtained a lower score of speech intelligibility compared to the mean score of same-age peers and demonstrated having negative feelings when talking to others. Unlike previous studies which report abnormal vocal characteristics [6, 42], we found that the child did not demonstrate hypernasality and hyponasality for his vocal performance.

This case report also demonstrated poor language skills among Vietnamese-speaking children with Prader-Willi Syndrome. All scores on language assessment tasks were below the mean scores of Vietnamese-speaking children with DLD in previous studies [33, 35]. Specifically, the child did not repeat correctly any item that was an 8-syllable sentence on the Sentence Repetition. He got very low scores on phonological awareness skills, rapid naming skills, nonword repetition, and story comprehension (see **Table 2**). These skills may influence his reading skills in the future [34]. He produced shorter MLU compared to the typically developing and DLD groups [33].

Children with Prader-Willi Syndrome may have problems in cognition, which are considered as the main factors influencing speech and language skills [16]. The Vietnamese child with Prader-Willi Syndrome scored average IQ scores at 82 from the PTONI test (the standard score was 75). However, he showed poor auditory short-term memory, temporal order procession and poor auditory verbal processing skills in narrative tasks as he could retell two details and answered two out of ten questions related to the story.

For the direct assessment tasks, the child was able to complete all the tasks. However, it took him a longer time to complete compared to same-age typically developing peers due to the child's low compliance during the direct assessment sessions. For example, the OMA task actually took about 5-10 minutes to complete with typically developing children [31] but it took him in total of about 30 minutes. The child understood how to perform the task but he sometimes kept silent or did not do the task by the verbal rejection "don't want to do". The assessment team broke the assessment sessions into many parts and were flexible with the order of the testing items wherever possible. They also praised him constantly after each attempt. Another example of the length

Table 2. Measures for language skills of a Vietnamese child with Prader-Willi Syndrome in comparison to typically developing and developmental language disorder Vietnamese children.

Measures	PWS Participant (Aged 5;11)	DLD	TD
Picture Identification (max = 60)	40	46.9 (*SD*=6.2) [a]	54.6 (*SD*=2.1) [a] 53.5 (*SD*=2.8) [b]
Picture Naming (max=60)	31	35.1 (*SD*=6.2) [a]	49.6 (*SD*=3.8) [a] 47.7 (*SD*=4.7) [b]
Rapid Naming (RAN)	*M*=0.41 items/s	-	*M*=0.86 (*SD*=0.3) [b]
Sentence Repetition	0%	*M*=56% [b]	*M*=82% [b]
Nonword Repetition	20%	22% (*n*=10) [c]	*M*=38% [c]
PhAMV	2 out of 36	-	12.1/36 (*SD*=6.0) [c]
Story Comprehension	2 out of 10	-	-
Story Structure (max=17)	2 out of 17	3.6 (*SD*=2.2) [a]	9.5 (*SD*=1.9) [a]
MLU	4.5	4.8 (*SD*=2.2) [a]	7.5 (1.4) [a]
Gram	30%	49% (*SD*=33%) [a]	88% (*SD*=11) [a]
TTR	0.3	-	-

Note. PWS = Prader-Willi Syndrome; DLD = Developmental Language Disorder; TD=typical development; M=mean; SD=Standard deviation; [a] Pham et al. [33] sampled from two groups of children aged 5;8; [b] Pham and Snow [34] sampled on 94 typically developing children and 10 DLD children, mean age 5;8; [c] Pham and Ebert [35] sampled on typically developing children (n=94) and DLD children (n=10) aged 5–6 years old. All measures in language skills showed that the participants had lower scores in all tasks when compared with the scores of those who were typically developing and even lower than those who were with developmental language disorder [33–35].

of time it takes when testing a Vietnamese child with Prader-Willi Syndrome was during the speech test. Normally, a speech test takes about 10 minutes to be administered by the VSA [27, 31]; however, it took 45 minutes to complete the VSA with the child. It was divided into 5 intervals as the child required to have small breaks during the speech testing administration. Normally, it takes about two to three hours to complete speech tasks (OMA, VSA, SPAAC) and language tasks (Vocabulary Receptive, Vocabulary Expression, Sentence repetition, Rapid Naming, Nonword repetition, Phonological Awareness Measure, MAIN stories) for children with typical development and language disorders. However, it took 5 visits with the child to complete these testing tasks.

