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Archives of Clinical Neuropsychology 00 (2020) 1-13

# Transposition and Normalization of the Mini-Mental State Examination in French Sign Language

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Received 8 May 2020; revised 15 October 2020; Accepted 2 December 2020

# Abstract

**Objective** Due to the lack of a validated translation, the mini-mental state examination (MMSE) cannot be used to screen for suspected dementia in deaf people who communicate in French Sign Language (FSL). Taking into consideration the cultural and linguistic features of this specific population, we transposed the validated French version of the MMSE into a version adapted to FSL users: mini-mental state-langue des signes (MMS-LS). The objective of our work was to obtain screening norms for the MMS-LS.

**Methods** The MMS-LS was tested on 194 deaf users of FSL with clinical dementia rating as the gold standard. Healthy and demented participants were seen for two or three consecutive testing sessions at 1-year intervals.

**Results** The MMS-LS exhibited excellent internal coherence validity (Cronbach's  $\alpha = .81$ ), unidimensionality (p = .002), and excellent sensitivity (p < .001). The MMS-LS score declined with overt and severe dementia.

**Conclusion** The percentiles obtained are useful norms for clinical assessment but must be interpreted with precaution due to the small number of participants (related to recruitment constraints) in the present study. In order to facilitate clinical use, the MMS-LS has been made available online, together with an instructions manual and clinical advice useful for improved awareness of the specific nature of this population.

Keywords: Mini-mental state examination; Deaf; Sign language; Cognitive decline; Cognitive screening; Dementia

## Introduction

The mini-mental state examination (MMSE) (Folstein, Folstein, & McHugh, 1975) is a screening test for cognitive impairment that has been translated into a wide range of oral languages. In France, the French version is used for the assessment of neurology and geriatric patients presenting symptoms suggestive of neurodegenerative disorders such as Alzheimer's or related diseases, but to our knowledge, no validated version of the MMSE has come forward in French Sign Language (FSL). Practitioners thus have to rely solely on their subjective clinical judgment to screen for dementia in this population, yielding an important risk of uncertain diagnosis. But practitioners, who cannot use their usual mode of oral communication with these

patients, are generally unfamiliar with the implications of deafness. They rarely encounter deaf people in their consultations and may be unaware of deaf culture and the neurolinguistic specificities involved. Here, we first reviewed the pertinent issues, with the goal of creating a new screening tool adapted to the deaf population.

# Cultural and Sociological Aspects of Deafness

*Family setting.* For the purpose of this article, we will not consider deafness per se (i.e., origin, type, and rehabilitation modalities) because the only relevant parameter here is the language used by the patient for everyday living. Although some Deaf people do communicate orally, speech is simply an acquired skill that complies with the communication mode used by healthcare professionals. It is not the Deaf patient's everyday language. The use of a capital D to designate the Deaf population emphasizes the cultural, linguistic, and sociological features of this specific group of individuals belonging to a common community (Leigh & Andrews, 2017) of people with prelingual deafness, defined as deafness present at birth or developing during the first years of life. Criteria of particular importance for the assessment of cognitive capacity in this population thus concern the use of sign language and their parents' hearing status are determining factors for affective and intellectual fulfillment (Courtin, Limousin, & Morgenstern, 2010): the earlier individuals learn to sign the better their language proficiency. But, 95% of deaf children have hearing parents (Courtin *et al.*, 2010), and the age of exposure to sign language is highly variable between families. Three types of signers are recognized among Deaf people: native signers (exposure to sign language from birth, deaf children with deaf parents), early signers (exposure to sign language at 5 to 8 years of age), and late signers (exposure after 13 years). Screening tests should take into account the linguistic variability observed in this population.

*Education.* The childhood of older Deaf people seen in our units for suspected cognitive impairment was quite different from that of Deaf children today. Nearly all of these older patients attended specialized schools for deaf children where FSL was not taught. These oralist schools did not have FSL interpreters. They banned the use of sign language and promoted the acquisition of speech (see history of Deaf people). When the patients we see were in school, sign language was less formalized and included a broad spectrum of old codes or signs created by family and friends, sometimes called homesign (Courtin et al., 2010) or dialect (Morere, 2013; Sutton-Spence & Woll, 1999). This led to disparate languages that varied from one school to another, sometimes within the same geographical area. Because certain old signs that differ from the academic FSL taught today are still used by these patients, a professional intermediary, known as Certified Deaf Interpreter (Drion & Buhler, 2016), is needed to understand them and adapt the healthcare message to their mode of communication. There are also regional variants of FSL, often related to the school attended. Experience has shown that to bridge the gap, a lot of consultations must be conducted with a Certified Deaf Interpreter. Today, it is not unreasonable nor discrediting to affirm that the educational level of older Deaf people is lower than that of their hearing compatriots (Moores & Martin, 2006). Because access to higher education was so difficult (no interpretation service) and because they were rapidly oriented toward vocational education programs, many Deaf people ended up with manual occupations: for example seamstress, cook, factory worker, shoemaker, painter, cabinet maker, and gardener (Moores & Martin, 2006). Many institutions for the Deaf still offer no other type of education. But as Deaf people became collectively aware that their language is like any other language—specific, complete, and autonomous—educational profiles began to change significantly (Mottez, 2006; Stokoe, 1960). This wake-up call led to educational reforms with adapted programs in FSL (Fabius law 1991, 2002 law). The age of the Deaf patient we see in consultation is thus an essential element to consider when interpreting clinical data, particularly if the tests proposed to evaluate cognitive capacity involve recall of academic knowledge.

Moreover, the overall educational background of older Deaf people in France is quite specific. For example, in the fields of arithmetic, French and geography, teachers used spoken language and were unable to communicate in their pupils' natural language. For many Deaf, the consequence was limited competency in reading, writing and arithmetic, although some were able to improve their skills later during their occupational career. This special context was further highlighted in 2005 when FSL was officially recognized as a separate language from French, with its own lexical properties and a completely different set of rules for grammar and syntax. For Deaf FSL signers, learning French is like learning a foreign language (Koutsoubou, Herman, & Woll, 2006). Obviously, new norms must be established for psychometric testing in the Deaf population.

