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Mentalization and cognitive skills in men with Klinefelter syndrome versus non-clinical controls

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ABSTRACT

We investigated cognition and mentalization skills, defined as the ability to understand one's own and others' intentions and emotions, in men with Klinefelter Syndrome (KS). The sample was 26 men with KS and 26 non-clinical male controls aged 19–65 years. We measured mentalization with the Reading the Mind in the Eyes Test (RMET) and cognition with neuropsychological tests. The results showed that men with KS had significantly lower scores on the RMET compared to controls. However, the group difference was not significant when controlling for IQ. There were more significant correlations between cognitive domains and mentalization skills in the KS group than for controls. In regression models, cognitive domains explained up to 54% of the variance in mentalization skills for men with KS, compared to 15% for controls. The men with KS struggled particularly with interpreting neutral and negative emotional states relative to the control group. We conclude that men with KS exhibit mentalization difficulties, which are strongly linked to their cognitive abilities, and especially their deficits in verbal learning. Interventions aimed at enhancing language and other neuropsychological functions, as well as mentalization skills, are warranted.

1. Introduction

Klinefelter syndrome (KS) is a male sex chromosome aneuploidy involving an additional X chromosome, with an estimated prevalence of 1:660 men (Bojesen and Gravholt, 2007). Men with KS are at increased risk of physical, psychological, and social challenges (Boada et al., 2009; Skakkebæk et al., 2017). They commonly have pronounced difficulties related to verbal intelligence, whereas non-verbal intelligence does not differ significantly from controls (Boada et al., 2009). The current study focuses on a socio-cognitive domain that is largely unexplored among men with KS, i.e., mentalization skills. Mentalization is the ability to interpret and understand one's own and others' inner states (Bateman and Fonagy, 2004). Mentalizing is associated with mental health (Luyten et al., 2020), verbal abilities (Peterson and Miller, 2012), and general intellectual abilities (Baker et al., 2014). The relationship between mentalization and verbal skills has been widely explored in autism spectrum disorders (ASD). A consistent finding is that persons with ASD struggle with mentalizing, even when they have adequate verbal skills (Baron-Cohen et al., 2001; Peñuelas-Calvo et al., 2019).

Examining mentalization and verbal skills has implications for treatment. If mentalizing difficulties are secondary to language difficulties or to general cognitive difficulties, this indicates early intervention targeting verbal and other cognitive skills. However, if mentalizing difficulties are more independent of cognitive functions, such as in ASD, this indicates interventions such as mentalizing-based therapy (Bateman and Fonagy, 2004). To better inform interventions, it is essential to examine mentalization and its' link to verbal and other cognitive functions in men with KS.

There is sparse research on mentalization skills among men with KS. Studies have examined related concepts, such as social cognition. One

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study found that men with KS struggled with tasks involving identifying and verbalizing feelings (van Rijn et al., 2006). A later study showed that men with KS had difficulties in identifying specific emotions in others, such as fear, contempt, and anger (Babinet et al., 2017). Men with KS have also exhibited difficulties with identifying correct affective states in face images, seemingly independently of general visuospatial perceptual skills (van Rijn et al., 2018). Thus, research suggests that men with KS struggle to understand and interpret their own and others' emotional states. This can lead to misunderstandings in social situations, and potentially inappropriate behavioral regulation. Such difficulties are associated with behavioral and emotional difficulties (Leist & Dadds, 2009, van Zonneveld et al., 2019). Hence, studies on the mentalization skills of men with KS are needed.

Mentalization tests, such as the Reading the Mind in the Eyes Test, Revised (RMET; Baron-Cohen et al., 2001), have facilitated empirical mentalization studies. In the RMET, participants are to interpret others' emotional states based on images of the facial area surrounding the eye. Ours is the first study to use the RMET to assess mentalization among men with KS. What we explicitly measure with the RMET is the ability to interpret other people's emotions based on their facial expression. A consistent finding is that people with ASD perform worse on the RMET than healthy controls, even when controlling for general intellectual abilities (Baron-Cohen et al., 2001; Peñuelas-Calvo et al., 2019). However, other studies indicate that general cognitive abilities play a role for mentalization skills (Baker et al., 2014; Peterson and Miller, 2012). Therefore, mentalization in men with KS should be evaluated in light of other cognitive domains.

