



Assessing the state of care for Huntington disease in the United States: Results from a survey of practices treating Huntington disease patients

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ABSTRACT

Background: No study to date has thoroughly examined US Huntington disease (HD) care delivery in a variety of clinic settings by HD specialists and non-specialists.

Objective: To obtain a clearer understanding of current care structure and delivery of care through a survey of representative US physicians treating HD patients.

Methods: We designed and fielded a survey of 40 closed-ended evaluative items and one open-ended item to a sample of 339 US practices. Unique to this survey was the inclusion of non-specialists.

Results: Responses were received from 156 practices (overall response rate 46.02%), with 52.6% from academic sites, 35.3% from private practices, and 12.2% from the VA. More than half (63.5%) of the practice leads were movement disorder trained or Directors of HD Centers of Excellence and 58.3% had an HD or multidisciplinary care clinic. However, 48.7% of the practices saw 1–25 HD patients, 28.2% saw 26–100 HD patients, and 23.1% served over 100 HD patients annually. Most practices (>69%) reported having difficulty providing social work, genetic counseling, care coordination and psychologists/psychiatrists. Increased HD practice size was associated with higher rates of pre-visit screenings, care navigator/care coordinators, routine monitoring of weight, and provision of genetic counseling by genetic counselors.

Conclusions: Not surprisingly, we found that HD care was inconsistently applied across the US. Practices led by neurologists trained in movement disorders, and higher HD volume practices, tended to be better equipped to provide multi-disciplinary staffing and procedures as compared to those with fewer numbers of HD patients.

1. Introduction

Huntington disease (HD) is a rare, autosomal dominant, disorder caused by a mutation in the *HTT* gene. While disease manifestations vary, there are core clinical features that characterize the disease including mood changes, cognitive decline, chorea and other disturbances of movement. Great breakthroughs have been made in understanding the molecular genetics of HD since 1993, however, there remain gaps in our approach to diagnosis and treatment. Clinicians who treat HD patients have differing levels of expertise in the management and treatment of disease as supported by a recent study of HD families

who felt that an increase in physician training and awareness were necessary to improve their overall care [1]. Furthermore, HD expertise may vary across practices, meaning that measures of progression and quality care may not be consistently applied. An international survey of HD specialty clinics in 2016 found that most practices provided multi-disciplinary care and saw >50 HD patients, annually [2]. Even so, the authors noted variability in provision of services and recommended more research to understand centers' organization and care delivery, coordination with local health care systems, and the way in which these features might impact quality of care.

HD-Net, formed in 2019 and supported by the Huntington Study

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Group, is an effort to better understand the state of HD care across the US so that we may identify gaps and strengthen HD care and access. No study to date has thoroughly examined US HD care delivery in a variety of clinic settings by HD specialists and non-specialists. The goal of this HD-Net study was to obtain a clearer understanding of current structure and delivery of care to HD persons through a survey of representative US physicians caring for those with HD.

2. Methods

We designed and fielded a survey to assess the current state of HD care in the United States.

2.1. Survey development

An initial survey draft was developed by the HD-Net team, informed by prior HD research studies and surveys [1,2], and supported by ten qualitative interviews conducted with selected HD providers representing various backgrounds, practice types and regions. We further improved the survey with feedback from our team and the HD-Net Steering Committee, which provided comment and oversight of the survey content.

The final survey instrument contained 40 closed-ended evaluative items and one open-ended item. (Appendix A).

2.2. Sample selection

We obtained claims data via the Symphony Health (SHA) database, a large, nationally representative data source which covers about 280 million lives annually and includes claims submitted to all payer types, including commercial plans, Medicare Part D, cash, assistance programs, and Medicaid. This data identified 3,478 U.S. physicians who had billed for HD care during 2018/19 (time frame of the database access). Practices that billed for fewer than 10 HD patients per year were removed, resulting in an initial sample file of 449 physician practices.

2.3. Practice classifications

We used three different approaches to classify physician practices in the sample. First, lead physicians within each practice were stratified into 5 mutually exclusive categories, based on the highest level of HD expertise of their practice lead:

A - Neurologist, at a university-based practice who is movement disorder (MD) trained and/or Director of a Huntington Study Group (HSG) research site or Huntington's Disease Society of America (HDSA) Center of Excellence.