## Neurolinguistic Specificities

*Sensory modality bias.* Because they have a visual representation of the world and have had little exposure to academic FSL during their period of development, older Deaf people use homesigns or old signs based on concrete elements perceived in the

environment. For categorization tasks, they generally opt for a schematic or functional justification (Courtin, 1997). Furthermore, for their daily needs, FSL signers use abstract notions (e.g., vegetables, vehicles, and insects) less often than lists of individual elements (e.g., carrots and potatoes, bicycles and cars, flies, and butterflies). Older Deaf people are often familiar with only a few specific categories (e.g., fruits and colors). Consequently, aptitude for categorial and conceptual reasoning cannot be considered strictly comparable between deaf and hearing people. Moreover, most tests used in neuropsychological explorations require conceptual reasoning and comprehension of oral or written information. Test instructions are often presented orally, without any visual support, particularly when evaluating verbal memory. This type of test induces sensory modality bias and is not adapted to deafness nor to visual-gestural-spatial language.

*Limitations of existing neuropsychological instruments designed for hearing patients.* For comparison, we can consider the report by Dean et al. (2009) who demonstrated the importance of using norms specifically adapted to the population under study to avoid interpretation bias. These authors gave the original version of the MMSE to a group of older (mean age 69 years) deaf American Sign Language (ASL) users recruited during a conference. Overall, the participants, who had a high level of education (14 years of schooling), did well on the test yet their mean MMSE scores were significantly lower than those obtained by their hearing counterparts (Dean, Feldman, Morere, & Morton, 2009). Such lower scores might have been interpreted as meaning the Deaf participants had suspected mild-to-moderate cognitive impairment when actually there was a testing bias related to misunderstood, or at least poorly adapted, test questions. This bias becomes clear when looking at the items used to evaluate language competency. These items require good reading skills (e.g., ability to read the instructions, to spell words) that many Deaf people do not have. Illiteracy rates can be very high in this population, as high as 80% as reported in 1998 in one French parliamentary district, but perhaps higher today. Many Deaf people can read words, but do not correctly understand the meaning of sentences, which leads to many interpretation errors (Drion & Semail, 2016). This is why we initially considered transposing the Montreal Cognitive Assessment (MoCA, Nasreddine et al., 2005). We found, however, that this would have been extremely complex due to the specificities of the target population as outlined earlier. Many of the MoCA questions require categorial reasoning (i.e., items involving similarities between two concepts) or verbal fluency (i.e., produce a list of words beginning with the same letter). Most Deaf people would probably be unable to understand this type of question and would thus give an erroneous answer. Some of the MoCA questions were nevertheless retained in the recent British Sign Language Cognitive Screening Test (BSL-CST) created by Atkinson et al. (Atkinson, Denmark, Marshall, Mummery, & Woll, 2015).

Published well after the creation of the MMS-LS, this study designed to set the norms for a new test in BSL demonstrated the importance of proposing an instrument adapted to the participants' everyday language. The test under study was found reliable enough to distinguish the presence or absence of dementia in all BSL users, irrespective of their sign language (dialect or academic). This study also described the heterogeneous nature of the British Deaf community and found that it would not be a bias for using this test in the clinical setting.

We thus concluded that the validated French version of the MMSE (Kalafat, Hugonot-Diener, & Poitrenaud, 2003) met two important criteria: It is the most widely used screening test in France and its results have been abundantly reported in the literature. We wanted to validate this tool in FSL so it could be appropriately used for screening the Deaf population in France. This meant developing an MMSE in sign language with psychometric properties ensuring reliable measurement of the desired dimension (i.e., the presence of dementia) with good sensitivity and acceptable internal validity.

Summarizing, a simple translation of the original MMSE cannot produce a pertinent screening test for cognitive disorders in a Deaf population. Transposition or adaptation would be a better way of describing the necessary transition (for review see Haug & Mann, 2008). In order to take into account the neurolinguistic specificity of Deaf people as well as their social and educational particularities, we modified certain MMSE test items, especially those soliciting language skills, to achieve this adaptation. The resulting MMSE in FSL (MMS-LS) was then tested as a screening tool for cognitive impairment in a Deaf population of FSL signers.

# **Materials and Methods**

# Study Population

Potential candidates for inclusion in this study attended medical visits conducted by the investigating physician in centers providing assistance and care for Deaf people. For inclusion, participants had to be over 18 years old, have prelingual deafness, and be FSL signers. Individuals who presented low visual acuity (<.5) or motor impairment perturbing the use of FSL were

not included. All participants gave their informed consent after receiving an information document and participating in an individualized interview where the general objectives of the study and the research protocol were explained in detail. The study protocol was approved by our institutional review board (CPP Nord-Ouest IV, approval 10/52, 11/9/2010) and registered with clinicaltrials.org (NCT02005679). We recruited 194 participants, 98 women and 96 men, in the following French centers: Lille Lille Catholic Hospitals, Lille Catholic University (n = 139), Pontchaillou Hospital, University of Rennes (n = 20), Psychiatrics Pole, Conception Hospital of Marseille (n = 29), and Saint Julien Hospital, University of Nancy (n = 6).

The participants had attended various schools in different regions of France or other countries. Their educational level was relatively low: no schooling (2.1%); highest level primary school (33%), middle school (13.3%). A few participants had finished high school or attended university (4.2%) but there were more who had had a vocational education (professional license 42%, apprenticeship 5.3%). Most of the participants were currently working (92.3%), mainly in a regular work environment and for some in a protected environment (i.e., a working environment specifically adapted to the employee's personal situation, as is available in France for disabled persons).

# Study Protocol

Participants and an accompanying person (e.g., spouse, descendant, and assistant) attended a first visit (t0) in the Deaf care unit of a participating center during which the study questionnaires—MMS-LS and other questionnaires and tests adapted as needed to ensure correct understanding—were administered. The participant and the accompanying person were invited to respond separately. First, the participant was met alone for an interview and the MMS-LS, the "patient" version for Clinical Dementia Rating (CDR) scale (Hughes, Berg, Danziger, Coben, & Martin, 1982) and digitalized linguistic tests. Later, the participant rested in a waiting room while the accompanying person was met for an individual interview and the "accompanying person" CDR (Hughes *et al.*, 1982). At the end of the visit, the neuropsychologist combined the information collected during the interviews and the tests to establish the final CDR. The participant and the accompanying person were then met together to answer questions concerning the participant's linguistic and schooling background.

