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The Prosodic Profile Of Individuals With Prader-Willi Syndrome

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THE PROSODIC PROFILE OF INDIVIDUALS WITH PRADER-WILLI SYNDROME

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ABSTRACT

Prosody is an important component of effective communication, playing a major role in language comprehension and expression. Despite its importance, little research has examined prosody in individuals with Prader-Willi syndrome (PWS), a population that struggles with communication. Therefore, the purpose of the present study was to compare prosodic skills in individuals with PWS to individuals with mixed-etiology intellectual and developmental disability (IDD) as well as determine patterns of prosodic strengths and weakness among individuals with PWS. Adolescents and adults with PWS (*n* = 9) were matched to adults with mixed-etiology IDD (*n* = 9) on nonverbal ability. Participants completed standardized assessments measuring IQ (Kaufman Brief Intelligence Test – 2nd edition), receptive vocabulary (Peabody Picture Vocabulary Test – 4 edition), and prosody (Profiling Elements of Prosody in Speech-Communication; PEPS-C).

Adolescents and adults with PWS performed better than adults with mixed-etiology IDD on the majority of the prosody subtasks. In addition, individuals with PWS demonstrated better prosody comprehension on word-level tasks versus phrase-level tasks. However, the opposite was true for phrase-level tasks; participants with PWS exhibited better prosody expression on phrase-level tasks versus word-level tasks. As the first study to examine prosody in PWS, these results provide foundational information for future research. Further, by identifying prosodic weaknesses common in PWS, the results will have important implications for speech and language therapy outcomes in this population.

Keywords: prosody, Prader-Willi syndrome, intellectual and developmental disability

DEDICATION

This thesis is dedicated to Windsor “Win” Drewry. May all your dreams come true.

LIST OF ABBREVIATIONS

GSV	Growth Score Value
IDD	Intellectual and Developmental Disability
IQ	Intelligence Quotient
KBIT-2	Kaufman Brief Intelligence Test, 2^{nd} Edition
PEPS-C	Profiling Elements of Prosody in Speech-Communication
PPVT-4	Peabody Picture Vocabulary Test, 4^{th} Edition
PWS	Prader-Willi Syndrome
SLP	Speech-Language Pathologist
TD	Typically Developing

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TABLE OF CONTENTS

LIST OF TABLES

I. INTRODUCTION

Approximately 65% of individuals with intellectual and developmental disabilities (IDD) struggle with speech and language (Brown & Percy, 2007), with deficits appearing in both spoken and written language and across all domains of language (i.e., phonology, morphology, syntax, semantics, and pragmatics; American Speech-Language-Hearing Association (ASHA), n.d.). Prosody, the rhythm and melody of speech, is an important component of effective communication. While prosody is typically categorized as an element of phonology, it also plays an important role in pragmatics and syntax (Gerken & McGregor, 1998). More specifically, prosody aids speakers in communicating both efficiently and appropriately by enhancing or changing the meaning of spoken utterances, segmenting speech, informing syntactic structure, emphasizing important information, and conveying emotional and mental states (Peppé et al., 2006). In contrast, impaired prosody can lead to breakdowns in spoken language comprehension, lower intelligibility ratings, and negative social consequences (Lewis et al., 2002; Skwerer et al., 2007). Despite its important role in communication, little research has examined prosodic abilities within or across different etiologies of IDD. Further, no research has examined prosody in Prader-Willi syndrome (PWS), a rare genetic etiology of IDD known to impact appetite, growth, metabolism, behavior, cognitive function, and communication (Lewis et al., 2002; PWS Association – USA, 2016). Thus, the purpose of the present study was to examine prosody within PWS.

Prosody

Prosodic Form and Function

Prosody is typically described in terms of form and function. Form refers to the auditory and perceptual characteristics of speech, while function refers to the pragmatic and linguistic meaning of an utterance (Järvinen-Palsey et al., 2008).

Prosodic Form. Prosodic form includes three acoustic cues: fundamental frequency (i.e., the acoustic correlate of pitch), intensity (i.e., the acoustic correlate of loudness), and duration (Järvinen-Pasley et al., 2008). Fundamental frequency refers to the rate of vocal fold vibration (Baker et al., 2008), and is perceived by listeners as changes in pitch (Baken & Orlikoff, 2000). Pitch is measured in hertz and can be characterized by pitch-height and pitch-range. Pitch-height refers to the highest fundamental frequency produced in an utterance. For example, stressed syllables tend to possess greater pitch heights than unstressed syllables (Campbell & Beckman, 1997). Pitch-range refers to the difference between the maximum and minimum fundamental frequencies in an utterance (Nadig & Shaw, 2011). For example, sad speech is conveyed using a small pitch range while happy speech is conveyed with a wider pitch range (Mozziconacci, 1998). Intensity refers to the level of speaking volume and is measured in decibels. Duration refers to the length of sound and is typically measured in milliseconds across syllables or speech segments (Peppé, 2009).

Prosodic Function. The cues conveyed through prosodic form combine to achieve several functions that facilitate the overall understanding of spoken utterances (Crystal, 1971; Gibbon & Smyth, 2001). More specifically, prosodic functions serve both linguistic (i.e., grammatical and pragmatic) and paralinguistic (i.e., index and affective) functions.

Grammatical functions of prosody are the verbal representations of written punctuation (Peppé, 2009). One important grammatical function is distinguishing the points where clauses, phrases, and utterances begin and end. This can be achieved by inserting pauses at a boundary (Boomer, 1965; Goldman-Eisler, 1972; Grosjean & Deschamps, 1975; Scott, 1982), lengthening the final syllable of a phrase (Copper et al., 1978; Huggins, 1974; Klatt, 1975; Lindblom & Rapp, 1973), implementing specific pitch movements (e.g., fall-rise intonation), or decreasing amplitude (Streeter, 1978). For example, these cues can be used to distinguish the phrase "coffee, cake, and jam" from "coffee-cake and jam" (Peppé, 2009). Grammatical functions also distinguish between types of utterances. In general, questions are conveyed using a rising pitch, while statements are conveyed using a falling pitch (Lieberman, 1967), e.g., "I will see you tomorrow?" versus "I will see you tomorrow." Finally, grammatical functions are also used to distinguish between word classes (e.g., nouns vs. verbs) by using variations in stress patterns. In general, bisyllabic nouns are often stressed on the first syllable while bisyllabic verbs are often stressed on the second syllable (Kelly & Block, 1988; Sereno, 1986), e.g., "*im*print versus im*print"* (Peppé et al., 2006).

