The Long-Term Outcomes of Childhood Tourette Syndrome: A Systematic Review

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Background

Tourette Syndrome (TS) is a neuropsychiatric condition characterized by motor and vocal tics, which onsets in childhood and affects almost 1% of the population. Unfortunately, many individuals with TS experience significant distress, impairment, and a poor quality of life. Although much is known about its clinical presentation and evidence-based treatment in childhood, little information exists on clinical outcomes in adulthood for individuals with a childhood onset. Further information on adult outcomes of TS and its predictors is critical to identify new treatment targets and develop new interventions to help optimize therapeutic outcomes for patients with TS across the lifespan.
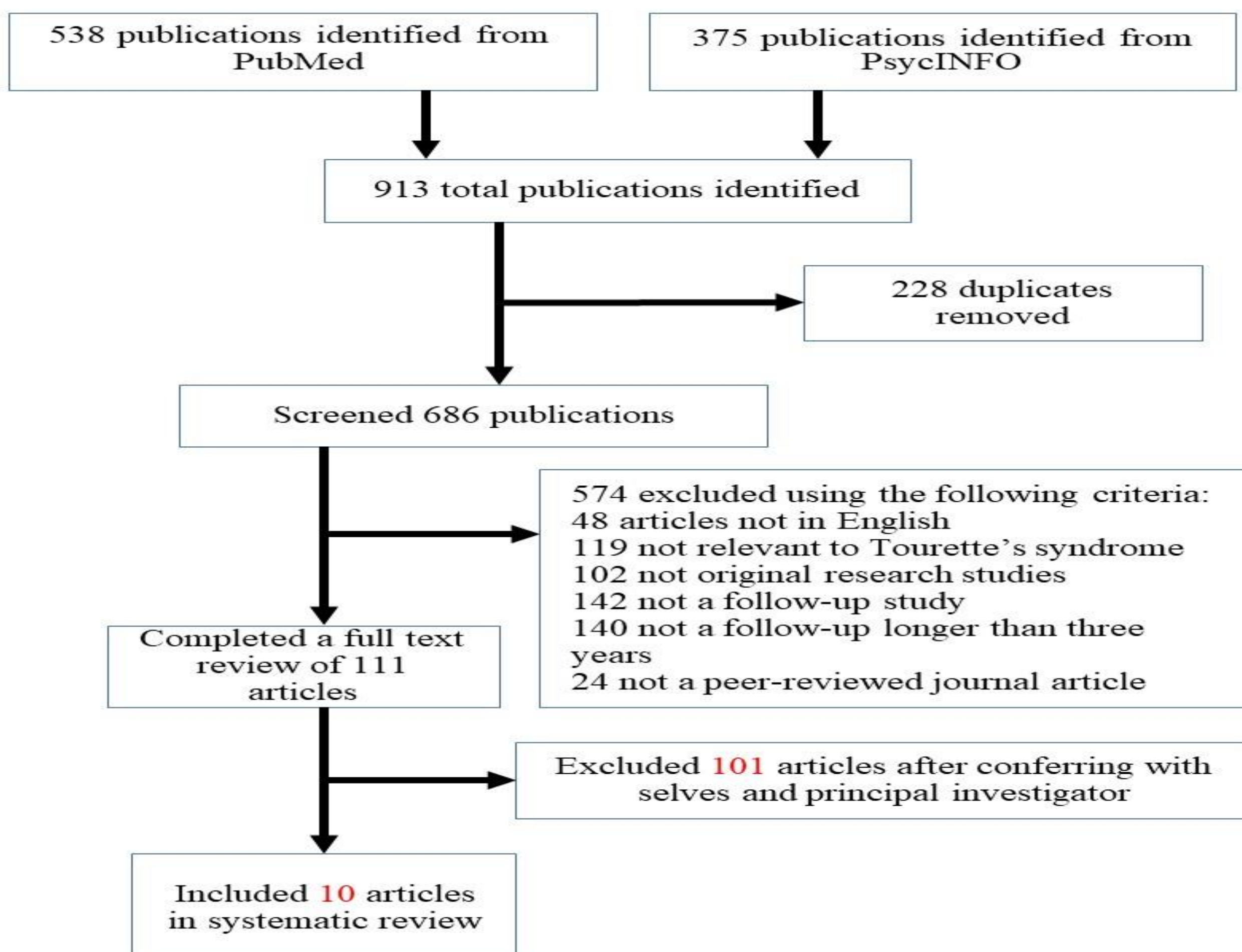
Objective

This study aimed to: (1) characterize tic remission rates into adulthood, and (2) determine childhood predictors of tic remission into adulthood.

