

Increasing Patient Self-Enrollment in the National Amyotrophic Lateral Sclerosis Registry: Lessons Learned From a Direct to Provider Campaign

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Abstract

Objective: To conduct educational and promotional outreach activities to general neurologists and to increase self-enrollment of persons with amyotrophic lateral sclerosis (ALS) in the National ALS Registry (Registry). **Methods:** A multicomponent project to educate neurologists and increase Registry self-enrollment was delivered. Project components consisted of phone calls, mailings, train-the-trainer presentations, and key informant interviews. Project-specific metrics, continuing education enrollment, and Registry self-enrollment data were analyzed to measure project efficacy. **Results:** Mailings were sent to 1561 neurologists in 6 states during 2015 to 2016. Sixty-five percent of responding neurologists remembered the mailing 3 months after receipt. Of providers who saw patients with ALS in the 3-month period, 60% read the provider guide, 22% distributed a patient guide, and 15% advised a patient to self-enroll. No changes in self-enrollment rates were observed. **Conclusion:** Targeted mailings to providers can be used to educate them about the Registry; however, most providers did not distribute materials to patients with ALS. Increases in providers receiving Registry material did not lead to increases in patient self-enrollment. **Practice Implications:** General neurologists have competing priorities, and they see patients with ALS infrequently. Neurologists could be the appropriate channel to distribute Registry information to patients, but they are not the appropriate resource to assist patients with self-enrollment. Engaging the support staff of busy specialists can help increase research response rates and information distribution. The lessons learned from this project can be applied to other rare conditions and disease specialists.

Keywords

amyotrophic lateral sclerosis (ALS), Registry, self-enrollment, provider education, provider–patient communication channel

Introduction

Amyotrophic lateral sclerosis (ALS), also known as Lou Gehrig’s disease, is a rare, fatal neuromuscular disorder. While the causes and risk factors of ALS are largely unknown and under investigation, much work has been conducted worldwide to better understand the epidemiology of the disease (1,2). In October 2008, the ALS Registry Act was signed into law, and the Agency for Toxic Substances and Disease Registry (ATSDR) was designated to develop and maintain the National ALS Registry (Registry) in the United States (3). The main goals of the Registry are to better describe the incidence and prevalence of ALS, to examine risk factors that might be associated with the disease, and to better outline demographic factors associated with the disease (4). The Registry uses two approaches to identify ALS

cases; the application of a validated algorithm to national administrative databases (5) and a voluntary, Internet-based, patient self-enrollment web portal.

The National ALS Registry has published estimates of ALS prevalence for 2010 to 2011, 2012, 2013, and 2014. The estimated prevalence for 2014 was 5.0 per 100 000

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population, the same as the 2013 estimate (4,6). The web portal, launched in October 2010, is of vital importance, as this mechanism allows patients with ALS who may not be found through the administrative data sets to enroll and be represented in the Registry (7). The web portal allows persons with ALS to voluntarily self-enroll, upon which they may elect to receive notifications regarding clinical trials and other studies, to join the National ALS Biorepository, and to take brief risk factor surveys about their disease. Only those who self-enroll through the web portal are eligible to complete the risk factor surveys as opposed to those who are only identified through the national administrative databases. Prevalence estimates are calculated from the overall registry (deduplicated data set of cases from self-enrollment web portal and national administrative data sets). In 2010 to 2011, the proportion of cases identified via the web portal only was 16%, web portal and administrative database was 15%, and administrative databases only was 69%. The majority of the 12 187 identified persons with “definite ALS” had not enrolled through the web portal, thus missing the opportunity to complete survey modules and be notified of research (7).

Continuous marketing through educational and promotional outreach activities is critical to the success of the self-enrollment component of the Registry. Agency for Toxic Substances and Disease Registry has created numerous recruitment materials that are distributed to persons with ALS at ALS gatherings such as Walks to Defeat ALS, patient symposiums, support groups, and health-care encounters. In addition, ATSDR has a comprehensive social media campaign to inform people about the Registry and encourage them to participate. Further, ATSDR has collaborated with stakeholders at 2 large ALS service organizations (Amyotrophic Lateral Sclerosis Association [ALSA] and Muscular Dystrophy Association [MDA]) and a regional organization (Les Turner ALS Foundation) to promote the Registry, and much of their work is conducted at the major ALS referral/specialty centers in the United States. Efforts to increase Registry awareness at general neurology practices and to increase self-enrollment of persons with ALS through the neurologist–patient communication channel at nonreferral/nonspecialty center neurology practices are lacking. This could be problematic in light of the fact that the ALS Surveillance Projects (Surveillance Projects) found 1 in 5 cases were reported by neurologists practicing at nonreferral centers. These Surveillance Projects, which were conducted in 3 states and 8 large metropolitan areas, were undertaken by ATSDR to help test the completeness of Registry data. Based on data collected in the Florida and New Jersey Surveillance Projects, this subset of cases was more likely to be nonwhite, male, and slightly older than cases reported from referral centers (8,9), which is the exact subset of persons with ALS that is underrepresented in the Registry (10).

It was hypothesized that knowledgeable and engaged health-care providers, including neurologists, would be more likely to advise their patients to self-enroll in the Registry.