Some emotional states may be easier to interpret than others. An earlier study found that men with KS had particular difficulties in recognizing negative emotions like fear and contempt (Babinet et al., 2017). Another study showed that 25% of males with KS failed to recognize neutral faces (van Rijn et al., 2018). Therefore, it is important to examine if men with KS perceive some types of emotions more accurately than other emotions.

In the current study, we investigated mentalization skills in men with KS while statistically controlling for general intellectual abilities and specific cognitive domains. We included a non-clinical control group. Our first research question was: Do men with KS differ from controls on the RMET? Our hypothesis was that men with KS would score lower than controls, based on previous findings of social cognition difficulties in the KS group (Bojesen & Gravholt, 2007, van Rijn et al., 2018). We controlled for full scale IQ in the group comparison. We also examined KS versus non-clinical control differences on the other cognitive tests. Our second research question was: Which cognitive domains are associated with mentalization skills in men with KS, and are these different for non-clinical controls? As language challenges are well-documented among men with KS (Boada et al., 2009; Skakkebæk et al., 2015), we expected that verbal abilities would be more strongly associated with mentalization skills in the KS group than in controls. Our third study question was: Are there certain emotional states that men with KS particularly struggle to mentalize? Based on previous findings, we hypothesized that men with KS would have more difficulties than controls with identifying negative/neutral than positive emotional states (Babinet et al., 2017, van Rijn et al., 2006).

2. Methods

2.1. Sample and procedures

The full sample comprised 30 men with KS and 28 healthy men without KS. Four of the participants with KS and two controls did not complete the RMET test and were therefore excluded from the current study. The KS group (n = 26) was aged 19–60 years (M = 37.0, SD = 10.9). The control group (n = 26) was aged 20–65 years (M = 37.8 years, SD = 13.7). There were significant background differences between the groups. (Table 1). The participants with KS had lower education, fewer worked, and they were less likely to have a romantic partner. Self-reported reading and writing difficulties were frequent among the participants with KS.

Table 1
Background characteristics for men with Klinefelter syndrome and non-clinical controls.

	Klinefelter syndrome	Controls
	N (%)	N (%)
	26 (100)	26 † (100)
Highest completed education*		
Primary school	1 (3.8)	0 (0)
Junior high school	2 (7.7)	1 (3.8)
High school	18 (69.2)	9 (34.6)
Higher education (>2 years)	5 (19.2)	16 (61.6)
Occupational status*		
Student	4 (15.4)	1 (3.8)
Working	11 (42.3)	24 (92.3)
Disability pension	8 (30.8)	0 (0)
Retired	3 (11.4)	0 (0)
Marital status*		
Single	14 (53.8)	4 (15.4)
Married/cohabiting	10 (38.5)	20 (76.9)
Divorced/separated	2 (7.7)	1 (3.8)
Reading/writing difficulties*		
Yes	11 (42.3)	1 (3.8)
No	13 (50.0)	24 (92.3)
Unsure	2 (7.7)	0 (0)

Note. † Missing data for one control person for occupational status, marital status, and reading/writing difficulties. *Significant difference $p < .05$.

We recruited participants with KS via the user register, website, and social media platforms of the Frambu resource centre for rare disorders, and through information posted on the Klinefelter Association website. Consent forms and information about the study were sent to users and members by regular mail or e-mail. The control group was recruited via advertisements in local newspapers, notices at educational institutions and shops, and through an advertisement on social media. The only exclusion criterion for participation was claustrophobia, as the participants had to undergo an fMRI examination (results not reported here). The test administrators were advanced clinical psychology students who had been trained by an experienced clinical neuropsychologist.

The study was carried out in accordance with the Declaration of Helsinki. The project was ethically approved by the institutional review board. All participants gave written informed consent and were offered a verbal presentation of the consent form. In the consent form, the RMET test was described to measure "your understanding of emotions". The participants took part in a draw for a universal gift card worth USD100.

