

Social attention, empathy, emotion recognition and emotional arousal in Klinefelter syndrome

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Abstract

The aim of this study was to expand the knowledge about the specific social-emotional difficulties within Klinefelter syndrome (47, XXY) by measuring attention to social cues, emotion recognition skills, empathy and emotional arousal. 14 participants with Klinefelter syndrome (age range 16-56) and 14 control participants (age range 22-60) were included. All participants watched four video clips designed to evoke empathy, and filled out a questionnaire about their own and the main character's emotions after each video.

Furthermore, they completed an emotion recognition task consisting of 80 pictures of faces with a neutral, happy, scared or angry expression. During both tasks eye movements and fixations were measured. Electrocardiogram and skin conductance measurements were done at baseline, during the video clips and during the emotion recognition task. Participants with Klinefelter syndrome had equal emotion recognition scores compared to the control group, but empathy scores were lower for the Klinefelter group. Participants with Klinefelter spent less time fixating on eyes and more time fixating outside the face during the emotion recognition task. No group differences in overall fixation times were found during the videos, but longer fixations on eyes and mouths and shorter on objects predicted better empathy scores. Psychophysiological responses differed between groups during the empathy videos: participants with Klinefelter showed somewhat stronger skin conductance reactions than controls. The current results can contribute to the development of interventions for Klinefelter syndrome. Moreover, the results can give insight into the role of the X-chromosome in the relation between (social) attention processes and social-cognitive functioning.

Keywords: Klinefelter syndrome, eye tracking, psychophysiology, empathy, emotion recognition

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Klinefelter syndrome is a chromosome disorder characterized by an additional X chromosome in boys and men (47, XXY), and is prevalent in 1:500 to 1:1000 male births (Morris, Alberman, Scott, & Jacobs, 2008). Three quarters of these cases goes unnoticed, only a quarter is diagnosed (Bojesen, Juul, & Gravholt, 2003). An explanation for this could be that the physical features of Klinefelter syndrome are mild compared to other chromosomal or genetic abnormalities and the behavioural symptoms vary. The IQ of boys and men with Klinefelter is mostly in the normal range, but especially their verbal IQ has often been found to be lower than that of comparison groups (Leggett, Jacobs, Nation, Sherif, & Bishop, 2010).

The social and emotional functioning of individuals with Klinefelter syndrome have only recently become a focus of research. Generally boys and men with Klinefelter are said to have difficulties with social situations and problems with social adjustment (Bender, Harmon, Linden, Bucher-Bartelson, & Robinson, 1999; Geschwind, Boone, Miller, & Swerdloff, 2000). They are described as shy, unassertive and socially withdrawn, but also as impulsive and showing socially inappropriate behaviour. Recent studies have found that men and boys with Klinefelter have a higher level of autistic traits and a higher risk for developing an autism spectrum disorder (ASD; Bishop et al., 2010; Van Rijn, Swaab, Aleman, & Kahn, 2008). The elevated level of autistic traits was found on the whole range of components of the autism phenotype: social skills, communication, attention to detail, attention switching and imagination.

On the emotional domain, men with Klinefelter syndrome report to be emotionally aroused more easily than controls (Van Rijn, Swaab, Aleman, & Kahn, 2006). Besides that, they report a higher distress than controls during social situations, for example when dealing with criticism or when starting a conversation with others (Van Rijn et al., 2008). The Klinefelter group also reported that they had fewer interactions in which they expressed negative feelings, although their overall amount of participation in social situations was equal to that of controls. Moreover, they have a weaker ability to identify and verbalize their own emotions (Van Rijn et al., 2006). The studies described above mainly concern observable behaviour and are based on self-report measures; little research has directly tested social-cognitive skills in individuals with Klinefelter. There is some evidence that men with Klinefelter syndrome have difficulties recognizing emotions: in one study they were less accurate at recognizing emotions from people's tone of voice and in another study they made

more errors in recognizing angry, but not other types of, facial expressions (Van Rijn, Aleman, Swaab, Krijn, Vingerhoets, & Kahn, 2007; Van Rijn et al., 2006).

From the above it can be concluded that individuals with Klinefelter often have problems in social and emotional functioning that persist into adulthood. They also seem to have a higher chance of developing (traits of) autism. A deviant social attention could be one of the bases of these problems. Social attention can be described as the process of directing attention to social cues in the environment, such as faces, eyes and hand gestures. The link between social attention and social adjustment or social-cognitive skills has been examined several times in children and adults with ASD. For example, when looking at video clips of social situations, males with autism spent less time focusing attention on eyes and more time focusing attention on mouths, bodies and objects (Klin, James, Schultz, Volkmar, & Cohen, 2002). Besides that, spending more time looking at objects predicted lower social adjustment, but spending more time looking at mouths predicted better social adjustment. Differences in eye gaze behaviour have also been found when individuals with and without autism looked at pictures of faces (Dalton et al., 2005; Pelphrey et al., 2002). Individuals with autism mainly paid less attention to the eye region. Kirchner, Hatri, Heekeren and Dziobek (2011) found that the more men with autism looked at the eye region of a face, the better their emotion recognition skills. The time they spent looking at the mouth was negatively related to their emotion recognition abilities. Another study found these same relations in a group of children with ASD and a typically developing control group: emotion recognition accuracy was positively related to fixation time on the eye region and negatively to fixation time on the mouth region (Bal et al., 2010). There is thus a well established link between attention to social cues, especially eyes, and social-cognitive skills. In a review, Itier and Batty (2009) even conclude that problems in processing information from eyes and gaze may be a crucial part of social cognition deficits.

