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ORIGINAL PAPER

Is Theory of Mind Understanding Impaired in Males with Fragile X Syndrome?

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Abstract Males with fragile X syndrome (FXS) have difficulties with social interaction and many show autistic features. This study examined whether the social deficits characteristic of FXS are associated with theory of mind difficulties. Two groups of boys with FXS participated: a group with few autistic features and a group with many autistic features. An intellectual disability control group also participated. In addition to using standard theory of mind tasks, new techniques were used that were able to separate out the various processing demands of the task (e.g., memory, inhibitory control). Overall, the findings indicate that both groups of boys with FXS have difficulty with theory of mind tasks compared to an intellectual disability control group. However, both groups with FXS also performed worse on comparison trials that required working memory but not theory of mind. Theory of mind difficulties are likely to be an important aspect of the FXS clinical profile, but are most likely the result from a more basic difficulty with working memory.

Keywords Fragile X syndrome · Autistic spectrum disorder · Theory of mind · Inhibition · Working memory

Introduction

Fragile X syndrome (FXS) is a well-recognized cause of intellectual disability and developmental delay in males and females. Numerous studies have attempted to identify a specific cognitive-behavioural phenotype associated with FXS (reviewed in Bennetto & Pennington, 2002). In affected males over 90% will present with intellectual disability usually in the mild-moderate range, compared with the majority of females with full mutation FXS who have IQs that fall within the low average range of normal ability (Bennetto & Pennington, 2002). Although FXS is present at birth, its behavioural manifestations are often not apparent until late in the first year of life. The fragile-X behavioural phenotype is still being defined but notable characteristics include social difficulties (e.g. deficits in social interactions with peers and social withdrawal) and autistic-like behaviours that include poor eye contact, stereotypic movements and odd communication, including echolalia and perseverative speech (e.g. Belser & Sudhalter, 2001; Sudhalter & Belser, 2001).

Much of the research into cognitive impairment in FXS has focused upon executive function. Executive function includes processes such as maintaining and updating information in working memory, cognitive flexibility, planning, initiation, inhibition and attentional regulation, in short, abilities that are engaged in the generation and monitoring of goal-directed behaviour. Numerous studies have reported impaired executive functioning abilities in males and females with FXS (e.g., Bennetto, Pennington, Porter, Taylor, & Hagerman, 2001; Loesch, Bui, Grigsby, Butler, Epstein, Huggins et al., 2003). Studies of children with autism and typically developing children have shown executive functioning to be related to social cognitive abilities and in particular, theory of mind abilities (e.g., Russell, 1997; Perner & Lang, 2000; Carlson & Moses, 2001). More recently, researchers have also begun to examine whether individuals with FXS might also have impaired theory of mind abilities.

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Theory of mind and Fragile X

It is increasingly recognised that reasoning about mental states such as beliefs, desires and knowledge (often referred to as "theory of mind") is central to a range of social-cognitive activities including the ability to communicate and to explain and predict behaviour (Astington, 2000; Sperber, 2000). A task commonly used to examine mental state reasoning in children requires the inference that someone has a false belief (Wimmer & Perner, 1983). For example, the child might be told a story where Simon puts his chocolate in the cupboard, then goes outside to play. While he is away his mother moves the chocolate to the fridge. The child is asked where Simon will first look for his chocolate when he returns. To answer correctly, they must infer that Simon thinks that the chocolate is still in the cupboard. Many four-year-olds answer correctly, while many three-year-olds judge incorrectly that he will look in the fridge (i.e., they answer from their own knowledge and not the perspective of the other person). Distinctive impairment on false belief, and other theory of mind tasks, is often also observed in individuals with autism and it has been suggested that this deficit could account for the triad of impairments in socialisation, communication and imagination (Baron-Cohen, 2000; Yirmiya, Erel, Shaked, & Solomonica-Levi, 1998). It is estimated that 15–25% of boys with FXS meet the diagnostic criteria for autism (e.g., Bailey et al., 1998; Dykens & Volkmar, 1997). An important issue for studies of theory of mind in FXS is whether any deficit in theory of mind is an artifact of the high co-morbidity between FXS and autism or if such deficits are characteristic of FXS irrespective of autistic features. Several studies have found that individuals with intellectual disability but without autism also perform poorly on false belief tasks (Yirmiya et al., 1998). The issue of whether the poor performance of individuals with intellectual disability reflects theory of mind deficits or difficulties with task demands is unresolved.

