Characteristics of Hypotonia in Children: A Consensus Opinion of Pediatric Occupational and Physical Therapists

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Characteristics of Hypotonia in Children: A Consensus Opinion of Pediatric Occupational and Physical Therapists

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Purpose: The term hypotonia is often used to describe children with reduced muscle tone, yet it remains abstract and undefined. The purpose of this study was to identify characteristics of children with hypotonia to begin the process of developing an operational definition of hypotonia. **Methods:** Three hundred physical and occupational therapists were systematically selected from the memberships of the Pediatric Section of the American Physical Therapy Association and the Developmental Delay Section of the American Occupational Therapy Association and asked to complete an open-ended survey exploring characteristics of strength, endurance, mobility, posture, and flexibility. **Results:** The response rate was 26.6%. Forty-six physical therapists and 34 occupational therapists participated. The criterion for consensus about a characteristic was being mentioned by at least 25% of respondents from each discipline. The consensus was that children with hypotonia have decreased strength, decreased activity tolerance, delayed motor skills development, rounded shoulder posture, with leaning onto supports, hypermobile joints, increased flexibility, and poor attention and motivation. **Conclusion:** An objective tool for defining and quantifying hypotonia does not exist. A preliminary characterization of children with hypotonia was established, but further research is needed to achieve objectivity and clarity. **(Pediatr Phys Ther 2005;17:275–282)** *Key words: child, child/preschool, data collection/questionnaire, diagnosis/differential, infant, muscle hypotonia/classification, occupational therapy, physical therapy*

INTRODUCTION

Hypotonia refers to an impairment that may be associated many different conditions, including those of neuromuscular, genetic, central nervous system, connective tissue, and/or metabolic origins.[1,2] Hypotonia is frequently mentioned as a component of disorders such as Down syndrome (DS), Prader-Willi syndrome, and cerebral palsy.[3–5] In other instances, hypotonia may be the only impairment with no clear etiology.[2] In these cases, either benign congenital hypotonia (BCH) or congenital hypotonia with favorable outcome have been suggested as diagnoses; however, the authors clearly do not agree on what to call this condition.[2,6,7]

While there is agreement that hypotonia is reduced muscle tone, the term "muscle tone" is vague and imprecise, as no objective definition of the term has been developed.[8] Clinically, muscle tone is evaluated by assessing resistance to passive stretch.[8,9] Muscle contraction and stiffness of muscles, tendons, and soft tissue are thought to contribute to muscle tone.[8] Pathology of the stretch reflex mechanism and decreased segmental motor neuron pool excitability have been suggested as the physiological basis for hypotonia.[8] According to Hunt and Virji-Babul,[8] the evaluation and interpretation of muscle tone is highly controversial and the lack of a specific measure for tone is the result of a lack of knowledge about the underlying mechanisms of tone. Thus, the term hypotonia remains abstract and poorly defined.

The Infant Neurological International Battery (INFANIB) has been purported to be useful to evaluate hypotonia using the French angles, which include the scarf

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sign, heel-to-ear, popliteal angle, and leg abduction maneuvers.[10] However, the author of the INFANIB acknowledges that the main problem with these items is reliability. Most clinicians visually estimate the angles and then compare them to the pictures in the INFANIB manual. Pilon et al[11] state that although the "construct of muscle tone may not be fully captured using the French Angles Factor of the INFANIB, it remains one of the few tools available for use."

In contrast, definitions and assessment techniques for hypertonicity have been published and accepted.[9,12] The purpose of developing these definitions was to enable reliable communication between clinicians, accurate diagnosis, and proper patient selection for medical and surgical interventions.[12] A clinical rating scale, the Modified Ashworth Scale, has been developed to help quantify hypertonia. While this tool has been criticized for subjectivity and inability to detect small changes in muscle tone, it is still considered the gold standard for assessing hypertonicity.[9] Sanger et al[12] state that the "ultimate goal is to provide a reliable method to characterize hypertonia and to establish effective treatment options for affected children."

