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ORIGINAL ARTICLE

How social media can help to understand treatment experiences of survivors of rare cancers: Findings from the Granulosa Cell Tumor (GCT) Survivor Sisters Facebook group member survey

Victoria M. White PhD[1] | Maria Alexiadis BSc(Hons)[2,3] | Kimberly D. Eroh Graduate Certificate Teaching[4] | M. Kaye Ackermann BA[4] | Sue Rodgers BS[4] | Linda M. Langdale FCCA (Retired)[4] | Natasha E. Armour[5] | Thomas W. Jobling MBBS, FRCOG, MD[6] | Peter J. Fuller MBBS PhD[2,3] | Simon Chu PhD[2,3]

[1]Deakin University, Burwood, Victoria, Australia

[2]Centre for Endocrinology and Metabolism, Hudson Institute of Medical Research, Clayton, Victoria, Australia

[3]Department of Molecular and Translational Sciences, Monash University, Clayton, Victoria, Australia

[4]GCT-Survivor Sisters, Facebook, Statesville, North Carolina, USA

[5]Rare Ovarian Cancer Inc, Shellharbour, New South Wales, Australia

[6]Monash Health, Clayton, Victoria, Australia

Correspondence

Victoria M. White, Deakin University, 221 Burwood Hwy, Burwood, VIC 3125, Australia.
Email: Vicki.white@deakin.edu.au

Peter J. Fuller, Hudson Institute of Medical Research, Monash Medical Centre, Clayton, VIC 3168, Australia.
Email: Peter.fuller@hudson.org.au

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Abstract

Background: Engaging with online social media consumer groups for rare cancers may help to develop collaborations between consumers and researchers. This study, a collaboration with the Granulosa Cell Tumor-Survivor Sisters (GCT-SS) Facebook group, explores the results of their survey of member's treatment and follow-up experiences.

Methods: Members of the closed multinational GCT-SS Facebook group completed a 43-item survey covering symptoms, diagnosis, treatment, recurrence, follow-up, and possible risk factors for GCT. Group members could have adult (*a*GCT) or juvenile (*j*GCT) disease. Data was collected via an online survey between 2014 and 2019.

Results: A total of 743 members (average 4.4 years [SD = 5.9] post-diagnosis) participated including 52 with *j*GCT. A total of 67% had stage I disease and 8% had stage III–IV at diagnosis, although 30% of *a*GCT and 25% of *j*GCT reported recurrent disease at survey completion. A total of 48% of *a*GCT had laparoscopic surgery, tumor encapsulation was reported by 49%, and tumor bagging reported by 29% overall (37% laparoscopic; 8% open). Recurrence rates were higher when the tumor was cut or ruptured (ruptured: $p < .001$; cut: $p = .01$). A total of 19% of *a*GCT had chemotherapy with this most common for stage II-III disease. Bleomycin, etoposide, and cisplatin protocols became less common over time (diagnosed before 2015: 47% vs. diagnosed post-2015: 21%).

See editorial on pages 000–000, this issue.

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Conclusions: This is one of the largest surveys of GCT treatment. Members of the GCT-SS group report treatment patterns generally in line with those found from clinical audits. Using naturally forming consumer groups may assist with developing the evidence base for care and supporting those living with GCT ovarian cancer.

Plain language summary

- This study is a collaboration between members of Granulosa Cell Tumor-Survivor Sisters (GCT-SS) Facebook group and researchers to assess members' experiences of treatment and follow-up.
- A total of 743 members (52 with juvenile GCT) completed an online survey.
- A total of 67% had stage I disease at diagnosis.
- Treatment patterns were generally in line with those found from clinical audits: 95% had surgery and 19% of those with adult GCT had chemotherapy.
- A total of 30% reported recurrent disease, with recurrence occurring within 5 years of diagnosis for 33%.
- Using naturally forming consumer groups may assist with developing the evidence base for care and supporting those living with GCT ovarian cancer.

KEYWORDS
consumer experiences, GCT ovarian cancer, Patient Public Involvement, survey, treatment

INTRODUCTION

Granulosa cell tumors (GCT) arising from the sex-cord stromal cells of the ovary are rare contributing approximately 2%-7% of malignant ovarian cancers.[1–3] The majority are of the adult subtype (*a*GCT) characterized by a specific pathognomonic mutation in the FOXL2 gene (*FOXL2*^{C134W}) and ~5% are a juvenile subtype (*j*GCT) which, as the name implies has an earlier age of onset and does not contain *FOXL2*.^{C134W} Although GCT are generally diagnosed at an early-stage and have a favorable 5-year prognosis, relapse can occur many years after initial diagnosis.[2,4] Although treatment recommendations for GCT exist[1,5] a lack of evidence from randomized trials regarding management means optimal treatment is not clear. The lack of certainty is most obviously seen in relation to chemotherapy, with limited evidence regarding its benefit for both early and advanced-stage disease.[2,6,7]

