

# **Dysphagia screening with the Dutch Signal List in adults with intellectual disabilities is not valid when compared to the Dysphagia Disorders Survey**

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## **Abstract**

*Background* Swallowing difficulties can lead to major health problems. Previous research shows that up to 50% of elderly with intellectual disabilities (ID) have dysphagia. Dysphagia is often not recognized, which underlines the importance of screening. A screening tool for caretakers, the Signal List, was developed.

*Aim* With this validation study we aim to determine the criterion validity of the Dutch Signal List for screening of dysphagia in adults with ID.

*Methods* The Dysphagia Disorders Survey (DDS) is a standardized screening tool often used by speech therapists for screening for dysphagia in people with ID. We assume the DDS as the golden standard for the screening of dysphagia. In this multi centre study we determined sensitivity and specificity of the Dutch Signal List compared to DDS in a population of adults with ID in residential setting.

*Results* The sensitivity of the Dutch Signal List compared with the DSS is 55%; the specificity 58%. 45% of the scores are false negative and 42% percent are false positive.

*Conclusions* In its current state, the Dutch Signal List (compared to the DDS) is not a valid screening tool for the screening of dysphagia in adults with ID. The Signal List is practical and easy to use. Future research should focus on adjusting the Signal List prior to further validation studies.

## **Introduction**

Swallowing requires multisystem skills. Any imbalance in sensory perception, autonomic integration and neuromuscular control in the area of the mouth, pharynx and oesophagus can lead to dysphagia ([Gallagher 2009](#)). According to the Royal College of Speech and Language Therapists, dysphagia is defined as 'eating and drinking disorders which may occur in the oral, pharyngeal and oesophageal stages of deglutition ([RCSLT 2006](#)). Dysphagia is associated with neurological, anatomical, gastrointestinal and pulmonary disorders ([Chadwick 2009](#); [Dobbelsteyn 2005](#); [Lazenby 2008](#)) which are more prevalent in

the population of people with intellectual disabilities (ID) ([Straetmans 2007](#)) when compared to the general population. Moreover, medication use is known to be a risk factor for dysphagia, especially antiepileptic drugs which are frequently used by people with ID ([Bastiaanse submitted](#); [Gallagher and Naidoo 2009](#)). Dysphagia can lead to a variety of health problems, including malnutrition, dehydration, and aspiration pneumonia as well as to choking and death ([Chadwick and Jolliffe 2009](#); [Samuels 2006](#)). The consequences of dysphagia demand adequate and timely intervention. In addition to the mentioned medical consequences, dysphagia can also cause behavioral disorders (rapid eating, food phobias), physiological disorders (reflux) and developmental delay disorders (poorly developing chewing skills) ([Sheppard 2013](#)). Screening for dysphagia is thus of great importance. However, valid and practical screening tools are not available, which is why in this study we provided information on the validity of a new screening tool for dysphagia.

The prevalence of dysphagia in people with ID widely ranges from 9% to 99% ([Calis 2008](#)). Prevalence studies were only conducted in selected groups, such as children with profound and multiple handicaps and adults over 50 years of age ([Calis 2008](#); [Chadwick and Jolliffe 2009](#)). In a recent Dutch study amongst people with ID of 50 years and over, moderate to severe dysphagia was found in 51.6% ([Bastiaanse submitted](#)).

The diagnosis of dysphagia is often missed amongst people with ID, while caregivers may not be aware of the symptoms and the consequences of dysphagia ([Calis 2008](#); [Helder 2010](#); [Leeuwen 2007](#)). In addition, Thacker et al. described an underdiagnosis of dysphagia ([Thacker 2008](#)). It was mentioned that caregivers and clinicians should benefit from awareness of choking risks and screen all people with ID on these risks during a routine health assessment. Screening for dysphagia will reduce secondary co-morbidity ([Calis 2008](#); [Leeuwen 2007](#)).

