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ORIGINAL PAPER

Pigmented villonodular synovitis: a crowdsourcing study of two hundred and seventy two patients

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Abstract
Purpose We aimed to ascertain the feasibility of crowdsourcing via Facebook for medical research purposes; by investigating surgical, oncological and functional outcome and quality-of-life (QOL) in patients with pigmented villonodular synovitis (PVNS) enrolled in a Facebook community (1112 members).
Methods Patients completed online open surveys on demographics, surgery and clinical outcomes (group 1); and patient-reported outcome measures (PROMs) including knee-injury osteoarthritis outcome score (KOOS), hip-disability osteoarthritis outcome score (HOOS), Toronto extremity salvage score (TESS) and SF-36 (group 2). Mean follow-up was 70 months (12–374). Consistency checks were performed with Cohen's kappa statistic for intra-rater agreement.
Results The first survey was completed by 272 patients (group 1) and 72 patients completed the second (group 2). In group 1, recurrence-rate was 58 % (69/118) after arthroscopic, 36 % (35/97) after open and 50 % (5/10) after combined synovectomy ($p = 0.003$). In group 2, recurrence-rate was 67 % (26/39) after arthroscopic and 51 % (17/33) after open synovectomy ($p = 0.19$). Recurrence-risk was increased for diffuse disease (OR = 16; 95%CI = 3.2–85; $p < 0.001$). Mean function and QOL did not differ after arthroscopic or open synovectomy: KOOS 49 *vs.* 58 ($p = 0.24$), HOOS 62 *vs.* 53 ($p = 0.56$), TESS 78 *vs.* 82 ($p = 0.86$), SF-36 61 *vs.* 66 ($p = 0.41$). Cohen's kappa statistic for intra-rater agreement was good to outstanding ($\kappa = 0.68$–0.95; $p < 0.001$).
Conclusion Local recurrence-risk was higher for diffuse-type disease and arthroscopic synovectomy. Functional outcome and QOL were comparable for both types of surgery. Gathering data via crowdsourcing seems a promising and innovative way of evaluating rare diseases including PVNS.

Keywords Crowdsourcing · Diffuse-type giant cell tumor · Facebook · Functional outcome · Pigmented villonodular synovitis · PVNS · Quality of life

Introduction

Pigmented villonodular synovitis (PVNS) is a rare benign but locally aggressive giant cell tumour deriving from synovium, either localized giant cell tumour of tendon sheath (GCT-TS) or diffuse-type giant cell tumour (Dt-GCT) [1, 2]. It is most commonly found in the knee (75 %; Fig. 1) and hip (15 %) [1, 3]. Treatment may consist of arthroscopic or open synovectomy, intra-articular radioactive colloids, radiation therapy or systemic targeted therapy [4, 5].

Rare diseases, including PVNS, are difficult to investigate as published case series are generally small, of retrospective nature and often describe research periods of several decades with a short follow-up duration. Therefore, these studies only provide levels of evidence

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Fig. 1 MR images of a 27 year old male patient with recurrent diffuse PVNS in the knee after previous anterior arthroscopic synovectomy and ^{90}Yttrium instillation show multiple intra-articular lesions dorsal to the posterior cruciate ligament, with low signal intensity on T1-weighted sequences (A) and heterogeneous intermediate signal intensity on T2-weighted sequences (B) with artefacts due to haemosiderin depositions being characteristic for PVNS.

III-IV, and clear evidence is lacking as meta-analysis of gathered data is often not warranted.

With a growing number of patients using social media as a source of medical information, online patient platforms can also be used as a powerful educational tool. Gathering patient data via crowdsourcing on social media can be especially promising and useful in the evaluation of rare diseases, as large study populations may become easily available to researchers. Crowdsourcing is the practice of obtaining services, ideas or content by collecting contributions from a large group of people from an online community rather than from traditional data suppliers. This is already increasingly being used for meeting the needs of consumers, and we wanted to investigate if it could be used to evaluate patient satisfaction, functional outcome and quality of life for patients with PVNS. Crowdsourcing to obtain big data can be useful in all fields of medicine, especially regarding the evaluation of rare diseases and patient reported outcome measures (PROMs). Open online surveys may also be more attractive and quick to complete for the modern patient than conventional questionnaires on paper via regular mail.