There has been growing acknowledgment of the benefits of involving patients and the public in health care services and research, with this move increasingly encouraged by consumer organizations and research bodies in the United States (US), the United Kingdom (UK), Europe, and Australia.[8,9] Increasingly known as patient and public involvement (PPI), this movement promotes collaboration between scientists and members of the public affected by the research questions or outcomes.[8,10] Multiple frameworks for PPI have been proposed[11] with all agreeing that engagement needs to be genuine and involve more than including people as research participants.[11–13] Although PPI is advocated at all stages of research, a systematic review of 27 studies reporting consumer involvement in cancer-related research found it mostly occurred at two stages: (1) identifying the research focus, and (2) recruitment.[11] No study at that time included PPI in data collection.[11] Beginning this work, a recent study used a consumer-led process to develop and implement a questionnaire examining the impact of colorectal cancer on sexual functioning after surgery.[14]

Online social network platforms such as Facebook and Twitter have allowed those with rare conditions including cancers to connect with similar others regardless of where they live. Although the utility of social network platforms for study recruitment is increasingly acknowledged,[15–17] there is also growing recognition that these groups of engaged consumers offer the opportunity for bidirectional engagement with researchers to facilitate collaboration on projects relevant to consumers.[18–20] The GCT Survivor Sisters (GCT-SS) Facebook group, currently has over 1600 members from across the globe connecting to provide support and information to women and girls affected by *a*GCT or *j*GCT. GCT-SS form active collaborations with researchers to improve outcomes for GCT survivors. GCT-SS also established a need to gather data on member's experiences of treatment for their cancer and those joining the GCT-SS Facebook group, complete a survey describing their diagnostic, treatment, and follow-up experiences. The administers of GCT-SS and the Australian Rare Ovarian Cancer Incorporated (ROC Inc) charity are collaborating with researchers from Melbourne, Australia to further understand the biology of GCT, and the treatment and care experiences of those with this cancer. As part of this collaboration, researchers and consumers have worked to undertake an analysis of the data collected through the GCT Survivor Sisters survey. In this article, we

report findings from the analyses of this data set describing the diagnosis, treatment, and follow-up care experiences of those with GCT and explore feasibility of using PPI in data collection.

MATERIALS AND METHODS

Our primary objective is to report the issues covered in a consumer-driven survey assessing care experiences for GCT. Data was collected through a survey administered to members of the private GCT-SS Facebook group established and managed by K.E., K.A., S.R. and L. M.L. Members are invited to complete the survey when they first join. K.E., K.A., S.R., L.M.L., N.E.A., V.W., and M.A. met regularly via video-link to discuss the data, its analyses, and findings.

Questionnaire

The questionnaire was developed by the GCT-SS team and administered online. The questionnaire consisted of 43 items covering symptoms, diagnosis, treatment, recurrence, follow-up, and family history of cancer as well as questions assessing menstrual history, fertility, and experience of menopause (see Table S1 for list of questions). Most questions were open ended with participants writing a short response. The question assessing stage of disease used the International Federation of Gynecology and Obstetrics (FIGO) staging system and asked respondents to select from one of the available FIGO stages or "don't know." Questions reflected the issues of interest to the GCT-SS membership and included: understanding diagnostic process, treatment, recurrence experiences, and prevalence of putative risk factors.

Ethics

As the study used data collected through an existing consumer-based survey, the study was exempt from ethical review. However, use of the data set was registered with Deakin University's Human Research Ethics Committee (2021-068). Informed consent was obtained from all people completing surveys.

Data analyses

Open-ended responses were reviewed by the study team and coded into agreed categorical responses or binary-type variables. Tumor size was collapsed into two groups: <8.5 cm and ≥8.5 cm. Some reported tumor size with reference to an object with those described as a prune, walnut, golf ball, and orange categorized as <8.5 cm whereas those described as a grapefruit, cantaloupe, softball/baseball, football (American), basketball or watermelon coded ≥8.5 cm.

Because many questions used an open-ended response, no response may indicate an absence of this issue rather than missing per se. Missing data was managed at a variable level and included as a response option in analyses when substantial. Statistical analysis was conducted using SPSS V28. Descriptive statistics (e.g., frequencies) were used to describe the sample in terms of individual, disease and treatment characteristics. Pearson χ^2 or Fisher exact test were used to compare proportions between groups (e.g., recurrence or not). Logistic regression was used to examine factors associated with recurrent disease.