While research has shown information alone may not be sufficient to induce a behavior change, the key audience in this project is invested in the topic area and helping those affected by ALS. Therefore, it was assumed that lack of information was the ultimate barrier to a successful patient–provider communication channel to advise patients of the Registry. This project aimed to educate and engage providers through already developed and available materials, as they had been previously reviewed for use with this population. This in turn could yield an increase in the likelihood of persons with ALS completing the self-registration process. Therefore, our objectives were to (1) implement a pilot project to conduct educational and promotional outreach activities at selected nonreferral/nonspecialty center neurology practices in the United States, to inform neurologists and their staff about the Registry, to encourage them to inform their patients about the Registry, and to increase self-enrollment in the Registry among persons with ALS through the web portal via the use of existing Registry brochures, pamphlets, and factsheets; and (2) to examine the effectiveness of educational and promotional outreach activities by reviewing self-enrollment rates among persons with ALS before and after the project period. By increasing self-enrollment rates, ATSDR will be able to produce more accurate prevalence estimates of ALS and collect risk factor survey data from a more representative sample of persons with ALS nationwide. These activities will help ATSDR fulfill its congressional mandate under the ALS Registry Act (3).

Methods

A logic model (Figure 1) was developed, and a 4 group educational and promotional outreach project using an interrupted time series design was completed. A subinvestigation of this study included a qualitative analysis of individual semistructured Key Informant Interviews (KIIs). The purpose of the KIIs was to characterize neurologists’ interactions with persons with ALS; to better understand their knowledge, attitudes, and beliefs about the Registry; and to gather feedback about Registry materials currently available and the self-enrollment process. The project followed the Quorum Review Institutional Review Board (IRB) reviewed project protocol which received an exempt determination.

Study Population

Groups 1, 2, and 3 consisted of 2 states each; group 1: New Jersey and Florida, group 2: New York and Virginia, and group 3: Ohio and Washington. Group 4 was the comparison group and consisted of the remaining 44 states. Group 1 states previously participated in the Surveillance Projects (8,9). Groups 2 and 3 states were chosen because they are similar to group 1 states by population sizes, percentage of population older than 60, percentage of minorities, and Registry enrollment rates.

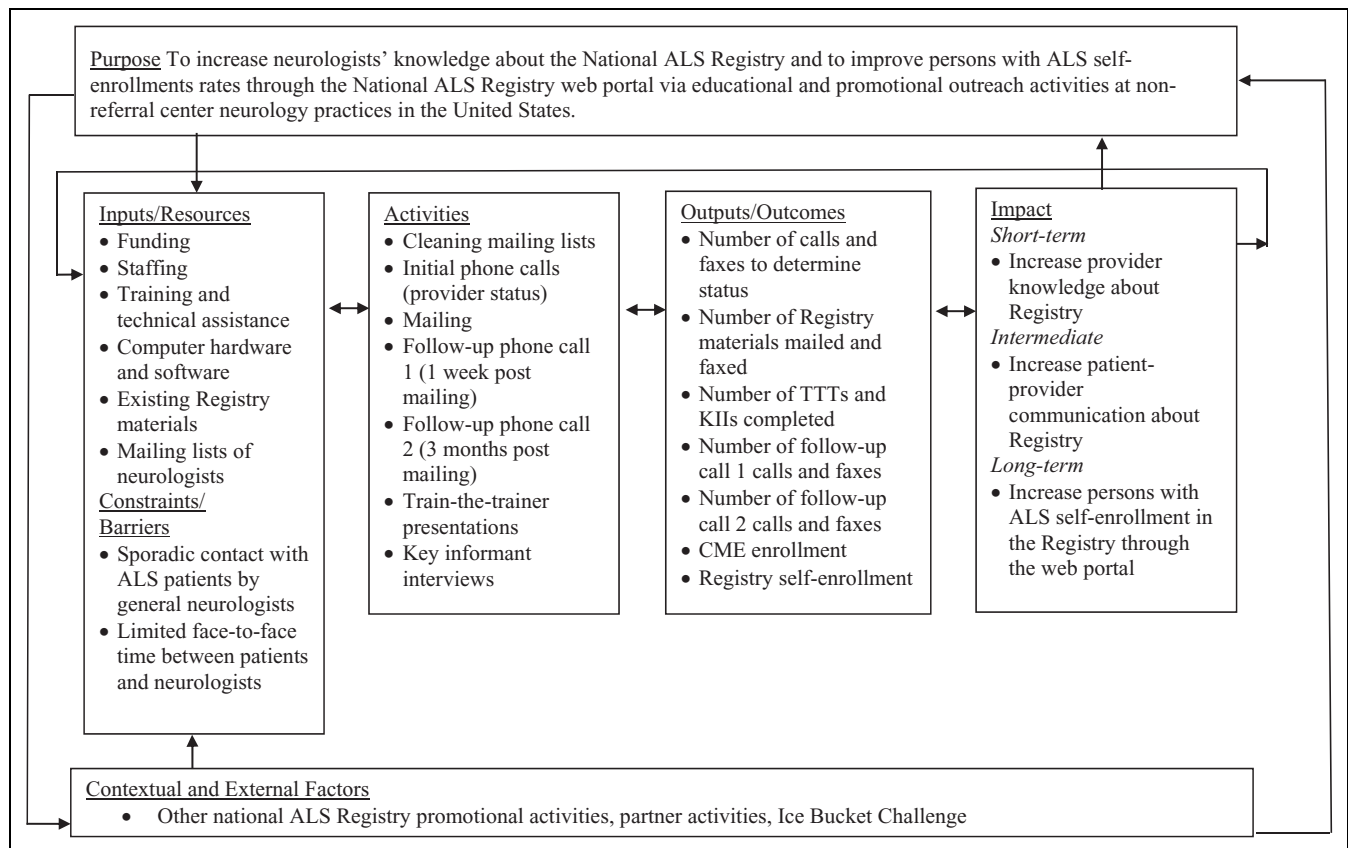


Figure 1. Logic model.