Several small communities of patients with PVNS exist on Facebook; however, there is only one community with over 1100 patients at the time of study (>2000 at the time of writing). On this forum, patients share information on their disease, experiences with treating physicians, surgery or other treatments and clinical outcome, and seek support from fellow patients. From this forum, we learned that adequate patient information on PVNS is deficient and that there is a desire among patients to interact with other patients and professionals and a growing demand for taking part in scientific research projects on PVNS. This is also reflected by recent initiatives such as PatientsLikeMe (www.patientslikeme.com), in which patients can track their outcomes by sharing their data and by participating in the design and implementation of patient-centred trials [6, 7]. Such online patient communities may allow researchers rapid access to large study populations that are otherwise hard to assemble in rare diseases.

The aim of this study was to investigate whether we could use crowdsourcing via Facebook and online surveys for medical research purposes on PVNS. More specifically, we set out to correlate functional outcome and quality of life (QOL) to patient characteristics, surgical procedures and oncological outcome in patients with PVNS who were enrolled in a patient community on Facebook.

Materials and methods

In this open survey observational study, patients were recruited to complete questionnaires on their disease through a patient-initiated campaign on a PVNS patient community on Facebook (1112 members at time of consulting; https://www.facebook.com/groups/91851410592/?ref=ts&fref=ts). A first online survey on demographics, clinical presentation, findings on imaging and biopsy material, type and localization of disease, surgical and adjuvant treatment, local recurrences, postoperative complications, functional outcome, quality of life (QOL) and follow-up was open from December 2012 to April 2014 (group 1). A second online survey using standardized and validated questionnaires on

patient-reported outcome measures (PROMs) [8], to evaluate functional outcome and QOL, was open from August 2013 to February 2014 (group 2). The latter included range of motion (ROM), knee injury and osteoarthritis outcome score (KOOS) [9, 10], hip disability and osteoarthritis outcome score (HOOS) [11], Toronto extremity salvage score (TESS) [12] and short form-36 health survey (SF-36) [13]. Mean follow-up after index surgery was 70 months (range 12–374). The surveys were developed via SurveyMonkey® (https://www.surveymonkey.com) and usability and technical functionality were tested by the researchers. The surveys were announced on the Facebook community wall with a first notice of the study, and repeated requests for compilation after one, two, five, six and ten months. It was a voluntary survey and no incentives were offered. All responses were anonymous and automatically captured into an SPSS 20.0 file, which was password protected and only accessible to the researchers. Completeness checks were performed through JAVAScript and only fully completed surveys were included in the analysis. Respondents were able to review and change their answers before submitting. Unique site visitors were determined by IP addresses, and in case of duplicate entries only the most recent ones were included in the analysis. Propensity scores to adjust for a non-representative sample were not used in this study, but results were compared with previous results from the literature. The checklist for Reporting Results of Internet E-Surveys (CHERRIES) was used (Appendix 1) [14, 15]. None of the patients were recalled for this study. In the present study, data were not verified by contacting treating physicians or patients. By completing the surveys, patients gave their informed consent for this study. All procedures performed were in accordance with the ethical standards of Dutch law (Medical Research involving Human Subjects Act) and with the 1964 Helsinki declaration and its later amendments. For this type of study formal consent is not required.

