TO THE EDITOR:

Dyadic communication in rare cancer: a registry-based study of patients with Erdheim-Chester disease and their caregivers

Anne S. Reiner,[1] Allison J. Applebaum,[2] Dana Bossert,[3] Justin J. Buthorn,[3] Allison M. Sigler,[3] Deanna Fournier,[4] Kathleen Brewer,[5] Thomas M. Atkinson,[2] Kathleen A. Lynch,[2] Jaime Gilliland,[2] Jun J. Mao,[6] Katherine S. Panageas,[1] and Eli L. Diamond[3]

[1]Department of Epidemiology and Biostatistics, [2]Department of Psychiatry and Behavioral Sciences, and [3]Department of Neurology, Memorial Sloan Kettering Cancer Center, New York, NY; [4]Histiocytosis Association, Pitman, NJ; [5]Erdheim-Chester Disease Global Alliance, DeRidder, LA; and [6]Department of Medicine, Memorial Sloan Kettering Cancer Center, New York, NY

A cancer diagnosis causes distress and instability not only to the affected patient but also to the psychosocial ecosystem of the patient, most notably their family and friend caregivers. Caregivers support patients in all components of their disease trajectory, from navigating the diagnosis to discussing the symptoms, treatments and side effects, and even advanced care planning. Investigating the dyad, that is, the patient-caregiver pair, is of critical importance. Open dyadic communication between a patient with cancer and their caregiver is associated with positive elements of dyadic coping, such as increased intimacy and emotional acceptance, fewer physical symptoms, and less psychological distress.[1,2] Conversely, avoidance of communication about illness is associated with adverse features of dyadic coping, such as decreased intimacy and relational satisfaction.[3]

Existing literature about dyadic communication in cancer has examined common cancers; there has been little examination of patient-caregiver communication about illness for dyads navigating a highly rare cancer. Although individual rare cancers are uncommon, rare cancers, in aggregate, constitute 20% of all cancer diagnoses in the United States,[4] rendering them an important and distinct entity to investigate from the viewpoint of patient, caregiver, and dyadic experience.

Erdheim-Chester disease (ECD) is a very rare blood cancer, with ~1500 cases since 1930. It is a disease with protean manifestations that can affect any structure, including bones, skin, eyes, and brain.[5] Additionally, it is nearly invariably defined by delayed diagnosis exceeding 2 years.[6] We have demonstrated dense, widely varied, and poor symptomatology, including frequent fatigue, pain, sadness, and anxiety in patients with ECD.[7,8] Among caregivers of individuals with ECD, we have identified a constellation of challenges emanating directly from the experience of providing support for a patient with a rare disease, such as heightened anxiety, distress, and unmet support needs related to protracted undiagnosed illness and scarcity of reliable information about ECD. Herein, we investigated communication about illness within the ECD patient-caregiver dyad; we hypothesized that factors salient to the experience of patients and caregivers with rare cancer would be associated with worse dyadic communication about illness.

This study consists of an institutional review board–approved registry study and caregiver study maintained at Memorial Sloan Kettering Cancer Center (#NCT03329274 and #NCT03990428). All data are contained in Research Electronic Data Capture, a platform for collecting data in a secure web-based interface.

Participants were patients with biopsy-confirmed ECD and their caregivers. Patient participants were self-referred or referred from within the Memorial Sloan Kettering Cancer Center cohort by their health care providers or from centers recognized by the ECD Global Alliance. Patients and caregivers provided informed consent to participate and complete self-reported assessments. Dyads of patient

Table 1. Caregiver-patient dyad characteristics (N = 63)

