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Mental Health of People with Retinitis Pigmentosa

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ABSTRACT

Purpose. The purpose of this study was to evaluate and compare the mental health of patients with retinitis pigmentosa (RP) with that of the general population of Korea.

Methods. Online surveys were completed by patients registered with the KRPS (Korean Retinitis Pigmentosa Society), an online organization that promotes research on RP and provides advocacy and online and offline support and information for patients with RP. Control population was selected from the fourth round of the KNHANES (Korean National Health and Nutrition Examination Survey). One hundred eighty-seven patients with RP were matched with the control population using the propensity-score method to optimize comparative analysis.

Results. Stress was reported in 51.9% of RP patients and 29.4% of controls ($p < 0.001$). Depressive mood of at least 2 weeks' duration in the previous year was reported by 34.8 and 17.1% of patients and controls, respectively ($p < 0.001$). Suicidal thoughts were reported by 38.5 and 12.9% of patients and controls, respectively ($p < 0.001$), although there was no significant difference in the number of suicide attempts between the groups (2.1 vs. 1.6%, $p = 0.703$). In multivariate analysis, disability rating was significantly associated with stress (adjusted odds ratio, 0.46; 95% confidence interval, 0.24 to 0.88).

Conclusions. People with RP had poorer mental health than the general population. Further investigations are warranted on the mental health of RP patients, and appropriate welfare services are needed to decrease the impact of mental illness in this population.

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Key Words: retinitis pigmentosa, visual impairment, young, mental health, depression, stress

Of all the sensory losses, loss of vision may cause the most severe social and psychological disabilities.[1] Retinitis pigmentosa (RP) is a leading cause of blindness in younger individuals, with most patients losing their sight by the age of 40 years.[2] Retinitis pigmentosa is a degenerative disease that leads to progressive and irreversible deterioration in visual function usually during a period of several decades.

The psychological impact of vision loss has been associated with reduced quality of life, anxiety, depression, and suicide, which have been investigated in numerous previous studies in older patients with age-related macular degeneration,[3–5] diabetes-related retinopathy,[6] and other age-related eye diseases.[7–9] However, relatively few studies have evaluated the effects of early-onset progressive vision loss on the emotional state of younger individuals. Of the few studies addressing the issue of mental health in this younger group of visually impaired patients, most were conducted in single centers without controls[10] or as qualitative studies with limited sample sizes,[11] whereas others used patients' personal statements describing their experiences with RP.[2,12] Furthermore, to date, no study has compared the mental health of RP patients with that of the general population. Thus, the purpose of our study was to compare the mental health of RP patients in Korea

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with that of the general population and to identify factors associated with mental health in RP patients.

METHODS

Study Design, Subjects, and Data Collection

The study was conducted as part of an investigation of obesity prevalence in RP patients. This particular study focused on the health behaviors of people with RP and compared them with those of the general population.

Participants were recruited from the Korean Retinitis Pigmentosa Society (KRPS), a patient advocacy group composed of approximately 200 active members with RP. With administrative and technical support from the KRPS, a Web-based survey was administered for 3 months from December 2010 through February 2011. During this period, a notice was generated to introduce the survey on access to the KRPS Web site. Those who were eligible for the study were encouraged to participate in the survey through phone calls and text messages from the KRPS and were provided with an online informed consent document. Patients were considered eligible for participation if they (1) were 19 years old or older and (2) had been diagnosed with RP and had received a disability rating from the government.

The online questionnaire was developed to ensure completion: if any question was left unanswered, the questions that followed were inaccessible, thus eliminating incomplete surveys. Audio instructions and surveys were provided for those who had severe visual impairment. The research followed the tenets of the Declaration of Helsinki, and the study protocol was approved by the institutional review board of Seoul National University Hospital (IRB No. C-1011-071-340). Informed consent was obtained from all participants. Although the patients were not required to sign a disclaimer, they were asked to click on the "agree" button on the Web site before proceeding to the survey and were made aware that the data were to be used for study purposes only.

Controls

Control participants were selected from the Korean National Health and Nutrition Examination Survey (KNHANES), a nationwide survey on the health and nutritional status of a nationally representative sample of civilian noninstitutionalized Korean people. For the control group, we used data from the fourth round of KNHANES, the most recent survey performed (from 2007 to 2009). Among the 13,009 participants aged 19 to 70 years (we applied an upper limit because the oldest patient with RP was 70 years old) who took part in the KNHANES, 136 reported visual disabilities and were excluded. Of the remaining 12,873 people, an individual control was matched to each RP patient using propensity-score matching, a methodology used to ensure comparability to the treatment group and provide unbiased estimation of treatment effects in the statistical analysis of observational data.[13] For this analysis, propensity scores were estimated through multiple logistic-regression analysis using baseline characteristics; this methodology was preferred to adjustment by multiple logistic regression analyses because of the large differences in the characteristics and sample sizes of the comparator groups. The baseline characteristics were age, sex, education level, marital status, income, and employment status.