B - Neurologist who is fellowship trained in movement disorders, but the practice does not meet the criteria of the "A" practices.

C - Neurologist who treats HD patients, but is not movement disorder-trained.

D - Clinical practitioner who treats HD patients but is not a neurologist. Provider may be a psychiatrist, primary care provider, physical therapist, or some other specialty.

E- Veterans Affairs Movement Disorder Center Director.

After reviewing initial response patterns, we combined several practice characteristics to identify the most meaningful comparisons of interest for our statistical analyses. Specifically, we merged the categories describing lead physician expertise into a dichotomous variable of either movement disorder trained or not. Second, practices were categorized as academic, non-academic or Veterans Affairs (VA). And third, we consolidated responses to two separate survey items into a single dichotomous variable indicating presence of an HD-only or multidisciplinary clinic vs absence of either of these clinics in the practice. We had initially separated out the VA (category E) from the others because it is a unique model of care which incorporates comprehensive services in a single payor system, with a defined patient population.

2.4. Survey Administration

Verification phone calls were made to each practice to assure correct contact information and identify the provider seeing HD patients at the practice. Removal of duplicates resulted in 421 practices that were sent surveys by multiple methods, including email, and/or mail (FedEx) with reminder emails, mail and phone calls. The participants were provided a modest monetary incentive for their participation.

2.5. Statistical analyses

First, descriptive analyses characterized the sample in terms of HD practice characteristics and summarized the responses to survey items measuring the current state of HD care. Most respondents (96 %) completed at least 80 % of the questions for which they were eligible. Missingness rates on individual survey items were minimal, ranging from 1 to 11 %. Thus, missing data was handled using a single data imputation via the proc MI (multiple imputation) command in SAS.

Second, we used principal components analyses (PCA) to identify meaningful groupings among key survey items capturing the state of HD care. Because endorsement was low on some survey items and we were limited by the number of items to include in the PCA, we consolidated some survey response categories to obtain a total of 15 items for use in our PCA. Figure B.1 in the Appendix provides further details on how we created the final 15 items to be used in the PCA. The PCA identified six latent factors underlying our 15 survey items and these six factors were used as outcomes in multivariable regression models to examine whether key practice characteristics were associated with differing states of HD care, even after multivariate adjustment.

Prior to fitting our final regression models, we assessed the bivariate relationships between HD practice characteristics and the 15 items entered into the PCA. We computed effect size (ES) differences between the different HD practice groups on each survey item (e.g., Cohen's d) to guide our selection of what might be considered meaningful bivariate relationships between HD practice characteristics and the 15 survey items. We identified moderate ES differences between 0.4 and 0.7 and large ES differences as those over 0.7. We relied on ES differences using Cohen's d rather than p-values from traditional t-tests to determine notable findings because our goal was to better understand the estimated size of the bivariate relationships, rather than identifying statistically significant differences between the groups. By reporting ES differences rather than p-values, we also avoided any issues related to multiple testing concerns.

We estimated multivariate regression models for each of the six latent variables identified in the PCA as we sought to understand how the current state of HD care varies as a function of practice characteristics. Because of our limited overall sample size relative to the number of comparisons under consideration, we removed two predictor variables before entering into the multivariable regression model: site involvement in HD research and US Census region. These decisions were made based on overlap with other practice characteristics, and the ability to affect change based on practice characteristics (e.g. a practice cannot easily change its Census region). The outcomes (dependent variables) were computed using results from the PCA (see Appendix B for details) and separate models were fit to each controlling jointly for practice setting, expertise of practice lead, HD practice size as measured by HD patients seen annually, and availability of multidisciplinary/HD clinic. As in our bivariate analyses, we standardized the current state of HD care scores to allow us to interpret the regression coefficients as ES and used criteria to determine when associations are moderate or large (0.4, and 0.7, respectively). Statistical analyses were implemented using SAS 9.4.