In contrast, pragmatic functions of prosody aid the speaker in emphasizing important words or syllables in an utterance through boosted pitch, increased length, and/or increased loudness (Crystal, 1969; Fry, 1958; Laver, 1994). In general, an utterance with a broad focus refers to neutral utterances in which no emphasis is added to any particular part of the utterance (Peppé et al., 2009), e.g., "He asked for coffee." An utterance with a narrow focus contains an accent placed on an important word or syllable, e.g., "No, he asked for *tea.*"

Paralinguistic functions add circumstantial information to utterances and include both index and affect. Index refers to an individual's unique speaking characteristics (e.g., speaking pitch, intonation patterns, and regional dialect). Affect allows the speaker to indicate their feelings and attitudes towards the spoken content as well as convey their likes and dislikes. Affect can be conveyed through changes in rate, pitch-height, pitch-range, and intensity (Mozziconacci, 1998, Banse & Scherer, 1996). In general, positive affect is marked by a wider and higher pitch range, whereas negative affect is marked by a narrow, lower pitch range (Banse & Scherer, 1996).

Measuring Prosody

Prosodic form and function are measured using both input and output tasks. Input tasks measure perception and comprehension (i.e., receptive ability) whereas output tasks measure speech generation and production (i.e., expressive ability). Measures of prosodic form often assess lower level phonetic processing, such as discriminating between prosodic variations in utterances or imitating prosodic cues. In contrast, measures of prosodic function may involve tasks that require higher level processing, such as expressing and understanding emotions and attitudes, dividing utterances into syntactic/linguistic units, using and distinguishing between types of closure, and assigning stress. The Profiling Elements of Prosody in Speech-Communication (PEPS-C) is a frequently used assessment for measuring prosody. The test measures prosody skills, including both receptive and expressive form and function, to identify prosodic strengths and weaknesses among individuals (Gibbon & Smyth, 2013; Peppé, 2015).

Prosodic Development

Prosody plays a critical role in language development. In fact, prosodic cues are among the first aspects of speech accessed prenatally (Gervain, 2018), and there is a strong relationship between children's intonation comprehension and future receptive and expressive language development (Wells et al., 2004).

Receptively, fetuses use the speech they hear in utero to develop the perceptual abilities and brain specialization needed for language acquisition, including information related to the lexicon and grammar system of their native language (Gervain, 2018). After birth, at approximately two to three months, infants become aware of the prosodic contrasts (e.g., pitch variation) directed towards them in adult utterances (Crystal, 1979). Infants as young as six months utilize “prosodic bootstrapping” in order to divide the utterances they hear into meaningful units (Gerken & McGregor, 1988) as well as to segment speech into words, phrases, and clauses. This allows them to learn the syntactic and semantic features that are necessary for language development (Morgan & Demuth, 1996). Prosody also influences reading development, including decoding speed, word-reading accuracy, and reading comprehension (Schwanenflugel et al., 2004).

Expressively, infant vocalizations allow children to express attitudes such as pleasure and recognition. At approximately six months of age, infants’ non-segmental features (e.g., pitch and intensity control; Kent et al., 1994) begin to resemble the prosodic patterns of the language they are learning. Infants also begin to configure their pitch, rhythm, and pauses (Crystal, 1979). Consequently, the typical errors produced in children’s early word production (e.g., weak syllable deletion) may be related to their misinterpretation of prosodic cues (Gerken & McGregor, 1988). While prosody is important for language development, it is not typically mastered until approximately 12 to 13 years of age (Wells & Peppé, 2003).

Prosodic Deficits

Prosodic deficits are one of the earliest and most prominent indicators of decreased communication and social skills detected by unfamiliar listeners (Paul et al., 2005b). Furthermore, prosodic deficits persist and show little development over time, even when other aspects of language, such as vocabulary and sentence structure, begin to improve (DeMyer et al., 1973; Kanner, 1971; Rutter & Lockyer, 1967; Simmons & Baltaxe, 1975).

Receptively, impaired prosody can lead to breakdowns in spoken language comprehension, impaired theory of mind (Baron-Cohen et al., 1985), and difficulty orienting to conversations (Peppé, 2006). In addition, individuals with receptive prosodic impairments may have difficulty understanding metaphors, as these individuals often utilize literal interpretations of language (Tager-Flusberg, 1999). They may also have difficulty recognizing the mental and emotional states of others, especially if they are different from their own (Peppé et al., 2006). Whether one demonstrates a receptive or expressive impairment, deficits in prosody can lead to negative social consequences such as poor social integration and limited participation in vocational, recreational, and learning activities (Lewis et al., 2002; Lewis et al., 2004; Paul et al., 2005b; Skwerer et al., 2007).

Expressively, a prosodic impairment may lead to overall lower speech intelligibility (Monsen, 1983). Furthermore, an individual with monotonous pitch may have difficulty conveying phrasing and emphasis, while an individual with an exaggerated pitch may be misinterpreted as patronizing or insincere (Peppé et al., 2006). Prosodic deficits can also decrease one's ability to convey desired intentions, which may lead to further social isolation (DePape et al., 2012; Järvinen-Palsey et al., 2008, Lord et al., 1999; Paul et al., 2005b). These impairments may contribute to a listener's impression of social oddness towards the speaker (Van Bourgondien & Woods, 1992). For example, a listener may develop an impression of an "overbearing insistence" when communicating with an individual who speaks with a high pitch (Shriberg & Widder, 1990).

Prader-Willi Syndrome

Prader-Willi syndrome (PWS) is a rare (i.e., occurs in approximately one in every 15,000 births), genetic neurodevelopmental disorder that results from a paternal abnormality of chromosome 15 (Cassidy et al., 2012). Prader-Willi syndrome can be diagnosed as early as the first month of life through genetic testing and can occur in one of three ways: 1) PWS by Deletion, 2) PWS by Uniparental Disomy, or 3) PWS by Imprinting Mutation (Cassidy et al., 2012). Deletion, the most common form of PWS, occurs when a segment of chromosome 15 is deleted. Uniparental Disomy occurs when an individual inherits two copies of chromosome 15. Imprinting Mutation, the least common cause of PWS, occurs when chromosome 15 is present but inactive.