Three research groups have investigated theory of mind ability in people with FXS, yielding inconsistent results. Mazzocco, Pennington, and Hagerman (1994) compared 19 adult females with FXS with a group of 27 women carrying the fragile X gene and a group of 56 control women who tested negative for the gene. All participants had IQ's of 70 or above and no participants were reported to have a diagnosis of autism. The study found that performance on a perspective-taking task was related to IQ rather than fragile X group status. However, the perspective-taking task used in this study was originally designed for children so may have lacked the sensitivity to detect subtle deficits in theory of mind in adult participants. Garner, Callias, and Turk (1999) addressed theory of mind in a group of eight boys with FXS (without autism) and reported that significantly more children with FXS failed a first order false belief task (Smarties task) than a comparison group of eight children with intellectual disability of unknown aetiology matched on chronological age and verbal mental age. However, on another first order false belief task (the Sally-Anne task) and a second-order false belief task there were no statistically significant group differences. Only seven of the eight children in each group completed the Sally-Anne task, of which five boys in the FXS group failed the task, compared to one boy in the control group. The authors suggested that their findings did not allow them to assess whether theory of mind deficits were a specific feature of the syndrome since performance on one of the tasks was related to overall level of ability. A third study by Cornish, Burack, Rahman, Munir, Russo, & Grant (2005) compared the performance of children with FXS (without autism) with children with Down's syndrome matched on chronological age and verbal mental age on two trials of the Sally-Anne false belief task. The children with FXS performed similarly to children with Down's syndrome, with just under half the children in each group passing the task. The authors concluded that while children with FXS did have difficulties with theory of mind, the deficit was not as severe as that previously reported for children with autism, who would have been expected to perform less well than children with Down's syndrome on false belief tasks. Taken together, these studies suggest that children with FXS often *do* make errors on false belief tasks. However, it is unclear whether this pattern is a distinctive feature of the FXS cognitive phenotype, or whether it only reflects the intellectual disability that is commonly apparent in children with FXS. A further issue is that the severity of autistic features in the samples employed in Garner et al. (1999) and Cornish et al. (2005) was not reported. Although these studies included participants with FXS who did not have a diagnosis of autism, some measure of autistic features would have been useful since boys with FXS who do not meet the diagnostic criteria for autism often still display significant autistic features (Dykens & Volkmar, 1997).

Why might individuals with FXS fail false belief tasks?

The rationale for the present investigation is that, besides the inconsistent findings in existing studies, there are also methodological reasons why these studies might not be able to give a clear picture of the existence or the origins of theory of mind difficulties in FXS. First, the standard false belief tasks used in these, and most other studies of belief reasoning, require participants both to reason about false beliefs (that Simon thinks the chocolate is still in the cupboard) *and* to resist interference from their own knowledge (that in fact the chocolate is now in the fridge). The need to resist inference places demands upon executive function. This confound is particularly relevant because children with FXS have well-attested impairments in executive function that are disproportionate to their general level of intellectual impairment

(Cornish, Sudhalter, & Turk, 2004). If children with FXS have disproportionate difficulty resisting interference from their own perspective, this alone could cause them to perform less well on standard false belief tasks than comparison groups matched simply on general intellectual disability.

A second reason why it is difficult to reach clear conclusions from existing studies of false belief reasoning in children with FXS is that false belief tasks make substantial demands on working memory. Although the standard false belief tasks used by Garner et al. (1999) and Cornish et al. (2005) included check questions to ensure that the participant could remember crucial facts about the story, remembering these facts is not the only demand that the false belief task makes on working memory. It is also necessary to remember the sequence of events, which is not measured adequately by check questions. Since children with FXS have impairments in working memory that are disproportionate to their general level of intellectual impairment (Munir, Cornish, & Wilding, 2001), it could be that any difficulty they have on false belief tasks arises from working memory impairment, rather than a more specific "theory of mind" impairment.

These considerations suggest two distinct questions about whether theory of mind problems are part of the cognitive phenotype of FXS. One question is whether children with FXS are impaired on theory of mind tasks (such as false belief tasks), in comparison to children with a similar general level of intellectual ability. This is an important clinical question addressed (albeit inconclusively) in existing studies. The second question concerns the nature of any theory of mind deficit. Some authors propose that theory of mind depends upon a dedicated cognitive system with a discrete neural basis (for a discussion of this issue see e.g., Apperly, Samson, & Humphreys, 2005; Frith & Frith, 2003; Saxe, Carey, & Canwisher, 2004). Theory of mind impairment in FXS might be the result of damage to such a system. It is also widely recognised that theory of mind abilities depend critically upon more general cognitive processes, such as working memory and inhibitory control. Thus, theory of mind impairment in FXS may be the indirect result of a primary impairment to these processes.

The present study

The present study employed two first order standard false belief tasks, plus two versions of a video-based, non-verbal false belief reasoning task. Inclusion of two first order standard false belief tasks (the location-change task and the deceptive box task) allowed us to gauge the performance of children in this sample in comparison with a large number of previous studies. The standard tasks included memory check questions within each trial, though for the reasons already discussed, these were considered a rather weak test of the contribution of working memory problems to the errors children with FXS might make on false belief tasks. Two versions of the video-based tasks were used, "reality known" and "reality unknown". Inclusion of the video-based tasks allowed us to address our first question by assessing belief reasoning over a wider range of tasks than in previous studies.

Importantly, the video-based tasks gave us two ways to investigate whether individuals with FXS showed a primary ToM deficit, or a deficit that was a consequence of impairment to more general cognitive resources such as working memory and inhibitory control. First, they included separate comparison trials that did not require belief reasoning, but that made demands on working memory that corresponded to specific incidental demands of the false belief trials. If individuals with FXS only showed impaired belief reasoning because they struggled to meet the incidental processing demands of false belief tasks, then we would expect similar levels of performance in false belief and comparison trials. Another possibility was that the same cognitive resources (e.g., working memory) that were necessary for handling the incidental processing demands of false belief tasks were also necessary for belief reasoning itself. If this were the case, we might observe greater impairment on false belief trials than on comparison trials (because false belief trials would make greater overall demands on impaired cognitive resources), but this impairment should be *in proportion* to the level of impairment on comparison trials. Finally, if impairment on false belief trials was *disproportionate* to impairment on comparison trials then this would be evidence of a primary deficit in "theory of mind" that was not merely the consequence of impaired working memory.

