

Initial severity of motor and non-motor disabilities in patients with facial palsy: an assessment using patient-reported outcome measures

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Abstract Patients with facial palsy (FP) not only suffer from their facial movement disorder, but also from social and psychological disabilities. These can be assessed by patient-reported outcome measures (PROMs) like the quality-of-life Short-Form 36 Item Questionnaire (SF36) or FP-specific instruments like the Facial Clinimetric Evaluation Scale (FaCE) or the Facial Disability Index (FDI). Not much is known about factors influencing PROMs in patients with FP. We identified predictors for baseline SF36, FaCE, and FDI scoring in 256 patients with unilateral peripheral FP using univariate correlation and multivariate linear regression analyses. Mean age was 52 ± 18 years. 153 patients (60 %) were female. 90 patients (31 %) and 176 patients (69 %) were first seen <90 or >90 days after onset, respectively, i.e., with acute or chronic FP. House–Brackmann grading was 3.9 ± 1.4 . FaCE subscores varied from 41 ± 28 to 71 ± 26 , FDI scores from 65 ± 20 to 70 ± 22 , and SF36 domains from 52 ± 20 to 80 ± 24 . Older age, female gender, higher

House–Brackmann grading, and initial assessment >90 days after onset were independent predictors for lower FaCE subscores and partly for lower FDI subscores (all $p < 0.05$). Older age and female gender were best predictors for lower results in SF36 domains. Comorbidity was associated with lower SF General health perception and lower SF36 Emotional role (all $p < 0.05$). Specific PROMs reveal that older and female patients and patients with chronic FP suffer particularly from motor and non-motor disabilities related to FP. Comorbidity unrelated to the FP could additionally impact the quality of life of patients with FP.

Keywords Facial nerve · Quality of life · Patient-oriented methods · Bell's palsy · Facial nerve reconstruction

Introduction

Nowadays it is self-evident and clinical routine to use at least a subjective grading system like the House–Brackmann grading system, the Sunnybrook Facial Grading System, or the Stennert index (popular in German-speaking countries) to score the severity of a peripheral facial palsy (FP) by an assessment of the facial motor function [1–3]. All these grading systems are applied by the involved physician, but do not take into account the perception of the patient with FP. This is of practical importance, however, as the patients may complain additionally or even primarily from the non-motor disabilities related to FP, i.e., social, communicative, and psychological difficulties [4, 5].

Patient-reported outcome measures (PROMs) are the ideal instruments to address these aspects of FP. Very well-known are generic health-related quality-of-life instruments

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like the Short-Form 36 Item Questionnaire (SF36) [6]. SF36 involves a broad multi-dimensional concept including self-reported measures of physical and mental health. SF36, like other generic instruments, allows a comparison for different diseases, but is not disease-specific [7]. Two validated patient-based facial grading instruments specifically addressing the non-motor disabilities of patients with FP are available but still not routinely used: one is the Facial Disability Index (FDI) and the other tool is the Facial Clinical Evaluation Scale (FaCE) [8, 9].

So far, only one larger recent study in US American patients with peripheral FP has analyzed which factors influence the results of the FaCE Scale instrument [10]. Therefore, we present herewith a first European single-center study on baseline non-motor function of patients with acute and chronic facial palsy analyzing predictors for the PROMs FaCE, FDI, and SF36.

Materials and methods

Patients

We performed a prospective, single-center, observational study among patients with peripheral and unilateral FP. The primary objective was to study baseline motor and especially non-motor disabilities of patients with FP using several PROMs. Data on predictors of changes of the PROMs during follow-up are presented elsewhere [11]. The assessments were performed from August 2012 until February 2015. Only the initial assessments and not the follow-up assessments are presented here. In total, 308 patients were screened for inclusion. Patients with the diagnosis of unilateral and peripheral FP who went completely through comprehensive diagnostics (patient's history, otorhinolaryngological examination, serology, ultrasonography of the neck and other imaging if needed, facial electrodiagnostics, hearing and balance tests, gustatory test) were eligible [12]. Patients with bilateral FP, with an FP arising from central causes, or patients <14 years of age (missing validation of the FaCE and FDI in children) were excluded. Two hundred and fifty-six patients could be included based on these inclusion criteria. Institutional review board approval was obtained from the University Ethics Committee of the Jena University Hospital, Thuringia, Germany.

