Examining the hikikomori syndrome in a French sample of hospitalized adolescents with severe social withdrawal and school refusal behavior

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Abstract

While the term hikikomori (HKM) has spread internationally to describe a chronic and severe form of social withdrawal, its place in current nosography and its transposition into non-Asian cultures are still debated. A retrospective chart review was conducted to determine the rate and the clinical profiles of HKM among a French sample of adolescent inpatients. Data were obtained from 191 adolescents aged 12–18 years (M = 15.0, 44% boys) consecutively admitted in two inpatient units from January 2017 to December 2019. Using a retrospective diagnosis of HKM based on Teo and Gaw's criteria, we compared socio-demographic characteristics, clinical features, and treatment outcomes between HKM patients and those with other forms of social withdrawal and/or school refusal (SW/SR). At admission, 7% of participants met HKM criteria (n = 14, M = 14.3, 64% boys), one out of six adolescents with SW/SR. Among those with SW/SR, HKM + vs. HKM- participants had higher rates of anxiety disorder (Odd Ratio, OR = 35.2) and lower disruptive behavioral disorder (OR = 0.03). A minority of the participants with anxiety and depressive disorders met the HKM criteria (respectively, 15% and 9%), but those with HKM had a longer duration of symptoms, longer hospitalization, and required more daily care facilities at discharge compared to HKM-. While HKM syndrome could not be delimitated from anxiety disorder, it was associated with specific clinical features and treatment outcomes. The clinical characteristics observed were consistent with the features reported in Asian HKM adults, supporting face validity of this clinical concept in adolescent inpatients with different cultural contexts.

Keywords

adolescent, anxiety disorder, hikikomori, hospitalization, social withdrawal

Introduction

Social withdrawal and school refusal (SW/SR) are among the most frequent reasons for referral in child and adolescent mental health services (Rubin et al., 2009). A wide range of individual and environmental factors contribute to SW/SR (Barzeva et al., 2019), including co-occurring psychiatric disorders. There are several ways in which cultural factors can affect the course of SW/SR and associated psychiatric disorders: (a) some family risk factors for SW/ SR are shaped by cultural heritages, e.g., the parental level of expectations for a child's academic achievement, (b) cultural factors could influence the parental capacity to seek treatment from mental health professionals for a youth with school-related problems, (c) the level of adherence to the most commonly provided treatments for SW/ ¹Child and Adolescent Psychopathology Services, Amiens University Hospital, Amiens, France

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SR may vary according to culturally specific values (Chen, 2015; Franceschini-Mandel et al., 2018). This research aimed to explore the clinical utility of the hikikomori (HKM) syndrome, initially described in Japanese adults, when applied to adolescents from non-Asian cultural backgrounds. For this purpose, we determined whether using definition criteria for HKM among a French sample of adolescents hospitalized for SW/SR identified a group of patients with a specific form of distress and impairment not captured by commonly diagnosed psychiatric disorders.

Initially described in Japan by Tamaki Saito (1998), the HKM syndrome is a particularly severe and chronic type of social withdrawal lasting for months, usually years. Individuals with HKM spend all their time at home avoiding any direct inter-personal situations with same-age peers in academic/professional domains, and sometimes with other family members (Kaneko, 2006; Saito, 1998). An expert group operationalized criteria for HKM (Teo & Gaw, 2010): (1) spending most of the time at home; (2) no interest in going to school or working; (3) persistence of withdrawal for more than six months; (4) exclusion of previously diagnosed schizophrenia, mental retardation, and bipolar disorder; and (5) exclusion of those who maintain personal relationships (e.g., friendships). The specificities of the social systems inherited from Confucianism, enhancing collective rather than individualistic values (e.g., humility or social cohesion), are frequently mentioned as cultural factors involved in the emergence of HKM in East-Asian countries. The HKM syndrome has been viewed as an extreme form of disengagement from conventional social roles and social expectations in response to painful emotions ("makeinu") (Norasakkunkit & Uchida, 2014; Toivonen et al., 2011). In this respect, the syndrome has been placed at the end of a clinical spectrum encompassing less severe forms of social withdrawal in Japan such as the "taijin-kvofu" (phobia of the interpersonal situation) or the student apathy (Suzuki, 2020).

The operationalizing of consensual definition criteria for HKM has helped to develop a clinical research framework in East-Asian countries (Wong et al., 2019). The life prevalence of HKM in adults would be about 1% in Japan, with comparable frequencies in nearby regions such as Hong Kong and Korea (Koyama et al., 2010; Wong et al., 2015). Revised guidelines were published in 2010 by the Japanese Ministry of Health, Labour and Welfare (2010).

While the term HKM has spread internationally in common language, its place in current nosography and its transposition into non-Asian cultures are still debated. Over the last decade, cases have been reported in North-American and European countries such as Spain (Malagon-Amor et al., 2015), Italy (De Michele et al., 2013), and France (Chauliac et al., 2015; Guedj, 2017). While some argue that the HKM syndrome exists across all cultures, in particular in modern globalized urban areas

(Kato et al., 2012), to date this assumption has received little scrutiny. Previously, case reports in non-Asian cultures stressed the very high rate of psychiatric disorders in HKM, questioning its relevance as a distinct clinical syndrome in non-Asian cultures.¹ It is also not clear how cultural factors specific to the social and family organizations in Asian countries can contribute to the HKM syndrome in other countries.