Prader-Willi syndrome is characterized by behavioral disabilities, mild to moderate intellectual disability, and facial abnormalities (Dimitropoulos et al., 2013). Behaviorally, individuals with PWS often show an excessive interest in food, skin picking, resistance to changes in routine, temper tantrums, obsessive and compulsive behaviors, mood fluctuations, and difficulties with socialization (Holland et al., 2003). In contrast, individuals with PWS demonstrate a relative strength in daily living skills (Holland et al., 2003) and are often described as good natured, affectionate, friendly, placid, and cheerful (Curfs & Fryns, 1992; Greenswag, 1987).

Cognitively, individuals with PWS demonstrate IQs that typically fall between 50 and 85 with a mean IQ of 65 - 70 (Debladis et al., 2019; Further Inform Neurogenetic Disorders [FIND], n.d.). Individuals with PWS also typically present with learning challenges and poor working memory when completing tasks that require simultaneous use of multiple cognitive functions (Curfs et al. 1991; Curfs & Fryns, 1992). In addition, individuals with PWS often display relative strengths in long-term memory but weaknesses in short-term memory (Hochhalter & Joseph, 2001; Stauder et al., 2005; Warren & Hunt, 1981; Whittington et al., 2004).

Regarding speech, individuals with PWS are likely to display a unique set of orofacial complications that may lead to reduced articulatory skills. These include a small mouth, narrow overjet, and narrow palatal arch (Lewis et al., 2002). In addition, individuals with PWS may experience altered larynx growth due to endocrine dysfunction, which may, in turn, lead to increased pitch variations (Lewis et al., 2002). Furthermore, hypotonia of the orofacial structures may lead to hyper/hypo nasality, variations in vocal quality, a slow rate of speech, and poor velopharyngeal movement (Lewis et al., 2002). Additional speech difficulties often include sound distortions, omissions, and vowel errors (Stein et al., 2006).

Within the language domain, the limited available research indicates that individuals with PWS also often demonstrate several language deficits when compared to typically developing (TD) peers, with notably more impairments in expressive versus receptive language (Stein et al., 2006). Difficulties in language form (i.e., phonology, morphology, and syntax) often include low mean length of utterances and poor reading comprehension skills (Lewis et al., 2002), while issues with language content (i.e., semantics) often include small vocabularies (Lewis et al., 2002). Individuals with PWS are also likely to exhibit impairments in language use (i.e., pragmatics) (Debladis et al., 2000; Dimitropoulos & Schultz, 2007; Lewis, et al., 2002). For example, individuals with PWS may struggle to recognize facial expressions and social intent when compared to TD peers of similar age (Tager-Flusberg et al., 1998). In addition, individuals with PWS may struggle with skills related to theory of mind, social ability and interactions, understanding emotions, and developing and maintaining peer relationships (Dimitropoulus & Schultz, 2007; Holland et al., 2003; Klin, 2000). These difficulties in social functioning are also observed when individuals with PWS are matched to TD peers on age and ethnicity. (Dimitropoulos et al., 2019).

Despite many difficulties with speech and language relative to typical development, individuals with PWS often outperform other etiologies of IDD on tasks related to some areas of pragmatics. For example, when matched on IQ and receptive and expressive language, individuals with PWS have performed better on tasks related to social functioning than individuals with autism spectrum disorder (Tager-Flusber & Sullivan, 2000). When matched on the same variables, individuals with PWS also demonstrated greater theory of mind skills when compared to individuals with Williams syndrome (Tager-Flushber & Sullivan, 2000). However, these results may reflect patterns of weakness in autism spectrum disorder and Williams syndrome, respectively, more so than strengths in PWS, and more research is needed to fully capture the linguistic profile of PWS.

Despite some notable difficulties with communication, no research has examined prosody in PWS. However, understanding the prosodic profile of PWS will provide a more detailed picture of their communication abilities. It will also identify possible underlying difficulties contributing to pragmatic challenges and identify targets for intervention.

Prosody in IDD

There is a small research base on prosody in other etiologies of IDD. However, this research is limited and has mostly focused on autism spectrum disorder, Williams syndrome, and Down syndrome. This research indicates prosodic difficulties are common in IDD but also that there are unique patterns of strength and weakness across different etiologies.

Individuals with autism spectrum disorder are described as having atypical expressive prosody, with particular difficulty using accents to indicate focus (Kanner, 1943). In addition, individuals with autism spectrum disorder often demonstrate atypical prosodic characteristics such as “sing-song” speech patterns (i.e., wide pitch range), poor volume control, unnatural stress patterns, and difficulty expressing and understanding emotions relative to their same-age, TD peers (Globerson et al., 2014; Grossman et al., 2010; Jarvinen-Pasley et al., 2008; Paul et al., 2005a; Peppé et al., 2011; Nadig & Shaw, 2012). These prosodic difficulties may lead to poor social integration and acceptance (Paul et al., 2005a). However, research examining prosody in autism spectrum disorder has yielded mixed results, and some research has indicated that individuals with autism with verbal IQs within normal limits demonstrate normal awareness and use of prosodic cues (Paul et al., 2005b).

Individuals with Williams syndrome, in turn, have difficulties with both expressive and receptive prosody. When matched on chronological age to TD peers, children with Williams syndrome often show a delayed onset in their ability to understand and use prosody to indicate the most important word in an utterance (i.e., focus), draw attention to certain words/syllables in an utterance (i.e., contrastive stress), segment complex noun phrases (i.e., boundary), and regulate conversations (i.e., turn-end) (Stojanovik, 2010). Individuals with Williams syndrome also utilize both a larger pitch range as well as a higher overall speaking pitch compared to both language-matched and age-matched TD peers (Setter et al., 2007). This results in individuals with Williams syndrome being perceived as more emotionally involved than TD speakers (Setter et al., 2007). However, when matched on mental age, the differences in pitch are much less pronounced (Stojanovik, 2010).

Finally, individuals with Down syndrome also struggle with aspects of prosody when matched to TD peers on chronological age, including the skills of affect, boundary, contrastive stress, and turn-end (Stojanovik, 2011). Difficulties with expressive turn-end are also seen relative to TD peers matched on developmental age and vocabulary (Zampini et al., 2016). In addition, individuals with Down syndrome, when matched to TD peers on receptive language and nonverbal abilities, demonstrate increased difficulty using prosody to express emotion and to indicate the most important word in an utterance (Stojanovik, 2011).