2.2. Measurements

2.2.1 The Reading the Mind in the Eyes Test, revised (Baron-Cohen et al., 2001) was used as a measure of mentalization. The test comprises 36 different photographs of the facial area surrounding the eyes of adult women and men showing different emotional states (Fig. 1). For each picture, there are four answer options, one with the "correct" emotional state. There are 16 positive (e.g., friendly), seven neutral (e.g., thoughtful), and 13 negative (e.g., hostile) images (Hudson et al., 2020). The answers are scored true (1) or false (0), yielding a maximum score of 36. Participants are asked to choose which word best describes what the person in the picture is thinking or feeling. The RMET comes with a synonym list of the words used to describe the images and an example sentence for each word. The test administrators provide the participants with this list and explains that they can use it for emotion words that are difficult to understand. The test has been developed to be a measure of mentalization, where the purpose is to indicate the degree of social sensitivity (Baron-Cohen et al., 2001). The RMET does not have standard norms. However, average scores from several samples have been published (Baron-Cohen et al., 2001; Kidd and Castano, 2013; Wilson et al.,

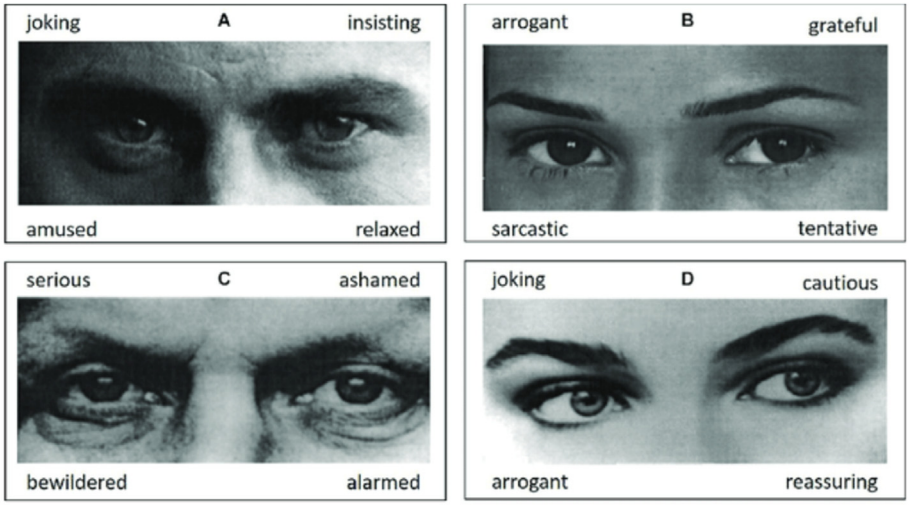

Fig. 1. Sample tasks from the Reading the Mind in the Eyes Test.
Note. Fig. 1 shows four example tasks from the Reading the Mind in the Eyes Test (RMET). The participant is instructed to choose one of the four words that best describes the mental/emotional state. The images are taken from the "Reading the Mind in the Eyes" Test Revised Version: A study with Normal Adults, and Adults with Asperger Syndrome or High-functioning Autism" by Baron-Cohen et al., 2001) Baron-Cohen et al., 2001), J. Child Psychol. Psychiat., 42 (2), pp 241–251, Cambridge University Press. Copyright 2001 Association for Child Psychology and Psychiatry.

2014). The reliability and validity of the test are considered to be good (Olderbak et al., 2015; Ventola and Friedman, 2015). We used the Norwegian translation of the RMET (Sommerfeldt and Skårderud, 2008).

2.2.2 The Wechsler Abbreviated Scale of Intelligence, Second Edition (WASI-II; Wechsler, 2011) was used to measure general intellectual abilities. We used four subtests, which combined provide an estimate of the general ability level (full scale IQ). Verbal IQ was assessed via the Vocabulary and Similarities subtests. In the Vocabulary subtest, the participant is asked to explain words. The test comprises a total of 31 words of increasing difficulty. The answers are scored as completely correct (2), partly correct (1), or 0 (wrong). Administration stops after 3 consecutive 0-point answers. The test measures expressive language, verbal knowledge, and general knowledge and is linked to other cognitive abilities such as memory, learning ability and language development, including concept formation (Wechsler, 2011). In the Similarities subtest, the participant is asked to explain the similarity between words. The test comprises a total of 28 word or concept pairs of increasing difficulty (i.e., abstraction level). The answers are scored as completely correct (2), partly correct (1) or 0 (wrong). Administration stops after 3 consecutive 0-point answers. The test measures verbal concept formation and abstract verbal reasoning (Wechsler, 2011).

