Anxiety and Spatial Navigation in Williams Syndrome and Down Syndrome

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ABSTRACT

Individuals with Down Syndrome (DS) and individuals with Williams syndrome (WS) present with poor navigation and elevated anxiety. The aim of this study was to determine the relationship between these two characteristics. Parent report questionnaires measured navigation abilities and anxiety in WS (N = 55) and DS (N = 42) as follows. Anxiety: Spence Children’s Anxiety Scale and a novel measure of navigation anxiety. Navigation: Santa Barbara Sense of Direction Scale (SBSOD) and a novel measure of navigation competence. Most individuals were not permitted to travel independently. A relationship between navigation anxiety and SBSOD scores (but not navigation competence) was observed for both groups.

Introduction

The ability to navigate in space is a crucial facet of human and animal development, allowing an organism to recognize its location, explore, and learn routes. In humans, the ability to learn a route relies on knowledge of landmarks (landmark knowledge), the sequences of turns (route knowledge) and integration of this information with perceptual-motor skills to form a cognitive map of an environment (configural knowledge) (Jansen-Osmann & Fuchs, 2006; Montello, 1998; Siegel & White, 1975). For individuals with Williams syndrome (WS) and Down syndrome (DS) there are marked deficits in the ability to learn routes and navigate in both real-world and virtual environments (e.g., Courbois et al., 2013; Farran, Blades, Boucher & Tranter, 2010; Farran et al., 2015; Lakusta, Dessalegn, & Landau, 2010).

DS is a genetic syndrome that results from partial or complete trisomy of chromosome 21 (Roubertoux & Kerdelhué, 2006). In Europe, DS has a prevalence of 10.1 per 10,000 live births (de Graaf, Buckley, & Skotko, 2021), although this varies by country dependent on the incidence of elective terminations. At a cognitive level, DS presents with mild to moderate intellectual disability, characterized by an uneven profile of relative deficits in verbal ability compared to visuospatial skills (Jarrold & Baddeley, 1997; Visu-Petra, Benga, Tincas, & Miclea, 2007). WS is a rare genetic syndrome with an estimated prevalence of live births of between one in 7,500 (Stromme, Biomstad & Ramstad, 2002) and 1 in 20,000 (Morris, Dempsey, Leonard, Dilts & Blackburn, 1988). WS results from a deletion of approximately 28 contiguous genes due to a hemizygous microdeletion of 1.6Mb on chromosome 7q11.23 (Tassabehji, 2003). WS is also characterized by mild to moderate learning difficulties, which also comprises an unusual cognitive profile. However, contrary to DS, the WS cognitive profile is typified by a disparity between relatively strong linguistic ability and poor visuospatial ability (Farran & Karmiloff-Smith, 2012).

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Despite different cognitive profiles for individuals with WS and DS, they share marked deficits in spatial navigation. Both groups rarely develop configural knowledge (Broadbent, Farran, & Tolmie, 2014; Courbois et al., 2013; Edgin et al., 2010; Farran et al., 2010, 2015; although see Banta Lavenex & Lavenex, 2021 for evidence of low-level configural knowledge). Both groups can, however, develop route knowledge (Davis, Merrill, Conners, & Roskos, 2014; Farran et al., 2015; Purser et al., 2015), with route knowledge typically poorer for individuals with DS, than for individuals with WS (Purser et al., 2015). This difference might relate to a strong reliance on non-verbal ability to learn routes in DS (Purser et al., 2015) and difficulties in recognizing and recalling landmarks (Courbois et al., 2013; Davis, Merrill, Conners & Roskos, 2014; Toffalini et al., 2018) in this group. Individuals with WS have a strong reliance on landmarks when learning a route (Broadbent et al., 2014), can use landmarks to orient themselves (Lakusta et al., 2010) and benefit from verbalizing the landmarks and turns along a route (Farran et al., 2010). However, they do not always choose appropriate landmarks (Farran, Courbois, Van Herwegen, & Blades, 2012; Farran, Formby, Daniyal, Holmes, & Van Herwegen, 2016; Nardini, Atkinson, Braddock, & Burgess, 2008). The above navigational difficulties in WS and DS have strong implications for independence. Without an understanding of the configuration of the environment, wayfinding is inflexible to change. A reliance on a fixed route makes it difficult to re-orient when lost, to take short cuts, or to find alternative routes if the usual route is not accessible.

A potentially important behavioral aspect to consider, is the impact of anxiety on navigation. For example, Mueller et al. (2009) compared navigation performance in children with an anxiety disorder relative to controls. They report poorer navigation performance in the disorder group, thus demonstrating a relationship between general anxiety and navigation. Evidence from the general population also suggests a negative association between anxiety and navigation performance in both childhood (Schmitz, 1997) and adulthood (Lawton & Kallai, 2002; Munoz-Montoya, Fidalgo, Juan, & Mendez-Lopez, 2019; Pazzaglia, Meneghetti, & Ronconi, 2018; Thoresen et al., 2016; Walkowiak, Foulsham, Eardley, 2015).