This goal is also important for children who have hypotonia, but the scientific community has yet to agree on the definition, diagnostic criteria, or assessment techniques for hypotonia. At this time, only a few studies have investigated children with hypotonia. These studies have discussed differential diagnosis in hypotonia of central versus peripheral origin,[13,14] long-term prognosis for children with hypotonia,[1,11] assessment techniques,[8,11,14] characteristics of children with hypotonia,[1] and suggestions for intervention.[11] However, even though hypotonia was an inclusion criterion for children in each of these studies or the topic of the report, the term was not operationally defined by any of the authors.

Currently, there is no way of knowing definitively if clinicians are using the term hypotonia to describe the same phenomenon. Without an operational definition of hypotonia, clinicians will have difficulty consistently identifying children with hypotonia, conducting further research on the efficacy of interventions for hypotonia, comparing study results, or applying study results to individual patients. Therefore, work needs to be done to define hypotonia and delineate the specific characteristics of hypotonia in children.

To begin to establish consensus among physical and occupational therapists on a definition of hypotonia in children, the first step in the research was to conduct an open-ended survey. Specifically, for the purposes of our study, professionals from the Pediatric Section of the American Physical Therapy Association and the Developmental Delay Section of the American Occupational Therapy Association were surveyed about the methods and criteria used to identify a child as having hypotonia. This step was important to begin the process of defining hypotonia so that pediatric health care professionals can be sure that they are describing the same phenomenon when they use that term.

METHODS

Subjects

Occupational therapists (OTs) in the Developmental Delay Section of the American Occupational Therapy Association and physical therapists (PTs) in the Pediatric Section of the American Physical Therapy Association were systematically selected to complete a survey on hypotonia. These groups of health professionals were used because of their direct experience with children and their participation in appropriate sections of each respective professional association. A list from each organization provided a systematic sample of the members of each of the two groups. One hundred fifty surveys were mailed to potential participants in each discipline, for a total sample population of 300 pediatric therapists. This study was approved by the University of Indianapolis Committee on Research Involving Human Participants.

Instrument

The survey consisted of both open- and close-ended questions (Appendix). The open-ended questions addressed a proposed set of attributes associated with hypotonia such as strength, mobility, posture, endurance and flexibility. Specifically, participants were asked to identify clinical features in each of these categories that they observe in children they believe have hypotonia. Respondents were given the opportunity to identify objective measures they use to quantify hypotonia and to list the medical diagnoses commonly associated with hypotonia. Additionally, participants were asked to state any differences in intervention techniques for children with hypotonia as compared to children with normal muscle tone or spasticity. They were asked to give their professional opinion about whether children outgrow hypotonia. Finally, clinicians were given the opportunity to define hypotonia and to discuss other issues observed in children with hypotonia. Although the survey also included questions about assessment, interventions, and outcomes, this paper focuses only on the characteristics of hypotonia as identified by the respondents.

Close-ended questions asked about personal demographics, such as degree(s) obtained, and years of experience. In addition, the survey included close-ended questions to attain information about the current practice setting of the respondent and the average age of patients the respondent treated.

Procedure

A pilot survey was created to test the validity and clarity of the survey. The pilot survey was sent to 10 OTs and 10 PTs chosen from the Indianapolis, IN metropolitan area using convenience sampling. Pediatric PT and OT faculty at the University of Indianapolis selected pilot study participants. A cover letter was included to explain the purpose of the research and indicate the time frame for reply. A statement about the confidentiality of the respondents was included in the cover letter, and all pilot surveys

were numerically coded for confidentiality and analysis purposes. After pilot surveys were returned, analysis of the data was performed to determine content validity. Revisions of the pilot survey were not necessary based on the data gathered as well as feedback from the participants.