Second, our video-based tasks allowed us to assess whether individuals with FXS had particular difficulty resisting interference from knowledge of reality when reasoning about false beliefs. Resisting interference from knowledge of reality is an inhibitory demand that is thought by many to be a key problem in false belief reasoning (Carlson & Moses, 2001), and this demand was present in the reality-known false belief task (as well as in the standard false belief tasks) but absent in the reality-unknown false belief task. If individuals with FXS performed proportionately better on false belief tasks when they did not need to resist interference from knowledge of reality this would suggest that a primary deficit with inhibitory control was responsible for at least some of their "theory of mind" problems.

In accordance with the findings of Garner et al. (1999) and Cornish et al. (2005) that males with FXS (without autism) performed similarly to other children with intellectual disability on tests of theory of mind, it was predicted that boys with FXS (without autism) will not have a specific theory of mind deficit. It was predicted that their performance would be related to the executive demands (i.e., working memory and inhibitory control) of the task. However, on the basis of studies of children with autism that have reported a specific deficit in theory of mind (Yirmiya et al., 1998), it was predicted that boys with FXS who scored above the clinical cut-off for autism on a screening questionnaire, would show theory of mind impairments and executive functioning deficits.

Method

Participants

Three groups of children took part in the study: 15 boys with fragile X syndrome (FXS) who displayed few autistic features (full mutation (FMR-1) and fully methylated); 15 boys with fragile X syndrome (FXS-A) with significant autistic features (full mutation (FMR-1) and fully methylated); 15 boys with intellectual disability (ID) of unknown aetiology who had no history of autism or FXS. Each participant was assessed for autism using the Social Communication Questionnaire (SCQ: Berument, Rutter, Lord, Pickles, & Bailey, 1999), which is a measure that can be completed by parents and can be done without the guidance of professionals. The SCQ was designed as a screening measure for the Autism Diagnostic Interview-Revised (ADI-R: Lord et al., 1994) and has been shown to have high diagnostic validity. The authors suggest a cut-off point for autism spectrum disorders of 15. This score was found to differentiate individuals with autism from other diagnoses, excluding individuals with intellectual disability, with a specificity of .80 and a sensitivity of .96. The same cut-off point was found to differentiate individuals with autism from individuals with intellectual disability without autism, with a specificity of .67 and a sensitivity of .96 (Berument et al., 1999). The screen is suitable for participants above the chronological age of 4 years and a mental age of 2 years.

Families of children with FXS were contacted through the Fragile X Society. Consenting participants completed an initial screening questionnaire that was designed to provide information on their child's developmental level and symptoms associated with autism (using the SCQ). The aim was to recruit boys who were of sufficient verbal ability to participate and to separate boys with FXS unlikely to have autism from those who are likely to meet the diagnostic criteria for autism. Boys who were rated by parents as verbal and able to use simple sentences were included. Whilst SCQ suggests that a score of 15 and above is a possible indicator of autism, the standardisation data showed that the mean score for children with autism was 24.2. Consequently, boys with FXS were included in the FXS group if they scored 14 or below on their total SCQ score. Boys with FXS with an SCQ score of 25 or above were included in the FXS-A group. From 110 completed screening questionnaires, 73 were selected on the basis of having sufficient developmental ability. A total of 33 boys who scored between 15 and 24 on the SCQ were not included in the main study and a further 10 children who were eligible for inclusion could not be tested due to a variety of factors including geographical location and child illness. Boys with intellectual disability were recruited from special needs schools. Teachers were asked to nominate boys who had no history of autism or FXS, were verbal and able to use sentences and provided a match for the chronological age of the boys with FXS. A total of 21 boys were assessed on measures of verbal and non-verbal ability. Of this initial sample of 21 boys, 15 boys were selected to complete the full battery on the basis that they provided a good match for the FXS group. All of the 15 boys selected scored 14 or below on the SCQ.

The three clinical groups were pairwise matched on Chronological age (CA) and verbal mental age (VMA) calculated using the long form of the British Picture Vocabulary Scale (BPVS) (Dunn, Dunn, & Whetton, 1997). The first edition of the test was used since the norms for this test are suitable for children up to the age of eighteen years. This test is designed to measure receptive vocabulary and does not require any reading, speaking or writing, only simple responses to picture cards. The child is presented with a series of plates containing 4 pictures and is asked to indicate which is the picture of the word that is said. Reliability and validity of the test are acceptable (see Dunn, Dunn, & Whetton, 1997). It is widely acknowledged that theory of mind development has a close connection with the development of language (e.g., Astington & Jenkins, 1999; De Villiers & Pyers, 2002; Slade & Ruffman, 2005; Smith, Apperly, & White, 2003), though the relative importance of syntax and semantics in this relationship remains controversial. Whilst the BPVS is primarily a measure of receptive vocabulary it has been shown to be a reliable estimate of general language ability in children with autism (Jarrold, Boucher, & Russell, 1997).

The Coloured Progressive Matrices (Raven, 1965) was used to assess non-verbal mental age. This test is designed to measure a person's ability to form perceptual relations and reason by analogy independent of language. It consists of 36 items arranged in three sets of 12 items each. Each item contains a figure with a missing piece. Below the figure are six alternative pieces to complete the figure, only one of which is correct. This test is known to have high internal consistency and high test-retest reliability (see Raven, 1965). Verbal ability rather than non-verbal ability was chosen to match the groups since verbal mental age has consistently been related to theory of mind development in typically developing children and children with autism (Happé, 1995). In order to be included in the study, participants had a verbal mental age of 4 years or above. Table 1 summarises mean CA, VMA, VIQ and NVMA for each group.