While general anxiety and situation-specific navigation anxiety are related to one another in the general population (Lawton & Kallai, 2002), there are some specificities with reference to the relationship between these two types of anxiety and navigation competence. Situation-specific navigation anxiety is consistently related to navigation competence (Lawton & Kallai, 2002; Pazzaglia et al., 2018; Walkowiak et al., 2015). However, evidence for a relationship between general anxiety and navigation competence is mixed with some finding no relationship (Walkowiak et al., 2015), some finding mixed evidence (Lawton & Kallai, 2002) and some finding support for a relationship (Schmitz, 1997; Thoresen et al., 2016). Viaud-Delmon, Berthoz, and Jouvent (2002) demonstrate no differences between high-anxious and low-anxious individuals when performing an assessment of route knowledge, but impaired performance in high-anxious individuals when asked to represent the configural structure of the environment. This might explain the mixed findings, and further suggests that general anxiety contributes to known individual differences in representations of spatial layouts (Weisberg et al., 2016).

Global rates of anxiety in the general population are estimated at 7% to 11% (Baxter, Scott, Vos, & Whiteford, 2013). In both WS and DS, this prevalence is elevated. A recent meta-analysis of anxiety in WS determined that 48% of individuals with WS meet criteria for at least one anxiety disorder (Royston, Howlin, Waite, & Oliver, 2017). In DS, anxiety disorders are less prevalent than in WS (Vicari et al., 2016), with approximately 14% of individuals with DS showing evidence of anxiety disorders (Myers & Pueschel, 1991; Vicari et al., 2016).

Mueller et al. (2009) also suggest a neural association between navigation and anxiety. The hippocampus plays a crucial role in navigation (Maguire et al., 1998). Specifically, it promotes learning of the spatial configuration of landmarks within an environment to develop a cognitive map and is involved in landmark recognition (Eichenbaum, 2017; O’Keefe & Nadel, 1978; Sharma et al., 2017). In parallel, evidence suggests a role of the hippocampus in anxiety. Specifically, the rodent literature reports a role of the Septo-Hippocampal System (SHS) in anxiety (see Mueller et al., 2009), and in research with humans, hippocampal lesions have shown an association between the hippocampus and
avoid-approach behavior (Bach, Hoffmann, Finke, Hurlemann, & Ploner, 2019), while a volume reduction in posterior hippocampus is associated with risk for anxiety in monozygotic twins discordant for high risk for anxiety (De Geus et al., 2007).

Individuals with WS and individuals with DS present with atypicalities in the structure and function of the hippocampus (Meyer-Lindenberg et al., 2004; Pinter, Eliez, Schmitt, Capone, & Reiss, 2001). Given the behavioral navigational limitations discussed above for individuals with WS and individuals with DS, knowledge that anxiety disorders feature in both WS and DS, the potentially shared neural associations with navigation and anxiety, and evidence that anxiety impacts navigation in the typical population, it is likely that anxiety is a contributing factor to navigational experiences in WS and in DS. This evidence forms the rationale for the current study. To our knowledge, there is just one study that has investigated anxiety during navigation in DS and no studies with WS. Yang, Faught, and Merrill (2017) focused on the influence of parental teaching of navigation skills and parental concerns on the navigational competence and navigational confidence of their son/daughter. They report that parental teaching positively predicts navigation competence in their son/daughter, and that lower parental concerns were associated with both higher navigation skills and confidence in those with DS. The aim of the current study is to further determine the relationship between both general and situation-specific anxiety, with navigation in DS and WS.

Understanding the nature of the role of both general and situation-specific anxiety in navigation has important implications for individuals with WS and DS. With only 7–12% of individuals with WS being in independent work (Stinton & Howlin, 2012), it is possible that this is impacted by poor navigation. This is supported by studies with individuals with Intellectual Disability which have demonstrated that limitations in independent travel present a barrier to community participation, social inclusion, and access to health care and leisure activities (McCausland et al., 2020; Mooney, Raﬁque, & Tilly, 2019; Wilson, Jaques, Johnson, & Broherton, 2017). It is important to better understand the complexity of limitations in navigation in WS and DS, specifically an understanding of whether anxiety inﬂuences navigation, and how it can be used to inform remediation strategies to improve navigation.

We measured general anxiety using the Spence Children’s Anxiety Scale (SCAS; Spence, 1997a) and situation-specific navigation anxiety using our novel measure of navigation anxiety. Navigation was measured using the Santa Barbara Sense of Direction Scale (SBSOD; Hegarty, Richardson, Montello, Lovelace, & Subbiah, 2002) and our novel measure of navigation competence. Sense of direction refers to the ability to orient within space (e.g., directions, configural knowledge) and has been associated with personality traits such as conscientiousness, intellect, emotional stability, and extraversion (Condon et al., 2015). We included a measure of sense of direction because it has been suggested that low sense of direction relates to uncertainty and low navigation-specific conﬁdence (Cornell, Sorenson, & Mio, 2003) and navigation anxiety (Lawton & Kallai, 2002). It has also been suggested that sense of direction can be improved through navigational experience (Condon et al., 2015). If navigational experience is limited due to anxiety, it follows that sense of direction might be poor.

