Paratonia: A Delphi Procedure for Consensus Definition

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ABSTRACT
Background and Purpose: Paratonia is a motor problem that develops during the course of dementia. Definitions of paratonia used in the literature differ considerably, which has clinical implications and may lead to an undesirable heterogeneity in study populations. For this reason, we initiated a Delphi procedure with known experts in the field to establish an operational consensus definition of paratonia. Methods: The Delphi procedure involved an anonymous and multi-stage approach presented as a questionnaire, with each stage building on the results of the previous one in order to reach consensus on the definition of paratonia. Results: Eight of 17 experts agreed to participate in the study. After 4 rounds, the participants reached consensus on the following definition: paratonia is a form of hypertonia with an involuntary variable resistance during passive movement. The nature of paratonia may change with progression of dementia (eg, from active assistance (aka Mitgehen) to active resistance). The degree of resistance depends on the speed of movement (eg, slow → low resistance, fast → high resistance). The degree of paratonia is proportional to the amount of force applied and increases with progression of dementia. The resistance to passive movement is in any direction and there is no clasp-knife phenomenon. Conclusion: The Delphi procedure resulted in a comprehensive, operational definition of paratonia. Future research should focus on the reliability and validity of this definition.

Key Words: paratonia, dementia, Delphi, movement disorders

INTRODUCTION
Dementia is becoming an increasing problem worldwide, with an estimated prevalence of 25 million people in the year 2000 and a projected prevalence of 63 million by 2030.1 Although movement disorders, which have a different underlying cause, are common in dementia, they are often not accurately described and are usually diagnosed as Parkinsonian or extrapyramidal signs, with rigidity, slowness, and impaired gait.2-6 Paratonia is a common motor problem seen in individuals with dementia. Dupré first described it in 1910 as the inability to relax muscles in combination with a mental disorder.7 In 1927, Kleist noted a similar phenomenon in his clinical observations of patients in a late stage of dementia.8 He observed motor negativism and called it Gegenhalten.

Carers and nurses find it difficult to wash and dress patients with paratonia because the problem is associated with a loss of mobility and with the development of contractures, especially in the late stages of the dementia.9,11 Passive movement therapy, to decrease high muscle tone and to sustain range of motion of affected joints, is the main therapy applied. However, in a pilot study of the efficacy of this intervention, we found that passive movement therapy seemingly worsened the joint and limb stiffness.12 Unfortunately, the study was underpowered, and because of the lack of a clear operational definition of paratonia, the study population was heterogeneous.

Paratonia differs from spasticity, which Lance defined as a motor disorder characterized by velocity dependent increase in the tonic stretch reflex with exaggerated tendon jerks. Parkinsonian rigidity on the contrary is defined as a resistance to passive movement of the limb whereby the degree of resistance is constant whether the limb is moved slowly or rapidly (like bending a lead pipe).6,13 However, differentiation with paratonia is hampered by inconsistent and even contradictory definitions of paratonia used in the various published studies. Most authors define paratonia as a resistance to passive movement or a sudden increase in muscle tone with accompanying elements like cogwheeling and Mitgehen (ie, actively assisting passive movement) and several factors that influence the degree of paratonia such as the amplitude and irregularity of passive movement, external stimuli (eg, sound and light), deep sleep, and the use of antipsychotic drugs.6,9,12-14,19,21-22 Furthermore, it is not known whether paratonia initially emerges in the lower or upper limbs or if it develops in a distinct pattern.17,19 Most authors mention that the degree of paratonia can be influenced by the amount of force applied and that it depends on the speed of movement, in which a forceful fast movement results in the most resistance.6,9,11,15,16,18,19,21 Some authors state that paratonia is more pronounced when the patient is instructed to relax and not by clinching of the contralateral fist and that it is characterized by the absence of a clasp-knife phenomenon.6,9,11,16,18,21

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practice and to differentiate it from other movement disorders, we initiated a Delphi procedure in which known experts in the field participated and reached consensus on the definition of paratonia. Here, we describe this procedure and list the most essential elements defining paratonia.

METHODS

The Delphi procedure is an instrument to reach consensus on a particular issue.\textsuperscript{23,24} The procedure entails a questionnaire for a panel of informed experts in a specific field. Once responses are collected the data are summarized and a new questionnaire is designed based on the former results. The respondents are asked to reconsider their initial opinion in light of the group results. This anonymous process is repeated at least once, yet preferably more often, in order to reach consensus.\textsuperscript{23,24}

We contacted clinicians with special expertise in paratonia and spasticity or rigidity and/or contractures in dementia, in order to get as broad a perspective on paratonia as possible. All authors of papers (in Dutch or English) identified by searches in MEDLINE, EMBASE, CINAHL, PsychINFO, and PEDro, in which paratonia was either the subject or was contrasted with spasticity or rigidity, were considered experts and possible participants. We preferred participants with a background in physical therapy.