Statistical analysis

Differences in dichotomous data were calculated with Chi-squared and Fisher's exact tests and in numerical data with Mann–Whitney U tests. Multivariable logistic regression analyses were performed to identify factors of influence on recurrence, including diffuse or localized disease, localization and type of surgery. To determine whether the results of this study are reliable, we performed consistency tests by evaluating intra-rater agreement for patients' answers between both online surveys. We used Cohen's kappa statistic to perform consistency checks by determining intra-rater agreement for patients that could be cross-linked by IP address in the two online surveys. Outstanding agreement was defined as $\kappa > 0.80$; substantial agreement as $\kappa = 0.60–0.79$; moderate agreement as $\kappa = 0.40–0.59$; and poor agreement as $\kappa < 0.40$ [16]. Data from the two surveys were not combined but analysed separately; hence, the occurrence of repeated measures was not accounted for. We used SPSS 20.0 (SPSS Inc, Armonk, NJ, USA) for statistical analyses. The level of statistical significance was set at $p < 0.05$.

Results

All 1112 members of the PVNS patient community on Facebook, at the time of consulting, could view the online open survey. Participation rate was 26.3 % (293/1112) in the first survey; and 11.8 % (131/1112) for SF-36, 5.3 % (59/1112) for KOOS, 0.8 % (9/1112) for HOOS and 3.1 % (34/1112) for TESS in the second survey. Completion rate was 92.8 % (272/293) in the first survey; and 84.7 % (111/131) for SF-36, 86.4 % (51/59) for KOOS, 100 % (9/9) for KOOS and 100 % (34/34) for TESS in the second survey.

From group 1, 272 patients were included (Table 1). They originated from 23 countries and underwent arthroscopic or open synovectomy between 1982 and 2012. Primary surgery was arthroscopic synovectomy ($n = 118$), open synovectomy ($n = 97$), combined arthroscopic and open synovectomy (hybrid synovectomy; $n = 10$) or unknown ($n = 47$). One hundred twenty two patients reported at least one local recurrence, 124 patients had no recurrences (Table 2). Mean number of surgeries was 2.6 (1–9). Final surgery was open synovectomy ($n = 52$), arthroscopic synovectomy ($n = 36$), hybrid synovectomy ($n = 10$) or unknown ($n = 24$). Total hip arthroplasty was required in 13/25 and total knee arthroplasty in 17/199 ($p < 0.001$). Adjuvant treatment was radiation therapy ($n = 30$), radioactive colloid instillation with ^{90}Yttrium ($n = 18$), M-CSFR targeted tyrosine kinase inhibitor imatinib ($n = 8$) or nilotinib ($n = 2$), cryosurgery ($n = 3$) or methotrexate ($n = 1$).

From group 2, 72 patients with PVNS in the knee or hip joint and a minimum follow-up of one year were included (Table 1). The knee was affected in 64 patients (52 diffuse; 12 localized) and the hip in eight (six diffuse; two localized). Other localizations ($n = 18$; e.g. ankle, elbow and shoulder) were excluded from PROMs analyses, as we wanted to focus on the two most common localizations; however, they were used for consis-

Table 1 Patient, tumour and treatment characteristics

	Group 1 ($n=272$)		Group 2 ($n=72$)	
	mean	range	mean	range
Age (years)	32	11–67	31	15–58
Follow-up (months)	U	U	68	12–374
Number of surgeries	2.6	1–9	2.5	1–9
	n	%	n	%
Gender				
Female	230	85	56	78
Male	42	15	16	22
Localization				
Knee	199	73	64	89
Hip	25	9	8	11
Ankle	31	11.5	–	–
Elbow	8	3	–	–
Foot	4	1.5	–	–
Shoulder	4	1.5	–	–
Hand	1	0.5	–	–
Type of disease				
Diffuse disease	U	U	58	81
Localized disease	U	U	14	19
Pre-operative complaints				
Pain	202	74	25	35
-At rest	U	U	19	26
-During exercise	U	U	19	26
-At night	U	U	16	22
Swelling	202	74	25	35
Stiffness/limited ROM	113	41	25	35
Locking	61	22	U	U
Surgical treatment				
Arthroscopic synovectomy	118	43	38	53
Open synovectomy	97	36	34	47
Combined hybrid synovectomy	10	4	–	–
Unknown	47	17	–	–

U unknown, *ROM* range of motion

Table 3 Factors of influence on recurrence rate after synovectomy for PVNS in the knee or hip