In France, the cultural factors contributing to the growing feeling of social isolation in patients with social withdrawal / hikikomori are rarely mentioned (De Luca, 2020). In their historical article entitled "Le confinement à domicile" (Home confinement), Gavral et al. (1953) used a phenomenological approach to describe the psychological mechanisms involved. The progressive retreat from all social activities was viewed as a defense mechanism against a "Self/World" or "Self/Society" conflict which results in the patient living in a shrunken and enclosed area, basically his/her room. Thereafter, authors have focused more on the family factors that maintain the patient's passive behavior through ill-adapted or overadaptive attitudes (De Luca, 2020; Jeammet, 1985),² although sociocultural risk factors for the withdrawal were not really questioned. More recently, the term "invisible social" (socially hidden) has been used in France for youths with social withdrawal / hikikomori based on the model of the epidemiological notion of NEET ("Neither in Employment nor in Education or Training").³ The French notion of "syndrome d'auto-exclusion" (self-exclusion syndrome) has been used to describe the interplay between the patient's perceived feeling of social isolation and his/her difficulties in seeking treatment, care, or support, in particular in adults suffering from mental health conditions and precarious social situations (Furtos, 1999).⁴

Essentially, three conceptual models have been proposed to characterize the place of HKM in current psychiatric classifications (Table 1). The proponents of a "strictly medical" approach view the HKM syndrome as an integral component of a preexisting psychiatric disorder (e.g., social phobia). Patients with HKM are seen as suffering from a particularly severe form of anxiety disorder, with predominant avoidance behaviors.⁵ The proponents of a "sociological" approach suggest that HKM is as maladaptive reaction of distress in young adults facing social phenomenon and cultural transition. The co-occurrence of psychiatric disorders in these youths is regarded to be purely contingent. For these authors, the interventions should focus on addressing the economic and social challenges facing these youths rather than medicalizing a so-called psychiatric syndrome. Finally, the proponents of a "medical-anthropological" approach view the HKM syndrome and psychopathology as dissociable, but ultimately correlated, dimensions. These authors noted a relative homogeneity in the risk factors, the clinical course, and the specificity of psycho-social treatments supporting the

	Medical	Sociological	Medical-anthropological	
Main view	The syndrome is only a fashionable and exotic relabeling of a preexisting psychiatric disorder, in particular schizophrenia, autistic syndrome, social phobia, and personality disorder.	HKM is a social phenomenon. It represents an extreme form of disengagement from society in response to an individual's painful experience.	HKM describes a specific constellation of family and social factors associated with atypical expression of psychiatric symptoms (such as anxiety and depression).	
Expected findings in empirical studies on associated diagnoses	All HKM subjects should be diagnosed with a psychiatric disorder.	HKM is not a source of distress by itself. Subjects may actually feel content in the initial stage of social withdrawal. Some subjects have pure HKM without co-occurring psychiatric disorder.	While a psychiatric disorder is frequently associated, it rarely accounts for the high level of functional impairment and symptom duration.	
Expected findings in empirical studies on etiological factors	Etiological factors and clinical features should not differ between patients with a psychiatric disorder with and without HKM.	Isolation and psychiatric disorders may share common family and socially mediated risk factors.	Etiological factors (for the psychiatric disorder) and maintenance factors (for the HKM process) should be distinguished.	
Expected findings in empirical studies on clinical course	The onset of the psychiatric disorder should systematically precede the isolation.	Social isolation and psychiatric disorders are not interweaved.	Social isolation precedes the onset of an associated psychiatric disorder. The HKM is a prognosis/moderator factor for the clinical course of the psychiatric symptoms.	
Expected findings in empirical studies on therapeutics	cal studies psychiatric disorder should are not effective, and can lead to		Combined interventions (including pee and family interventions) are needed, providing only treatment for the co-occurring psychiatric disorder lead to a high risk of relapse.	
Implication for psychiatric classification	HKM is an integral component of a preexisting psychiatric disorder. Making space for HKM in classification can hide underying treatable psychiatric disorders.	A distinction should be made between authentic/primary and secondary "pseudo-HKM" where chronic social withdrawal is due to psychiatric symptoms.	HKM is a cultural concept of distress ^a that affects the natural course of associated symptoms.	

Table I. Conceptual models to explai	the overlap between	n HKM and psychiatric disorders
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Note. ^a The status of HKM as a cultural-bound syndrome or a global phenomenon is still debated. Several authors stress the importance of considering the globalization of many sociocultural factors suspected to play a role in the emergence of HKM, e.g., urbanization, breakdown of social cohesion, downward social mobility, or technological progress. In line with this, some have argued that the HKM could be included in the new category of "cultural concepts of distress" in the DSM-5, stressing the importance of cultural factors in shaping the way these subjects communicate individual distress without implying cultural exclusivity (American Psychiatric Association, 2013).

utility of the notion of HKM, whether a psychiatric disorder co-occurs or not.

It is not so surprising that the term HKM has been applied to adolescents, since the first clinical expression of this syndrome frequently occurs at this age (Koyama et al., 2010; Teo, 2010). In line with the observation made by Saito (1998) of a "never-ending adolescence," clinicians from various theoretical backgrounds noted that the psychological mechanisms involved in the withdrawal from the social world in HKM are typical from this life period. This behavior was understood in terms of reactivation of an insecure/anxious attachment (Hattori, 2006), unresolved identity crisis (Guedj, 2017; Jeammet, 1985), maladaptive responses to social expectations (Furlong, 2008), or family dysfunctions (Lee et al., 2013). The relation between the duration of SW/SR and the overall prognosis of this behavior also supports the importance of providing early interventions, i.e., before adulthood, that could alter the course of this syndrome (Malagon-Amor et al., 2018).