5. Conclusions

In conclusion, people with Prader-Willi Syndrome require different services including speech and language support from infancy to adulthood [6]. It is relevant for professionals to be aware of the speech and language characteristics of individuals with Prader-Willi Syndrome. This case report involved an individual child, which may be insufficient to represent Vietnamese-speaking individuals with Prader-Willi Syndrome. Given that the speech and language profession is growing in Vietnam [41], further research is needed to investigate speech and language characteristics from a larger sample size or at different ages to distinguish differences in speech and language characteristics associated with different types of genetic abnormality. Further research is also needed to investigate the effectiveness of speech and language services and interventions provided for individuals with Prader-Willi Syndrome in the Vietnamese language.

Author Contributions

Y.N. made the following contributions to the paper: conceptualisation of the paper; review and interpretation of the literature; design of the research questions; and writing, editing and revision of the manuscript. B.P. made the following contributions to the paper: Conceptualisation of the paper; Collection and analysis of data; writing, editing and revision of the manuscript; and writing cover letters and submitting the manuscript to the journal. H.P., V.P., T.D., P.N. and T.N. have made the following contributions to the paper: Collection and analysis of data and writing, editing and revising of the manuscript.

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Institutional Review Board Statement

This case report was part of our research project managed by the Institute of Clinical Psychology Vietnam under

the grant number NCUD.05-2019.67. Ethical approval for the research project was obtained from the Institute board (number: 2021/05, dated 5 March 2021).

Informed Consent Statement

Written consent forms were obtained from the child's parents for volunteering involvement to provide information about their child. They also permitted for their child to attend all assessment sessions required. The child's parents allowed the assessment team to use their child's assessment information for any forms of publications including this case report. We also asked permission from the child participant by obtaining his assent to participate after hearing a child-friendly description of the research.

Data Availability Statement

This is a case report study; therefore, data collected for a child who was diagnosed with Prader-Willi Syndrome. Data that were collected included: (1) medical and developmental history; (2) reports from parents for the Intelligibility in Context Scale: Vietnamese (ICS-VN) and the Focus on Communication Under Six: Vietnamese (FOCUS-34); (3) all score forms of direct assessment sessions: Oral Motor Assessment (OMA), Vietnamese Speech Assessment (VSA), Speech Participation and Activity Assessment of Children Vietnamese (SPAA-C), Receptive Vocabulary, Expressive Vocabulary, Rapid Naming (RAN), Phonological Awareness Measures: Vietnamese (PhAMV), Sentence Repetition, Nonword Repetition, Cat Retell Story, Baby Goat Tell Story, and Voice Rating Scale. These forms were in printed formats filling by handwriting. Filling forms were scanned and stored in external hard disc drive for the research group only and were publicly unavailable due to privacy. However, the instruments used in the study are mostly available online so that other professionals can access to.

- ICS-VN: https://cdn.csu.edu.au/__data/assets/pdf_file/0004/2175988/ICS-Vietnamese-English-Final-Nov-2015.pdf.
- FOCUS-34-Vietnamese: https://bdc.sdsu.edu/wp-content/uploads/2019/04/FOCUS-34_Clinician_Final-Vietnamese-version.pdf.
- SPAA-C-Vietnamese: https://cdn.csu.edu.au/__data/assets/pdf_file/0011/2755406/SPAA-C-Child-Vietnamese-English.pdf.
- Language tasks for (1) Picture Identification, (2) Picture Naming, (3) Rapid Naming (RAN), (4) Sentence Repetition, (5) Nonword Repetition (NWR), and (6) Phonological Awareness Measure in Vietnamese (PhAM-V) available at: https://vietslp.sdsu.edu/assessment-tools/.
- Cat Retell Story and Baby Goat Tell Story, as part of the Multilingual Assessment Instrument for Narratives (MAIN), Vietnamese version are available at: https://www.leibniz-zas.de/fileadmin/media/Dokumente/ZASPiL/ZASPiL_63/MAIN_Vietnamese_2020_Revised.pdf.

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Conflict of Interest

The authors declare that no competing interests existed at the time of publication.

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