The final survey was sent by U.S. mail to all 300 subjects identified earlier, along with the cover letter describing the purpose of the study and how confidentiality would be protected. All surveys were coded with a three-digit number prior to mailing so that the nonrespondents could be identified. Surveys were remailed to nonrespondents three weeks after the initial mailing.

Data Analysis

The open-ended questions of each survey were analyzed and coded by seven raters. The raters included five student PTs, a pediatric PT, and an expert in PT research. After reviewing the same 10 surveys individually, the raters discussed common themes discovered and developed the coding sheet. Raters used the coding sheet to organize the descriptive terms for the clinical features, definitions, and specific tests used by each respondent. Two raters coded each question of every survey, and then each pair discussed the reasoning for coding the specific questions with the rest of the research team. Differences in coding were negotiated until a consistent and accurate code was determined for each answer listed on the surveys. A database was then formed to analyze the responses in each category.

Statistical analysis was completed using SPSS version 10.0 for Windows. The percentage of respondents in each discipline was calculated. Descriptive statistics consisting of means and standard deviations were calculated to portray demographic information.

RESULTS

Response Rate

A total of 86 of the 300 surveys were returned, which generated a return rate of 28.6%. Of the surveys returned, 80 were completed (return rate of 26.6%) and six were not completed by individuals who cited a lack of experience or inadequate training in pediatrics. Incomplete surveys were not included in the data analysis. Physical therapists returned 46 of the 80 complete surveys, and OTs returned the remaining 34 surveys.

Statistical Analysis

Demographic data of the respondents are presented in Table 1. The coded survey responses were analyzed, and characteristics that were noted by at least 25% of all respondents are presented in Table 2. Table 3 represents common medical diagnoses associated with hypotonia that were reported by the respondents.

Consensus of Characteristics

Our criterion for consensus about a characteristic of hypotonia was that it had to be mentioned by at least 25% of respondents in each discipline. Based on this, our respondents agreed that a child with hypotonia displays the following characteristics: decreased strength, decreased activity tolerance, delayed motor skill development, rounded shoulder posture, leaning onto supports, hypermobile joints, increased flexibility, and poor attention and motivation.

TABLE 1
Demographics of Survey Respondents

	N-PT	% PT	N-OT	% OT
Experience (yr)				
<1	1	2.2	0	0
1–3s	4	8.7	4	11.8
3–10	15	32.6	10	29.4
>10	26	56.5	20	58.8
Degree obtained				
Bachelor	35	76.1	22	64.7
Entry-level master's	16	34.8	7	20.6
Entry-level doctorate	1	2.2	0	0
Postprofessional master's	12	26.1	10	29.4
Postprofessional doctorate	2	4.3	1	2.9
Certificate	6	13.0	1	2.9
Setting				
School setting	21	45.7	19	55.9
Outpatient facility	21	45.7	11	32.4
Inpatient facility	6	13.0	1	2.9
Home care	15	32.6	9	26.5
Other	12	26.1	15	44.1
Weekly pediatric case load, no. of patients				
<5	5	10.9	3	8.8
5–9	4	8.7	3	8.8
10–14	5	10.9	7	20.6
15–20	10	21.7	5	14.7
>20	21	45.7	16	47.1

N-PT = number of physical therapist responses; % PT = percentage of physical therapist responses; N-OT = number of occupational therapist responses; % OT = percentage of occupational therapist responses.

DISCUSSION

Characteristics

The available literature both supports and contradicts the findings of this survey. Several of the eight characteristics for which we found consensus indeed are mentioned in the literature about hypotonia and some are not. In the literature that addressed some of the characteristics our respondents identified, opinions varied and sometimes contradicted what our respondents reported.

Strength

In regards to the characteristic of decreased muscle strength, most of the literature reports that hypotonia and generalized muscle weakness are correlated.[2,8,15,16] However, this correlation is not universally accepted. Jacobson[16] stated that muscle weakness and hypotonia typically coincide in children with peripheral muscle or nerve disease but that children with central motor problems may be significantly hypotonic but have reasonable strength.

