Original Article

Family History of Stuttering among Kurdish Children Who Stutter Near the Age of Onset

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Abstract

Background: The family history of stuttering is one of the most important factors for the prediction of recovery from stuttering. There is a high controversy about the rate of positive family history of stuttering among people who stutter (PWS). In the present study, the family history of stuttering in the first-, second-, and third-degree relatives was investigated among a group of Kurdish children who stuttered (CWS) close to the onset of the disorder.

Materials and Methods: One hundred and seventy-two CWS, including 46 females (24.60%) aged 2–5 years (mean age: 3.71 years, SD = 0.75) were consecutively recruited. They were first examined close to the onset of the stuttering. Data about current and former stuttering among first-, second-, and third-degree relatives were collected using interviews with both parents. Data was described and analyzed by independent sample *t* test or Chi-square.

Results: Among 172 CWS (mean age: 3.71 years, SD = 0.75), 119 (69.20%) reported a family history of stuttering. According to the result, the most family history of stuttering was reported among third-degree relatives (32.60%). Sixty-five participants (37.80%) reported just one relative with stuttering. CWS with a positive family history of stuttering had no significant difference with CWS without a positive family history of stuttering in sex ($P = 0.48$) and age of stuttering onset ($P = 0.96$).

Conclusion: The rate of positive family history of stuttering among Kurdish CWS is similar to previously reported data in the same age group. There was no significant association of family history of stuttering with sex and age on the onset of the disorder.

Keywords: Childhood-onset fluency disorder, family medical history, heredity, stuttering

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Introduction

Developmental stuttering is a speech-motor control condition that disrupts the normal flow of speech because of involuntary prolongation, block, repetition, and abnormal hesitation and pause.[1,2] The disorder often starts between 2 and 5 years of age, but few cases have been reported outside this age range.[3] It affects 4%–8% of children during early childhood but the prevalence will be gradually decreasing with age because of considerable recovery during childhood either spontaneously or with the aid of speech therapy services.[3] It is one of the most benign developmental disorders with 80% spontaneous recovery. The overall prevalence of the disorder has been reported as 1%.[4] But despite the high rate of spontaneous recovery and low overall prevalence, it puts lots of adverse impacts on the sufferer's psychological well-being, social activity, achievements, education, and occupation.[5,6]

Investigating different behavioral, cognitive, and neurological aspects of stuttering over several decades has been determining

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many pieces of the puzzle of stuttering and sometimes led to behavioral and cognitive methods for efficacious intervention. Although these methods seem to be effective,[7,8] a holistic scenery revealed a disappointing situation. In 1978, Porfert and Rosenfield emphasized the need to investigate organic causes of stuttering because of the lack of a decline in stuttering prevalence over decades despite the increased number of speech therapy clinicians.[9] Unexpectedly, new reports have not indicated any reduction in stuttering prevalence compared with older data despite the considerable enhancement of speech therapy services all over the world. Therefore, investigating the basic cause of stuttering remains the most important problem in the field. It seems that some important pieces of the puzzle have been remained unknown and some well-known pieces need to be rearranged.

Although the cause of the disorder has not been well-established yet, many psychological, familial, sociodemographic, and neurodevelopmental factors have been related to the higher risk of stuttering.[1,10] On the other hand, several demographics, linguistic, motor, and psychological factors have been linked to persistence or recovery from the disorder.[11-14] Gender and family history of stuttering are well-known factors in the assessment of the risk of stuttering and the recovery from the disorder.[3] Previous findings have confirmed the inheritance of stuttering.[10] A positive family history of stuttering has been related to the earlier age of onset, the persistence of, and recovery from stuttering.[15,16] It has been estimated that around 50% of people who stutter (PWS) have first- and second-degree relatives who also suffer from stuttering. The rate was even increased to 70% among studies that considered third-degree relatives too.[15,17] This is significantly higher than the incidence of stuttering reported in families of normally fluent controls.[18,19] The family history of stuttering is the best-known question that clinicians usually ask in the first examination worldwide, maybe without any idea about its clinical implications, or agreement about the exact rate of the positive family history among PWS for clinical advice and prognostic judgments.

Many studies have focused on the family history of stuttering with different methodologies and a wide range of results has been reported. Some studies used parental interviews to determine any family history of stuttering.[15,20-24] Some other studies used parental or self-reported questionnaires.[6,9,10] A few studies recruited more detailed familial pedigrees.[16,25,26] In addition, there has been a wide range of findings from 10% to 70% between different studies, and research has not completely clarified the rate of family history of stuttering among PWS so far. Some previous findings relied on data obtained primarily from adults who stutter (AWS) or a population that was far from the age of onset and maybe encountered several biases, including inadequate representation.[9,27-29] In addition, the finding that makes up our mind about the issue usually comes from the Western population and data from other parts of the world is scarce.[6,25] On the other hand, many studies did not consider all relatives and just gathered data about the immediate family members.[23] In the present study, the family history of stuttering was investigated, for the first time, among a group of Kurdish children who stutter (CWS), who were first examined close to the onset of the disorder. In addition, the data about family history of stuttering were collected for immediate and extended family members including the first-, second-, and third-degree relatives.