Variable	Level	n	%	Median	Mean	SD	Min	Max
Caregiver age at assessment	Continuous, y	63	100	54.0	52.9	12.2	20.0	79.0
Patient age at assessment*	Continuous, y	63	100	55.7	54.1	12.7	20.2	73.5
Caregiver CCAT†	Continuous total score	63	100	11.0	11.1	4.0	5.0	24.0
Caregiver CCID‡	Continuous mean score	63	100	2.2	2.3	1.1	1.0	4.2
Caregiver sex	Male	15	24					
	Female	48	76					
Patient sex	Male	41	65					
	Female	22	35					
Caregiver race/ethnicity	White	50	79					
	Latino	3	5					
	White, Latino	3	5					
	API	3	5					
	API, Latino	2	3					
	API, White	1	2					
	Indian/Aleut/Eskimo	1	2					
Patient race/ethnicity	White	55	87					
	Black or African American	1	2					
	Asian	5	8					
	Unknown	2	3					
Caregiver education	High school diploma/GED	7	11					
	Vocational school or some college	8	13					
	College degree	28	44					
	Professional or graduate school	20	32					
Patient education	Some high school	1	2					
	High school diploma/GED	8	13					
	Vocational school or some college	10	16					
	College degree	25	40					
	Professional or graduate school	19	30					
Patient income	<$75 000	17	27					
	≥$75 000	37	59					
	Unknown	9	14					
Caregiver income	<$75 000	21	33					
	≥$75 000	39	62					
	Unknown	3	5					
Caregiver employment	Paid full-time	27	43					
	Paid part-time	4	6					
	Self-employed	4	6					
	Student full-time	1	2					
	Homemaker	6	10					
	Unemployed, disabled	1	2					
	Retired	14	22					
	Unemployed, looking for work	1	2					
	Unemployed, not looking for work	4	6					
	Unknown	1	2					

API, Asian/Pacific Islander; GED, graduate equivalency degree; LCH, Langerhans cell histiocytosis; Max, maximum; Min, minimum; MSKCC, Memorial Sloan Kettering Cancer Center; RDD, Rosai-Dorfman disease.

*Using the assessment date closest to that of the caregiver's assessment.

†The CCAT family communication subscale comprises 5 items scored on a 6-point Likert scale ranging from 1, "Strongly disagree" to 6, "Strongly Agree," with higher scores reflective of poorer communication. The CCAT family communication subscale score is the total of all 5 responses.

‡The CCID comprises 5 items scored on a 5-point Likert scale ranging from 1, "not at all" to 5, "to a large extent," with higher scores indicative of poorer communication. The CCID total score is the average of all 5 responses.

Table 1 (continued)

Variable	Level	n	%	Median	Mean	SD	Min	Max
Patient employment	Paid full-time	17	27					
	Paid part-time	2	3					
	Self-employed	6	10					
	Student full-time	2	3					
	Homemaker	1	2					
	Not employed-disabled	17	27					
	Retired	11	17					
	Unemployed, looking for work	1	2					
	Unemployed, not looking for work	6	10					
Ever seen at MSKCC	No	25	40					
	Yes	38	60					
Diagnosis	ECD	54	88					
	Mixed ECD/LCH	7	11					
	Mixed ECD/RDD	2	3					

API, Asian/Pacific Islander; GED, graduate equivalency degree; LCH, Langerhans cell histiocytosis; Max, maximum; Min, minimum; MSKCC, Memorial Sloan Kettering Cancer Center; RDD, Rosai-Dorfman disease.

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participants with ECD who enrolled from 2018 to 2021 and their caregivers with completed assessments were included.

The main outcome was caregiver-assessed dyadic communication, which was evaluated using 2 caregiver-reported measures: (1) The Cancer Communication Assessment Tool for Patients and Families (CCAT[9]) family communication subscale and (2) The Caregivers' Communication with Patients about Illness and Death (CCID[10]) scale.

Patient and caregiver demographics and patient disease characteristics were recorded. For patients only, self-reported assessments included the Functional Assessment of Cancer Therapy–Cognitive assessment, ECD Symptom Scale, Instrumental Activities of Daily Life, Brief Fatigue Inventory, and Brief Pain Inventory. For caregivers only, self-reported assessments included the Duke–University of North Carolina Functional Social Support Questionnaire, University of California, Los Angeles Loneliness Scale, Difficulties in Emotional Regulation Scale, and Caregiver Reaction Assessment. For both patients and caregivers, assessments included the Supportive Care Needs Survey and the Hospital Anxiety and Depression Scale.