After reviewing unadjusted differences by key HD practice characteristics, there were stark differences between the VA system and other healthcare systems. Because VA practices are structurally and financially unique (e.g. separate structure and payment mechanism

compared with health care writ large), and as many of our outcome variables relate to practice structures supporting HD patients, we decided not to include VA practices in the PCA. Thus, we showcase the differences between the VA and other practice settings as part of our bivariate analyses but did not enter VA practices into the PCA and final multivariate outcome models.

3. Results

We fielded our survey to 421 clinical practices over a 13 week survey period ending November 2020. During the fielding period, 82 were identified as ineligible for the survey because they: 1) represented duplicate practices that had not previously been identified (n = 12); 2) reported that they had not treated an HD patient within the past year (n = 59); 3) provided inpatient care only with no outpatient continuity or the practice had closed (n = 11). This brought the final eligible sample to 339.

We received completed surveys from 156 practices, a 46.02 % overall response rate with the following notable differences by HD expertise of the lead physician: Group A = 80.00 % (n = 68); Group B = 43.18 % (n = 19); Group C = 30.60 % (n = 41); Group D = 34.62 % (n = 9); Group E = 38.00 % (n = 19). Approximately 72 % of responses were received via the web mode and 28 % via mail.

Of the 156 responses, 52.6 % were from academic practices, 35.3 % from private practices, and 12.1 % from the VA (Table 1). More than half (63.5 %) of the practice leads were movement disorder trained or Directors of HDSA Centers of Excellence (ratings A or B) and 58.3 % reported that they had an HD or multidisciplinary care clinic for patients with HD. HD practice size varied, with 48.7 % of the practices seeing 1–25 HD patients, 28.2 % seeing 26–100 HD patients, and 23.1 % serving over 100 HD patients, annually. 41.7 % of the practices reported active involvement in HD research. Finally, practices were evenly spread across the U.S.

With regards to practices' use of telehealth, it is notable that approximately 70 % of practices did not use telehealth pre-COVID, although the VA used more telehealth than other practices. In contrast, 99 % of practices reported using telehealth during the COVID-19 pandemic. Half of practices reported a "significant" increase in the number of telehealth visits as a result of COVID-19. The most striking expansion in use of telehealth was seen among medical and psychiatric practitioners where visits escalated from 24 to 90 %, and also social workers where visits rose from 8 to 41 %. However, genetic counselors, care coordinators, and physical therapists also reported substantial increases in use of telehealth during the COVID-19 pandemic.

Table 2 highlights findings from our bivariate analysis examining unadjusted differences between key HD practice characteristics and the

15 variables that entered into the PCA. Overall, we identified high rates of endorsement on items related to monitoring disease progression (at least 68 % across practices for weight, activities of daily living and behavioral health, at least 81 % for monitoring motor function, and between 62 and 77 % for cognitive functioning). It also appeared that the large majority of practices (>79 %) were seeing HD patients as often as deemed necessary. Most practices (>69 %) reported having difficulty providing social work, genetic counseling, care coordination or psychologists/psychiatrists. It was relatively less common for practices to report measuring patient and family experience as a care outcome (9–36 %) or being able to seamlessly provide future intravenous and intrathecal therapies (13–25 % of practices). It was also generally uncommon (<50 %) to engage in pre-visit screenings by phone, except for the largest HD practices where 72 % of practices reported conducting such screenings. In terms of bivariate relationships, we highlight cases where the ES relationship between practice characteristics and a survey item were >0.7 (a large effect size difference). Specifically, we found three notable findings for practice setting: first, academic practices had the highest rates of providing genetic counseling with genetic counselors (67 %) followed next by the VA (47 %) and last by non-academic practices (only 25 %). Second, prior to Covid-19, the VA offered telehealth visits at a much higher rate of 63 % compared to academic (35 %) and non-academic practices (11 %). Finally, the VA had the lowest percent of prescription medications, treatments and tests that required insurance follow-up (22 %) compared with academic (39 %) and non-academic (46 %) practices. For site involvement in HD research, the greatest differences in responses were seen in whether or not practices conducted a pre-visit screening, with practices involved in HD research more likely to conduct pre-visit screenings compared with practices not involved in HD research (51 % versus 18 %). For expertise of practice leads, the most notable differences (ES > 0.7) highlight that practices with MD trained leads are more likely to provide genetic counseling by genetic counselors than practices without MD trained leads (64 % versus 26 %, respectively) and more likely to monitor weight (96 % versus 68 %, respectively).