In order to achieve consensus on the definition of paratonia we designed a questionnaire in which we classified available information on paratonia into 3 categories: short descriptions, influencing factors, and differentiating elements. First, we compiled a list of 12 commonly used short descriptions of paratonia. Then we selected 8 factors that influence the degree of paratonia. And finally, we identified 15 features that potentially differentiate paratonia from muscle spasm, contractures, and Parkinson rigidity.

We asked the participants to rate these 35 items on a 5-point Likert scale (1 = Not important at all, 2 = Somewhat important, 3 = Moderately important, 4 = Very important, and 5 = Extremely important) and to provide additional items for each category. In the subsequent consultation rounds, the participants were asked to rate the newly provided items and to review their initial rating in the light of the calculated group median for each item. Furthermore, we asked the participants to provide a cut-off score for each category. Items with a score equal to or higher than this cut-off score were considered essential for a proper definition of paratonia. All items with a score lower than this cut-off score or with ratings with a wide range (ie, no consensus between the participants) were discarded. In the fourth round, the final results were presented and the participants were asked if they agreed that the remaining items reflected a proper definition of paratonia (Figure 1). In all 4 rounds, the participants were invited to comment on the items presented, their own rating, the calculated group medians, and the final result.

RESULTS

We contacted 17 experts by e-mail and telephone, of whom 8 agreed to participate (see appendix; 3 from the Netherlands, 2 from the United States of America, 2 from the United Kingdom, and 1 from Australia). The other experts did not participate for reasons not related to the topic of this survey (ie, time constraints). All participants completed the project and were blind to the identity of the other participants during the Delphi procedure. The interval between each of the 4 rounds was about 5 weeks.

In the first round, the participants provided 2 additional short descriptions, 3 additional influencing factors, and 7 new differentiating elements. In the second and third rounds, 7 of the participants changed some of their ratings, which resulted in a greater similarity of scores. All participants added comments on several of the items during the first 3 rounds (Table 1). The final results are presented in Table 2. Four short descriptions of paratonia had scores higher than the group median cut-off score of 3.5: “A resistance to passive movement” (median score of 5), “An involuntary resistance” (median score of 5), “A form of hypertonia” (median score of 4.5) and “A variable resistance during passive movement” (median score of 3.5). No consensus was reached on the item “A combination of spasticity and rigidity in different grades.”

The factors identified as influencing the severity of paratonia had a group median cut-off score of 3. After analysis, 4 factors remained: “The nature of the paratonia may change with progression of the dementing illness (eg, early in the course of degenerative dementias, active assistance [Mitgehen] is more common and later in the disease active resistance is more common)” (median score of 5), “The degree of resistance varies depending on the speed of movement; slow \(\rightarrow\) decrease, fast \(\rightarrow\) increase” (median score of 4.5), “The degree of paratonia is proportional to the amount of force applied” (median score of 4) and “Paratonia increases with progression of dementia” (median score of 4). Three items were discarded because there was no agreement on their relevance (scores had a large range): “Improves with distraction,” “The degree of paratonia is proportional to the amplitude of passive movement,” and “Hypertonia decreases in deep sleep.” Only 2 of 22 differentiating items had scores higher than the median cut-off score of 3.75: “No clasp-knife phenomenon” and “The resistance to passive movement is in any direction.” Both had a group score of 5.0.

In the concluding fourth round, all participants agreed on the generated consensus description of paratonia. The description is presented in Figure 2.

DISCUSSION

This Delphi procedure established a useful and comprehensive definition of paratonia. However, our study had some limitations. First, the small number of participants may have influenced the validity of the definition. Second, this Delphi procedure was conducted in English, yet 3 participants were not native English speakers, which could be a source of bias. Third, the participants work with patients in different stages of dementia. Some participants work solely with patients with early dementias while others work only with patients in the
Description of phenomenon: Paratonia is

| An alteration of tone to passive movement \cite{14,16,22} | 3 | 3 | 3 |
| A resistance to passive movement \cite{6,10,14-17,19,20} | 5 | 5 | 5 |
| A sudden increase in muscle tone \cite{6,11,15,16} | 2 | 2 | 2 |
| An involuntary resistance \cite{10,11,19} | 4.5 | 5 | 5 |
| A progressively increasing resistance \cite{18} | 2.5 | 2 | 2 |
| An irregular resistance \cite{6,15,18} | 3 | 3 | 3 |
| An active resistance to changes in limb position \cite{6,18-20} | 3.5 | 3 | 3 |
| An active resistance to, or an active assistance (Mitgehen) of, passive movement \cite{6,14} | 2.5 | 2.5 | 2.5 |
| A variable resistance during passive movement \cite{9,15} | 3.5 | 3.5 | 3.5 |
| A form of hypertonia \cite{11} | 3.5 | 3.5 | 4 |
| A form of rigidity \cite{6} | 2.5 | 2.5 | 2.5 |
| Frozen in place \cite{18,19,27} | 1 | 1 | 1 |