The CCAT family communication subscale and average CCID scale were correlated using Pearson correlation coefficient. Demographics, patient disease characteristics, patient-reported assessments, and caregiver-reported assessments were individually associated with caregiver-assessed dyadic communication, with univariable linear regression models using the CCAT family communication subscale and the CCID scale separately as outcomes. Tests were 2 sided with statistical level of significance <.05. Analyses were performed in SAS version 9.4.

Sixty-three dyads contributed to analyses (Table 1). Patients were aged 54.1 years, on average, at assessment, and caregivers were 52.9 years. Patients were mostly male (65%), and caregivers

mostly female (76%). The mean and standard deviations (SD) of the CCAT family communication subscale and average CCID scale were 11.1 (SD, 4.0) and 2.3 (SD, 1.1), respectively, and the measures were moderately correlated ($\rho = 0.45$; 95% confidence interval [CI], 0.22-0.62).

Modeling the CCAT family communication subscale, caregivers of patients on treatment were more likely to report worse caregiver-assessed dyadic communication (Table 2). Caregivers of patients with unmet psychological needs ($\beta_{CCAT} = 2.25$; 95% CI, 0.27-4.23) or greater impact of perceived cognitive impairments ($\beta_{CCAT} = -0.21$; 95% CI, −0.42 to −0.01) were more likely to report worse caregiver-assessed dyadic communication, as were caregivers of patients rating their top 3 ECD symptoms occurring almost constantly ($\beta_{CCAT} = 7.25$; 95% CI, 1.69-12.80). If a patient reported more severe pain, the caregiver was more likely to report worse dyadic communication ($\beta_{CCAT} = 0.44$; 95% CI, 0.04-0.84). Worse caregiver anxiety ($\beta_{CCAT} = 0.30$; 95% CI, 0.08-0.52) and depression ($\beta_{CCAT} = 0.41$; 95% CI, 0.19-0.62) were associated with worse caregiver-assessed dyadic communication, as were more unmet caregiver work and social needs ($\beta_{CCAT} = 2.75$; 95% CI, 0.73-4.77) and informational needs ($\beta_{CCAT} = 2.32$; 95% CI, 0.35-4.29). Lower caregiver-perceived self-esteem ($\beta_{CCAT} = -0.28$; 95% CI, −0.55 to −0.005) and greater caregiver challenges with emotional regulation ($\beta_{CCAT} = 0.12$; 95% CI, 0.07-0.17) were associated with poorer caregiver-assessed dyadic communication.

Modeling the CCID scale mean, caregivers of patients with more anxiety reported worse communication (Table 2). Caregivers of patients with severe and interfering fatigue reported poorer communication ($\beta_{CCID} = 0.14$; 95% CI, 0.07-0.22; $\beta_{CCID} = 0.10$; 95% CI, 0.02-0.18, respectively). Similar to when the CCAT family communication subscale was the outcome, worse caregiver