Finally, increasing HD practice size was associated with higher rates of conducting a pre-visit screening, a practice having a care navigator/care coordinator, genetic counseling provided by genetic counselors and a practice routinely monitoring weight. Finally, there were two notable differences based on presence or absence of an HD or multidisciplinary clinic. Practices with an HD or multidisciplinary clinic were more likely to report conducting pre-visit screenings (49 % vs 6 %) and to provide genetic counseling with genetic counselors (65 % vs 29 %).

Our PCA showed that variations in our 15 observed variables mainly reflected variations in six latent (underlying) variables. Practices scoring highly on Factor 1, capturing sufficient HD specialized staffing, were more likely to have a care navigator or care coordinator, provide genetic counseling by genetic counselors and to have had any telehealth in place prior to the COVID-19 pandemic. Practices scoring highly on Factor 2, capturing the provision of routine HD care, were more likely to routinely monitor weight, behavioral health and motor function and to report that HD patients were able to visit the practice as often as needed. Practices scoring highly on Factor 3, capturing the ability to provide pre-visit screening and staff such as social workers, were more likely to conduct a pre-visit screening and less likely to have difficulty providing social work, genetic counseling, care coordination or psychologists/psychiatrists. Practices scoring highly on Factor 4 were more likely to routinely monitor cognitive function. Practices scoring highly on Factor 5, capturing access to complex, novel therapeutic interventions, were more likely to report being able to seamlessly incorporate new intravenous and intrathecal therapies into their practice and to report a lower percentage of prescriptions, treatments and tests that need insurance follow up. Practices scoring highly on Factor 6, capturing the provision of patient-centered care, were more likely to report routine monitoring activities of daily living and patient and family experiences, and to report a longer average wait time for new HD patients.

Table 1
Characteristics of the survey sample (N = 156).

Practice Setting N (%)	
Academic	82 (52.6)
Non-academic	55 (35.3)
Veterans Administration	19 (12.2)
Site involvement with HD Research N (%)	65 (41.7)
Expertise of practice lead N (%)	
Movement disorder trained	99 (63.5)
Not Movement Disorder Trained	57 (36.5)
HD patients seen annually N (%)	
1–25 HD patients	76 (48.7)
26–100 HD patients	44 (28.2)
>100 HD patients	36 (23.1)
HD or multidisciplinary clinic N (%)	91 (58.3)
Census Region N (%)	
Midwest	39 (25.0)
Northeast	34 (21.8)
South	52 (33.3)
West	31 (19.9)

Table 2

Bivariate analysis effect sizes (ES) for key Huntington disease practice characteristics and variables entered into the Principal Component Analysis (PCA).