Measures of Mental Health

To assess the mental health of the study population, the surveys provided to the participants included the same questions as those in the KNHANES. Three dimensions within the domains of health status and mental health were determined: stress, depression, and suicidal thoughts and attempts. Participants reported their level of stress as none, mild, moderate, or severe. To assess depression, participants answered either "yes" or "no" to a question on whether they had experienced depressed mood for two or more continuous weeks during the previous year. A "yes" or "no" response was also used to determine if they had suicidal thoughts, and if they answered "yes," they were asked about their suicide attempts, if any.

The following demographic characteristics were collected: age, sex, highest level of education (dichotomized as less than high school vs. college and above), marital status, household income (≥2 million Korean won per month [$US 1800 per month] vs. <2 million Korean won per month), and employment status. To assess the degree of disability, patients were asked for their disability rating as issued by the government. According to the Korean Disability Act, visual disability is graded according to the government criteria after being subjected to a visual acuity test, visual field test, fundoscopic examination, and an objective medical assessment by a board-certified ophthalmologist thereafter. Disability grading is based on the best-corrected visual acuity of the better eye and the degree of central field remaining (for specific criteria, see Appendix 1 available at http://links.lww.com/OPX/A119). A visual acuity examination using the Snellen test and a visual field test using Goldmann perimetry were performed for each patient. An ophthalmologist or a specially trained technician recorded isopters III-4e for each patient using the Goldmann perimeter. If the patient had a best-corrected visual acuity of less than 20/100, the isopters "V" was used. Fundoscopic findings suggestive of RP were necessary for the diagnosis of RP. The disability rating system is used by the government when providing a social welfare allowance or assistance. For the analyses, we classified RP participants with a disability rating of 1 to 3 as having severe disability and those with a rating of 4 to 6 as having mild disability. While evaluating the effects of duration of the disease on the study end points, the duration of visual disability from the year 2010 (>1 year or <1 year) was calculated; if the visual impairment was present for less than 1 year, we classified the RP as recent.

Statistical Analysis

The demographic characteristics of the RP participants and controls in unmatched and matched samples were compared using t tests for continuous variables and χ^2 tests for categorical variables. Differences in mental health status between RP participants and controls in the matched sample were evaluated using χ^2 tests. Multiple logistic regression analyses were performed to identify factors, including demographic characteristics and the year of diagnosis, associated with adverse mental health in people with RP.

TABLE 1.
Characteristics of the general population and people with retinitis pigmentosa

	Unmatched samples					Matched samples				
	Controls		RP subjects			Controls		RP subjects		
	n = 12,873		n = 194			n = 187		n = 187		
	n	%	n	%	p	n	%	n	%	p
Age, mean ± SD, yr	45.6 ± 13.8		40.0 ± 11.2		<0.001	40.5 ± 13.4		40.1 ± 11.0		0.355
Sex										
Male	5140	39.9	129	66.5		123	65.8	124	66.3	
Female	7733	60.1	65	33.5	<0.001	64	34.2	63	33.7	0.913
Education										
College and above	3554	27.7	132	68.0		112	59.9	125	66.8	
High school and below	9295	72.3	62	32.0	<0.001	75	40.1	62	33.2	0.163
Marital status										
Married	9738	75.9	117	60.3		111	59.4	114	61.0	
Unmarried	3091	24.1	77	39.7	<0.001	76	40.6	73	39.0	0.751
Income										
≥2 million won	7496	62.3	72	37.1		59	31.6	72	38.5	
<2 million won	4536	37.7	122	62.9	<0.001	128	68.4	115	61.5	0.159
Job										
Yes	7851	61.3	122	62.9		125	66.8	120	64.2	
No	4959	38.7	72	37.1	0.650	62	33.2	67	35.8	0.587
The degree of disability										
1st degree			55	28.4				53	28.3	
2nd degree			5	2.6				5	2.7	
3rd degree			49	25.3				49	26.2	
4th degree			44	22.7				41	21.9	
5th degree			18	9.3				17	9.1	
6th degree			23	11.9				22	11.8	
Year of diagnosis										
Before 2010			126	64.9				119	63.6	
2010			68	35.1				68	36.4	

p values were calculated by *t* test (for continuous variables) and χ^2 test (for categorical variables).