One year ( $\pm 4$  months) later, a second visit (t1) was proposed for a comparative neuropsychological evaluation designed to determine the stability of the test results. It was assumed that in the absence of a dementia disorder, cognitive function would remain stable over time, and consequently the MMS-LS scores would be similar at t0 and t1.

## Transposing the MMSE into the MMS-LS

*Phase of conception.* A group of clinicians, researchers, and specialists (geriatricians, neurologists, Deaf care clinicians, neuropsychologists, and deaf and hearing linguists) working in the field of language were solicited to undertake the transposition of the French version of the MMSE into the FSL version, MMS-LS. Volunteer Deaf patients participated in pretests to adapt test items. For various reasons detailed subsequently, certain items in the initial French version of the MMSE were modified or replaced for the MMS-LS.

*The screening team.* Each investigating center hosted a team of three persons to administer the screening tool: a neuropsychologist, a Certified Deaf Interpreter who was also an FSL teacher and a research assistant, and a certified French-FSL interpreter. Preliminary work involved discussions among the members of the team to establish a standardized way to ask the MMS-LS questions. Some questions issuing from other questionnaires were not standardized but adapted for proper comprehension by Deaf people.

Concerning the MMS-LS test to be standardized, test items were not administered with a video as in Atkinson *et al.* (2015), but rather presented to the participant in FSL by the Certified Deaf Interpreter. Unlike an impersonal video, the physical presence of the test administrator took advantage of the tridimensional aspect of FSL and provided a reassuring setting for the participants. The neuropsychologist carefully monitored the screening sessions, making sure the questions were asked correctly, noting the participants' responses, and intervening as needed.

*Categorization problems with registration/recall.* In the French version of the MMSE, the registration/recall test lists words belonging to different semantic categories. When the participant fails to recall a particular word spontaneously, the investigator is supposed to prompt the participant to help recall the registered information. For an older Deaf person, the signs GAME or PLANT are not very effective as prompts for BALL or FLOWER because they correspond to very abstract notions not used in everyday life. Nevertheless, the clinical experience of the investigating team working on creating the MMS-LS led to a

consensus concerning certain specific categories, that is FRUITS and ANIMALS, that are regularly used by older Deaf people. Based on this observation, it became apparent that recall could be prompted in the MMS-LS in the same way as in the original version of the MMSE as long as the prompts belonged to these specific types of categories regularly used by the population under study. Moreover, because in FSL certain signs have visual similarity (e.g., CIGAR is visually similar to TO SMOKE), the list of signs to remember was selected so there would be little visual similarity between a single categorial member and the corresponding categorial concept. SOMETHING ROUND OR SPHERICAL could not be used as a prompt for BALL. For the comparative evaluation to determine result stability, the signs GOAT, DOOR, and CHERRY were selected for the first visit and SHEEP, WINDOW, and APRICOT for the second visit. If the participant had difficulty recalling the registered information, the administrator prompted with: "What was an animal, ... part of a house, ... a fruit?". These semantic categories were selected because they involved concrete concepts frequently encountered in FSL (Courtin, 1997).

# Language items and problems related to written language acquisition.

*Interference task (between three-word registration and recall).* Spelling world (Fr: *monde*) backward could not be used as the interference task for several reasons. First, FSL is not a written language, and besides the fact that knowledge of written language is nominal in many FSL signers, few know how to use dactylology. In order to avoid a floor effect, spelling backward was replaced by a sequence of numbers: the participant was asked to repeat "11, 12, 13, 14, 15" backward. FSL users are familiar with these numbers and sign them the same way throughout the country, unlike the names of days or months that can be signed differently in different regions of France. This backward sequence of numbers is less automatic than would be "10, 9, 8, so forth." and implies activation of attention and working memory. It is nevertheless a simple task easier to perform than the mental arithmetic task (i.e., items of calculation).

*Comprehension of written instructions.* As mentioned earlier, many older Deaf people learned to read under very suboptimal conditions. In order to avoid the penalizing bias this would imply for a majority of participants, we chose to test a specific grammatical function of FSL. For example, repeating a sentence in FSL requires correct visual–spatial "inversion," which thus signals good comprehension and execution of a transfer. A person who does not understand FSL would mimic a mirror image of the gestures without grasping the meaning. For this task, the participant was asked to repeat the sentence "walk along a road, with a mountain to the left and a river to the right" signed by a deaf person in a video. The point was awarded if the participant repeated the sentence using correct FSL grammar. Marshall *et al.* (2015) demonstrated that a signed question is repeated correctly if and only if it is understood semantically. This repetition of a significant sentence thus tests the participant's comprehension of FSL.

*writing test.* The participant was not asked to write. This task was replaced by a signing skill test. Initially, the participant was asked to sign something spontaneously in FSL, but the preliminary period showed that participants were confused by this instruction, not knowing what to sign. It was thus decided to propose a supporting image of a daily life activity with the following (signed) instruction: "describe this image, telling us what you see." The participant's production was analyzed by the expert in FSL, taking into consideration FSL's rules of spatial grammar (e.g., positional rules). This task evaluated the participant's signing skill in describing a visual scene.

#### Language items and problems related to FSL.

*Naming task.* The two objects generally used in the MMSE naming task were not retained for the FSL version because of their strong iconicity that would probably mask any phasic disorder. In FSL, the signs PENCIL and WATCH are easy to find, simply by chance. The participant could easily respond in FSL that the object is used to write (and thus be given the point) without naming PENCIL. The sign WATCH is made simply by pointing to a watch on the wrist. These objects were replaced by WATER and HANDKERCHIEF, signs that have weak iconicity and are only understandable by FSL signers.

