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1	French translation and validation of the Synkinesis Assessment Questionnaire
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23 Financial disclosure: None to declare 24 25 Conflict of interest: None to declare 26 27 Funding: This study did not receive any specific grant from funding agencies in the public, 28 commercial or not-for-profit sectors. 29 30 Word count of Main Text: 1484 31 Word count of Abstract: 100 32 Abstract 33 Synkinesis is a distressing sequela of peripheral facial palsy (PFP). This study aimed to 34 translate and validate the Synkinesis Assessment Questionnaire (SAQ), a reliable patient-35 reported outcome evaluation tool for synkinesis, in French. 36 The SAQ was translated following a standard forward-backward translation procedure. After a cognitive debriefing with 10 PFP patients, the SAQ-F was assessed amongst 50 patients for 37 38 internal consistency, known-group validity, construct validity, criterion validity and test-retest 39 reliability. 40 Results demonstrated that the SAQ-F was valid, reliable and had a unidimensional structure. 41 The SAQ-F should be accompanied by clinician-based scales, to provide valuable additional

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information on the severity of synkinesis.

43 Facial synkinesis is among the most invalidating consequences of peripheral facial palsy (PFP). 44 It is defined as abnormal muscle contractions of one or many facial areas during volitional facial 45 movement¹. Synkinesis has numerous functional and cosmetic adverse effects as it limits 46 several day-to-day activities like speaking and eating². Potential mechanisms for the development of synkinesis could be due to aberrant reinnervation, either by stimulation of 47 48 neighbor axons in the context of myelin loss or due to hyperexcitability of the facial nucleus¹. 49 From a research perspective, the use of a validated universal grading system for synkinesis 50 would allow appropriate data pooling and help in establishing valid recommendations for clinical 51 decision making³. From a clinical perspective, the evaluation of synkinesis through a patient-52 reported outcome measure (PROM) is critical to grasp the scope of the handicap that it causes⁴. 53 Observer-based evaluation of facial function often leads to an incomplete description of patient 54 psychological distress and functional impairments that are caused by the sequelae of facial 55 palsy⁵. The Synkinesis Assessment Questionnaire (SAQ)² was developed as a specific and validated 56 57 PROM for synkinesis. While the original English version has demonstrated to be a reliable and 58 valid instrument, there is no existing French equivalent. The purpose of the present study was to 59 create a validated French version of the SAQ in accordance with international guidelines of 60 translation and cultural adaptation. 61 62 The present study was approved by the ethics review board of the Centre-intégré-universitaire-

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consent was obtained from all participants.

The translation and cultural validation process respected international guidelines³. A standard 65 66 forward and backward-translation procedure was adopted, with two independent certified 67 translators who produced distinct translations from the original to the target language. Those 68 two translations were merged by the senior researcher of the study. A third translator back-69 translated the reconciled version for review and identification of discrepancies. 70 The preliminary version was administered by the first author to 10 native French patients with 71 PFP (women: 8; mean age: 47.4 (15.6)) for cognitive debriefing³. Appropriate minor changes 72 were then made to the preliminary version and the resulting French version of the SAQ (SAQ-F) 73 was used for validation (Figure 1). 74 Validation of SAQ-F was conducted with a prospective cohort study including 25 patients with 75 PFP and 25 controls who visited the Otolaryngology clinic for other indications than a PFP (ear 76 infection, dysphonia, tonsillitis, etc.), from February to April 2020. Inclusion criteria were having 77 a PFP and being 18 years old and older. Exclusion criteria were: 1) history of neurological 78 disorders: 2) active psychiatric disease: 3) cognitive disorder: 4) inability to understand written 79 and oral French. For the PFP participants, the severity of facial palsy was assessed using the 80 Facial Nerve Grading System 2.0 (FNGS 2.0; also known as the House-Brackmann 2.0 score)⁶ and the Sunnybrook Facial Grading System (SB)⁷. These were chosen because each has been 81 82 shown to have high inter-observer agreement and validity⁸. Specific subscores of synkinesis can 83 be calculated from either scale to allow for more specific analyses. Patients completed the SAQ-84 F twice within a two-week interval for test-retest reliability. None of the PFP patients were 85 subject to changes in their treatment. Of 50 respondents, 25 were PFP patients and 25 controls (Table 1), with 20 men (40%) and 30 86 87 women (60%). The average age was 51.6 (18.4) years for the entire sample, 52.7 (18.6) years in PFP and 50.6 (18.4) years in controls. The mean total SAQ score was 18.5 (95% CI 15.7 to