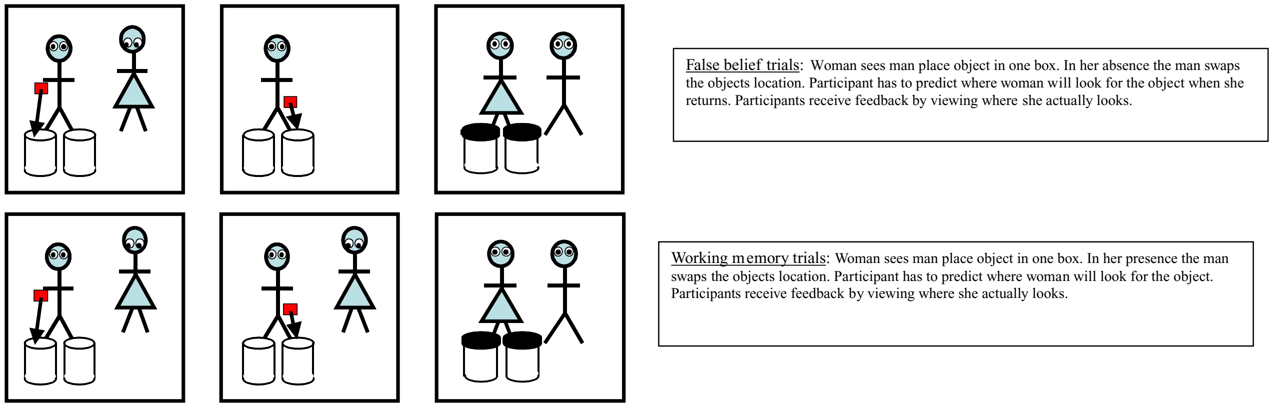

Fig. 1 Reality known task (This figure is adapted and extended from Apperly, Samson, & Humphreys, 2005)

Table 1 Participant characteristics: mean chronological age (CA), verbal mental age (VMA), verbal IQ & non-verbal mental age (NVMA) in years and months (standard deviations are shown in brackets)

Group	C A	VMA	VIQ	NVMA
FXS ($n = 15$)	13:8 (3:0)	6:11 (2.0)	53.9 (13.2)	6:0 (1.3)
FXS-A ($n = 15$)	12:5 (2:6)	6:8 (2.7)	53.4 (12.8)	6:4 (1.9)
ID ($n = 15$)	13:9 (2:9)	6:11 (1.8)	55.8 (13.8)	6:8 (1.10)

Note. ID: Intellectual disability group, FXS: fragile X group, FXS-A: fragile X group with many autistic features.

Materials

The video-based tasks used videos of human actors and were presented on a laptop computer using Windows Media Player software. The location change task (also known as the Sally-Anne task; Baron-Cohen, Leslie, & Frith, 1985) employed two easily distinguishable play-mobile figures, a ball, a small plastic box, and a small plastic cupboard. The deceptive box task (Perner, Frith, Leslie, & Leekam, 1989) employed an egg box and a pen.

Procedure

The boys with FXS were tested in their homes, with the exception of one boy who was tested at school. The group with intellectual disability were contacted through their school and participants were tested at school. Every effort was made to ensure that testing conditions within the home and school environments were similar. Each participant was individually tested in a quiet room, with no interruption and testing was conducted at a table or desk. All the tasks were given in one session, which typically lasted one-hour, inclusive of regular breaks. All participants completed an assessment of verbal and non-verbal ability and a battery of theory of mind tests (two video-based tasks, one location change and one deceptive box task). The order of administration of tasks was BPVS, video-based false belief task (reality unknown or reality known counter-balanced across participants), Deceptive box task, Standard location change task, Raven's Coloured Progressive Matrices and video-based false belief task (reality unknown or reality known counter-balanced across participants).

In the "reality known" version of the video-based task, participants watched a series of short (∼45 s) videos in which they were asked to predict where a character will search for a hidden object (see Samson, Apperly, Kathirgamanathan, & Humphreys, 2005). The task principles were explained to the participant at the beginning of the task, and comprehension was checked with a number of warm-up trials on which corrective feedback was given. Video presentation was controlled manually by the experimenter, enabling the time allowed for responding and the rate of progress to the next video to be adapted to the needs of the participant. Predicting where the woman would look on false belief trials required the participant to follow and remember the transfer of the object from one box to another, and to take account of the woman's false belief while resisting interference from their own knowledge of the object's true location (see Fig. 1). Predicting her search on memory comparison trials also required the participant to follow and remember the transfer of the object, but did not require belief reasoning, since the woman's belief was true. After responding to the test question about where the woman will first look for a hidden object, the participant received feedback by viewing the end of the video clip where the woman opens a box. False belief and memory comparison trials were mixed with two anti-strategy filler trials, designed to minimise the possibility that the participant could learn to solve test trials using superficial strategies. In the "woman-absent" filler trial, the woman left the room and the object was first removed and then replaced in its original box. Thus, a participant who succeeded on false belief trials by pointing to the empty box whenever Character 1 has left the room would fail this trial. In the "woman-present" filler trial, the woman remained in the room and the object was first removed from and then

