

Original Research Article

Assessment of health-related quality of life (HRQoL) in patients with spasmodic dysphonia

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ABSTRACT

Background: There is paucity of literature regarding health related quality of life in Spasmodic Dysphonia (SD) especially from India. This study assessed HRQoL in it's global and disease specific aspect by previously validated instruments in patients with SD.

Methods: The study was performed in AIIMS, New Delhi. Subjects with SD as well as age and gender matched healthy controls were enrolled from Movement Disorder and botulinum toxin clinic, Department of Neurology, AIIMS, New Delhi. Uneducated patient, those could not read questionnaires & cases who had received botulinum toxin within 6 months were excluded from the study. Each patient filled SF-36, BDI, VoiSS-30 and VPQ questionnaire.

Results: 14 pts of Spasmodic Dysphonia (SD) were enrolled. Compared with controls SD patients suffered from statistically significant impaired global health related quality of life (SF36) in areas of role physical ($p = 0.007$), general health ($p = 0.004$), social functioning ($p = 0.024$), role emotion ($p = 0.008$) and mental health ($p = 0.039$). Patients with SD scored much worse in BDI scale than their control group (12.57 ± 8.0 vs. 4.71 ± 5.0 , $p = 0.005$). 65% pt of SD had depression out of whom 14% had moderate depression. SD patient group showed statistically significant impaired scores in disease-specific QOL (VoiSS) in 2 out of 3 subscales, impairment ($p < 0.001$) and emotional ($p < 0.001$) but not in physical ($p = 0.44$). Voice quality of patients with SD was severely affected compared to controls (mean 37 ± 8.0 vs. 12.9 ± 1.5).

Conclusions: This study clearly demonstrated that patients with SD suffered from significant impairment in HRQoL as compared to controls. Higher proportion of patients with SD suffered from mild to moderate depression compared to their control.

Keywords: BDI, HRQoL, SD, SF-36, VPQ, VoiSS

INTRODUCTION

Dystonia is defined as a “neurologic syndrome characterized by involuntary, sustained, patterned contractions of opposing muscles, causing twisting and repetitive movement or abnormal postures” may be associated with tremor (dystonia tremor) or myoclonus

(myoclonic dystonia).¹ Spasmodic dysphonia is a focal dystonia involving involuntary contractions of the vocal cords causing abnormal excessive adduction or abduction of vocal cords leading to interruptions of speech and abnormal voice quality. One of the most characteristic features of spasmodic dysphonia is the patterned, repeated “breaks” in speech.

Primary dystonia is one of the most prevalent movement disorders. Worldwide prevalence of focal dystonia varies from 3-732 per 1,00,000 population from various studies.²⁻⁴

In only Indian community-based study, crude prevalence rate of focal dystonia is 43.9 per 1,00,000 population. In majority of studies cervical dystonia (CD) and blepharospasm (BSP) are found to be common dystonia accounting for about 75% of cases with primary focal dystonia while Spasmodic dysphonia was very rare type of focal dystonia. Indian study by Das et al, shows that writer's cramp and blepharospasm are the most common focal dystonia.⁵

Very little is known about impact of spasmodic dysphonia on quality of life. In most cases life expectancy is not reduced however it may be responsible for considerable morbidity in terms of pain, low self-esteem, depression, embarrassment and poor social interaction. Health-related QoL (HRQOL) is a multi-dimensional concept that encompasses the subjective assessment of the impact of illness or treatment across the physical, psychological, social and somatic domains of functioning and the well being.⁶

There is paucity of literature regarding health related quality of life in patient of spasmodic dysphonia (SD) especially from India. Little is known about the clinical and demographic factors associated with poor HRQoL and depression in patients with SD.

Since spasmodic Dysphonia have a life time visible chronic disability, it is important to identify the factors that influence quality of life in these patients to optimize the goal directed therapy. This study was conducted to assess HRQoL in it's global and disease specific aspect by previously validated instruments in patients with spasmodic Dysphonia (SD).