	OR	95 % CI	*p*-value
Diffuse disease	16	3.2–85	0.001
Arthroscopic synovectomy	2.2	0.74–6.6	0.16
Localization in knee	1.3	0.27–5.9	0.77
Arthroscopic synovectomy	1.7	0.67–4.5	0.26

OR odds ratio, *CI* confidence interval

tency checks between both surveys. Primary surgery was arthroscopic synovectomy ($n=38$) or open synovectomy ($n=34$). Recurrence rates are listed in Table 2. Mean number of surgeries was 2.5 (1–9). Recurrences were treated with open synovectomy ($n=16$), arthroscopic synovectomy ($n=15$) or complete resection and joint arthroplasty ($n=12$). Total hip arthroplasty was required in 6/8 and total knee arthroplasty in 7/64 ($p=0.004$). Adjuvant treatment was radiation therapy ($n=12$), ^{90}Yttrium ($n=9$), imatinib ($n=1$) or nilotinib ($n=1$). Diffuse disease increased recurrence risk (odds ratio [OR] = 16; 95 % confidence interval [CI] = 3.2–85; $p=0.001$; Table 3). Mean functional and QOL results did not differ significantly after primary arthroscopic or open synovectomy (Table 4). Final surgery resulted in a mean TESS of 88 after arthroscopic synovectomy, 68 after open synovectomy and 69 after total joint arthroplasty ($p=0.017$). Joint replacement surgery resulted in lower functional scores compared with joint salvage: TESS 69 vs. 82 ($p=0.010$), KOOS 34 vs. 55 ($p=0.031$) and SF-36 49 vs. 66 ($p=0.020$). Mean ROM was lower for patients with diffuse disease (117 vs. 151 degrees; $p=0.024$) and for recurrent disease requiring repeat surgery (113 vs. 138 degrees; $p=0.046$).

Consistency checks were performed with Cohen's kappa statistic for intra-rater agreement; 66 patients could be cross-linked between both surveys through IP address. Cohen's kap-

Table 2 Local recurrences after synovectomy for PVNS in the knee or hip

Group 1 ($n=272$)	Arthroscopic synovectomy		Open synovectomy		Combined synovectomy*		*p*-value
	%	n	%	n	%	n	
Total group	58 %	69/118	36 %	35/97	50 %	5/10	0.003
Group 2 ($n=72$)							
Total group	67 %	26/39	51 %	17/33	–	–	0.19
Localized disease	25 %	2/8	0 %	0/6	–	–	0.31
Diffuse disease	77 %	23/30	64 %	18/28	–	–	0.23

*Combined arthroscopic and open synovectomy

Table 4 Functional outcome and QOL after primary synovectomy for PVNS in the knee or hip

	Arthroscopic synovectomy		Open synovectomy		*p*-value
	mean	range	mean	range	
ROM (degrees)	124	80–170	129	65–170	0.65
KOOS	49	8–92	58	34–92	0.24
HOOS	62	51–72	53	31–67	0.56
TESS	78	33–100	82	63–97	0.86
SF-36	61	11–100	66	21–98	0.41

ROM range of motion, *TESS* Toronto extremity salvage score, *KOOS* knee injury and osteoarthritis outcome score, *HOOS* hip disability and osteoarthritis outcome score, *SF*-36 short form 36 health survey

pa statistic for the two online surveys was outstanding for tumour localization with $\kappa = 0.95$ ($p < 0.001$), joint arthroplasty with $\kappa = 0.88$ ($p < 0.001$) and adjuvant treatment with $\kappa = 0.82$ ($p < 0.001$); and substantial for primary surgical treatment with $\kappa = 0.78$ ($p < 0.001$) and recurrences with $\kappa = 0.68$ ($p < 0.001$).

