

IT-based Psychosocial Distress Screening in Patients with Sarcoma and Parental Caregivers *via* Disease-specific Online Social Media Communities

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Abstract. *Background:* Psychosocial distress can be frequently observed in patients with sarcoma, depicting a relevant clinical problem. However, prospective data collection on psychosocial distress in patients with rare tumors is often time-consuming. In this context, social media such as Facebook can serve as a potential platform to expand research. The aim of this study was to assess the feasibility of psychosocial distress screening in patients with osteosarcoma and Ewing's sarcoma via social media. *Materials and Methods:* For this study an online questionnaire including general information and self-assessment distress measurement tools for patients and parents was created. The link to the questionnaire was then posted on the main page of the two largest disease-specific Facebook communities on osteosarcoma and Ewing's sarcoma. *Results:* Within 2 months, 28 patients and 58 parents of patients were enrolled. All patients with osteosarcoma and Ewing's sarcoma, as well as the majority of parental caregivers of such patients, showed relevant psychosocial distress levels. *Conclusion:* Crowdsourcing via disease-specific patient communities on Facebook is feasible and provides great potential for acquisition of medical data of rare diseases.

Psychosocial distress can be observed in 30-45% of all patients with cancer during the course of their disease, and is an important clinical problem (1-3). In particular, orthopedic patients with primary malignant bone tumors, such as osteosarcoma (OS) and Ewing's sarcoma (ES), experience high levels of distress due to the malignancy of these tumors and often disabling surgical treatment (4-6). However, few patients with distress are detected early in the treatment process and offered adequate psychological support (7, 8). This low detection rate may be explained by low correlations between the objective characteristics of the disease and the patient's subjective perception (9, 10). Furthermore, only 25% of patients discuss emotional concerns with their physician without being invited to do so (11).

International guidelines suggest routine psychosocial distress screening for all patients with cancer and provision of psychological support as needed (8, 12). However, routine psychosocial distress screening is not an integral component of sarcoma treatment guidelines despite the high demand (6).

Most common screening tools are long questionnaires with multiple items, which are not generally accepted in clinical practice. Consequently, short forms of validated distress screening tools have been developed (2, 7, 8, 13). A popular tool for self-assessment of psychosocial distress in daily life among patients with cancer is the Questionnaire on Stress in Cancer Patients (QSC-R23). This tool was originally developed in 1985 by Herschbach and colleagues, who demonstrated high reliability and validity in a sample of 1,645 patients with cancer (14, 15). In order to increase clinical acceptance, Book *et al.* adapted a 10-item short form of the QSC-R23 (QSC-R10) (2).

Parents of these predominantly young patients also suffer from severe psychosocial distress, particularly fear of disease progression, as described by Schepper *et al.* (10) and Fidika

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et al. (16, 17). These authors used the short form (12 items) of the Fear of Progression Questionnaire (FoP-Q-SF), another self-assessment tool, and modified it to capture the parental perspective (FoP-Q-SF/PR) (16-18).

A major problem in determining psychosocial distress in large cohorts of patients with OS or ES is their rarity, as they account for fewer than 1% of all cancers diagnosed in the United States per year (19). Prospective data collection on psychosocial distress in these patients is time-consuming and often limited to a single institution or single country in multicenter studies. In this context, social media such as Facebook are potential platforms to expand research (20). Specific topic-based groups bring together patients with rare diseases (*e.g.* sarcomas) from all over the world, providing unique potential for collecting medical data for large cohorts (crowdsourcing).

Therefore, we hypothesized that (i) social media platforms would facilitate data collection on rare diseases with inclusion of significantly more participants than those undergoing treatment in a single institution during the same time period, (ii) self-assessment questionnaires would be particularly suitable for crowdsourcing, and (iii) patients with OS or ES, as well as parents of such patients, would exhibit high levels of psychosocial distress.

Materials and Methods

Online questionnaire and social network. For this study, we developed an online questionnaire including general sociodemographic and medical information. The questionnaire was combined with previously published self-assessment distress measurement tools for patients and parents, as described below. The link to the questionnaire was posted along with detailed information about the purpose of the study and instructions for completing the questionnaire on the main page of the two largest specific topic-based Facebook communities. To evaluate patients with osteosarcoma and parental caregivers, we chose the Facebook group "Osteosarcoma-Survivors, Family, and Friends", with 2649 current members. For Ewing's sarcoma, we selected the group "Ewing's Sarcoma Survivors," with 1839 members. The survey was closed after 2 months (December 2014 and January 2015) and the collected data were analyzed.