RESULTS

Sample

Data was collected between 2014 and 2019. A total of 744 GCT-SS members (of approximately 1200–1300) completed the survey including 52 with *j*GCT. Missing data varied per question ranging from 3% to 4% for items assessing year diagnosed, and surgical procedure, 14%–15% for items assessing symptoms, and tumor size to 67% for an item assessing whether the tumor was bagged. One respondent with substantial missing data (>85% of questions) was removed. Although 98% of respondents reported on their own experiences of GCT, 11 surveys were completed for a person with GCT 16 years old or younger with the survey completed generally by a family member (i.e., mother). Respondents were on average 4.4 years (SD, 5.9; median, 2; range, 0–38 years) post-diagnosis when completing the survey, with no significant difference between *a*GCT and *j*GCT. The majority of respondents with *a*GCT were diagnosed between 30 to 49 years (60.4%). For those with *j*GCT, one-third (33%) were diagnosed before the age of 20, with 21% diagnosed between the ages of 20 and 30 years (Table 1). Most participants were diagnosed in the 2010s (73%) with 43% diagnosed between 2015–2019. Reflecting participants' age, most were premenopausal at diagnosis (88%). For the 190 respondents (including four with *j*GCT) who had gone through menopause, menopause was rated on average as slightly easy (mean, 2.25). Sixteen percent had been diagnosed with polycystic ovarian syndrome (PCOS), and 12% had a mother or sister with a history of breast or gynecological cancer (Table 1).

Symptoms and disease characteristics

The most common symptoms were abdominal pains (45%), bloating (27%), excessive bleeding (24%), or amenorrhea (24%). Symptoms were similar for *a*GCT and *j*GCT. Just over half (54%) reported symptom onset between the ages of 30–49 years and most (85%) were diagnosed in the year their symptoms presented.

Sixty-seven percent reported they were diagnosed with stage I disease including 35% having stage IA and 29% having stage IC. However, 17% did not report their stage of disease. Tumor size was reported by 74%, with most (40%) having tumors ≥8.5 cm. Tumors were larger for *j*GCT than *a*GCT ($p = .015$).

TABLE 1 Personal characteristics, diagnosis stage, size, and diagnostic time period of participants by adult-type or juvenile-type disease.

	Adult (*N* = 691), %	Juvenile (*N* = 52), %	Total (*N* = 743), %
Age at diagnosis, years			
0–12	0.1	15.4	1.2
13–19	0.4	17.3	1.6
20–29	7.8	21.2	8.7
30–39	27.5	23.1	27.2
40–49	32.9	9.6	31.2
50–59	21.7	1.9	20.3
60–69	6.8	0.0	6.3
Missing	2.7	11.5	3.4
Premenopausal at diagnosis	86.8	100.0	87.8
Regular periods	42.1	32.7	41.5
Periods: light (1) or heavy (5) mean (SD)	3.53 (1.13)	3.14 (1.22)	3.50 (1.14)
Diagnosed with PCOS	15.6	15.4	15.6
Diagnosed with HPV	11.9	9.6	11.7
Mother or sister had breast or a gynecological cancer	12.4	7.7	12.1
Symptoms (*n* = 639)	(*N* = 594)	(*N* = 45)	(*N* = 639)
Amenorrhea[a]	25.4	11.1	24.2
Excessive bleeding	24.9	15.6	24.3
Fatigue	4.5	6.7	4.7
Abdominal Pains	44.8	42.2	44.6
Bloating	26.9	22.2	26.6
Increased sex drive	0.7	4.4	0.9
Age when symptoms started, years			
<20	1.3	26.7	3.1
20–29	9.4	20.0	10.2
30–39	24.2	24.4	24.3
40–49	31.3	13.3	30.0
50–59	17.8	2.2	16.7
60+	5.1	0.0	4.7
Not sure	10.8	13.3	11.0
Period of diagnosis			
Diagnosed 2018 or 2019	12.7	13.5	12.8
Diagnosed 2015 thru 2017	30.4	28.8	30.3
Diagnosed 2010–2014	29.7	32.7	29.9
Diagnosed 2005–2009	15.9	9.6	15.4
Diagnosed 2000–2004	7.3	5.8	7.2
Diagnosed 1990 thru 1999	2.9	7.7	3.2
Diagnosed 1981 thru 1989	1.2	1.9	1.3
Stage at diagnosis			
IA	35.3	34.6	35.3
IB	3.6	0.0	3.4

TABLE 1 (Continued)

	Adult (*N* = 691), %	Juvenile (*N* = 52), %	Total (*N* = 743), %
IC	28.2	32.7	28.5
IIA	2.7	1.9	2.7
IIB	3.3	0.0	3.1
IIC	1.3	5.8	1.6
III	6.9	9.6	7.1
IV	1.3	0.0	1.2
Missing/don't know	17.2	15.3	17.1
Tumor size			
<8.5 cm	35.6	19.2	34.5
≥8.5 cm	38.5	57.7	39.8
Not sure	25.9	23.1	25.7

Abbreviations: HPV, human papilloma virus; PCOS, polycystic ovarian syndrome.