TABLE 2

Hypotonia Characteristics

	N	%	N-PT	% PT	N-OT	% OT
Decreased strength	55	68.8	28	60.9	27	79.4
Decreased proximal strength	31	38.8	24	15.2	7	20.6
Hand skills	24	30	7	15.2	17	50.0
Muscle recruitment	21	26.3	13	28.3	8	23.6
Antigravity movements	20	25	13	28.3	7	20.6
Endurance						
Decreased activity tolerance	74	92.5	40	87.0	34	100
Mobility						
Delayed motor skills/ development	35	43.8	26	56.5	9	26.5
Gait deviations	21	26.3	15	32.6	6	17.6
Posture						
Rounded shoulders	53	66.3	33	71.7	20	58.8
Leaning onto supports	34	42.5	12	26.1	22	64.7
Forward head posture	21	26.3	17	37.0	4	11.8
Posterior pelvic tilt	20	25	12	26.1	8	23.6
Flexibility						
Hypermobile joints	44	55	25	54.3	19	55.9
Increased flexibility	30	37.5	15	32.6	15	44.1
Other						
Poor attention/Motivation	26	32.5	13	28.3	13	38.2

N = number of responses; % = percentage of responses; N-PT = number of physical therapist responses; % PT = percentage of physical therapist responses; N-OT = number of occupational therapist responses; % OT = percentage of occupational therapist responses.

TABLE 3

Diagnoses Associated with Hypotonia

	N	%	N-PT	% PT	N-OT	% OT
Down syndrome	55	68.8	36	78.3	19	55.9
Cerebral palsy	46	57.5	28	60.9	18	52.9
Autism/PDD spectrum	26	32.5	11	23.9	15	44.1
Developmental delay	26	32.5	19	41.3	7	20.6
Other genetic syndromes	23	28.8	14	30.4	9	26.5
Sensory integration dysfunction	11	13.8	2	4.3	9	26.5
No specific diagnosis	9	11.3	7	15.2	2	5.9
Prematurity	9	11.3	6	13.0	3	8.8
Spina bifida	7	8.8	6	13.0	1	2.9
ADD/ADHD	6	7.5	0	0.0	6	17.6
Muscular dystrophy	4	5	3	6.5	1	2.9

N = number of responses; % = percentage of responses; N-PT = number of physical therapist responses; % PT = percentage of physical therapist responses; N-OT = number of occupational therapist responses; % OT = percentage of occupational therapist responses; PDD = pervasive developmental disorder; ADD = attention deficit disorder; ADHD = attention-deficit/hyperactivity disorder.

Steifel[15] stated that a central etiology for the abnormal muscle tone may indeed lead to weakness, but he also wrote that weakness is absent in BCH. Prasad and Prasad[2] noted that children with central hypotonia had significant axial weakness and that weakness was common with peripheral nerve involvement. Respondents in our survey did not address a difference between central and peripheral disease as a cause of hypotonia. Finally, Jacobson[16] notes that children with hypotonia are weak, but cautions that weakness and hypotonia represent two different aspects of motor control and are not interchangeable terms. Given the lack of agreement in the literature, we conclude that it is unwise to assume that a child with hypotonia will have weakness without formally examining the child's strength.

Several authors addressed the decreased strength issue specifically with individuals with DS. Hunt and Virji-Babul[8] found in individuals with DS decreased peak torque using electromyography. Lauteslager et al[17] did not directly state that children with DS have decreased strength, but they did note that decreased proximal stability and muscle cocontraction contribute to the motor delay seen in this population. Caution must be used in applying these results to any child with hypotonia because even though hypotonia is an accepted component of DS, the other neurological aspects of DS could possibly account for decreased strength.[18]

Activity Tolerance

In our survey, decreased activity tolerance was the most often reported characteristic of children with hypotonia, yet none of the previous reports mentions this issue. Perhaps this is because most of the literature available on hypotonia focuses on differential diagnostic procedures or prognosis. We surveyed practicing physical and occupational therapists, and poor endurance may be a common issue that they have to face in their treatment sessions.