METHODS

The study was performed between Jan 2007 to July 2008 in All India Institute of Medical Sciences, New Delhi. Subjects with spasmodic dysphonia as well as age and gender matched healthy controls were enrolled from Movement Disorder and botulinum toxin clinic, Department of Neurology, AIIMS, New Delhi.

All the patients aged > 15 years with clinical diagnosis of spasmodic dysphonia by movement disorder specialist, were screened for the enrollment in the study. Uneducated patient, those could not read questionnaires, cases that had associated other neurological or debilitating systemic disorders, secondary / pediatrics dystonias, pregnancy or received botulinum toxin within 6 months or underwent surgical treatment were excluded from the study.

Ethical clearance was taken from institutional ethical committee (IEC) of All India Institute of Medical Sciences, New Delhi. Written informed consent was taken after explaining nature and need of the study. Finally, study patients were enrolled after fulfilling the inclusion and exclusion criteria.

Study Questionnaire

After enrolment, demographic and clinical details of cases were noted down in a present form designed for the study. Each patient filled SF-36 (for HRQoL), BDI (Beck Depression Inventory for depression), VPQ and VoiSS.

Global HRQoL

SF-36^R (short form 36) is acceptable, internally consistent, valid and reliable measure of the health status of patients SF36^R a 36 item, self-report generic measure that provides a profile assessment of health-related quality measuring multi-item variables, which includes; physical functioning (PF, 10 items), role limitation due to physical problem (RP, 4 items), bodily pain (BP, 2 items), general perception of health (GH, 5 items), vitality (VT, 4 items), social functioning (SF, 2 items), role limitation due to emotional problem (RE, 3 items) and mental health (MH, 5 items).^{7,8} A score ranging from 0 (worst health) to 100 (best health) is generated for each domain/subscale.

Disease specific quality of life scales

Disease specific quality of life in patients with Spasmodic Dysphonia was assessed with voice symptoms scale (VoiSS-30), a 30 item, self-report generic measure. The voice symptom scale is a 30-item scale with three content domains (Impairment, Emotional, Physical) and a total score, the reliability and validity of which have been assessed in a series of studies involving over 800 subjects.^{9,10}

Disease severity scales

In patients of SD, voice quality was assessed by Voice performance questionnaire (VPQ) consisting of 12 items which address the physical aspects of the voice problem and its social and emotional impact.¹¹

Statistical analysis

Comparison between all the variables described earlier for patients vs. control was carried out using t- test and χ^2 (chi-square) test for continuous and categorical variables, respectively. Association between SF 36 and VPQ subscales and variables addressing disease duration, age of onset and severity were evaluated by Pearson correlation coefficients. P values of 0.05 or less (2-sided) were considered statistically significant. All data were analyzed using SPSS 12 software.

RESULTS

Demographic characteristics

This study included 14 patients of SD. Demographic details of these patients are described in Table (1) There was no significant difference in demographic details between patients with spasmodic dysphonia and control.

Table 1: Demographic characteristics among patients with spasmodic dysphonia and SD- Controls.

Demographic Characteristics	SD (n=14)	Controls (n=14)	p value
Age mean (yrs) (SD)	45.9 (15.3)	46.7(8.0)	0.88(NS)
Age groups (%)			
<39yrs	35.8	21.4	0.17(NS)
40-49yrs	21.4	35.8	
50-59yrs	21.4	42.8	
60-69yrs	21.4	0	
>70 yrs	0	0	
M:F(n)	5:9	4:10	0.69(NS)
Education (%)			
≤ 12thstandard	21.4	21.4	0.54(NS)
Graduate	57.2	71.4	
Post graduate or higher	21.4	7.2	
Marital status (%)			
Married	85.6	78.6	0.62(NS)
Unmarried	14.4	21.4	
Current employment status (%)			
Currently employed	50	78.6	0.13(NS)
Retired	7.1	0	
House wives	35.7	21.4	
Unemployed	7.1	0	
Age of onset mean (yrs), SD	40.9(14.9)	NA	
Age of onset groups (%)			
≤ 39yrs	57.2	NA	
40-49 yrs	7.2	NA	
50-59yrs	21.4	NA	
60-69yrs	14.4	NA	
>70 yrs	0	NA	
Duration mean (years), SD	4.86(2.3)		
Duration groups (%)			
≤2yrs	21.4	NA	
3-5yrs	35.7	NA	
>5yrs	42.9		