	Practice Setting [□]			Site Involved with HD Research		Expertise of Practice Lead [□]		HD Patients Seen Annually			HD or multi-discip clinic		Census Region [□]			
	Acad	Non Acad	VA	Yes	No	MD train	No MD train	1–25	26–100	>100	Yes	No	MW	NE	S	W
N	82	55	19	65	91	99	57	76	44	36	91	65	39	34	52	31
Practice conducts a pre-visit screening to discuss symptoms, health history, and care needs prior to first visit	0.41	0.22	0.16*	0.51	0.18**	0.4	0.16*	0.11	0.34	0.72**	0.49	0.06**	0.28	0.29	0.37	0.29
Practice has care navigator/care coordinator	0.77	0.51	0.68*	0.78	0.58*	0.76	0.51*	0.53	0.73	0.89**	0.78	0.51*	0.67	0.59	0.6	0.87*
Genetic counseling provided by genetic counselors	0.67	0.25	0.47**	0.69	0.36*	0.64	0.26**	0.32	0.52	0.86**	0.65	0.29**	0.51	0.56	0.48	0.45
Practice routinely monitors weight	0.91	0.76	0.89*	0.94	0.8*	0.96	0.68**	0.78	0.89	1**	0.92	0.77*	0.82	0.94	0.77	0.97*
Practice routinely monitors activities of daily living	0.93	0.84	0.79	0.92	0.85	0.91	0.82	0.83	0.95	0.89*	0.92	0.82	0.85	0.79	0.94	0.9*
Practice routinely monitors behavioral health	0.89	0.76	0.74	0.94	0.75*	0.89	0.72*	0.72	0.93	0.92*	0.91	0.71*	0.82	0.76	0.83	0.9
Practice routinely monitors cognitive function	0.7	0.67	0.63	0.62	0.73	0.69	0.67	0.66	0.64	0.78	0.73	0.62	0.77	0.5	0.67	0.77*
Practice routinely monitors motor function	0.96	0.84	0.89*	0.95	0.88	0.97	0.81*	0.84	0.98	0.97*	0.95	0.86	0.9	0.85	0.9	1*
Practice routinely monitors patient/family experience	0.27	0.16	0.11*	0.25	0.19	0.26	0.12	0.12	0.25	0.36*	0.27	0.12	0.28	0.09	0.27	0.16*
Practice had any telehealth prior to COVID-19	0.35	0.11	0.63**	0.32	0.29	0.39	0.14*	0.26	0.27	0.42	0.35	0.23	0.26	0.32	0.25	0.42
Average wait time for new HD patients (weeks)	6.24	7.69	5.11	6.54	6.67	6.49	6.82	7.53	6.55	4.78*	6.1	7.34	7.15	5.15	7.67	5.77
HD patients visit the practice as often as needed	0.85	0.89	0.84	0.89	0.85	0.87	0.86	0.82	0.91	0.92	0.9	0.82	0.87	0.79	0.88	0.9
Future intravenous and intrathecal therapies would fit seamlessly into current practice	0.13	0.22	0.21	0.18	0.16	0.16	0.19	0.14	0.25	0.14	0.21	0.12	0.23	0.15	0.17	0.13
Percent of prescription medications,	39.02	46.09	22.37**	33.46	43.79	34.39	48.33*	44.61	32.73	36.9	35.11	45.62	36.54	35.59	42.5	42.42

(continued on next page)

Table 2 (continued)

	Practice Setting [□]			Site Involved with HD Research		Expertise of Practice Lead [□]		HD Patients Seen Annually			HD or multi-discip clinic		Census Region [□]			
	Acad	Non Acad	VA	Yes	No	MD train	No MD train	1-25	26-100	>100	Yes	No	MW	NE	S	W
treatments and tests that require insurance follow up	0.77	0.84	0.79	0.8	0.79	0.8	0.79	0.84	0.8	0.69	0.73	0.89*	0.72	0.79	0.83	0.84
Practice has trouble providing social work, genetic counseling, care coordination, or psychologists/psychiatrists																

*Maximum effect size difference > 0.4, which denotes moderate sized differences.

**Maximum effect size difference > 0.7, which denote large differences.

□MW = Midwest; NE = Northeast; S = South; W = West.

□Acad = Academic; Non Acad = Non-academic; VA = Veterans Administration.

□MD train = Movement disorder trained; No MD train = Non movement disorder trained.

Fig. 1 shows the overall mean level of endorsement of the survey items used in our PCA that aimed to capture the current state of HD care in our sample. The colors in the bar chart indicate how the 15 variables clustered into 6 latent variables based on their highest factor loadings.

However, we note that all 15 items were included in the final calculation for a given factor with weights proportional to their factor loadings. Further details of the PCA results can be found in Appendix B.