Questionnaires and other outcomes

The FaCE questionnaire is a validated quality-of-life instrument that is used to assess facial impairment and disability after facial paralysis [9]. It involves 15 statements, each using a five-item Likert scale, whereby 1 corresponds to the lowest level of functioning and 5 corresponds to the highest level of functioning. These statements are

subsequently grouped into six independent domains: social function, facial movement, facial comfort, oral function, eye comfort, and lacrimal control. A total score incorporates all of these domains. Using a specific formula, a score from 0 (worst) to 100 (best) is calculated. A German version of the FaCE questionnaire has been validated recently [13].

The FDI questionnaire is composed of 10 Likert-type questions divided into two domains and includes physical function and social/well-being function [8]. The physical function scale is scored from -25 (worst) to 100 (best), while the social/well-being function scores from 0 (worst) to 100 (best). A German version of the FDI questionnaire has been validated recently [13]. The 36-item SF-36 is a well-established and validated general health status instrument [6]. SF-36 is divided into eight domains and is scored according to published algorithms on a conversion scale from 0 (worst) to 100 (best).

The facial palsy was graded according to the House–Brackmann six-point facial grading system, and also according to the Stennert Index [1, 2]. The Stennert Index separately classifies the face at rest (0–4 points; 0 = normal to 4 = complete loss of resting tone) and during motion (0–6 points; 0 = normal to 6 = no motion). Clinically, the palsy was defined as complete if the patient presented with a complete loss of motor function in the affected hemiface or if the palsy deteriorated to a complete palsy during the inpatient course of treatment. Otherwise, the palsy was defined as incomplete.

Statistical analysis

All statistical analyses were performed using IBM SPSS (version 22.0.0; IBM, New York). If not otherwise indicated, data are presented as mean \pm standard deviation (SD). Pearson's correlation coefficients were calculated to measure the strength of association between the different subscales of the three questionnaires (FDI; FaCE; SF-36) and several patient characteristics. Significant associations ($p < 0.05$) were included into a multivariate linear logistic regression model for the analysis of potential independent predictors of these changes. If both clinical grading scales, House–Brackmann grading and Stennert index, were significantly associated to a subscore, only House–Brackmann grading was included in the regression model. In general, all p values are results of two-sided tests. The significance level was set at $p < 0.05$.

Results

Patient characteristics and severity of facial palsy at baseline

Patient and clinical characteristics of all 256 patients with FP are listed in Table 1. The majority of the patients were

women (60 %). The mean age was 52 ± 18 years. The majority was married or lived in a permanent partnership, was employed, and had no relevant comorbidity. Nearly half of the patients had an idiopathic FP (45 %). Traumatic lesions were the second most frequent reason (36 %). About two-thirds of the patients presented with a long-term FP (assessment >90 days after onset; 69 %) and about one-third early after onset of the FP (assessment <90 days after onset; 31 %). Most patients presented with a clinically incomplete palsy (82 %). Of the patients with chronic FP, 35 % showed a defective healing of the FP. Baseline House–Brackmann grading was 4 ± 1.3 , initial Stennert index at rest 1.6 ± 1.2 , and Stennert index in motion 3.8 ± 1.8 . Details on the patients who underwent facial reanimation surgery prior to the assessment are given in Supplement Table 1.

Baseline non-motor disabilities: FaCE, FDI, and SF36 subscores

All patients filled out the three questionnaires (FaCE, FDI, SF36). The results are presented in Fig. 1 and Supplement Table 2. The lowest scores were recorded for the FaCE Facial movement subscore (41 ± 28), the FaCE Eye comfort subscore (48 ± 34), the FDI Physical function (65 ± 20), the SF36 Vitality (52 ± 20), and the SF36 General health perception subscore (57 ± 22). There were strong correlations between the FaCE Total score and the FDI Total score ($r = 0.681$; $p < 0.0001$; Supplement Table 3), the SF36 General health perception and the FaCE Total score ($r = 0.473$; $p < 0.0001$), and the SF36 General health perception and the FDI Total score ($r = 0.480$; $p < 0.0001$).