However, the use of the term HKM in subjects as young as 15 or even 12 years old, as suggested by Wong et al. (2015), raises several questions. If HKM adolescents represent the most severely impaired patients with SW/SR, without any specific features, what is the need for providing a new label? Adding a new term may be even more confusing for clinicians, patients, and families if it does not cover relevant clinical information.⁶ A direct comparison between patients with HKM and those with other types of SW/SR at these ages could help to determine how specific this syndrome is. In addition, the criteria for HKM were developed in Japanese samples of young adults (Ministry of Health, Labour and Welfare, 2010; Teo & Gaw, 2010) and it is not clear how these criteria might apply to non-Asian samples.

Here, we conducted a retrospective chart review to determine whether a group of adolescents with matching criteria for the HKM syndrome can be identified among a sample of French adolescent inpatients. We aimed to determine the rate of the syndrome in this inpatient sample and how these subjects differ from those with other types of SW/ SR in terms of socio-demographic and clinical characteristics and treatment outcomes. On a methodological ground, caution is however required when interpreting information provided from the direct comparison between a suspected cultural concept of distress and psychiatric disorders in a specific sample (Kohrt et al., 2014). This research can be, therefore, regarded as worthy in providing a preliminary proof of concept for the use of the HKM syndrome in non-Asian countries.

Literature on HKM in adolescents in non-Asian countries stresses the high level of parental burden and family dissatisfaction with mental health care providers in these situations (Guedj, 2017, p. 2019; Malagon-Amor et al., 2015). A mismatch is frequently reported between the responses provided by most outpatient psychiatric services where these patients are referred (expecting that the patient is capable of formulating his/her own request) and the family demands (stressing the patient's passivity and his/her incapacity to ask for any help). This research aims to better understand such discrepancy in order to not limit our mental health care in treating only children and adolescents with typical expression of psychiatric disorder.

Method

Study design and setting

Data from the medical records of all patients hospitalized in the adolescent units of a tertiary care university hospital from January 1, 2017 to December 31, 2018 were extracted (N = 191). The chart review database was initially prospectively developed to examine the relation between substance use and various clinical and therapeutic outcomes in adolescent inpatients. Two researchers extracted information for all consecutive patient admittances based on the available clinical and paramedical chart using a 72-item computerized questionnaire. The assessment of the interrater reliability was based on the analysis of 10 randomly selected files ($\kappa = .80$). The data collected encompassed socio-demographic characteristics, clinical features, symptom severity, developmental history, associated medical conditions, school-functioning, and psychosocial factors. The study was funded by a competent national authority (project IReSP-15-Prevention-11) and received the approval of an ethics committee (Committee for the protection of persons CPP II de France 05/17/2018 ref: 71-2017/ C16-68 / ID RCB 2017-A02952-51). A waiver of consent was allowed in this chart review study due to its retrospective design, patient de-identification, and the use of routine questionnaires.

Participants

The identification of patients with HKM was made in three steps.

Phase 1: Screening for social withdrawal and school refusal. The presence of SW/SR for at least three months was assessed in all participants during the chart review process. Abstractors rated the item based on all information available on the medical charts. Both abstractors were health care professionals, blind to the research hypothesis, with scheduled meetings between the abstractors and the investigator to resolve data conflicts.

Phase 2: Identification of probable HKM. The medical files of all adolescents screened positively for SW/SR were reviewed by a senior psychiatrist directly involved in inpatient care (the first and third authors). A specific instrument based on the French translation of the criteria for HKM, made operational by Teo and Gaw (2010), was used. Adolescents with SW/SR were rated as "probable HKM" if they matched all the following criteria:

- Criteria A: socially withdrawn for at least six months
- Criteria B: avoidance of any interpersonal relations, i.e., at school AND peer relations AND familial relations AND professional, if any
- Criteria C: does not suffer from previously identified psychotic disorder or severe neurodevelopmental disorder (intellectual disability, autistic spectrum disorder, motor disabilities). Social isolation could not result from the lack of adequate educational facilities for patients with physical or mental disabilities.

Phase 3: Confirmation of HKM patient. The medical chart of all subjects previously identified as "probable HKM" was reviewed by another psychiatrist (the second author) with specific clinical expertise in SW/SR, who was not included in the care.

Other measures

The psychiatric diagnoses were based on discharge diagnosis. The level of functioning and symptom severity were assessed using questionnaires routinely used in the care center: the Children-Global Assessment of Functioning scale (C-GAF) (Jones et al., 1995) and the Clinical Global Impressions-Severity scale (CGI-S) (Busner & Targum, 2007). The DEP-ADO questionnaire was used to document substance use in the previous 12 months. The screening question was "During the last 12 months, how often have you [has X] used one of the following substances: alcohol, cannabis, cocaine, inhalant/solvent, stimulant, hallucinogen, or heroin"; examples and trivial names were provided for each substance (Germain et al., 2007).