In order to identify dysphagia, different assessments can be used. Assessments are classified as screening tests, clinical assessments, severity scales and risk assessments. Videofluoroscopy is considered to be the gold standard ([Helder 2010](#); [Penning 2003](#); [Sheppard 1991](#)), while it is being criticized. Not only is videofluoroscopy time-consuming and expensive, the necessity to visit a hospital, to cooperate and sit up during measurements can be a burden for people with ID.

In this validation study of the Signal List, the Dysphagia Disorders Survey (DDS) was used to determine the risk of dysphagia. The DDS is a standardized observation of swallowing and feeding functions. The DDS requires training of professional observers (such as speech

therapists) ([Sheppard 1991](#); [Sheppard 2013](#); [Sheppard 1988](#); [Sheppard 1995](#)). In comparison with other assessments and screening tools the DDS gives well detailed information about the pathophysiology of swallowing. The DDS is non invasive, cheap and regularly used in daily practice and the field of dysphagia research with people with ID. We assume the DDS as the golden standard for the screening of dysphagia, because it is a common used tool in practice.

Recently, a short Dutch observational screening questionnaire (Signaleringslijst Verslikken, further referred to as Signal List) has been developed. It can be completed by untrained staff (like caregivers). Helder ([Helder 2010](#)) developed this Signal List and tested it in a group of people with ID (n=83), aged 50 years and over. She compared the outcome with the DDS. Due to the restricted study population and the fact that Helder did not publish her data, a validation study is necessary prior to daily use of the Signal List.

With the intention to lower the underdiagnosis of swallowing disorders, the aim of this study is to determine the validity and specificity of the Signal List for screening for dysphagia in adults with ID. With this information, the utility and reliability of the Signal list as a screening tool for adults with ID can be debated. We want to determine if the Signal List is a good practical alternative for the DDS.

## **Method**

### ***Study design***

Cross-sectional validation study.

### ***Study population***

In this study 388 adult residents with different levels of ID of three Dutch care provider services for people with ID were randomly selected. Participating care providers were: Ipse de Bruggen, 's Heeren Loo and Daelzicht. These three care providers provide supported living, work and day care. Inclusion criteria for participation were: minimum age 18 years and receiving formal care from one of the above-mentioned care providers. Exclusion criteria were: complete tube feeding and participants who refused cooperation while there was permission from the legal guardians.

## ***Recruitment methods***

To obtain a representative sample of adults with ID, a randomized selection within the residents of the three care provider services was performed. Inclusion of the participants was between 23.05.2013 and 03.09.2013. A random selection was used to obtain a balanced distribution of different levels of cognitive functioning and associated morbidities of the research population. Randomisation was performed using the random number generator at [www.random.org](http://www.random.org).

## ***Instruments***

### *Dysphagia Disorders Survey*

The Dysphagia Disorders Survey (DDS) is a screening assessment tool for dysphagia developed by Joan Sheppard ([Sheppard 1998](#)) and evaluated in children and adults with intellectual and developmental disabilities. Only trained professional observers are certified to administer the DDS. The DDS consists of two parts with 15 items in total ([Calis 2008](#)). The first part of the DDS contains seven items of factors that have been found to be associated with dysphagia. The second part contains eight task-related items to assess signs of neurologic, motor and behavioral competency of swallowing. This task analysis is applied to three food types: liquid, solids that require chewing and solids that do not require chewing. Five of the items in the second part address task components of the oral preparatory phase of swallowing, and respectively two and one item address the oral-pharyngeal and esophageal phase. A score in the top range of the DDS (maximum score being 38) indicates signs of dysphagia. The final evaluation study for the DDS resulted in a correlation between total DDS score and an expert speech therapist's opinion of 0.92. Inter-rater reliability in previous research was 97% (Sheppard 1988, unpublished data). To compare the outcome of de DDS with the outcome of de Signal List, a cut-off value was needed. Sheppard determined that 3 was an optimal cut-off point, yielding a sensitivity of 95% and a specificity of 94% (Joan Sheppard, personal communication). A summed score of 3 points or more of the DDS indicates that there is an increased risk of dysphagia. The DDS has not been evaluated using imaging diagnostic methods, such as video fluoroscopy.