Current Study

Despite its important role in communication, little research has examined prosodic abilities within or across different etiologies of IDD. The research that is available has primarily focused on individuals with autism spectrum disorder, with a few studies on Williams syndrome and Down syndrome (e.g., Nadig & Shaw, 2012; Stojanovik, 2010; Stojanovik, 2011). Given the pattern of speech and language difficulties documented in PWS, it is likely that these individuals also struggle with some aspects of prosody, including indicating focus (i.e., contrastive stress), regulating conversations (i.e., turn-end), and expressing and understanding emotions (i.e., affect). However, given the unique behavioral, cognitive, and linguistic profile of PWS, it is also likely that individuals with PWS possess a unique prosodic profile. Identifying areas of difficulty can provide therapy targets that could be used to improve intelligibility, comprehensibility, and social interactions among individuals with PWS. It is also possible that individuals with PWS have some areas of relative prosodic strength that could be leveraged to improve overall communication abilities. Thus, the purpose of the present study is to examine the prosodic profile of individuals with PWS. Specifically, the present study assessed the following research questions:

a) How do adolescents and adults with PWS compare to adults with mixed-etiology IDD on measures of expressive and receptive prosody?

Based on previous research indicating that individuals with PWS demonstrate language strengths compared to individuals with different etiologies of IDD, we hypothesize that adolescents and adults with PWS will have greater expressive and receptive prosody skills than adults with mixed-etiology IDD.

b) Are there patterns of prosodic strength and weakness among adolescents and adults with PWS?

Based on prior research that indicates individuals with PWS have stronger receptive than expressive language skills, we hypothesize that adolescents and adults with PWS will perform better on measures of receptive prosody than expressive prosody.

II. METHODS

Design

This study was conducted utilizing a mixed-group design. The between-group component compared a group with PWS to a group with mixed-etiology IDD, matched on nonverbal ability, on measures of expressive and receptive prosody. The within-group component compared expressive and receptive prosodic abilities within the group with PWS.

Participants

PWS

Participants for the present study included adolescents and adults with PWS (n = 9; 7 males, 2 females; all Caucasian Americans). Five participants with PWS came from a larger study examining language in IDD. These participants were recruited through North Mississippi Regional Center (NMRC). Additional participants with PWS (n = 4) were recruited specifically for the present study and were recruited through the University of Alabama Intellectual Disabilities Participant Registry, social media, and word of mouth.

IDD

Participants with PWS were matched to a comparison group of participants with mixed-etiology IDD (n = 9; 4 males, 5 females; 8 Caucasian Americans and 1 African American) on nonverbal ability via the Kaufman Brief Intelligence Test, 2nd ed. (KBIT-2; Kaufman & Kaufman, 2004). All participants with mixed-etiology IDD came from the larger study examining language in IDD and were selected from that sample ($n = 31$) on a 1:1 basis to serve as matches for the participants with PWS.

Inclusion Criteria

Inclusion criteria required participants to be adolescents or adults diagnosed with PWS or another etiology of IDD (e.g., autism spectrum disorder, Down syndrome, intellectual disability), use speech as their primary method of communication, and have the comprehension skills needed to understand the tasks. This included the ability to sustain attention and follow multi-step directions. Each participant's caregiver confirmed that they met the inclusion criteria.

Participant Matching

Participants were matched using nonverbal standard scores from the KBIT-2. First, nonverbal standard scores were calculated for each of the nine participants with PWS. Next, a participant with mixed-etiology IDD was selected as a match for each participant with PWS. To be considered a match, each participant with IDD who was selected had to be within 10 points of the target participant with PWS. This created nine pairs of participants matched on nonverbal standard scores. Nonverbal ability was selected as the matching variable because it typically remains less impacted in individuals with IDD in comparison to other abilities, such as expressive language (Phillips et al., 2014). Nonverbal ability also allowed for the closest match between participants, given the profile of participants that had already been collected for the larger study.

An independent samples *t*-test was used to confirm that there were no significant differences between groups on nonverbal ability using the nonverbal standard scores on the KBIT-2, t (16) = .18, $p = 0.86$. This effect was small, Cohen's $d = .09$. We also confirmed that there were no statistically significant differences between groups on nonverbal raw scores on the KBIT-2, t (16) = 1.23, p = 0.24.

See Table 1 for participant demographics and scores on the matching variable (i.e., nonverbal standard scores).

Table 1

Descriptive information and matching variable.

	PWS			IDD		
	Mean	Range	SD	Mean	Range	SD
Age	31.11	17 - 51	11.55	47.44	29 - 69	15.12
KBIT-2 IQ	60.22	43 – 88	14.28	54.44	42-77	9.96
KBIT Nonverbal Standard Scores	58.33	40 - 84	14.61	57.22	42 - 75	10.93
KBIT Matrices Raw Scores	18.80	11 – 29	5.30	16.20	13 – 24	3.40
PPVT-4 Vocabulary Standard Scores	73.11	55 – 101	14.07	60.56	45 – 80	11.18
PPVT-IV Vocabulary GSV	185.22	162 - 225	20.04	158.78	131 - 200	23.57

Note. Age = Chronological age; IQ = KBIT-2 intelligence quotient composite; Nonverbal standard score = KBIT-2 matrices raw score; Vocabulary GSV = PPVT-4 vocabulary growth scale value and standard scores.

Procedure

This study was approved by the University of Mississippi Institutional Review Board (IRB). Before administering assessments, participants' caregivers provided written consent, and the participants themselves provided verbal assent. Testing for the larger study took place at the NMRC main campus or a NMRC community home. Testing for the additional participants with PWS, recruited specifically for the present study, took place in their homes. Participants from the larger study received a completion certificate and new video games to share at the NMRC recreation center. Additional participants with PWS received a $10 Amazon gift card for completing the study. All participants completed three assessments in the following order: 1) KBIT-2, 2) Peabody Picture Vocabulary Test, 4^{th} ed. (PPVT-4; Dunn & Dunn, 2007), and 3) the Profiling Elements of Prosody in Speech-Communication (PEPS-C; Peppé, 2015). Testing at NMRC was completed in two to three sessions while testing for the additional participants with PWS was completed in a single session. To combat potential fatigue, participants were allowed to take breaks between each of the three tasks as well as between each of the PEPS-C subtasks.