Multivariate analysis of predictors for FaCE, FDI, and SF36 subscores

The results of the univariate correlation analyses are summarized in Supplement Tables 4–6. Some of the parameters of the univariate analyses were confirmed as independent predictors in the multivariate linear regression analyses (Tables 2, 3): complete palsy ($p = 0.027$), higher House–Brackmann grading ($p < 0.0001$) and an interval onset to assessment >90 days ($p = 0.021$) were independent predictors for worse FaCE Facial movement scoring. An interval onset to assessment >90 days was also a predictor for lower FaCE Facial comfort ($p = 0.001$) and lower FaCE Total score ($p = 0.026$). Lower FaCE Oral function was independently predicted by higher House–Brackmann grading ($p = 0.003$). Higher age was an independent predictor for lower FaCE Eye comfort ($p = 0.003$), FaCE Lacrimal control ($p = 0.037$), FaCE Social function ($p = 0.044$), and FaCE Total score

($p = 0.020$). Female patients had a lower FaCE Social function ($p = 0.021$) and a lower FaCE Total score ($p = 0.029$). Age was also an important independent predictor for the FDI scoring and SF36 results: older patients had a lower FDI Physical function ($p = 0.003$), FDI Social/well-being function ($p < 0.0001$), SF36 Physical functioning ($p < 0.0001$), SF36 Physical role ($p < 0.0001$), SF36 General health perception ($p = 0.023$), SF36 Social functioning ($p = 0.023$), and SF36 Mental health ($p = 0.036$). Females had a lower FDI Social/well-being function ($p = 0.024$) and SF36 Physical functioning ($p = 0.028$). A higher baseline House–Brackmann grading was independently correlated to poorer FDI Physical function ($p = 0.008$), SF36 Physical functioning ($p = 0.015$), and SF36 Social functioning ($p = 0.016$).

