



Published in final edited form as:

*Epilepsy Res.* 2009 March ; 84(1): 1–5. doi:10.1016/j.eplepsyres.2008.11.014.

## The Accuracy of Self-Reported History of Seizures in Danish, Norwegian and U.S. Twins

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### Summary

Questionnaire surveys provide an efficient means of identifying potential seizure cases in large population-based cohorts. Concerns exist, however, with regard to the reliability of self-reported information both with respect to the validity of the results obtained and with regard to the usefulness of this approach in identifying true cases. Information on history of seizures obtained by questionnaire from members of 47,626 twin pairs included in the Mid-Atlantic (MATR), Danish (DTR) and Norwegian (NTR) Twin Registries was verified using medical records and detailed clinical and family interviews. The accuracy of these reports was assessed. Self-reported epilepsy was verified in 81.9% of twins overall (86.1% (DTR), 75.6% (NTR) and 80.7% (MATR)). However, when both pair members reported a history of epilepsy in the affected pair member, epilepsy was verified in >90% of cases. Among MATR twins with a verified history of epilepsy, 21.5% reported other seizures but not epilepsy and 18.5% of verified Norwegian epilepsy cases reported no history of epilepsy themselves and were identified only through their co-twin. The results of this study indicate that the accuracy of self-reported epilepsy and febrile seizures among those who provided information on health history was high across all populations. However, the relatively large percentage of twins with a verified diagnosis who did not acknowledge epilepsy suggests that the frequency of epilepsy may be under-estimated in self-reported samples.

### Keywords

self-reported seizures; questionnaire surveys; twin studies

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## 1. Introduction

The creation of large population-based samples of individuals with a history of seizures can be approached in two ways; either prospectively, by following a large cohort of subjects and identifying those who have a seizure or develop epilepsy through surveillance of hospital admissions or physician records, or retrospectively using surveys in which history of seizures is self-reported. While prospectively identified cases are most likely to be accurately diagnosed, prospective studies are labor intensive, can be very expensive, and omit cases either who do not receive treatment for seizures or receive treatment from a provider that is not part of the surveillance system. Further, the available sample is limited to those with active seizures or epilepsy. This can have significant implications with regard to sample size.

Questionnaire surveys provide an efficient and inexpensive means of identifying seizure cases from members of large population-based cohorts. Concerns exist, however, with regard to the accuracy of self-reported information, both with respect to the validity of results obtained and with regard to the usefulness of this approach in identifying true seizure cases. Although adults with epilepsy have been found to report reasonably accurately on epilepsy in their parents and siblings (Ottman, et al., 1993), little information is available about the accuracy of self-reported seizures in unselected samples. We investigated the accuracy of self-reported history of seizures in subjects from three large population-based twin registries who provided information about their medical histories.

## 2. Materials and Methods

Twin pairs reporting a history of seizures were ascertained from among participants in the population-based Mid-Atlantic (MATR), Norwegian (NTR) and Danish (DTR) Twin Registries based upon a positive response to queries included in a mailed health history questionnaire survey. The MATR includes all twin pairs born in Virginia and North Carolina between 1915 and 1997, the NTR all same-sex pairs born in Norway between 1915 and 1979, and the DTR all pairs born in Denmark between 1952 and 1982. Participants in the registries were ascertained from birth records. The surveys queried about a number of health problems and included four questions related to seizures that included queries about a history of “epilepsy”, “febrile seizures”, “other seizures” or “staring spells” either in the twin or their co-twin. MATR and NTR twins were surveyed twice between 1982 and 1999 while DTR twins were surveyed in 1996. MATR and NTR pairs were asked to provide information on history of seizures in themselves and their co-twin. Due to restrictions on the type of information that could be collected imposed by the Danish Ethics Committee, Danish twins were not asked to provide information on the seizure history of their co-twin.

As an initial screen of questionnaire responses, twins or their parents, (in the case of juveniles), who reported a positive seizure history on the survey were contacted by telephone to verify their response and to exclude those whose medical history was not consistent with a history of seizures. The telephone interview was constructed in such a way as to identify breath-holding in children and syncopy in adults and carried out by trained study personnel. Twins where a history of seizures could be excluded either because they now denied ever having had a seizure or on the basis of the additional information obtained were omitted from further study. Twins, where a history of seizures could not be excluded, were asked to provide detailed information about seizure history, including age at onset, seizure characteristics (clinical signs, length, and recurrence), medication history, and family history, in addition to, a blood sample for DNA extraction and a signed release for pertinent medical records. Epileptologists, who were blinded to subject identity, then verified these cases using information contained in medical records and from detailed family interviews as outlined in Figure 1. Self-/co-twin-reported cases were

classified as verified seizures, possible seizures, another medical condition (e.g., syncope in adults or breath-holding in children), a mistaken report, or as impossible to verify based upon available information.