Measures

IQ, Nonverbal Ability, and Verbal Ability

The KBIT-2 (30 minutes) was used to assess verbal and nonverbal cognitive abilities and overall IQ. The KBIT-2 is normed for children and adults from ages 4;0 to 90;11. It is scored objectively as participants respond to questions using one-word responses or through pointing.

The KBIT-2 includes three subtests. The Verbal Knowledge and Riddles subtests combine to provide a verbal standard score, and the Matrices subtest provides a nonverbal standard score. Verbal and nonverbal standard scores are then combined to calculate an overall IQ composite for each participant, which was used to describe this study's sample.

The KBIT-2 demonstrates good test-retest reliability on the verbal scale (*r* = .91), nonverbal scale (*r* = .83), and IQ composite (*r* = .90) across children, adolescents, and adults. This assessment also demonstrates good concurrent validity, correlating with the Wechsler Adult Intelligence Scales, 3rd ed. on the verbal scale at *r* = .82, nonverbal scale at *r* = .83, and IQ composite at *r* = .89 (Kaufman & Kaufman, 2004; Wechsler, 1997).

Receptive Vocabulary

The PPVT-4 (30 minutes) was used to assess receptive vocabulary. The PPVT-4 is normed for children and adults from ages 2;6 to 90;11. The test requires participants to point to a picture that matches the meaning of a word spoken by the experimenter. The PPVT-4 provides raw, standard, and growth score values (GSV; raw scores weighted for item difficulty). The present study used GSV and standard scores to describe the study's sample.

The PPVT-4 demonstrates good test-retest reliability (*r* = .87 - .93) and good validity (correlates with Clinical Evaluation of Language Fundamentals, 4th ed. at *r* = .67 - .75; Pearson, 2019).

Prosody

The PEPS-C (1-2 hours) was used to assess receptive and expressive prosody. The PEPS-C is a computerized task that targets both prosodic form and function via paired expressive and receptive tasks. Six paired tasks (one receptive, one expressive per pair) assess prosodic function, four of which were included in the present study (i.e., turn-end, affect, boundary, contrastive stress). An additional paired task (i.e., discrimination/imitation) was used to assess prosodic form. Thus, across receptive and expressive form and function subtasks, a total of 5 paired tasks (i.e., 10 subtasks) were administered. Each subtask yields a percentage correct score out of 16 items (or less if a test item was not scorable). To combat order effects, the PEPS-C was administered in two different orders that were alternated between participants. The remaining two tasks (i.e., lexical stress and phrase stress) were deemed too difficult by the research team and were not included in the present study. Table 2 outlines each of the administered PEPS-C subtasks.

Table 2

Description of PEPS-C Subtasks (Peppé, 2015).

Task	Prosodic Target	Purpose	Example	
			Receptive	Expressive
Turn-End	Grammatical/ Pragmatic Function	Identify or express utterances as questions vs. statements	Indicate if the word “carrots” was used as a question or statement	Produce the word “carrots” as a question or statement based on the visual cue provided by the computer
Affect	Affect Function	Identify or express likes vs. dislikes	Indicate if the computer likes or dislikes cheese based on how it was said	Use prosody to say the word “cheese” to indicate whether they like it or not
Boundary	Grammatical Function	Understand or use prosody for chunking speech into syntactic units	Identify a picture that depicts “fruit, salad, and milk” as opposed to “fruit-salad and milk”	Produce the correct list of foods shown on the screen
Contrastive Stress	Pragmatic Function	Identify and express emphasis	Indicate which color socks the computer forgot to buy based on which color was stressed, e.g., “I wanted *blue* and black socks.”	Use stress to correct the computer about what color/animal has the ball in a soccer game. e.g., “No, the green *cow* has the ball.”

(table continues)

Table 2 *(continued)*.

Task	Prosodic Target	Purpose	Example	
			Receptive	Expressive
Discrimination /Imitation	Prosodic Form	Identify if utterances are the same or different (discrimination) or repeat utterances (imitation) said by the computer, exactly how the computer said it	Indicate if a muffled version of the phrase “I saw a blue bird today” sounded the same or different as the muffled phrase “I saw a blue-bird today”	Repeat the word “intern” using the same prosody that was used by the computer

Scoring and Reliability. Receptive subtasks on the PEPS-C required the participant to point to a picture on the computer that corresponded with their answer. The tester then entered the participant's response by clicking on the chosen picture. These responses were scored automatically once the tester clicked on the participant's pointed response. Expressive subtasks can also be scored in real time. However, the research team decided to score these subtasks after the testing session using stored audio files. This was done in an effort to increase scoring accuracy and to allow for reliability checks. Expressive subtasks were scored based on the researcher's perception of various prosodic cues (e.g., presence of rising intonation to indicate a question).

To ensure reliability, each expressive subtask was scored by two independent scorers (i.e., the primary researcher and a research assistant). Consensus coding was then used to discuss and resolve any discrepancies between scores. This resulted in a single, agreed upon score for each expressive item (see Bradley et al., 2007). When reviewing discrepancies during consensus coding, the raters' initial scores were hidden, making them blind to their initial score. This was done to minimize any bias towards the original score.

Analytic Plan

For research question one, a series of paired-samples *t*-tests were used to compare groups on each of the ten PEPS-C subtasks. Paired-samples *t*-tests were selected instead of independent samples *t*-tests because the participants were individually matched on nonverbal ability. Some of the dependent variables were not normally distributed (e.g., imitation and expressive affect tasks for the group with PWS; expressive and receptive boundary and receptive turn-end for the group with IDD), so these subtasks were also examined using the nonparametric alternative: Wilcoxon

Signed Ranks test. The pattern of results did not change, and thus we chose to report the paired-samples *t*-test results below.

For research question two, a series of paired-samples *t*-tests were used to compare performance within the group with PWS on each of the paired expressive-receptive tasks (e.g. imitation vs. discrimination, expressive turn-end vs. receptive turn-end, etc.). Again, because the imitation and expressive affect subtasks were not normally distributed for this group, we also examined these results using the nonparametric alternative: Wilcoxon Signed Ranks test. Because the pattern of results did not change, we reported the paired samples *t*-test results below.

