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Original Article

Outcome of Severe Obsessive-compulsive Disorder With Schizotypal Features: A Pilot Study

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Background/Purpose: Long-term outcome of patients with severe obsessive–compulsive disorder (OCD) and schizotypal features has been rarely studied. We investigated this issue in this retrospective pilot study. **Methods**: Twenty-two patients with severe OCD and schizotypal features were identified by chart review. Another 22 OCD patients without schizotypal features (OCD-NS) served as the comparison group. Those with schizotypal features must not fulfill a diagnosis of schizophrenia or schizotypal disorder. After an average follow-up of 6.6 years, each patient received a re-diagnosis clinical interview. Relevant demographic and clinical data were collected. Patients with schizotypal features were classified into two groups after re-diagnosis: those with schizophrenia or schizotypal disorder (OCD-SS group, n=9) and those with only schizotypal traits (OCD-ST group, n=13) that did not fulfill a well-formed schizophrenia-spectrum disorder. Demographic data, family history, clinical symptoms, and OCD course were compared among the three patient groups.

Results: Compared with the OCD-NS group, the OCD-SS group was significantly less educated, less likely to be married or female, and had earlier onset of illness and poorer OCD course (p < 0.05). There was no significant difference in any demographic and clinical variables between the OCD-SS and OCD-ST groups except that the OCD-ST group had a significantly better OCD course (p < 0.01).

Conclusion: The findings suggest that a substantial proportion of the patients with severe OCD and schizotypal features evolve into schizophrenia spectrum disorder and are associated with a poor long-term outcome, whereas the OCD-NS group might stay with limited manifestations of schizotypal features and have a better outcome.

Key Words: long-term outcome, obsessive–compulsive disorder, psychotic disorder, schizophrenia, schizotypal disorder

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It is now recognized that obsessive–compulsive disorder (OCD) affects approximately 2–3% of the world's population, and it is a chronic and often disabling mental disorder. OCD has heterogeneous manifestations and can be associated with schizotypal features. The estimated prevalence of schizotypal features in patients with OCD ranges from 0 to 50%, depending on various definitions and measurement tools, as reviewed by Poyurovsky et al. For example, in a large series of 475 OCD patients, 4% had concomitant schizotypal personality disorder. Another study found that 50% of 119 OCD patients had positive schizotypal symptoms.

Previous studies have shown that schizotypal features in OCD are associated with early onset of illness, comorbid diagnoses, special types of obsessive-compulsive symptoms (OCSs),⁵ increased impairment of cognitive function,⁶ and reduced gray matter volume.⁷ Several reports have demonstrated that the presence of schizotypal personality disorder (SPD) predicts a poorer response to drug or behavior therapy.⁸⁻¹¹ Furthermore, SPD has been found to be more common in OCD patients whose insight remains poor even after treatment.¹²

In the era of the International Classification of Diseases 9th edition (ICD-9), some OCD patients with obvious schizotypal features, such as concomitant eccentric behavior, anomalies of affect, or inconsequent personality, were diagnosed with latent schizophrenia.¹³ In severe cases of OCD, insight can become tenuous as obsessions progress to overvalued ideas or delusions, which has prompted the special diagnostic specifier in the Diagnostic and Statistical Manual of Mental Disorders 4th edition of OCD with poor insight.¹⁴ In daily practice, there is a subgroup of OCD patients who have severe OCSs as well as concomitant schizotypal features. Little is known about the long-term course and outcome of these patients. In fact, there has been sparse research into the long-term outcome of OCD with schizotypal features, as demonstrated by the limited number of reviews of OCD outcome.¹⁵ Given the paucity of literature in this area, we conducted a pilot study

to investigate the long-term outcome of severe OCD with schizotypal features. We aimed to trace the course and outcome of severe OCD with schizotypal features, and identify relevant clinical correlates associated with different outcome.