[a]Based on those diagnosed while premenopausal: adult, $n = 512$; juvenile, $n = 45$.

Initial Treatment experiences

Most respondents had surgery (Table 2), with approximately the same proportions having laparoscopic (48%) and open surgery (47%) for those with *a*GCT. Of those with *j*GCT, 56% had open surgery. Those diagnosed after 2010 were more likely to report laparoscopic surgery (56% vs. 25%) ($p < .01$). *j*GCT respondents were less likely to report hysterectomy (13% vs. 42%, $p < .01$) or bilateral oophorectomy (8% vs. 43%, $p < .01$) than *a*GCT respondents. For *a*GCT, bilateral oophorectomy and hysterectomy was inversely related to age, with 64% and 77% of those over 50 years reporting hysterectomy and bilateral oophorectomy, respectively. No one under 30 years reported these procedures ($p < .001$).

Of those having surgery, 49% reported their tumor was encapsulated, 13% indicated the tumor was cut, and 29% reported tumor rupture. Twenty-two percent reported tumor bagging with this higher for laparoscopic (37%) than open (8%) surgery ($p = .03$).

Of all *a*GCT, 19% indicated they had chemotherapy. Chemotherapy was most common for those with stage II or III disease (Table 3). Two main protocols were reported: bleomycin, etoposide, and cisplatin (BEP) (25%) and carboplatin and paclitaxel (39%). The protocol used related to diagnosis period ($p < .01$), with 21% diagnosed from 2015 receiving BEP compared to 47% of those diagnosed before 2015. Of *j*GCT respondents, 13 reported chemotherapy.

Experiences of recurrent disease

Overall, 30% of *a*GCT participants reported recurrent disease with proportions for stage I-III disease not significantly different. Of *j*GCT, 25% ($n = 13$) reported recurrent disease. The most common symptoms leading to diagnosis of recurrence were abdominal issues relating to bloating and pain (31%). Although for 10% recurrence was detected through elevated serum/plasma markers, 4% noted it was detected through routine surveillance scans, and 20% indicated no discernible symptoms at time of recurrence. Timing of recurrence differed between *a*GCT and *j*GCT: 50% of recurrent *j*GCT occurred within 12 months of diagnosis compared to 11% of recurrent *a*GCT ($p < .01$). Of those with recurrent *a*GCT disease, 26% had their recurrence diagnosed 5–9 years post-diagnosis, with 22% diagnosed 3–5 years post-diagnosis. The most common treatment for recurrent disease was surgery with chemotherapy (41%) or surgery alone (35%). Chemotherapy was most commonly carboplatin and paclitaxel (53%), with 38% reporting a BEP protocol.

Those with recurrent disease were more likely to be premenopausal at diagnosis, have later stage disease, have a tumor that ruptured during surgery, and be more years post-diagnosis (Table 4). Not having the tumor bagged was associated with recurrence in univariate analyses. Focusing on those reporting tumor rupture (cut or spontaneous), there was no association between bagging and recurrence. However, recurrence rates when the tumor was cut or ruptured were significantly higher than when the tumor was encapsulated (ruptured: $p < .001$; cut: $p = .01$). Associations between recurrence and stage and tumor rupture were maintained after adjusting for time since diagnosis.

Follow-up care for respondents without recurrence

Of the 522 respondents without recurrent disease, 431 (83%) provided follow-up information: 60% reported monitoring of inhibin A&B, 26% reported monitoring of inhibin B, and 9% reported monitoring of inhibin A. Other common procedures included regular scans (51%; computed tomography scans or ultrasounds), and monitoring CA-125 levels (33%).