To control for family-wise error and the possibility of Type 1 errors, we utilized the Holm-Bonferroni method (Holm, 1979). This is a modification of the Bonferroni correction in which the criteria for rejecting the null hypothesis (i.e. the alpha level) is adjusted for each individual comparison (for our purposes, each *t*-test). First, the number of tests (for our purposes, 10 for the between-groups analysis and five for the within-group analysis) is subtracted by the rank number of each pair, plus one. This value is then divided by the target alpha level (.05). Thus, for research question one, which included 10 separate analyses, the alpha level for significance was .005 for the first comparison with the smallest *p*-value, .0056 for the second comparison, .00625 for the third comparison, et cetera, up to .05 for the tenth comparison. For research question two, which included five separate analyses, the alpha level ranged from .01 to .05. To assess significance, obtained *p*-values for each individual *t*-test are ranked from smallest to largest and then compared to the Holm-Bonferroni corrected alphas of increasing stringency. Obtained *p*-values and Holm-Bonferroni corrected alphas used to determine statistical significance are reported in Tables 3 and 4.

Table 3

Between-Groups Comparison of PEPS-C Scores.

Subtask	Mean (SD)		p	Holm-Bonferroni Corrected Alpha	Eta squared
	PWS	IDD			
Imitation	.77 (.18)	.76 (.13)	.82	.025	.003
Turn-End – E	.74 (.18)	.52 (.25)	.02	.005	.36
Affect – E	.70 (.28)	.61 (.22)	.35	.0083	.05
Boundary – E	.79 (.15)	.72 (.12)	.32	.007	.06
Contrastive – E	.79 (.16)	.78 (.18)	.97	.05	.0001
Discrimination	.68 (.11)	.59 (.17)	.28	.00625	.07
Turn-End – R	.77 (.16)	.62 (.22)	.13	.0056	.14
Affect – R	.73 (.18)	.68 (.14)	.42	.01	.04
Boundary – R	.66 (.16)	.61 (.11)	.44	.0125	.04
Contrastive Stress – R	.59 (.18)	.62 (.14)	.69	.0167	.01

Note. E = Expressive subtask; R = Receptive subtask.

Table 4

Within-Groups Comparison of PEPS-C Scores.

Paired Task	Mean (SD)		p	Holm-Bonferroni Corrected Alpha	Eta squared
	E	R			
Imitation/Discrimination	.77 (.12)	.68 (.11)	.12	.0167	.27
Turn-End	.74 (.18)	.77 (.16)	.51	.025	.06
Affect	.70 (.28)	.73 (.18)	.76	.05	.01
Boundary	.79 (.15)	.66 (.16)	.01	.01	.56
Contrastive Stress	.79 (.16)	.59 (.18)	.05	.0125	.40

Note. E = Expressive subtask; R = Receptive subtask.

III. RESULTS

Preliminary Analyses

Scores on each of the PEPS-C subtasks are reported in Tables 3 and 4. In addition, Pearson's *r* was used to assess correlations between each groups' scores on the various PEPS-C subtasks, nonverbal standard scores, and vocabulary GSVs. Within the group with PWS, a significant, positive correlation was found between vocabulary GSV and performance on the imitation subtask ($r = .67$). Interestingly, a significant, negative correlation was found between vocabulary GSV and expressive boundary skills ($r = -.70$). Within the group with IDD, significant, positive correlations were found between vocabulary GSV and expressive ($r = .80$) and receptive ($r = .83$) turn-end, receptive affect ($r = .76$), and expressive boundary ($r = .76$) skills. Table 5 outlines correlations between variables.

Table 5

Correlations between Nonverbal Ability, Vocabulary, and PEPS-C Subtasks (Group with PWS presented above the diagonal; group with IDD presented below the diagonal).

	1	2	3	4	5	6	7	8	9	10	11	12
1. Nonverbal Standard Score	--	.77*	.62	.33	.52	-.66	-.56	.28	.29	.04	-.43	.01
2. Vocabulary GSV	.14	--	.67*	.04	.59	-.70*	.28	.14	-.15	.08	-.41	.05
3. Imitation	-.08	.36	--	.09	.71*	-.58	-.23	.16	-.04	.45	.05	.38
4. Turn-End – E	-.06	.80**	.64	--	-.26	-.37	-.45	.46	.73*	.22	-.28	.50
5. Affect – E	.40	.33	.48	.66	--	-.45	.20	.08	-.05	.34	-.12	.21
6. Boundary – E	.43	.69*	-.16	.50	.38	--	.02	.11	.12	.17	.69*	.02
7. Contrastive Stress – E	-.43	-.47	-.20	-.14	-.03	-.52	--	-.60	-.48	-.26	-.06	-.13
8. Discrimination	-.02	.55	.42	.51	.20	.22	-.36	--	.73*	.74*	.23	.66
9. Turn-End – R	.39	.83**	.56	.73*	.52	.53	-.55	.55	--	.51	.03	.58
10. Affect – R	-.10	.76*	.66	.89*	.40	.35	-.13	.33	.73*	--	.44	.85**
11. Boundary – R	-.24	.52	.60	.51	.19	.14	-.48	.30	.63	.53	--	.41
12. Contrastive Stress – R	.39	.31	.13	.43	.49	.40	-.09	.47	.61	.35	.09	--

**p* < .05, ***p* <.01.

PWS versus IDD

Research question one compared the performance of individuals with PWS to individuals with mixed-etiology IDD on measures of expressive and receptive prosody. Examining group means, the group with PWS appeared to score higher than the group with mixed-etiology IDD on the majority of the PEPS-C subtasks, with the exception of the receptive contrastive stress subtask. However, paired samples *t*-tests indicated that participants with PWS did not perform statistically significantly better than the group with IDD on any subtasks. After the Holm-Bonferroni correction, there was a marginally significant difference between groups on the expressive turn-end subtask, t (df) = 3.08, p = .015, with a large effect, eta squared = .36. In addition, a large effect was found on the receptive turn-end subtask, eta squared = .14. Medium effects were also found on the expressive boundary, eta squared = .06, and discrimination, eta squared = .07, subtasks. In all cases, the group with PWS performed better than the group with IDD.

