Health services utilization of Chinese patients with Huntington's disease: A cross-sectional study

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Research article

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Abstract

Background

Huntington's disease (HD) is a hereditary disease which could have a large impact on patients' quality of life. As the neurodegenerative disorders progress, HD patients are expected to regularly take follow-up medical visits for proper treatment. This study aimed to analyze the general situation of health services utilization of Chinese HD patients and factors associated with their adherence to follow-up medical visits.

Methods

We collected data from a questionnaire-based investigation conducted by Chinese Huntington's Disease Association. Data from 232 respondents were included to investigate whether they adhered to regular follow-up medical visits and the cause and correlates. Based on Andersen's behavioral model, the independent variables were categorized into predisposing, enabling and need factors. The variables were analyzed by chi-square test and stepwise logistic regression analysis.

Results

31.9% of the respondents had regular follow-up medical visits over the past year. Univariate analysis showed there were significant differences with 6 factors ($P < 0.05$), among which, according to logistic regression, 2 enabling factors (reimbursement of health insurance, need for accompanying family members to follow-up visits) and 3 need factors (perceived stage of disease, perceived effectiveness of drugs, self-care ability) were independent influencing factors of follow-up medical behaviors of Chinese HD patients. The predisposing factors investigated here did not play a part in determining patients' adherence to follow-up visits.

Conclusions

Poor adherence to medical visits among Chinese HD patients is derived from multiple factors, including reimbursement of health insurance, perceived stage of disease and effectiveness of drugs, need for accompanying family members and self-care ability. To promote HD patients' health services utilization, the improvement of health insurance system, the enhancement of social support and the development of therapeutic approaches still have a way to go.

Background

Huntington's disease (HD) is an autosomal dominantly inherited neurodegenerative disease characterized by cognitive, motor and psychiatric disorders. The onset of symptoms is generally in middle-age, followed by an irreversible progression of symptoms over 15–20 years until patients' premature death [1]. HD is

caused by CAG trinucleotide repeat expansion in the huntingtin (*HTT*) gene on chromosome 4. The normal *HTT* gene contains a sequence of 6–26 CAG repeats in general population. Mutation carriers with > 39 CAG repeats manifest the disease with full penetrance, whereas reduced penetrance is seen with 36–39 CAG repeats. The intermediate range (27–35 CAG repeats) is not associated with the disorder, but may lead to expansion and increased likelihood of disease onset in successive generations [2].

The prevalence of HD varies across the globe. In Western populations, 10.6–13.7 individuals per 100,000 are affected [3]. The prevalence rate is considered much lower in China. An epidemiological study in Taiwan estimated the average annual incidence rate of HD at 0.1/100,000 [4]. Among East Asians, the prevalence of HD is similarly reported to be 0.1–1 per 100,000 [5]. However, the precise prevalence rate in mainland China is yet to be investigated.

As HD is a chronic neurodegenerative disease with progressive manifestation of a spectrum of symptoms, patients would require multidisciplinary interventions and long-term disease management. Therefore, proper utilization of medical services is essential to HD patients for monitoring disease progression and receiving symptomatic treatments, which could also influence clinical outcomes and health care budgets. Health services utilization not only depends on individual characteristics, but also associates with public health policies and socioeconomic trends. However, evidence on the situation of health services utilization of HD patients is scarce, especially for China. Herein, this study aimed to determine the adherence to follow-up medical care among Chinese HD patients surveyed by Chinese Huntington's Disease Association. We used Andersen's model, a theoretical basis broadly used to analyze determining factors of health services utilization, to examine correlates and predictors of follow-up medical visits among Chinese HD patients. Our results have significance in providing concrete statistics on patients' use of healthcare services, determining the obstacles they might encounter in seeking medical services, and developing measures to ameliorate patients' health-services-utilization experiences.

Methods

Data collection

This was a cross-sectional study, using data from a questionnaire survey oriented towards HD patients registered in Chinese Huntington's Disease Association. From October to December 2019, information was gathered from a structured questionnaire completed by 236 diagnosed HD patients from all provinces in China except for Hainan, Taiwan and Macao. Among all the respondents, a total of 232 patients provided complete and valid information without omitted variables. Informed consent was obtained from the participants prior to the questionnaire-based investigation.