Statistical analysis

Bivariate analyses were conducted to compare the sociodemographic features, associated psychiatric diagnoses, clinical features, and therapeutic outcomes of HKM patients ("HKM + group") compared to those with SW/SR without HKM ("other SW/SR group"). Then, exploratory analyses were conducted to compare youths with various psychiatric disorders with regards to their HKM status. Since normal distribution was not confirmed for most variables, the nonparametric Mann Whitney test was used to compare the HKM + group and the other SW/SR group for continuous variables and the Fisher exact test for categorical variables. Cramer's ϕ (Phi) correlation coefficient was calculated to determine effect sizes. A Cramer's score (ϕ) between 0.10 and 0.30 was generally considered as a low effect size, between 0.30 and 0.50 as a medium effect size, and above 0.50 as a large effect size (Peacock & Peacock, 2010). The Cliff's Delta score (δ) was used to measure the effect size of the Mann-Whitney tests; the score varied between -1and +1, the higher and stronger the effect size. Analyses were run on R 3.4.0, with a p-value less than 0.05 being considered significant.

Results

Identification of adolescents with HKM

Among the 191 inpatient adolescents consecutively admitted during the study period, 83 were identified as having SW/SR as follows:

- n = 38 adolescents with school refusal without withdrawal from family or peer relations

- n = 7 adolescents with school refusal and withdrawal from peer relations but not from family relations

- n=4 adolescents with intermittent SW/SR over the past three months

- n = 18 adolescents with severe neurodevelopmental disorders, e.g., autism or intellectual disability, staying at home waiting for a place in a special education facility

Finally, 16 adolescents were considered as probable HKM. The status of HKM was not confirmed for two patients due to the lack of information in the medical files, resulting in 14 adolescents with a confirmed HKM (Figure 1).

Comparisons of socio-demographic, clinical features and therapeutic outcomes between HKM patients and those with other forms of SW/SR

Most of the HKM adolescents were male (sex ratio 2:1), with a mean age at 14.29 ± 1.53 . As presented in Table 2, youths with HKM did not significantly differ from youths with other forms of SW/SR in terms of demographic factors, academic performance, or psychosocial factors.

All inpatient adolescents with HKM had a discharge diagnosis of anxiety disorder. Half of them suffered from performance anxiety and 21% from panic disorder (Table 3). Depressive disorders were the second most frequent diagnosis found in youths with HKM. In the majority of cases, the depressive disorder diagnosed was a persistent depressive disorder (8/9). HKM patients were less likely to report any disruptive behavioral disorder. No patient with HKM had a psychotic disorder, however the differences with the control group were not statically different.

The frequency of suicidal thoughts, suicidal attempts, nonsuicidal self-injury, repeated runaway, and substance misuse, and the duration of the current clinical symptom, did not significantly differ between youths with HKM and those with other SW/SR (Table 4). However, youths with HKM were less likely to report chronic irritability-aggressive behaviors and at least two previous inpatient admissions compared to their counterparts. Among youths with HKM, 21% had psychotic symptoms. None of the adolescents with HKM reported at least one use of illicit drugs or alcohol in the last 30 days, compared to 25% of the youths with other SW/SR. The two groups were not statistically different in terms of length of stay, CGI-S and CGI-I scores, or C-GAF scores at admission and at discharge.

Delimitation of HKM and SW/SR from associated psychiatric disorders at discharge

As illustrated in Figure 2, all adolescents with HKM had an internalized disorder. We conducted exploratory analyses to determine to what extent the youths with anxiety or depressive disorders differed with regards to HKM status.

Only 15% of youths with anxiety disorders had an HKM syndrome, although all adolescents with HKM had an anxiety disorder. Compared with adolescents with anxiety disorder but not HKM, patients with HKM tended to stay longer in hospital (respectively, 55 days vs. 96 days, p = .028) and

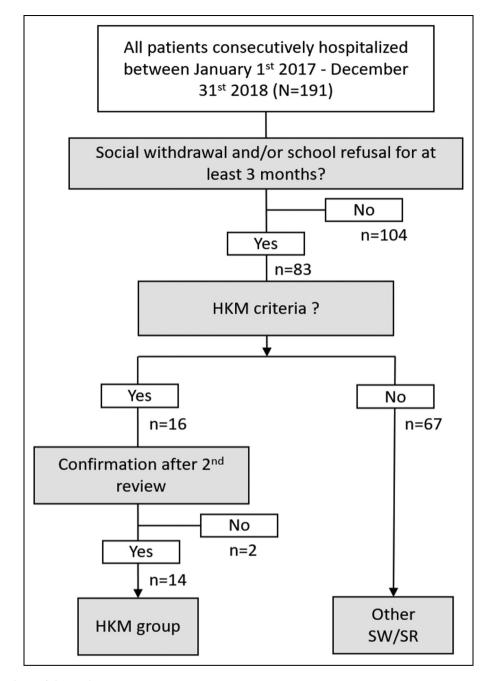


Figure 1. Flow-chart of the study.

had a longer evolution of psychiatric symptoms (respectively, 11.5 months vs. 6 months, p = 0.018). The two groups did not differ in terms of severity level and response to treatment, although youths with HKM were more likely to be discharged in a daily care facility compared to those without HKM (respectively, 57% vs. 10%, p < .001).

Only 9% of adolescents with depressive disorders had an HKM syndrome, whereas 64% of adolescents with HKM had a depressive disorder. The frequency of suicidal thoughts did not differ between youths with HKM and those with a

depressive disorder without HKM (respectively, 50% and 74%, p = .108). However, compared to the depressive group, youths with HKM were less likely to report a lifetime history of suicidal attempt (respectively 42% vs. 14%, p = 0.049) or non-suicidal self-injury (39% vs. 7%, p = .030).