Attention to social cues, as measured with eye gaze patterns, could also be related to the amount of emotional arousal evoked by the stimuli. Very little research has been done to investigate the relation between eye movements or fixations and physiological arousal. In one study, people with PTSD showed more and stronger skin conductance responses to initial fixations on trauma words than controls (Felmingham, Rennie, Manor, & Bryant, 2011). Their physiological reaction when paying attention to something threatening is thus stronger than for most people. However, nothing is known about social attention, nor about arousal regulation within Klinefelter syndrome.

In the general population, the presentation of faces, especially those with an emotional expression, typically leads to a short increase in skin conductance, as well as a brief deceleration followed by an acceleration in heart rate (Bradley, Lang, & Cuthbert, 1993; Fusar-Poli, Landi, & O'Conner, 2009; Jönsson, & Sonnby-Borgström, 2003). These reactions are less strong when you have seen the stimulus before (the so called habituation effect). The strength of heart rate and skin conductance responses varies from person to person and is different for certain clinical groups: children and adolescents with autism for example have been found to show a stronger skin conductance reaction to faces with direct and averted gaze compared to the control group (Joseph, Ehrman, McNally, & Keehn, 2008). Other studies have however found that children and adults with autism show an equal skin conductance response to objects as to faces, whereas the healthy control children and adults show a stronger reaction to faces than to objects (Hirstein, Iversen, & Ramachandran, 2001; Hubert, Wicker, Monfardini, & Deruelle, 2009). Emotional video clips typically also induce physiological reactions, mainly an increase in heart rate, in skin conductance level and in the amount and amplitude of skin conductance responses (SCRs; for an overview, see Kreibig, Wilhelm, Roth, & Gross, 2007). Again, some clinical groups have been reported to show devious reactions (Giesbrecht, Merckelbach, Van Oorsouw, & Simeon, 2010).

Physiological arousal could have an influence on emotional processing and social cognition, as is stated by the somatic marker hypothesis (Damasio, 1994). Having a relatively strong arousal to social stimuli, such as emotional faces or the start of a conversation with someone, may make these stimuli threatening or overwhelming, in a way you would rather avoid them. Higher emotional arousal may also lead to the fear around and avoidance of emotional situations, like expressing negative feelings or receiving criticism. As mentioned above, men with Klinefelter syndrome reported to experience less situations in which they express negative feelings (Van Rijn et al., 2008). On the other hand, having a relatively weak arousal to social and emotional stimuli, may make the urge to pay attention to (relevant aspects of) these stimuli weaker. Abnormal arousal regulation, in either direction, could thus have an effect on social-emotional functioning.

To conclude, there is evidence that boys and men with Klinefelter syndrome have several difficulties in the domain of social-emotional functioning, such as problems with identifying their own and other's emotions and an elevated level of autistic traits. This evidence is however mainly based on self-report measurements. In this research, the knowledge about the specific social-emotional difficulties of people with Klinefelter is

expanded by directly measuring their social attention, emotion recognition skills and emotional arousal.

Research questions

The first focus of this research concerns social attention. More specifically, it was investigated how much time males with and without Klinefelter spontaneously pay attention to social cues when watching video clips that are selected to evoke empathy, and to relevant aspects of faces when watching photographs of facial expressions. Also, it was examined whether social attention (eye fixation patterns) while watching videos predicts the level of empathy evidenced from participants' reactions to the videos. Paying less attention to social cues could be a basis for the difficulties in social functioning of people with Klinefelter. The link between attention to social cues and emotion recognition and social functioning has already been established in boys and men with autism (Kirchner, et al., 2011; Klin, et al., 2002).

The second focus of the study was on physiological arousal. The difference between people with and without Klinefelter syndrome in physiological arousal (heart rate and skin conductance) evoked by the video clips and photographs of faces was investigated. In a previous study (Van Rijn et al., 2006) men with Klinefelter syndrome showed a higher emotional arousal on self-reports, but it is not known if this is due to the perception of their arousal or if they also show more actual, physiological arousal. Since men with Klinefelter syndrome have difficulties identifying and describing their own emotions (Van Rijn et al., 2006), their self-reported arousal may not be in accordance with their actual arousal. Also, among adults from the general population rather weak correlations (between .27 and .35) between these two types of arousal have been found (Sze, Gyurak, Yuan, & Levenson, 2010). Therefore, the relation between participants' heart rate and skin conductance and the subjective ratings of their emotional arousal during the video clips was also examined.

