1. **Review title**

Epidemiology Of Dysphagia In Adults With Atypical Parkinsonian Disorders:

A Systematic Review And Meta-Analysis

2. **Anticipated or actual start date:** 01 April 2021
3. **Anticipated completion date:** 14 May 2021
4. **Stage of review at time of this submission** *mandatory*

Preliminary searches started

5. **Named contact** *mandatory*

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9. **Organisational affiliation of the review** *mandatory*

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10. **Review team members and their organisational affiliations** *mandatory*

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11. **Funding sources/sponsors** *mandatory*

None

12. **Conflicts of interest** *mandatory*

None known.

13. Collaborators

None.

14. Review question *mandatory*

1. What is the prevalence of dysphagia in adults with atypical parkinsonian disorders (APD)?
2. Which symptoms characterise dysphagia in the subtypes of APD?
3. What are the risk factors for developing dysphagia in APD?

15. **Searches** *mandatory*

Electronic bibliographic databases to be searched are PubMed, EMBASE, CINAHL, Web of Science Core. Grey literature will be included by searching ProQuest Dissertations & Theses A & I, and OpenGrey. All databases will be searched from inception to April 2021 with ongoing searches to May 2021. The search strategy will also include checking of included studies reference lists by JR to identify further relevant studies. No date, time or language restrictions will be set. Additional details of the search strategy can be found in the attached file.

16. URL to search strategy.

An example of a search strategy for PubMed will be supplied in an electronic file.

17. **Condition or domain being studied** *mandatory*

Atypical parkinsonian disorders (APD) are primarily caused by the neurodegenerative processes progressive supranuclear palsy (PSP), multiple system atrophy (MSA), Dementia with Lewy bodies (DLB), and Corticobasal syndrome (CBS) (Litvan, 2005). Both, idiopathic Parkinson's disease (PD) and APDs are aggregated under the term Parkinsonism. In contrast to PD, APDs progress faster (Müller et al., 2000), affected patients' survival time is shorter (Litvan et al., 1996; Watanabe et al., 2002; Wenning et al., 1995; Wenning et al., 1998), and the treatment differs substantially (Levin et al., 2016). However, distinction of the various causes of Parkinsonism is difficult which leads to underdiagnosis, delay of diagnosis, and even misdiagnosis (Joutsa et al., 2014; Litvan et al., 1996; McFarland, 2016; Wenning et al., 1995). The faster progression of APDs becomes apparent in a more rapid functional decline (McFarland, 2016). This manifests in more severe complications occurring earlier on, such as dysphagia, with a shorter latency of dysphagia onset in APD than in PD (Müller et al., 2001).

Dysphagia is defined as difficulty in swallowing saliva, food, or liquid, due to sensory and/ or motor deficits. For the APDs in general an impairment of the oral and pharyngeal phase, and with some syndromes of the oesophageal phase is reported, but the characteristics vary between the different neurodegenerative processes (Grunho et al., 2015; Higo et al., 2005; Higo et al., 2003; Johnston et al., 1997; Larsson et al., 2017; Lee et al., 2018; Litvan et al., 1997; Sulena et al., 2017). In general,

an impaired swallowing process increases the risk of aspiration. As the dysphagia characteristics vary, so do the reported risk of aspiration from no observed aspiration up to 90.5% between patients with PSP, MSA, DLB, and CBD (Grunho et al., 2015; Higo et al., 2003; Johnston et al., 1997; Lee et al., 2018; Yamamoto et al., 2010). Dysphagia is related with aspiration pneumonia in MSA and DLB (Higo et al., 2003; Yamamoto et al., 2010) and is named as the most common cause of death in PSP patients (Maher & Lees, 1986).

18. **Participants/population** *mandatory*

Inclusion: Adults with dysphagia due to an atypical parkinsonian disorder (MSA, PSP, DLB, CBD) diagnosed based on neurological investigations, regardless of severity level, disease duration or context

Exclusion: Adults with any other underlying diseases or comorbidities that might also cause dysphagia, such as nonprogressive neurological or oncological conditions

19. Intervention(s), exposure(s). *mandatory*

Due to this review examining the epidemiology of dysphagia in form of determining prevalence and describing its nature, details regarding interventions are not relevant to data extraction.

20. **Comparator(s)/control.** *mandatory*

Not relevant.

21. **Types of study to be included.** *mandatory*

Quantitative research, such as observational studies, including cohort, case-control, case reports, case series, and cross-sectional studies will be included. Randomised and non-randomised controlled trials will be included as well since they might also provide prevalence data and description about the nature of the condition of interest. Grey literature, such as conference abstracts, reports, dissertations, and theses are deemed eligible to ensure full inclusion of any relevant information. Errata and letters are included as they potentially include information regarding critical weaknesses and retractions. Secondary research in form of literature reviews will be included as a source to identify further eligible primary research but will be excluded from data extraction to reduce risk of bias from duplication of data.