TABLE 2 Initial treatment and recurrence history by self-reported disease stage at diagnosis for adult-type disease.[a]

	Stage at diagnosis										
	IA	IB	IC	IIA	IIB	IIC	III	IV	Don't know	Missing	Total
(*N*)	(244)	(25)	(195)	(19)	(23)	(9)	(48)	(9)	(89)	(30)	(691)
	%	%	%	%	%	%	%	%	%	%	%
Surgery	98.8	100.0	95.4	100.0	95.7	100.0	100.0	88.9	91.0	66.7	95.4
Tumor encapsulated?											
Encapsulated	81.1	64.0	22.1	36.8	47.8	0.0	27.1	22.2	38.2	40.0	48.6
Cut during surgery	5.7	16.0	22.1	10.5	13.0	22.2	14.6	11.1	10.1	10.0	12.7
Ruptured	6.6	16.0	50.3	36.8	34.8	77.8	41.7	44.4	31.5	26.7	28.9
Tumor bagged	27.9	20.0	19.5	10.5	21.7	11.1	16.7	0.0	23.6	13.3	22.0
Any recurrence	22.5	32.0	31.3	36.8	21.7	11.1	37.5	77.8	46.1	16.7	30.1
Of those with recurrence—timing of recurrence post-diagnosis											
0–1 years	9.1	0.0	14.7	0.0	7.1	0.0	12.7	0.0	14.3	0.0	9.1
1–2 years	10.9	0.0	13.1	0.0	20.0	0.0	5.6	14.3	4.9	20.0	9.6
2–3 years	9.1	0.0	3.3	0.0	0.0	0.0	0.0	0.0	7.3	0.0	4.8
3–5 years	25.5	25.0	21.3	0.0	0.0	0.0	27.8	0.0	24.4	20.0	21.6
5–9 years	21.8	12.5	19.7	42.9	20.0	100.0	33.3	57.1	31.7	20.0	26.0
10+ years	9.1	37.5	9.8	28.6	0.0	0.0	11.1	14.3	17.1	40.0	13.5
Missing data	14.5	25.0	18.0	28.6	40.0	0.0	11.1	14.3	9.8	0.0	15.4

[a]Note small cell size for some disease stages, % reported with caution.

TABLE 3 Chemotherapy by stage of disease for participants with adult GCT.

	Stage at diagnosis						
	IA	IB	IC	II	III-IV	Don't know/missing	Total
(*N*)	(244)	(25)	(195)	(51)	(57)	(119)	(691)
Chemotherapy							
No. (%)	4 (1.6)	4 (16.0)	42 (21.5)	33 (64.7)	36 (63.2)	11 (9.2)	130 (18.8)
Protocols for those having chemotherapy[a]							
(*N*)	(4)	(4)	(42)	(33)	(36)	(11)	(130)
BEP, No. (%)	2 (50)	2 (50)	14 (33)	11 (33)	18 (50)	1 (9.1)	48 (36.9)
Carboplatin and paclitaxel, No. (%)	2 (50)	2 (50)	23 (54.8)	12 (36.4)	10 (27.8)	6 (54.5)	55 (42.3)

Abbreviations: BEP, bleomycin, etoposide and platinum; GCT, granulosa cell tumor.

[a]% does not add to 100% because only the most common protocols shown.

Table 5 shows the frequency of blood tests and scans for those without recurrent disease by time since diagnosis. Of respondents reporting these tests, blood tests ($p < .01$) and scans ($p = .023$) were more frequent for those more recently diagnosed, with 56% of those diagnosed in the previous year reporting blood tests every 1–3 months. Just fewer than 50% of those without recurrent disease had missing data or reported no plans for follow-up scans. Approximately 25% of those diagnosed in the previous year and 20% of those diagnosed within the previous 2 years reported having scans every 2–4 months or every 6 months (Table 5).

Monitoring of recurrent disease

Forty-seven percent of those with recurrent disease had monitoring blood tests every 1–3 months, 21% had monitoring blood tests every 3–6 months, and 7% had monitoring blood tests every year. Inhibin A&B (49%), inhibin B (27%), and CA-125 (34%) were most commonly monitored. Seventy-five percent had scans as part of their monitoring, with 24% having scans every 2–4 months, 18% having scans every 6 months, and 13% having scans when indicated by blood test results. Blood tests ($p < .001$) and scans ($p < .001$) were more frequent for those with recurrent disease than for those that were disease-free.

TABLE 4 Primary tumor and treatment characteristics of participants by recurrence experience.