Strengths and Weaknesses within PWS

Research question two sought to identify patterns of prosodic strengths and weaknesses among adolescents and adults with PWS. On the boundary task, participants with PWS performed better on the expressive versus receptive subtask, though this finding was only marginally significant after the Holm-Bonferroni correction, t (df) = 3.18, p = .013, but with a large effect, eta squared = .56. Participants also performed better on the expressive versus receptive contrastive stress subtask, though again this was only marginally significant, t (df) = 2.29, p = .05, with a large effect, eta squared = .40. A large effect size was also found on imitation versus discrimination subtasks, with participants with PWS performing better on the expressive versus receptive subtask, eta squared = .27. In addition, a medium effect size was found on the turn-end subtask, with participants with PWS performing better on the receptive subtask, eta squared = .06.

IV. DISCUSSION

The purpose of the present study was to examine prosody in PWS. Research question one examined differences between adolescents and adults with PWS and adults with mixed-etiology IDD, matched on nonverbal ability, on measures of receptive and expressive prosody. Research question two examined prosodic strengths and weaknesses, focusing specifically on receptive versus expressive abilities, among individuals with PWS.

Preliminary Analyses

Correlations were used to examine the relationship between both groups' scores on the various PEPS-C subtasks, nonverbal standard scores, and vocabulary GSVs. Interestingly, a significant, negative correlation was found between vocabulary GSV and the expressive boundary subtask within the group with PWS. This did not appear to be driven by any single participant. Furthermore, given the small sample size of the current study, this correlation should be interpreted with caution, and more data is needed to fully understand this relationship.

PWS versus IDD

Research question one examined differences between groups by comparing scores on each of the ten PEPS-C subtasks. While the differences between groups were not statistically significant, the pattern of results, including medium and large effect sizes, indicate that individuals with PWS may possess some greater prosodic skills than individuals with mixed-etiology IDD.

More specifically, a large effect size was found on the turn-end task, suggesting that individuals with PWS may be better at regulating conversations than individuals with IDD, when matched on nonverbal ability. A large effect was also found on the expressive boundary subtask, with individuals with PWS performing better than the group with IDD, indicating a relative strength in segmenting speech to accurately convey intended messages. Lastly, the participants with PWS outperformed, with a large effect, participants with IDD on the discrimination subtask, which indicates that individuals with PWS may be better at distinguishing between various prosodic cues than individuals with other etiologies of IDD. These increased discrimination skills may, in turn, promote stronger receptive prosody skills, such as those needed to distinguish between questions and statements.

Interestingly, both groups demonstrated a higher level of performance on the expressive versus receptive contrastive stress subtasks, with groups performing almost identically on the expressive subtask. This suggests that individuals with IDD, regardless of etiology, may struggle to perceive the important components of an utterance, but are able to successfully convey the important components of their own productions.

The observed relative strengths in prosody in individuals with PWS aligns with previous research indicating that individuals with PWS demonstrate greater skills in some aspects of language when compared to individuals with other etiologies of IDD (Tager-Flusber & Sullivan, 2000). While the differences between groups were not statistically significant, the medium to large effect sizes suggest that the differences between groups may become significant in a larger sample.

Strengths and Weaknesses within PWS

Research question two examined the difference between expressive and receptive prosody skills within adolescents and adults with PWS. The results indicate that participants with PWS performed better on the expressive versus receptive subtask on three of the five tasks. This finding does not align with previous research examining language in PWS that has reported stronger receptive versus expressive language skills (Stein et al., 2006). Again, while these differences were not statistically significant, medium to large effect sizes were found on several of the subtasks, discussed below.

First, the imitation/discrimination task was used to measure participants' ability to understand and express prosodic form. The results of the current study indicate that individuals with PWS are better at imitating prosodic form than perceiving it. However, the difficulty experienced by individuals with PWS on the receptive subtask may have been due to the receptive subtask being more challenging than the expressive subtask. During the receptive subtask, participants were asked to use prosody to discriminate between muffled utterances, whereas on the expressive subtask, participants were asked to repeat words and phrases using the same prosody modeled by the computer. Therefore, the poorer performance on the receptive subtask may reflect that participants struggled to assign prosodic cues to meaningless utterances, but they may, in fact, be able to accurately apply this skill in a meaningful context.

Next, the boundary task measured participants' ability to use prosody to segment utterances. The results suggest that individuals with PWS are better at segmenting spoken utterances during speech than using this skill while perceiving utterances. However, during the receptive subtask, participants were asked to determine if the phrase spoken the computer best matched a picture displayed on the right or left side of the screen. Therefore, participants had to determine the meaning of both pictures while simultaneously listening to, and remembering, the phrase spoken by the computer. As a result, this receptive subtask may have taxed the participants' working memory skills, a known difficulty for individuals with PWS (Curfs et al. 1991), more so than their prosody skills and at least more than the paired expressive subtask.

Lastly, the contrastive stress task measured participants' ability to use prosody to understand and express key words within in an utterance. The results of these subtasks suggest that individuals with PWS may be better at emphasizing important words in utterances than perceiving important words. However, expressive contrastive stress is usually one of the first components of prosody acquired by TD children (Peppé, 2011), a pattern that may also be present in individuals with PWS. Future research may wish to further examine this possibility across the other PEPS-C subtasks.

In contrast, comprehension seemed to exceed production on tasks involving single-word items (i.e., turn-end, affect). Notably, the paired tasks in which participants performed better on the expressive subtask (i.e., boundary, contrastive stress), both involved multi-word items. This may indicate that individuals with PWS are better able to access prosody to comprehend single words than longer utterances. This pattern of results is consistent with prior research indicating that individuals with autism spectrum disorder have difficulty perceiving prosodic changes over longer speech stimuli versus shorter stimuli (Järvinen-Pasley et al., 2008). In contrast, individuals with PWS are better able to use prosody to express longer utterances than single words. This may suggest that individuals with PWS rely on additional verbal content to accurately convey intended messages, a characteristic that is also observed in individuals with autism spectrum disorder (Lindner & Rósen, 2006).