### *Signal List*

The Signal List was designed to be filled out by untrained caregivers with the aim to screen for risk of dysphagia in people with ID ([Helder 2010](#)), as a basis for referral for further diagnostic assessment. It contains eight dichotomous questions that mainly include medical conditions related to dysphagia (see Appendix 1). In accordance with the procedure followed

by the author, the Signal List is filled out by a caregiver who knows the client for at least two months and who observed at least one meal in the last two weeks. Affirmative answers are scored 1. Each question is weighted differently in the calculation of the total score ([Helder 2010](#)).

The Signal List has been tested in 87 adults aged 50 years and older with ID and was compared with the DDS. A cut-off total score of 12 is used to determine the risk on dysphagia, with a sensitivity and specificity of 92.3% and 65.9% respectively. The correlation between the Signal List and the DDS was 0,7 (Annemarie Helder, personal communication). The used cut-off score in this validation study is 12.

### ***Procedure***

The study protocol was approved by the Medical Ethics Committee of the Erasmus University of Rotterdam (Protocol number MEC-2013-171). Informed consent of all participants and their legal guardians was obtained. Caregivers and their management were informed before appointments were booked,

Between one and seven days before the DDS was performed, a caregiver filled out the Signal List. Physicians and speech therapists, all certified by J Sheppard, working in the ID care, performed the DDS observations. All participants were observed during one meal in accordance with the procedure of the DDS within daily routine of participants. Only food and drink suited for the diet of the participants were offered. When there was not enough variation in consistency in the regular meals, extra food was offered to the participant.

When the DDS could not fully be performed due to the lack of cooperation of the participant, the observation was put on hold. If desired in agreement with the participant the DDS was continued later. Completing the DDS took no more than 30 minutes. The physicians and the speech therapists were blinded for results of the Signal List. The results of the DDS were reported to and discussed with the primary treating physician.

### ***Statistical analysis***

Data analysis was performed using the Statistical Package for the Social Sciences version 20. Correlation between the results of the DDS and Signal List were determined with Spearman's rho. Sensitivity and specificity were calculated and 95% confidence intervals were provided. To demonstrate the effect of changing the cut-off point of the Signal List, a cut-off point of 12 and 10 will be used.

To indicate the extent to which the Signal List may classify respondents as having dysphagia or no dysphagia according to the DDS, the area under the receiver operator characteristic (ROC) curve was calculated. Ninety-five percent confidence intervals were provided and deviation from .50 was statistically tested.

## Results

Of 388 contacted legal guardians, 139 agreed on participation. No response from the legal guardians, was the most common reason for not participating in the study. Reasons by which legal guardians decided not to participate were; overburden and assumed absence of swallowing problems. In 13 participants (9%) the DDS was not administered due to no permission by the participant. Absence of participant for medical reasons or holidays were noted as well. 126 people participated and fulfilled the Signal List and the DDS. Mean age of the participants is 52.7 years (range 20-98). 75 (60%) of the participants are male and 51 (40%) were female.

83 participants (66%) have a score of three or higher on the DDS, indicating an increased risk of dysphagia. Using the Dutch Signal List, 64 participants (51%) have an increased risk on dysphagia. The correlation between the Signal List and DDS is 0.51 ( $p < 0.01$ ).