Methods

Subjects and design

To study severe OCD with schizotypal features, we retrospectively reviewed the admission records (1986–1996) of the National Taiwan University Hospital and collected data on patients with severe OCD with schizotypal features. We only selected patients who fulfilled three inclusion criteria: (1) severe OCSs as defined by a Yale-Brown Obsessive-Compulsive Scale (Y-BOCS)^{16,17} score of ≥24, which usually necessitates hospitalization; (2) 1-3 of the nine symptom criteria of the ICD-10 schizotypal disorder¹⁸ for at least 3 months before receiving any antipsychotic treatment, yet not severe enough to fulfill a diagnosis of schizophrenia or schizotypal disorder; and (3) an OCD course of at least 6 months, that is, not an acute transient episode. The retrospective rating of the Y-BOCS was done by two psychiatrists (T.J. Hwang and Y.J. Lee) simultaneously to reach a consensus. If there was any doubt, the patient was excluded. Exclusion criteria included the presence of mental retardation, schizophrenia, schizoaffective disorder, persistent delusional disorder, a history of head injury, or active diagnosis of substance abuse or dependence. A total of 27 patients were found. As a result of severe clinical symptoms and functional impairment, all the patients had undergone vigorous pharmacotherapy (including antiobsessive and antipsychotic drugs for at least 3 months), and supportive psychotherapy. Some had also received cognitivebehavioral therapy or electroconvulsive therapy for refractory symptoms. For comparison, we selected another 22 OCD patients during the same period as the control group. The inclusion criteria were: (1) a confirmed diagnosis of OCD; and (2) no significant concomitant schizotypal features.

These patients also received retrospective rating of the Y-BOCS.

The patients were contacted to participate in follow-up interviews at an average of 6.6 years after they fulfilled the enrollment criteria. Of the 27 patients with severe OCD and schizotypal features, three were not contactable, and two refused to participate in the study. The remaining 22 patients with severe OCD and schizotypal features and the other 22 pure OCD patients agreed to join the study and signed an informed consent statement after the detailed procedure was explained to them. To confirm the current clinical status and long-term course of our sample, all 44 subjects received a re-diagnosis clinical interview, based on ICD-10 criteria, to determine whether schizophrenia or schizotypal disorder occurred during the follow-up period. To focus on the evolution of schizotypal features, we did not assess other concomitant depressive or anxiety symptoms that usually fluctuated during the course. The clinical interview was done by a psychiatrist (T.J. Hwang) to ensure proper detection of nuances of behavior and history that were necessary for the correct diagnosis. The results were discussed with another senior psychiatrist (Y.J. Lee) to reach a final decision based on their consensus. For a diagnosis of ICD-10 schizotypal disorder, at least four of the nine criteria must have persisted for 2 years. Those who fulfilled ≤ 3 criteria were classified as only having schizotypal traits. The interview also recorded demographics, family history, OCSs, and other clinical data. Measurement of socioeconomic status (SES) was based on the Hollingshead Index.¹⁹ The Hollingshead Index ranges from 1 (families of wealth, graduate education, top-rank social prestige) to 5 (unskilled and semi-skilled workers, elementary education). Subjects who had a Hollingshead Index score of 4 or 5 were classified as low SES.

At the end of follow-up, out of 22 patients with severe OCD and schizotypal features, four met the ICD-10 criteria for schizophrenia, five met the ICD-10 criteria for schizotypal disorder, and 13 met the criteria for schizotypal traits after a re-diagnosis procedure. Thus, the first nine patients formed

the schizophrenia spectrum disorder group (OCD-SS), and the latter 13 patients with schizotypal traits formed the OCD-ST group. Of the 22 patients with an initial diagnosis of pure OCD without schizotypal features, none developed significant schizotypal features during follow-up; therefore, they formed the non-schizotypal group (OCD-NS).

Classification of symptoms and outcome

The OCSs were classified into six subgroups: contamination/washing/avoidance; doubt/checking; obsessive thought without compulsion; symmetry/precision with compulsive slowness; mixed (at least two of the above-mentioned OCS subgroups); and other (such as peculiar sexual obsessions and rituals). Degree of insight was assessed using the insight question of the Y-BOCS. The insight question of the Y-BOCS was rated on a 0-4-point scale that represented "excellent insight" (0) to "lacks insight, delusional" (4). Subjects who had a score raging of 3 or 4 were considered to have "poor insight". The course of OCD was based on previous research⁴ and defined as follows: (1) deteriorative with no remission: gradual functional deterioration with or without OCS; (2) continuous with no remission: maintaining similar level of OCS severity and function, without improvement or deterioration; (3) waxing and waning course: periods of partial remission of OCSs and function for > 6 months; or (4) episodic course: periods of complete remission of OCSs and function > 6 months. Subjects who had a waxing and waning course or episodic course were considered to have a "fair course".