Delayed Development

Almost half of our respondents reported that hypotonia is associated with a delay in motor skill development. The literature is both supportive and contradictory about this point. Several authors noted that either normal motor development or mild motor retardation is associated with BCH and/or congenital hypotonia with favorable outcome.[2,6] Shuper et al[1] noted that while none of the infants in their study were diagnosed as hypotonic at birth, they were referred for examination and ultimately diagnosed with BCH because of motor delays. However, Steifel[15] wrote that developmental delay is usually absent in BCH. Pilon et al[11] specifically investigated the relationship between hypotonia, joint laxity, and motor development. These authors found that in a population-based study of full-term infants, there was no significant relationship between hypotonia and motor development or between joint laxity and motor development. Thus, the available literature continues to provide contradictory information about whether delayed motor development is related to hypotonia.

Posture

In the category of posture, our respondents noted that children with hypotonia have a rounded shoulder posture and tend to lean against external supports. The characteristic of rounded shoulder posture was not specifically mentioned in the literature we reviewed; however, Lauteslager et al[17] note that children with DS have decreased postural

control and insufficient cocontractions for joint stabilization. This could be viewed as the cause of the postural deviations noted by our respondents. Thus, while the characteristic of rounded shoulder posture is not specifically mentioned in the literature, there does seem to be support for it. The characteristic of leaning against external supports could potentially be viewed as a compensatory strategy for decreased postural control or decreased strength. Whether leaning is truly a characteristic of hypotonia or just a compensatory strategy needs to be further clarified in future studies.

Flexibility

Under the category of flexibility, our respondents agreed that hypermobile joints and increased flexibility were characteristics of children with hypotonia. However, differentiation should be made between laxity of the ligaments and increased extensibility in the muscle itself.[16] Methods for assessing joint laxity have been proposed,[19,20] but no formal method of assessment of flexibility in children has been published. In a study by Livingstone and Hirst,[21] joint laxity and orthopedic problems were examined in children with DS. Although all 39 children were assumed to have hypotonia because of their diagnosis of DS, only 23 had signs of joint laxity.[21] Lauteslager et al[17] concluded that joint stability is negatively influenced not only by joint laxity but also by decreased postural tone and insufficient cocontraction around a joint. In their report of a longitudinal study of children with congenital hypotonia with favorable outcome, Carboni et al[6] stated that there is a correlation between joint hypermobility and hypotonia. In fact, these authors suggest that joint hyperlaxity in adolescents as described by orthopedic specialists corresponds to congenital hypotonia with favorable outcome in children described by pediatric neurologists. In our opinion, the distinction between joint laxity and increased flexibility is unclear in both the literature and our survey findings. Some respondents distinguished between muscle flexibility and joint laxity, but most did not. While clinically the two characteristics are frequently found to coexist and the literature supports that they both are related to hypotonia, we suggest that they are two separate entities, and a thorough assessment is needed to distinguish between them.

Poor Attention and Motivation

Another characteristic identified by respondents to our survey was poor attention and motivation. We did not find anything in the literature to either support or contradict this result. We also did not find any literature that would perhaps offer a rationale as to why our respondents included this as a characteristic of a child with hypotonia. Future studies should attempt to clarify this reported aspect of hypertonia.

Medical Diagnoses

DS and cerebral palsy were the two most commonly identified medical diagnoses associated with hypotonia (Table 3). Developmental delay, autism, and other genetic syndromes were also listed by at least 25% of respondents. Interestingly, the diagnoses BCH and congenital hypotonia with favorable outcome were not reported by our respondents. As Table 3 illustrates, our respondents associated the term hypotonia with a number of other diagnoses but did not consider hypotonia a diagnosis by itself.