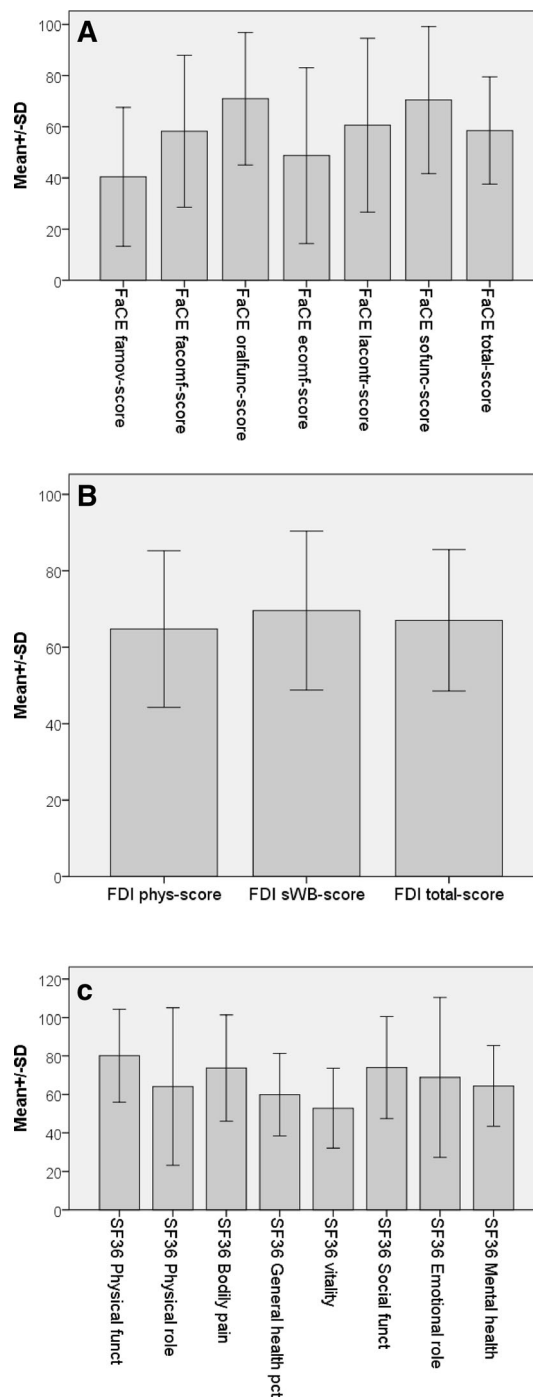
Discussion

The PROMs used show that patients with FP not only severely suffer from motor, but also non-motor disabilities. FaCE and FDI measure the facial movement disorder from the patient's perspective. Nevertheless, the number of studies on patients with FP using such PROMs is very limited (overview of the publications at the current time in Supplement Table 7). Mostly, as in the present study, the total scores as well as the subscores of these PROMs show a high negative correlation to the classical grading systems like House–Brackmann grading, Stennert index, or Sunnybrook grading (cf. Supplement Table 7). Not surprisingly, the correlation with the classical grading system results was higher for those subscores of the PROMs directly addressing physical aspects of the movement disorder than for the non-motor subscores [7, 14, 15]. In the present analysis, only a low or non-significant association was found between clinical grading, as an expression of facial motor dysfunction, and FaCE Facial Comfort, FDI Social/well-being function, SF36 Physical role, SF36 Bodily Pain, SF36 General health perception, SF36 Vitality, SF36 Emotional role, and SF36 Mental health. This underlines that the severity of the facial movement disorder of patients with FP is not (directly or indirectly) associated with all complaints and impairments of the patients—i.e., some complaints, especially social, mental, and psychological disabilities, can occur in patients with weak FP or can even be unrelated to the palsy but related to their comorbidity.

When we compare the absolute magnitude of the different baseline subscores of the PROMs in the present studies to the results given in the literature (cf. Supplement Table 7, last column), it soon becomes clear that the variability in the results of the PROMs cannot only be explained by the variability of the severity of the motor disabilities in the different study samples of patients with

Table 1 Patients' characteristics ($n = 256$)

Parameter	Absolute	Relative (%)
Gender		
Female	153	60
Male	103	40
Marital status		
Married/permanent partnership	92	36
Single/divorced/widowed	41	16
Unknown	123	48
Occupational group		
Employee	97	38
Unemployed	2	1
Retiree	58	23
Student	9	4
Other	2	1
Unknown	88	34
Charlson Comorbidity Index		
0	73	61
1	18	15
2	12	10
3	4	3
Unknown	13	11
Affected side of facial palsy		
Left	141	55
Right	115	45
Etiology of facial palsy		
Idiopathic	116	45
Traumatic/postsurgical	93	36
Inflammatory/infectious	33	13
Neoplastic	36	2
Congenital	6	2
Other	2	1
Severity		
Complete palsy	47	18
Incomplete palsy	209	82
Defective healing at baseline		
No	166	65
Yes	90	35
Interval onset of facial palsy to assessment		
<90 days	80	31
>90 days	176	69
	Mean \pm SD	Median, range
Age (years)	52 \pm 18	54, 14–93
Interval onset to 1st assessment (days)	1447 \pm 3188	287, 0–23,690
House–Brackmann scale, baseline	3.9 \pm 1.4	4, 2–6
Stennert index at rest, baseline	1.4 \pm 1.3	1, 0–4
Stennert index in motion, baseline	3.5 \pm 1.8	3, 0–6

**Fig. 1** Results of the initial FaCE (a), FDI (b), and SF-36 (c) scoring for the different subscores and domains in mean \pm SD. *FaCE* Facial Clinimetric Evaluation Scale, *FDI* Facial Disability Index, *SF36* Short-Form health survey 36

FP. There are other independent predictors: older age, female gender, longer duration of the palsy (>90 days, i.e., a chronic palsy in most cases) were negative predictors for the PROMs. Moreover, comorbidity was an independent predictor for some of the SF36 subdomains, but did not

Table 2 Linear regression analysis for independent predictors of the FaCE subscores