Study measures

Characteristics of interest in this study were investigated in the questionnaire, including age (8–17, 18–44, 45–64, ≥65 years), gender (male, female), education level (under elementary school, junior high school, senior high school, junior college, university or above), marital status (single, married, without children,

married, with children, separated, divorced or widowed), employment status (employed, or enrolled student, retired, unemployed or unenrolled, unable to work or study), residence registration (urban, rural), annual household income (¥, <50,000, 50,000-149,999, ≥150,000), health insurance type (basic medical insurance for urban employees, new rural cooperative medical insurance, without health insurance), perceived stage of disease (early stage, middle stage, late stage, unaware), etc. The utilization of health services, the dependent variable in this study, was based on the question “How often have you had follow-up medical visits over the past year?”. Patients who reported to have taken follow-up medical visits at least once over the past year were considered to have adhered to regular follow-up visits, while the remainder were considered non-adherent to follow-up visits.

Research model and variables

This study referred to Andersen’s behavioral model, which has been extensively applied in the investigation of health services utilization, as the theoretical basis [6, 7]. This model explains use of health care services by predisposing, enabling and need factors, into which the independent variables were categorized accordingly (Fig. 1).

Independent variables were categorized into predisposing, enabling and need factors. The dependent variable was whether the patient adhered to regular follow-up medical visits (at least once over the past year).

Statistical method

The characteristics of the individuals were summarized by descriptive analysis and the results were presented as absolute frequencies and percentages. To identify the influencing factors of patients’ adherence to follow-up visits, univariate analysis and logistic regression analysis were used successively, missing data being excluded. The former was performed with chi-square test to examine the impacts of different variables. Logistic regression analysis with backward selection was further conducted to determine independent influencing factors (removal criteria=0.05). All analyses were performed with SPSS version 17.

Results

Among the 232 respondents, 74 (31.9%) had regular follow-up medical visits, and 158 (68.1%) didn’t seek health care over the past year. As shown in Table 1, 53.9% were middle-aged (between 45 and 64). Over half (55.6%) were female, 70.7% were currently married and had children, 37.1% were unable to work or attend school, and urban residents accounted for 54.3%. The median annual household income was approximately ¥50,000.

Comparing the patients who adhered to regular follow-up visits with those who didn’t, univariate analysis demonstrated that there were significant differences with the following 6 factors ($P < 0.05$): medical expenses, reimbursement of health insurance, need for accompanying family members to follow-up

visits, perceived stage of disease, perceived effectiveness of drugs, self-care ability (Table 1). On the other hand, none of the predisposing factors (age, gender, education level, marital status, employment status, residence registration, annual household income) showed significant differences ($P > 0.05$), nor did health insurance type, access to medical care, or impact of the disease on daily life (Table 1). These results suggested that between the two groups of patients, disparities existed for the 6 aforementioned enabling or need factors, but not the others.

Table 1

Baseline characteristics and univariate analysis of adherence to follow-up visits in HD patients

Characteristic	N (%)	Adherent to follow-up visits	Non-adherent to follow-up visits	χ^2	P
Predisposing factors					
Age				4.011	0.260
8–17	6 (2.6)	3 (50.0)	3 (50.0)		
18–44	80 (34.5)	31 (38.8)	49 (61.2)		
45–64	125 (53.9)	34 (27.2)	91 (72.8)		
≥ 65	21 (9.0)	6 (28.6)	15 (71.4)		
Gender				0.276	0.599
Male	103 (44.4)	31 (30.1)	72 (69.9)		
Female	129 (55.6)	43 (33.3)	86 (66.7)		
Education level				7.237	0.124
Under elementary school	45 (19.4)	13 (28.9)	32 (71.1)		
Junior high school	72 (31.0)	27 (37.5)	45 (62.5)		
Senior high school	50 (21.6)	10 (20.0)	40 (80.0)		
Junior college	39 (16.8)	17 (43.6)	22 (56.4)		
University or above	26 (11.2)	7 (26.9)	19 (73.1)		
Marital status				1.271	0.736
Single	23 (9.9)	6 (26.1)	17 (73.9)		
Married, without children	12 (5.2)	4 (33.3)	8 (66.7)		
Married, with children	164 (70.7)	51 (31.1)	113 (68.9)		