*Repeating a nonsense sequence task.* The principle of a nursery rhyme initially chosen for the English version of MMSE was not retained in the French version. The French version that uses "*pas de mais, de si, ni de et*" (no but, no yes, no and) was transposed into FSL in an adapted form in order to avoid asking the participant to recall too many signs. It was important to avoid the word "et" (and) because there is no equivalent in FSL. The sequence to repeat was "*mais oui, après non, jamais là*" (but yes, after no, never there).

*Comprehension task and execution of a triple order.* The instructions in the official French version of the MMSE are "Take this sheet of paper in your right hand. Fold it in two and throw it on the floor" (Kalafat *et al.*, 2003). Translation of this into FSL would have led to the use of very iconic signs that could be understood without any knowledge of FSL. Thus, in order to create an item capable of testing the participant's comprehension of FSL, we decided to use a less iconic vocabulary. Participants were thus

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asked to execute a triple order: "Take this newspaper, open it, and tear out a page." To properly execute these unusual and rather incongruous orders, the participant had to understand the instructions signed in FSL.

Globally, the choice of items to memorize or process was a delicate problem because of regional variability and the etymological evolution of certain signs (Atkinson *et al.*, 2015).

Items of orientation, copy a drawing and calculation. These items were not changed.

## Other Tests and Questionnaires Administered

*Clinical dementia rating.* CDR has two complementary sub-scores, one for the accompanying person and one for the patient. The patient is not present when the accompanying person answers the questionnaire so the patient's behavior in different spheres of everyday life can be analyzed in more detail. The investigator asks the accompanying person to describe any memory problems the patient might have as well as how the patient copes with orientation to time and space in a familiar environment. The participant's aptitude for judgment in case of an unexpected event and adaptation to social conventions are also analyzed, as well as the patient's capacity to execute daily life activities for eating and personal hygiene. The analysis of the participant's and the accompanying person's answers to different items on the CDR questionnaire are analyzed by a neuropsychologist who then completes the evaluation chart for memory, orientation, social skills, autonomy in daily life activities, and level of dependency.

At the end of the interview, the neuropsychologist investigator determined whether the domains memory, orientation, judgment and problem solving, family and recreational activities, and personal hygiene were affected or preserved, then established the participant's CDR. An algorithm, available online at https://www.alz.washington.edu/cdrnacc.html was used to calculate the total CDR; CDR = 0: absence of dementia disorder; CDR = .5: suspected disorder (questionable disorder); CDR > .5: mild (CDR = 1), moderate (CDR = 2), or severe (CDR = 3) dementia.

*Linguistic, familial, schooling, and occupational background.* One final questionnaire created for the purpose of this study was administered to the participant and the accompanying person. This questionnaire was used to learn more about the participant's background, including whether or not family members (parents, grandparents, siblings, and spouse) were FSL signers. Data on the participant's age at first contact with FSL and schooling were collected, as was information concerning occupational history and use of social aids specifically designed for Deaf people. The participant's social-occupational status was noted to complement available information about the participant's life experience or personal and family history.

*Linguistic level.* Considering the diversity of linguistic abilities in the study population, two tests were used at the first visit to evaluate the participant's knowledge of FSL. Performance level and links with the MMS-LS results were studied to construct a calibration grid. These two tests had been created before initiating the study. They were inspired by the work of British and American (Bencie Woll *et al.*) and French (Courtin) linguistic research. Unfortunately, tools evaluating comprehension (e.g., BSL Receptive Skills Tests, Herman, Holmes, & Woll, 1999) and production (e.g., ASL—Proficiency Assessment, Maller, Singleton, Supalla, & Wix, 1999) have not been validated in FSL. The two tests described subsequently and proposed in this study are currently in the process of being validated in the Deaf population in France. Test 1 was designed to evaluate capacity for lexical reception and Test 2 (sentence repetition) to evaluate syntaxial expression in FSL. For these two tests, the signs to be understood, and the sentences to repeat were signed by a Deaf expert in FSL (video recording in a neutral environment). The 10 items in each test were presented in random order. All recordings were presented in a uniform way as a slide show on a laptop computer screen set about 50 cm in front of the patient.

For the lexical reception test, the patient was instructed to point to the image corresponding to the sign on the screen. Images were colored drawings made by an independent graphist and incrusted in random order around the edge of the screen. The patient was asked to match the target image with the visual sign. In addition to the target image (e.g., train station), four distractors were proposed as possible solutions, including semantic distractors (e.g., bus stop) and distractors created with minimal pairs, that is signs differing only by one FSL parameter (position, movement, orientation, or configuration). For example, in FSL the signs CAKE and SOAP can only be distinguished by movement. In addition, one of the distractors was a sign with high iconicity value that would be more likely chosen by individuals who do not understand FSL. The maximal score for Test 1 was 10.

For the syntax test, the patient was asked to repeat exactly a signed sentence displayed on the screen. The patient had to understand the meaning of the sentence and repeat each syntaxial unit (subject, verb, and complement) using correct FSL grammar. The team's Certified Deaf Interpreter, a linguistic expert, analyzed the participant's production. Sentences of different

Table 1.	Demographic	data in healthy	participants	s and demei	ntia patients at t0

	Healthy $(n = 108)$	Dementia $(n = 29)$	<i>p</i> value
Age (year) m $\pm$ SD [range]	56.5 ± 14 [22-83]	78.1 ± 10.4 [57–92]	<.0001
Age at exposure to FSL (year) $m \pm SD$ [range]	$5.5 \pm 4$ [0–17]	$7.3 \pm 6.3 \ [0-30]$	.14
MMS-LS score (max 28 points) m $\pm$ SD [range]	$24.5 \pm 3$ [16–28]	$9.8 \pm 5.5$ [0–18]	<.0001
Other deaf family member $(n; \%)$	18; 19.8	4; 17.4	1
Education ( <i>n</i> ; %)			
Middle school, professional license	81; 75	15; 51.7	.028
No diploma	27; 25	14; 48.3	

length and complexity were presented. Several criteria for FSL grammar were evaluated pointing, directional verbs, and marks of plural. The maximal score for Test 2 was 10.