Measure	β	95 % CI lower	95 % CI upper	Stand. ^a β	<i>p</i>
FaCE Facial movement; $R^2 = 0.396$, $p < 0.0001$					
Occupational group (0 = unemployed; 1 = employed)	-0.002	-0.008	0.004	-0.037	0.504
Severity of palsy at onset (1 = incomplete; 2 = complete)	9.799	1.122	18.477	0.142	0.027
House-Brackmann at onset	-13.023	-15.573	-10.474	-0.658	<0.0001
Interval onset to 1st assessment (1 \leq 90 days; 2 \geq 90 days)	-7.783	-14.399	-1.168	-0.134	0.021
Surgical treatment (0 = no; 1 = yes)	-2.706	-11.022	5.610	-0.037	0.522
FaCE Facial comfort; $R^2 = 0.046$, $p = 0.001$					
Interval onset to 1st assessment (1 \leq 90 days; 2 \geq 90 days)	-13.922	-21.809	-6.035	-0.215	0.001
FaCE Oral function; $R^2 = 0.037$, $p = 0.003$					
House-Brackmann at onset	-3.793	-6.244	-1.343	-0.192	0.003
FaCE Eye comfort; $R^2 = 0.256$, $p < 0.0001$					
Age (years)	-0.329	-0.547	-0.111	-0.168	0.003
Severity of palsy at onset (1 = incomplete; 2 = complete)	5.224	-6.744	17.192	0.059	0.391
House-Brackmann at onset	-11.162	-14.629	-7.696	-0.441	0.0001
Surgical treatment (0 = no; 1 = yes)	-11.495	-22.315	-0.674	-0.122	0.037
FaCE Lacrimal control; $R^2 = 0.077$, $p = 0.009$					
Age (years)	-0.326	-0.632	-0.020	-0.167	0.037
Comorbidity (0 = no; 1 = yes*)	-7.428	-18.524	3.668	-0.103	0.188
Stennert index** at onset	-1.275	-3.076	0.526	-0.110	0.164
Surgical treatment (0 = no; 1 = yes)	-6.375	-20.526	7.776	-0.069	0.375
FaCE Social function; $R^2 = 0.112$, $p < 0.0001$					
Gender (0 = male; 1 = female)	-8.519	-15.722	-1.317	-0.144	0.021
Age (years)	-0.206	-0.406	-0.006	-0.125	0.044
Occupational group (0 = unemployed; 1 = employed)	-0.006	-0.014	0.002	-0.100	0.125
House-Brackmann at onset	-3.655	-6.261	-1.049	-0.171	0.006
Interval onset to 1st assessment (1 \leq 90 days; 2 \geq 90 days)	-7.968	-15.980	0.044	-0.127	0.051
FaCE Total score; $R^2 = 0.208$, $p < 0.0001$					
Gender (0 = male; 1 = female)	-5.569	-10.550	-0.588	-0.129	0.029
Age (years)	-0.165	-0.304	-0.027	-0.137	0.020
Occupational group (0 = unemployed; 1 = employed)	-0.004	-0.009	0.002	-0.081	0.187
Severity of palsy at onset (1 = incomplete; 2 = complete)	5.019	-2.621	12.659	0.092	0.197
House-Brackmann at onset	-6.057	-8.235	-3.880	-0.390	<0.0001
Interval onset to 1st assessment (1 \leq 90 days; 2 \geq 90 days)	-6.292	-11.817	-0.768	-0.138	0.026

p-values <0.05 are shown in bold

FaCE Facial Clinimetric Evaluation Scale, CI confidence interval

* If Charlson comorbidity score >0

** Sum of Stennert index at rest and index during motion

^a Standardized β

play a role for the FP-specific PROMs FaCE and FDI. Age was also an important independent predictor for SF36 results in FP patients after facial nerve reconstruction [16]. Older age, female gender, and duration of FP were also independent predictors for the FaCE in a mixed sample of US American patients with FP [10]. In contrast, age and duration were no predictors for FaCE in a sample of patients with FP after vestibular schwannoma surgery [17]. In another study on patients with Bell's palsy and patients with FP after vestibular schwannoma surgery, age and

gender also did not predict FaCE, but a better informed consent about the sequences of FP at onset of FP [18]. The present study only contained 10 patients with FP after vestibular schwannoma surgery. Therefore, a meaningful subgroup analysis for these patients cannot be performed within the present sample. Predictors for FDI beyond clinical grading were so far not analyzed in other studies.

The detected predictors which are independent of the motor disabilities of the patients show how important it is to apply PROMs in patients with FP. Moreover, it seems

Table 3 Linear regression analysis for independent predictors of the FDI and SF36 subscores