Discussion

This research focused on a special sample of inpatients with severe and chronic forms of psychiatric disorders associated

	Total (N = 191)	HKM + (n = 14)	Other SW/SR $(n = 67)$	p value	Effect size
Demographics					
Sex, female, n (%)	74 (41%)	5 (36%)	41 (62%)	.890 †	$\phi = 0.19$
Age (y) (mean \pm SD)	14.71 ± 1.71	14.29 ± 1.53	15.08 ± 1.73	.572 †	$\delta = -0.10$
Socio-economic status, high and middle, n (%)	138 (72%)	10 (71%)	41 (62%)	.518†	$\phi = 0.08$
Academic performance					
Prior grade repetition, n (%)	40 (21%)	3 (21%)	17 (25%)	I †	$\phi = 0.07$
School adaptations, n (%)	102 (53%)	7 (50%)	42 (62%)	.414 †	$\phi = 0.10$
Special educational facilities (MEI, PETI), n (%)	27 (14%)	0	16 (24%)	.061 †	$\phi = 0.23$
Outpatient psychiatric structure (daily care facilities), n (%)	58 (30%)	8 (57%)	25 (37%)	.170 †	$\dot{\phi} = 0.15$
Psychosocial factors					-
Maltreatment exposure, n (%)	54 (28%)	3 (21%)	21 (32%)	.537 †	$\phi = 0.08$
Foster care placement, n (%)	33 (17%)	2 (14%)	19 (28%)	.501 †	$\dot{\phi} = 0.12$
Home educative assistance / report to child protection agencies, n (%)	85 (44%)	5 (36%)	35 (52%)	.261 †	$\dot{\phi} = 0.12$

	Table 2.	Comparisons of socio-der	ographic features between HKM	patients and those with other forms of SW/SR
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Note. * indicates p < .05. ** indicates p < .01. † Fisher exact test; †† Mann-Whitney test.

Table 3. Comparisons of associated psychiatric diagnoses between HKM patients and those with other forms of SW/SR.

Diagnoses at discharge, %	Total (N = 191)	HKM + (n = 14)	Other SW/SR $(n = 67)$	p value	Effect size
Any anxiety disorders	91 (47%)	14 (100%)	22 (33%)	** [†] 100. >	$\phi = 0.51$
Performance anxiety	54 (28%)	7 (50%)	13 (19%)	.035 [†] *	$\dot{\phi} = 0.27$
Panic disorder	I8 (9%)	3 (21%)	4 (6%)	.092 †	$\dot{\phi} = 0.21$
Any depressive disorder	95 (49%)	9 (64%)	34 (51%)	.354 †	$\dot{\phi} = 0.10$
BD I/II	13 (7%)	I (7%)	6 (9%)	l †	$\dot{\phi} = 0.02$
AD + PTSD	55 (29%)	3 (21%)	19 (28%)	.751 †	$\dot{\phi} = 0.06$
ADHD	33 (17%)	0	11 (17%)	.204 †	$\dot{\phi} = 0.18$
ODD+CD	57 (30%)	0	27 (41%)	.002 [†] **	$\dot{\phi} = 0.32$
Any psychotic disorder	26 (14%)	0	9 (Ì3%)́	.583 †	$\dot{\phi} = 0.16$
Eating disorder	7 (4%)	I (7%)	2 (3%)	.430 †	$\dot{\phi} = 0.08$
OCD	3 (2%)	0	5 (7%)	l †	$\dot{\phi} = 0.12$
ID	23 (12%)	0	11 (16%)	.197 [†]	$\dot{\phi} = 0.18$
ASD	21 (11%)	0	7 (10%)	.596 [†]	$\dot{\phi} = 0.14$
Learning/language disorder	78 (41%)́	8 (62%)	27 (40%)	.161 †	$\dot{\phi} = 0.13$

Note. * indicates p < .05. ** indicates p < .01. † Fisher exact test; †† Mann-Whitney test.

BD: Bipolar disorder; AD: Adjustment Disorder; PTSD: Post-traumatic stress disorder; ADHD: Attention deficit hyperactivity disorder; ODD: Oppositional defiant disorder; CD: Conduct disorder; OCD: Obsessive compulsive disorder; ID: Intellectual disability; BPD: Brief psychotic disorder; ADS: Autistic spectrum disorder, ID.

with SW/SR. While these patients were clearly not representative of all adolescents who may match the definition of HKM in a general population, such a long period of observation represented an opportunity to better understand the needs that should be addressed during their care. Considering its limitations, this research does not provide direct evidence for the validation of the concept of HKM but rather suggests some information about its utility and generates testable hypotheses for future research.

Interpretation of the main findings

The frequency of the HKM syndrome among these adolescent inpatients was 7%. This represented one out of six adolescents

with SW/SR at admission. In East-Asian countries, the prevalence in community-based samples is estimated at approximately 1–2%, although the studies do not differentiate between adolescents and young adults (Koyama et al., 2010; Teo, 2010; Wong et al., 2015). The main socio-demographic features observed in the adolescents identified as HKM in this research were consistent with data reported in young adults, i.e., a predominance of males and patients from various socioeconomic backgrounds (Koyama et al., 2010; Malagon-Amor et al., 2018; Teo, 2010; Wong et al., 2015, 2019; Wu et al., 2019). The mean duration of symptoms in adolescents with HKM was in the same range as reported in the literature, ranging from one to four years (Koyama et al., 2010; Li & Wong, 2015b; Stip et al., 2016; Wong et al., 2019).