Methods

Participants

14 men and adolescents with Klinefelter syndrome (age range 16-56, $M=39.6$) were included in this study, as well as 14 men and adolescents (age range 22-60, $M=34.9$) without Klinefelter in the control group. The mean age did not differ between groups, but the mean

education level was higher for the control group (age: $t(26) = -1.00, p=.33$; education level: $t(26) = 4.76, p<.001$). Participants with Klinefelter were recruited through the Klinefelter society or were in the existing database of the university and control participants were found using informal recruitment. All participants had cognitive abilities within the normal range and normal or corrected-to-normal vision. Written informed consent was obtained from all participants.

Instruments

Eye tracking video clips. After a calibration procedure four empathy evoking video clips of approximately 90 seconds were presented in randomized order. The main character of the video clip showed either pain and surprise, sadness and upset, fear and anxiety or happiness and cheerfulness. Eye movements and fixations were measured with the Tobii T120 eye tracker. The areas of interest (AOIs) were grouped into the following categories: *face, eyes, mouth, hands, body, objects* and *other*. *Face* is defined as a fixation within the face but outside the eyes and mouth. *Hands* are only part of the AOI *hands* if they have a communicative function (for example hand gestures or putting a hand on someone's shoulder to show support). *Other* is defined if the fixation is on the screen but outside the areas of interest of all other categories. After each video clip participants completed two questionnaires, one concerning the emotions of the main character and the other concerning their own emotions while viewing the video. By putting a cross on a continuous line of 10 cm they indicated how strong they or the main character felt a range of 10 emotions. Participants were asked the reason of the one or two emotions that they felt most strongly.

Eye tracking emotion recognition task. In this task 80 greyscale pictures of faces with a neutral, happy, scared or angry expression were shown in randomized order. The three emotions were all shown in four intensity levels (5, 10, 15, 20) and all neutral faces were part of the fifth intensity level (0). The pictures were each depicted for 15 seconds, after which the four answer categories are shown and the participant can respond by pressing one of four buttons, each corresponding to one of the emotions. A white screen is visible during the 2s interval between pictures. Eye movements and fixations were again measured with the Tobii T120 eye tracker. The areas of interest are grouped into the following categories: *eyes, mouth, face* and *other*. *Face* is defined as a fixation within the face but outside the eyes and mouth. *Other* is defined as a fixation on the screen but outside the areas of interest of the other categories.

Psychophysiology. Electrocardiogram (ECG) and skin conductance (SC) measurements were done at baseline, during the video clips and during the emotion recognition task. During the baseline measurement participants watched a relaxing video clip of an aquarium lasting three minutes. Heart rate (HR) and SC were acquired with the Biopac System and analyzed using Acqknowledge software. Before electrodes were attached, participants washed their hands. SC electrodes were placed on the middle phalanges of the ring and middle fingers of the participant's non-dominant hand. HR electrodes were placed on the participant's chest, one on the lower left side and one on the upper middle side.

Procedure

The tests mentioned above were part of a larger test battery. The relaxing video clip was shown first, then the other video clips and finally the emotion recognition task. Before the relaxing video participants were instructed to sit quietly, watch the video and try to relax. As instruction before the empathy evoking video clips, participants were told they were going to watch four short videos and would have to answer several questions about each video. Also, they were instructed to sit quietly and minimize their movements during the empathy videos and during the emotion recognition task.

Data analysis

ECG. The electrocardiogram signal was high-pass filtered (0.5 Hz). The peak of the R-wave was detected online and transferred to heart rate signal in beats per minute (BPM). Data was visually inspected and corrections were made for extreme heart periods (due to missing or false R-waves). The mean BPM during each empathy evoking video clip was compared to the mean BPM during the baseline measurement using repeated measures analyses. Furthermore, the root mean square of successive differences (rMSSD) was used as a measure of heart rate variability. This measure is sensitive to fluctuations in heart rate in the respiratory frequency range and can be used as an indicator of vagal cardiac control (Task Force of the European Society of Cardiology and the North American Society of Pacing and Electrophysiology, 1996). The mean rMSSD in ms during the baseline video was calculated and compared to the mean level during each empathy evoking video. Besides that, a change score of BPM and rMSSD was created for each video by subtracting the baseline level of BPM and rMSSD from the mean level during the video.