Performance IQ was assessed via the Block Design and Matrix Reasoning subtests. In Block Design, the participant is asked to use blocks to recreate two-color patterns shown by the test administrator. Each block has two white sides, two red sides, and two half red/half white sides. The test comprises 13 items of increasing difficulty. The tasks have a time limit. The answers are scored from 0 to 7 according to execution and time spent. Administration stops after 2 consecutive 0-point answers. The test measures aspects of perceptual and motor function, as well as tempo (Wechsler, 2011). In Matrix Reasoning, image matrices are shown where a bit is missing, and the participant is asked to complete the matrices by pointing out which image is missing from five possible options. The test comprises 30 matrices of increasing difficulty. The answers are scored as right (1) or wrong (0). Administration stops after 3 consecutive 0-point answers. The test consists of five forms of non-linguistic reasoning tasks: pattern completion, classification, judgment of similarity, and sequential reasoning (Wechsler, 2011). The WASI-II is widely used both in research and in clinical practice as it provides a reliable and valid estimate of IQ (McCrimmon and Smith, 2013; Wechsler, 2011).

2.2.3 The Verbal Fluency Test from the Delis Kaplan Executive Function System (D-KEFS; Delis et al., 2001) was used to measure phonemic fluency. The participant is asked to say as many words as possible that start with a certain letter (i.e., F, A, S) within 60 s. The participant is instructed not to repeat the same word, not to state words that are numbers, names, or the same word with different inflections. The test measures the retrieval of semantic information from long-term memory after a phonemic category or cue, and the ability for rapid verbal production. The test requires short-term memory for the instructions and words said. The test is mainly used to measure executive functions, but also provides information about the semantic system that accounts for the storage of words and their associations (Delis et al., 2001). The Verbal Fluency Test has demonstrated good reliability (Shunk et al., 2010) and had larger correlations with language measures such as Vocabulary and Similarities than with executive functions in a study of children with language difficulties (Henry et al., 2015). In the current study, we used the sum score of the total number of correct words in the analyses.

2.2.4 The California Verbal Learning Test, 2nd edition (CVLT-II; Delis et al., 2000) was used to measure verbal memory. The CVLT-II is used to map strategies and processes when learning and recalling verbal material (Delis et al., 2001). In this test, 16 words are read out by the test administrator, after which the participant is asked to repeat the words. This is repeated until all 16 words are repeated, or maximum five times. The glossary can be divided into four semantic categories (i.e., fruits, animals, means of transport, furniture), but this is not explained by the administrator. Thus, the participant may use semantic learning strategies to remember the words more easily. The total number of words remembered over five trials is summed to provide a measure of verbal learning. We used the Norwegian version of the CVLT-II (Lundervold and Sundet, 2004). A literature review showed that the psychometric properties of the Norwegian version are similar to the American version, and that the test has adequate properties as a measure of verbal learning (Siqveland, 2014).

2.3. Data analytic plan

We used chi-square tests and t-tests for independent samples to examine group differences. We used Cohen's *d* (M_2-M_1/SD pooled) as an effect size measure (Cohen, 2013). We used two-tailed Pearson's *r* correlation tests to examine associations between variables. We then employed two sets of linear multiple regression analyses. First, to examine group differences while controlling for IQ, we used KS status and full scale IQ as predictors of the RMET scores. KS versus non-clinical control status was dummy coded as 1 and 2, respectively. Then, we ran a series of multiple linear regressions adding cognitive domains (i.e., language tests and performance IQ) as predictors of the RMET scores, separately for the KS group and the controls. We used the Akaike Information Criterion (AIC) to assess model fit. We examined collinearity by considering the variance inflation factor (VIF) and tolerance values, where VIF >10 or tolerance <0.10 was considered problematic. IBM

SPSS 27.0 was used for all analyses with alpha at $p < .05$. As the study is exploratory, we did not adjust the p-level for multiple comparisons.

3. Results

3.1. Group differences

The KS group had significantly lower scores than controls on all measures except Verbal Learning (Table 2). The effect size difference between the groups on the RMET was moderate. The other significant group differences were large. We predicted the RMET scores based on KS status while controlling for IQ. The significant group difference in RMET scores disappeared when we controlled for full scale IQ (Table 3).

3.2. Relations between mentalization and other cognitive domains

For the KS group, the RMET score was significantly correlated with all the nine other cognitive domains (Table 4). All the correlations were positive, indicating better RMET scores with better cognitive test results. For the non-clinical controls, the RMET was only significantly correlated with four of the other nine cognitive domains. These exploratory correlation patterns suggest that there may some differences in the magnitude and patterns of correlations between mentalization and cognitive domains for men with KS and non-clinical controls.