Measure	β	95 % CI lower	95 % CI upper	Stand. ^a β	<i>p</i>
FDI Physical function; $R^2 = 0.084$, $p < 0.0001$					
Age (years)	-0.217	-0.358	-0.077	-0.189	0.003
Severity of palsy at onset (1 = incomplete; 2 = complete)	0.231	-7.595	8.056	0.004	0.954
House–Brackmann at onset	-2.986	-5.202	-0.771	-0.201	0.008
FDI Social/well-being function; $R^2 = 0.125$, $p < 0.0001$					
Gender (0 = male; 1 = female)	-5.766	-10.751	-0.782	-0.136	0.024
Age (years)	-0.268	-0.404	-0.133	-0.230	<0.0001
Occupational group (0 = unemployed; 1 = employed)	-0.004	-0.009	0.002	-0.083	0.182
Interval onset to 1st assessment (1 \leq 90 days; 2 \geq 90 days)	-8.000	-13.511	-2.490	-0.178	0.005
FDI Total; $R^2 = 0.113$, $p < 0.0001$					
Age (years)	-0.265	-0.394	-0.137	-0.247	<0.0001
House–Brackmann at onset	-1.432	-3.093	0.229	-0.103	0.091
Interval onset to 1st assessment (1 \leq 90 days; 2 \geq 90 days)	-7.586	-12.406	-2.766	-0.187	0.002
SF36 Physical functioning; $R^2 = 0.282$, $p < 0.0001$					
Gender (0 = male; 1 = female)	-7.362	-13.938	-0.787	-0.151	0.028
Age (years)	-0.526	-0.729	-0.324	-0.371	<0.0001
Comorbidity (0 = no; 1 = yes*)	-7.439	-14.607	-0.272	-0.146	0.042
Etiology (0 = not idiopathic; 1 = idiopathic)	4.026	-2.783	10.836	0.083	0.245
Severity of palsy at onset (1 = incomplete; 2 = complete)	8.534	-1.817	18.885	0.137	0.105
House–Brackmann at onset	-3.686	-6.652	-0.720	-0.208	0.015
Surgical treatment (0 = no; 1 = yes)	-7.794	-17.657	2.068	-0.114	0.121
SF36 Physical role; $R^2 = 0.058$, $p < 0.0001$					
Age (years)	-0.557	-0.847	-0.268	-0.241	<0.0001
SF36 General health perception; $R^2 = 0.074$, $p = 0.002$					
Age (years)	-0.203	-0.381	-0.025	-0.175	0.026
Comorbidity (0 = no; 1 = yes*)	-6.942	-13.522	-0.361	-0.162	0.039
SF36 Social functioning; $R^2 = 0.051$, $p = 0.002$					
Age (years)	-0.225	-0.419	-0.031	-0.147	0.023
House–Brackmann at onset	-3.052	-5.536	-0.568	-0.155	0.016
SF36 Emotional role; $R^2 = 0.061$, $p = 0.017$					
Gender (0 = male; 1 = female)	-9.842	-22.358	2.674	-0.119	0.122
Age (years)	-0.274	-0.645	0.097	-0.117	0.147
Comorbidity (0 = no; 1 = yes*)	-13.232	-27.109	0.646	-0.152	0.062
SF36 Mental health; $R^2 = 0.019$, $p = 0.036$					
Age (years)	-0.163	-0.315	-0.011	-0.137	0.036

p-values <0.05 are shown in bold

FDI Facial Disability Index, SF36 Short-Form health survey 36, CI confidence interval

* If Charlson comorbidity score >0

^a Standardized β

that using the FaCE and the FDI together may detect more facets of the non-motor disabilities than just one of the two FP-specific scores. Finally, the present study shows how valuable it is also to use a generic and non-specific PROM like SF36. The patients may have other diseases, i.e., comorbidities, which influence their well-being and quality of life and thereby might intensify the morbidity related to the non-motor disabilities of the FP.

The present study is not without limitations. Due to the retrospective design, some subgroups of patients like patients after vestibular schwannoma surgery may be underrepresented (see above) or completely missing (like patients with facial reanimation by free flaps [19]). Furthermore, FaCE, FDI, and SF36 may not cover all important non-motor disabilities of patients with FP, like for instance self-perception of facial appearance, procedure-related symptoms, and

procedural satisfaction [20]. Nevertheless, such a comprehensive analysis of initial disabilities of patients with FP as in the present study including FACE, FDI, and SF36 at the same time has not yet been performed [21–33].

What should be the consequence of the presented results? Others have recommended that a psychologist, specialized in the psychosocial consequences of FP, should be part of the multidisciplinary FP team to counsel all patients suffering from FP [10]. Whether such psychological counseling can indeed ameliorate potential psychosocial dysfunction in these patients should be the subject of future investigations. Any future controlled clinical trial in patients with FP, whether analyzing a drug effect in Bell's palsy or results of a special surgical facial reanimation procedure, should include PROMs as outcome measures, as the non-motor disabilities are no less important for patients with FP than the motor disabilities.

Conclusion

This prospective, single-center cohort study of 256 patients with FP confirms that the patients also suffer severely from social and psychological disabilities. Patients with chronic FP showed more disabilities than patients with acute FP. Older age and female gender were robust predictors for more social and psychological disabilities using the more general quality-of-life score SF36 as well as when using the FP-specific scores FaCE and FDI. The age and gender effect has to be examined more profoundly in future trials.

Compliance with ethical standards

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Conflict of interest All authors declare that they have no conflict of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from all individual participants included in the study.

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