This study has three aims. First, to determine the absolute levels of anxiety and navigation competence for individuals with WS and DS and how they compare between groups. We predict, consistent with the literature, that general anxiety will be high and that sense of direction and navigation competence will be poor in both groups, with a higher frequency of elevated general levels of anxiety in WS than in DS (e.g., Farran et al., 2015; Royston et al., 2017; Vicari et al., 2016). Navigation-specific anxiety has hitherto not been measured in these groups and so no predictions are made; however, given the association between general anxiety and situation-specific anxiety in the general population, it is likely that navigation-specific anxiety will be high, akin to our predictions for general anxiety levels.

Second, we aim to determine the relationship between anxiety (situation-specific and general) with sense of direction and navigation competence in these groups. In doing so, we will also determine whether any relationships between anxiety and navigation are driven by particular subdomain(s) of
general anxiety (owing to the lack of research at this granular level, we do not have specific predictions relating to anxiety subdomains). Based on the studies above, we predict significant associations between anxiety and navigation (e.g., Lawton & Kallai, 2002). For all associations, because this is cross-sectional data, the causal directions are unknown, and we do not rule out bi-directional relationships. Based on Viaud-Delmon et al. (2002), who showed that general anxiety is associated with configural knowledge, but not route knowledge, the association between general anxiety and navigation competence might be weaker than between situation-specific anxiety and navigation competence for our groups. This is because our navigation competence measure encompasses both route knowledge and configural knowledge. Due to sense of direction being related to personality trait and its association with navigation confidence (Cornell et al., 2003), we specifically predict the strongest association between navigation-specific anxiety and sense of direction.

Finally, using qualitative analysis, we aim to determine whether anxiety is a theme within parental discussion of their son’s/daughter’s independent travel (or lack of) and perceived training needs. Based on Yang et al. (2017), we predict that there will be a strong focus on anxiety in relation to independent travel and training needs.

**Method**

**Participants**

Parents/caregivers of individuals with WS (N = 55) were recruited through the Williams Syndrome Foundation UK. Parents/caregivers of individuals with DS (N = 42) were recruited through charities and support groups in the South East of England. All individuals with WS and individuals with DS had a genotypic and phenotypic diagnosis confirmed by a clinician. In practice, all responders were parents (there were no non-parent caregivers). Ethical approval was obtained from the University ethics committee. Written consent was provided by parents. For both groups, the minimum age of the individual with WS or DS was 12 years. This was specified because some form of navigation experience was deemed possible at this age. For the WS group, there was no upper age limit. However, for the DS group an upper age limit of 35 years for the individual with DS was specified due to the risk early onset dementia from 40 years (Head, Powell, Gold, & Schmitt, 2012). Details of the individuals with WS and individuals with DS are shown in Table 1.

**Design and procedure**

This study had a questionnaire design. The questionnaire comprised of sections which included demographic questions and questions relating to travel training experience, and three main sections to assess: navigation ability (Santa Barbara Sense of Direction Scale; SBSOD; Hegarty et al., 2002), general anxiety (Spence Children’s Anxiety Scale; SCAS; Spence, 1997a) and navigation-specific anxiety and navigation competence (novel measures, designed for this study). The questionnaire was designed for caregivers of individuals with WS or individuals with DS to complete. The questionnaires were provided as physical copies at WS or DS events, were sent to respondents by e-mail or post, or were completed online via Qualtrics.

| Group       | Chronological Age (year; month): Mean (range) | Gender (M:F) | Has received travel training | Travels independently |
|-------------|-----------------------------------------------|--------------|------------------------------|-----------------------|
| WS (N=55)   | 28.8 (12.3 to 51.4)                           | 27:28        | 32/55 (58%)                 | 22/55 (40%)           |
| DS (N=42)   | 21.0 (12.1 to 33.10)                          | 21:21        | 23/42 (55%)                 | 10/39 (26%)           |
**Santa Barbara Sense of Direction Scale (SBSOD; Hegarty et al., 2002)**

The SBSOD is a standardized measure of sense of direction which has been previously used successfully with individuals with DS (Yang et al., 2017). The SBSOD is a 15-item scale that assesses interaction with the environment such as giving directions, navigation, and understanding of space. Responses are given on a seven-point Likert scale (1 exactly like me – 7 not at all like me). In adults, the scale has been shown to be associated with measures of reorientation, formulation of a cognitive map and directional pointing in virtual and real-world environments (Hegarty et al., 2002). Test–retest reliability is .91 and internal consistency is high (coefficient α = .88). The wording of all items was changed from first person to third person reference, and some items were adjusted to UK wording or make it more relevant for caregivers of individuals with WS or DS (see Table 2).

**Spence Children’s Anxiety Scale (SCAS; Spence, 1997a)**

The SCAS is a standardized measure of anxiety, which is suitable for use with individuals with neurodevelopmental disorders in childhood through to adulthood (e.g., White, Oswald, Ollendick, & Scahill, 2009), and has been successfully used with individuals with WS (Rodgers et al., 2012) and individuals with DS (Zadeh et al., 2019). Additionally, the SCAS is recommended by the National Institute for Health and Care Excellence (NICE) as a measure to identify children and young adults with anxiety disorders (NICE, 2013).