In Tables 5 and 6 we present the results of four regression models for the KS group and controls, respectively. We added new predictors in each model. The first three predictors were Verbal IQ, Verbal Fluency, and Verbal Learning. In the fourth model, we added Performance IQ. For the KS group, all the models were significant, and collectively the cognitive measures explained 54% of the variance in scores on the RMET. Based on the AIC values, adding more cognitive measures enhanced model fit and the full model showed best fit. For the control group, only models 1 and 2 were significant. The cognitive measures explained 15% of the variance in the RMET scores, which is substantially lower than for the KS group. Further, the AIC values indicated that adding cognitive measures did not enhance model fit in the control group. Thus, it appears that the verbal and performance measures play a larger role for mentalization skills in men with KS than among controls.

3.3. Specific emotional states

The KS group had lower scores than the controls on all the RMET categories (i.e., negative, positive, and neutral emotional states). The differences were only significant for negative and neutral emotional states (both $p < .05$). The group difference in effect size was large for negative emotional states ($d = 1.35$) and moderate for neutral emotional states ($d = 0.55$).

4. Discussion

We examined mentalization skills among men with KS compared to non-clinical controls. Our first hypothesis was supported in that men with KS had lower scores on the mentalization test (RMET) than controls. The KS group performed equally to or slightly poorer than high-functioning ASD samples in other studies (Baron-Cohen et al., 2001; Wilson et al., 2014). Note, however, that the difference between the KS group and controls disappeared when controlling for Full Scale IQ. Thus, general intelligence outweighs KS status as a potential explanation for the group difference. We cannot know how the mentalization skills of men with KS would compare to controls who were matched on general intellectual ability.

We also examined associations between cognitive domains and mentalization skills. Our results showed that the scores on mentalization skills among men with KS were linked to both verbal and non-verbal cognitive abilities. Men with KS typically display the asymmetry of relatively weaker verbal than non-verbal abilities as persons diagnosed with ASD (Mayes and Calhoun, 2003). A meta-study showed that verbal IQ did not correlate with the RMET scores in an ASD group, whereas it did for healthy controls (Peñuelas-Calvo et al., 2019). Our findings suggest that for men with KS, like non-ASD controls, there is a strong association between verbal cognitive skills and RMET performance.

Based on our findings and others (Peñuelas-Calvo et al., 2019, 2019van Rijn et al., 2014), there is reason to assume that men with KS also use other cognitive abilities to solve the RMET than both ASD groups and the normal population. In the KS group, higher performance IQ was significantly related to higher RMET scores. This correlation was non-significant in the control group. This could reflect a difference between the groups in how they approach mentalization tasks. Performance IQ reflects broad visual and spatial abilities. Hence, men with KS may have used more visual analysis and logical reasoning related to details in and around the depicted eyes rather than affect-oriented "gut feeling". One study found that RMET scores depended on controlled processing, such as executive functions (Malaei et al., 2020). Executive difficulties are common among men with KS (Lee et al., 2011; Skakkebæk et al., 2014), whereas visual-analytical abilities are considered a relative strength (Boada et al., 2009; Skakkebæk et al., 2015). A meta-study found that performance IQ correlated significantly and negatively with the RMET in an ASD group (Peñuelas-Calvo et al., 2019). Our findings therefore also point to a possible difference in how people with ASD and KS process social stimuli. This is supported by a brain imaging study showing that men with KS had more activation in the frontal areas of the brain than people with ASD and non-clinical controls when solving social tasks (Brandenburg-Goddard et al., 2013). Such increased engagement of the frontal cortex may be due to more “voluntary” logical reasoning compared to a relatively more “intuitive/automatic” manner of solving the task, possibly as a compensatory strategy.

Whereas there was a significant association between verbal IQ and the RMET score also for the non-clinical controls, adding further verbal measures (or performance IQ) to the RMET prediction model did not enhance model fit in the control group. Further, the exploratory analyses of the correlation patterns overall showed more and larger correlations between verbal cognitive domains and mentalization for the KS group compared to controls. Thus, in terms of our second hypothesis, we take these findings to indicate support for a *possibly* stronger link between

Table 2
Test Scores in Men with Klinefelter Syndrome and Controls.