The institutional review board at each of the institutions involved in the study has reviewed and approved the study protocol.

### 3. Results

Information on seizure history was available on 81,798 twins included in 47,626 pairs (Table 1). Since twins living in Denmark and Norway must have a current address on file with authorities in order to receive health care, current addresses were available for 80% of twins. It was much more difficult to trace twins born in the MATR catchment area since the only information available was name and date of birth and place of residence of the twins' parents at the time of their birth. This is reflected in the relatively small proportion of pairs among those born in the MATR catchment area that could be traced. Overall, among twins who could be traced, a little less than half of those contacted agreed to become a twin registry member and provided health survey information. The Norwegian and Danish Twin Registries were much more successful in recruiting twins than the MATR as is reflected in the increased percentages of twins who were willing to participate in twin-related research projects. Although 4,231 twins reported a history of seizures of some type, this study is limited to the 2,645 pairs not requiring additional tracing due to funding limitations and those who were willing to participate in the study. All twins who reported a history of epilepsy or febrile seizures, where current contact information was available or could be provided by a co-twin, were included in the study. No effort was made to verify self-reports of other seizures or staring spells by Danish twins or of staring spells in Norwegian twins after initial efforts indicated that few twins reporting these conditions had actually had an epileptic seizure of any type. As shown in Table 1, among those who reported a history of seizures on the survey, the percentage of twins who did not wish to participate in the study when asked to do so, ranged from 14.7% in Norway, to 27.6% in Virginia/North Carolina, to 28.2% in Denmark.

The accuracy of self-reported history of seizures was estimated for each population. Table 2 provides the percentage of non-duplicated self-/co-twin reported seizure cases partitioned by type verified in each sample. Self-reported information was most reliable in the Danish sample whose ages ranged from 30–50 years and least reliable in the Norwegian sample (age range 42–80 years) when queried. In most cases, it was possible to verify the occurrence of seizures in twins reporting a history of epilepsy or febrile seizures, but verification was much less likely if other seizures or staring spells were reported. A significantly greater number of self-reported seizure cases were verified in Denmark compared to Norway (epilepsy and febrile seizures,  $p < 0.01$ ) and the US (febrile seizures,  $p < 0.01$ ).

As shown in Table 3, a higher percentage of epilepsy cases were verified compared to self-reports alone when both pair members reported the case to be affected. In two of the three unverified epilepsy cases where both twins reported the affected twin to have epilepsy, the twin was found to have had febrile seizures. The percentage of febrile seizure cases verified when both pair members reported the case twin to be affected was also significantly increased over that based on self-reports alone in the Norwegian, but not in the MATR sample. Both of the reported unverified febrile seizure cases in the Norwegian sample were found to be as cases of epilepsy, while two of five MATR cases that could not be verified as febrile seizures were found to be epilepsy and two were declared to be mistakes by the twins. Approximately five percent of verified epilepsy cases in the MATR sample and 18.5% of verified epilepsy cases in the Norwegian sample were twins who denied any history of seizures in the survey and were ascertained through their unaffected co-twin who reported them to be affected.

The Danish sample was characterized by significantly smaller percentages of both self-reported epilepsy and febrile seizure cases that were later denied upon telephone contact compared to both the Norwegian and American samples where no significant differences in the proportions denying either epilepsy or febrile seizure were observed (Table 4). In contrast to epilepsy, where similar percentages of those whose reported epilepsy was found to be another medical condition were observed in the three populations, a significantly lower percentage of self-reported Danish febrile seizure cases was found to be another condition ( $p < 0.01$ ).

#### 4. Discussion

The primary focus of this study was an assessment of the accuracy of self-reported information history of seizures provided by twins, who were unselected for history of seizures. Because information on history of seizures was unavailable for a large number of the twins born in the catchment areas of each of the twin registries included in this study, either because they could not be traced or did not wish to participate in a twin registry, it was not possible to identify all twins who had a history of seizures either within or across registries. For this reason, non-responder bias is likely to be an issue and no attempt was made to estimate the prevalence of seizures in the base twin populations. Non-responder bias is unlikely to be an issue with regard to the whether or not self-reports of a positive history of seizures are accurate. It is also unlikely to be an issue with regard to the accuracy of positive reports of a history of seizures in a twin by their co twin.