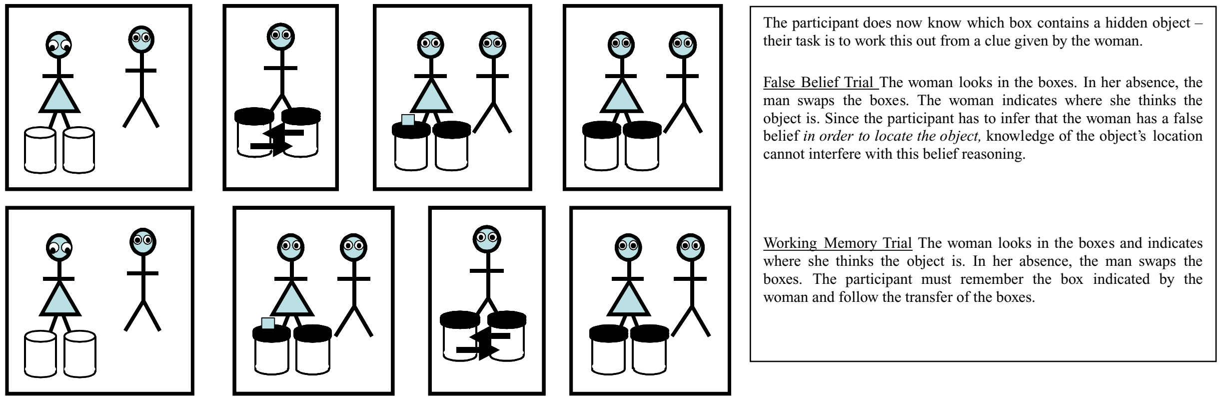

Fig. 2 Reality unknown task (This figure is adapted and extended from Apperly, Samson, & Humphreys, 2005)

replaced in its original box. If a participant passed memory comparison trials by always pointing to the opposite box from the one in which the object was initially placed then they would fail this trial. Thus, in total participants viewed two false belief trials, two memory comparison trials, and two trials of each anti-strategy filler trial. Trials of the same kind were always presented consecutively, and the order of trial presentation was counterbalanced across participants.

In the "reality unknown" false belief task, participants again watched a series of short videos (from Apperly, Samson, Chiavarino, & Humphreys, 2004). This time, although they knew that there was an object in one of two identical boxes, participants did not initially know which. The participant's job was to locate the object, and on each trial a female character in the video gave them a helpful clue by placing a marker on top of the box she thought contained the object (see Fig. 2). The principles of the task were explained and practised in a warm-up phase at the beginning of the task. Locating the object in false belief trials required the participant to take account of the movement of the target object when the boxes were swapped and to take account of the woman's false belief when interpreting her clue about the object's location (see Fig. 2). However, unlike the reality-known false belief task, participants did not know the object's true location, so did not have to resist interference from this knowledge when reasoning about the woman's false belief. Locating the object in memory comparison trials also required the participant to take account of the woman's clue (at a point when the woman had a true belief) and also required the participant to track the movement of the target object when the boxes were swapped. However, the memory comparison trial did not require reasoning about false beliefs. After pointing to where they thought the object was located the participant received feedback by viewing the end of the video clip where the boxes are opened.

False belief and memory comparison trials were mixed with two filler trials designed to check that participants understood the task and could not succeed on false belief trials by adopting superficial strategies. On "inhibition" filler trials the participant already knew the location of the object when the woman (who had a false belief) returned to the room and indicated the wrong box. Thus, to point correctly participants had to resist any tendency to point to the box just indicated by the woman. On "true belief" filler trials the woman had a true belief and so gave a correct clue about the object's location. Since participants received feedback at the end of each trial, these "true belief" trials provided feedback that should have prevented participants from adopting the strategy of always pointing to the opposite box from the one indicated by the woman. In total, participants viewed two trials of each type. Trials of the same type were always presented consecutively, and the order of trial presentation was counterbalanced across participants.

Participants also completed one trial of the standard location change task (Baron-Cohen et al., 1985) and one trial of the deceptive box task (Perner et al., 1989). Details of these tasks are presented in Appendix A.

Results

Participant characteristics

There were no significant differences between groups on Chronological Age ($F(2, 42) = 1.15, p = .32$), Verbal Mental Age ($F(2, 42) = 0.05, p = .95$), Verbal IQ ($F(2, 42) = 0.13, p = .88$) or Nonverbal Mental Age ($F(2, 42) = 0.51, p = .61$).

Comparison of performance on belief reasoning tasks

We initially analysed false belief scores without taking account of performance on control questions for standard false belief tasks or comparison trials for the video-based tasks. This analysis served two objectives. First, it provided a comprehensive test of whether individuals with FXS simply showed an overall impairment in belief reasoning in comparison with matched controls (regardless of whether this impairment was primary, or the result of impairments to executive function or working memory). Second, this analysis allowed us to assess whether the simple absence of the need to resist interference from knowledge of reality (in the reality-unknown video-based task) enabled children to reason more successfully about false beliefs than when they also needed to meet this incidental task demand (in the standard false belief tasks and reality-known video-based task). Scores across the two standard false belief tasks were summed to give one Standard task score. This yielded group means of 0.60 ($SD = .74$), 0.40 ($SD = .63$) and 1.20 ($SD = .77$) for the FXS, FXS-A and Intellectual Disability groups respectively. The mean scores (out of two trials) for each group on the reality known and reality unknown false belief tasks are shown in Table 2. To examine whether performance on false belief tasks was disproportionately impaired in children with FXS, and whether performance varied across the three types of false belief task, a mixed ANOVA with Group (FXS, FXS-A, Intellectual Disability) as the between-participants factor and performance on the three false belief tasks (Standard, reality-known, reality-unknown) as repeated measures was conducted. There was a significant main effect of group, $F(2, 42) = 10.91$, $p < .0001$. Post-hoc LSD comparisons showed both FXS and FXS-A groups to have lower scores than the intellectual disability group ($p = .003$, $p < .001$ respectively), but scores for the FXS and FXS-A groups did not differ from each other ($p = .184$). There was no significant effect of task, $F(2, 84) = .575$, $p = .565$, and no significant interaction between task and group, $F(4, 84) = .263$, $p = .901$. Thus, both the FXS and FXS-A groups performed worse on the three types of false belief tasks than the Intellectual Disability control group.