Table 3 shows the final multivariate regression model results for our six outcomes. After multivariate adjustment, several notable findings remained. For factor 1, sufficient HD specialized staffing, we found a moderately sized positive association between higher ratings on this scale and practices seeing > 100 HD patients annually (effect size [ES] association = 0.64; 95 % CI = 0.15, 1.13), and moderately sized negative associations with lower ratings on this scale with non-academic practices (ES = -0.47; 95 % CI = -0.84, -0.09) and practices without a

movement disorder trained lead physician (ES = -0.45 95 % CI = -0.85, -0.04). For factor 2, provision of routine HD care, there was a moderate positive trend towards higher ratings on this scale and practices seeing 26-100 HD patients annually (ES = 0.40 95 % CI = -0.06, 0.86) and practices seeing >100 HD patients annually (ES = 0.51 95 % CI = -0.03, 1.06). There was also a moderately sized negative trend for this scale for practices without a movement disorder trained lead (ES = -0.45 95 % CI = -0.90, 0) having lower ratings. For factor 3, the ability to provide pre-visit screening and staff such as SW, there was a moderately strong positive association between higher ratings on this scale and practices seeing > 100 HD patients annually (ES = 0.59 95 % CI = 0.05, 1.13) and a negative association with higher ratings with practices without an HD or multidisciplinary clinic (ES = -0.45 95 % CI = -0.89, -0.01). For factor 5, access to complex, novel therapeutic interventions, there was a potential small negative trend for this scale and practices without an HD

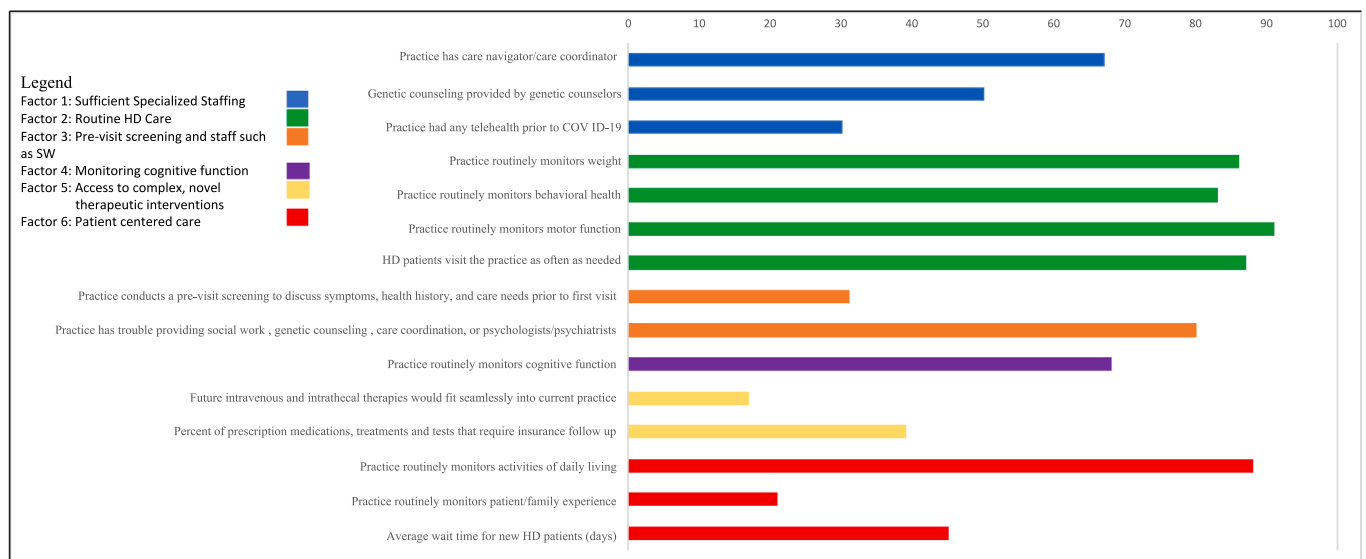


Fig. 1. Mean level of response for each survey item along with information on how the survey items grouped into six latent variables based on their highest factor loading in the Principal Component Analysis.

Table 3

Regression results examining the association between Huntington disease (HD) practice characteristics and our key state of care factors.