General information. General information was subdivided by patients and parents. Items for patients included information on age, sex, tumor location, treatment, offered psychological support, and satisfaction with treatment and psychological support (if offered). For parents, only age and sex were collected.

QSC-R10. The QSC-R10 is a 10-item psycho-oncological screening instrument for self-assessment of distress, previously published by Book *et al.* (2). It is based on the QSC-R23 screening tool developed and validated by Herschbach and colleagues (15). Item selection for the QSC-R10 was based on intercorrelations and factor loadings.

Each item is scored from 0-5 points, with 0 points indicating no distress and 5 points indicating a high level of distress. The total score is calculated by summing the item scores. A value above 14 points indicates a relevant level of distress, possibly requiring psychooncological treatment (2).

FoP-Q-SF/PR. The FoP-Q-SF/PR comprises 12 items and is based on the FoP-Q-SF for partners, developed by Zimmermann *et al.* (18). The item content was appropriately modified to reflect the parents' perspective.

Response categories range from 1 (never) to 5 (very often), resulting in a total score between 12 and 60 points. A value above 32 points indicates a relevant level of distress, potentially requiring psycho-oncological treatment.

Statistical analyses. Statistical analyses were performed with SPSS Statistics (IBM, Armonk, NY, USA). Descriptive statistics were calculated for sample characteristics in patients and parents. For each QSC-R10 and FoP-Q-SF/PR item, we calculated means and standard deviations as well as skewness and kurtosis. Internal consistency was assessed with Cronbach's alpha. Results for OS and ES, patients and parents, were compared using a *t*-test for unpaired samples. Additionally, regression analysis was performed to evaluate the influence of age, gender or the presence of metastases on psychosocial distress levels. A *p*-value of 0.05 or less was considered statistically significant.

Results

Study sample. During the 2-month study period, data were collected for 28 patients with OS (*n*=14) and ES (*n*=14). We also collected data for 58 parents of patients (not necessarily parents of the included patients) (OS: 17; ES: 41), including 45 mothers and 13 fathers (Tables I and II).

There was an equal sex distribution of patients with OS, whereas those with ES were mainly female (79%). The mean age at diagnosis was 24.8 (range=6-50; SD=13.0) years for patients with OS (median=21.5 years) and 28.8 (range=16-24; SD=9.7) years for those with ES (median=30 years). Time since initial diagnosis was 13.7 years for those with OS and 5.5 years for those with ES. In over 50% of all cases, the period from first symptoms to initial diagnosis was more than 3 months. In our sample, OS was predominately located around the thigh (50%) and lower leg (21%). ES mainly presented in the lower leg (21%) and in extraskeletal locations (50%). A majority of patients underwent chemotherapy and surgical tumor resection (OS=93%; ES=79%). Additional radiotherapy was applied in 50% of patients with ES. Occasional or permanent disability in daily life was indicated by 86% of patients with OS and 50% of those with ES. Psycho-oncological treatment was offered in less than 50% of all cases (OS=43%; ES=36%).

Parental caregivers of patients with OS had a mean age of 47.5 years and those of patients with ES had a mean age of 47.0 years. Parents were predominately mothers (OS=65%; ES=83%).

Internal consistency. Cronbach's alpha was 0.77 ($\alpha=0.78$ to 0.76 for OS and ES subsamples) for the QSC-R10 and 0.80 for the FoP-Q-SF/PR ($\alpha=0.77$ to 0.82 for OS and ES subsamples) indicating a good reliability.

Table I. Sociodemographic and medical characteristics of the participants (patients with osteosarcoma and Ewing's sarcoma) (n=28).