Materials and Methods

Participants

One hundred and eighty-seven Kurdish CWS including 46 females (24.60%) aged 2–5 years were consecutively recruited to participate in this cross-sectional study from May 2018 to June 2022. The first examination was done close to the onset of the stuttering in university-based or private speech therapy clinics in Kermanshah, west of Iran. The sample was considered close to the onset of the disorder to reduce the age variability and inadequate representation biases. Participants were included in the study if the diagnosis was confirmed by parents and two speech-language pathologists. In addition, the time interval between the onset of stuttering and data collection should not exceed 6 months. Participants were excluded from the study if they had a history of seizure, brain trauma, diagnosed metabolic condition, and diagnosis of Down syndrome. Unsolved disagreement between speech-language pathologists about stuttering diagnosis was excluded. If speech dysfluency was a sign of other condition such as cluttering and Tourette's syndrome, the participant was excluded too. Data about the date of birth, age, gender, and age of the onset of stuttering was collected by asking parents and the same was recorded in a checklist. Two males with the final diagnosis of cluttering and Tourette syndrome were excluded in the first step. In addition, one male who was born from a donated embryo and two participants (one male and one female) who resulted from donated eggs were also excluded. Data of 10 participants were also ignored because information about the family history of stuttering from one parent was unavailable. Finally, 172 CWS were included in the study.

Stuttering diagnosis

All participants were admitted or referred to the clinics as having stuttering. Participants were involved in the study if stuttering was confirmed by both parents and by two speech-language pathologists who were experienced in the field of stuttering based on spontaneous and conversational speech samples. If there was a disagreement between the two speech and language pathologists, the final decision was made by discussing the condition and making a final diagnosis together. Finally, no disagreement occurred between clinicians.

Family history data

Data about the family history of stuttering was collected using interviews. Clear criteria for definition of stuttering were gotten to parents with examples. It included part-word repetition, blocks, prolongation, and abnormal silent or audible pause. Both parents were interviewed separately by an expert

speech-language pathologist and were told to report any history of stuttering (both recovered and persistent stuttering) among first-degree biological relatives of their kids; including parents and siblings. Then any history of recovered or persistent stuttering among second biological relatives, including uncles, aunts, nephews, nieces, and grandparents was asked one by one. In addition, data about the history of stuttering among third-degree relatives, including cousins and great grandparents were also collected from both parents.

In case only one parent was available, the data on the family history of stuttering was obtained from the available parent. In Kermanshah, Iran, where the study took place, there is very strong interaction within families, and usually, one spouse can provide such information on the family of the other spouse. In the case that the parent could not provide data on the other parent's family history, he/she was asked to call his/her spouse and ask for the information. If only one parent was available and could not finally give information on the family history of the other parent, the participant was excluded. If at least one first-, second-, or third-degree relative was reported to have ever stuttered either recovered from or persistent on the disorder, the history was considered positive.

Statistical analysis

Data was described in tables. Statistical comparison of the demographic variable between different groups was performed by either independent sample *t* test or Chi-square using SPSS®, version 28.0 (IBM Corporation, Armonk NY; USA) for Windows Microsoft.

Results

Among 172 CWS (mean age: 3.71 years, SD = 0.75), 119 (69.20%) reported to have at least one stuttering relative among first-second and/or third-degree family members. According to the result, the most family history of stuttering was reported among third-degree relatives, whereas 28 participants (16.20%) reported a positive family history among first-degree relatives [Table 1].

Results indicate that 37.8% of CWS had just one relative who ever stuttered. Then, the number of CWS with two or more relatives who have stuttered gradually decreased. Finally, just one CWS had five or six relatives with stuttering [Table 2]. Among 119 participants with a family history of stuttering, 45 (37.8%) reported the history among the mother's relatives, 51 (42.9%) among the father's relatives, and 20 (16.8%) among both parents' relatives.

The age of stuttering onset was not significantly different between CWS with a family history of stuttering (Mean, 40.46; SD, 12.05 months) and CWS without a family history of stuttering (Mean, 40.55; SD, 16.32 months) ($P = 0.96$). The distribution of sex was not significantly different between the two groups. There were 90 males (75.6%) among CWS with a positive family history in comparison with 41 males (77.4%) in the group without a family history of stuttering ($P = 0.48$, $X^2 = 0.06$). In addition, a positive family history of stuttering was reported among 90 (68.70%) male participants and 29 (70.70%) female participants with no significant difference.