In order to be included in the study population, neurologists had to practice at a nonreferral center location in one of the participating states. Medical doctors and doctors of osteopathic medicine were included; medical residents and fellows were excluded.

The promotion intervention used neurologists as the recipients of the educational materials as opposed to other health-care team members (eg, nurses). While nurses or other health-care professionals may have more time to spend with patients, not all practice locations have nurses to provide health education and the responsibilities of nurses and nurse-patient interaction in each medical office differs. Also, the National ALS Registry provider materials were designed to target the neurologist as opposed to other members of the health-care team.

Train the Trainer (TTT) sessions provided information about the Registry and its importance as well as described the role of the provider in communicating the Registry to patients. A section of the training was dedicated to preparing the provider and/or staff members to talk to patients about the Registry, including how they can describe the Registry and how they can assist a patient through the self-enrollment process, including a walk-through of the Registry website. The TTT participants were invited via a clustered random sample with replacements for providers who were uninterested in

participating. Train the Trainer sessions lasted approximately 60 minutes.

Key Informant Interviews were conducted in a subsample of neurologists, independent of the sample selected for the TTT sessions. Key Informant Interviews were used to evaluate an increase in provider knowledge as well as to assess provider attitudes and beliefs surrounding the Registry and its promotional materials. To identify possible participants, a cluster sampling technique was used which facilitated the distribution of respondents across geographic regions in Florida and New Jersey. Neurologists on each regional list were randomly selected for participation in the KIIs and had an equal chance of being selected. A purposeful snowballing technique was also used in an attempt to increase the number of participants. Participating providers were invited to identify additional neurologists as potential participants.

Data Collection

Methods to identify and communicate with neurologists in groups 1 to 3 were similar to the surveillance projects (8,9,11,12). Briefly, American Medical Association mailing lists of all neurologists in groups 1 to 3 were purchased from Medical Marketing Services. Information was verified and expanded through public data sources, such as state licensing board websites. Neurologists known to be practicing in a

Table 1. Educational and Promotional Outreach Components.

Project Components	Group 1		Group 2		Group 3		Group 4
	Florida	New Jersey	New York	Virginia	Ohio	Washington	Comparison Group of 44 Remaining States
Initial phone call to determine status	✓	✓	✓	✓			
Additional initial phone calls and faxes (as needed)	✓	✓	✓	✓			
Mailing	✓	✓	✓	✓	✓	✓	
Follow-up phone call #1 (1-week postmailing)	✓	✓	✓	✓			
Additional follow-up phone call #1 and faxes (as needed)	✓	✓	✓	✓			
Train-the-trainer site visits	✓	✓					
Key informant interviews	✓	✓					
Follow-up phone call #2 (3-months postmailing)	✓	✓	✓	✓			
Additional follow-up phone call #2 and faxes (as needed)	✓	✓	✓	✓			
Self-enrollment data obtained from ATSDR	✓	✓	✓	✓	✓	✓	✓

Abbreviation: ATSDR, Agency for Toxic Substances and Disease Registry.

specialty unlikely to treat patients with ALS (eg, child neurologist) and known medical residents were excluded from the list. The final list contained 4089 neurologists in groups 1 to 3.

Table 1 describes the intervention components of this project. An initial phone call to neurologists in groups 1 and 2 was conducted to characterize providers' status as currently diagnosing/treating (Yes), would diagnose/treat (Would), would never diagnose/treat patients (No) with ALS, or ineligible (eg, practicing at a referral center) to determine how many patients with ALS are seen per year and to confirm contact information.

Neurologists identified as Yes and Would in groups 1 and 2, and all neurologists in group 3 received a mail containing a cover letter, ATSDR endorsement letter, and ATSDR-created resource materials, including posters, continuing medical education (CME) announcements, provider and patient guides, and infographics. The number of brochures sent was scaled based on number of patients with ALS seen per year. All promotional materials that were used have been designed by ATSDR and are currently in use by the National ALS Registry. Four states had mailings sent in 1 wave, Florida was sent in 2 waves, and New York was sent in 3 waves. The mailings were sent over a 7-month period in 2015 and 2016.

Follow-up phone calls to groups 1 and 2 were completed 1 week after the mailings were sent to determine whether the informational packet was received, to answer any questions, and to encourage neurologists to distribute Registry information to their patients with ALS. The materials were faxed to the office if a practice did not remember receiving a packet in the mail. All phone calls and faxes were tracked by provider name. A maximum combination of 5 phone call attempts and 2 faxes were used to complete this round of phone calls.

Additional follow-up phone calls to groups 1 and 2 were completed 3 months after the mailing was sent. Three

Table 2. Sample Key Informant Interview Guide Questions.

Sample Questions and Follow-Up Probes

What can you tell me about the National ALS Registry?

Do you have any concerns about the National ALS Registry?

What do you think the benefits of the National ALS Registry are?

Can you describe a situation when you would hand out the patient guide?

Can you tell me about these types of conversations where you inform patients of the Registry?

What was that experience like for you when telling a patient about the Registry?