	Osteosarcoma	Ewing's sarcoma
N	14	14
Sociodemographic characteristics		
Gender (n, %)		
male	7 (50)	3 (21)
female	7 (50)	11 (79)
Age at diagnosis (years)		
Mean (SD)	24.8 (13.0)	28.8 (9.7)
Min.	6	16
Max.	50	44
Time since diagnosis (years)		
Mean (SD)	13.7 (10.1)	5.5 (5.1)
Min.	1	1
Max.	31	17
Medical characteristics		
Time to diagnosis (n, %)		
4 weeks or less		1 (7)
1-3 months	2 (14)	0
3-6 months	4 (29)	0
over 6 months	1 (7)	5 (36)
not specified	7 (50)	5 (36)
Location of the tumor (n, %)		
Upper arm/Shoulder	0	0
Forearm	1 (7)	0
Hand/Wrist	0	0
Pelvis	0	0
Thigh	0	0
Lower leg	7 (50)	0
Foot/Ankle	3 (21)	3 (21)
Spine	0	1 (7)
Other	0	0
not specified	3 (21)	7 (50)
Metastases (n, %)	0	3 (21)
Therapy (n, %)		
Chemotherapy	1 (7)	0
Radiotherapy	0	0
Surgery	0	0
Endoprosthesis	0	0
Biological Reconstruction	7 (50)	0
Resection only	3 (21)	3 (21)
Rotationplasty	0	1 (7)
Amputation	0	0
Not specified	2 (14)	0
Handicapped in daily life (n, %)		
No	0	3 (21)
Sometimes	2 (14)	3 (21)
Always	8 (57)	5 (36)
Severely handicapped	4 (29)	2 (14)
Not specified	0	1 (7)
Psycho-oncological treatment offered (n, %)		
Yes	0	3 (21)
No	6 (43)	5 (36)
Not specified	8 (57)	6 (43)
Not specified	0	3 (21)

Table II. Sociodemographic characteristics of the participating parents of patients with osteosarcoma and Ewing's sarcoma (n=58).

	Osteosarcoma	Ewing's sarcoma
N	17	41
Gender (n; %)		
Male	6 (35)	7 (17)
Female	11 (65)	34 (83)
Age (years)		
Mean (SD)	47.5 (6.8)	47.0 (7.7)

Item analyses. The item analysis of the QSC-R10 is presented in Table III. Mean scores for the items ranged from 1.54-4.69 in patients with OS and from 1.56-4.22 in patients with ES. The highest mean was found for item 8 "It is harder for me to take part in recreational activities (e.g. sports) now than it was before I became ill" and the lowest was for item 9 "I do not feel well informed about my disease/treatment" in both the OS and ES groups.

The item analysis of the FoP-Q-SF/PR is presented in Table IV. In the OS group, means for each item ranged from 2.50-4.75. The ES group showed values from 2.34-4.61. In both groups of parents, item 10 "I worry that the medication could damage my child's body" had the highest mean, and item 8 "I am worried that at some point in time, because of my child's illness, I will no longer be able to pursue my hobbies" had the lowest mean.

Psychosocial distress level. The QSC-R10 for the OS patient group had an overall mean of 29.46±9.41 (range=15-43). The ES group had a mean of 32.00±9.25 (range=21-47). All results for both groups were above the cut-off value of ≤14 points (Figure 1a). Psycho-oncological treatment was offered in 43% of OS cases and 36% of ES cases. No difference in distress level was identified between patients with OS and ES ($p=0.54$) nor between male and female patients with OS ($p=0.20$) or ES ($p=0.53$).

Overall results for the FoP-Q-SF/PR showed a mean of 48.19±7.71 for the OS parent group and of 47.03±6.71 for the ES parent group. The lowest individual result for the OS parent group was 31 and for the ES parent group was 32, and a maximum of 60 was identified in both groups (cut-off value 34 points) (Figure 1b). In total, 93.8% of the OS parent group and 97.4% of the ES parent group were classified as having high fear of disease progression. Again, no difference in distress level was found between parents of patients with OS and those with ES ($p=0.60$), nor between mothers and fathers of patients with OS ($p=0.78$) or ES ($p=0.35$) patients.

No significant influence of age, gender or presence of metastases on psychosocial distress levels was found in the

Table III. Item characteristics and component loading of the QSC-R10 questionnaire for osteosarcoma and Ewing's sarcoma patients.

Item	Osteosarcoma				Ewing's sarcoma			
	Mean	SD	Skewness	Kurtosis	Mean	SD	Skewness	Kurtosis
1. I feel tired and weak.	2.92	1.50	0.15	-1.20	3.44	1.59	-1.43	2.02
2. I have pain.	2.69	1.89	-0.44	-1.38	2.33	1.87	-0.03	-1.69
3. I feel physically imperfect.	3.92	1.04	-0.35	-1.16	4.11	1.05	-1.09	0.61
4. I have too few opportunities to speak to a professional about my psychological distress.	2.69	1.84	-0.13	-1.38	2.44	1.67	-0.29	-0.40
5. I am afraid that my disease will spread/recur.	2.69	1.65	-0.07	-1.28	4.33	1.66	-2.80	7.97
6. It is difficult for my partner to empathize with my situation.	2.54	2.07	0.07	-1.75	2.44	1.81	-0.21	-1.32
7. My sleep is disturbed.	3.00	1.96	-0.47	-1.47	3.00	2.06	-0.55	-1.40
8. It is harder for me to take part in recreation activities (e.g. sports) now than it was before I became ill.	4.69	0.63	-2.05	3.71	4.22	1.64	-2.63	7.24
9. I do not feel well informed about my disease/treatment.	1.54	1.66	1.52	1.55	1.56	1.81	1.18	0.27
10. I feel tense and/or nervous.	2.77	1.42	0.27	-1.09	4.11	1.05	-1.09	0.61