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21.2, median 17, range 17-34) points in PFP group, and 9.0 (no variance) points in controls with a difference of -9.5 (95% CI -12.2 to -6.8) points. and *p*-value <0.0001. Of the PFP patients, 80% were diagnosed with Bell's palsy and the remaining 20% were diagnosed with a PFP secondary to Ramsay-Hunt's Syndrome, facial nerve schwannoma or traumatic injury. Severity of facial palsy was generally rated as light to moderate with both FNGS 2.0 and SB scales: mean FNGS 2.0 score was 9.4 (4.2) and mean SB score was 73.5 (21.5).

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The analyses were performed using Stata/IC Statistical Software: Release 16. College Station (StataCorp LP, TX, USA). The internal consistency of SAQ-F was assessed by using a Chronbach's alpha along with its lower 95% confidence limit (95% CL). Alpha ≥ 0.9 was considered excellent, ≥0.8 good, ≥0.7 acceptable, ≥0.6 questionable and ≥0.5 poor. The knowngroup validity (PFP vs. controls) was assessed by using a t-test for independent groups in case of total score, and the Kruskal-Wallis nonparametric test for the items' ordinal scores. A twotailed p-value ≤ 0.05 was considered statistically significant. The test-retest validity of the SAQ-F scale was assessed by employing a Spearman rank correlation coefficient. Exploratory factor analysis (EFA) was used to approximate the construct structure of the SAQ-F and included only PFP patients. The goal was to determine whether the SAQ-F measures only one latent trait (= signs of facial paralysis) or if there are other possible significant latent variables affecting the results. The results were analyzed graphically. After the orthogonal varimax rotation was applied, retained and excluded factors were explored visually on a scree plot along with a parallel analysis. Pearson's product-moment correlation was used when comparing the SAQ-F total score with the synkinesis subscores obtained from the Sunnybrook and FNGS 2.0 scales. Fisher's transformation was used for both Spearman and Pearson's tests. Correlation <0.2 was considered poor, from 0.21 to 0.4 fair, from 0.41 to 0.6 moderate, from 0.61 to 0.8 substantial, and >0.8 perfect.

Results showed that the internal consistency of the SAQ-F was good with alpha of 0.87 (lower 95% CL 0.82). Results of the test-retest reliability were substantial to perfect for the total score as well as for all nine items individually (0.96, 95% CI 0.93 to 0.98) (Table 2). Known-group validity of SAQ-F appeared to be high as there were significant differences between groups in the total score and in seven out of nine items' scores (p<0.01) (Table 3). Construct validity of the SAQ-F was tested by an exploratory factor analysis (Table 4 and Figure 2). The parallel analysis of the scree plot showed that SAQ-F had three factors with positive eigenvalues above the parallel analysis line. However, the eigenvalues of the second and third factors were as low as 1.2 and 0.6 respectively and were disregarded for retaining. Thus, the SAQ-F was considered to have a unidimensional structure with one factor, whose eigenvalue was 3.1. When assessing the criterion validity based on the 25 PFP patients, Pearson's product-moment correlation of the SAQ-F total score and the FNGS 2.0 synkinesis subscore was not significant (r=-0.23; 95% CI: -0.57 to 0.18). The Spearman's rank correlation of SAQ-F total score with Sunnybrook synkinesis subscore was also not significant (r=-0.19; 95% CI: -0.55 to 0.22). In this study, we presented the translation and validation of the SAQ-F, a French patientcentered questionnaire based on the original English SAQ². The SAQ scale allows to quantify the patient's perception of synkinesis' severity and thus allows to adapt the management and overall care of synkinesis, to fit the patient's expectations. Cross-cultural adaptation and validation is necessary in the use of PROM questionnaires, to avoid misinterpretation while using questionnaires developed in other countries³. To our knowledge, no other study validated the SAQ scale in French. Thus, the SAQ-F will be highly relevant for many clinical and research settings in Quebec and other French-speaking regions. We translated and validated the SAQ-F according to the best practice's international guidelines³. Our results showed that the SAQ-F has good internal consistency, a high test-retest reliability, a high known-group validity, allowing to distinguish between controls and PFP