False belief versus comparison trials on video-based tasks

To assess whether children in the sample had impairments to belief reasoning that were disproportionate to any impairment of executive function (e.g., working memory and inhibitory control), we conducted further analyses taking account of performance on comparison trials. Because the comparison trial data and false belief trial data from the video-based tasks came from independent trials we were able to pursue the analytic strategy of assessing statistically whether impairment on false belief trials was disproportionate to impairment on comparison trials.

Table 2 shows mean scores by group on the video-based reality known and reality unknown belief reasoning tasks. The data were analysed using a mixed ANOVA with Group (FXS, FXS-A, Intellectual Disability) as the between- participants factor and trial type (false belief and working memory comparison) as the within-participants factor.

Table 2 Mean scores by group on video-based reality unknown and reality known false belief tasks (standard deviations are shown in brackets)

	Reality known		Reality unknown	
Group	False belief	Working memory	False belief	Working memory
FXS ($n = 15$)	0.40 (0.74)	1.87 (0.35)	0.73 (0.70)	1.20 (0.86)
FXS-A ($n = 15$)	0.27 (0.59)	1.53 (0.64)	0.33 (0.62)	0.93 (0.88)
ID ($n = 15$)	1.13 (0.92)	1.87 (0.35)	1.13 (0.92)	1.73 (0.46)

Note. ID: Intellectual disability group, FXS: fragile X group, FXS-A: fragile X group with many autistic features.

For the *reality known false belief task*, there was a significant main effect of group ($F_{(2,42)} = 5.32$, $p = .009$, $\eta_p^2 = .20$) and trial type ($F_{(1,42)} = 106.94$, $p < .001$, $\eta_p^2 = .72$), and a significant interaction ($F_{(2,42)} = 3.84$, $p = .03$, $\eta_p^2 = .15$). Simple effects analyses showed this interaction to be due to differences between groups on false belief trials, with both FXS and FXS-A performing worse than the Intellectual Disability group. Thus, the FXS and FXS-A groups performed worse than the Intellectual Disability control group on the false belief trials.

To check whether participants learned from the feedback provided at the end of false belief trials we compared performance on the first false belief trial with performance on the second false belief trial. Performance did not change significantly from trial 1 to trial 2 for any group (all $ps > .5$ by sign test).

We also checked performance on filler trials to assess whether children showed a strong tendency for adopting incorrect response strategies. On "woman present" filler trials there were 4/30 errors in the FXS group, 5/30 errors in the FXS-A group, and 4/30 errors in the ID group. On "woman absent" filler trials there were 5/30 errors in the FXS group, 4/30 errors in the FXS-A group, and 0/30 errors in the ID group. This indicated that participants were not generally succeeding on test trials by adopting strategies linked superficially to the presence or absence of the woman at the point when the object was manipulated.

For the *reality unknown false belief task*, there was a significant main effect of group ($F_{(2,42)} = 7.89$, $p = .01$, $\eta_p^2 = .27$) and trial type ($F_{(1,42)} = 12.98$, $p = .01$, $\eta_p^2 = .24$). There was no significant interaction ($F_{(2,42)} = 0.83$, $p = .92$, $\eta_p^2 = .004$). Pair-wise comparisons showed that the FXS group differed from the Intellectual Disability group ($p = .026$) but not from the FXS-A group ($p = .11$). The FXS-A group also differed from the Intellectual Disability group ($p = .001$). Thus, the FXS and FXS-A groups performed worse than the Intellectual Disability control group on both false belief trials and working memory comparison trials.

Table 3 Number of participants by group passing or failing standard false belief memory and test questions

Performance	FXS ($n = 15$)		FXS-A ($n = 15$)		ID ($n = 15$)	
	Deceptive box	Location change	Deceptive box	Location change	Deceptive box	Location change
Passed FB, passed memory	6	3	3	3	9	9
Failed FB, passed memory	8	11	9	10	6	6
Failed FB, failed memory	1	1	3	2	0	0

Note. ID: Intellectual disability group, FXS: fragile X group, FXS-A: fragile X group with many autistic features.

To check whether participants learned from the feedback provided at the end of false belief trials we compared performance on the first false belief trial with performance on the second false belief trial. Performance did not change significantly from trial 1 to trial 2 for any group (all *ps* $> .5$ by sign test).

We also checked performance on filler trials to assess whether children showed a strong tendency for adopting incorrect response strategies. On true belief filler trials there were 4/30 errors in the FXS group, 3/30 errors in the FXS-A group, and 5/30 errors in the ID group. This pattern confirmed that participants understood the meaning of the woman's clue and were not achieving correct responses on false belief trials by pursuing a general strategy of pointing to the opposite box from that indicated by the woman. On "inhibition" filler trials there were 4/30 errors in the FXS group, 5/30 errors in the FXS-A group, and 3/30 errors in the ID group. This indicated that participants were not generally failing on false belief trials merely because they always pointed to the box indicated by the woman.

Standard false belief tasks

Since the control questions of the Standard false belief tasks were asked within the same trial as the false belief question itself, failure on the control questions may have directly affected responses on the false belief question. Consequently, we did not include data from children who answered control questions incorrectly.

Performance on the two individual standard false belief tasks was analysed and the number of participants by group passing these tasks is shown in Table 3. A small number of children made errors on the memory check questions of the location change and deceptive box false belief tasks. Data from these children were excluded for the following analyses.