	No recurrence, No. (%)	Recurrence, No. (%)	Total, No. (%)	Univariate *p*	*p* Adjusting for time since diagnosis
Total	522 (70.3)	221 (29.7)	743 (100)		
Type of disease					
Adult	691 (92.5)	483 (94.1)	208 (93.0)		
Juvenile	39 (7.5)	13 (5.9)	52 (7.0)	.438	.634
Menopausal at diagnosis					
Postmenopausal	73 (14.0)	18 (9.1)	91 (12)		
Premenopausal	449 (86.0)	203 (91.9)	652 (87.8)	.026	.198
Tumor size					
<8.5 cm	187 (35.8)	69 (31.2)	256 (34.5)		
8.5+ cm	214 (41.0)	82 (37.1)	296 (39.8)		
Missing	121 (23.2)	70 (31.7)	191 (25.7)	.05	.391
Tumor stage					
IA	204 (39.0)	58 (26.0)	262 (35.0)		
IB	17 (3.3)	8 (3.6)	25 (3.4)		
IC	148 (28.4)	64 (29.0)	212 (28.5)		
II	41 (7.9)	14 (6.3)	55 (7.4)		
III-IV	36 (6.9)	26 (11.8)	62 (8.3)		
Not reported/missing	76 (14.6)	51 (23.1)	127 (17.1)	.002	.004
Type of surgery					
Laparoscopic	258 (49.0)	96 (43.0)	354 (47.6)		
Open	234 (44.8)	119 (53.8)	353 (47.5)	.057	.02
Tumor rupture					
No	287 (55.0)	77 (34.8)	364 (49.0)		
Cut during surgery	60 (11.5)	31 (14.0)	91 (12.2)		
Ruptured	126 (24.1)	88 (39.8)	214 (28.8)		
Not sure/missing	49 (9.4)	25 (11.3)	74 (10.0)	.000	.001
Bagged					
Yes	121 (23.2)	44 (19.9)	165 (22.2)		
No	48 (9.2)	34 (15.4)	82 (11.0)		
Not sure/missing	353 (67.6)	143 (64.7)	496 (66.8)	.042	.102
Bagged if tumor cut or rupture					
No	27 (14.5)	28 (23.5)	55 (18.0)		
Yes	35 (18.8)	17 (14.3)	52 (17.0)		
Missing	124 (66.7)	74 (62.2)	198 (64.9)	.113	.126
Chemotherapy					
None	415 (79.5)	185 (83.7)	600 (80.8)		
Yes	107 (20.5)	36 (16.3)	143 (19.2)	.183	.176
Time since diagnosis					
Same year	206 (41.0)	5 (2.3)	211 (29.4)		
1 year	110 (21.9)	11 (5.1)	121 (16.9)		
2 years	45 (8.9)	15 (7.0)	60 (8.4)		

(Continues)

TABLE 4 (Continued)

	No recurrence, No. (%)	Recurrence, No. (%)	Total, No. (%)	Univariate *p*	*p* Adjusting for time since diagnosis
3–4 years	54 (10.7)	24 (11.2)	78 (10.9)		
5+ years	88 (17.5)	160 (74.4)	248 (34.5)	.000	na

Abbreviation: na, not applicable.

TABLE 5 Frequency of blood tests and scans for participants without recurrent disease reporting that they had regular follow-up tests/scans by time since diagnosis.

	Time from diagnosis to survey					
	Same year	1 year	2 years	3–5 years	6+ years	Total
(*N*)	(206) %	(110) %	(45) %	(71) %	(71) %	(503) %
Blood tests frequency						
Every 1–3 months	28.2	55.5	37.8	22.5	4.2	30.8
Every 3–6 months	17.5	24.5	37.8	56.3	21.1	26.8
Once a year	0.0	0.9	13.3	8.5	47.9	9.3
Other	13.6	8.2	6.7	7.0	12.7	10.7
Missing	40.8	10.9	4.4	5.6	14.1	22.3
Scan frequency[a]						
Every 2–4 months	8.7	11.8	6.7	5.6	2.8	8.0
Every 6 months	8.3	12.7	13.3	11.3	4.2	9.5
Once a year	5.3	8.2	20.0	25.4	19.7	12.1
As needed	3.9	8.2	8.9	5.6	12.7	6.8
Other	5.8	15.5	6.7	16.9	15.5	10.9
Missing/no follow-up	63.6	37.3	42.2	33.8	43.7	48.9

[a]Scans can include ultrasounds, computed tomography, x-rays, positron emission tomography, and magnetic resonance imaging.