Data analysis

We compared the demographic data, psychopathology (insight and OCSs), and outcome variables (OCD course) among the three groups of patients: OCD-NS, OCD-SS, and OCD-ST. Oneway analysis of variance was conducted on continuous variables, and the χ^2 (or Fisher's exact) test was performed on categorical variables. When there was a significant difference (p<0.05) of a continuous variable, the Tukey honestly significant difference or Games–Howell test was adopted for

post hoc multiple pairwise comparisons depending on test of homogeneity of variances. Rates and proportion with a two-tailed 95% confidence interval were calculated for the variables of interest. All data were analyzed using SPSS version 10.0 (SPSS Inc., Chicago, IL, USA).

Results

Nine (41%) of the 22 patients with severe OCD and schizotypal features developed schizotypal disorder or schizophrenia during the follow-up period. The mean time to conversion was 3.1 years. Table 1 compares demographic profiles, clinical correlates and family history among the three groups. Compared with the OCD-NS group, the OCD-SS group was significantly younger, less educated, less likely to be married or female, and had earlier onset of illness (p<0.05). Compared with the OCD-NS group, the OCD-ST group was also significantly younger and less likely to be married (p<0.05). By contrast, there was no significant difference on any demographic variable between

the OCD-SS and OCD-ST groups. The baseline Y-BOCS score of the OCD-SS and OCD-ST groups was significantly higher than that of the OCD-NS group, but there were no significant differences in the follow-up Y-BOCS scores among the three groups. No significant differences among the three groups were found for duration of illness, years of follow-up, SES, family history of OCD or psychotic disorders.

There was no significant difference in OCSs among the three OCD groups (Table 2). The OCD-SS and OCD-ST groups tended to have more "other symptoms" than the OCD-NS group, and there was a borderline significance among the three groups (p = 0.09). Those with other symptoms included two patients in the OCD-NS group: impulsive staring/compulsive avoidance of male genital organs, ritualized bathing and eating behaviors; three in the OCD-SS group: pervasive contamination obsessions with undoing behaviors and magical thinking, counting compulsions, pervasive undoing thoughts and behaviors with magic thinking; and five in the OCD-ST group: pervasive pathological doubting, pervasive religious obsessions

Table 1. Comparison of demographic data, clinical correlates, and family history in three different obsessive—compulsive disorder groups^a

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	OCD-NS (n=22)	OCD-SS (n = 9)	OCD-ST (n=13)	χ^2 or F
Demographic data and clinical correlates				
Age (yr)	35.5 ± 13.2	23.5 ± 4.0^b	25.0 ± 5.9^{b}	6.66 ^c
Age of onset (yr)	24.0 ± 12.8	15.1 ± 2.8	16.3 ± 6.3	3.79 ^b
Duration of illness (yr)	11.5 ± 9.1	8.5 ± 3.6	8.7 ± 5.3	0.85
Years of follow-up (yr)	7.4 ± 6.5	6.3 ± 2.3	5.7 ± 2.9	0.51
Male sex	9 (40.9)	8 (88.9) ^d	8 (61.5)	6.16 ^e
Married	10 (45.5)	0 (0) ^d	1 (7.7) ^d	9.99 ^c
College education	12 (54.5)	0 (0) ^b	3 (23.1)	9.45 ^c
Low SES	6 (27.3)	6 (66.7)	6 (46.2)	4.31
Baseline Y-BOCS	17.6 (5.7)	26.9 (5.4) ^b	26.3 (4.7) ^b	15.3 ^c
Follow-up Y-BOCS	8.5 (4.9)	13.6 (10.4)	9.7 (6.5)	1.8
Family history				
OCD	3 (13.6)	0 (0)	2 (15.4)	1.48
Psychotic disorders	3 (13.6)	3 (33.3)	3 (23.1)	1.60

^aData presented as mean \pm standard deviation or n (%); ${}^bp < 0.01$: versus OCD-NS; ${}^cp < 0.01$: (two-tailed) for comparison among the three groups; ${}^dp < 0.05$: versus OCD-NS; ${}^ep < 0.05$. OCD= obsessive-compulsive disorder; OCD-NS=OCD with no schizotypal features; OCD-SS=OCD with schizophrenia-spectrum disorders (schizophrenia or schizotypal disorder); OCD-ST=OCD with schizotypal traits; SES= socioeconomic status; Y-BOCS= Yale-Brown Obsessive-Compulsive Scale.