In the SCAS participant rate, the frequency of experiencing 39 anxiety-related feelings such as “My daughter or son can’t seem to get bad or silly thoughts out of his/her head.” The SCAS provides an overall anxiety score as well as subscale measures of generalized anxiety, panic/agoraphobia, social phobia, separation anxiety, obsessive compulsive disorder, and physical injury fears based on DSM-IV diagnostic criteria. Participants are scored for each item based on the frequency of occurrence (never [0], sometimes [1], often [3], always [4]) and so lower scores indicate lower levels of anxiety; the scores can then be categorized as “normal” or “elevated” for the participants’ age/sex. The scale has excellent reliability (Cronbach’s α = .93) and a good six-month test–retest reliability of .60 (Spence, 1997b).

| Table 2. List of original items and their revised version in the SBSOD. |
|-----------------|-----------------|
| **SBSOD original item** | **revised item** |
| 2. I have a poor memory for where I left things. | 2. My son/daughter is not very good at remembering where they left things. |
| 5. I tend to think of my environment in terms of cardinal directions (N, E, S, W). | 5. My son/daughter thinks of the world around them in terms of the directions North, South, East and West. |
| 6. I very easily get lost in a new city. | 6. My son/daughter very easily gets lost in a new place. |
| 10. I don’t remember routes very well while riding as a passenger in a car. | 10. My son/daughter doesn’t remember the route very well if they are a passenger in a car. |
| 13. I usually get someone else to do the navigational planning for long trips. | 13. My son/daughter usually lets someone else plan how to get to places when they go on long trips. |
| 14. I usually remember a new route after I have traveled it only once. | 14. My son/daughter can usually remember how to get somewhere even if they have only been there once before. |

| Table 3. Correlations for SCAS subscales with SBSOD, navigation, and navigation anxiety for navigation in Williams syndrome. |
|----------------|-----------------|----------------|----------------|----------------|----------------|----------------|----------------|----------------|----------------|
| **2** | **3** | **4** | **5** | **6** | **7** | **8** | **9** | **10** | **11** |
| 1. SBSOD | .64** | -.26 | -.14 | -.32* | -.27* | -.15 | -.12 | -.12 | -.39* | .38** |
| 2. Navigation | / | -.28* | -.16 | -.33* | -.31* | -.14 | -.015 | -.26 | -.36* | .33* |
| 3. SCAS | / | -.74** | .81** | .78** | .80** | .49** | .88* | .74** | -.18 |
| 4. OCD | / | -.56** | .67** | .46** | .034 | .62** | .37* | -.30* |
| 5. Social phobia | / | -.49** | .48** | .32* | .70** | .73** | -.08 |
| 6. Panic or Agoraphobia | / | -.53** | .17 | .67** | .50** | -.23 |
| 7. Separation anxiety | / | -.48** | .67** | .59** | -.21 |
| 8. Physical injury fear | / | -.28* | .45** | .08 |
| 9. Generalized anxiety | / | .65** | -.12 |
| 10. Navigation anxiety | / | -.20 |
| 11. Age of individual | / | 1.0 |

Note. N = 91 *p < .05, **p < .001. Correlations for SBSOD (n = 90).
The parent version of the task was used, which is identical to the child version but does not contain seven filler items; this shorter version was used to reduce the overall length of the questionnaire to ensure that caregivers did not become fatigued when completing the questionnaire. The parent version of the scale has been shown to have good reliability and validity (Nauta et al., 2004) so we can be confident that parental reports accurately reflect levels of anxiety in individuals with WS or DS. Minor alterations were made to the original form of the questionnaire to make the instrument more suitable for older participants who may be living away from parents and not attending school, as follows: item 15 “my daughter or son has trouble going to school in the mornings because (s)he feels nervous or afraid” was changed to “my daughter or son has trouble getting to school/work/ out of the house in the mornings because he/she feels nervous or afraid."

**Feelings about wayfinding**

This novel section of the questionnaire, broadly labeled as “feelings about wayfinding” comprised of 31 questions of mixed format that specifically related to behaviors and emotions when navigating. These comprised open-ended questions, likert scales, and categorical variables.

**Navigation behaviors.** Questions 1 to 5 were categorical questions where the caregiver ticked boxes to indicate which behaviors were most associated with the individual, with reference to what they find difficult and the strategies employed for navigation experiences. We chose not to analyze one question regarding the modes of transport used when traveling because it was not central to our current research questions. The remaining four questions in this section were: 1) “My daughter/son finds it hard to understand.” (with five tick box items); 2) “If my son/daughter gets lost they:” (with five tick box items); 3) “When my daughter/son needs to find their way they:” (with five tick box items); 4) “My son/daughter can plan on their own how to get to where they want to go:” (with three tick box items).

Question 31 was a categorical (yes/no) question asking parents whether their son or daughter travels independently. This was followed by further questions relating to a “Yes” response, to determine when, how and where individuals traveled to (not reported due to low participant numbers), and further questions relating to a “No” response to establish why independent travel was not permitted.