	Klinefelter syndrome			Controls			Effect size
Tests	*M*	*SD*	*Range*	*M*	*SD*	*Range*	*Cohen's d*
Mentalization	20.77	4.30	11–28	23.69	4.29	13–30	0.68*
Full scale IQ	98.36	14.67	63–119	117.50	11.29	88–134	1.46**
Verbal IQ	92.69	14.70	55–114	115.15	11.29	88–133	1.71**
Performance IQ	103.62	15.36	62–121	116.27	12.41	92–135	0.91**
Verbal Fluency	30.58	10.48	12–54	44.15	14.88	25–85	1.05**
Verbal Learning	49.50	11.34	30–71	51.73	9.72	35–72	0.21

Men with Klinefelter Syndrome and Non-Clinical Controls' Test Scores.
Note. Mentalization = Reading the Mind in the Eyes Test. IQ = Intelligence Quotient. *the difference was significant at the p < .05 level. **the difference was significant at the p < .001 level.

Table 3
Klinefelter status and intelligence as predictors of mentalization skills.

	Predictors	B	SE	Beta	95% CI	t	Adj.R^2	F	VIF	AIC
Model 1							0.09	6.18*		153.77
	(Constant)	17.66	1.93		(13.80–21.54)	9.17**				
	Group	3.01	1.21	0.34	(0.58–5.45)	2.49*			1.00	
Model 2							0.36	15.18**		134.39
	(Constant)	3.05	3.54		(-4.06-10.16)	0.86				
	Group	−0.52	1.29	−0.06	(-3.07–2.03)	−0.41			1.56	
	Full scale IQ	0.18	0.40	0.66	(0.11–0.26)	4.65**			1.56	

Note. SE = Standard error. CI = confidence interval. Adj. = adjusted. VIF = Variance inflation factor.
AIC = Akaike information criterion.*significant at the $p < .05$ level- **significant at the $p < .001$ level.

Table 4
Correlations between cognitive domains for 26 men with Klinefelter syndrome and 26 non-clinical controls.

	RMET	Full scale IQ	Verbal IQ	Performance IQ	Verbal Fluency	Matrix Reasoning	Block Design	Vocabulary	Similarities	Verbal Learning
RMET		.42*	.42*	.28	.40*	.08	.23	.31	.45*	.15
Full scale IQ	**.68****		.84**	.84**	.47*	.39*	.36	.92**	78**	.00
Verbal IQ	**.49***	**.90****		.41*	.46*	.13	.36	.92**	92**	.18
Performance IQ	**.64****	**.87****	**.53****		.33	.63**	.94**	.33	.39	−.18
Verbal Fluency	**.53****	**.38**	**.23**	**.43***		.27	.22	.47*	.46*	−.01
Matrix Reasoning	**.68****	**.82****	**.53****	**.94****	**.41***		.57**	.05	.07	−.09
Block Design	**.40***	**.63****	**.30**	**.85****	**.46***	**.72****		.27	.36	−.30
Vocabulary	**.58****	**.90****	**.91****	**.67****	**.27***	**.62****	**.35**		.74**	.16
Similarities	**.40***	**.80****	**.93****	**.46***	**.25**	**.45***	**.33**	**.73****		.13
Verbal Learning	**.59****	**.55****	**.39**	**.38**	**.35**	**.39***	**.17**	**.56****	**.28**	

Note. Correlations for the KS group are given in the lower quadrant. Correlations for the control group are given in the upper quadrant. The mentalization test RMET = Reading the Mind in the Eyes Test, Revised, Full-scale IQ (Verbal IQ + Performance IQ), and the individual subtests (Vocabulary, Similarities, Matrix Reasoning, and Block Design) are from the WASI-II (Wechsler Abbreviated Scale of Intelligence, Second Edition), Verbal Fluency is a test of phonemic fluency from the D-KEFS (Delis-Kaplan Executive Function System). Verbal Learning refers to the total number of words learned over the five trials of the CVLT-II (California Verbal Learning Test), ** = $p < .01$, * = $p < .05$.

Table 5
Predicting the reading the mind in the eyes test in men with Klinefelter syndrome.