	Total (N = 191)	HKM + (n = 14)	Other SW/SR $(n=67)$	þ value	Effect size
Clinical features					
SI	111 (58%)	7 (50%)	38 (57%)	.654 †	$\phi = 0.05$
SA	55 (29%)	2 (14%)	19 (29%)	.334 †	$\dot{\phi} = 0.12$
NNSI	54 (28%)	l (7%)	19 (29%)	.102 †	$\dot{\phi} = 0.19$
Psychotic symptoms	61 (32%)	3 (21%)	18 (27%)	.751 †	$\dot{\phi} = 0.05$
Chronic irritability	67 (35%)	l (7%)	31 (46%)	.006 ^{† **}	$\dot{\phi} = 0.30$
Aggressive behaviors	43 (22%)	0	21 (32%)	.016 ^{† *}	$\dot{\phi} = 0.27$
Repeated runaway	38 (20%)	2 (14%)	15 (23%)	.724 †	$\dot{\phi} = 0.08$
Substance misuse	27 (14%)	0	17 (25%)	.101 †	$\dot{\phi} = 0.24$
Duration of the current clinical symptoms (months) (mean \pm SD)	23.14 ± 0.94	11.5 ± 8.87	12±8.87	. 94 2 ^{††}	$\delta = 0.01$
History of two admissions in inpatient structure, n (%)	55 (28%)	I (7%)	28 (42%)	.013 † *	$\phi = 0.27$
Severity level and treatment outcome					•
CGI-S at admission (mean \pm SD)	4.99 <u>+</u> 1.08	5 <u>+</u> 0.76	5 <u>+</u> 0.93	.632 ††	$\delta = -0.08$
CGI-I at discharge (mean \pm SD)	2.51 ± 0.88	2.5 ± 0.68	2 ± 0.72	.784 ††	$\delta = 0.04$
C-GAF at admission (mean \pm SD)	37.07 <u>+</u> 12.42	35 <u>+</u> 9.36	35 ± 11.09	.927 ††	$\delta = 0.02$
C-GAF at discharge (mean \pm SD)	60.67 <u>+</u> 12.41	60.5 <u>+</u> 5.44	60 ± 10.84	.400 ††	$\delta = 0.14$
Δ C-GAF (mean ± SD)	23.95 <u>+</u> 12.99	25.5 <u>+</u> 11.53	22 <u>+</u> 9.61	.552 ††	$\delta = 0.10$
Length of stay, days (mean \pm SD)	96 ± 101.00	96 <u>+</u> 56.58	71 <u>+</u> 47.29	.294 †	$\delta = 0.18$

Table 4. Comparisons of clinical features, severity level, and treatment outcomes between HKM patients and those with other forms of SW/SR.

Note. * indicates p < .05. ** indicates p < .01. † Fisher exact test; †† Mann-Whitney test.

SI: Suicidal ideation, SA: Suicidal attempt, NNSI: Non-suicidal self-injury, Children-Global Assessment Scale (CGAS); Clinical Global Impressions-Severity (CGI-S); Clinical Global Impressions-Improvement (CGI-I). Aggressive behaviors referred to the use of physical restraints and/or intramuscular PRN medications and/or admission via intensive psychiatric care unit.

Unlike adolescents with other forms of SW/SR, those with HKM tended to present fewer irritability-aggressive behaviors prior to and during their hospitalization. This was in line with the low rate of associated disruptive behavioral disorder found in HKM patients (Koyama et al., 2010; Li & Wong, 2015b; Wong et al., 2015). Case reports and qualitative studies reported that aggressive behaviors in HKM patients were rare, generally occurring in response to acute stressors or frustration (e.g., during family attempts to discuss the patient's behavior or to encourage them to seek treatment) (Chong & Chan, 2012; Furlong, 2008; Guedj, 2017; Hattori, 2006; Lee et al., 2013; Stip et al., 2016). A low level of anger-aggression and a restriction of their emotional expression have instead been described as a clinical landmark of HKM (Li & Wong, 2015b; Masataka, 2002).

Based on previous reports in clinical patients in Europe (Chauliac et al., 2015; De Luca, 2020; De Michele et al., 2013; Guedj, 2017; Malagon-Amor et al., 2018), it was hypothesized that a substantial proportion of these adolescents with HKM would have a diagnosis of schizophrenia. Our findings contradicted this hypothesis, as none of the adolescents with HKM had a discharge diagnosis of a psychotic disorder. Thus, a known diagnosis of schizophrenia was an exclusion criterion for the status of HKM in this study, in line with expert guidelines (Ministry of Health, Labour and Welfare, 2010; Teo & Gaw, 2010; Wong et al., 2019). Even without applying such exclusion criteria, we did not find any such adolescents. However, many of the patients were not "naïve" from previous clinical psychiatric assessment, as most of them had been addressed after a second or third line of outpatient and inpatient treatments.

We found that 20% of youths with HKM had psychotic symptoms before or during the hospitalization. In our personal experience, such psychosis symptoms did not systematically require pharmacological treatment and decreased as long as the patients were re-socialized, unlike patients with an early-onset schizophrenia. Authors such as Stip et al. (2016) or Guedj (2017) remind us that sensory deprivation in patients with social withdrawal may be accompanied by acute and transitory psychotic symptoms. The crosssectional nature of the data precludes us from empirically testing this assumption.