Discussion

In this study, we investigated whether we could use crowdsourcing via Facebook and online surveys for medical research purposes on PVNS. We set out to correlate functional outcome and QOL to patient characteristics, surgical procedures and oncological outcome in patients with PVNS who were enrolled in a patient community on Facebook. We concluded that the recurrence risk was highest for diffuse disease and after arthroscopic synovectomy. Lower functional results were reported for patients with diffuse disease, for patients with recurrences requiring repeat surgery, and for patients eventually requiring joint replacement surgery.

To determine whether the results of this study are reliable and whether crowdsourcing via social media is feasible in medical research, we performed consistency tests by evaluating intra-rater agreement for patients' answers between both online surveys, which was substantial to outstanding for most study variables. The somewhat lower agreement in follow-up questions can be explained by the time interval between the two surveys as patients may have experienced a recurrence after filling out the first survey. In addition, we compared our results with previous publications on PVNS to assess the representativeness of our sampling frame [17] (Tables 3 and 5). An advantage of this comparison may be the determination of accurateness of the use of crowdsourcing and the reliability of the obtained data. Possible disadvantages of crowdsourcing may include selection bias through the sampling method, inhibiting comparison of results with previously published reports. The average age of 32 years at onset of disease in this study matches known epidemiology of PVNS (<40 years) [1, 2, 36]. Localization of PVNS in this study (73 % knee, 9 % hip, 18 % other) was also comparable to percentages described in literature (75 % knee, 15 % hip, 10 % other). Multifocal PVNS is rare and was not reported in this study [36]. Women were over-represented (78 %) when compared with known epidemiology of PVNS; possibly indicating that women are more likely to be on Facebook and to seek information via support groups [37]. All study variables were distributed equally between men and women. Women scored somewhat lower on SF-36 subdomains physical functioning, pain and PCS; all other functional and QOL scores did not differ between men and women (results not shown). Proportions of arthroscopic and open synovectomy [4] and total knee or hip arthroplasty were comparable to those previously published [38, 39].

Mean follow-up of approximately six years (range 1–31) was longer when compared with the majority of previously published reports [3, 19, 23, 29, 31, 32]. Together with the relatively high recurrence rates in this study (58–77 % after arthroscopic and 36–64 % after open synovectomy), this may indicate that to date, an underestimate of the true local recurrence rate of PVNS has been published.

Functional results after surgical treatment for PVNS have been reported (Table 5) [3, 18–32], but comparison is difficult, as functional results were not specified for diffuse and localized disease, type of surgery, primary or recurrent disease and different localizations. In this study, there was no difference in functional outcome and QOL between patients who underwent primary arthroscopic or open synovectomy. However, the long course of disease and the need for multiple surgeries has previously been reported to result in worse functional results in a large number of patients [23]; and in this study, lower functional results were also reported for patients with diffuse disease, with recurrences requiring repeat surgery,

Table 5 Literature overview on oncological and functional results after arthroscopic and open synovectomy for PVNS