All the reported p values are two-sided, and values of $p < 0.05$ were considered statistically significant. We used SAS 9.1 (SAS Institute, Inc., Cary, NC) for propensity-score matching and STATA 11.1 (StataCorp., College Station, TX) for statistical analyses.

RESULTS

Baseline Characteristics

A total of 194 eligible RP patients voluntarily responded to the online survey. One hundred eighty-seven individuals were matched with controls from the general population using propensity score matching. The characteristics of the unmatched and matched participants who completed the online questionnaire are presented in Table 1. Compared with the general population, RP participants were more likely to be young, male, highly educated, and earning a lower monthly income than controls. However, propensity matching successfully eliminated these differences and, as a result, there were no significant differences between the matched sample groups.

Differences in Mental Health

Table 2 presents the differences in mental health between the study population and the control group. The proportion of patients reporting moderate to severe stress was 51.9% compared with 29.4% of the controls ($p < 0.001$). Depression was reported in 34.8% of patients and 17.1% of controls ($p < 0.001$), whereas suicidal thoughts were reported by 38.5% of participants and 12.9% of controls ($p < 0.001$). There were no significant differences between the groups in the number of suicide attempts during the previous year (2.1 vs. 1.6%, $p = 0.703$).

Factors Associated with Mental Health in RP Patients

In multivariate analysis, patients' disability rating was associated with stress, such that RP patients with a mild degree of visual impairment reported more stress than patients with a higher disability rating (adjusted odds, 0.46; 95% confidence interval, 0.24 to 0.88) (Table 3). No other variables, including age, sex, education level, marital and employment status, income level, and

TABLE 2.
Mental health of RP patients and matched controls

	Controls		RP patients		
	n = 187		n = 187		
	n	%	n	%	p
Stress					
Moderate to severe	55	29.41	97	51.87	
None to mild	132	70.59	90	48.13	<0.001
Experiences of depressive mood for 2 or more continuous weeks					
Yes	32	17.11	65	34.76	
No	155	82.89	122	65.24	<0.001
Suicidal thoughts during the previous year					
Yes	24	12.90	72	38.50	
No	163	87.10	115	61.50	<0.001
Suicidal attempts during the previous year					
Yes	4	2.14	3	1.60	
No	183	97.86	184	98.40	0.703

p values were calculated using χ^2 test (for categorical variables).

the duration of visual impairment were significantly associated with any domain of mental health.

DISCUSSION

To our knowledge, this is the first study to compare the mental health of RP patients with that of the general population of Korea. As expected, RP participants reported more stress, depression, and suicidal thought compared with that of the general population. This result is consistent with that of previous studies that have reported increased stress, depression, and suicidal ideation among visually impaired older people.[3–9,14–16]

Participants with vision loss from RP reported more stress compared with the controls. A previous qualitative study suggested that stress may begin when an objective event involving a loss, challenge, or threat (e.g., the diagnosis of a disease) requires adaptation.[11] In that sense, a slow and progressive disease such as RP may require continuous adaptation to the disease and therefore result in stress. Furthermore, difficulties with mobility, increased fear of falling, and loss of independence have been suggested to contribute to stress in patients with vision loss from RP.[11] Although we did not analyze stress factors in depth, our study similarly demonstrated that vision loss from RP represents a significant risk factor for stress.

A higher proportion of the subject population reported at least one depressive mood during the past year compared with the controls. This is consistent with previous studies that have reported a high prevalence of depression in visually impaired persons.[4,8] Previous studies assessing depression using the Beck Depression Inventory have reported a prevalence of depression as high as 25.7% in RP patients.[10] Other studies have reported the association of depression as a result of a reduced level of

TABLE 3.
Factors associated with the mental health of RP patients*

	Stress (moderate to severe) aOR (95% CI)	Depression aOR (95% CI)	Suicidal ideation aOR (95% CI)
Demographic characteristics			
Age >40 yr (vs. <40 yr)	0.62 (0.30–1.26)	0.77 (0.36–1.64)	0.83 (0.40-1.74)
Male (vs. female)	0.95 (0.48–1.87)	1.03 (0.51–2.11)	0.94 (0.47–1.88)
Married (vs. unmarried)	0.81 (0.39–1.70)	0.71 (0.33–1.52)	0.63 (0.30–1.33)
Highly educated (vs. less educated)	0.78 (0.40–1.51)	0.76 (0.38–1.51)	0.58 (0.30–1.13)
High income (vs. low income)	1.37 (0.67–2.78)	1.78 (0.85–3.76)	1.27 (0.62–2.63)
Employment (vs. unemployment)	1.02 (0.53–1.97)	0.81 (0.41–1.61)	0.91 (0.47–1.77)
Patient characteristics			
Severe disability rating (vs. mild disability rating)	0.46 (0.24–0.88)	0.66 (0.32–1.34)	0.89 (0.45–1.74)
Year of diagnosis before 2010 (vs. after 2010)	0.91 (0.49–1.69)	1.67 (0.83–2.97)	1.37 (0.74–2.57)

*Multiple logistic regression analysis adjusted for all variables in the table.
aOR, adjusted odds ratio; 95% CI, 95% confidence interval.

functioning and increased fluctuation of visual acuity and visual field loss in RP patients.[12,17] However, our study is unique in that we have compared the prevalence of depression in RP patients relative to that of the general population.