#### Evaluation Times (t0, t1, and t2)

Among the 194 participants evaluated at the first visit (t0), 33% dropped out of the study for various reasons (geographical distance, personal decision, death, etc.). Thus, there were 133 participants evaluated at t1 ( $12 \pm 4$  months after inclusion) and 40 at t2 (about 2 years after inclusion). By t2, many healthy participants grew weary of taking tests and had little motivation for a third round of MMS-LS. It was thus decided that MMS-LS re-evaluation and neuropsychological follow-up would be proposed at t2 only for patients with degenerative disorders (dementia score in groups CDR > .5) at t1. As this decision was made during the course of the study, data collected at t2 were limited. Demographic data for patients with dementia and healthy participants are given in Table 1.

# Statistical Analysis

Comparison between Healthy and Dementia patients was done using Chi<sup>2</sup> tests for qualitative variables, or in case of low numbers, Fisher exact tests. For numeric variables, Wilcoxon–Mann–Whitney tests were used because of non-normal distributions.

At each visit, internal validity was assessed using Cronbach's alpha coefficient. In accordance with common interpretation,  $\alpha > .7$  was considered satisfactory (Bland & Altman, 1997). Confirmatory factorial analysis was applied to check items for unidimensionality. Considering that a significant change in CDR would not be expected between t0 and t1, MMS-LS repeatability was evaluated in participants whose CDR remained stable between these two visits. All questionnaires were administered by the same investigator. Intra-investigator reliability was evaluated with the interclass correlation coefficient (ICC) using the 2-factor random model scored with the Shrout and Fleiss convention (ICC [2.1]). According to the Koo and Li (2016) interpretation grid, > .75 intra-investigator reliability was considered to be good and > .9 excellent.

Concurrent validity of a measure is its capacity to measure its target, and sensitivity is its capacity to differentiate two individuals with different states of dementia. Certain diagnosis is lacking due to the absence of a gold standard. Nevertheless, the coherence of results obtained with available tools was evaluated by determining Spearman's correlation coefficient between MMS-LS and CDR for the three visits.

#### Floor or Ceiling Effect

MMS-LS item scores were analyzed to detect those with a floor or ceiling effect, that is questions that were too easy (or too difficult) to answer such that the point was awarded (or not) for all participants irrespective of their CDR.

#### Factors Having an Impact on the MMS-LS

We searched for a link between the MMS-LS result (maximum 30 points) and a certain number of factors, independently of dementia status. In addition to age, gender, and manual dominance for signing, determining factors studied were related to familial and social context: deafness in family (mother, father, or grandparents), age at exposure to FSL (native, early, late), use of FSL in the family (with parents, siblings and/or spouse). We also searched for a link between MMS-LS and Test 1 and

Variable	Modality	n	Mean $\pm$ <i>SD</i>	Median [IQR]	<i>p</i> value
-	Total	108	$24.5 \pm 3$	25 [22; 27]	
Gender	Woman	60	$24.5\pm3.1$	25 [22; 27]	.76
	Man	48	$24.4\pm2.9$	25 [23; 27]	
Educational level	With diploma	81	$24.9 \pm 2.7$	25 [23; 27]	.029*
	No diploma	27	$23.2\pm3.5$	24 [20.5; 26]	
Dominant hand	Right	99	$24.5 \pm 3$	25 [22; 27]	.81
	Left	9	$24.3 \pm 3$	26 [24; 26]	
Age at exposure to FSL	Before 6 years	38	$24.1 \pm 2.9$	25 [22; 26]	.59
	At school	39	$23.6 \pm 3.1$	24 [21.5; 26.5]	
	Later	12	$24.3 \pm 3.3$	25 [23.8; 27]	
Occupational activity	Ordinary environment	47	$24.4 \pm 3.3$	25 [23; 27]	.12
	Protected environment	5	$21.8\pm3.3$	20 [20; 23]	
	None	6	$23.3 \pm 2.5$	22.5 [22; 23.8]	
Other deaf family member	No	73	$24.1 \pm 3.1$	25 [22; 27]	.00058**
	Yes	18	$26.7 \pm 1.5$	27.5 [25.2; 28]	
FSL used in the family	No	7	$25 \pm 3.3$	26 [23.5; 27.5]	.63
	Yes	96	$24.5\pm3.1$	25 [22; 27]	

Table 2. Impact of discrete variables on the mini-mental state - langue des signes (MMS-LS) score at t0

*Note:* Significance threshold: \**p* < .05; \*\**p* < .01; \*\*\**p* < .001.

**Table 3.** mini-mental state - langue des signes (MMS-LS) score percentiles in all healthy participants (clinical dementia rating = 0) at t0 and indicative results by sub-groups (maximum score 28 points)

	Percentiles						
	1%	5%	10%	25%	50%	75%	90%
All healthy participants $(n = 108)$	17	19	20	22	25	27	28
Sub-group analyses							
With diploma $(n = 81)$	18.6	20	21	23	25	27	28
Without diploma $(n = 27)$	16.26	17.6	19	20.5	24	26	27.4
With other Deaf family members	24	24	24.7	25.25	27.5	28	28
(n = 18)							
Without other Deaf family members	16.7	19	20	22	25	27	28
(n = 73)							
All healthy participants $(n = 108)$	17	19	20	22	25	27	28

2 results evaluating level of FSL fluency. For educational and occupational background, the following factors were studied: schooling completed with a diploma (or not), occupational activity (regular vs. protected environment/none).

For discrete variables, mean MMS-LS scores were compared using Student's t test for normal distributions or the Wilcoxon-Mann–Whitney test otherwise. For continuous variables (age, Test 1 and Test 2 results) Spearman's nonparametric coefficient of correlation was determined. Only data from participants rated CDR = 0 were analyzed (Table 2).

# Norms

The norms for MMS-LS scores in a healthy Deaf population were described by the distribution percentiles (Table 3).

# Results

# Psychometric Properties of the MMS-LS

*Internal validity.* The internal validity of the MMS-LS test was excellent: the Cronbach's coefficient at t0 (n = 194,  $\alpha = .81$  [95% confidence interval (CI) .78; .84]); at t1 (n = 133,  $\alpha = .83$  [95% CI .80; .85]); and at t2 (n = 40,  $\alpha = .81$  [95% CI .77; .83]).