	Sufficient specialized Staffing	Provision of routine HD care	Provision of PVS and staff such as SW	Monitoring cognitive function	Access to novel, complex therapeutic interventions	Provision of patient-centered care
r^2	0.30	0.15	0.16	0.01	0.07	0.02
Parameter	Coefficient (95 % CI)	Coefficient (95 % CI)	Coefficient (95 % CI)	Coefficient (95 % CI)	Coefficient (95 % CI)	Coefficient (95 % CI)
Intercept	0.10 (−0.32, 0.52)	−0.11 (−0.58, 0.35)	−0.02 (−0.48, 0.44)	0.13 (−0.37, 0.63)	−0.09 (−0.58, 0.39)	0.19 (−0.31, 0.69)
Practice Size						
1–25 HD patients	reference group	reference group	reference group	reference group	reference group	reference group
26–100 HD patients	0.17 (−0.25, 0.29)	0.40 (−0.06, 0.86)	0.06 (−0.39, 0.52)	−0.15 (−0.65, 0.34)	0.36 (−0.12, 0.84)	−0.14 (−0.64, 0.35)
>100 HD patients	0.64 (0.15, 1.13)	0.51 (−0.03, 1.06)	0.59 (0.05, 1.13)	−0.23 (−0.81, 0.36)	0.16 (−0.41, 0.72)	−0.31 (−0.89, 0.27)
HD/MD Clinic						
Yes	reference group	reference group	reference group	reference group	reference group	reference group
No	0.08 (−0.32, 0.48)	−0.06 (−0.50, 0.38)	−0.45 (−0.89, −0.01)	−0.08 (−0.55, 0.40)	−0.37 (−0.83, 0.10)	0.01 (−0.47, 0.48)
Practice Setting						
Non-academic	−0.47 (−0.84, −0.09)	0.11 (−0.30, 0.53)	0.09 (−0.32, 0.50)	−0.03 (−0.48, 0.42)	0.29 (−0.14, 0.72)	0.08 (−0.36, 0.53)
Academic	reference group	reference group	reference group	reference group	reference group	reference group
Expertise of Practice Lead						
Movement disorder trained	reference group	reference group	reference group	reference group	reference group	reference group
Not movement disorder trained	−0.45 (−0.85, −0.04)	−0.45 (−0.90, 0)	−0.02 (−0.46, 0.42)	0.05 (−0.43, 0.53)	−0.07 (−0.53, 0.40)	−0.27 (−0.75, 0.21)

*reference group is the group with the largest frequency of respondent.

or multidisciplinary clinic (ES = −0.37; 95 % CI = −0.83, 0.10) having lower ratings. Notably, we did not identify any significant associations or trends for two factors: factor 4, routinely monitoring cognitive function; and factor 6, provision of patient-centered care, suggesting no evidence of a meaningful association between our HD practice characteristics and these factors. The estimated effect size relationships between HD practice characteristics and these two factors are small, ranging from 0.01 to 0.31.

4. Discussion

To our knowledge, this is the largest survey of clinical services for HD ever undertaken in the U.S. Similar to a previous survey by Frich et al (2), we found that almost 35 % of the practices we surveyed saw >50 HD patients per year and about a quarter >100. However, unlike the Frich study, which focused on Enroll-HD research centers, it was our intention to also examine provision of clinical services in smaller settings and almost 50 % of surveyed practices saw ≤25 HD patients annually. Importantly, of the practices surveyed, more than a third were non-academic and >10 % were VA-based.

Not surprisingly, we found that HD care was inconsistently applied across the US. Practices led by neurologists trained in movement disorders, and higher HD volume practices, tended to be better equipped to provide multi-disciplinary staffing as compared to practices with fewer numbers of HD patients. In addition, many practices had insufficient in-house staffing to meet the variety of needs of an HD patient. About two-thirds of all practices surveyed did not conduct a pre-visit screening to discuss patients' symptoms and care needs prior to the first visit; it was those with a dedicated or multidisciplinary clinic who were more likely to do so. About half of all practices surveyed cited the lack of dietitians, genetic counselors, psychologists, psychiatrists, care coordinators, or occupational or speech therapists. Dietitians and care coordinators were the functions least likely to be available no matter what the size of the

practice. Likewise, Occupational Therapy, Physical Therapy, Psychiatry, and Speech Therapy were the multidisciplinary functions most likely to be referred out.

On the other hand, there was a high overall rate of endorsement as regards monitoring motor function, weight, activities of daily living, behavioral health, and even cognitive functioning, of HD patients no matter the practice size. The level of assessment and implementation of that monitoring was not, however, scrutinized and may have had different meanings to different size practices. Less than a third of practices, no matter how experienced in HD care, validated monitoring patient experience of care.