Table 2. Comparison of symptoms and course in different obsessive-compulsive disorder groups^a χ^2 or F OCD-NS (n=22)OCD-SS (n=9)OCD-ST (n=13)OCS subgroup 9.48 Contamination/washing 5 (22.7) 1 (11.1) 1 (7.7) 0(0)Doubt/checking 2(9.1)3(23.1)Obsessive thought 3 (13.6) 2 (22.2) 2 (15.4) Symmetry/precision 1 (4.5) 1 (11.1) 0(0)Mixed 9 (40.9) 2(22.2)2(15.4)Other 2 (9.1) 3 (33.3) 5 (38.5) Insight Poor insight 4 (44.4) 9 (69.2)^c 11.23^b 3 (13.6) 23.47^b OCD course 5 (55.6)^d Deteriorative 1 (4.5) 2 (15.4) Continuous 4 (44.4) 0(0)3 (13.6) Waxing and waning 12 (54.5) 0(0)7 (53.8) 4 (30.8) **Episodic** 6 (27.3) 0(0)

^aData presented as n (%); bp <0.01: (two-tailed) for comparison among the 3 groups; cp <0.01: OCD-ST vs. OCD-NS; dp <0.01: OCD-SS vs. OCD-NS; OCD-SS vs. OCD-ST. OCD=obsessive—compulsive disorder; OCD-NS=OCD with no schizotypal features; OCD-SS=OCD with schizophrenia-spectrum disorders (schizophrenia or schizotypal disorder); OCD-ST=OCD with schizotypal traits; OCS=obsessive—compulsive symptom.

with undoing behaviors, dysmorphophobia, dysmorphophobia + needs to ask/touch, peculiar sexual obsessions + AIDS phobia + pervasive pathological doubting. Compared with the OCD-NS group, the OCD-ST group was significantly less likely to have better insight. With respect to the outcome measures, the OCD course most commonly found was waxing and waning in the OCD-NS and the OCD-ST group, and deteriorative without remission in the OCD-SS group (Table 2). Compared with the other two groups, the OCD-SS group was significantly more likely to have a deteriorative course of illness (p < 0.01). By contrast, there was no significant difference between the OCD-ST and OCD-NS groups for the distribution of OCD course. When episodic and waxing and waning courses were combined into a fair course, the OCD-SS group was significantly less likely to have a fair course (OCD-SS, 0%; OCD-NS, 81.8%; OCD-ST, 84.6%; $\chi^2 = 21.9$, p < 0.001).

Discussion

Our pilot study had two main findings. First, a substantial proportion (41%) of the patients

with severe OCD and schizotypal features progressed to full-blown schizophrenia or schizotypal disorder. Second, although the OCD-ST group was similar to the OCD-SS group in terms of age, age of onset, marital status, and baseline Y-BOCS score, the long-term outcome might not have been as poor as that of the OCD-SS group.

The presence of schizotypal features is a risk factor for development of schizophrenia or schizotypal disorder, as demonstrated by the early psychosis projects. 20,21 Those with some schizotypal or quasi-psychotic symptoms were at greater risk of developing into schizophrenia or schizotypal disorder when compared with those without. Compared with OCD patients without schizotypal features, OCD patients with schizotypal features have been shown to have more severe reduction of gray mater volume⁷ and frontal cognitive dysfunction. 6 These findings are in line with the results of studies in at-risk subjects who had frontal cognitive dysfunction and gray matter reduction during development towards schizophrenia.²² In addition, severe OCSs might be associated with poor outcome and presence of SPD. 10,23 Taken together, these factors can explain why a higher proportion of our patients with severe OCD and

schizotypal features evolved into schizophrenia spectrum disorder.