Unidimensionality. Factorial analysis confirmed unidimensionality (p = .002).

*Repeatability.* The interclass correlation coefficient (ICC), an estimation of intra-observer reliability, was excellent: ICC = .91[.87; .94] (n = 103). Participants' MMS-LS scores were considered to be equivalent at t0 and t1.

Sensitivity. The link between MMS-LS and CDR at the three visits was studied with Spearman's coefficient of correlation. The results showed good correlation at t0 (n = 189,  $\rho = -.64$  [-.74; -.55], p < .0001), at t1 (n = 129,  $\rho = -.66$  [-.76; -.52], p < .0001), and at t2 (n = 39,  $\rho = -.82$  [-.93; -.76], p < .0001). MMS-LS score was lower for more severe dementia.

*Changes made in the MMS-LS.* The statistical analysis of the recorded data showed that most of the participants failed two items on the MMS-LS test: at t0 95% and 91% of participants failed the repeating a nonsense sequence task and repeating a signed sentence task, respectively. Internal validity and repeatability remained unchanged when these two items were removed from the analysis (at t0 Cronbach's  $\alpha = .82$ , ICC (2.1) = .92). In the final version of the MMS-LS, the maximum total score was thus 28 instead of 30.

*Norms.* The results showed a link between the MMS-LS score and the results of the fluency tests (Test 1:  $\rho = .31$ , p < .01; Test 2:  $\rho = .40$ , p < .0001). There was also a link between the MMS-LS score and age of nondementia participants ( $\rho = -.27$ , p < .01), having a diploma (p = .029) and the presence of another Deaf family member (p < .001). For the fluency tests and age, the coefficients of correlation showed a weak link that did not appear to be clinically pertinent for determining population norms. Similarly, although there was a statistically significant difference between participants with and without a diploma, it was only a small 1-point variation from the median. The norms proposed in Table 3 as percentiles were determined using data from only nondementia participants (CDR = 0). Similarly, percentiles were determined for sub-groups defined by significant discrete variables (with and without a diploma, presence or not of other Deaf family members). Because of the small sample sizes (n = 27 for patients without a diploma and n = 18 for participants with another Deaf family member), the corresponding percentiles are to be interpreted with caution. No difference was observed in the MMS-LS scores from the different investigating centers (p = .68). This confirms that the test is well adapted to the Deaf population in the different regions of France, independently of linguistic particularities.

## Discussion

The purpose of this study was to obtain a standardized tool for dementia screening in a population of Deaf FSL signers and to establish norms for its use in routine practice. A sample of this neurolinguistically specific population was studied to improve the reliability of the psychometric measurements so that patients with a potential risk to develop neurodegenerative disorders can be detected early. The French version of the 30-point MMSE was transposed and adapted for administration to Deaf people in their natural language. Two test items were found to provide little information and were removed from the final version. Thus, the MMS-LS presented here had a maximum score of 28 points. The analysis of psychometric properties revealed excellent internal validity and good reliability and sensitivity.

# Contribution of the Study and Clinical Implications

The MMS-LS will undoubtedly improve screening practices in the Deaf population. Earlier detection of potential cognitive impairment will allow healthcare professionals to initiate adapted care as early as in the hearing population. Established norms will be useful in avoiding misdiagnosis, for example false-positive diagnosis of dementia. In the Deaf population, this is a real risk if screening test questions are not administered in everyday language. Otherwise what is actually a straightforward problem of linguistic incompatibility might be interpreted as a sign of cognitive impairment. Because the MMS-LS results were found to be independent of social-demographic, educational, and familial variables, the test can be used as a global assessment tool despite its characteristic heterogenicity. A French-FSL interpreter must be present to administration manual that provides appropriate instructions to be given to the participant in FSL. The interpretation manual details the specificities of the target population and offers advice for interpreting results.

## Limitations of the Study

In order to better orient future research, the choices made when elaborating the MMS-LS may now, a posteriori, be questioned. For instance as outlined in the introduction, the specific conceptual context in the Deaf population compromised

the use of categorial cues to prompt memory recall. We could have opted for the solution proposed in the BSL-CST (Atkinson *et al.*, 2015), that is to prompt with the first of three signs to recall. This would, however, involve a methodology bias because the storage of episodic memory could not be evaluated for patients who had spontaneously recalled the first sign, but not the second and third. For future development, another way of prompting recall would be needed to evaluate verbal memory in Deaf people. The use of associative memory tests (Maillet *et al.*, 2016; Maillet *et al.*, 2017) might be proposed.

The interference test that used backward repetition of "11, 12, 13, 14, and 15" might have to be replaced. Even though the mental manipulation involved in repeated a series of numbers backward involves processes of information updating and inhibition of the automatic series (Friedman & Miyake, 2004), the task may have been too simple. The evaluation of working memory might have been usefully completed with a complementary test inviting the subject to recite the months of the year in inverse order, as was proposed by Atkinson *et al.* (2015). But the fact that older Deaf people in France use different signs for the months in different geographical regions would potentially complicate result interpretation.