Table IV. Item characteristics and component loading of the FoP-Q-SF/parents questionnaire for parents of patients with osteosarcoma and Ewing's sarcoma.

Item	Osteosarcoma				Ewing's sarcoma			
	Mean	SD	Skewness	Kurtosis	Mean	SD	Skewness	Kurtosis
1. I become anxious if I think the disease may progress.	4.63	0.50	-0.57	-1.93	4.58	0.79	-2.85	10.59
2. I am nervous prior to doctors' appointments or periodic examinations.	4.13	1.26	-1.41	1.23	4.47	0.73	-1.02	-0.29
3. I am afraid that my child may have pain.	4.38	0.96	-1.42	1.10	4.39	0.97	-1.82	3.26
4. I have concerns about my child reaching his/her professional goals because of his/her illness.	4.25	1.00	-1.03	-0.14	4.39	0.89	-1.87	4.52
5. When I am anxious, I have physical symptoms (e.g. rapid heartbeat, stomach ache, nervousness).	4.06	1.06	-0.90	-0.26	4.13	0.96	-0.85	-0.25
6. The possibility of my other children contracting the disease disturbs me.	3.31	1.49	-0.20	-1.50	2.76	1.55	0.23	-1.44
7. It disturbs me that my child may have to rely on strangers for activities of daily living.	3.31	1.45	-0.33	-1.38	3.00	1.38	-0.07	-1.04
8. I am worried that at some point in time, because of my child's illness, I will no longer be able to pursue my hobbies.	2.50	1.46	0.44	-1.10	2.34	1.34	0.61	-0.65
9. I am afraid of severe medical treatments over the course of the illness.	4.63	0.62	-1.50	1.58	4.39	0.79	-1.20	0.92
10. I worry that the medication could damage my child's body.	4.75	0.58	-2.38	5.31	4.61	0.82	-2.81	9.46
11. I worry about what will become of my family if something should happen with my child.	4.38	1.15	-2.07	4.26	4.24	0.85	-0.77	-0.39
12. The thought that my child might not be able to work due to his/her illness disturbs me.	3.88	1.09	-0.43	-1.10	3.71	1.18	-0.54	-0.46

OS ($r=-0.31, p=0.30$; $r=0.41, p=0.16$; $r=-0.46, p=0.11$, respectively) and ES ($r=0.38, p=0.32$; $r=-0.28, p=0.47$; $r=0.09, p=0.82$, respectively) patient group. Similarly, no significant correlation between distress level and age or gender was identified for parents of patients with OS ($r=0.43, p=0.09$; $r=0.07, p=0.81$, respectively) and ES ($r=-0.19, p=0.25$; $r=-0.14, p=0.39$, respectively).

Discussion

In the present study, we assessed psychosocial distress in patients with a history of OS or ES and in parents of patients with OS or ES. Our survey used validated psychoncological screening tools and was conducted online via specific topic-based groups in the Facebook social network.