Study	n	Sex	Age (years) mean (range)	Follow-up (months) mean (range)	Localization	PVNS type	Surgery	Recurrence rate %	Functional outcome	
										mean (range)
De Visser et al. [18]	38	F18, M20	32 (12–72)	48 (12–228)	knee, hip, ankle	29 diffuse 9 localized	unspecified	unspecified	MSTS	24 (15–30)
Zvijac et al. [19]	14	F7, M7	35 (19–64)	42 (8–83)	knee	12 diffuse 2 localized	arthroscopic synovectomy	diffuse 14 % localized 0 %	other	10 excellent/good, 2 fair, 2 poor[1]
Shabat et al. [20]	10	F2, M8	NR (15–49)	72 (30–144)	knee, ankle, hip	diffuse	unspecified	10 %	MSTS	9 excellent, 1 unknown[1]
Chin et al. [21]	40	F17, M23	35 (14–68)	60 (18–96)	knee	diffuse	open synovectomy	17 %	KSS other[2]	92 (55–100) 92 (0–100)
De Ponti et al. [22]	19	F10, M9	59 (37–83)	60 (12–128)	knee	15 diffuse 4 localized	arthroscopic synovectomy	diffuse 50 % localized 0 %	other[3]	Complete arthroscopy: excellent[1] Partial arthroscopy: good[1]
Chiari et al. [23]	42	F27, M15	40 (6–76)	80 (26–194)	knee, ankle, hip, foot, shoulder, hand	19 diffuse 23 localized	open synovectomy	overall 24 %	MSTS	28 (18–30)
Wu et al. [24]	9	F4, M5	31 (19–51)	67 (37–103)	knee	diffuse	open synovectomy	11 %	KSS-knee KSS-function	94 (86–98) 97 (80–100)
Dines et al. [25]	26	F11, M15	36 (12–68)	66 (46–123)	knee	localized	14 open synovectomy 12 arthroscopic synovectomy	0 %	LKS[4]	95 (71–100)
Ozturk et al. [26]	7	F4, M3	45 (20–68)	48 (24–97)	knee	diffuse	4 arthroscopic synovectomy 3 open synovectomy	0 %	MSTS	21 (12–26)
Nassar et al. [27]	12	F4, M8	NR (19–49)	27 (20–36)	knee	diffuse	open synovectomy	0 %	MSTS	25.5 (24–27)
Liu et al. [28]	22	F14, M8	24 (16–35)	22 (18–28)	knee	localized	arthroscopic synovectomy	14 %	LKS IKDC	95 (SD 3.5) 93 (SD 2.4)
Akinci et al. [29]	19	F10, M9	43 (NR)	80 (15–156)	knee	15 diffuse 4 localized	open synovectomy	overall 26 %	KSS	8 perfect, 9 good, 2 bad[1]
Griffin et al. [30]	50	F30, M20	38 (18–74)	94 (19–330)	knee, ankle, hip, foot, hand, wrist	diffuse	unspecified	6 %	MSTS-87[5] MSTS-93[5] TESS[5] other[5]	31 (25–35) 28 (19–30) 90 (65–99) 7 excellent, 34 good, 5 fair, 4 poor[1]
Nakahara et al. [31]	17	F7, M10	33 (SD 17.2)	65 (10–146)	knee	diffuse	open synovectomy	12 %	KSS	97 (76–100)
Chen et al. [3]	19	F10, M9	43 (29–59)	98 (42–143)	knee	diffuse	open synovectomy	11 %	TLKS	93 (86–100)
Loriaut et al. [32]	30	NR	46 (23–71)	75 (12–144)	knee	localized	arthroscopic synovectomy	20 %	LKS	86 (83–88)
Van der Heijden et al. [33]	30	F17, M13	34 (6–73)	95 (24–403)	knee	diffuse	14 open synovectomy 16 arthroscopic synovectomy	open 28 % arthroscopy 94 %	KOOS MSTS TESS SF-36	59 (12–99) 21 (8–30) 80 (45–100) 70 (26–98)
Jain et al. [34]	40	F9, M31	44 (21–76)	84 (24–120)	knee	29 diffuse 11 localized	arthroscopic synovectomy	41 % 0 %	LKS	31 excellent, 8 good
Ma et al. [35]	75	F48, M27	46 (15–80)	41	knee, hip, ankle	67 diffuse 8 localized	open or arthroscopic synovectomy with or without artroplasty	16 %	NR	
Current study	272	F230, M42	32 (SD 12)	NR	knee, hip, ankle, elbow, foot, shoulder, hand	diffuse and localized	97 open synovectomy 118 arthroscopic synovectomy	open 36 % arthroscopy 58 % hybrid 50 %	NR	

Table 5 (continued)

Study	n	Sex	Age (years) mean (range)	Follow-up (months) mean (range)	Localization	PVNS type	Surgery	Recurrence rate %	Functional outcome	mean (range)
							10 combined open and arthroscopic synovectomy 47 unknown			
	72	F56, M16	31 (SD 12)	70 (12–374)	knee, hip	58 diffuse 14 localized	34 open synovectomy 38 arthroscopic synovectomy	open 51 % arthroscopy 67 %	KOOS HOOS TESS SF-36	58 (34–92)/49 (8–92)[6] 53 (31–67)/62 (51–72)[6] 82 (63–97)/78 (33–100)[6] 66 (21–98)/61 (11–100)[6]