Others (Atkinson et al., 2015) have administered screening tests in the Deaf population using a standardized video presentation. For our study, a neuropsychologist supervised the tests ensuring that all questions were administered in the same way to all participants. The Deaf participants reported that the testing process was stressful, many comparing it with tests in school. Moreover, those suffering from cognitive disorders were confronted with their failures in the arithmetic and memory tasks and had to be re-assured. The presence of a Deaf investigator who asked the questions in FSL, not only had an appeasing effect, but was also a constraint, limiting potential use of the MMS-LS in routine practice, especially because specialist consultations with a deaf professional are rare. Nevertheless, a Deaf investigator might be usefully integrated into the teams of the more than 20 centers providing care for Deaf people in France. To improve test administration, we would like to propose training sessions for healthcare professionals administering the MMS-LS. Moreover, the study population was far from uniform due to the wide age range (Table 1) and the participants' highly variable usage of FSL. For us, it would be important to have test norms for older people (>65 years) and for a younger population (<65 years). In this way, the test could be used over a wide age range yet be adapted to age-related linguistic characteristics. The norms obtained here can be used as a starting point, gaining in robustness as a larger number of participants are evaluated. It is also important to recognize that the use of FSL is in constant evolution so that changes can be expected to develop in the future. The correlations observed here show that the higher age at which the participant encountered FSL, the lower the linguistic proficiency (for the lexical reception Test 1; Spearman's correlation = -.24, p = .0011; sentence repetition Test 2, Spearman's correlation = -.36, p < .0001). Although this variable had no impact on the MMS-LS score, it did affect the performance level on test items we have modified. These items were created and pretested by Deaf people with higher education who were highly proficient in FSL (i.e., FSL teachers), which was not the case for the majority of the study participants. Retaining these two repetition items did not have a pertinent impact because of their lack of sensitivity. We can also remark that the "no ifs, no buts, no ands" phrase in the English version of the MMSE may be a familiar expression in English but that its translation, literally "pas de mais, de si, ni de et" is rather nonsensical in French. The result is even worse in FSL and cannot be used to evaluate language function (Jacus & Martin, 2000). What is more pertinent is the observation that the 28-point version of the MMS-LS was not correlated with Deaf participants' FSL proficiency nor with their linguistic background because it is a screening test for dementia independently of linguistic abilities. Nevertheless, Table 3 provides percentiles for sub-groups of patients based on individual characteristics that could be useful for interpreting test scores.

Ideally, a larger sample size would have been useful. Having scores for a larger number of patients with dementia would have enabled a matched pair design based on sex, age, education, age, and FSL exposure and potentially helped reduce the impact of individual variability. But such patients belong to a very isolated and vulnerable population, making recruitment rather difficult, even for units providing care for the Deaf population.

Due to the lack of a validated tool for the diagnosis of dementia in this population, CDR was taken as the gold standard for comparisons. CDR is based on a combination of items evaluating cognitive deficiencies and their impact on daily life activities, but remains a subjective judgment made by a neuropsychologist. Thus, at the present time, the validity of the MMS-LS cannot be fully confirmed. Nevertheless, the screening tool transposed into FSL, the MMS-LS, evaluates the designated dimension (dementia) independently of social and demographic variables; specifically the goal of this study. Because the classical MMSE has not been translated into FSL and thus not validated, there is no possible comparison with the MMS-LS results. Correlation analyses between two versions (MMSE translated into FSL and MMS-LS) might have been performed to provide further information on the pertinence of our adaptation.

The statistical analysis was conducted with rigor despite the smaller number of data items at the t2 visit compared with the t1 and t0 visits. Score stability was analyzed using data from the first two visits (t0 and t1) collected from nondemented participants. Moreover, we found that MMS-LS results showed good discriminating power between CDR groups: CDR = 0 no



Fig. 1. Mini-mental state - langue des signes (MMS-LS) scores (modified) (maximum 28 points) at the three evaluation times (t0, t1, t2) as a function of the clinical dementia rating score.

dementia; CDR = .5 suspected cognitive disorder, and CDR > .5 overt cognitive disorder (Fig. 1). The stability of the MMS-LS scores at 1 year in the participants free of dementia (CDR = 0) and the fall in the score in patients with dementia, demonstrated the tool's the good sensitivity.

#### **Research Perspectives**

Though the MMS-LS described here proposes a global measure of cognitive function, it remains a screening test and does not explore all domains of cognition and thus must not be considered as a diagnostic tool. An improved version of the MMS-LS based on data obtained from other tests administered within the framework of this study is in the development phase and should lead to a more complete screening test to be published later. Furthermore, the Deaf participants also responded to neuropsychological tests evaluating visual episodic memory and capacity for visuospatial and visuoconstructive organization. Corresponding norms will be published shortly. The possibility of transposing into FSL the test created by Atkinson *et al.* (2015) that is more complete and recently validated in a large population was also examined. This would require a much larger cohort because of the wide variability of linguistic and educational profiles.

Indeed, in order to establish an evidence-based diagnosis of cognitive disorders in Deaf patients, clinicians need validated norms for psychometric screening tests conducted in the patient's natural language, particularly important for evaluating linguistic competency.

# Conclusion

Created in 1975 by Folstein *et al.*, the MMSE is a well-recognized screening test for cognitive disorders. Translated into a whole series of oral languages, it has become a highly useful routine screening tool in many countries. Nevertheless, its usefulness in the Deaf population, even when translated into sign language, is questionable. This lack of pertinence arises because of the cultural and linguistic specificities of the Deaf population. Consequently, screening for cognitive impairment and diagnosis of dementia is hazardous in this small population (about 1 per 1,000 persons in the general population). We propose a transposition

of the French version of the MMSE into FSL: MMS-LS. This new tool has excellent internal validity and good reliability and sensitivity for the diagnosis of potential cognitive impairment in Deaf signers of FSL.

Conflict of Interests

None declared.

#### Funding

The study was promoted by GHICL and funded by a French public grant obtained in 2010 (PHRC n°19-08). It received the approval of the French ethics committee CPP Nord-Ouest IV (n°10/52) and is registered on clinicaltrial.org (NCT02005679).

## Acknowledgements

This study would not have been completed without the contributions and advice of international researchers. The authors thank in particular Bencie Woll, DCAL, London and Laurence Meurant, FNRS, and University of Namur. The authors also thank the members of the GHICL Delegation for Clinical Research and Innovation and the members of the Deafness and Health Network and the Deaf Care units in Rennes, Marseille and Nancy for their active recruitment of participants. Finally, this work could not have been completed without the generous participation of the Deaf individuals to whom we are most grateful. Link to the website https://www.psysourds.fr for more information.