	Predictors	B	SE	Beta	95% CI	t	Adj.R^2	F	VIF	AIC
Model 1							0.21	7. 65*		72.18
	(Constant)	7.43	4.88		(-2.64–17.50)	1.52				
	Verbal IQ	0.14	0.05	0.49	(0.04–0.25)	2.77*			1.00	
Model 2							0.37	8.32*		69.57
	(Constant)	4.73	4.48		(-4.54–13.10)	1.06				
	Verbal IQ	0.11	0.05	0.39	(0.02–0.21)	2.40*			1.06	
	Verbal Fluency	0.18	0.07	0.43	(0.04–0.32)	2.66*			1.06	
Model 3							0.47	8.89*		65.82
	(Constant)	1.59	4.23		(-7.18–10.37)	0.38				
	Verbal IQ	0.08	0.05	0.27	(-0.01–0.17)	1.78			1.16	
	Verbal Fluency	0.14	0.06	0.35	(0.01–0.27)	2.31*			1.11	
	Verbal Learning	0.15	0.06	0.39	(0.03–0.27)	2.50*			1.20	
Model 4							0.54	8.22*		63.34
	(Constant)	−2.03	4.48		(-11.34–7.28)	−0.45				
	Verbal IQ	0.04	0.05	0.14	(-0.06–0.14)	0.82			1.47	
	Verbal Fluency	0.11	0.06	0.26	(-0.03–0.26)	1.67			1.25	
	Verbal Learning	0.12	0.06	0.35	(-0.01–0.25)	2.28*			1.23	
	Performance IQ	0.09	0.05	0.33	(-0.01–0.19)	1.84			1.69	

Note. Dependent variable: The mentalization test RMET (Reading the Mind in the Eyes Test, Revised), SE = Standard error. CI = confidence interval. Adj. = adjusted. VIF = Variance inflation factor. AIC = Akaike information criterion.*significant at the $p < .05$ level- **significant at the $p < .001$ level. Verbal IQ and Performance IQ are from the WASI-II (Wechsler Abbreviated Scale of Intelligence, Second Edition), Verbal Fluency from D-KEFS (Delis-Kaplan Executive Function System). Verbal Learning is from the CVLT-II (California Verbal Learning Test, Second Edition).

verbal abilities and mentalization skills in the KS group than in the control group. However, we cannot demonstrate a significant difference in magnitude of the overlap between the samples. It is also likely that the considerable variation in IQ range in the KS sample led to more significant associations for this group than for controls.

Our third hypothesis was that the participants with KS would particularly struggle with identifying negative and neutral emotional states on the RMET. Our results were consistent with this, in that men with KS diverged most from the control group on the negative emotion items. Struggling to interpret negative emotional states such as fear, contempt, and anger can lead to problems in social situations, as nonverbal cues convey information about the emotional state of the sender. Misunderstanding such signals can affect how one understands the environment and thus how one regulates behavior. Challenges in identifying negative emotional states are associated with several types of personal and inter-personal difficulties. Such mentalizing difficulties are

Table 6
Predicting the reading the mind in the eyes test in non-clinical controls.

	Predictors	B	SE	Beta	95% CI	t	Adj.R^2	F	VIF	AIC
Model 1							0.14	5.02*		74.26
	(Constant)	5.51	8.15		(-11.31–22.34)	0.68				
	Verbal IQ	0.16	0.07	0.42	(0.01–0.30)	2.24*			1.00	
Model 2							0.16	3.39*		74.26
	(Constant)	7.47	8.19		(-9.48–24.41)	0.91				
	Verbal IQ	0.11	0.08	0.29	(-0.50–0.27)	1.43			1.27	
	Verbal Fluency	0.08	0.06	0.26	(-0.05–0.20)	1.28			1.27	
Model 3							0.17	2.67		74.26
	(Constant)	3.52	8.95		(-15.04–22.08)	0.39				
	Verbal IQ	0.11	0.08	0.28	(-0.06–0.27)	1.35			1.28	
	Verbal Fluency	0.08	0.06	0.27	(-0.04–0.20)	1.33			1.27	
	Verbal Learning	0.09	0.08	0.20	(-0.08–0.25)	1 .08			1.01	
Model 4							0.15	2.09		74.26
	(Constant)	−0.89	10.90		(-23.57–21.78)	−0.08				
	Verbal IQ	0.09	0.08	0.23	(-0.09–0.26)	1.03			1.43	
	Verbal Fluency	0.07	0.06	0.25	(-0.06–0.20)	1.17			1.31	
	Verbal Learning	0.10	8.15	0.23	(-0.07–0.28)	1 .22			1.08	
	Performance IQ	0.05	0.07	0.15	(-0.10–0.21)	0.73			1.33	