F female, *M* male, *NR* not reported, *KSS* Knee Society score, *TLKS* Tegner-Lysholm knee score, *LKS* Lysholm knee scale, *IKDC* International Knee Documentation Committee, *SD* standard deviation

[1] Functional outcome was not further specified

[2] Functional outcome was based on pain, walking status, joint swelling, effusion, crepitus, locking, instability and ROM

[3] Functional outcome was based on pain, synovitis, joint swelling and ROM

[4] Functional outcome was obtained from 10/26 patients

[5] Functional outcome was obtained from 14/50 patients

[6] Functional results after open synovectomy/arthroscopic synovectomy

and for patients eventually requiring joint replacement surgery.

Our study has several limitations. First, the low participation rate in this study (26.3 %) may be explained by several arguments, including the voluntary character of the survey, and the attention paid to the survey by the Facebook members; maybe they didn't see the requests on their wall, didn't have time to compile the questionnaires or didn't feel the need to add to a scientific research project. This is also of concern when similar studies involving social media are performed in medicine, as it may induce selection bias. In regards to this potential selection bias, patients who are currently unsatisfied with their situation may be more likely to enrol in a patient community on Facebook and to complete online surveys concerning functional and QOL results. However, the opposite may also be true for satisfied patients who are willing to share their experience and to improve awareness on this rare disease. Second, patients were not uniformly diagnosed and treated for PVNS, as they were collected from 23 different countries and treated by various surgeons without distinguishing between peripheral and tertiary referral centres. However, we believe that it represents a randomly chosen group of patients, perfectly reflecting the current worldwide situation and underlining the importance of centralization of care for musculoskeletal tumours [40]. Third, the accuracy of surgical data is subjective to the understanding of patients; data obtained from patients were not verified by contacting treating physicians. We considered this beyond the scope of the current study but we deem it valuable in future crowdsourcing studies via social media. Yet, PROMs are considered more reliable in reporting subjective patient outcomes when compared to the evaluation by treating physician, and are more often required by the FDA [8, 41].

In addition to using social media for crowdsourcing purposes in rare diseases, it may also increase the readership and impact of scientific publications apart from the indexed impact factor.

An increasing number of scientific papers can be found through social media such as Facebook, Twitter, LinkedIn and ResearchGate; and journals should explore these platforms and use it in a constructive way to increase the scientific exposure [42].

In conclusion, gathering data via crowdsourcing in a patient community on Facebook seems a promising and innovative way of evaluating rare diseases, as it provides for a representative and large sample of patients with long-term follow-up and valid clinical outcome data. The results of this study suggest that local recurrence risk and functional outcome were both negatively influenced by diffuse disease, which comprises a large part of the joint, is difficult to resect completely and often requires repeat surgery, especially after arthroscopic synovectomy.

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Compliance with ethical standards

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

Appendix: Data reporting guidelines

Appendix 1: CHERRIES checklist

Table 6 Checklist for reporting results of internet E-surveys (CHERRIES)