Additional short descriptions provided by the participants in the first Delphi-round. Paratonia is:

An umbrella term to denote phenomena of muscle activity observed during functional activities requiring postural control that cannot be ascribed to forms of spasticity and rigidity

A combination of spasticity and rigidity in different grades

Influencing factors:

The degree of paratonia is proportional to the amount of force applied \cite{9,15,16,18,19,21} | 4 | 4 | 4 |

The degree of resistance varies depending on the speed of movement; slow → decrease, fast → increase \cite{6,9,11,15} | 4.5 | 4.5 | 4.5 |

The degree of paratonia is proportional to the amplitude of the passive movement \cite{9,16,21} | 3 | 3 | 3 |

The degree of paratonia is proportional to the degree of irregularity of the passive movement \cite{9,16} | 2 | 2 | 2 |

More pronounced by instructing the patient to relax \cite{9,11,16,18} | 2 | 2 | 2.5 |

External stimuli (eg, sound and light) elicit a paratonic response \cite{15} | 2 | 2 | 2 |

Hypertonia decreases in deep sleep \cite{15} | 3 | 3 | 3 |

Paratonia increases with progression of the dementia \cite{9-11,15,21,25,26} | 4 | 4 | 4 |

Additional influencing factors:

Improves with distraction (eg, asking the patient to count or to clench the contralateral fist) | 3 | 3 |

With the first movement you experience the most resistance, the second and third movement with the same limb and in the same direction the resistance is less | 2.5 | 2.5 |

The nature of the paratonia may change with progression of the dementing illness (eg, early in the course of degenerative dementias, active assistance is more common and later of the disease active resistance is more common) | 5 | 5 |
advanced stages of the disease. This diversity meant that it was not possible to achieve agreement on the relevance of some items, specifically those items in which paratonia changes over time. For example, in early dementia it is still possible to communicate with the patient and to observe if paratonia improves when the patient is distracted by asking him/her to count or to clench the contralateral fist. However, in an advanced stage this becomes impossible. Conversely, in the final stages of the disease a distinct pattern becomes visible with head and trunk in extension, arm in adduction/flexion, and legs in extension; whereas this pattern is not obvious in the early stages of dementia. Yet this diversity may be a strength of the study in that the definition covers paratonia of different stages of dementia. In this way, it became obvious that while paratonia can give the impression of being a combination of spasticity and rigidity of different severity, spasticity is not a part of paratonia and there is no clasp-knife phenomenon.

This project highlights the uncertainties surrounding the problem of paratonia. For example, one of the most discussed items that emerged from this Delphi procedure involved

<table>
<thead>
<tr>
<th>Differentiating elements:</th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Different grades of hypertonia are present in different parts of the body</td>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>No distinct pattern</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>The presence of Mitgehen (= actively assisting passive movement) suggests that the quality of rigidity is paratonic</td>
<td>3</td>
<td>2</td>
</tr>
<tr>
<td>Not on antipsychotic drug therapy</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>No exacerbation by movement of the contralateral fist</td>
<td>2.5</td>
<td>3</td>
</tr>
<tr>
<td>Cog wheeling can occur</td>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>No cog wheeling</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>No clasp-knife phenomenon</td>
<td>4.5</td>
<td>5</td>
</tr>
<tr>
<td>Occurs usually in the lower limbs</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Occurs usually in the upper extremities</td>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>Paratonia is independent of the starting position of the joint</td>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>The increased muscle tone is throughout the range of motion</td>
<td>3.5</td>
<td>3.5</td>
</tr>
<tr>
<td>The resistance to passive movement is in any direction</td>
<td>4.5</td>
<td>5</td>
</tr>
<tr>
<td>Correlates highly with echopraxia (= a tendency to imitate movements of others)</td>
<td>1.5</td>
<td>2</td>
</tr>
<tr>
<td>Correlates highly with the inability to inhibit eye-movements to peripheral stimuli</td>
<td>1</td>
<td>1</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Additional differentiating elements:</th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>A distinct pattern: head and trunk in extension, arms in adduction/flexion and legs in extension (with possible flexion component)</td>
<td>2.5</td>
<td>2.5</td>
</tr>
<tr>
<td>May observe variation with time of day</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>May observe day-to-day variation</td>
<td>2.5</td>
<td>2.5</td>
</tr>
<tr>
<td>Can change with different positions in relation to size of base; smaller base of support‡ increase of paratonia</td>
<td>3</td>
<td>2</td>
</tr>
<tr>
<td>The presence of eyelid paratonia (resistance to passive raising of the eyelids) suggests that the quality of limb rigidity is paratonic</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Contralateral reinforcement increases tremor, bradykinesia and rigidity but reduces paratonia.</td>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>These patients have a general high muscle tone on which the resistance to passive movement is superimposed</td>
<td>2</td>
<td>2</td>
</tr>
</tbody>
</table>