Previous family and twin studies have shown that OCD is a familial disorder,²⁴ and that OCD and schizophrenia can be inherited together but with different severity of schizotypal symptoms and outcome.²⁵ Our study found that the OCD-ST and OCD-SS groups were similar in age of onset, marital status, and baseline Y-BOCS score but different in long-term outcome. One explanation for this is related to the concept of "schizotaxia", ²⁶ which suggests that the genetic liability for schizophrenia could be manifested, even without the full manifestations of schizophrenia. This liability is characterized by neurological, neurobiological, psychiatric, neuropsychological, and psychosocial impairments, in non-psychotic first-degree relatives of people with schizophrenia.²⁷ By modification of the concept of schizotaxia, Tsuang and colleagues have proposed that the predisposition to develop schizophrenia in non-prodromal, nonpsychotic adult relatives of patients with schizophrenia might be expressed as a meaningful, diagnosable, clinical syndrome or set of traits.²⁸ Moreover, there is evidence that the schizotaxic subjects might have a positive response following treatment with low-dose antipsychotics.²⁸ Given that our non-schizophrenic OCD-ST group exhibited substantial rates of a family history of psychotic disorders (23.1%), poor social-occupational functioning, poor insight, more unusual OCSs, and a positive response to treatment, this group of individuals might represent schizotaxic characteristics to some extent. This could have accounted for their better outcome after treatment. It would be interesting to establish baseline factors among these OCD patients with schizotypal features that could predict future development into the OCD-SS or OCD-ST group. Unfortunately, our study could not find any significant predictor since there was no significant difference in the baseline demographic data and clinical correlates between the two groups. Clearly, further studies with a larger sample size and prospective design will be needed to elucidate the real nature of these patients.

Our results were in accordance with previous findings that OCD patients with psychotic features are more likely to be single and have a deteriorative course, 4 at least in the OCD-SS group. The symptoms used to define OCD are diverse and include a range of obsessions and compulsions. Some authors have described the increased rate of a certain type of OCSs subgroup (e.g. counting compulsions) in OCD patients with psychotic features.⁵ Our data showed no significant difference in the OCSs subgroup of the three OCD groups. The OCD-SS and OCD-ST groups tended to have more other symptoms than the OCD-NS group (p =0.09). This could have been due to the coexistence of OCD and schizotypal features in the former two groups, which made the manifestation of OCSs become more unusual.

A generalization of our results must be viewed with caution, considering the retrospective and observational nature of the analysis and the small sample size. Given the limited number of OCD patients with and without schizotypal features, there was possible selection bias when enrolling the study sample. The retrospective review of baseline OCD severity and schizotypal features might not be sufficiently accurate, although this could be less problematic for the OCD-SS and OCD-ST groups because > 90% of these patients were admitted with detailed records of their clinical manifestations. The outcome measure (OCD course) was clinical rather than scale-based. The medications were not well-controlled due to the retrospective and observational study design, although the patients with severe OCD and schizotypal features always received vigorous treatment. It is possible that some patients' schizotypal features were masked by antipsychotics, so they were classified in the OCD-ST rather than OCD-SS group. Another limitation is that we did not assess other comorbid disorders such as anxiety, depressive, or personality disorders. The baseline Y-BOCS score was not comparable between the OCD-NS and the other two groups; therefore, it could have been a confounder of outcome measurement. However, past studies have already shown that OCD patients with schizotypal features have poorer outcome than those without such features, even when both groups have similar severity of OCSs.^{8,11,29} The value of our study is to demonstrate the different outcome between the OCD-SS and OCD-ST groups. Clearly, further research needs to be undertaken with a larger sample size, standardized scales, and a prospective, systematic design to clarify the subtyping and clinical outcome of severe OCD with schizotypal features.

In summary, our study identifies the clinical course and outcome of severe OCD with schizotypal features. Our results suggest that a substantial proportion of these patients evolve into schizophrenia spectrum disorder and are associated with poor long-term outcome, whereas other patients might stay with limited manifestations of schizotypal features and have a better outcome following vigorous treatment.

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