Skin conductance. Both tonic and phasic components of skin conductance were examined: skin conductance level (SCL), skin conductance responses (SCRs) and area under

the curve (AUC). The mean SCL (in μS) for each empathy evoking video clip was compared to the mean SCL during baseline with a repeated measures analysis. The number of SCRs per minute (with a minimum of $0.05 \mu\text{S}$) and AUC (in $\mu\text{S} / \text{s}$, after applying a 0.5 - 2 Hz band-pass filter) were also calculated for each video clip separately and compared to the number of SCRs per minute and AUC during baseline, using repeated measures analyses. Furthermore, a change score of SCL, number of SCRs per minute and AUC was created for each video by subtracting the baseline level of these variables from the mean level during the video. The natural log of the mean level of SCL and of AUC were calculated to normalize the data distribution of these variables.

Specific SCRs during the emotion recognition task were defined as the maximum amplitude in μS between 1 and 6 s after stimulus onset, using the mean between 0 and 1 s as a baseline and a minimum increase criterion of $0.05 \mu\text{S}$. The number and mean amplitude of these specific SCRs was calculated and compared between neutral faces and those with an emotional expression. Besides that, the magnitude of skin conductance responses was calculated, which is equal to the amplitude but without the criterion of a minimum increase. The natural log of the amplitude and magnitude of SCRs were calculated to normalize the data distribution of these variables. All statistical analyses are conducted with the normalized variables, but means and standard deviations of the original variables are given to increase interpretability.

Self rating of empathy videos. An empathy score was calculated based on the self rating questionnaires that participants completed after each empathy video. This empathy score consists of three components: recognizing the two most important emotions of the main character, feeling emotions concurrent with the emotions of the main character and giving an empathic reason for one's own emotion(s). If participants gave the two most important emotions of the main character a higher score than the other emotions on the list, they received two points (one point per emotion). Furthermore, they received two points if the emotions they indicated as strongest and second strongest for themselves, were the same emotions as those which they indicated as strongest and second strongest for the main character (one point for each emotion). Finally, they received a point if the reason they gave for their own emotion(s) was empathic, for example when they said they felt bad or happy for the main character or they wondered what it would be like for him/her to experience that. This leads to a maximum score of 20 points, 5 for each video. The reasons participants gave were coded by a second coder who was blind to the clinical status of the participants. Interrater agreement was 94.6%. Disagreements were settled through discussion.

Statistical analyses. Participants who scored at least three standard deviations above or below the mean were considered outliers and removed from the analysis. Heart rate data of one control participant and part of the heart rate data of one participant with Klinefelter syndrome and of one control participant were eliminated due to technical difficulties during recording. Skewed variables, with a skewness or kurtosis outside the range of -3 to 3, were analyzed with non-parametric tests, unless they could be normalized with transformations. Since education level differed between the Klinefelter and control group, the relation between this variable and all outcome variables was examined. Only for the three outcome variables that were related to education level ($r < .05$), analyses were done including *Education* as a covariate.

Psychophysiology variables of the empathy videos (BPM, rMSSD, SCL, AUC and number of SCRs per minute) were analyzed using repeated measures analyses with two levels. The average during the baseline measurement was included as the first level and the average during the empathy video as the second level. Main effects of condition (change from baseline to the video) were examined as well as interactions between group (Klinefelter group versus control group) and condition. Repeated measures was also used to analyze the psychophysiology variables of the emotion recognition test (number, mean amplitude and magnitude of SCRs), with the average during neutral faces as level one and the average during emotional faces as level two. Again, main effects of condition and group by condition interactions were examined.

Results

Empathy videos

Fixations. All empathy videos taken together, there were no differences in fixation time between the Klinefelter group and the control group on any of the areas of interest (*Body*: $t(26) = .37, p = .71$; *Face*: $t(26) = .74, p = .47$; *Eyes*: $t(26) = 1.41, p = .17$; *Mouth*: $t(26) = -.75, p = .46$; *Hands*: $t(26) = -.83, p = .41$; *Objects*: $t(26) = -.40, p = .70$; *Other*: $t(25) = .36, p = .73$). See Table 1 for the means and standard deviations per AOI.

When each empathy video was analyzed separately, there were some differences in fixation times between the groups. These differences were all on the border of significance but had medium effect sizes. In the video with as main emotions pain and surprise participants with Klinefelter seemed to spend more time fixating on objects compared to controls ($t(25) =$

Table 1. Total fixation time in seconds per area of interest

	Fixation time <i>Body</i> (<i>SD</i>)	Fixation time <i>Face</i> (<i>SD</i>)	Fixation time <i>Eyes</i> (<i>SD</i>)	Fixation time <i>Mouth</i> (<i>SD</i>)	Fixation time <i>Hands</i> (<i>SD</i>)	Fixation time <i>Objects</i> (<i>SD</i>)	Fixation time <i>Other</i> (<i>SD</i>)
Control group	69.33 (6.45)	71.88 (13.16)	37.41 (25.52)	34.22 (25.94)	3.08 (1.81)	17.68 (4.08)	95.37 (8.30)
Klinefelter group	67.95 (12.34)	68.15 (13.61)	25.48 (18.79)	41.71 (27.10)	3.61 (1.55)	18.37 (5.07)	93.90 (13.04)