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139 patients, as well as good construct validity. Compared to the original version, the SAQ-F 140 presented a slightly higher internal consistency (0.87 for SAQ-F and 0.80 for SAQ) and test-141 retest reliability (0.96 in our study and 0.881 in the original one). 142 Correlations with synkinesis subscores of clinician-based questionnaires were not significant. 143 Other studies already reported discrepancies between PROM and clinician-based physical examination⁵. A high correlation between both measures is probably not to be expected, and 144 both of them should be taken for a complete overview of the synkinesis severity^{2,1}. 145 146 This study is not without limitation. As the data comes from a small number of patients, non-147 significant results regarding criterion validity could be due to lack of power. Due to practical 148 reasons, the group size was limited to 25 patients, which is nonetheless comparable with many other PROM studies in the literature about PFP9. Further research may reveal valuable 149 150 information about SAQ-F psychometric properties by employing, for example, item response theory analysis (IRT)¹⁰. 151 152 The SAQ-F was found to be a reliable, easy-to-use and valid unidimensional scale to assess 153 synkinesis after PFP. The SAQ-F should be accompanied by clinician-based scales to provide 154 valuable additional information on the severity of synkinesis. 155 Acknowledgements: The authors would like to thank Anne-Marie Chouinard for her 156 contribution in the development of the research protocol. 157 Statement of Authorship: SM & LG participated in the development of the research protocol, 158 recruitment of patients and the redaction of the article. AR and KM participated in the redaction 159 of the article. MS participated in the analysis of data and redaction of the article. SPM 160 supervised each step of the research.

162 Figure Captions:

SAQ

Veuillez répondre aux questions suivantes concernant la fonction faciale <u>du côté du visage atteint par la paralysie faciale</u>, sur une échelle de 1 à 5, selon les classements suivants :

- 1 = rarement ou pas du tout
- 2 = occasionnellement, ou très légèrement
- 3 = parfois, ou légèrement
- 4 = la plupart du temps, ou modérément
- 5 = tout le temps, ou sévèrement

Question	Score (1 À 5)
1 Lorsque je souris, mon œil se ferme	
2 Lorsque je parle, mon œil se ferme	
3 Lorsque je siffle ou que j'arrondis les lèvres, mon œil ferme	
4 Lorsque je souris, mon cou se tend	
5 lorsque je ferme les yeux, mon visage se contracte	
6 lorsque je ferme les yeux, le coin de ma bouche bouge	
7 lorsque je ferme les yeux, mon cou se tend	
8 Lorsque je mange, mes yeux pleurent	
9 Lorsque je bouge mon visage, des creux apparaissent sur mon mentor	1

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Figure 1. Synkinesis Assessment Questionnaire- French

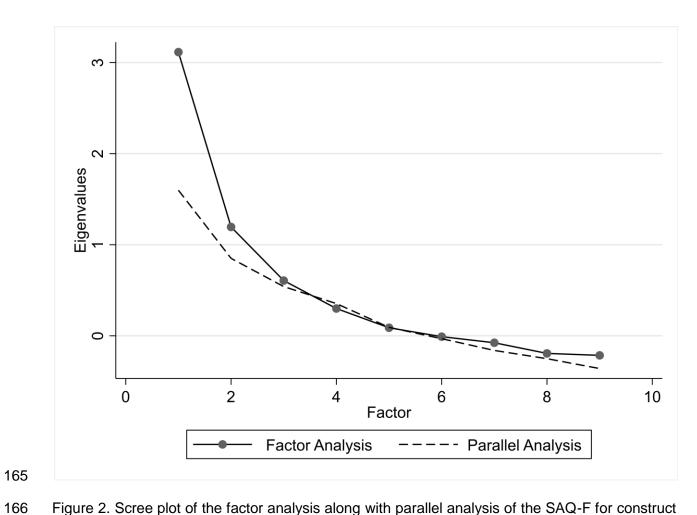


Figure 2. Scree plot of the factor analysis along with parallel analysis of the SAQ-F for construct validity