Characteristic	N (%)	Adherent to follow-up visits	Non-adherent to follow-up visits	χ ²	P
Separated, divorced or widowed	33 (14.2)	13 (39.4)	20 (60.6)	4.993	0.172
Employment status					
Employed, or enrolled student	41 (17.7)	11 (26.8)	30 (73.2)		
Retired	46 (19.8)	17 (37.0)	29 (63.0)		
Unemployed, or unenrolled	58 (25.0)	24 (41.4)	34 (58.6)		
Unable to work or study	86 (37.1)	22 (25.6)	64 (74.4)		
Residence registration				0.053	0.819
Urban	126 (54.3)	41 (32.5)	85 (67.5)	3.063	0.216
Rural	106 (45.7)	33 (31.1)	73 (68.9)		
Annual household income (¥)					
< 50,000	116 (50.0)	31 (26.7)	85 (73.3)		
50,000-149,999	89 (38.4)	34 (38.2)	55 (61.8)		
≥ 150,000	25 (10.8)	8 (32.0)	17 (68.0)		
Enabling Factors					
Health insurance type				0.507	0.776
Basic medical insurance for urban employees	110 (47.4)	38 (34.5)	72 (65.5)		
New rural cooperative medical insurance	87 (37.5)	26 (29.9)	61 (70.1)		
Without health insurance	29 (12.5)	9 (31.0)	20 (69.0)		
Access to medical care				0.567	0.753
Local hospitals	77 (33.2)	30 (39.0)	47 (61.0)		

Characteristic	N (%)	Adherent to follow-up visits	Non-adherent to follow-up visits	χ^2	P
Cross-city	43 (18.5)	14 (32.6)	29 (67.4)		
Cross-province	67 (28.9)	26 (38.8)	41 (61.2)		
Medical expenses(¥)				8.824	0.012
< 10,000	105 (45.3)	24 (22.9)	81 (77.1)		
10,000–49,999	88 (37.9)	35 (39.8)	53 (60.2)		
≥ 50,000	31 (13.4)	14 (45.2)	17 (54.8)		
Reimbursement of health insurance				10.22	0.001
Yes	51 (22.0)	26 (51.0)	25 (49.0)		
No	170 (73.3)	46 (27.1)	124 (72.9)		
Need for accompanying family members to follow-up visits				6.735	0.034
No	28 (12.1)	8 (28.6)	20 (71.4)		
1 accompanying family member	110 (47.4)	43 (39.1)	67 (60.9)		
2 or more accompanying family members	94 (40.5)	21 (22.3)	73 (77.7)		
Need factors					
Perceived stage of disease				15.701	0.001
Early stage	48 (20.7)	20 (41.7)	28 (58.3)		
Middle stage	101 (43.5)	41 (40.6)	60 (59.4)		
Late stage	52 (22.4)	8 (15.4)	44 (84.6)		
Unaware	31 (13.4)	5 (16.1)	26 (83.9)		
Impact of the disease on daily life				4.324	0.229

Characteristic	N (%)	Adherent to follow-up visits	Non-adherent to follow-up visits	χ^2	P
No	13 (5.6)	3 (23.1)	10 (76.9)		
Mild	32 (13.8)	8 (25.0)	24 (75.0)		
Moderate	51 (22.0)	22 (43.1)	29 (56.9)		
Severe	136 (58.6)	41 (30.1)	95 (69.9)		
Perceived effectiveness of drugs				37.71	< 0.001
Effective	27 (11.6)	18 (66.7)	9 (33.3)		
Not very effective	77 (33.2)	34 (44.2)	43 (55.8)		
Not effective	66 (28.4)	17 (25.8)	49 (74.2)		
Unaware	62 (26.7)	5 (8.1)	57 (91.9)		
Self-care ability				8.635	0.035
Completely able	63 (27.2)	16 (25.4)	47 (74.6)		
Able for the majority	49 (21.1)	23 (46.9)	26 (53.1)		
Able for the minority	74 (31.9)	15 (20.3)	59 (79.7)		
Completely unable	46 (19.8)	10 (21.7)	36 (78.3)		

Data are presented as n (%). Data missing exists for the following factors: employment status, annual household income, health insurance type, access to medical care, medical expenses, reimbursement of health insurance.