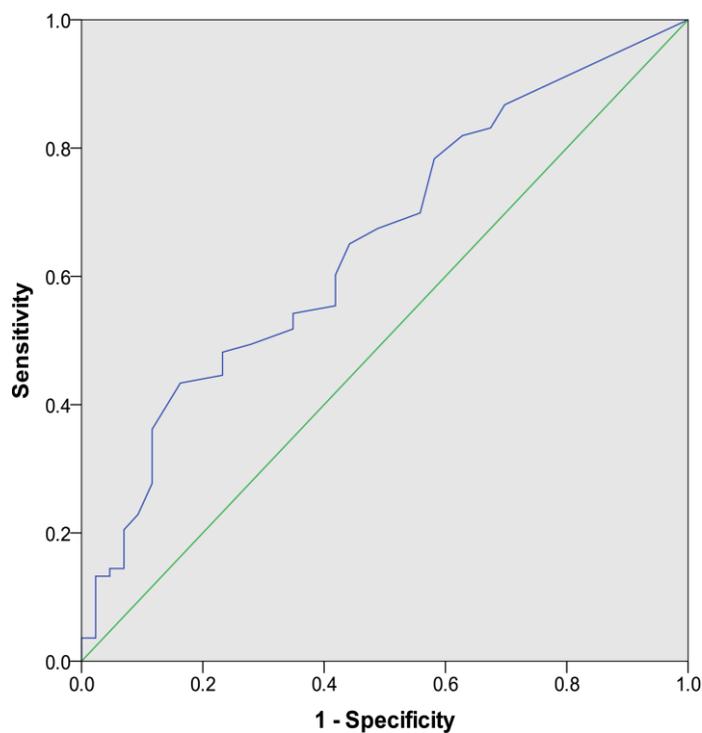
In table 1 the sensitivity and specificity of the Signal list compared to the DDS are shown, which are 55% (CI=45-66%) and is 58% (CI=43-72%) respectively. The false negative outcomes with the Signal List are in 45% (CI=34-55%) and the false positive outcomes are in 42% (CI=28-57%).

**Table 1. Outcome DDS and Signal List (cut-off point 12 and 10)**

	Number (n=126)*	Signal List (cut-off 12)		Signal List (cut-off 10)	
		Positive	Negative	Positive	Negative
Dysphagia	83	46	37	50	33
No dysphagia	43	18	25	18	25
Sensitivity		55% (95% CI 45-66%)		60% (95% CI 49-70%)	
Specificity		58% (95% CI 43-72%)		58% (95% CI 43-72%)	

\*diagnosis by DDS

The receiver operator characteristic (ROC) curve is shown in Figure 1. The relative proximity of the ROC line to the reference line indicates suboptimal classification. The area under the ROC curve is .65 ( $p=.005$ ; CI =.55 - .75), which can be classified as 'poor'. Meaning that the Signal List is a poor discriminator for dysphagia. An optimal cut-off point of the Signal List is 10, which corresponds with a sensitivity of 60% (CI=49-70%) and a specificity of 58% (CI=43-72%). A cut-off value of 6.5 provides a better sensitivity (.70) in favour of specificity (.44).



**Figure 1**

*The blue line represents the Receiver Operator Characteristic (ROC) curve, indicating the performance of the Signal List in classifying the respondents as having dysphagia or no dysphagia according to the DDS. The straight diagonal green line represents classification by chance.*

## **Discussion**

The aim of this study is to assess the validity of the Signal List for dysphagia compared with the DDS in a population of adults with ID. In total 126 participants were included in our study. Detection of high risk of dysphagia, scored by the DDS and Signal List differed vastly, being

45% and 66% of participants respectively. The correlation of 0.51 between the two screening tools was moderate (statistical approach according Cohen), the area under the receiver operator characteristic (ROC) curve was poor and the sensitivity (55%) and specificity (58%) of the Signal List in detecting dysphagia were poor.