-1.76, $p = .09$, Cohen's $d = .69$). Also, there was a trend towards a difference in fixation time within the AOI *face* ($t(26) = 1.87$, $p = .07$, Cohen's $d = .71$), with the Klinefelter group spending less time fixating on the face than controls. In the video that mainly showed happiness and cheerfulness, as well as in the video with sadness and upset as main emotions, the Klinefelter group seemed to spend less time fixating on eyes than the control group ($t(26) = 1.74$, $p = .09$, Cohen's $d = .66$ and : $t(19.27) = 1.93$, $p = .07$, Cohen's $d = .75$ respectively).

Empathy score. Participants with Klinefelter syndrome had a lower empathy score than participants without Klinefelter ($t(25) = 3.38$, $p < .01$, Cohen's $d = 1.32$). The groups differed in their score for similarity between own and character's emotion and in their score for the reason they gave for their emotion(s) ($t(26) = 2.09$, $p < .05$, Cohen's $d = .79$ and $t(21.21) = 3.63$, $p < .01$, Cohen's $d = 1.37$ respectively). There was trend towards a difference between the Klinefelter group and controls in their score around recognizing the main character's emotions ($t(25) = 1.81$, $p = .08$, Cohen's $d = .70$).

Linear regression analyses showed that participants' fixation patterns, mainly their fixation time on eyes, mouths and objects, were related to their empathy scores (Klinefelter group and control group taken together). Controlled for fixation time on other AOIs, the time spent looking within the eye region and within the mouth region were positive predictors of the total empathy score ($\beta = .89$, $t(19) = 2.08$, $p = .05$, part $r = .40$ and $\beta = 1.10$, $t(19) = 2.21$, $p < .05$, part $r = .42$ respectively). Also, controlled for fixation time on all other AOIs, fixation time on eyes positively predicted recognition of the main character's emotion(s) ($\beta = .83$, $t(19) = 2.09$, $p = .05$, part $r = .38$). Fixations time on the eye and mouth region were also positive predictors of the empathy score around the reason participants gave for their emotion(s), whereas fixation time on objects was a negative predictor of this score (again controlled for fixation time on all other AOIs; $\beta = 1.00$, $t(20) = 2.56$, $p < .05$, part $r = .45$ and $\beta = 1.33$, $t(20) = 3.01$, $p < .01$, part $r = .53$ respectively).

Table 2. Means and standard deviations of psychophysiology variables during each video and comparisons of empathy videos with baseline (main effects of condition)

	Baseline-video		Happiness-video		Sadness-video			Fear-video			Pain-video		
	<i>M</i> (<i>SD</i>)	<i>M</i> (<i>SD</i>)	<i>F</i> ^a	η^2	<i>M</i> (<i>SD</i>)	<i>F</i> ^a	η^2	<i>M</i> (<i>SD</i>)	<i>F</i> ^a	η^2	<i>M</i> (<i>SD</i>)	<i>F</i> ^a	η^2
SCL (μ S)	4.68 (3.35)	7.11 (4.66)	55.25***	.68	7.46 (5.20)	68.42***	.73	6.81 (3.90)	66.20***	.72	8.15 (6.18)	90.90***	.78
AUC (μ S /s)	1.73 (3.70)	1.25 (1.88)	.13	.01	1.35 (1.84)	.13	.01	1.41 (2.21)	.18	.01	3.16 (4.47)	28.46***	.52
Number of SCRs (/min)	3.05 (3.29)	4.83 (3.55)	45.84***	.64	4.89 (3.31)	52.75***	.67	4.33 (2.61)	62.11***	.71	8.97 (4.61)	92.69***	.78
BPM	67.12 (9.66)	68.16 (9.78)	2.96 ⁺	.11	66.86 (9.66)	.51	.02	67.65 (9.28)	.40	.02	65.47 (8.41)	5.93*	.19
rMSSD (ms)	43.01 (22.79)	36.93 (17.27)	7.05*	.23	40.33 (21.14)	.78	.03	40.00 (20.86)	.84	.03	45.67 (26.40)	.35	.01

Notes. Means and SDs of original variables are presented, but F-tests are conducted with normalized variables.

^a Degrees of freedom are 1 and 26 for all analyses of SCL, AUC and number of SCRs; 1 and 25 for BPM analyses of the happiness-, fear- and pain-video; 1 and 24 for rMSSD analyses of the happiness-, fear- and pain-video and for the BPM analysis of the sadness-video; and 1 and 23 for rMSSD analysis of the sadness-video.