Limitations and Future Directions

There are several potential limitations that may have impacted the results of this study. Most notable is the study's small sample size. Although small sample sizes are common among studies investigating rare etiologies of IDD, the medium and large effect sizes seen across the results indicate that more research with larger samples is warranted. In addition, the participants in the present study were limited to adolescents and adults with PWS, and future longitudinal research may wish to examine the development of prosody in PWS. Additional limitations include the different number of sessions implemented across different participants. This is the result of the travel and resources needed to collect data from the additional participants with PWS. Furthermore, the decision to match on nonverbal standard scores may also have impacted the results, and a different pattern of performance may have been observed if groups were matched on different skills. However, matching presents a unique challenge as matching on one variable may lead to a mismatch on others, and this study was an important first step toward understanding the syndrome specificity of prosody in PWS. Future research may also wish to examine the way prosodic productions differ within individuals with IDD, even when they are interpreted correctly by the listener. Targeting these subtle differences may reduce the perception of social oddness associated with expressive prosodic deficits. Lastly, it may be beneficial to examine the relationship between prosodic strengths and other areas of communication, social skills, and literacy among individuals with IDD.

Implications

The pattern of strengths and weaknesses observed in this sample may provide initial therapy goals for speech-language pathologists (SLPs) targeting prosody within this population. Though participants with PWS performed better than participants with IDD on some measures of prosody, the results of the present study indicate a need for services aimed at increasing prosodic skills among both populations. For individuals with PWS, this includes both receptive and expressive prosody. For example, SLPs may wish to first target skills related to understanding contrastive stress, as this subtask represented the highest level of difficulty among the participants in this study.

Prosodic strengths could also be leveraged to improve areas of weakness. For example, the ability to use stress to emphasize important information could be used to improve comprehension of the same skill. More specifically, a client may complete an activity similar to the expressive contrastive subtask on the PEPS-C. For example, the SLP may ask the client a question about their class schedule in which part of the question is incorrect, e.g., Do you go to lunch after recess? The client then uses stress to correct the utterance, e.g., No, I go to recess *before* lunch. Following a correct response to a target item, the SLP and client may then discuss the difference in prosodic cues between the client's correct production compared to an incorrect production. Once the client has identified the difference between correct and incorrect productions, the client may then complete an activity similar to the receptive contrastive subtask, in which the client uses the knowledge gained in the production activity to assign the same prosodic cues in a comprehension activity. For example, the SLP may tell the client they forgot to pack an item in their lunch, e.g., I meant to pack a peanut butter and *jelly* sandwich. Using their knowledge of contrastive stress, the client then indicates which item the SLP forgot to pack.

Conclusions

The results of the present study indicate that adolescents and adults with PWS may have strengths in some areas of prosody when compared to adults with mixed-etiology IDD matched on nonverbal ability. These areas of relative strength include regulating conversations (i.e., turn-end), segmenting utterances (i.e., boundary), and identifying differences in prosodic form (i.e., discrimination). Furthermore, individuals with PWS show stronger expressive versus receptive prosody skills at the utterance-level, a finding that aligns with some previous research (e.g., Järvinen-Pasley et al., 2008). Interestingly, individuals with PWS demonstrated the greatest strength in using stress to emphasize important information with the greatest weakness comprehending stress. Individuals with PWS also demonstrated relative strengths in prosody compared to nonverbal matched individuals with IDD, particularly on skills related to regulating conversations and segmenting utterances. However, individuals with PWS may also have weaknesses in some aspects of both receptive and expressive aspects of prosody, specifically on skills related to comprehending utterance segments and stress. While individuals with PWS appeared to demonstrate a relative strength compared to individuals with IDD when distinguishing prosodic cues, this skill still presents as a challenge for individuals with PWS as their performance on this subtask was among the lowest compared to other subtasks. Given the importance of prosody for effective communication (Gerken & McGregor, 1998), the results of this study suggest a need for interventions focused on both components of prosody.

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VITAE

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EDUCATION

B.S. Communication Sciences and Disorders, University of Mississippi, 2018
Sally McDonnell Barksdale Honors College

PROFESSIONAL EXPERIENCE

2019 – 2020 Graduate Research Assistant: Directed Research Practicum, Neurogenic Disorders Lab

2018 – 2020 Graduate Teaching Assistant: Research Methods, Introduction to Phonetics, Fluency and Voice Disorders, Phonological Disorders, Neurogenic Disorders of Language

2018 – 2020 Graduate Practicums

RESEARCH PRESENTATIONS

2019 Thome, E. K. *The Prosodic Profile of Individuals with Prader-Willi Syndrome*. Oral presentation given as a competitor in the Three-Minute Thesis Competition.

2019 Thome, E. K., Loveall, S., Hawthorne, K. *The Prosodic Profile of Individuals with Prader-Willi Syndrome.* Poster session: American Speech-Language-Hearing Association Convention.

2019 Crenshaw, K., Thome, E. K., Loveall, S., Hawthorne, K. *The Prosodic Abilities of Individuals with Intellectual and Developmental Disabilities.* Poster session: Mississippi Speech-Language-Hearing Association Annual Continuing Education Conference.

2018 Thome, E. K., Loveall, S, & Henderson, D. E. *A Survey of Speech-Language Pathologists' Use and Understanding of Evidence-Based Practice.* Poster session: American Speech-Language-Hearing Association Convention.

2018 Thome, E. K., Loveall, S, & Henderson, D. E. *A Survey of Speech-Language Pathologists' Use and Understanding of Evidence-Based Practice.* Guest lecturer: CSD 495: Introduction to Clinical Science.

2018 Thome, E. K., Loveall, S, & Henderson, D. E. *A Survey of Speech-Language Pathologists' Use and Understanding of Evidence-Based Practice.* Presenter: Communication Sciences and Disorder's Research Colloquium.

2018 Thome, E. K., Loveall, S, & Henderson, D. E. *A Survey of Speech Language Pathologists' Use and Understanding of Evidence-Based Practice.* Poster session: Mississippi Speech-Language-Hearing Association Annual Continuing Education Conference.

PUBLICATIONS

Under Review Thome, E. K., Loveall, S., Henderson, D. E., *A Survey of Speech Language Pathologists' Use and Understanding of Evidence-Based Practice*

LEADERSHIP

2019 – 2020 Undergraduate Student Mentor, Sally McDonnell Barksdale Honors College Mentoring Network

2018 – 2019 Founding Member, Vice Chair, & Department Representative, Interprofessional Education Student Liaison Advisory Council

GRANTS, SCHOLARSHIPS, HONORS, AND AWARDS

2019 Graduate Student Council Research Grant
2018 Who's Who Among Students
2018 Tommy and Suzan Thames Communicative Disorders Endowment Award
2018 Phi Kappa Phi Honor Society
2018 Order of Omega Honor Society