The accuracy of self-reported epilepsy and febrile seizures was high across populations, with that of seizures in one twin reported by both pair members being very high. In most cases, a history of seizures could be verified in twins reporting either epilepsy or febrile seizures, but verification of positive seizure history was much less likely if these seizures were reported as 'other seizures' or 'staring spells'. The term 'other seizures' was used in order to identify those with a history of epilepsy who might not answer positively to a direct query about epilepsy. The term 'staring spells' was used following the method of Santilli and Dreyfus (1979) to elicit identification of absence, complex partial and other non-convulsive epileptic activity. While the inclusion of these terms in the English version of the questionnaire permitted the successful identification of twins with a history of epilepsy that, otherwise, would not have been found, they were not meaningful descriptors for epileptic seizures in either Norway or Denmark. The Norwegian and Danish languages simply could not convey the meaning of these terms in a manner analogous to that in English. This resulted in few positive responses to the questionnaire query regarding history of staring spells and only eight positive responses to the 'other seizures' item in the Danish sample. Although larger numbers of Norwegian twins reported a history of "other seizures", only a small proportion of these could be verified as epileptic. While only 41.8% (66/158) of self-reported 'other seizures' in the US sample were verified as epileptic, this group accounted for 21.5% of those with verified epilepsy and 7.1% of those with verified febrile seizures. Since these twins are limited to those who indicated a history of 'other seizures' but not "epilepsy" or 'febrile seizures', this group represents a subset of the American sample whose history of epileptic seizures would have been missed had this question not been included in the questionnaire survey.

Ten percent or less of epilepsy self-reports and 7.9% or less of reports of febrile seizures were determined to be false positives. The degree to which epilepsy was under-reported in this sample is more difficult to determine. However, this does appear to be an issue given the number of self-reported cases that were later denied upon direct contact with the twin, the percentage of verified Norwegian and American epilepsy cases that were ascertained through reports by the unaffected co-twin and the relatively high frequency of twins with verified epilepsy who reported this history not as a history of epilepsy but as a history of other seizures. A history of epilepsy can fail to be reported either because of a lack of awareness of the disorder

or because of a desire to withhold this information because of a fear of stigmatization. It would appear that the later may be the more likely explanation in this case, given that >25% of verified epilepsy cases were twins who either reported no history of seizures of any type or reported a history of other seizures but not epilepsy in the American sample and 18.5% of verified Norwegian epilepsy cases reported no history of seizures of any type in themselves and were ascertained through surrogate reports provided by unaffected co-twins. This is consistent with the findings of perceived stigma reported by other authors (Scambler and Hopkins, 1986, Jacoby, 1994, Jacoby 1999, Scambler, 1994).

Differences between populations in the percentage of reports verified demonstrates the difficulty encountered in assessing retrospective reports of epileptic seizures that occurred in subjects more than 40 years ago. Seizure history could not be verified in many cases because the relevant medical records were either unavailable or inadequate, and relatives who may have observed the seizures are no longer living. These factors were less problematic in the younger Danish sample as reflected in the higher percentages of self-reported seizure cases verified.

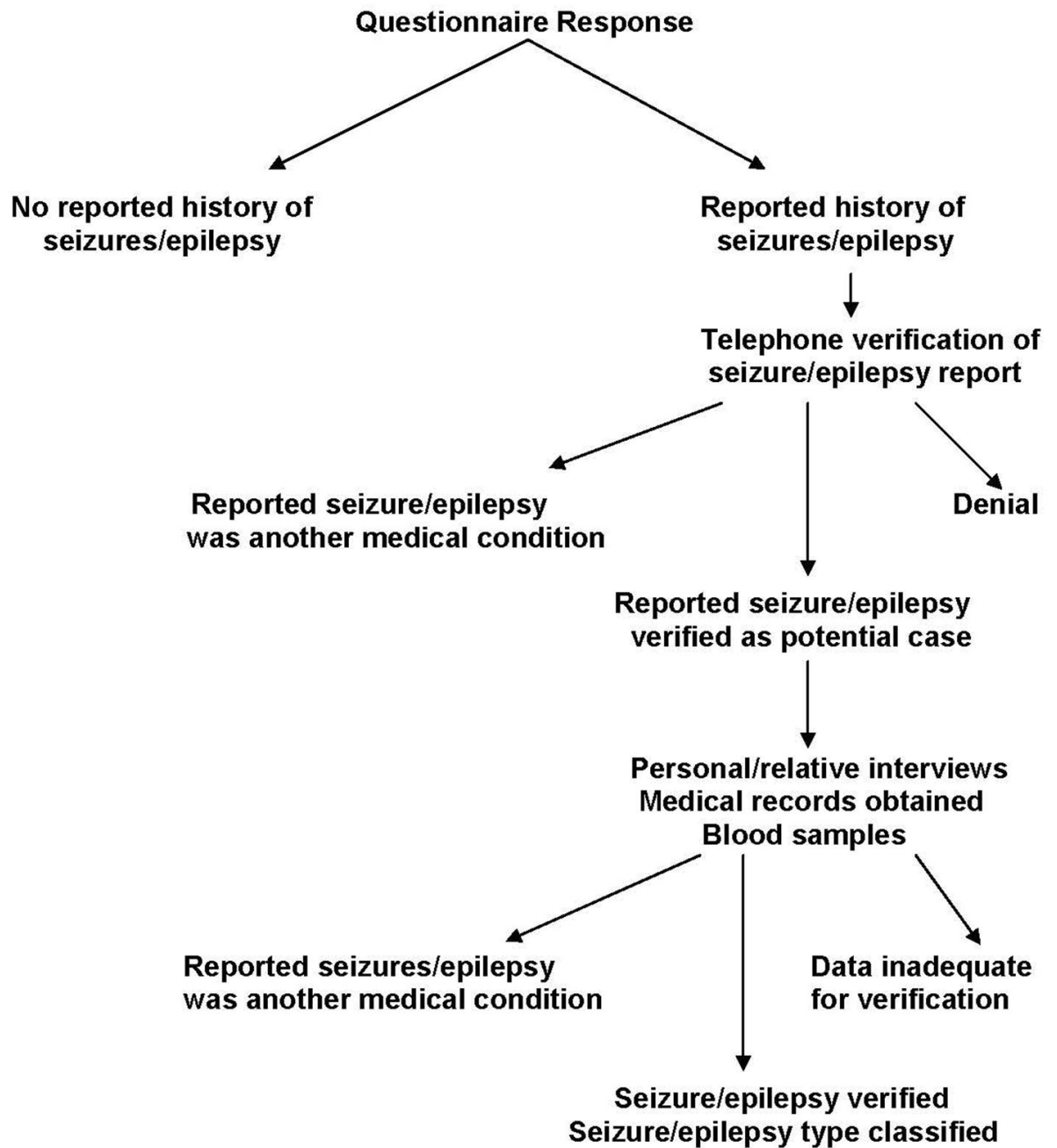
The results of this study document both the accuracy of self-reported information on epilepsy and the difficulty in identifying those with a history of seizures using retrospective information. The proximity of data collection to seizure occurrence was an extremely important determinant of the ability to verify self-reported epilepsy. Even in this highly motivated and somewhat self-selected sample of twin registry participants, denial of epilepsy history occurred. Multiple factors are likely to contribute to this denial.