Table 1: Family history of stuttering among Kurdish CWS close to the age of onset

	Frequency	Percent
No family history	53	30.8
First-degree relatives	15	8.7
Second-degree relatives	20	11.6
Third-degree relatives	56	32.6
First-, second-, and third-degree relatives	3	1.7
First- and third-degree relatives	6	3.5
Second and third-degree relatives	15	8.7
First- and second-degree relatives	4	2.3
Total	172	100.0

CWS, children who stuttered

Table 2: The number of relatives with a family history of stuttering among Kurdish CWS near the age of onset

	Frequency	Percentage
No family history	53	30.8
One relative	65	37.8
Two relatives	31	18.0
Three relatives	15	8.7
Four relatives	6	3.5
Five relatives	1	0.6
Six relatives	1	0.6
Total	119	100.0

CWS, children who stuttered

Discussion

The family history of stuttering has gotten specific attention from the beginning of stuttering research and is usually asked both in clinical and research contexts. It has an important predictive value for recovery from stuttering among CWS specifically near the age of onset.[16] Many studies focused on family history, either epidemiologically or as a predictive value for recovery from the disorder using different methods for data gathering and on different age groups. Studies reported a wide range of estimates for positive family history of stuttering among PWS. The results are different based on the age of participants, the time from the onset, the methods, and the level of relativeness. In addition, rare data were reported from non-Western communities. In the present study, for the first time, data about the family history of stuttering among Kurdish CWS were investigated by a precise method near the age of onset.

Based on the results, 119 of 172 CWS (69.20%) were reported to have at least one stuttering relative among first-second and/or third-degree family members. This overall rate is similar to studies that recruited the same age group using parental interviews for data collection of family history.[15,19,21,30]

The results are also similar to the study by Ambrose *et al*., 1993 who investigated the same age group using a more precise family pedigree method.[26] In addition, other studies which investigated the same age group with methods other than interviews reported similar results.[10,31] It seems that regardless of method, the results of studies that recruited CWS close to the age of onset are similar. Though some studies that used questionnaires reported a lower rate of positive family history of stuttering among CWS.[28,32,33] Generally, the present study provided nice confirmatory evidence of familiality that is consistent with previous family and pedigree studies. Table 3 summarizes the finding, sample size, sample's age, and methods for gathering data about family history from previous investigations and the present study.

The chance of persistence from stuttering is higher among PWS with a positive family history compared with those who have no family history of stuttering.[16] Therefore, we may expect a higher rate of positive family history among older PWS. However, studies did not confirm the expectation. Based on the result of the present study and similar studies that recruited a younger population, the rate of positive family history is higher among studies that recruited participants near the age of onset.[10,15,19,26,30,31] On the other hand, studies that recruited older populations reported a lower rate of positive family history of the disorder.[9,27,34] Based on these findings, the farther the age of the sample was from the age of the stuttering onset, the lower the rate of family history of the disorder. It seems that studies that relied on data collected far from the age of onset may encounter inadequate representation and therefore underestimate the positive family history of stuttering.[9,27-29] Clinically, this issue should be considered when older PWS, for example, adolescents and adults, are examined. Clinicians are recommended to use more precise methods such as family pedigree instead of just asking about "any positive family history." Reilly *et al*. (2009) indicated that the results of a positive family history of stuttering varied based on the time of data collection. In their study, an extremely low number of participants reported a positive family history if data were collected before stuttering onset. But, when family history data were collected after stuttering onset, 71 (51.80%) participants reported a family history of the disorder.[21] Similarly, studies that investigated school-aged CWS[6,20,27,34] reported a lower positive family history compared with studies that recruited preschool CWS.[10,15,19,21,22,26,32,35] In addition, the present study confirmed that using a comprehensive interview with both parents could give information about family

Table 3: Summarization of studies which reported family history data of stuttering among PWS