On the location change task, the FXS and FXS-A groups both performed poorly compared to the Intellectual Disability group ($\chi^2 = 4.441$, df $= 1$, $p = .035$; $\chi^2 = 3.877$, df $= 1$, $p = .049$ respectively). Performance of the FXS, and FXS-A groups did not differ (Fisher's exact $= 0.11$, df $= 1$, $p = .638$). On the deceptive box task, there were no significant differences between groups passing/failing the false belief test question ($\chi^2 = 3.325$, df $= 2$, $p = .19$). Thus, after excluding participants who failed the memory control questions, both the FXS and FXS-A groups performed worse than the Intellectual Disability control group on the location change task, but there were no statistically significant differences between groups on the deceptive box task.

Discussion

Our first question was whether children with FXS would be disproportionately impaired on theory of mind tasks (such as false belief tasks). Our findings suggest that children with FXS do have a clear impairment on false belief tasks that is disproportionate to their general level of intellectual disability. The absence of learning from feedback on our tasks indicated that this difficulty is robust, and not the result of a simple misunderstanding of the point of the tasks. Importantly, this was the case whether or not the children with FXS had significant autistic features, suggesting that theory of mind difficulties in FXS are not merely an artifact of the high co-morbidity of FXS and autism. Indeed, while there was a numerical trend for the FXS group to make fewer errors on false belief tasks than the FXS-A group, this was non-significant for the sample size reported here. It is possible that differences between the FXS and the FXS-A groups would emerge using a larger sample size. This remains an important area of investigation for future studies.

The findings of the present study are broadly in line with those reported by Garner et al. (1999), whose findings suggested that boys with FXS performed poorly on tests of first order false belief compared with mental age matched controls. Fewer boys with FXS passed the deceptive box task and the location change task than the control group, although this difference only reached statistical significance on the deceptive box task. The FXS group reported in Cornish et al. (2005) performed better on tests of first order false belief than the FXS groups reported in our study and the FXS group reported in Garner et al. (1999). Just under half the FXS group passed the location change task in Cornish et al. (2005), which was similar to the pass rate for the comparison group of children with Down syndrome included in the study. It is unclear why the FXS group in the Cornish et al. (2005) performed comparably to the control group whilst in the present study and the Garner study the FXS groups were out-performed by controls. One possibility is that the control group in the Cornish et al. (2005) had an unusually poor performance. A meta-analysis of some 40 studies by Yirmiya, Erel, Shaked, and Solomonica-Levi (1998) has highlighted that people with Down syndrome show a unique strength on tests of theory of mind. It is therefore puzzling why less than half of the group with Down syndrome passed the first order false belief tests despite a relatively high verbal mental age of 6 years 7 months.

Our second question was whether any theory of mind impairment was primary, or a consequence of impaired inhibitory control or working memory. We noted that standard false belief tasks confound the need to reason about beliefs (e.g., Simon thinks the chocolate is in the cupboard) with the need to resist interference from one's own knowledge of the correct answer (that in fact the chocolate is in the fridge). Since difficulty with executive processes such as resisting interference is known to be a distinctive feature of the FXS cognitive phenotype (Cornish, Sudhalter, & Turk, 2004), children with FXS might have made errors on such false belief tasks merely because they failed to meet this executive demand. This hypothesis was tested by comparing performance on traditional "reality-known" false belief tasks with performance on the relatively novel "reality-unknown" false belief tasks, where this executive confound is removed. The results showed that, compared with the Intellectual Disability control group, children with FXS made more errors on all types of false belief task (although on the standard tasks this only reaches statistical significance when the tasks are combined), and that neither the Intellectual Disability nor FXS children found the reality-unknown false belief task any easier than reality-known false belief tasks. This result suggests that difficulty resisting interference from knowledge of reality could not explain difficulty on the false belief tasks for any group. It is also noteworthy that the standard false belief tasks required children to comprehend verbal narratives acted out with puppets, whereas the video-based false belief tasks made relatively few demands on verbal comprehension. The absence of any difference in performance across these types of task suggests that the verbal comprehension demands of standard false belief tasks may not be a significant source of difficulty for children with FXS.

It was also noted that false belief tasks make substantial demands on working memory. Since impaired working memory is known to be a feature of the FXS cognitive phenotype, it is important to test whether impaired working memory alone could explain any errors made on false belief tasks. The commonly used location change and deceptive box false belief tasks only allow relatively weak checks to be made, by testing whether children remember crucial facts about the story. When data from children who failed these memory checks were discounted, children with FXS still performed less well than the ID control group on the location change false belief task. However, for the deceptive box task, once data from children who failed the check question were discounted, differences between the groups' performance were not significant.

The video-based reality-known and reality-unknown false belief task allowed a stronger test of the role of working memory because they incorporated *independent* comparison trials. For the reality-known task, the comparison trial was analogous to the "reality" control question of the location change false belief task, and merely tested whether the participant had followed the visible displacement of the object from one container to the other. Compared with Intellectual Disability controls, children with FXS were still disproportionately impaired on false belief trials, even when these memory comparison trials were taken into account. Memory comparison trials for the reality-unknown task were more demanding, and more closely matched to the incidental memory demands of the reality-unknown false belief trials. For both false belief and comparison trials, participants did not see the critical hidden object, but had to realize that it had changed locations when boxes were swapped. Analyses showed that children with FXS had significantly more difficulty than Intellectual Disability controls on *both* the false belief and the memory comparison trials. That is to say, the difficulty that children with FXS experienced on false belief trials was proportionate to this group's impairment on working memory trials. This pattern of results can be interpreted in two ways. One possibility is that children with FXS have independent impairments of theory of mind and working memory. The second possibility is that children with FXS made errors on false belief trials because of the substantial demands that belief reasoning makes on their impaired working memory processes, not because of independent impairment to a neuro-cognitive system devoted to belief reasoning. Evidence to discriminate between these alternatives might come from assessing individual differences in working memory and theory of mind in a much larger sample of children with FXS. However, in the absence of such evidence, we favour the more parsimonious account that attributes theory of mind impairment in children with FXS to a primary deficit in working memory.