Table 2. Associations with caregiver-assessed communication

Variable	Level or construct	n	%	Outcome: CCAT			Outcome: CCID		
				Estimate	95% CI	P value	Estimate	95% CI	P value
Patient age	Continuous, y	63	100	−0.003	−0.08-0.08	.94	0.00	−0.02-0.02	.70
Patient sex	Female	22	35	Ref			Ref		
	Male	41	65	0.26	−1.89-2.42	.81	0.004	−0.56-0.56	.99
Patient education	Other	19	30	Ref			Ref		
	College, professional, or graduate	44	70	−0.11	−2.35-2.13	.92	0.26	−0.32-0.84	.37
Patient income	<$75 000	17	27	Ref			Ref		
	≥$75 000	37	59	−0.62	−3.04-1.79	.61	0.11	−0.52-0.75	.72
Patient neurologic illness	No	13	21	Ref			Ref		
	Yes	47	78	0.09	−2.49-2.66	.95	−0.42	−1.08-0.24	.21
ECD duration	Continuous, y	63	100	−0.06	−0.27-0.15	.56	−0.02	−0.08-0.03	.40
Patient on treatment	No	14	22	Ref			Ref		
	Yes	49	78	2.49	0.10-4.88	.04	−0.12	−0.76-0.52	.71
Patient unmet needs (SCNS)	Met health care service needs	40	63	Ref			Ref		
	Unmet health care service needs	21	33	1.77	−0.29-3.82	.09	0.31	−0.25-0.88	.27
	Met psychological and emotional needs	30	48	Ref			Ref		
	Unmet psychological and emotional needs	32	51	2.25	0.27-4.23	.03	0.25	−0.29-0.79	.36
	Met physical and daily living needs	42	67	Ref			Ref		
	Unmet physical and daily living needs	19	30	1.25	−0.89-3.38	.25	0.30	−0.28-0.89	.30
	Met care and support needs	52	83	Ref			Ref		
	Unmet care and support needs	9	14	1.64	−1.17-4.45	.25	0.37	−0.39-1.13	.34
	Met sexual needs	46	73	Ref			Ref		
	Unmet sexual needs	14	22	0.28	−2.13-2.69	.82	−0.20	−0.84-0.45	.55
	Met total needs	21	33	Ref			Ref		
	Unmet total needs	41	65	1.90	−0.22-4.03	.08	0.11	−0.46-0.68	.71
Patient FACT-Cog	Perceived cognitive impairments	62	98	−0.06	−0.13-0.006	.07	−0.01	−0.03-0.01	.17
	Impact of perceived cognitive impairments	62	98	−0.21	−0.42 to −0.01	.04	−0.05	−0.10-0.00	.053
	Comments from others	62	98	−0.09	−0.48-0.31	.66	−0.02	−0.13-0.08	.64
	Perceived cognitive abilities	62	98	0.009	−0.15-0.17	.91	−0.02	−0.07-0.02	.24
Patient ECDSS	Top 3 symptoms, continuous score	61	97	0.27	−0.10-0.65	.15	0.09	−0.003-0.19	.056
	Top 5 symptoms, continuous score	61	97	0.19	−0.21-0.58	.35	0.09	−0.01-0.19	.09
	Top 3 symptoms: at least 1 not almost constantly	55	87	Ref			Ref		
	Top 3 symptoms: all almost constantly	6	10	3.92	0.51-7.33	.03	0.41	−0.50-1.32	.37
	Top 5 symptoms: at least 1 not almost constantly	57	90	Ref			Ref		
	Top 5 symptoms: all almost constantly	2	3	7.25	1.69-12.80	.01	0.18	−1.32-1.68	.81
Patient IADL	Continuous score	62	98	−0.34	−0.78-0.10	.13	−0.04	−0.16-0.08	.48

BFI, Brief Fatigue Inventory; BPI, Brief Pain Inventory; ECD-SS, ECD Symptom Scale; FACT-Cog, Functional Assessment of Cancer Therapy–Cognitive assessment; FSSQ, Functional Social Support Questionnaire; HADS, Hospital Anxiety and Depression Scale; IADL, Instrumental Activities of Daily Life; Ref, reference category; SCNS, Supportive Care Needs Survey; UNC, University of North Carolina.

Table 2 (continued)