#### References

- Atkinson, J., Denmark, T., Marshall, J., Mummery, C., & Woll, B. (2015). Detecting cognitive impairment and dementia in deaf people: The British Sign Language Cognitive Screening Test. Archives of Clinical Neuropsychology, 30(7), 694–711. doi: 10.1093/arclin/acv042.
- Bland, J. M., & Altman, D. G. (1997). Statistics notes: Cronbach's alpha. British Medical Journal, 314(7080), 572–572. doi: 10.1136/bmj.314.7080.572.
- Courtin, C. (1997). Does sign language provide deaf children with an abstraction advantage? Evidence from a categorization task. *The Journal of Deaf Studies* and *Deaf Education*, 2(3), 161–171. doi: 10.1093/oxfordjournals.deafed.a014322.
- Courtin, C., Limousin, F., & Morgenstern, A. (2010). Évaluer les compétences linguistiques des enfants en langue des signes française: Une expérience pionnière. Language, Interaction and Acquisition, 1(1), 129–158. doi: 10.1075/lia.1.1.08cou.
- Dean, P. M., Feldman, D. M., Morere, D., & Morton, D. (2009). Clinical evaluation of the mini-mental state exam with culturally deaf senior citizens. Archives of Clinical Neuropsychology, 24(8), 753–760. doi: 10.1093/arclin/acp077.

Drion, B., & Buhler, L. (2016). Access to care in sign language: The French experience. Public Health, 137, 200–203. doi: 10.1016/j.puhe.2016.01.018.

- Drion, B., & Semail, J. (2016). La maltraitance méconnue des sourds locuteurs de la langue des signes en maison de retraite. NPG Neurologie Psychiatrie Gériatrie, 16(91), 17–23. doi: 10.1016/j.npg.2015.08.002.
- Friedman, N. P., & Miyake, A. (2004). The relations among inhibition and interference control functions: A latent-variable analysis. *Journal of Experimental Psychology: General*, 133(1), 101–135. doi: 10.1037/0096-3445.133.1.101.
- Folstein, M., Folstein, S., & McHugh, P. (1975). Mini-mental state: A practical method for grading the cognitive state of patients for the clinician. *Journal of Psychiatric Research*, *12*, 189–198.
- Haug, T., & Mann, W. (2008). Adapting tests of sign language assessment for other sign languages a review of linguistic, cultural, and psychometric problems. *Journal of Deaf Studies and Deaf Education*, 13(1), 138–147. doi: 10.1093/deafed/enm027.
- Herman, R., Holmes, S., & Woll, B. (1999). Assessing British Sign Language development: Receptive skills test. Coleford, UK: The Forest Bookshop.
- Hughes, C., Berg, L., Danziger, W., Coben, L., & Martin, R. (1982). A new clinical scale for the staging of dementia. *British Journal of Psychiatry*, 140(6), 566–572. doi: 10.1192/bjp. 140.6.566.
- Jacus, J. P., & Martin, C. (2000). Cognitive impairment: Interest and limitations of psychometric evaluation. La Revue de Gériatrie, 25(2), 105-113.
- Kalafat, M., Hugonot-Diener, L., & Poitrenaud, J. (2003). The mini mental state (MMS): French standardization and normative data [version GRÉCO]. *Revue de Neuropsychologie*, 13(2), 209–236.
- Koo, T. K., & Li, M. Y. (2016). A guideline of selecting and reporting Intraclass correlation coefficients for reliability research. *Journal of Chiropractic Medicine*, 15(2), 155–163. doi: doi.org/10.1016/j.jcm.2016.02.012.
- Koutsoubou, M., Herman, R., & Woll, B. (2006). Bilingual language profiles of deaf students: An analysis of the written narratives of three deaf writers with different language proficiencies. *Deafness & Education International*, 8(3), 144–168. doi: 10.1179/146431506790560139.
- Leigh, I. W. & Andrews, J. F. (2017). Deaf people and society: psychological, sociological, and educational perspectives. New York: Routledge. doi:10.4324/9781315473819
- Maillet, D., Matharan, F., Le Clésiau, H., Bailon, O., Pérès, K., Amieva, H. et al. (2016). TNI-93: A new memory test for dementia detection in illiterate and low-educated patients. Archives of Clinical Neuropsychology. doi: 10.1093/arclin/acw065.
- Maillet, D., Narme, P., Amieva, H., Matharan, F., Bailon, O., Clésiau, H. et al. (2017). The TMA-93: A new memory test for Alzheimer's disease in illiterate and less educated people. *American Journal of Alzheimer's Disease & Other Dementias*, 32(8), 461–467.
- Maller, S. J., Singleton, J. L., Supalla, S. J., & Wix, T. (1999). The development and psychometric properties of the American Sign Language Proficiency Assessment (ASL-PA). Journal of Deaf Studies and Deaf Education, 4(4), 249–269. doi: 10.1093/deafed/4.4.249.

Marshall, C., Mason, K., Rowley, K., Herman, R., Atkinson, J., Woll, B. et al. (2015). Sentence repetition in deaf children with specific language impairment in British sign language. *Language Learning and Development*, *11*(3), 237–251. doi: 10.1080/15475441.2014.917557.

Mottez, B. (2006). Les Sourds existent-ils ? Bernard Mottez, Textes réunis et présentés par Andréa (Benvenuto. ed.). Paris: L'Harmattan.

Moores, D. F., & Martin, D. S. (Eds.) (2006). Deaf learners: Development in curriculum and instruction. Washington DC: Gallaudet University Press.

- Morere, D. A. (2013). The signed verbal learning test: Assessing verbal memory of deaf signers. Sign Language Studies, 14(1), 39–57. doi: 10.1353/sls.2013.0025.
- Nasreddine, Z. S., Phillips, N. A., Bédirian, V., Charbonneau, S., Whitehead, V., Collin, I. et al. (2005). The Montreal cognitive assessment, MoCA: A brief screening tool for mild cognitive impairment. *Journal of the American Geriatrics Society*, 53(4), 695–699.
- Stokoe, W. C. (1960). Sign language structure: An outline of the visual communication systems of the American deaf. In *Studies in linguistics: Occasional papers (No. 8)*. Buffalo, NY: University of Buffalo, Department of Anthropology and Linguistics.

Sutton-Spence, R., & Woll, B. (1999). The linguistics of British Sign Language: An introduction. Cambridge: Cambridge University Press.