QOL Characteristics.

Compared with controls SD patients suffered from statistically significant impaired global health related quality of life (SF36) in areas of role physical (p = 0.007), general health (p = 0.004), social functioning (p = 0.024), role emotion (p = 0.008) and mental health

(p=0.039), while there was no significant difference in areas of physical functioning (p=0.45), vitality (p=0.87) and bodily pain (p = 0.15) (Table 2).

Table 2: QOL Characteristics among patients with spasmodic dysphonia and SD-Controls.

	SD	Controls	p value
SF 36 subscale, mean (SD)			
Physical Functioning (PF)	85.7 (17.2)	83.6 (15.2)	0.45 (NS)
Role Physical (RP)	48.2 (22.9)	75 (21.9)	0.007
Bodily Pain (BP)	72.6 (24.3)	85.6 (20.7)	0.15 (NS)
General Health (GH)	35.0 (21.9)	64.3(23.7)	0.004
Vitality (VT)	64.3 (16.4)	62.3 (23.6)	0.87 (NS)
Social Functioning (SF)	60.7 (16.9)	78.6 (19.9)	0.024
Role Emotional (RE)	50.0 (21.7)	77.3 (26.0)	0.008
Mental Health (MH)	58.0 (20.1)	74 (14.4)	0.039
BDI mean (SD)	12.57 (8.0)	4.71 (5.0)	0.005
BDI groups, %			
1-10: Normal	35.8	85.6	
11-16: Mild mood disturbance	35.8	7.2	
17-20: Borderline depression	7.2	7.2	
21-30: Moderate Depression	14.4	0	
31-40: Severe depression	0	0	
Over 40: Extreme depression	0	0	
VPQ, mean (SD)	37.4 (8.0)	12.9 (1.54)	<0.001
VOiSS			
Im	48.3 (9.8)	19.1 (4.3)	<0.001
Em	21.4 (6.8)	8.8(1.8)	<0.001
Phy	11.1 (2.3)	11.8 (2.9)	0.44 (NS)
Sum	26.9 (5.6)	13.3 (2.5)	<0.001

Patients with SD scored much worse in BDI scale than their control group (12.57±8.0 vs. 4.71±5.0, p=0.005) (Table 2). 65% patients of SD had depression (as compared to 15% of controls) out of whom 14% had moderate depression (as compared to 0% of controls) (Table 2). SD patient group showed statistically significant impaired scores in disease-specific QOL (VOiSS) in 2 out of 3 subscales, impairment (p= <0.001)

and emotional ($p < 0.001$) but not in physical ($p=0.44$). Voice quality (assessed by VPQ) of patients with SD was severely affected compared to controls (mean 37 ± 8.0 vs. 12.9 ± 1.5).

There was no correlation of VoiSS subscales and voice quality (VPQ) with age, gender, marital status, education, occupational status, duration of disease and age at onset of disease ($p > 0.05$). BDI score was significantly associated with age (younger better $p=0.02$), marital status (married better, $p=0.011$) and education (educated better, $p=0.011$).

Significant correlation of Quality of voice was found with BDI (poorer voice more depression score, $p=0.04$) and mental health domain of SF 36 (poorer voice more poor mental health score, $p=0.003$). There was no statistically significant correlation of Quality of voice with any other domains of SF36 or VoiSS.