DISCUSSION

Although there is a growing recognition of the need for involvement of patients and the public across the entire research process,[8,9] to date, consumer involvement has mainly focused on setting research questions and/or focus and recruitment.[11] The development of information and support groups on social media platforms has provided a unique opportunity for greater collaboration between consumers and researchers. This may be of particular benefit when the disease is rare because the global reach of these platforms allows relatively large numbers of people with the condition to connect.[18,19] Our study suggests the feasibility of engaging with communities that have developed via social media platforms. The current study used data from a survey of over 700 members of the GCT-SS Facebook group and represents one of the largest data sets assessing experiences with treatment and follow-up for those with GCT. The study is one of the few to use a PPI approach for data collection. Survey questions reflect the issues of interest to those living with *a*GCT or *j*GCT. Strong participation rates and minimal missing data suggest good levels of engagement in the survey by GCT-SS members. Treatment, follow-up, and recurrence experiences were key issues but questions relating to menstrual history, PCOS, and family history of cancer, reflect interest in understanding the role of putative risk factors for GCT. Although some participants in the study had been living with GCT for over 20 years, most were diagnosed within the previous 5 years, suggesting treatment patterns reported broadly reflect current practices. However, because the diagnostic period of those in the study ranged over a number of years, changes in treatment practices could also be examined. This showed greater use of carboplatin and paclitaxel protocols after 2015 reflecting a change in practice recommendations to reduce toxicities.[2] Although limitations of our data, including a selective sample and lack of validation, means it needs to be interpreted cautiously, the patterns of primary treatment found reflect those reported in several long-term follow-up studies.[21–24] The range of follow-up care experiences reported here including frequency of blood tests and scans and the type of markers monitored highlights the need for further work to understand follow-up practices for survivors of this rare cancer.

Most of those completing the survey were under 50 years of age at diagnosis. This finding contrasts patterns seen from treatment center databases where the average age at diagnosis is approximately 50 years.[3,25] The younger age of participants likely reflects

that the survey was administered to members of a specific Facebook group, with Facebook users known to skew to a younger age.[26] However, the younger age suggests that ovarian cancer and GCT should not be considered only a disease of older people. Despite the younger age, treatment patterns were similar to those reported in case-series from specific treatment centers.[21,22,24,25]

Approximately 30% of those with *a*GCT in this study reported recurrent disease, with this most commonly occurring within 10 years of diagnosis. The overall recurrence rate was slightly lower than the 35% reported in a long-term follow-up of cases in several European countries.[3,5] Reviews suggest a recurrence rate for aGCT of approximately 33% that is comparable to rates reported here.[2,4] Although the recurrence rate for those with stage I disease is slightly higher than reported elsewhere,[4,27] our data along with studies reporting a relapse rate of approximately 20% for those with stage I disease, suggest that monitoring and follow-up is needed for all stages of this disease. Relapse has been reported to generally occur within a range of 4 –8 years of diagnosis,[2,4,24] with the study from the three European countries finding a median time to recurrence of 7.2 years.[3] Although these findings suggest long-term follow-up of GCT patients is warranted, others have noted that most guidelines or recommendations discuss 5-year follow-up protocols.

Identification of prognostic factors for *a*GCT has been identified as a key area of research.[2] Consumers are also keen to identify these factors as evidenced by a number of questions in the survey assessing putative risk factors. Although the survey used in the current study asked a number of questions assessing family history of cancer, reproductive, and menstrual history, other potential risk factors were not assessed. Further work is needed in this area and our study suggests that working with social media groups may provide one mechanism of engaging with relevant populations to gather information in this area.

Previous studies have suggested that tumor rupture is associated with increased risk of relapse,[21,24] and a question assessing whether the tumor ruptured or was cut during surgery was included in the GCT-SS survey. The proportion of respondents reporting tumor rupture during surgery (either spontaneous or cut) in the current study (41%) was slightly higher than reported elsewhere (36%)[24] that may reflect biases in our sample and limitations in data. Like others,[21] we found an association between tumor rupture and relapse, although we note this association is not found in all reports.[27] Similar to others,[24] the current study did not find any difference in risk of relapse between surgically or spontaneously ruptured tumors. The interest in this issue by consumers and the associations found in studies to date suggest further work is needed to understand how to avoid this potential risk factor and to identify best management practices if a tumor ruptures.[24]

GCT-SS were interested in understanding whether tumor bagging reduced the risk of recurrence when the tumor was cut or ruptured. Although the current study found those reporting recurrence were less likely to report tumor bagging when all respondents were included in the analyses, there was no association when this was examined in only those with ruptured tumors (either cut or spontaneous). There was a large amount of missing data regarding tumor bagging in our study and this is likely to influence findings. The amount of missing data for this variable contrasts with the smaller amount found for questions assessing tumor rupture and may suggest many are not informed about or forget this aspect of their surgery. Given consumers interest in this area, future studies using medical record data could report the prevalence of tumor bagging for those treated surgically for ovarian GCT.