Abbreviation: ALS Registry, Amyotrophic Lateral Sclerosis Registry.

months was selected as the follow-up time period, as many providers only see a few patients with ALS a year; therefore, the time period needed to be long enough to increase the chance the neurologist saw an patient with ALS while not being too long that any impact of the intervention would not be seen. The purpose of this call was to determine whether patients with ALS had been seen during the past 3 months, whether the Registry materials were used, whether the neurologists took the free CME course, and to answer any questions about the Registry. During the phone call, researchers asked each neurologist to report whether they had or had not advised patients to self-enroll in the Registry, helped any patients enroll in the registry, provided patients with ALS Registry materials, or read the ALS Registry Provider Guide. All phone calls and faxes were tracked by provider name, and a maximum of 5 call attempts and 2 faxes were used to complete this round of phone calls.

A semistructured qualitative interview guide was used to lead the KIIs (see example questions in Table 2). The interview guide contained 3 parts: (1) questions to characterize neurologists' interactions with patients with ALS; (2) questions to determine neurologists' knowledge, attitudes,

beliefs, and practices pertaining to the Registry; and (3) questions to review and critique the Infographic, Provider Guide, and Screenshots of the enrollment process. The interview guide was pretested with 4 individuals representative of the population. The pretest was to determine whether the guide was of appropriate length and to determine whether the questions were clear and accurately captured the data as intended. The interview guide was designed to be completed in 45 to 60 minutes, as this would allow the data sought to be captured while not being a significant time burden on participants.

The KIIs were conducted in English either in person or over the telephone by the first and second authors. At the beginning of each interview, participants were provided written and verbal information about the study, asked if they had any questions or concerns to discuss, and provided verbal consent prior to commencing the KII. Researchers read an informed consent statement to participants who had been reviewed by the Quorum Review IRB. Interviews were voice-recorded without names or personal identifiers. All transcripts were identified by a study ID number rather than a name, and transcriptionists used “speaker 1” and “speaker 2” to indicate text. A small amount of descriptive information was collected for each interviewee, including sex, status as either currently diagnosing/treating (Yes) or would diagnose/treat (Would), how many patients with ALS were seen per year, type of practice (individual vs group), and if they reported cases to the ALS Surveillance Projects. Participants received a \$100 gift card for participation. Nine interviews were originally scheduled, as similar research has found 7 to 9 interviews may be sufficient to reach saturation (13).

Analysis Plan

As seen in the logic model (Figure 1), there were 3 theorized outcomes from this project: increased provider knowledge and access to materials, increased patient–provider communication regarding the Registry, and increased patient enrollment in the Registry. First, to evaluate the increase in provider knowledge and access to materials, the number of calls and faxes, the number of Registry materials mailed and faxed, and the number of TTTs and KIIs completed were tabulated. The monthly CME completion rates were reviewed to identify any increases that could be attributed to the CME flier included in the mailing. Second, it was hypothesized that this increase in access to materials would result in an increase in provider–patient Registry communication. As such, the number of providers that used Registry materials and number of neurologists that aided persons with ALS in self-enrolling were assessed. Third, ultimately, this project aimed to increase Registry self-enrollment, and thus self-enrollment counts by states and groups were compared. To analyze the change in Registry self-enrollment, rates were compared between groups 1, 2, 3, and 4 for the period October 2015 to September 2016. Monthly Registry self-

enrollment rates were calculated and then assigned a change score based on the increase or decrease from the previous month’s enrollment (eg, the difference between monthly enrollment in January 2016 and February 2016) and the change from the number enrolled in that month of the previous year (eg, the change in enrollment from January 2015 compared to January 2016). Persons with ALS self-enrollment rates in the other 44 states were monitored for comparison purposes on a monthly basis to control possible threats to internal validity of this study design. Descriptive statistics are used to describe the differences, if any, between the groups. Data cleaning and statistical analysis were performed in Microsoft Excel 2013.

To analyze the KIIs, the data were transcribed verbatim, and transcripts were reviewed for missing words and/or errors. A codebook was developed and edited throughout the coding process. This study was exploratory in nature, and codes were created both deductively and inductively. Initially, codes were deduced via the team’s understanding of the target population and the Registry. As the data were reviewed, additional codes were developed through an inductive, iterative process based on individual interviews. The transcripts were coded by 2 independent coders. The coding was compared and discrepancies were reviewed, discussed, and revised as needed. The KII transcripts were analyzed for common themes and categories across the interviewee’s responses using ATLAS.ti qualitative data management software. Coding began after the first 5 interviews were completed. Saturation was reached after the seventh interview; however, 2 additional interviews were already scheduled and thus completed. No additional themes emerged in reviewing interviews 8 and 9.

Results

Initial Phone Call to Determine Status

A total of 4562 phone calls were made to neurologists in groups 1 and 2 to determine their status. The response rate was 98%. On average, it took 1.62 calls to make this determination (1.88 in Florida; 1.78 in New Jersey; 1.51 in New York; 1.3 in Virginia). An additional 248 faxes were sent to provider offices to facilitate this process. In groups 1 and 2, 21.5% ($n = 564$) of neurologists currently diagnose/treat patients with ALS (Yes) and 14.7% ($n = 385$) would diagnose/treat a patient (Would) if they presented at their office (Table 3). The proportion of providers who do or would diagnose/treat patients with ALS was statistically significantly ($P < .05$) different in Florida (52%) compared to New Jersey (38%), New York (26%), and Virginia (36%). Of all “Yes” neurologists, 81.9% ($n = 456$) see less than 5 patients with ALS per year.