Our findings on the changes in telehealth utilization as a result of the COVID-19 pandemic are also germane. Of note, our survey was conducted early in the COVID-19 pandemic, providing a unique opportunity to examine how care changed in response. Extant studies [12] indicate that in-person visits decreased dramatically at the beginning of the COVID-19 pandemic with a corresponding increase in telehealth during the pandemic. Our data indicate that the practices in this study appear to have followed general patterns seen in telehealth utilization in the United States. This is particularly important given that HD patients are a vulnerable population for whom continuity of care is vital, but who likely also sought to limit their exposure to the extent possible during the COVID-19 pandemic, making telehealth an important resource.

As expected, the VA had a higher use of telehealth prior to Covid 19, and lower percent of prescription medications, treatments and tests that require insurance follow up. However, we were somewhat surprised that this unique model of care was less likely than academic practices to offer care navigation and genetic counseling services by genetic counselors.

We were also interested in the ability of practices, as currently configured, to provide more complex disease modifying intravenous and especially intrathecal therapies were they to become available. Here we found that academic vs non-academic (58 % v 25 %); movement disorder-led vs non-movement disorder-led (57 % v 27 %); and high HD

patient volume vs low HD patient volume (47 % v 28 %) practices indicated that they would require new equipment, additional space, or additional staff/training to provide intrathecal therapy, for example, to their HD patients.

There is a lack of evidence based treatments for the majority of symptoms in HD, with the exception of chorea. Expert consensus guidelines for many features of disease have been published by individuals with significant expertise in HD [3–7], and some international groups [8–11] but will need to be updated and broadened with regard to stage of disease, care burden, geographic differences, and technological changes, to truly provide international consensus-based guidelines and benchmarks for quality improvement and a wider delivery of care.

Going forward, we also need to better elucidate and address barriers to care provision that currently exist in our health care system. While beyond the scope of this survey, factors such as cost of medical care, distance to centers of expertise, and transportation, for example, have been reported as important structural barriers by others [1,5] that will need to be incorporated into any plan for wider quality HD care delivery.

Limitations of this study include a non-responder rate that may have introduced bias toward specialty HD clinics with higher patient volumes and multi-disciplinary staffing; however, to our knowledge, this is the first survey to include large numbers of non-academic and smaller HD practices in its scope. Our survey was also limited to U.S. practices and therefore provides no understanding of organization and provision of HD care and services outside the U.S.

Nevertheless, this study adds to the current literature by providing significant information about the organization of clinical care and services for HD patients in the U.S. We found that HD care was inconsistently applied and that higher HD volume practices tended to be better equipped to provide multi-disciplinary staffing. Although multidisciplinary care or multispecialty care is considered by many to be the “gold standard”, we cannot conclude from this survey that it is necessarily the best care delivery model for HD. There is a need for additional understanding of care provision practices and gaps, proficient organization of clinical services, and the impact of consensus-based standards on care delivery improvement in HD.

CRediT authorship contribution statement

Lauren Seeberger: Conceptualization, Investigation, Project administration. **Jody Corey-Bloom:** Conceptualization, Investigation, Project administration. **Michael O'Brien:** Conceptualization, Investigation, Project administration, Supervision. **Diana Slowiejko:** Conceptualization, Investigation, Project administration, Funding acquisition. **Danielle Schlang:** Data curation, Formal analysis, Investigation, Methodology. **Marika S. Booth:** Data curation, Formal analysis, Investigation, Methodology. **Beth Ann Griffin:** Data curation, Formal analysis, Investigation, Methodology, Project administration. **Peggy G. Chen:** Data curation, Formal analysis, Investigation, Methodology, Project administration.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Ethical compliance statement

The Institutional Review Boards for the New England IRB and Rand Corporation approved the study. Informed patient consent was not necessary for this work. We confirm that all authors have read the Journal's position on issues involved in ethical publication and affirm that this work is consistent with those guidelines.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.prdoa.2022.100165>.

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