Note. Dependent variable: The mentalization test RMET (Reading the Mind in the Eyes Test, Revised), SE = Standard error. CI = confidence interval. Adj. = adjusted. VIF = Variance inflation factor. AIC = Akaike information criterion.*significant at the $p < .05$ level- **significant at the $p < .001$ level. Verbal IQ and Performance IQ are from the WASI-II (Wechsler Abbreviated Scale of Intelligence, Second Edition), Verbal Fluency from D-KEFS (Delis-Kaplan Executive Function System). Verbal Learning is from the CVLT-II (California Verbal Learning Test, Second Edition).

common among individuals with emotional burdens (Leist and Dadds, 2009) and in young people with behavioral problems (Hubble et al., 2015). Men with KS have an increased risk of a number of mental disorders (Bruining et al., 2009; Cederlöf et al., 2014; Skakkebæk et al., 2018). Possibly, more research on specific types of mentalization difficulties among men with KS can provide answers that can help sharpen interventions.

The participants with KS also struggled more than the control group to understand neutral facial expressions. For positive emotional states, the participants with KS were on a par with the control group, although the latter group had significantly higher IQ. This suggests that men with KS may have a relative strength when it comes to identifying positive emotional states. Difficulties in identifying neutral expressions have been found in people with vulnerability to depression (Leppänen et al., 2004). Men with KS have a higher risk of developing depression (Skakkebæk et al., 2018). We did not control for depression, but there is a possibility that depressive symptoms may have influenced our findings. Another possible explanation is that neutral expressions are often more ambiguous and involve less facial expression. Understanding neutral expressions may thus require a higher degree of social sensitivity than positive and negative expressions. An earlier study showed that healthy control subjects had more errors on neutral expressions than clear positive and negative expressions (Hudson et al., 2020). Struggling to put emotional states into words can be psychologically strenuous and can possibly help to understand why men with KS have increased stress levels during certain mentalizing tasks (van Rijn et al., 2006). However, the connection between mentalization difficulties related to neutral emotional states and depression among men with KS is undocumented and should be investigated further.

The current study has limitations. There were several differences between the men with KS and the controls, most importantly in general intellectual ability. This complicates interpretation of our findings. In particular, because the group difference between KS and controls on overall mentalization skills disappeared when controlling for IQ, we cannot conclude about the role of KS status for mentalization. Second, the sample size was relatively small. KS is a rare disorder, and an overview of 19 studies showed that the mean number of KS participants was 29 (Skakkebæk et al., 2015). A sample of 26 is therefore on par with other KS studies. Third, we recruited participants from non-clinical settings, so the generalizability to men who are more severely affected by KS may be limited.

The current study has implications for practice and research. Although the main reason appears to be lower intellectual abilities, the current study provides new findings showing that men with KS have more difficulties than controls in processing and interpreting certain emotional states, such as anger and other negative emotions, and more neutral or ambiguous facial expressions. This is important information for practitioners and counselors. Men with KS may need psychoeducation about these difficulties, for enhanced awareness and potentially, training. Because the mentalization skills of men with KS are linked to multiple cognitive domains, thorough neuropsychological examination can inform case formulations and subsequent interventions for men with KS.

The RMET was suited to identify that men with KS have difficulties with labelling certain emotional states in others. It is nevertheless possible that the RMET is not an optimal measure of social cognition or mentalization for clinical groups such as KS, and that the results are strongly affected by cognitive domains. There is reason to suspect that the extent to which cognitive factors influence mentalization varies between the clinical group and controls. Previous studies have also shown differences in which cognitive abilities affect performance on the RMET for various clinical groups (Babinet et al., 2017; Morel et al., 2018).

To investigate the connection between cognitive abilities and (broader) mentalization difficulties further, studies with matched control groups, both in terms of IQ and educational level, would be useful. One possibility could be to set a lower limit for intellectual ability level. However, this would be at the expense of generalizability. With larger samples, it will be possible to create subgroups based on functional level. Our findings support previous recommendations of enhanced training for people with KS. Neuropsychological interventions should encompass the social dimensions of cognition.

Author contributions

KWF contributed to conceptualization; data curation; formal analysis; funding acquisition; investigation; methodology; project administration; supervision; and writing, SKF contributed to data curation; formal analysis; and writing, NB contributed to data curation; formal analysis; and writing, RH contributed to onceptualization; data curation; project administration; supervision; and writing, CHG contributed to onceptualization and writing, A-KS contributed to conceptualization; data curation; Methodology; Project administration; Supervision; Validation; and writing.

Declaration of competing interest

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