⁺<.10, *<.05, ***<.001

Psychophysiology. For all empathy videos there was a main effect of condition on skin conductance level (SCL): the videos led to an increase in SCL compared to the level during baseline measurement (for these and all other main effects of condition, see Table 2). The group by condition interactions for SCL were not significant, meaning that the increases in SCL did not differ between the Klinefelter and control group (happiness-video: $F(1,26) = .14, p = .71$; sadness-video: $F(1,26) = .39, p = .54$; fear-video: $F(1,26) = .04, p = .84$; pain-video: $F(1,26) = .58, p = .45$).

With regard to number of skin conductance responses there was a main effect of condition for the happiness-video: this video caused an increase in the number of SCRs (see Table 2). The increase in number of SCRs seemed stronger for the control group and this interaction was on the border of significance ($F(1,26) = 3.35, p = .08, \eta^2 = .11$). The video depicting sadness as main emotion led to an increase in the number of SCRs, which was equal for both groups (thus a main effect of condition, see Table 2, but no interaction: $F(1,26) = 2.15, p = .16$). The fear-video also caused an increase in the number of SCRs (see Table 2) that did not differ between groups ($F(1,26) = 1.88, p = .18$). Concerning AUC there was a significant group by condition interaction for this video: it led to a decrease in AUC for the control group, but an increase for the Klinefelter group ($F(1,26) = 4.76, p < .05, \eta^2 = .16$, see Figure 1). This resulted in a non-significant main effect of condition (see Table 2). The video with pain as main emotion caused an increase in both AUC and the number of SCRs, the increase in AUC was stronger for the Klinefelter group compared to the control group (a significant group by condition interaction: $F(1,26) = 4.81, p < .05, \eta^2 = .16$, see Figure 2).

Heart rate decreased from baseline to the pain-video (see Table 2; equal effect for both groups: $F(1,25) = 1.23, p = .28$). Heart rate did not change from baseline to any of the other videos. Heart rate variability (rMSSD) decreased from baseline to the happiness-video (equal effect for both groups: $F(1,24) = 1.23, p = .31$), but did not change for any of the other videos.

Relation between psychophysiology and self-report. Participants' skin conductance reactivity to the empathy videos was significantly related to their own reports of emotionality (the average score of the two strongest felt emotions per video), but only in the control group. For example, stronger increases in AUC from baseline to the pain-video were in the control group associated with higher self-reports of emotionality about that video ($\rho(12) = .68, p < .01$). The same was true for the video with fear as main emotion ($\rho(12) = .55, p < .05$). Furthermore, the increase in the number of SCRs during the fear-video was in the control group positively related to self-reported emotionality about that video ($r(12) = .75, p < .01$). Also, a stronger increase of SCL during the happiness-video was associated with stronger self-reported

emotions of that video ($r(12) = .54, p < .05$). In the Klinefelter group, however, there were no significant relations between participants' skin conductance reactivity and their own reports of emotion for any of the videos. Changes in heart rate and rMSSD were not related to self-reports in either of the groups.

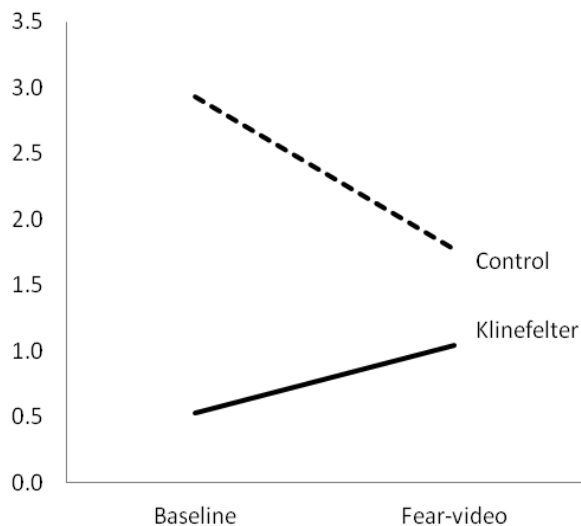


Figure 1. Change in AUC ($\mu S/s$) from baseline to fear-video for both groups separately.

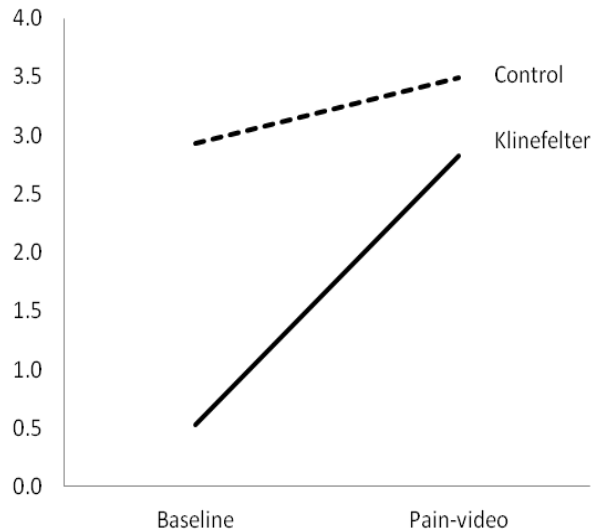


Figure 2. Change in AUC ($\mu S/s$) from baseline to pain-video for both groups separately.