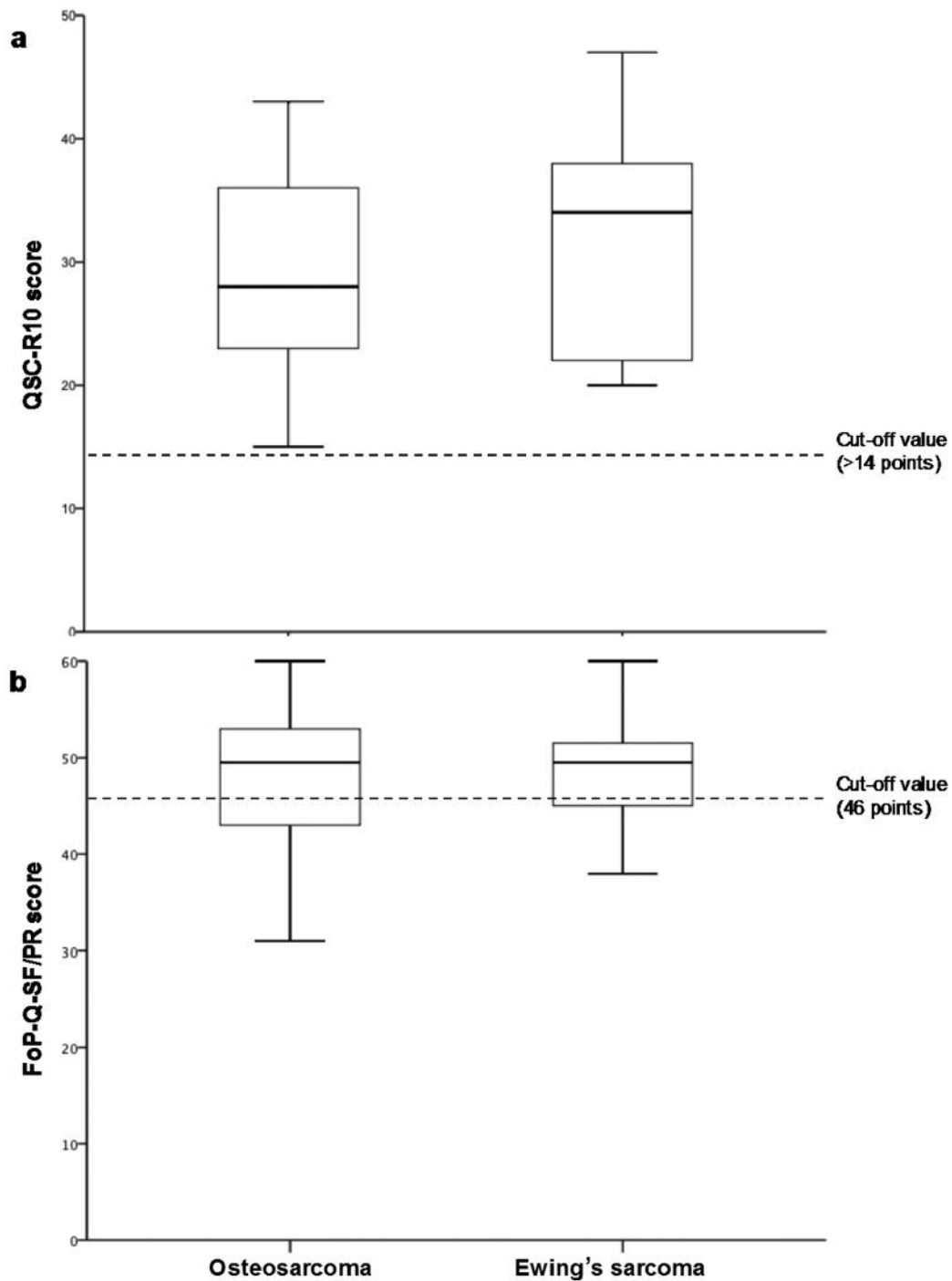


Figure 1. Psychosocial distress levels in patients with osteosarcoma ($n=14$) and Ewing's sarcoma ($n=14$) according to the questionnaire on distress in cancer patients – short form (QSC-R10) (a) and in parents of patients with osteosarcoma ($n=14$) and Ewing's sarcoma ($n=14$) according to the fear of progression questionnaire – short form (FoP-Q-SF/PR) (b).

We observed that (i) a relatively high number of participants were enrolled in the 2-month study period in relation to the low prevalence of OS and ES, and (ii) that all participating

patients and a high percentage of parental caregivers suffered from relevant psychosocial distress and potentially required psycho-oncological treatment.

OS and ES represent the predominant musculoskeletal tumor entities, especially during childhood and adolescence, but account for only 1% of all cancer diagnosed in the United States each year (19). Despite a lack of fixed criteria for the definition of rare cancer, Gatta *et al.* proposed an incidence of fewer than six cases per 100,000 people per year (21). With only five cases per 1,000,000 children aged 19 years and younger per year, OS is an extremely rare disease, and it is difficult and time-consuming to conduct clinical studies with cohorts of sufficient sample size (22). Therefore, in a recent methodology article, Billingham and Steven suggested the creation of national and international networks, as well as maximization of trial duration, to overcome poor patient recruitment in clinical studies of rare cancer (23).