We found that all adolescents with HKM had an anxiety disorder and half of them a depressive disorder. The association between HKM, anxiety, and depressive symptoms is the object of a large consensus (Chauliac et al., 2015; Chong & Chan, 2012; Koyama et al., 2010; Lee et al., 2013; Malagon-Amor et al., 2018; Masataka, 2002; Stip et al., 2016; Teo & Gaw, 2010; Wong et al., 2015, 2019). However, it is not clear whether such internalized symptoms pre-exist the social withdrawal. However, even when an HKM patient has an associated psychiatric

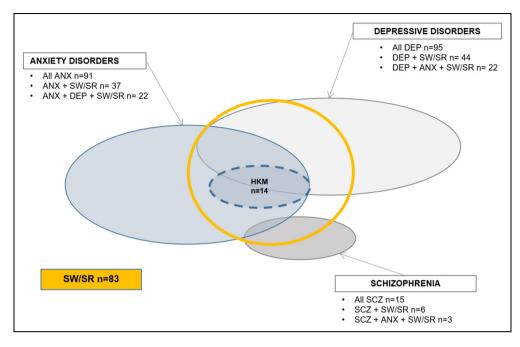


Figure 2. Delimitation of HKM and SW/SR from psychiatric disorders at discharge.

Note. ANX=Anxiety disorders, SW/SR=Social withdrawal and/or school refusal, DEP = Depressive disorders, SCZ = Schizophrenic disorder and other related psychotic disorders.

disorder (e.g., anxiety, depressive, autism spectrum disorders), the severity of clinical symptoms was not necessarily correlated with the severity or the duration of the social withdrawal (Li & Wong, 2015b).⁷

Clinical experiences in caring for adolescents with HKM

While essentially subjective, few comments could be made about our clinical experiences in caring with these 14 adolescents with HKM and SW/SR.

Cultural aspects of HKM. Most of the adolescent inpatients with HKM did not have an Asian ethnic origin (12/14). One patient was born in Japan and raised in South Korea until the age of 13; another was born in France with a deceased father from the Philippines. Some of these youths had a special interest in East-Asian culture, such as K-pop music or manga, but no more than other patients. When an adolescent with HKM had a specific interest in some aspects of Asian culture, this was never shared by other family members. In addition to French culture, the family might have other ethnic backgrounds (e.g., Spain, Italy, Maghreb countries, Sub-Saharan Africa), reflecting the diversity of the French population. We did not observe here the predominance of one culture or another.

Family functioning. Considering the retrospective design of this study, no standardized assessment of family functioning was provided. The weekly family interviews conducted during the hospitalizations were an opportunity to gain more knowledge on aspects of the family dynamic that may contribute to the natural course of the social withdrawal. Two characteristics of family functioning were systematically observed: an avoidance of emotional expression within the family and a strong feeling of isolation shared by all family members.

Parents commonly reported a constant fear of their adolescent's reactions in response to the slightest frustrations. Uncertainty and negative anticipation led such families to progressively avoid any potentially conflicting situation or discussion. In this respect, parents as well as siblings seemed to have got used to not overtly expressing their feelings at home, which in turn led many of them to express a sense of resignation or being incompetent as parents.⁸

All of the parents of HKM adolescents described a progressive reduction of the family and extra-family networks. The adolescent's difficulties were frequently hidden from distant family members, increasing the parental feeling of loneliness. The progressive withdrawal of all family members from their usual social network seemed to mirror the social withdrawal of the patient, a process elsewhere called a "double HKM" (Guedj, 2017). This feeling of being misunderstood also echoes the relations with the different mental health professionals they met.

Individual psychopathology. All of the adolescent inpatients with HKM in this sample had an associated psychiatric disorder. The association between a specific diagnostic and HKM status is difficult to affirm as it may reflect local

clinical practice (Kohrt et al., 2014). For example, anxiety and depressive disorders were also the most common diagnoses reported in all inpatients (Benarous, Cravero, et al., 2020). By considering this specific sample of adolescents with very severe forms of psychiatric disorders, our findings support the "type 3" approach of HKM described in Table 1. Put differently, the distinction between a primary and secondary HKM was less relevant here. Our finding was in line with the view of HKM as a concept, distinct from the categories of psychiatric disorders but otherwise clinically relevant to identify a constellation of individual/ familial/cultural factors influencing the health care trajectories of adolescents with SW/SR.

The most striking observation when caring for adolescent inpatients with HKM is the discrepancy between the long duration of SW/SR and the relatively low level of psychiatric symptoms observed during their stay. The vast majority of adolescent inpatients with HKM were not notably anxious during interactions with peers or professionals at the hospital. In these HKM adolescents, passive personality traits seemed more predominant than anxiety traits, in line with the observation by Gayral et al. (1953). While they never consented to the hospitalization, it was exceptional when they actively asked to be discharged, unlike most adolescent inpatients. Of note, no inpatients with HKM repeated a form of reclusion during the hospitalization (e.g., remaining all the time in their room).

Psychiatric symptoms were "unveiled" when HKM adolescents faced feared situations, for example when permissions were organized to enable the patient to return to school. Passivity and avoidance (both at the emotional and behavioral levels) seemed two important clinical dimensions for these patients. While severe anxiety was rarely observed, an intense need for perfection and control was systematically reported both in academic domains and in peer relations. There was little expression of conflicting or ambivalent emotions during their stay, and some of them seemed to adopt a hyper-mature adult-like behavior.

It is hard to say to what extent these characteristics reflect premorbid functioning or consequences of social isolation. At the end of their hospitalization, almost of the patients expressed feeling more confident to be able to reconnect with peers, although their improved ability to express feelings and conflicting views was not always replicated outside the hospital. According to our experience, adolescents with SW/SR and HKM were more likely to relapse after discharge compared to those without HKM. The continuation of family-focused interventions after patient discharge seems to have been an important aspect of the treatment success of adolescents with SW/SR (Benarous, Garny de La Riviere, et al., 2020).