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Table 1. Patients demographics

	Contr	ols					PFP pa	atients				
	n	%	Mean	SD	Median	Range	n	%	Mean	SD	Median	Range
Gender												
Male	11	44					9	36				
Female	14	56					16	64				
Age (years)			50.6	18.4	56	23-90			52.7	18.6	53	19-95
Diagnosis												
Bells							20	80				
Ramsay Hunt							2	8				
Facial nerve Schwannoma							2	8				
Traumatic injury							1	4				

Side										
Left					11	44				
Right					14	56				
FNGS 2.0 total score							9.44	4.2	9	9-24
Sunnybrook total score							73.5	21.5	76	5-97
SAQ-F total score	9	0	9	9			18.5	17	17	34

Note: PFP= peripheral facial palsy; n = number; SD = standard deviation; Facial Nerve Grading Scale 2.0 (FNGS 2.0) scores: 24 = total palsy; 4 = no facial palsy. Sunnybrook (SB) scores: minimum possible = 0 or total palsy; maximum possible = 100% or normal; SAQ scores: 9 = no synkinesis; 45 = severe synkinesis.

Table 2. Test-retest validity of the SAQ-F (including both PFP and control groups)

	r	95%CI			
		Lower limit	vs upper limit		
Total score	0.96	0.93	0.98		
Item 1	0.78	0.65	0.87		
Item 2	0.92	0.87	0.96		
Item 3	0.93	0.88	0.96		
Item 4	0.92	0.85	0.95		
Item 5	0.69	0.51	0.81		
Item 6	0.90	0.82	0.94		
Item 7	0.60	0.38	0.75		
Item 8	0.89	0.81	0.94		
Item 9	0.81	0.69	089		

Note: SAQ-F= Synkinesis Assessment Questionnaire – French; PFP = peripheral facial palsy; CI = confidence interval .

Table 3. Know-group validity – differences in SAQ-F scores between PFP and control groups.

	<i>p</i> -value
Total score	<0.0001 ^a
Item 1	0.0002 b
Item 2	0.0015 ^b
Item 3	0.0006 ^b
Item 4	0.0033 ^b
Item 5	0.0015 ^b
Item 6	0.0001 b
Item 7	0.0500 ^b
Item 8	0.0041 ^b
Item 9	0.2207 ^b

Note: ^a Independent groups t-test; ^b Kruskal-Wallis test; SAQ-F= Synkinesis Assessment Questionnaire – French; PFP = peripheral facial palsy; CI = confidence interval .

Table 4. Rotated factor loadings (pattern matrix) and unique variances

Factors →	1	2	3	4	5	Uniqueness
Variable ↓						
Item 1	0.10	0.28	0.70	0.02	0.00	0.42
Item 2	0.10	0.87	0.21	-0.10	-0.09	0.16
Item 3	0.19	0.88	0.01	0.08	0.12	0.17
itom 5	0.13	0.00	0.01	0.00	0.12	0.17
	0.00	0.40	0.44	0.40	0.00	0.20
Item 4	0.68	0.12	0.44	0.13	80.0	0.30
Item 5	0.74	0.33	0.09	-0.04	0.08	0.33
Item 6	0.54	0.40	0.13	0.01	0.28	0.45
Item 7	0.84	0.05	-0.05	0.01	-0.07	0.28
Item 8	0.03	-0.15	0.05	-0.34	-0.01	0.86
Item 9	0.13	-0.18	0.19	0.44	0.01	0.72
	0.10	0.10	0.10	0.11	0.01	0.72