DISCUSSION

This study was designed to provide information regarding impact of spasmodic dysphonia (botulinum toxin naïve patients) on QoL consisting of physical, psychological and social aspects of life as well as disease specific aspects of life. As effect of Botulinum toxin usually last for 2-4.5 months, all the patients who received botulinum toxin within 6 months were excluded from study.^{12,13}

HRQoL is a tool to assess impact of disease and treatment on QoL. Studies regarding impact of SD on HRQoL are very limited in numbers (Table 3). There is no study in patients with SD from India addressing the issue of QoL. As Meige’s syndrome is very rare disorder, we were able to enroll only 14 botulinum toxin naïve patients. In One study from India investigator S.K.Singh et al, looked for effect of botulinum toxin on voice quality.¹⁴

The previous research addressing HRQOL in SD had methodological limitations, like; (1) using questionnaire instruments whose psychometric properties are unknown, (2) no control group to compare, (3) studies have included patients treated with botulinum toxin [as effect of botulinum toxin on QoL is still controversial in SD.¹⁵⁻¹⁷

Present study clearly demonstrated that patients with SD suffered from significant impairment in HRQoL as compared to controls. In SF-36, patients with SD had impaired HRQoL in 5 out of 8 domains (except vitality, physical functioning and bodily pain). Studies in patients with SD showed poor HRQoL (global health) and decreased efficiency at work.¹⁸ Significant effect of spasmodic dysphonia on mental and social life could be because of voice change and feeling that they are drawing unwanted attention of others due to this disability. Physical function domain did not show significant changes compared to controls as SD usually do not directly affect physical activities like walking, lifting objects, bathing etc.

Higher proportion of patients with SD suffered from mild to moderate depression compared to their control. Higher incidence of depression could be due to impaired mental and social life and voice change. Study in patients with SD showed associated psychiatric co-morbidity and depression.

Regarding disease specific QoL, in patients with SD, voice related quality of life was found to be affected in 2 of 3 domains of impairment and emotional (except physical sub scale). Similar observation was found in few previous studies, but most of which were conducted as a part of study designed to judge effect of botulinum toxin on voice related quality of life and no comparison with normal population was done.^{19,20}

Table 3: Reviews of studies of BS and SD.

Author, Year	n	Scale used	Comments
Indian study			
Singh SK et al ¹⁴	SD= 10	Voice handicap index scoring	To see effect of botulinum toxin on quality of voice Did not assess QoL in patients
International study			
Bhattacharyya N et al ²²	SD: 18 OMD: 5 Total: 23	GBI	No general or disease specific quality of life scale used. OMD/SD: Significant benefit with Btx Treatment with Btx for these conditions is effective on the basis of quality of life criteria
Baylor CR et al ²³	Six participants	Participants’ self-rated voice quality and interview	No controls were taken, Communication related quality of life was significantly affected
Watts CR et al ²⁴	SD=148	Behavior Assessment Battery (BAB-Voice)	External controls were taken speakers with SD experienced substantial impact of their voice disorder on communication attitude, coping behaviors, and affective reactions

There was no correlation of disease severity, depression (BDI) or disease specific QoL with gender, occupational status, age at onset and duration of disease in patients with SD. Younger, educated and married patients have less depression. Significant correlation of Quality of voice was found with depression (poorer voice more depression) and mental health domain of SF 36 (poorer voice more poor mental health). Jones et al also found significant correlation of voice quality and disease specific quality of life in patients with SD.²¹

CONCLUSION

The present study's result regarding HRQoL in its global and disease specific aspect provides further evidence for profound impact of spasmodic dysphonia on physical, psychological and social aspect of quality of life. This study indicates that psychological counseling of patients, their family members and treatment aiming to treat depression may be a part of comprehensive treatment approach for these patients. Treatment should also improve quality of life of patients which is found lacking in several treatment trials of botulinum toxin indicating requirement of much more broad and comprehensive approach.

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