Mailing

There were 529 neurologists in group 1, 420 neurologists in group 2, and 620 neurologists in group 3 eligible to receive a

Table 3. Results of Provider Status Phone Calls and Faxes.^a

	Group 1		Group 2		Group 3		Total
	Florida	New Jersey	New York	Virginia	Ohio	Washington	
Providers on list, n (%)	828 (100)	504 (100)	1570 (100)	422 (100)	448 (100)	317 (100)	4089 (100)
Ineligible providers, n (%) ^b	134 (16)	77 (15)	392 (25)	98 (23)	100 (22)	45 (14)	846 (21)
Eligible providers, n (%)	694 (84)	427 (85)	1178 (75)	324 (77)	348 (78)	272 (86)	3243 (79)
“Yes” providers, n (%)	232 (33)	60 (14)	211 (18)	61 (19)	–	–	564 (17)
Small size practice, n (%) ^c	166 (72)	50 (83)	179 (85)	61 (100)	–	–	456 (81)
Medium size practice, n (%) ^c	66 (28)	10 (17)	32 (15)	0 (0)	–	–	108 (19)
“Would” providers, n (%)	133 (19)	104 (24)	92 (8)	56 (17)	–	–	385 (12)
“No” providers, n (%)	320 (46)	263 (62)	831 (71)	201 (62)	–	–	1615 (50)
Don't know providers, n (%) ^d	9 (1)	0 (0)	44 (4)	6 (2)	348 (100)	272 (100)	679 (21)

^aPercents have been rounded.

^bMarked ineligible if practicing at referral center, a medical resident, out-of-state, other specialty, license expired, or deceased.

^cAll Yes practices were either small (sees 0-4 patients/year) or medium (sees 5-19 patients/year).

^dMaximum call/fax attempts made to determine status or were not contacted due to group assignment.

mailed packet of information, 8 of which voluntarily opted out (Table 3). A total of 1561 packets, containing over 16 800 pieces of Registry materials, were mailed. Of them, 67 were returned to sender, 46 were successfully resent, 11 were sent to two addresses with no successful delivery, and the others were deemed to no longer be eligible for the project due to not meeting inclusion criteria (eg, moved out of state and changed locations to a referral center).

Follow-Up Phone Call #1

It took an average of 1.87 calls per office/doctor (2.2 in FL, 1.7 in NJ, 2.0 in NY, and 1.6 in VA) to complete this round of follow-up, yielding a total of 1871 phone calls. An additional 67 faxes were sent. A larger percentage of neurologists in New Jersey and New York (74% and 86%, respectively) remembered receiving the mailing compared to Florida and Virginia (68% and 71%, respectively). Faxes of the materials were sent to the 192 providers who did not remember receiving the mailing. Overall, 96% of providers confirmed receipt of the faxed materials.

Follow-Up Phone Call #2

There were 2143 calls made for an average of 2.2 attempts per completion (2.8 in Florida, 1.9 in New Jersey, 1.8 in New York, and 2.3 in Virginia). An additional 199 faxes were sent to these neurologists. Follow-up phone call 2 was completed for 90% (n = 854) neurologists. Only respondents who completed the entire call were considered participants. Of the 854 respondents who completed this follow-up call, 65.6% (n = 560) remembered receiving the mailing (Table 4). There were statistically significant differences in the percentage of respondents who remembered the mailing by state but not by group. Virginia had the largest percentage (84%) of respondents remember the mailing, while NY had the lowest (59%). Nearly half (46%) of respondents read the provider guide, with the largest proportion being in New

Jersey (63%) and the fewest in New York (28%). Thirty-one percent (n = 262) of respondents reported seeing patients with ALS in the previous 3 month. Of these providers, 60% (n = 157) reported reading the guide; 23% handed out the Guide, 15% advised a patient to self-enroll, and 1% helped a patient self-enroll (Table 4). In comparing group 1 (Florida and New Jersey) with group 2 (New York and Virginia), there were no statistically significant differences at follow-up 2 ($p > .05$), for the number of providers reported remembering the mailing, seeing patients with ALS, reading, or handing out the Guide.

Continuing Medical Education Module Enrollment

While information about the online CME module was included in the mailing, <1% of respondents indicated completing the course. There were no statistical differences between group 1 and group 2 ($P > .05$) regarding the number of neurologists who indicated completing the CME course.

Self-Enrollment Changes

Monthly self-enrollment rates did not appear to be influenced by the activities of this project. Monthly self-enrollment remained relatively constant in the 44 control states for the period October 2015 to September 2016 (average change ± 1 enrollee each month). In the group 1 and 2 states, the enrollment increased (New Jersey) or remained constant (New York, Florida, Virginia) the month after the mailing was sent. In group 3, the enrollment decreased (OH) and remained constant (WA) the month after the mailing was sent. Self-enrollment rates continued to vary 2 and 3 months after the mailing reached providers.

When comparing the monthly enrollment to the previous year, the control states vary by <1 enrollee at each monthly time point. Overall, the enrollment was slightly lower in the control group during the intervention year compared to the previous year. All 3 intervention groups had an overall

Table 4. Results of Follow-Up Phone Call #2.