Item category	Checklist item	This study
Design	Describe survey design	Target population is 1112 patients enrolled in "PVNS is pants!!" community on Facebook
IRB approval and informed consent process	IRB approval	Approval of the ethics committee of our institution was waived because this study did not fall under the scope of the Dutch law on Medical Research Involving Human Subjects Act (WMO).
	Informed consent	Informed consent was included in the questionnaires. Length of survey was explained on the first page of survey, investigators PhD student and consultant orthopedic oncologist from a tertiary centre in the Netherlands, purpose of study to increase knowledge and awareness on this rare disease including PROMs.
	Data protection	Results from survey visible to researchers only, password protected.
Development and pre-testing	Development and testing	Survey developed via surveymonkey.com, usability and technical functionality was tested by researchers.
Recruitment process and description of the sample having access to the questionnaire	Open survey versus closed survey	Open survey on community on Facebook
	Contact mode	Contact with participants was made through the Facebook community, links to the surveys were provided here.
	Advertising the survey	The e-survey was announced on the Facebook community wall with a first notice of the study, and repeated requests after 1, 2, 5, 6 and 10 months. See Appendix I (below).
Survey administration	Web/E-mail	E-survey posted on a website (i.e. Facebook community on PVNS). Responses were automatically captured into an SPSS 20.0 file, which was only accessible to the researchers.
	Context	Facebook community on PVNS, mostly patients but also family members and the research from this study. Normally looking to share information and personal experiences concerning this rare disease, its (surgical) treatment, recovery duration, tips and tricks; so best summarized as an information and support group. *Women are somewhat overrepresented when compared with known epidemiology of PVNS, and may be more likely to seek information via support groups?* *Patients from countries all over the world, including developing and developed countries, age groups of Facebook users matches known epidemiology.*
	Mandatory/voluntary	Voluntary survey
	Incentives	No incentives were offered
	Time/date	December 2012–April 2014 May 2013–May 2014
	Randomization of items or questionnaires	No randomization, standardizes PROMs
	Adaptive questioning	No adaptive questioning, standardized PROMs
	Number of items	General survey = 20 SF36 = 36 KOOS = 42 HOOS = 40 TESS = 30
	Number of screens (pages)	1 webpage per survey, if applicable
	Completeness check	Yes, completeness checks were done through JAVAScript, consistency checks were not performed, not applicable options were included, general questions included some open and some multiple option questions, PROMs were standardized validated questionnaires with enforced one-response-option.

Table 6 (continued)

Item category	Checklist item	This study
	Review step	Respondents were able to review and change their answers
Response rates	Unique site visitor	Unique site visitors were determined by IP addresses.
	View rate (ratio of unique survey visitors/unique site visitors)	View rate Total 274/1112 = 24,6 % SF36 131/1112 = 11.8 % KOOS 59/1112 = 5.3 % HOOS 9/1112 = 0.8 % TESS 34/1112 = 3.1 %
	Participation rate (ratio of unique visitors who agreed to participate/unique first survey page visitors)	Participation rate Total 270/274 = 98.5 % SF36 131/131 = 100 % KOOS 59/59 = 100 % HOOS 9/9 = 100 % TESS 34/34 = 100 %
	Completion rate (ratio of users who finished the survey/users who agreed to participate)	Completion/completeness rate SF36 111/131 = 84.7 % KOOS 51/59 = 86.4 % HOOS 9/9 = 100 % TESS 34/34 = 100 %
Preventing multiple entries from the same individual	Cookies used	No cookies were used
	IP check	Duplicate database entries having the same user ID/IP address were eliminated before analysis; the most recent entries were included.
	Log file analysis	See above
	Registration	N. A.
Analysis	Handling of incomplete questionnaires	Only completed questionnaires were analyzed.
	Questionnaires submitted with an atypical timestamp	N.A.
	Statistical correction	Propensity scores to adjust for the non-representative sample were not used in this study

Appendix I: Survey announcements 12-05-2013

Announcement of study initiative: Knowledge on QOL and functional results after treatment of PVNS is lacking; validated questionnaires will be launched here soon and your valuable data will be used to create awareness and improve treatment protocols as were recently published [REF JSO, JBJS Br].

01-08-2013 Announcement including link to survey: Please fill in this first Facebook-based research initiative on functional outcome and quality of life of patients with PVNS and allow us to evaluate your quality of life and limb or joint function in order to improve the current treatment protocols and therewith the functional outcome and quality of life of patients with PVNS in the future. All data will be handled with care and are only used according to the rules of good clinical and research practice. Data stored will not be traceable to your personal details, all will be anonymized.

Further announcements including link to survey were provided after 1, 2, 5, 6 and 10 months. (19-08-2013, 31-08-2013, 03-09-2013, 17-09-2013, 22-09-2013, 22-12-2013, 31-12-2013, 29-01-2014, 23-05-2014).

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