Limitations

Inherent bias may have been one limitation of this study. The opinions of the investigators may have introduced bias in the content and style of the questions. A pilot study was conducted in an attempt to decrease this bias. Participants in the pilot study were enlisted to provide feedback ensuring that the questions were free from bias, encompassed all views, and were worded in a way as not to lead survey respondents toward one particular answer.

Respondents who had a strong position toward or against the topic of children with hypotonia may have been more likely to return the survey. This could have potentially affected the outcome of the surveys since those who did not respond may have had different opinions. Our response rate was also low, and this in part may be because of the demanding schedules of the clinicians, even though the survey only took about 15 minutes to complete. The difficulty of the task of defining hypotonia may also have limited the response rate.

Rater bias could have been present when coding and summarizing the open-ended question portion of the survey as pertinent information may have been omitted when analyzing the data collected. However, seven raters compared notes and negotiated final decisions to limit this type of bias.

Further Research

This survey was the first step in characterizing and defining hypotonia from the perspective of physical and occupational therapists. While we attempted to attain a general idea of what clinicians see in children identified as having hypotonia, the characteristics identified are still imprecise, and there is currently no objective means for assessing or quantifying hypotonia. More studies need to be done to further clarify these characteristics, validate them, and ensure agreement among practitioners. Additional research is warranted to further objectify hypotonia. This work could then lead to studies that address intervention, long-term outcomes, and prognosis for children with hypotonia.

CONCLUSION

A preliminary characterization of hypotonia indicates that PTs and OTs concur that a child with hypotonia displays decreased strength, decreased activity tolerance, delayed motor skill development, rounded shoulder posture with a tendency to lean onto supports, hypermobile joints, increased flexibility, and poor attention and motivation. Further research into these characteristics should lead to a better definition with measurable and objective criteria in

order to efficiently and effectively diagnose and treat children with hypotonia.

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APPENDIX A

Operational definition of hypotonia in children: a consensus opinion of pediatric physical and occupational therapists

Krannert School of Physical Therapy, University of Indianapolis

Please complete the questionnaire and return in the envelope enclosed. Record the time to complete the questionnaire.

1. What specific clinical features do you identify in a child with hypotonia or low tone? Please list specific examples of what you see clinically in each of these categories.

Strength

a._____

b._____

c._____

Posture

a._____

b._____

c._____

Endurance

a._____

b._____

c._____

Flexibility

a._____

b._____

c._____

Mobility

a._____

b._____

c._____

Other

a._____

b._____

c._____

2. What objective measures do you utilize to define or quantify hypotonia?

3. What are the common diagnoses of children that you have treated for hypotonia?

4. How are treatment options for a child with hypotonia different from a child with normal tone or spasticity? (Please list no more than three)

5. Based on your clinical experience and professional opinion, do children "outgrow" hypotonia?

6. Based on your clinical experience and professional opinion, how would you define hypotonia within children?

7. What other issues do you observe in children with hypotonia?

8. How many years of experience do you have as a pediatric therapist?

__ Less than 1 year
__ 1-3 years
__ 3-10 years
__ Over 10 years

9. What degrees have you obtained? (Check all that apply)

__ Bachelor degree
__ Entry-level doctorate degree
__ Post professional doctorate degree
__ Entry-level masters degree
__ Post professional masters degree
__ Certificate

10. What is your current practice setting? (Check all that apply)

__ School System
__ Inpatient
__ Other
__ Outpatient Clinic
__ Home Care
_____(specify)

11. What ages of children do you typically treat based on your normal caseload? (Indicate proportion of patients within each age range)

__ % Birth – 3 years old
__ % 3-6 years old (preschool)
__ % School age
__ % Adolescents

12. What is your average pediatric caseload per week?

__ Less than 5 pediatric patients
__ 5-10 pediatric patients
__ 10-15 pediatric patients
__ 15-20 pediatric patients
__ > 20 patients

Thank you for taking the time to participate in our study.

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