Strengths and limitations

These findings should be considered with regard to the following limitations. The identification of HKM patients

in this chart review was retrospective, therefore subject to measurement bias. This risk was mitigated using a multistep identification procedure. In a context of long duration of stay in inpatient units, we were able to provide detailed clinical observations in various contexts (e.g., during family's interviews, with other patients, in recreational activities, in a school center). The interpretation of our findings should also take into account the specificity of a tertiary care university hospital center, particularly enriched in adolescents with severe and resistant forms of psychopathology. Results may not be generalized to other clinical settings or to community samples. Finally, the low size of the sample precludes us from providing any multivariate analysis. Future research might help to confirm these trends in another database and therefore to be more confident in generalizing these findings. Unlike previous reports (Stip et al., 2016), we did not consider in our study the category of prodromal phase of schizophrenia. However, the association of HKM and psychosis disorder is less likely to be as significant in adolescents as in adults considering the lower prevalence of this condition at a younger age (Giannitelli et al., 2018).

Clinical and research implications

It has been said that HKM has become a global phenomenon in young adults (Li & Wong, 2015a; Wu et al., 2019) but observational studies outside Asian countries are needed to confirm this assumption. Case reports and observational studies published in the last decades in non-Asian countries have focused on late adolescence and young adults (Chauliac et al., 2015; De Michele et al., 2013; Guedj, 2017; Malagon-Amor et al., 2015, 2018), while there is consensus regarding a teenage onset of social withdrawal (Li & Wong, 2015b; Wong et al., 2019). The emergence of consensual definition criteria for HKM will help to develop community-based studies to determine the prevalence of HKM in these countries. In particular, the validation of a French version of Teo and Gaw's criteria for HKM represents a next step for developing clinical research. Further studies with repeated assessments and multiple informants would help to determine to what extent associated psychiatric symptoms are primary or secondary to social withdrawal. Considering cumulative data stressing the possible atypical socioemotional developmental history in HKM (Chong & Chan, 2012; Lee et al., 2013; Wong et al., 2015), the association with childhood-onset psychiatric disorder (e.g., separation anxiety disorder), temperamental traits (e.g., behavioral inhibition), or attachment patterns (e.g., anxious-avoidant attachment style) could be better explored.

In response to the widespread use of the term "HKM" by family to describe youth with SW/SR, a clinical working group was formed in France in 2020 involving professionals from different backgrounds (e.g., home visits programs, university hospital, emergency structures, private practice) (AFHIKI [French Association for the Clinical and Research Study on Hikikomori], 2021). The proponents of a strictly medical view of HKM (approach 1 in Table 1) have interpreted the democratization of the HKM concept in non-Asian countries in terms of mental illness stigma and reluctance to receive a dreaded diagnosis of chronic psychiatric disorder, especially schizophrenia. Our clinical experience with patients and families who referred to themselves as HKM supports a more nuanced view. The progressive course of social withdrawal involving first the patient and then extending to the whole family system was a common denominator for patients with varied socio-economic, ethnical, or cultural backgrounds. Sharing some aspects of the family burden (feelings of shame, self-depreciation, and mistrust) with the patient but also during monthly family group sessions seems useful to help them gain knowledge about these interactive dynamics. The findings presented here are derived from a very special clinical practice, however we found this worth sharing with other professionals. A first clinically relevant step would be to better inform clinicians about the possible benefit of providing, in addition to traditional care, familyfocused interventions to help families with HKM to break down the logic of shame and social isolation impacting care trajectories.

Conclusion

We found a 7% prevalence of HKM syndrome in a French adolescent inpatient sample. This rate represents one in six adolescents with SW/SR at admission. Inpatients with HKM presented distinct clinical features compared with those with other SW/SR, roughly comparable to those reported in an Asian adult population. Further studies are needed to document the validity and clinical relevance for the HKM syndrome in adolescence. A highlight on the developmental and systemic aspects of the HKM syndrome can help our understanding of this syndrome.

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Notes

- 1. The notion of HKM as a distinct clinical syndrome without an associated psychiatric disorder has also been referred to as a primary form of HKM in the literature.
- Many of these authors were inspired by a psychoanalytical view of family dynamics. In particular, the implications of transgenerational values (aims, ideals, aspirations) in the illor over-adapted parental attitudes has been discussed.
- 3. The term NEET was coined in English-speaking countries to refer to young people hidden from employment statistics as they rarely proceed to any administrative procedure to be recognized. This is more a descriptive term useful for future cross-cultural observational studies than a clinical concept.
- 4. While social disaffiliation in these patients usually led to vagrancy rather than confinement, this concept sheds light on the relations between psychiatric, political, and social aspects of social withdrawal in France.
- 5. In this view, HKM is regarded as an anecdotal description of typical psychiatric disorders, such as anxiety disorder, in the same way that the Cotard syndrome is a historical description of a subtype of depressive disorder with melancholic features.
- 6. This assumption is endorsed by the proponent of a "strictly medical approach," as shown in Table 1.
- 7. The causal relationships between internalized symptoms and social withdrawal are related to the question of whether the HKM syndrome has to be considered as primary or secondary for a specific patient (see Table 1). If the two clinical dimensions belong to the same clinical construct, a dose effect relation between social withdrawn and internalized symptoms is expected.
- 8. The fact that a large part of the family interviews was conducted without the presence of the HKM patient probably helped these parents to express the burden represented by the patient's difficulties in a more overt and authentic way.

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