Variable	Level or construct	n	%	Outcome: CCAT			Outcome: CCID		
				Estimate	95% CI	P value	Estimate	95% CI	P value
Patient anxiety	Continuous HADS anxiety score	63	100	0.18	−0.10-0.47	.20	0.07	0.00-0.15	.04
Patient depression	Continuous HADS depression score	63	100	0.16	−0.07-0.40	.17	0.04	−0.02-0.10	.20
Patient BPI	Severity construct	62	98	0.44	0.04-0.84	.03	0.10	−0.002-0.21	.055
	Interference construct	61	97	0.24	−0.13-0.61	.20	0.05	−0.04-0.15	.26
	Total	61	97	0.32	−0.07-0.70	.11	0.07	−0.03-0.17	.17
Patient BFI	Severity construct	62	98	0.28	−0.02-0.59	.07	0.14	0.07-0.22	.0002
	Interference construct	62	98	0.22	−0.09-0.54	.17	0.10	0.02-0.18	.01
	Total	62	98	0.26	−0.06-0.58	.11	0.12	0.04-0.20	.003
Caregiver age	Continuous, y	63	100	−0.006	−0.09-0.08	.89	−0.01	−0.04-0.003	.09
Caregiver sex	Female	48	76	Ref			Ref		
	Male	15	24	−0.63	−3.04-1.78	.60	0.26	−0.36-0.89	.40
Caregiver education	Other	15	24	Ref			Ref		
	College, professional, or graduate	48	76	1.007	−1.33-3.47	.38	0.47	−0.14-1.09	.13
Caregiver anxiety	Continuous HADS anxiety score	63	100	0.30	0.08-0.52	.009	0.09	0.03-0.14	.002
Caregiver income	<$75 000	21	33	Ref			Ref		
	≥$75 000	39	62	−0.007	−2.21-2.20	.99	−0.002	−0.58-0.57	.99
Caregiver depression	Continuous HADS depression score	63	100	0.41	0.19-0.62	.0003	0.08	0.02-0.14	.009
Caregiver unmet needs (SCNS)	Met health care service needs	33	52	Ref			Ref		
	Unmet health care service needs	30	48	1.82	−0.18-3.83	.07	0.10	−0.43-0.63	.71
	Met psychological and emotional needs	24	28	Ref			Ref		
	Unmet psychological and emotional needs	39	62	1.68	−0.40-3.75	.11	0.22	−0.33-0.77	.42
	Met work and social needs	40	63	Ref			Ref		
	Unmet work and social needs	23	37	2.75	0.73-4.77	.008	0.17	−0.39-0.72	.55
	Met informational needs	32	51	Ref			Ref		
	Unmet informational needs	31	49	2.32	0.35-4.29	.02	0.27	−0.26-0.80	.31
	Met total needs	15	24	Ref			Ref		
	Unmet total needs	48	76	2.29	−0.05-4.63	.06	−0.19	−0.82-0.43	.54
Caregiver Duke-UNC FSSQ	Continuous score	63	100	−0.85	−1.90-0.20	.11	−0.04	−0.32-0.23	.75
Caregiver Loneliness	Continuous score	63	100	0.16	−0.07-0.40	.17	0.05	−0.01-0.11	.12
Caregiver Difficulties in Emotional Regulation	Nonacceptance of emotional response	62	98	0.34	0.14-0.54	.001	0.07	0.02-0.13	.01
	Difficulty engaging in goal-directed behavior	63	100	0.28	−0.0004-0.57	.05	0.10	0.02-0.17	.01
	Impulse control difficulties	63	100	0.44	0.19-0.69	.0008	0.07	0.01-0.14	.04
	Lack of emotional awareness	63	100	0.35	0.17-0.52	.0002	0.09	0.04-0.13	.0003
	Limited access to emotional regulation strategies	63	100	0.49	0.29-0.68	< .0001	0.11	0.05-0.16	.0002
	Lack of emotional clarity	63	100	0.46	0.18-0.73	.001	0.12	0.05-0.19	.0015
	Total	62	98	0.12	0.07-0.17	< .0001	0.03	0.02-0.04	< .0001

BFI, Brief Fatigue Inventory; BPI, Brief Pain Inventory; ECD-SS, ECD Symptom Scale; FACT-Cog, Functional Assessment of Cancer Therapy–Cognitive assessment; FSSQ, Functional Social Support Questionnaire; HADS, Hospital Anxiety and Depression Scale; IADL, Instrumental Activities of Daily Life; Ref, reference category; SCNS, Supportive Care Needs Survey; UNC, University of North Carolina.

Table 2 (continued)

Variable	Level or construct	n	%	Outcome: CCAT			Outcome: CCID		
				Estimate	95% CI	P value	Estimate	95% CI	P value
Caregiver Burden	Self-esteem	63	100	−0.28	−0.55 to −0.005	.046	−0.07	−0.14-0.003	.06
	Impact on schedule	63	100	0.11	−0.09-0.30	.27	−0.01	−0.06-0.04	.61
	Lack of family support	63	100	0.14	−0.06-0.35	.17	−0.02	−0.08-0.03	.38
	Impact on finances	63	100	0.14	−0.14-0.43	.32	0.09	0.02-0.16	.02
	Impact on health	63	100	0.30	−0.17-0.77	.20	−0.01	−0.13-0.11	.87