We asked the participants to rate these items on a 5-point Likert scale (1 = Not important at all, 2 = Somewhat important, 3 = Moderately important, 4 = Very important, and 5 = Extremely important) and we calculated the group median after each round.
Mitgehen. Most participants agreed that Mitgehen is in some way a part of paratonia and is mainly present in the early stages of dementia, but some participants questioned this, maintaining that it is impossible to distinguish Mitgehen from a normal inability to relax. The contribution of neurophysiological and biomechanical factors to paratonia was also a matter of discussion. Paratonia is hypothesized to develop centrally but to exert an effect on peripheral biomechanics. However, in clinical practice, it is difficult to distinguish between central neurophysiological and peripheral biomechanical factors. This was illustrated by the very different scores given to the item on the effect of the amplitude of the passive movement on the degree of paratonia. Furthermore, it is unclear whether paratonia can be felt throughout the whole range of motion, whether it is independent of the starting position of the joint, and whether cogwheeling can occur with paratonia.

This operational definition of paratonia should be seen as a first step to a better understanding of the motor disturbances of dementia. With this definition differentiation between paratonia and Parkinsonian rigidity and spasticity should be possible. Contrary to paratonia, Parkinsonian (lead pipe) rigidity has a constant degree of resistance which is not influenced by the speed of the movement. In contrast with the Lance definition of spasticity, there are in paratonia no exaggerated tendon jerks (no clasp-knife phenomenon). With this consensus definition it becomes clear that most authors used a less restrictive definition, especially according to the differentiation with spasticity, making the results of these studies difficult to interpret. The definition used by Paulson et al, Souren et al, and Kurlan et al are very close to our established description of paratonia.

CONCLUSIONS

By using the Delphi procedure we have established a comprehensive, operational definition of paratonia. This operational
Paratonia is a form of hypertonia with an involuntary variable resistance during passive movement. The nature of paratonia may change with progression of the dementing illness (e.g., active assistance (aka Mitgehen) is more common early in the course of degenerative dementias, whilst active resistance is more common later in the course of the disease). The degree of resistance varies depending on the speed of movement (e.g., a low resistance to slow movement and a high resistance to fast movement). The degree of paratonia is proportional to the amount of force applied. Paratonia increases with progression of dementia. Furthermore, the resistance to passive movement is in any direction and there is no clasp-knife phenomenon.

Figure 2. The consensus definition of paratonia.

We asked the participants to provide a cut-off score for each category. Items with a score equal to or higher than this cut-off score were considered essential for a proper definition of paratonia.

<table>
<thead>
<tr>
<th>Group median</th>
<th>Description of phenomenon: Paratonia is (Group median cut-off point 3,5)</th>
</tr>
</thead>
<tbody>
<tr>
<td>5</td>
<td>A resistance to passive movement</td>
</tr>
<tr>
<td>5</td>
<td>An involuntary resistance</td>
</tr>
<tr>
<td>4</td>
<td>A form of hypertonia</td>
</tr>
<tr>
<td>3.5</td>
<td>A variable resistance during passive movement</td>
</tr>
</tbody>
</table>

Influencing factors (Group median cut-off point 3)

| 5            | The nature of the paratonia may change with progression of the dementing illness (e.g., Early in the course of degenerative dementias, active assistance (Mitgehen) is more common and later of the disease active resistance is more common) |
| 4.5          | The degree of resistance varies depending on the speed of movement; slow → decrease, fast → increase |
| 4            | The degree of paratonia is proportional to the amount of force applied    |
| 4            | Paratonia increases with progression of the dementia                     |

Differentiating elements (Group median cut-off point 3,75)

| 5            | No clasp-knife phenomenon                                               |
| 5            | The resistance to passive movement is in any direction                   |

The definition of paratonia should be seen as a first step to a better understanding of the motor disturbances of dementia. More research is needed. For instance, cross-sectional or preferably longitudinal research should focus on the reliability and validity of this definition and on the ambiguous items, to clarify whether they contribute to the description of paratonia. Only when these uncertainties are removed will it be possible to search for an effective intervention.

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REFERENCES


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