The present study employed participants recruited from the Fragile X Society and schools, rather than from a clinical setting. Recruiting participants from these sources rather than recruiting from a clinic was undertaken to ensure the study employed a representative sample. In this respect, employing a non-clinical sample was a strength of the study. However, employing a non-clinical sample also meant that the FXS children were divided into FXS and FXS-A groups using the SCQ (Berument et al., 1999), a parent report measure of symptoms associated with autism rather than being divided into groups using a clinical diagnosis of autism. The SCQ is an autism screening measure derived from the

Autism Diagnostic Interview-Revised (ADI-R: Lord et al., 1994), which is one of the most widely used instruments in the diagnosis of autism. A recent study by Howlin and Karpf (2004) used the SCQ to identify autism spectrum disorders in Cohen Syndrome. The findings suggested that while the SCQ cut-off score of 15 and above outlined by Berument et al. (1999) was successful at identifying cases of autism spectrum disorder, there were some false negatives (individuals who did not meet the criteria on the SCQ but met criteria for autism spectrum disorders on the ADI-R). The implication of these findings for the present study is that the individuals in the FXS-A group are likely to meet the diagnostic criteria for autism on the ADI-R. However, it is possible that some of the individuals in the FXS group would also meet the diagnostic criteria for autism on the ADI-R. A further issue is that in forming the FXS groups, we excluded boys who had scores on the SCQ between 15 and 24. This group was excluded in an attempt to reduce the likelihood of false positives for autism in the FXS-A group. Consequently, we cannot conclude from our data how this group would perform on theory of mind measures. These methodological limitations should be addressed in future research.

Whilst acknowledging that there are only a handful of studies of theory of mind in boys with FXS, there are some issues identified that may be of value to consider in a clinical setting and to explore in future research. There is evidence that theory of mind is impaired in boys with FXS and it is possible that this deficit plays a role in the social difficulties associated with FXS. In particular, males with FXS experience difficulties with social interaction and communication, development of which is positively associated with theory of mind ability in typically developing children (e.g., Astington, 2000). Interventions aimed at improving theory of mind understanding in young children with FXS may therefore help to improve the developmental outcomes of these children. Work undertaken to improve theory of mind in children with autism has found that thought-bubbles are a successful strategy for developing mental-state understanding (Wellman, Baron-Cohen, Caswell, Gomez, Swettenham, Toye et al., 2002). The authors cautioned that whilst this technique did enable the majority of the children with autism to pass tests of false-belief and to generalize their understanding to other tests, some children learnt very little. Moreover, the generalizability of this training to real-life social situations has been disappointing (Hadwin, Baron-Cohen, Howlin, & Hill, 1997).

Overall, the findings of the research indicate that boys with FXS have difficulty with theory of mind tasks compared to an intellectual disability control group. However, boys with FXS also perform worse on comparison trials. This means that theory of mind difficulties are likely to be an important aspect of the FXS clinical profile. However, it is probable that the deficits in theory of mind understanding stem from general information processing deficits (e.g., working memory), not from a primary deficit in theory of mind, and that this is the case whether or not children with FXS have significant autistic features.

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Appendix A

Standard location change false belief task. Based upon the Sally-Anne task (Baron-Cohen et al., 1985), the participant is introduced to two play-mobile figures and is invited to give them names (for example James and Carol). The experimenter then narrates the following story: “This is James and this is Carol. James has just come into the house from playing football outside. He takes his ball and puts in into his toy box and goes to play upstairs”. The participant sees the play-mobile figure put his ball into a box and exit the area. The narrative is then continued. “While James is playing upstairs, Carol decides to move the ball. Carol takes the ball out of the box and puts it in the cupboard”. The participant sees the second play-mobile figure remove the ball from the box and put it into the cupboard, where it is out of sight. This second figure then exits the area. The participant is then asked two control questions about the original and the current location of the ball: Memory Question: “Where did James put his ball in the beginning?” and the Reality Question “Where is the ball now?” The first play-mobile character is then brought back into the area, and the participant is told “James is going to play outside again now and he wants his ball”. Then the participant is asked the Test Question, “Where will James look for his ball?” The participant scores one for a correct response or zero for an incorrect response on the test question.

Standard Deceptive box false belief task. In this variant of the deceptive box task (Perner, Frith, Leslie, & Leekam, 1989), the participant is shown a sealed egg carton and asked, “What do you think is inside here?” The participant answers “eggs” or “those” (pointing to the picture of the eggs on the box). The box is then opened up and the participant is shown that the box actually contains a pen. The participant is asked to name the pen to check that they know what it is. The pen is put back in the box and the box is closed. The participant is then asked, “In a minute X is going to come and work with me. (S)he hasn’t seen this box yet, or what’s inside it. When (s)he comes in, I am going to show her/him this box, closed just like this (participant is shown box again). I am going to ask her/him “X, What’s in this box?” The participant is then asked the Test Question “What will (s)he say?” and a Reality Question, “What is really inside the box?” The participant scores one for a correct response or zero for an incorrect response on the test question.

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