BFI, Brief Fatigue Inventory; BPI, Brief Pain Inventory; ECD-SS, ECD Symptom Scale; FACT-Cog, Functional Assessment of Cancer Therapy–Cognitive assessment; FSSQ, Functional Social Support Questionnaire; HADS, Hospital Anxiety and Depression Scale; IADL, Instrumental Activities of Daily Life; Ref, reference category; SCNS, Supportive Care Needs Survey; UNC, University of North Carolina.

anxiety and depression as well as greater caregiver challenges with emotional regulations were all associated with worse caregiver-assessed dyadic communication ($\beta_{CCID} = 0.09$; 95% CI, 0.03-0.14; $\beta_{CCID} = 0.08$; 95% CI, 0.02-0.14; and $\beta_{CCID} = 0.03$; 95% CI, 0.02-0.04, respectively). Finally, higher caregiver financial burden was associated with worse caregiver-assessed dyadic communication ($\beta_{CCID} = 0.09$; 95% CI, 0.02-0.16).

In this cross-sectional study, we have identified several patient- and caregiver-related factors affecting dyadic communication in a large, paired patient-caregiver cohort of a very rare neoplasm, a field neglected by existing literature. This aligns with the Theory of Dyadic Illness Management of Lyons et al, which describes illness management, whether rare or common, as a dyadic experience.[11] The involvement of caregivers is inherent to the adult cancer experience, but the factors that influence dyadic illness management vary. A prior study by Vukcevic et al of patients and paired caregivers with diffuse large B-cell lymphoma demonstrated that patients experience caregivers as instrumental to their comprehension of treatment-related discussions, and, furthermore, that developing tailored educational materials for caregivers could aid in increasing the proportion of patients (71%) satisfied with their treatment experience.[12] In a dyadic study of patients with more common hematological cancers and their caregivers, Siminoff et al demonstrated that dyadic communication was most discordant for dyads with lower levels of education and income and for cancers diagnosed at later stages.[13] In this current study, education and income level were not associated with communication, and, although ECD is not traditionally staged, ECD duration, a potential proxy, was also not associated with communication. We demonstrated in the rare cancer setting that dyads in which patients suffered more frequent disease symptoms, greater cognitive difficulties, and more severe pain and fatigue demonstrated worse dyadic communication. Caregivers with more frequent anxiety and unmet supportive care needs, higher financial burden, and less social support experienced poorer communication about illness.

Our results can be viewed in the context of prior psychosocial studies about patients with myeloproliferative neoplasms (MPNs). US-based and international MPN studies have evaluated patient-reported outcomes in patients with MPN and demonstrated reliance on caregivers, impact of MPN upon the patient-caregiver relationship, and the need for decisional and social support.[14-19] However, to our knowledge, the existing literature did not address dyadic communication. We investigated dyadic communication among patients with ECD and their caregivers and identified factors that may be unique to rare diseases, for example, unmet informational needs. The relatively young age of our caregivers and patients, in a disease now considered chronic, underscores the importance of durable interventions to improve dyadic communication in this population. A recent meta-analysis of 14 couple-based communication intervention studies in common cancers found improvements in individual functioning and mutual communication.[20] Furthermore, dyadic web-based interventions[21] that can customize content have also shown to affect psychological health and the dyadic relationship.

Although our cross-sectional design yielded participants heterogeneous in disease trajectory and treatment, our study represents a large dyadic cohort for a rare disease and, to our knowledge, the largest dyadic cohort in ECD investigation. We have identified

meaningful patient and caregiver factors associated with communication that are amenable to interventions.

Based on successes reported for web-based interventions of dyadic communication in common cancers, we propose that intensive disease-specific remote interventions focused upon disease education, specialized supportive care needs, and management of symptoms and anxiety be tested in ECD and other rare diseases. The current post-COVID era provides unprecedented opportunity to evaluate and implement such interventions.

ORCID profiles: A.S.R., 0000-0002-1258-6112; A.J.A., 0000-0002-2140-5635; K.A.L., 0000-0002-2174-1604; J.J.M., 0000-0001-9229-0380; E.L.D., 0000-0001-5456-5961.

Correspondence: Eli L. Diamond, Department of Neurology, Memorial Sloan Kettering Cancer Center, 160 East 53rd St, 2nd Floor Neurology, New York, NY; email: diamone1@mskcc.org.

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