**Navigation competence composite score.** Questions 6 to 19 were 7-item Likert scale questions (“Yes this is exactly like him/her” to “No, this is not at all like him/her”) which measured individuals’ wayfinding tendencies and were used to form the Navigation competence variable. Wayfinding is a spatial cognitive ability which uses goal-directed decision-making to reach a destination (Montello, 2005). Landmark knowledge and route knowledge and configural knowledge draw on wayfinding skills (Blades, 1991; Montello, 2005), and here we collectively refer to these skills as navigation competence. The Navigation competence composite score had a low Cronbach’s alpha reliability of \( \alpha = .48 \) when all questions were included. Inter-item correlation demonstrated that questions 9, 12, 13, 14, 15, 16, and 18 were not measuring the same construct as the other questions (correlations were close to zero or negative) and so were excluded. This is likely because some questions touched on emotions related to navigation, rather than navigation competence per se (e.g., Q12 “My son/daughter does not like leaving the house and avoids travelling”). The remaining questions (questions 6, 7, 8, 10, 11, 17, 19) had a Cronbach’s alpha reliability of \( \alpha = .78 \), which is considered a good level of reliability. Thus, only these items were retained in the measure; these items asked questions related to Executive Functions (distraction, memory, focusing on the task, planning), independence, understanding directions and configural knowledge.

**Navigation anxiety composite score.** Questions 20 to 28 were 5-item Likert scale questions (My son/daughter feels “Very Happy” to “Very worried” when . . .) which measured the feeling aspect of individuals within navigation situations and were used to form the Navigation Anxiety variable. In
contrast to the SCAS, this anxiety variable was situational, i.e., specific to anxiety associated with navigating. The navigation anxiety composite score had an above acceptable level of internal consistency with a value of $\alpha = .89$.

**Training needs and training received.** Questions 29 and 30 were qualitative questions which gave respondents the opportunity to describe any travel training needs and detail travel training received that they felt had been of benefit. These were: “Has your son/daughter received any help with finding their way that you think has really benefitted them?” and “What sort of help or training do you think your son/daughter would benefit from to help them find their way?”

**Data analysis**

Some participants did not provide responses for all questions. For variables with missing data, analyses were conducted with the relevant participant response(s) excluded. Specifically, of the three continuous variables, there was missing data for 1 individual with WS and 5 individuals with DS for navigation anxiety, 3 individuals with DS for navigation and 1 individual with DS for SBSOD. Of the categorical questions, data was missing for 3 individuals with DS for the independent travel question.

All continuous variables were normally distributed (Kolmogorov-Smirnov, $p > .05$) except for the WS group for navigation anxiety ($p < .001$). As ANOVA is robust to violations of assumptions of normality, parametric tests were carried out for all analyses (Blanca, Alarcón Postigo, Arnau Gras, Bono Cabré, & Bendayan, 2017). Furthermore, the sample size was large enough for the central limit theorem to apply (Field, 2013).

For each of the four continuous variables, group comparisons were conducted via t-tests, and cutoff scores for the SCAS were used to categorize the prevalence of “normal” and “elevated” anxiety in each group. The relationship between anxiety and navigation was analyzed using bivariate correlations and multiple regression. Categorical variables were presented descriptively, and analyzed using chi-squared (group comparisons) and Cochran’s Q tests with posthoc Dunn’s tests (profile analyses). Qualitative analysis of the two open-ended questions regarding navigation training was via thematic analysis (Braun & Clarke, 2006).

**Results**

**Absolute levels of navigation, anxiety, and navigation anxiety, per group**

Descriptive statistics for SCAS, SBSOD, navigation, and navigation anxiety are displayed in Figure 1. For the SBSOD and navigation competence variables, higher scores reflect stronger navigation. For SCAS and navigation anxiety, higher scores reflect higher levels of anxiety. Independent samples t-tests determined that the groups differed significantly for all four variables. The DS group had significantly stronger navigation (higher SBSOD: $t(94) = -2.20, p = .03$; and higher navigation scores: $t(90) = -2.70, p = .01$) and lower anxiety (lower SCAS: $t(95) = 2.83, p = .01$; and lower navigation anxiety scores: $t(89) = 2.55, p = .01$) than the WS group.

For the SCAS, raw scores can be categorized as “normal” or “elevated” and cutoffs were based on Muris, Schmidt, and Merkelbach (2000). As all individuals with WS or DS were above 12 years the 13–19-year-old cut off category was used (≥25 for males; ≥36 for females). This demonstrated that 53% of the WS group and 36% of the DS sample fell into the “elevated anxiety” category on the SCAS.

**Navigation behaviors**

We asked whether individuals were permitted by their caregiver to engage in independent travel, and whether they had received travel training. Responses are shown, as percentages in Table 1.
Two chi-squared analyses identified that the proportion of individuals permitted independent travel and who had received travel training was similar for both groups: independent travel: $\chi^2(2) = 2.10, p = .15$; travel training: $\chi^2(2) = 0.11, p = .74$. A further chi-squared analysis, collapsed across group, revealed an association between independent travel and travel training such that receiving travel training was associated with being permitted to travel independently: $\chi^2(2) = 17.93, p < .001$. For those who had received travel training, this was extremely variable in length (from a single on-off talk on road safety to 3 years/ongoing support) and in those who had delivered it (responses were: school/college, social services, parent, local council, day center, support/social group, social worker/carer, and charities).