Contributory to this study are: performance of the DDS by properly trained observers, and blinding of the DDS observers for the results of the Signal List. The inclusion process of this study might be slightly biased due to rejections by legal guardian of the selected adults with ID. Medical conditions of participants could have contributed to decision making, which could have resulted in over- or under presentation of healthy or ill participants. The reported prevalence of dysphagia are therefore only partially be generalized to the ID population. This lack of representativeness is not expected to account for variation in the associations between the DDS and the Signal List. In addition, the underreporting of swallowing problems by legal guardians is a common problem in daily practice ([Calis 2008](#); [Helder 2010](#); [Leeuwen 2007](#)). Thus, the low consent rate of 36% was expected. The reported prevalence of 45% of participants in risks for dysphagia found is comparable to recent Dutch research in older adults with ID (50%) ([Bastiaanse submitted](#)). This result was expected, even though participants in this study are younger, since a higher number of participants in this study were living in institutionalized settings when compared to in the Bastiaanse et al. study.

A screening tool with a false negative rate of 40% is of little use in clinical practice. ([Andermann 2008](#); [Wilson 1968](#)). Sensitivity (55%) and specificity (58%) presented in this study are lower than those found by Annemarie Helder: sensitivity and specificity were 92% and 66% respectively (personal correspondence).

There is a lack of useful dysphagia screening instruments in the ID population. The DDS is commonly used in ID research to map problems of swallowing disorders. Also in practice the DDS is commonly used by speech therapists and physicians in ID care. Although video fluoroscopy is considered to be a true golden standard, the choice to use the DDS in clinical research can be justified. If ethical reasoning can be argued, high participation rates cannot be expected using invasive instruments. The DDS can be criticized for its focus on just one medical factor, i.e. the Body Mass Index (BMI). The BMI is used as a measure for nutritional state, which means that it is an indirect and arguably imprecise measure for dysphagia. The Signal List is more focused on medical situations include: lung problems, having fever, epilepsy, cerebral vascular incident and dementia. The DDS and Signal List give both a partially indirect measurement of the swallowing function. The lack of correlation between the

Signal List and the DDS could currently still be caused by the measurement of different (in)direct parameters of dysphagia.

To further evolve the Signal List a properly validated Golden Standard is needed. Since the DDS shows a high sensitivity and specificity in the studies performed by Joan Sheppard, it is recommended to validate its reliability on screening for dysphagia evidence based.

We would like to propose a study to research the criterium validity of the DDS compared with videofluoroscopy, which is considered the true Golden Standard. Consensus on the feasibility of videofluoroscopy is lacking; its feasibility should first be tested among people with ID. Besides, it would be valuable to investigate the validity of the DDS in different research groups, since there are only a few specialists in this area so far.

## **Conclusion**

This study compared the ability of the Signal List to screen for dysphagia compared with the Dysphagia Disorders Survey (DDS). The Signal List is, in its current state, proven not to be useful as a proxy for the DDS in the population of adults with ID. However, the Signal List is practical and easy to use. Given the high prevalence's and morbidity of dysphagia in people with ID, future research needs to focus on validation of dysphagia diagnostics and evolving of screening tools for dysphagia. The Signal List needs to be further evolved before application in practice.

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## Appendix 1. Signal List (translated from the Signaleringslijst Verslikken)

	Question	Answer	Weighting Score
1	The client chokes sometimes.  <i>"Yes," with choking once a month or more. Fill with light and / or serious choking.</i>	Yes/No	6
2	The client coughs or splutters during or after eating and drinking.	Yes/No	6
3	The client takes more than 25 minutes for a meal for themselves	Yes/No	2
4	There is reflux or the client uses medication for reflux	Yes/No	3
5	One or more of the following factors apply:		
5.1	Lung problems	Yes/No	5
5.2	Regular fever	Yes/No	5
5.3	Epilepsy	Yes/No	5
5.4	CVA (stroke)	Yes/No	5
5.5	Dementia	Yes/No	5
5.6	Sitting in a wheelchair	Yes/No	5
5.7	Swallowing problems in the past	Yes/No	5
6	There is drowsiness or tiredness during the meal. Whether there is restlessness and possibly much talking.	Yes/No	3
7	Eating or drinking is adjusted.  <i>For instance: finely cut, without crusts, giving thickening drink, offering dosed, etc.</i>	Yes/No	6
8	The client refuses to eat or drink.	Yes/No	2