Social networks with specific topic-based groups bring together affected people (patients and relatives) and provide unique potential to collect medical data (crowdsourcing) from large cohorts with rare diseases in a relatively short time period. Substantiating this hypothesis in the field of orthopedic oncology, van der Heijden *et al.* investigated outcomes and quality of life in patients with pigmented villonodular synovitis *via* Facebook, using patient-reported outcome measures (24). That study enrolled 293 patients within a 16-month study period. A large study cohort was generated despite the low percentage of participants (26.3%) compared with 1112 group members. In contrast, recent studies using conventional recruitment strategies showed significantly smaller cohorts (25, 26). Similarly, in the present study, we enrolled 28 patients with OS or ES and 58 parents of patients with OS or ES in only 2 months. In contrast, only four patients underwent treatment at our Institution in the same time period.

To determine the representativeness of the collected data and thus the feasibility of crowdsourcing *via* social media in medical research, demographic and medical results were compared with previous publications on OS and ES. In the present study, the mean age at primary diagnosis of OS was 24.8 ± 13.0 years (range=6-50 years), confirming a bimodal age distribution with a first peak in the second decade of life and a second peak over age 40 years (27, 28). Similarly, our finding that more than 70% of OS occurred in the distal femur and proximal tibia is consistent with previous studies (27-29). Surgical tumor resection (100%) and (neo)-adjuvant chemotherapy (93%) were identified as the main therapeutic strategies, substantiating the modern therapy regimen (30, 31). The mean age of patients with ES was 28.8 ± 9.7 years (range=16-44 years), and was higher than proposed by previous authors for skeletal manifestations (27, 32). Baldini *et al.* found 49% extraosseous tumor manifestations in a study of ES in adults (33). Confirming their results, we also identified 50% extraosseous tumor manifestations, which might be associated with the higher age of our study cohort.

Results for therapeutic modalities were similar to the OS patient group, with a high percentage of (neo)-adjuvant chemotherapy (79%) and surgical tumor resection (79%). In our study population, 50% of patients with ES underwent additional radiotherapy as suggested in modern multimodal treatment concepts (34).

Adjacent to oncological and surgical treatment, supplementary psycho-oncological therapy has often been underestimated in the past, although it is of paramount importance (6). In this context, Book *et al.* found that 34.9% of their cohort (1,850 patients with different cancer types) suffered from relevant psychosocial distress, based on the QSC-R10 (2). Compared with their results, we observed that all patients in the OS and ES groups in our cohort were above the cut-off value of 14 points. To some extent this might be due to the higher mortality of OS or ES compared to more common malignancies such as breast or prostate cancer. In contrast, potentially dissatisfied patients or patients with high psychosocial distress levels may be more likely to enroll in a patient community on Facebook and participate in an online survey regarding outcomes and quality of life (24). We recorded relevant psychosocial distress levels in most parents of patients with OS and ES (93.8% and 97.4%, respectively). In contrast, Schepper *et al.* identified psychosocial distress in only 36.4% of participants in a similar study involving parents of patients with childhood cancer (16). Similar to the patient group, our results might be explained by the higher mortality of sarcomas compared with other malignancies, and a potential selection bias in the topic-based online communities.

In this context, our study has some limitations. Firstly, the participants might have been subject to selection bias as indicated above. Secondly, the patients enrolled in our study were not uniformly treated in sarcoma centers but also underwent therapy in secondary and tertiary referral centers, possibly contributing to unfavorable outcomes and increased psycho-oncological distress levels (35). However, in our opinion, the present study cohort was a randomly selected group reflecting the current situation of sarcoma therapy worldwide, and the findings emphasize the need for international treatment guidelines including psycho-oncological support (6). Thirdly, the accuracy of medical data was subject to the understanding of each patient, possibly influencing study results. However, patient-reported outcome measures, as used in the present study, are easy-to-use tools for crowdsourcing studies *via* social media and for subjective patient outcomes, and are even more reliable than an evaluation by the treating physician (36, 37).

In conclusion, crowdsourcing *via* specific topic-related patient communities on Facebook provides potential for expanding medical research in rare diseases. This novel approach is feasible for the acquisition of valid medical data

and especially self-assessment quality of life measures with larger cohorts than may potentially be enrolled in a single institution, with little effort. Moreover, we identified high distress levels in both the patient and parent groups, indicating a high demand for psycho-oncological treatment. However, evaluation of subjective quality of life measures should be planned carefully due to potential selection bias, with dissatisfied or distressed patients being more likely to enroll in these communities in social media.

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