Stepwise multivariate logistic regression analysis was then conducted based on all the variables investigated (definition of the variables in Additional file 1). Hosmer and Lemeshow test verified good fitness of the model ($P > 0.05$). 5 variables remained in the model after backward selection, including reimbursement of health insurance, need for accompanying family members to follow-up visits, perceived stage of disease, perceived effectiveness of drugs, and self-care ability ($P < 0.05$, Table 2), revealing that these were the independent influencing factors of follow-up medical behaviors of HD patients.

Specifically, with the reimbursement of health insurance for their medical costs, the patients were inclined to adhere to follow-up medical visits (OR = 3.276). The patients who needed only 1 accompanying family member for their medical visits were more likely to regularly visit their doctors than those who required 2 or more accompanying family members (OR = 3.488). Moreover, compared with the patients who believed themselves to be at the late stage of the disease, those self-estimated to be at the early or the middle stage tended to stick to follow-up visits (OR = 13.664, 3.746, respectively). Compared with the patients who considered the drugs ineffective, those unaware of whether the drugs were effective or not were even less likely to take follow-up visits (OR = 0.157). Besides, the patients who were partly able to take care of themselves adhered more to follow-up visits than those who were completely able (OR = 6.784, 4.632, for “Able for the majority” and “Able for the minority” respectively).

Table 2

Stepwise multivariate logistic regression analysis of adherence to follow-up visits in HD patients

Variables	β	OR	95%CI	P
Coefficient	-4.012	0.018	-	< 0.001
Reimbursement of health insurance (No)				
Yes	1.187	3.276	(1.353, 7.929)	0.009
Need for accompanying family members to follow-up visits (2 or more accompanying family members)				
No	1.374	3.950	(0.879, 17.745)	0.073
1 accompanying family member	1.249	3.488	(1.545, 7.875)	0.003
Perceived stage of disease (Late stage)				
Unaware	0.989	2.688	(0.478, 15.118)	0.262
Early stage	2.615	13.664	(2.584, 72.246)	0.002
Middle stage	1.321	3.746	(1.174, 11.954)	0.026
Perceived effectiveness of drugs (Not effective)				
Unaware	-1.849	0.157	(0.040, 0.617)	0.008
Not very effective	0.313	1.368	(0.550, 3.403)	0.501
Effective	0.657	1.930	(0.593, 6.281)	0.275
Self-care ability (Completely able)				
Able for the majority	1.915	6.784	(1.750, 26.292)	0.006
Able for the minority	1.533	4.632	(1.123, 19.110)	0.034
Completely unable	1.166	3.210	(0.604, 17.068)	0.171
OR = odds ratio; S.E.=standard error				

Discussion

This is the first study to our knowledge to investigate the situation of health services utilization of Chinese HD patients and to analyze the determinants. Our initial finding was that less than 1/3 of the respondents took regular follow-up medical visits over the past year, suggesting extensive non-adherence to follow-up among HD patients. The utilization rate established in the current study was lower than that reported in a previous study in the US, with 42% of HD patients receiving long-term care services [8]. As suggested by the World Health Organization, improvement of adherence interventions may be far more efficient to promote the health of the population than advances in biomedical treatments [9]. To enhance

adherence, it is necessary to examine the origins of loss of follow-up. Using logistic regression, we determined 2 enabling factors (reimbursement of health insurance, need for accompanying family members to follow-up visits) and 3 need factors (perceived stage of disease, perceived effectiveness of drugs, self-care ability) as the main factors influencing HD follow-up practices. The predisposing factors, namely the demographic characteristics of the patients, were not among the correlates of health services utilization, which indicated that regardless of age, gender, education and income levels, marital and employment status, residence, Chinese HD patients generally had poor adherence to follow-up medical visits.

The reimbursement of health insurance largely promoted patients' health services utilization, with an OR of 3.276 (Table 2). However, up to 73.3% of the respondents claimed not to benefit from health insurance for their medical costs (Table 1). With substantial hospital services and pharmacy costs, HD burdens patients with significant medical expenses in the long run [10], as are most rare diseases [11], whereas it seemed that basic medical insurance failed to improve the affordability of medical costs of Chinese patients with rare diseases [12]. China is still exploring policies regarding health insurance system for rare diseases. Although in areas such as Shanghai and Zhejiang, basic health insurance covers some orphan drugs, patients from remote areas have to disburse cross-provincial medical costs and seek reimbursement afterward. Thus, the unavailability of reimbursement of health insurance could partly impede regular healthcare utilization of patients.