For those who ticked “no” to the independent travel question, they were asked why, using five further tick boxes. This revealed overwhelmingly that the confidence of the parent (WS: 49%; DS: 60%) was the main reason. This was closely followed by confidence of the individual (WS: 33%; DS: 21%). Other categories were more practical in nature and received the following responses: they have no need to travel alone (WS: 29%; DS: 26%); where they need to go is too far to travel on foot (WS: 11%; DS: 12%); where they need to go is not accessible by public transport (WS: 7%; DS: 7%).

For each item of the four tick box questions in the novel section of the questionnaire, we were interested in group differences in the frequency of responses (i.e., evidence of syndrome-specific navigation difficulties or behaviors), and the profile of frequencies of responses across the items in each question (i.e., documentation of peaks and troughs in navigation behavior engagement for individuals with learning difficulties). Only two items showed syndrome-specific associations, both
of which related to asking for help. That is, individuals with WS were significantly more likely to ask someone to help them as a navigation strategy, $\chi^2 (2) = 4.79, p < .05$; and to ask a safe person for help them as a strategy when lost, than were individuals with DS, $\chi^2 (2) = 10.86, p < .001$. All other chi-squared analyses were non-significant ($p > .05$). Profiles were analyzed using Cochran’s Q tests, with posthoc Dunn’s tests. The two questions with syndrome-specific associations were analyzed for each group separately on account of group differences in the “help” items. For the remaining two questions, profiles were collapsed across group for analyses.

There was a significant main effect of item for “My daughter/son finds it hard to understand;,” $\chi^2 (4) = 150.75, p < .001$. As shown in Figure 2, this was due to high frequencies of difficulties with car speed (significantly higher than all other items; $p < .05$) and standard road crossing (significantly higher than all remaining items; $p < .05$). The rest of the pairwise comparisons were non-significant ($p < .05$ for all).

There was a significant main effect of item for both groups for “If my son/daughter gets lost they;” (WS: $\chi^2(4) = 44.89, p < .001$; DS: $\chi^2(4) = 19.05, p = .001$). As shown in Figure 3, for individuals with WS, the highest frequencies were for asking a safe person and texting/calling someone (significantly higher than stop and think/look around, and sending a photo of their location to someone; $p < .05$ for all), followed by walking around to find their way (significantly higher than sending a photo of their location to someone, $p < .05$). All other pairwise comparisons were non-significant ($p > .05$ for all). For individuals with DS, the highest frequency was for walking around (significantly more frequent than asking a safe person and using photos; $p < .05$), followed by texting/calling (higher than using photos; $p < .05$). All other pairwise comparisons were non-significant ($p > .05$ for all).

![Figure 2](image_url). Percentage of responses for navigation situations that individuals with WS or DS find hard to understand.
Figure 3. Percentage of responses for strategies/solutions individuals with WS or DS use when getting lost.

Figure 4. Percentage of responses for strategies used by individuals with WS or DS when needing to find their way.
There was a significant main effect of item for both groups for “When my daughter/son needs to find their way they” (WS: $\chi^2(4) = 117.87, p < .001$; DS: $\chi^2(4) = 54.91, p < .001$). As shown in Figure 4, both groups showed the same pattern, with significantly higher frequencies for getting someone’s help and remembering what they have seen before compared to all other items ($p < .05$).

There was a significant main effect of item for “My son/daughter can plan on their own how to get to where they want to go,” $\chi^2(2) = 68.86, p < .001$. As shown in Figure 5, the highest frequency was for being unable to plan (higher than both remaining items, $p < .05$), followed by planning when going to a familiar place, and finally, planning to go to an unfamiliar place ($p < .05$).

### Table 4. Correlations for SCAS subscales with SBSOD, navigation, and navigation anxiety for navigation in Down syndrome

|   | 2   | 3   | 4   | 5   | 6   | 7   | 8   | 9   | 10  | 11  |
|---|-----|-----|-----|-----|-----|-----|-----|-----|-----|-----|
| 1. SBSOD | 0.57** | 0.55* | 0.42* | 0.12 | 0.56** | 0.56** | 0.46* | 0.37* | 0.58** | 0.10 |
| 2. Navigation | / | 0.49* | 0.32 | 0.084 | 0.42* | 0.56** | 0.46* | 0.32 | 0.27* | 0.34* |
| 3. SCAS | / | 0.78** | 0.58** | 0.83** | 0.79** | 0.75** | 0.88** | 0.48* | 0.02 |
| 4. OCD | / | 0.20 | 0.67** | 0.56** | 0.40* | 0.70** | 0.30 | 0.08 | 0.11 |
| 5. Social phobia | / | 0.36* | 0.27 | 0.46* | 0.57** | 0.24 | 0.01 | 0.06 |
| 6. Panic or Agoraphobia | / | 0.63** | 0.56** | 0.70** | 0.46* | 0.20 | 0.11 | 0.06 |
| 7. Separation anxiety | / | 0.45* | 0.60** | 0.51** | 0.38* | 0.11 | 0.06 |
| 8. Physical injury fear | / | 0.57** | 0.38* | 0.29 | 0.20 |
| 9. Generalized anxiety | / | 0.29 | 0.03 | 0.11 | 0.06 |
| 10. Navigation anxiety | / | / | / | / | / |
| 11. Age of individual | / | / | / | / | / |