Emotion recognition task

Accuracy. Overall accuracy on the emotion recognition task did not differ between the Klinefelter group and controls (Klinefelter group $M = .69, SD = .05$, control group $M = .71, SD = .06$; $t(26) = .78, p = .44$). Accuracy for the different emotions did not differ between the groups either (happy: $t(19.26) = .03, p = .98$; fearful: $t(26) = .51, p = .61$; angry: $t(26) = .94, p = .36$; neutral: $t(26) = -.69, p = .50$). A repeated measures analysis showed that accuracy increased with increasing intensity of the emotion ($F(2.04, 53) = 443.78, p < .001, \epsilon = .68$, partial $\eta^2 = .95$), which applied equally for both groups ($F(2.04, 53) = .93, p = .21$).

Fixations. Participants with Klinefelter syndrome differed from control participants on fixation times in the eye region and outside the face ($t(25) = 2.91, p < .01$, Cohen's $d = 1.12$; $t(25) = -2.07, p < .05$, Cohen's $d = .80$). Participants with Klinefelter syndrome spent less time fixating within the eye region of the face and more time fixating outside the face (but on the screen) compared to controls (see Table 3). The groups did not differ in fixation time on the mouth or on other areas of the face ($t(25) = .92, p = .64$; $t(25) = .80, p = .57$). Besides that, the area in which participants made their first fixation differed between the Klinefelter group and the control group. Participants with Klinefelter had less first fixations on the eye region and more first fixations outside the face ($t(25) = 2.47, p < .05$, Cohen's $d = .95$; $t(25) = -2.74$,

$p < .05$, Cohen's $d = 1.06$, see Table 4). There were no differences between groups concerning the percentage of first fixations on mouth and other areas of the face ($t(25) = .47, p = .37$; $t(25) = -.58, p = .43$).

Table 3. Mean fixation time in seconds per area of interest

	Fixation time Eyes (SD)	Fixation time Mouth (SD)	Fixation time Face (SD)	Fixation time Other (SD)
Control group	4.79 (2.06)	1.73 (1.28)	1.20 (.54)	1.89 (1.33)
Klinefelter group	2.46 (2.10)	1.30 (1.16)	1.03 (.56)	3.33 (2.15)

Table 4. Percentage of first fixations per area of interest

	% of first fixations on Eyes (SD)	% of first fixations on Mouth (SD)	% of first fixations on Face (SD)	% of first fixations on Other (SD)
Control group	48.08 (23.25)	18.65 (16.35)	7.31 (4.70)	25.96 (16.24)
Klinefelter group	27.50 (19.98)	15.63 (16.90)	8.93 (8.97)	47.95 (24.35)

Psychophysiology. Participants with and without Klinefelter showed similar patterns of skin conductance reactivity during the emotion recognition task. The number and mean amplitude of specific SCRs as well as the magnitude of SCRs did not differ between the two groups ($t(26) = 1.40, p = .17, t(25) = .63, p = .53$ and $t(26) = 1.17, p = .25$ respectively). Repeated measures analyses showed that the number of SCRs to neutral faces was equal to that of faces with an emotional expression (Klinefelter group and control group taken together, $F(1,26) = .07, p = .80$). The mean amplitude and the magnitude of SCRs, on the other hand, was higher for faces with an emotional expression than for neutral faces (main effect of condition for amplitude of SCRs: $F(1,20) = 4.46, p < .05$, partial $\eta^2 = .18$, and for magnitude of SCRs: $F(1,24) = 5.54, p < .05$, partial $\eta^2 = .19$, see Table 5). There were no condition by group interactions, meaning that the two main effects of condition were equal for the Klinefelter and control group ($F(1,20) = .01, p = .94$. and $F(1,24) = .13, p = .73$ respectively).

Table 5. Means and standard deviations of skin conductance variables for neutral faces and emotional faces (average of all emotions)

	Number of SCRs (SD)	Mean SCR amplitude in μ S (SD)	Mean SCR magnitude in μ S (SD)
Neutral faces	5.79 (4.43)	.27 (.37)	.12 (.24)
Emotional faces	5.96 (2.92)	.34 (.39)	.13 (.23)

Discussion

From previous research, it is known that individuals with Klinefelter syndrome often have problems in social and emotional functioning that persist into adulthood, such as problems with identifying their own and other's emotions and an elevated level of autistic traits. However, this knowledge is mainly based on self-report measurements. In this research the social-emotional difficulties of men with Klinefelter were explored further by measuring their social attention, empathy, emotion recognition skills and emotional arousal.