Besides, we concluded that the patients self-considering to be at the late stage of HD, as well as those who were not convinced of the effectiveness of drugs were less likely to take follow-up visits. Previous studies showed that patients with poorer self-rated mental health were significantly more likely to receive treatment for emotional problems [13], which was not the case with the HD patients in our study. Indeed, it was suggested that Health-Related Quality of Life (HRQoL) was lowest in HD patients with advanced disease stage. Meanwhile, patients with a high level of unmet healthcare needs had lower HRQoL [14]. Faced with the gradual progression of the disease and limited approach of treatment, the patients who were pessimistic or lost faith in the possibility to be cured could be less willing to comply with HD long-term management. Our examination of the need factors revealed how patients' view of their health status and the cure played a part in determining their adherence to follow-up visits.

Other influencing factors of patients' health services utilization include their need for accompanying family members to follow-up visits and self-care ability. We speculated that as the motor disorders progressively impair patients' self-care ability, they might sense the urgent need to seek medical care for the alleviation of symptoms. On the other hand, when a patient is disabled to the extent that he or she needs to be accompanied by 2 or more family members, follow-up visits may be too troublesome and thus the frequency of medical visits declines.

Based on the research findings, we propose that to promote follow-up medical visits in HD patients, the rate of health insurance reimbursement of rare diseases such as HD should first be elevated. Second, social and community support plays an essential part in long-term disease management. Though social

support could help patients retain hope and facilitate their access to medical care, this service highly warranted by HD patients remains a need that is currently unmet [15]. Third, despite research advances over the past two decades and the emergence of therapeutic strategies and clinical trials, there has been unfortunately minimal success to date and few approaches are available for HD treatment. As the shortage of therapeutic means has been shown to contribute to the low utilization rate of health services among patients with rare diseases in China [16], the development of drugs and new therapeutic approaches is also indispensable.

This study has the following limitations. First, due to the cross-sectional nature of the study, the causality or temporal relation between the frequency of healthcare utilization and the variables could not be determined. Second, due to the limitation of the investigation, the variables that we have included in this study may be incomplete. Third, the data were collected by questionnaire survey in which information was self-reported by patients and was subject to recall errors. Finally, there has been data missing for several factors, especially access to medical care (local hospitals, cross-city, cross-province) as respondents who didn't have follow-up medical visits opted for "did not adhere to follow-up visits". This could have attenuated the influence of accessibility to medical care on health services utilization. Despite these limitations, our results highlight the determining role that enabling and need factors play in health services utilization of Chinese HD patients by applying the Andersen's model. This study also has significance in that it provides baseline data of one specific type of rare disease in China, and may serve as a reference for health service policy-making.

Conclusion

This study analyzed the situation of health services utilization of Chinese HD patients and associated factors, using data from Chinese Huntington's Disease Association and Andersen's behavioral model as theoretical basis. Our results suggested that non-adherence to follow-up medical visits is frequent among Chinese HD patients. Reimbursement of health insurance, need for accompanying family members to follow-up visits, perceived stage of disease, perceived effectiveness of drugs, and self-care ability were the 5 independent influencing factors of their follow-up medical behaviors. Therefore, measures to improve the medical practice and quality of life of HD patients should focus on a more developed health insurance system, broader social support for rare disease sufferers, and researches for more effective treatments.

Abbreviations

HD: Huntington's Disease; OR: odds ratio; S.E.: standard error; CI: Confidence Interval; HRQoL: Health-Related Quality of Life

Declarations

Ethics approval and consent to participate

Access to the data records was granted by Chinese Huntington's Disease Association. Ethics approval for this study was not necessary since it was based exclusively on data provided by Chinese Huntington's Disease Association and the study subjects were not directly approached (Act of the People's Republic of China on Ethics Review of Biomedical Research Involving Human Subjects, December 2016 [17]).

Consent to publish

Not applicable.

Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Competing interests

The authors declare that they have no competing interests.

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Authors' contributions

YS and LC conceived and designed the study. XC and LC acquired the data. HK, YS and LC analyzed and interpreted the data. HK wrote the manuscript draft. All authors critically reviewed and approved the final version.

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Figures

Figure 1

Research model and variables

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