22. Context.

There will be set no restrictions regarding the context. Patients with an APD regardless of the context will be included.

23. **Main outcome(s).** *mandatory*

Dysphagia: defined as difficulties in swallowing saliva, food, liquid, including impairment of oral, pharyngeal, and/or oesophageal phase, diagnosed either instrumentally, clinically or based on patient reported outcomes

- Dysphagia prevalence
- Dysphagia symptoms determined by an instrumental assessment
- Dysphagia symptoms determined by a clinical swallowing examination
- Dysphagia symptoms reported by the patient

24. **Additional outcome(s).** *mandatory*

- Risk factors for developing dysphagia
- Latency of onset of dysphagia

25. Data extraction (selection and coding). *mandatory*

Data extraction will be executed independently by two authors (JR and ET). COVIDENCE will be used for data management, including study selection and data extraction.

Selection: Following the search, duplicate texts will be removed. Title and abstract screening to exclude obvious ineligible results, will precede full text screening of potentially relevant results. JR will check reference lists of included studies to identify further relevant studies. Any disagreements will be resolved by discussion. Disagreements will be resolved by a third authors (MW).

Coding: The data extraction form will be created within COVIDENCE. JR will pilot the data extraction form on two randomly selected studies. Subsequently, JR and ET will independently extract data regarding study characteristics, eligibility, condition, population, and context.

26. Data extraction

JR will contact authors to obtain missing data if the article is published within a 5 year timeframe. Following a period of one month after no response with two contact attempts, studies will be listed as studies awaiting classification. Data will be extracted independently by JR and ET using the data extraction form generated in COVIDENCE. Disagreements will be resolved by discussion. Persisting disagreements will be resolved by a third reviewer (MW).

Category	Type of Data
Study description	▪ Authors/ Year of Publication ▪ Title ▪ Study Design ▪ Database
Eligibility	▪ Yes/No

	▪ Reasons for Exclusion
Population	▪ Number of participants
	▪ Age
	▪ Sex
	▪ Disease duration
	▪ Disease severity
	▪ Type of APD
	▪ Co-morbidities
Context	▪ Country
	▪ Setting
Condition	▪ Definition
	▪ Assessment-type
	▪ Prevalence
	▪ Dysphagia latency
	▪ Survival time
	▪ Symptoms (oral, pharyngeal, oesophageal phase)
	▪ PEG/RIG or NG-Tube
	▪ Self-perception
	▪ History of Pneumonia
	▪ Concurrent speech and language-related syndromes (e.g., dysarthria)
	▪ Risk factors

27. **Risk of bias (quality) assessment.** *mandatory*

The methodological quality will be assessed independently by JR and ET. Disagreements will be resolved by discussion. Any persisting disagreements will be resolved by a third reviewer (MW). The JBI Critical Appraisal Checklist for Studies Reporting Prevalence Data (Munn et al., 2015) will be used to assess the methodological quality of included studies. With 9 items the JBI checklist assesses studies that report prevalence data. Furthermore, it is applicable across various study designs. “Yes” answers will be scored ”1”, and the answers “No”, “Unclear”, and “Not applicable” will be scored “0”. Quality will be rated using a summary graph generated from JBI Critical Appraisal Checklist for Studies Reporting Prevalence Data (Munn et al., 2015).

28. **Strategy for data synthesis.** *mandatory*

The characteristics of included studies will be presented narratively and will be complemented by a series of tables. Data synthesis will be based on the data extraction form generated with COVIDENCE. The unit of analysis will be individual patients in the included studies. If data is sufficient, a meta-analysis will be carried out calculating prevalence of dysphagia. The heterogeneity will be assessed with the I^2 statistic. Results of the analysis will be presented quantitatively with 95% confidence intervals and graphically in a forest plot. If possible, stratification by methodological quality will be conducted.

29. **Analysis of subgroups or subsets.** *mandatory*

If data is sufficient, subgroup analysis of different types of APD (PSP, MSA, DLB, CBD) will be conducted regarding dysphagia prevalence.

30. **Type and method of review.** *mandatory*

Systematic Review. Neurological

31. Language

English.

32. Country

Ireland

33. Other registration details

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34. Reference and/or URL for published protocol

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35. Dissemination plans

This systematic review is being conducted in the course of a master's thesis at the Department of Clinical Speech and Language Studies of Trinity College Dublin. Subsequently, a paper will be submitted to a relevant journal in this field and presented at conferences such as the European Society for Swallowing Disorders or the Dysphagia Research Society.

36. Keywords

Systematic review; meta-analysis; atypical parkinsonian disorders; atypical parkinson's disease; dysphagia; progressive supranuclear palsy; multiple system atrophy; dementia with Lewy bodies; Lewy body dementia; corticobasal degeneration; prevalence; risk factors; deglutition disorders;

37. Details of any existing review of the same topic by the same authors.

None

38. Current review status. *mandatory*

Review about to commence.

39. Any additional information.

None.

40. Details of final report/publication(s) or preprints if available.

n/a

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