Note. $N = 91$ *$p < .05$, **$p < .001$. Correlations for SBSOD (n = 90)
Table 5. Regression model for SBSOD (N = 90).

|                | β    | t    | p   | F    | df  | p    | Adj. R² |
|----------------|------|------|-----|------|-----|------|---------|
| Group          | .54  | 3.64 | <.001 | 4.97  | 11, 78 | <.001 | 33      |
| Age of individual | .21  | 2.12 | .04  |      |      |      |         |
| Gender of individual | -.03  | -.21 | .84  |      |      |      |         |
| OCD            | -.09 | -.65 | .52  |      |      |      |         |
| Social Phobia  | -.25 | 1.80 | .08  |      |      |      |         |
| Panic or Agoraphobia | -2.9 | -1.07 | .29  |      |      |      |         |
| Separation Anxiety | .64  | 2.20 | .03  |      |      |      |         |
| Physical Injury Fears | -.11  | -2.96 | .004  |      |      |      |         |
| Generalized Anxiety | .30  | 1.73 | .09  |      |      |      |         |
| Navigation anxiety | -.7 | -2.52 | .01  |      |      |      |         |
| Separation Anxiety *Group | -.70  | 2.52 | .01  |      |      |      |         |

**Associations between anxiety and navigation**

Bivariate correlations were conducted for each group to assess the relationships between SCAS anxiety subdomains, chronological age of the individual and navigation anxiety, with SBSOD and navigation (see Tables 3 and 4 for correlation matrices).

To determine the proportion of variation in SBSOD and navigation scores that can be explained by anxiety, two hierarchical regression models were used. All variables were converted to z-scores prior to analyses. Variables were entered in three steps with the enter method used for steps 1 and 2 and a stepwise method for step 3. For step 1, group was entered. Chronological age of the individual and gender of the individual were also entered in step 1 in order to control for any effects of these two variables on SBSOD and navigation scores. For step 2, predictors that significantly correlated with the outcome variable for one or both groups (see Tables 3 and 4) were entered. For step 3, interaction terms of group with each predictor in step 2 were entered. Because the stepwise method was used for step 3, only significant interaction terms were retained in the model. The collinearity statistics indicated appropriate Tolerance (all > .02; Menard, 1995). VIF scores were appropriate (< 10; Myers, 1990), with one exception; VIF values for separation anxiety were higher than 10. This is discussed below, in the context of each regression model.

**Santa Barbara Sense of Direction (SBSOD)**

The final model accounted for 32.9% of the variation in SBSOD scores (see Table 5). For step 1, group (WS or DS), age and gender accounted for 9.4% of the variation. For step 2, z-scores of all six anxiety subscales and navigation anxiety explained a further 19.0% of the variation. Finally, for step 3, only the interaction term between group and separation anxiety was retained by the model. This accounted for a further 4.5% of the variation. Note that the VIF values were above 10 for separation anxiety and the separation anxiety*group interaction. This is not problematic since it simply reflects that separation anxiety is highly correlated with the product of separation anxiety (in this case, separation anxiety*group). The p-values of separation and separation anxiety*group are not affected by this multicollinearity (https://statisticalhorizons.com/multicollinearity). Observation of Tables 3 and 4 demonstrates that this interaction was due to a relationship between SBSOD and separation anxiety for the DS group, but not the WS group. The final model is shown in Table 5 and shows that group, age, separation anxiety, navigation anxiety, and the group by separation anxiety interaction term were significant predictors.

**Navigation competence**

The final model accounted for 30.5% of the variation in navigation competence scores (see Table 6). For step 1, group, age, and gender accounted for 15.8% of variation in the model. For step 2, z-scores of the significant anxiety subscales (social phobia, panic or agoraphobia, separation anxiety, and physical injury fears) and navigation anxiety explained a further 8.5% of variation. Finally, for step 3, only the interaction term between group and separation anxiety was retained by the model. This
Table 6. Regression model for navigation (N = 91).

|                | \( \beta \) | t     | p     | F    | df  | p    | Adj. \( R^2 \) |
|----------------|-------------|-------|-------|------|-----|------|---------------|
| Group          | .69         | 4.03  | <.001 | 5.39 | 8, 81| <.001| .31           |
| Age of individual | .31       | 3.16  | .002  |      |      |      |               |
| Gender of individual | −.11     | −1.26 | .21   |      |      |      |               |
| Social Phobia  | .09         | −.76  | .45   |      |      |      |               |
| Panic or Agoraphobia | −.20     | −1.67 | .10   |      |      |      |               |
| Separation Anxiety | .74      | 2.54  | .01   |      |      |      |               |
| Physical Injury Fears | −.08     | −.75  | .46   |      |      |      |               |
| Navigation anxiety | −.12     | −.95  | .35   |      |      |      |               |
| Separation Anxiety *Group | −.80     | −2.88 | .01   |      |      |      |               |

accounted for a further 6.2% of the variation. Note that the VIF value was above 10 for separation anxiety. This is not problematic, for the reasons explained above. As above, observation of Tables 3 and 4 demonstrates that the group by separation anxiety interaction was due to a relationship between navigation and separation anxiety for the DS group, but not the WS group. In the final model (Table 6), group, age, separation anxiety, and the group by separation anxiety interaction term were significant predictors.