	Group 1				Group 2				Train the Trainer Office Sessions ^a	Key Informant Interviews	
	Florida		New Jersey		New York		Virginia				Total
	Saw ALS Patients in Last 3 Months, n = 283 (%)	Saw ALS Patients in Last 3 Months, n = 152 (%)	Saw ALS Patients in Last 3 Months, n = 27 (%)	Overall, n = 302 (%)	Saw ALS Patients in Last 3 Months, n = 113 (%)	Overall, n = 302 (%)	Saw ALS Patients in Last 3 Months, n = 117 (%)	Overall, n = 854 (%)			
Did You ...	Overall, n = 283 (%)	Overall, n = 152 (%)	Overall, n = 27 (%)	Overall, n = 302 (%)	Overall, n = 113 (%)	Overall, n = 302 (%)	Overall, n = 117 (%)	Overall, n = 854 (%)	Overall, n = 262 (%)	Overall, n = 28 ^a	Overall, n = 6
Remember the mailing?	171 (60)	113 (74)	24 (89)	178 (59)	93 (82)	178 (59)	98 (84)	560 (66)	208 (79)	24 (86)	6 (100)
Read the provider guide?	147 (52)	96 (63)	24 (89)	84 (28)	55 (49)	84 (28)	68 (58)	395 (46)	157 (60)	23 (83)	6 (100)
Hang the poster?	2 (1)	0 (0)	0 (0)	3 (1)	3 (3)	3 (1)	0 (0)	5 (1)	5 (2)	0 (0)	0 (0)
Take CME?	1 (1)	0 (0)	0 (0)	2 (1)	2 (2)	2 (1)	0 (0)	3 (1)	3 (1)	0 (0)	0 (0)
Hand out patient guide?	21 (7)	9 (6)	9 (33)	17 (6)	17 (15)	17 (6)	12 (10)	59 (7)	59 (23)	6 (19)	1 (21)
Order additional materials?	1 (1)	0 (0)	0 (0)	3 (1)	2 (2)	3 (1)	1 (1)	5 (1)	4 (2)	1 (3)	0 (0)
Advise anyone to self-enroll?	8 (3)	16 (11)	16 (60)	2 (1)	2 (2)	2 (1)	13 (11)	39 (5)	39 (15)	7 (22)	1 (21)
Help anyone self-enroll?	0 (0)	0 (0)	0 (0)	2 (1)	2 (2)	2 (1)	1 (1)	3 (1)	3 (1)	0 (0)	0 (0)

Abbreviations: ALS Registry, Amyotrophic Lateral Sclerosis Registry; TTT, Train the Trainer.

^aThis count represents the offices that completed the TTT session and also answered follow-up call 2.

lower enrollment in the intervention year compared to the previous year.

Train the Trainer Sessions

A total of 22 neurologists and 37 staff members from New Jersey and Florida participated in the TTT sessions, and 26 additional neurologists were invited and declined participation. As shown in Table 4, of the providers who participated in the TTT sessions and completed follow-up phone call, 2 most remembered receiving the mailing (86%) and read the provider guide (83%). Nearly one-quarter (21%) of train the trainer participants advised patients to self-enroll.

Key Informant Interviews

A total of 72 neurologists were contacted, and 9 agreed to participate as interviewees. At the time of recruitment, 7 participants were currently diagnosing patients with ALS, and 2 would diagnose/treat a patient with ALS if one presented at their offices. Six were males and 3 were females. Interviewees noted they diagnose patient with ALS and conduct follow-up appointments with these patients in their offices, see patients when they round at the hospital, send patients for second opinions at Universities and/or referral centers, and/or they send patients to referral centers for follow-up care. Overall respondents indicated that following the interview, they were likely to recommend patients to self-enroll in the Registry but previously were unaware of the Registry or did not have a clear understanding of the Registry's objectives.

Only 2 respondents had heard about the Registry prior to their interview. Respondents had a vague understanding that patients are encouraged to enroll through a website and that the Registry is trying to better estimate the prevalence of ALS in the United States. No respondents remembered seeing the Registry booth at scientific meetings (eg, American Academy of Neurology Annual Meeting), and 2 recalled receiving a packet of information in the mail prior to receiving the information sent with specific instructions about their interview. They had not handed out the patient guide, although 1 respondent noted creating and handing out ALS-related materials from a different source. Respondents did not know how the Registry collects information about patients.

When asked to elaborate on benefits to their patients, respondents described several benefits and described why patients might feel compelled to participate. Respondents confirmed the Registry can link patients to research, "... [it] provide[s] some clinical trials information, which is what most patients ask these questions," and that by making this connection, it "may give them a sense of participation... It may not necessarily help a particular patient at a particular time, but on the other hand, accumulating information can be beneficial long term basis." Further, respondent thought the Registry provides patients the opportunity to play an active

role in their disease, despite whether or not their contribution to the Registry will cure their disease,

I think they feel that they're playing a more active role rather than a passive role, trying to see if changes can be made in the disease progression and more things can be found out from the disease by the ALS registry, hopefully, coming to some possible better treatment options besides Rilutek.

Respondents pointed out that, "... they have the attitude [that] if it's not going to work for them, it might work for someone else and they're willing to take part."

All respondents said they would have referred confirmed patients with ALS to the Registry if they had known about it. Further, given this newly acquired knowledge, all respondents said they would hand out the patient guide to patients with a confirmed ALS diagnosis, "Once I've explained to them the diagnosis there is absolutely no reason not to." Similarly noted, "I think I would always make it available to patients. It'd be up to them if they didn't want to become part of the Registry." However, one respondent cautioned,

"Well..., practices like ours, we don't see ALS patients on daily basis. Maybe like, once a year or once, once every 2 years, you know, we'll get a real ALS patient so, sometimes I wonder whether we are going to forget about it... So unless we get some, like some sort of periodic reminders..."

Respondents thought promoting the benefits of the Registry might be an effective way to engage their patients and to get them motivated to self-enroll. One respondent said,

I would say, well you know the by doing this is important so that we know exactly where who patients are, where the patients are, and then when the trials come up we know where to locate the patients. And eventually, sort of promise for them some trials may lead to some treatment during your disease process and, more likely it'll help for other patients in the future.

One respondent said they have doctors and nurses who could help a patient self-enroll if the patient decided that is what they wanted to do.