Previous studies examining the relationship between reported visual impairment and suicide mortality have indicated a possible increased risk of suicide associated with visual impairment mediated by effects on self-rated health.[14] Postmortem case reports of suicides involving sensory losses (hearing and visual) also revealed that fear of blindness may be associated with severe psychological distress that can lead to suicide.[18] Correspondingly, the number of RP participants who reported having suicidal thoughts in our study was three times higher than that in the controls. However, no significant difference was observed between the groups in the actual number of suicide attempts.

Patients with mild visual impairment (disability grades 4 to 6) experienced more stress than those with more severe impairment (disability grades 1 to 3). Knowledge of the progressive nature of RP and its poor prognosis may cause stress to be more pronounced in those with earlier and milder disease. In addition, because individuals with more severe visual disability receive more benefits from the government (The Ministry of Health and Welfare), the less severely impaired patients may possibly feel underserved, thus experiencing more stress. A previous study reported that environmental factors, as well as individual factors, can influence a person's reaction to a disability.[19] Those with disabilities may experience more stress because of social rejection, discriminatory employment practices, physical barriers, and inaccessibility to transport. Lesser amount of benefit from the government could be another such environmental factor explaining the higher level of stress in mildly visually impaired patients compared with more severely impaired patients.

Another interesting finding was the lack of a significant association between recent RP diagnosis and the degree of depression, stress, and suicidal thoughts. This result differs from that of a previous study conducted in older patients with retinal diseases, such as age-related macular degeneration and diabetic retinopathy. In that study, a shorter disease duration was significantly associated with emotional distress, suggesting that participants may adapt to their vision loss over time.[9] The discrepancy between this study and ours may be because many RP patients are severely visually impaired at the time of diagnosis. In addition, RP is an unpreventable and progressive disease, with no known effective treatment. Subsequently, we cannot ignore the various difficulties RP patients must face while having to continuously adapt to new levels of visual impairment as the disease progresses. Another possible reason for the discrepancy is that RP affects younger people who would otherwise have maintained a more socially active lifestyle if they did not have the disease.

There are several limitations to this study. First, the questions examining mental health, such as depression, stress, and suicidal ideation, were simplified and not formally validated; therefore, we could not conduct a deeper investigation into the mental health status. In addition, our study relied on self-reported questionnaires, and therefore, the results may not be very reliable because of the effects of social desirability and recall bias. However, the fact that both the patient and control groups answered the same questions on mental health provides the study with a greater degree of reliability. Another limitation of our study is that the RP patients were recruited from an online patient association; therefore, their characteristics may differ from those of the overall sample of RP patients in Korea. Association membership may boost feelings of empowerment and perhaps lead to better mental health, lower levels of depression, and better coping and adaptation. Therefore, it is possible that the mental health of the general population of RP patients may have been underestimated. On the other hand, the mental health status of the RP patients may have been overestimated because those who seek support of a patient advocacy group might do so because they have greater levels of stress and depression. In addition, the sample size was not large enough to allow highly sensitive detection of differences between the groups or investigation of the factors associated with poor mental health. However, RP is a rare disease, and our sample size was larger than that of most previous studies dealing with similar issues.[14] Lastly, because our survey was conducted online rather than by a face-to-face interview, access to and the ability to use the Internet may have skewed recruitment toward younger and computer-competent participants.

Despite these limitations, our study has important implications for both clinical practice and research. Patients with RP report greater levels of stress, depression, and suicidal thoughts than the general population. Therefore, it is important for practicing general physicians, low-vision rehabilitation practitioners, optometrists, and ophthalmologists to be aware that RP patients are at risk of significant mental health problems. These health care workers should be encouraged to screen for and promptly diagnose these problems and refer patients to appropriate mental health professionals and services. In addition, as this study was conducted in Korea, a country with a relatively low prevalence of RP, it may be necessary to conduct mental health research in RP patients in other countries using a better validated questionnaire.

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APPENDIX

The appendix is available online at http://links.lww.com/OPX/A119.

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