Methods

- A systematic review was applied using in PubMed and PsycINFO databases with the search terms below.

Search Terms: (Tourette*) AND ((Long-term) OR (Prospective) OR (Follow-up))

538 publications identified from PubMed

375 publications identified from PsycINFO

913 total publications identified

228 duplicates removed

Screened 686 publications

574 excluded using the following criteria:
48 articles not in English
119 not relevant to Tourette's syndrome
102 not original research studies
142 not a follow-up study
140 not a follow-up longer than three years
24 not a peer-reviewed journal article

Completed a full text review of 111 articles

Excluded **101** articles after conferring with selves and principal investigator

Included **10** articles in systematic review

Results

- Articles that met inclusion criteria were examined, and key variables were extracted. When multiple articles reported data from the same study, findings were grouped together.

- Rates of tic remission in adulthood varied from 0% to 47.2%.

- While no difference in design methodology was observed, studies that used in-person assessment appeared to have lower tic remission rates.

- Common childhood predictors of tic reduction remission in adulthood included: Age of tic onset between 6-8 years, lower baseline tic severity, longer length of follow-up, and greater neurobiological/ neurocognitive functioning.

Study/Studies	Number of participants	Average length of follow-up (in years)	Design Methodology	Follow-up Assessment Methodology	Percentage of tic remission	Predictors of tic symptom reduction / remission in adulthood
Corbett et al. 1969	73	5.4	Retrospective	Self-report survey	40.0%	Longer length of follow-up, age of TS onset between 6-8 years, presence of sleep disturbances, and lower level of obsessionality
Bruun et al. 1976	78	2.7	Prospective	In-person clinician rating at follow-up	6.4%	None reported
Leckman et al. 1998	36	7.5	Prospective	In-person clinician rating (i.e., YGTSS)	47.2%	Lower worst ever tic severity
Burd et al. 2001	39	12.7	Retrospective	Phone administration of tic severity ratings	44.0%	No specific times of day (temporal tic morphology) or age when tic symptoms peaked
Pappert et al. 2003	31	12	Prospective	In-person video coded observation	10.0%	None reported. Interestingly, authors identified that adult patients who considered themselves "tic-free" were often inaccurate in their self-assessment as 50% had objective evidence of tics.
Bloch et al. 2005; Bloch, Peterson, et al. 2006; Bloch, Sukhodolsky, et al. 2006	46	7.5	Prospective	In-person clinician rating (i.e., YGTSS)	33.0%	Lower baseline tic severity, larger total caudate nucleus volume at follow-up, larger right putamen volume at follow-up, higher dominant hand, higher non-dominant hand, and higher bimanual Purdue pegboard scores
Shprecher et al. 2014	10	7.5	Prospective	In-person clinician rating (i.e., YGTSS)	0.0%	None reported
Byler et al. 2015	83	15	Retrospective	Self-report survey	13.6%	None reported
Groth et al. 2017; Groth et al. 2019	227	5.6	Prospective	In-person clinician rating (i.e., YGTSS)	17.0%	Lower baseline tic severity, and absence of either a family history of TS+OCD+ADHD or teasing
Lowe et al. 2018	45	>25	Retrospective	Self-reported CGI-Severity at follow-up	21.0%	Younger age of onset, and presence of either academic impairment and/or diagnosis of ADHD

Discussion

- Prior reports of tic remission vary greatly. Earlier reviews suggesting that full tic remission, partial tic remission, and tic persistence are near equivalent.

- These findings suggests that most patients exhibit tics into adulthood. In-person and observation assessment methods were more often associated with lower tic remission rates.

- Common baseline predictors of tic severity into adulthood included were identified and warrant further investigation.

- Limitations included small sample sizes in most studies.

Future Directions

- Conduct meta-analysis of tic remission rates to determine predictors and moderators across studies.

- Investigate baseline predictors to understand factors associated with tic remission in adulthood.

- Conduct large scale follow-up studies that include in-person assessments.

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