We have some nurses in offices that are helping the patients. But, it's really up to the patient to make the decision to self-enroll, and if they need help, either a caregiver or a doctor, that's when one of us could step in and give them some help.

Discussion and Conclusions

Promotion of the Registry has primarily consisted of targeted outreach to clinicians at scientific conferences and the distribution of materials to persons with ALS at promotional events, support groups, patient forums, and health-care encounters at major ALS referral centers typically located in large metropolitan areas. A targeted approach to general, nonreferral center neurology practices located outside these

metropolitan areas and general neurologists was lacking. This study is the first of its kind to promote the Registry to general neurologists, in a large, geographically diverse area.

Discussion

It appears that sending a targeted mailing to this group was an effective method to provide information about the Registry; however, the retention and later dissemination of this information may be lacking. Most neurologists received the mailing and were able to confirm receipt of the package 1 week after it was sent. Additionally, 3 months later, most practices still remembered receiving the packet of information. Yet, the majority of neurologists did not report reading or using the materials in their practice. This may be due to the relevancy of the topic for these providers, the interest providers have in the topic, or the way the information is presented in the materials. Of those who had an opportunity to use the materials, that is, saw a patient with ALS, a majority did read the patient guide. However, of those providers that saw patients with ALS during the 3-month follow-up period, most did not advise or assist patients with the self-enrollment process. It appears as though general neurologists can serve as an information liaison to patients, but they may not be best suited to dedicate time and resources to facilitate the self-enrollment process, perhaps due to the infrequency of seeing patients with ALS, limited time with each patient, or lack of resources available (eg, dedicated computers in the waiting room) as noted in the KII. Perhaps more regular follow-up and engagement with these providers will aid in reminding them of the Registry. Once the neurologist had a clear understanding of the Registry, they reported being supportive of it and willing to inform patients that it exists.

The KII participants indicated in the future they would refer confirmed patients with ALS to the Registry; however, in the overall study group, most providers did not do so. It appears as though the materials alone do not provide sufficient engagement to providers to take action. This discrepancy between KII participants all indicating they would refer patients and most providers who received the mailings not doing so may be due to the need for elaboration on the materials or further cues to action such as an in-person conversation.

Although the overall rates of self-enrollment did not significantly increase during this intervention, it is possible that rates of registry self-enrollment at nonreferral centers changed. The data currently collected by the National ALS Registry does not capture the source of the enrollment or where the patient learned about the Registry. It is certainly possible that the rate of self-enrollment among patients seen at nonreferral centers changed during our study period, but being that the proportion of patients this represents is relatively small, it may not have led to an increase in the overall self-enrollment rate. Future studies would benefit from the knowledge of the patient referral source to determine where interventions should be focused.

There were differences detected by state, but it is unclear as to why. It could be that gatekeeper staff members in 1 state were more likely to intercept the mailing on behalf of the provider, and the provider may not have even opened the packet of materials themselves. Further research may be warranted to better understand these office processes and to determine how best to get information about the Registry to the neurologists in ways they are most likely going to receive and review it. While this study was successful in providing the information to neurologists, successful utilization of the materials was limited. As literature indicates, a change in knowledge does not lead to a behavior change, as such provider receipt and reading of the materials alone may not affect enrollment rates. As discussed by Woolf (14), knowledge is the first step in changing a physician's behavior but should be followed by attitude changes and reminders. It is possible a mixed approach, including traditional postal mail, in conjunction with mass e-mailing messages, and on-site delivery of materials may increase the likelihood of doctors receiving and reading the information. We found that providers who engaged face-to-face with a research team member were likely to have read and use the messaging as well as further acting on the materials and advising patients to self-enroll. As such, information dissemination should be done in person at the clinicians' office over a webinar or at an on-site meeting or conference.

Overall, small numbers of respondents reported handing out the patient guide and advising patients to self-enroll. This may be influenced by the fact that most neurologists reported that they see patients with ALS sporadically. As uncovered in the KII interviews, these general neurologists may or may not keep Registry materials at their disposal, and if they do, they may forget the materials exist because ALS patients are not seen in their offices on a weekly basis. This is in comparison with ALS specialist neurologists who may see patients with ALS on a weekly basis. While effective and informed patient-provider communication is particularly important in rare diseases such as ALS, general neurologists, practicing at nonreferral centers, may not be experts in the care of ALS due to various reasons, including a paucity of health-care interactions with these patients (15). Unfortunately, prior to this work, it was unclear whether nonreferral center neurologists were aware, knowledgeable, and supportive of the Registry. It is possible that other health-care providers and staff, such as community health workers and health educators, would be better suited to deliver the materials and assist with self-enrollment.

Key Informant Interview session participants who were not lost to follow-up, all reported reading the provider guide and remembering the mailing. While the number of participants this represents is not large enough to determine a statistical significance, this suggests the importance of in-person follow-up to increase the use of provided materials. Additionally, the rate of remembering the mailing and reading the provider guide was higher in the TTT participant group compared to the overall population groups again