Current follow-up recommendations include gynecological examinations with ultrasound and blood samples for 3–5 years with serum inhibin B and/or anti-Müllerian hormone (AMH) recognized as key tumor markers for monitoring.[2] The current study shows that follow-up mostly involved monitoring of inhibin B with few reporting AMH monitoring. There was substantial variation in the reported frequency of follow-up tests; reasons for this warrant investigation. Because GCT is associated with late relapse,[2,21,28] follow-up longer than 5 years may be warranted.[3] Regardless of recurrence status, approximately one-third of respondents reported having CA-125 tests despite little evidence for the efficacy of this test for GCT. The inclusion of questions assessing follow-up care indicates consumers' interest in understanding the range of experiences in this area. Because most studies reporting follow-up for ovarian GCT do not detail follow-up schedules,[3,21–23] further investigation is needed to understand experiences in this area and to determine the optimal follow-up protocols for those with and without recurrent disease.

*j*GCT is rare and has different molecular and clinical features to *a*GCT.[4] The inclusion of 52 respondents with *j*GCT represents one of the largest surveys of treatment experiences and outcomes for those with this tumor type. Although 50% of those with *j*GCT were diagnosed before the age of 30, a number were diagnosed later, suggesting a range of experiences for those with these tumors. Respondents with *j*GCT were more likely to report larger tumors at diagnosis. Although further work is needed to understand the reasons for this difference, perceptions of cancer risk for younger people may influence decisions regarding the need for investigations. Treatment of *j*GCT was generally similar to *a*GCT, although reflecting their younger age, fertility sparing treatment was more common for those with *j*GCT. There is little information around recurrence for *j*GCT. In this study, 25% of those with *j*GCT had recurrent disease, with this most likely within 12 months of their initial diagnosis. However, with only approximately 50 respondents with *j*GCT in our study, further work is needed to understand treatment patterns and outcomes for this group.

Although our study reflects an example of consumer-directed and -collected data, a number of limitations need to be noted. We relied on respondents' self-report of treatment and follow-up experiences. Although results reflect findings from other studies, details on some elements of treatment were not assessed, and verification of procedures could not be undertaken. In addition, the survey did not assess common tumor markers at diagnosis that may limit understanding of follow-up care patterns and the use of serum and/or plasma markers for detecting recurrence. Because dates for diagnosis and recurrence were not obtained, survival analyses could not be

undertaken. Analyses are largely bivariate and differences in stage and time since diagnosis may influence some findings. Because of the survey being completed by members of a Facebook group, the sample may be biased toward younger participants and this needs to be noted in the treatment patterns reported. Additionally, because only survivors could be members of this Facebook group, a survival bias is inherent in the sample. Because questions were developed to address the concerns and interests of members of GCT-Sisters Facebook group, questions clinicians or researchers may want to know may not have been collected. This may highlight potential differences in interest areas of people living with different conditions and researchers working in the area. Ensuring dialogue between these groups by PPI in research can assist in reducing differences.

Despite these limitations, the study provides useful insight into the questions those living with GCT are interested in answering. Others have noted that the low incidence of GCT makes it difficult to develop the evidence needed to direct management practices.[4,5,21] The larger studies needed to determine best practice care for these rare cancers will likely need an international approach. This study demonstrates that consumers are keen to be partners in research. Adopting a PPI approach to research and using naturally forming consumer groups on social media platforms may assist with developing the evidence base for care and for supporting those living with GCT ovarian cancer.

AUTHOR CONTRIBUTIONS

Victoria M. White: Conceptualization, analysis, results interpretation, writing–original draft, writing–review and editing, and funding acquisition. **Maria Alexiadis:** Conceptualization, results interpretation, and writing–review and editing. **Kimberly D Eroh:** Conceptualization, survey construction, results interpretation, writing–review and editing, and funding acquisition. **M. Kaye Ackermann:** Conceptualization, survey construction, results interpretation, writing–review and editing, and funding acquisition. **Sue Rodgers:** Conceptualization, survey construction, results interpretation, writing–review and editing, and funding acquisition. **Linda M. Langdale:** Conceptualization, survey construction, results interpretation, writing–review and editing, and funding acquisition. **Natasha E. Armour:** Conceptualization, survey construction, results interpretation, writing–review and editing, and funding acquisition. **Thomas W. Jobling:** Results interpretation, writing–review and editing, and funding acquisition. **Peter J. Fuller:** Conceptualization, results interpretation, writing–review and editing, and funding acquisition. **Simon Chu:** Conceptualization, results interpretation, writing–review and editing, and funding acquisition. All authors have read and agreed to the published version of the manuscript.

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CONFLICT OF INTEREST STATEMENT

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ORCID


Victoria M. White https://orcid.org/0000-0001-6619-8484

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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