Social attention differed between the Klinefelter group and the control group mainly while they were watching photographs of faces and less clearly while they viewed the empathy evoking video clips. The differences found in this study are in the same direction as those found in previous research with people with autism (Dalton et al., 2005; Klin, et al., 2002): the clinical group looked less at the most important social cue, namely eyes, and more at objects or other parts of the screen that hold no social information. The fact that the differences in fixation patterns were more clearly visible during the emotion recognition task than while watching the videos is unexpected. Previous research on social attention of individuals with autism have found differences in fixation patterns for both static (photographs of faces) and dynamic stimuli (videos) (Boraston, & Blakemore, 2007). The difference in instructions given to the participants ('just watch' versus 'recognize the emotion') could have an effect on viewing behaviour, but in a research by Pelphrey et al. (2002) differences in fixation time between people with and without autism were visible irrespective of the type of instruction. Another explanation for the result in the research presented here could be that during the empathy videos the maximum time that participants could look at eyes or mouths was less than 400 seconds, but during the emotion recognition task eyes and mouths were visible for 1200 seconds in total. When the AOIs are present on the screen for a longer period of time, this increases the power to detect differences between groups in fixation time.

People who spent more time looking at eyes and mouths and less time looking at objects had better empathy scores. This proves that social attention is important for social-cognitive skills. Also, it is in line with the results from studies on autism, which showed that higher fixation time on eyes was associated with better social functioning (Klin et al., 2002).

The Klinefelter group had difficulty responding in an empathic way to video clips, as reflected in their own emotions being less concordant with the main character's emotions and in showing less empathy in the reasons they gave for their emotion(s). Besides that, the

Klinefelters' self-reports of emotionality did not relate to their psychophysiological responses, whereas these relations were present in the control group. This indicates that men with Klinefelter might have difficulty determining and describing their own emotionality, which is in line with results of previous self-report research (Van Rijn, et al., 2006). These findings together form further proof that men with Klinefelter have deficits in social-cognitive functioning. These deficits can be a good focus of an intervention for men with Klinefelter syndrome; strengthening social-cognitive skills might improve their adaptive functioning and quality of life.

Emotion recognition skills, on the other hand, were equal for the Klinefelter group and the control group, although differences have been found in earlier research (Van Rijn, et al., 2006; Van Rijn, et al., 2007). Possible explanations for the lack of differences in the current study are the fact that participants watched each face for a relatively long time (15 seconds), they could not respond until those 15 seconds had passed and reaction times could therefore not be analyzed. Future research examining the emotion recognition skills of people with Klinefelter syndrome might better use a design that includes a brief presentation of each face and a time pressure element in responding. Also, a broader range of emotions can be studied. This way possible differences in the speed of emotion recognition or in the recognition of certain (more complex) emotions could be detected.

Psychophysiological responses to the video clips and faces were quite similar for participants with and without Klinefelter. The few differences that were found, during the empathy videos, indicated stronger skin conductance responses for the Klinefelter group. Men with Klinefelter syndrome thus seem to be more aroused by other people's emotions in videos. This is in line with the result from a previous study in which men with Klinefelter reported to be more distressed during social situations (Van Rijn et al., 2008). Higher arousal may be a reason to avoid situations in which you or the other person can become emotional, for example during the expression of negative feelings. People with Klinefelter syndrome have been found to have fewer interactions in which they express negative feelings (Van Rijn et al., 2008).

However, while watching photographs of faces with an emotional expression the arousal of men with Klinefelter was not stronger than that of controls. This might be due to the static and less ecologically valid nature of these stimuli. More research is needed to replicate these findings and to further investigate the psychophysiological responsiveness within Klinefelter syndrome. Also, because the XXY genotype is associated with low testosterone levels, a large part of adults with a diagnosis of Klinefelter takes medication to

increase testosterone levels (Bojesen, et al., 2006). Therefore, in future research it is important to take into account medication use to examine if this has an influence on the cardiac or skin conductance reactivity of those with Klinefelter syndrome.

Finally, the number of participants in the current research was small, so findings must be replicated in larger groups. Also, only a quarter of boys and men with an extra X-chromosome receive the diagnosis of Klinefelter syndrome (Bojesen, et al., 2003), and nothing is known about the social-emotional functioning of the other three quarters. It might therefore not be possible to generalize the results to those who have an extra X-chromosome but no diagnosis of Klinefelter.

This study found proof for social-cognitive difficulties in Klinefelter syndrome, and a deviant social attention that might be one of the bases of these difficulties. The results can give insight into the role of the X-chromosome in the relation between (social) perception and attention processes and social-cognitive functioning. Moreover, it proves the importance of addressing social-cognitive skills in interventions for Klinefelter syndrome. Stronger skin conductance reactivity to emotional situations might be another factor underlying the social-cognitive problems, but future studies should investigate this factor further as well as its relation to social-emotional functioning at the behavioural level.

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