suggesting the importance of in-person follow-up. Perhaps having an in-person follow-up scheduled tipped the provider to be engaged in the materials more so than seeing a package of materials arrive in the mail. One element to increase success of education and outreach is to use reminders (cues to action) (16). The key cues used in this project were the mailing including the Registry poster, CME announcement, and patient guide in the mailing, phone calls at 1-week post-mailing to encourage neurologists to read and use the materials and conducting TTTs and KIIs. While the data trends indicated a face-to-face intervention (ie, TTT and/or KII) may be a large enough cue to action, as those providers were more likely to have spoken to a patient about the registry, it is not feasible to conduct such visits with all providers, as they are quite costly and time consuming. It is possible that sending periodic reminders as cues to action to the “Yes” and “Would” neurologists might be a good strategy (17). To accomplish this, lists of neurologists would need constant review and refinement, and costly phone calls would need to be made to determine which neurologists do or do not see cases with ALS. Ongoing replication of this process on a nationwide scale to cultivate lists of “Yes” and “Would” doctors may not be worth the time, effort, and money, particularly given the scarcity of these neurologists interaction with persons with ALS; only a third of neurologists who do or would treat patients with ALS reported interacting with patients with ALS within a 3-month window of time. To this end, it may be more cost effective to procure, verify, and expand mailing lists of all neurologists, regardless of their status, on an on-going basis, as was done with group 3. Mass mailings could then be sent to all neurologists within a defined geographic area based on self-enrollment performance or other factors of interest. Being that even neurologists who do care for people with ALS may not see those patients on a regular basis, it might be as effective to mail to all providers, rather than target those who do or may see patients with ALS.

Another issue to consider is what method to use to remind neurologists of the Registry. Sending out blast e-mails, mailings of postcards or brochures via postal mail or express mail, or faxes should be considered. Additionally, a dedicated section for neurologists and researchers will be deployed on the Registry website, which will allow for e-mail communications to be used with those who have registered with the website. Research has demonstrated that use of multiple methods of outreach and promotion with health-care providers increases success of changing provider knowledge and behavior (18). To this end, we created a multicomponent promotion and outreach project and expected states, where more methods were used to outperform their counterparts in patient self-enrollment. Unfortunately, self-enrollment did not differ by group during our project time period. This could be because only about one-third of “Yes and “Would” neurologists saw patients with ALS in the 3-month period following the mass mailing. Perhaps if more time had passed and if these neurologists

had seen more patients, they could have promoted the Registry, which could have led to increases in the number of self-enrollees in the intervention states.

To further complicate the determination of whether our project components led to any changes in self-enrollment were the competing marketing and promotion activities simultaneously occurring. The project team requested and reviewed reports of partner, such as ALSA and MDA, activities. It is hard to pinpoint which promotional activities completed on whose behalf may have triggered a person with ALS to self-enroll in the Registry. Even if all neurologists who reported seeing patients in our follow-up period had assisted a patient self-enroll in the Registry, it would not explain the fluctuations shown in the state level enrollment data. Clearly, the results of this study show that self-enrollment rates did not change dramatically over the course of this project. This may be due in part to the fact that the neurologists in our groups did not see the critical mass of persons with ALS during the 3-month follow-up period that would translate to an increase in self-enrollment rates, or information to a provider is not impactful enough to increase patient behavior in the context of self-enrolling in the ALS Registry.

Most respondents had never heard of the Registry, despite having been sent materials as part of the greater Registry Promotion project. Careful consideration must be given to further promote the Registry to neurologists on an ongoing basis. As pointed out by 1 respondent, previously quoted, due to the small number of annual patient with ALS encounters, provider may not remember to promote the Registry due to an overabundance of other information, diseases, and patient needs. Respondents were genuinely interested in the Registry and willing to promote it to their patients, perhaps regular e-mailed reminders would serve beneficial to general neurologists.

Limitations. The goal of this study was to conduct educational and promotional outreach activities to general neurologists. Through our KIIs, it became clear that, in some settings, nurses may have a greater ability to build rapport with patients and have more time to spend with them. Perhaps the study results would have differed if not only general neurologists but also nurses at each practice were provided the Registry educational materials and asked to present them to eligible patients.

Conclusion

Registry self-enrollment is vital for identifying new and prevalent cases with ALS. Only self-enrolled persons with ALS are eligible to complete the risk factor surveys and to be eligible for notification about research projects such as the National ALS Biorepository. Promoting the Registry via the patient-provider communication channel seems ideal in theory, and while the information was successfully provided to the neurologists, it was difficult to encourage provider

utilization of the materials. Future promotion campaigns should consider novel approaches including cues to action among neurologists and other health-care providers with pretested materials that are relevant and interesting, which may lead to an improved likelihood of Registry information uptake and engagement with patients.

Practice Implications

This project has demonstrated that continued marketing, promotion, and educational initiatives to neurologists and office staff are important activities. At minimum, general neurologists serve as an informational resource clearinghouse for patients with ALS. However, these providers have competing priorities, and they see patients with ALS infrequently. The current project attempted to promote the Registry to providers in direct contact with persons with ALS. While this study focuses on one health issue, we believe the lessons learned from this project can be incorporated into future work for those working in areas of other rare conditions/diseases or those attempting to engage busy specialists in other research projects.

While a large proportion of providers did not help their patients with self-enrollment, they distributed the materials that were provided to them. Comprehensive interactions with providers (ie, KII and TTT) increased the likelihood of message retention. While ALS referral centers continue to be the main focus for outreach endeavors, due to the frequency and quantity of patients seen there, general neurology practices may be the only touchpoint for a subgroup of patients. Therefore, it would be a disservice to ignore this group of neurologists. The results of this study will assist ATSDR in future endeavors to promote the National ALS Registry and can provide a model for others attempting to increase enrollment in nonmandated registries.

Authors' Note

The findings and conclusions presented in this article are those of the authors and do not necessarily represent the views of the Agency for Toxic Substances and Disease Registry, the Centers for Disease Control and Prevention, or the United States Department of Health and Human Services.

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