	Authors	Sample size	Age	Method	Rate of positive family history of stuttering (%)
1.	Ambrose *et al*., 1993	69	2 to 6 years	Pedigree	71.00
2.	Devi *et al*., 2018	180	2.5 to 16 years	Pedigree	56.00
3.	Okasha *et al*., 1974	79	6–12 years	Interview	18.00
4.	Kiziltan *et al*., 1996	23	14 to 36 years	Interview	69.50
5.	Mansson, 2000	12	5	Interview	67.00
6.	Reilly *et al*., 2009	137	3	Interview	51.80
7.	Buck *et al*., 2002	61	2.7 to 6.5 years	Interview	72.10
8.	Howell *et al*., 2011	132	8 to 10	Interview	48.50
9.	Mohammadi *et al*., 2016	22	7 to 14 years	Interview	90.00
10.	Mohammadi *et al*., 2020	83	3 to 9 years	Interview	66.30
11.	Present study	172	3-6	Interview	69.20
12.	Porfert and Rosenfield, 1978	131	median age was 20.8 years	Questionnaire	22.10
13.	Janssen *et al*., 1990	78	8 to 17 years	Questionnaire	39.74
14.	Cavenagh *et al*., 2015	42	2 years 6 months to 5 years 10 months	Questionnaire	67.70
15.	Abou Ella *et al*., 2015	90	7 to 12 years	Questionnaire	31.11
16.	Ghazavi, *et al*., 2020	30	4 to 6 years	Questionnaire	43.30
17.	Bleek *et al*., 2011	87	Mean age 34.9 years	Self-report	58.60
18.	Howell *et al*., 2018	15	22 to 58 years	Self-report	66.00
19.	Boyce *et al*., 2022	987	7 to 93 years	Self-report	49.90
20.	Choi *et al*., 2018	25	36 to 71 months	Caregivers report	24.00
21.	Poulos and Webster, 1991	169	14 to 60 years	Asking	66.00
22.	Donaher and Richels, 2012	36	3.9 to 17.2 years	Asking	61.00
23.	Walsh *et al*., 2021	51	41 to 68 months.	Asking	35.00
24.	Singer *et al*., 2022	67	3 to 5 years	Asking	50.74
25.	Druker *et al*., 2019	185	2.5 to 6.2 years	Detailed case history	82.70
26.	Ismail *et al*., 2017	30	8 to 18 years	Not mentioned	23.30
27.	Ozgur 2019	64	Mean age; 7.36±3.76 years)	Not mentioned	21.90

Studies were sorted based on the methods of data collection about family history. PWS, people who stuttered

history as accurately as family pedigree as a more rigorous time-consuming method.

In the present study, the more distant the family relatedness, the more the family history of stuttering. The most family history of stuttering was reported among third-degree relatives, comprising 46.50% of participants. While just 28 participants (16.20%) reported a positive family history in first-degree relatives and 24.30% among second-degree relatives. The figure differed from previous studies that reported the highest positive family history among first-degree relatives.[10,36] Some parents might not have disclosed their stuttering in the past or mild present stuttering in themselves. This condition may be also arisen from the difference in family size between societies. The size of families in Iran has considerably decreased in the past decades. Therefore, children have a large community as their second- and third-degree relatives but a small, often one-sibling family. It seems that considering all levels of relativeness is very important when data about a family history of stuttering is collected in both clinical and research settings. Studies that considered only first-degree relatives reported a lower rate of family history.[27,33]

Some studies reported a more positive family of stuttering among male PWS compared with females.[10,26] Other studies reported a more family history of stuttering among female PWS compared with male PWS.[15,37] No significant association between the sex and family history of stuttering was observed among the Kurdish CWS. A positive family history of stuttering was reported among 90 (68.70%) male participants and 29 (70.70%) female participants with no significant difference. The controversies about the association between the gender of affected people and the family history of stuttering have remained, and further investigations are needed for understanding this association.

In the present study, the age of stuttering onset was approximately 40 months, which was higher than the findings reported from Western countries.[15,36] There is no national or local precise screening system for speech-language development in Iran, and the exact age of the onset of stuttering may be missed by parents in the present study, especially if the extreme fluctuations of disfluencies during the onset of the disorder are considered. There was no association between the family history of stuttering and the time of the disorder's onset in the present study. Some previous studies reported a slight nonsignificant impact of positive family history of stuttering on the earlier onset of the disorder.[15] But it seems that a positive family of stuttering has no association with the age of onset of the disorder.

The present finding should be interpreted with some caution. First of all, the size and composition of the families has not been accounted for. The second concern is about the informant-response approach. People may not be certain about their older adult's childhood stuttering. Third, some cultural factors may directly impact the findings of the study. Parents in Kermanshah may not generally be open and comfortable discussing the topic of stuttering within the family. In addition, there is a stigma attached to stuttering in Kurdish culture, as is the case in other cultures.

Conclusion

The rate of positive family history of stuttering among Kurdish CWS is consistent with existing data if the age of participants and method of data collection is considered. Most of the family's history of stuttering was reported among third-degree relatives. There was no significant association between sex and family history and between the age of onset and the family history of stuttering

Ethical approval

Written informed consent was obtained from the participant's parent to participate in the study. The study protocol was reviewed and approved by the Ethics Committee of the Kermanshah University of Medical Sciences (KUMS), approval no. [IR.KUMS.MED.REC.1401.261].

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Conflicts of interest

There are no conflicts of interest.

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