## Acknowledgements

This work was supported by the National Institute of Neurological Disorders and Stroke (NS31564).

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**Figure 1.**  
Procedure used in verifying reported seizures/epilepsy

**Table 1**

Sample characteristics by ascertainment source

	<b>DTR (1952 – 1982)</b>	<b>MATR (1915 – 1997)</b>	<b>NTR (1915 – 1960)</b>	<b>Total</b>
Total pairs born in catchment area	20,888	131,470	23, 917	176,275
Pairs traced	18,512	44,729	19,101	82,342
Twins providing health histories	29,179	29,789	22,830	81,798
Twins reporting history of seizures	1,309	1,552	1,370	4,231
Twins contacted	1,124	867	654	2,645
Twins not wishing to participate	317	239	96	652
Twins/pairs evaluated	807 twins/685 pairs	628 twins/531 pairs	558 twins/498 pairs	1993 twins/1714 pairs
Seizures verified Twins/pairs	724 twins/617 pairs	413 twins/360 pairs	349 twins/305 pairs	1,486 twins, 1,282 pairs

(DTR= Danish Twin Registry; MATR = Mid-Atlantic Twin Registry; NTR=Norwegian Twin Registry)

\* Date of birth



**Table 2**

Percentage of self-reported seizures verified partitioned by population and report

Report	Reports verified			Overall
	DTR	MATR	NTR	
Epilepsy	86.1%	80.7%	75.6%	81.9%
Febrile Seizures	92.8%	79.4%	75.2%	86.7%
Other Seizures	88.9%	41.8%	18.7%	35.3%
Staring Spells	-	16.2%	-	16.2%
Total	89.9%	62.1%	63.6%	75.1%



**Table 3**  
Percentage of self- and co-twin reported cases of epilepsy and febrile seizures verified

Report	MATR			NTR		
	Self	Co-Twin	Both*	Self	Co-Twin	Both
Epilepsy	80.7%	72.7%	94.4%	75.6%	97.4%	98.0%
Febrile Seizures	78.3%	53.1%	83.9%	76.3%	94.7%	95.1%

\* Both twins reported the affected twin to be affected.

**Table 4**  
 Summary of validation of non-duplicated epilepsy/febrile seizure self-reports

Reported seizure	Epilepsy					Febrile Seizures				
	DTR	MATR	NTR	Overall	DTR	MATR	NTR	Overall		
Verified	86.1%	80.7%	75.6%	81.9%	92.8%	79.4%	75.2%	86.7%		
Denied	0.6%	8.6%	4.5%	3.8%	5.2%	10.3%	11.3%	7.5%		
Other medical condition	6.7%	10.0%	0.5%	7.1%	0.9%	7.9%	7.8%	3.7%		
Inadequate information	6.6%	0.7%	13.4%	7.2%	1.1%	2.4%	5.7%	2.1%		