Training needs and training received

Two open-ended questions were included in the novel “feelings about wayfinding” section of the questionnaire related to navigation training needs and training received. With reference to both training needs and training received, we were interested in the extent to which anxiety was a theme, and also what other themes were present. We also examined whether there was a difference in the frequency of anxiety as a theme according to group, and according to the categorical variable of individuals who traveled independently and those who did not. Answers from both questions were analyzed together, collapsed across group, for the purpose of deriving the themes (N = 76 parent responses for training needs; N = 73 parent response for training received).

The formal phase of coding focused on identifying pertinent codes in the data. As recommended by Braun and Clarke (2006) this stage included systematically reading through the entire data set. This was carried out by three researchers. One researcher generated the codes, and a further two researchers independently reviewed the codes and the fit of the codes to the data. Once the codes were agreed and the data had been fully reviewed, themes were identified independently by each researcher and responses from participants were described as relating to a theme. Reflexive thematic analysis was used to identify patterns of meaning in the data and group similar themes into wider, super-ordinate themes. Three thematic maps were created to determine the most relevant themes and to highlight the relationships between them (Braun & Clarke, 2006). Themes that were not related to our central three super-ordinate themes were discarded at this stage.

The thematic analysis revealed three key themes relating to independent travel and training needs. These themes, although discussed independently below, are all interlinked and are therefore not isolated concepts. These themes have been labeled as follows: safety and anxiety about independent travel; parental influence; and learning opportunities.

Theme 1: Safety and anxiety about independent travel

Throughout all responses the overwhelmingly present theme was safety and anxiety about independent travel. This was the predominant driving force behind parents’ behaviors and emotions. Most respondents mentioned safety for their son or daughter. This aligns with the categorical data reported above in which confidence of the parent was the most frequently ticked category for why parents did not permit their son or daughter to travel independently. The qualitative data revealed that lack of safety was often cited as the main reason for not allowing their son or daughter to travel independently
and/or that any training should not only include independent travel skills but also focus on staying safe when traveling alone. Concerns about safety and risk were associated with anxiety in both parents and in their sons/daughters. Safety can mean different things for different parents but there is a consensus in our qualitative data that safety in independent travel is about being safe around traffic and about social vulnerability, i.e., who may be a danger to the individual when traveling alone. For example, when asked about perceived training needs, respondents answered:

“Crossing the road safely. Judging speed. Personal safety - making friends, who is ‘safe’ ” (caregiver of an individual with DS who does not travel independently)

“You could not trust him to keep safe. He has to be with someone at all times.” (caregiver of an individual with WS who does not travel independently)

Parents were worried about the fact that when traveling alone their son/daughter may be more vulnerable and so were reluctant to allow independence. For example, when asked about perceived training needs, one respondent answered:

“[they are] considered vulnerable therefore [they are] always accompanied” (caregiver of an individual with DS who does not travel independently)

Parents’ worry was considered a major barrier to individuals being permitted to travel independently. Parents worry that, although some individuals may have the skills to travel independently, their son’s/daughter’s ability to cope with anxiety, fear of traffic, and coping with unexpected situations limit their ability to do so. Interestingly, unlike parents own worries, parents report that their son’s/daughter’s anxiety is broadly acute and situational and seems to increase their risk level. For the individual with WS or DS, it is often about escaping a perceived dangerous situation, which paradoxically can have the opposite effect. Fears of getting lost and unexpected changes (e.g., missing the bus) were reported to play a pivotal role in the maintenance of anxiety and panic symptoms. For example, when asked about perceived training needs, respondents answered:

“She knows how to do it but when left to do it alone gets panicky. Because she is worried she will get run over and killed” (caregiver of an individual with WS who does not travel independently)

“Not to be scared of birds which can make him panic and run into the path of incoming car” (caregiver of an individual with DS who does not travel independently)

“She has shown erratic behaviour e.g., running into roads after missed buses” (caregiver of an individual with DS who does not travel independently)

When separating the data according to independent travel status (for “training needs” responses, N = 49 do not travel independently, N = 27 travel independently) there is no evidence that anxiety and panic differs according to independent travel status. Anxiety and/or panic was mentioned 6 times for those who do not travel independently (12.2% of N = 49) and 3 times for those who travel independently (11.1% of N = 27). For “training needs” responses, anxiety and/or panic were cited more frequently by caregivers of individuals with William Syndrome (8 mentions out of 39 responders; 20.5%) than caregivers of individuals with Down Syndrome (1 mention out of 37 responders; 2.7%).

**Theme 2: Parental influence**

Parents whose son/daughter were able to travel independently reported that they were proactive in teaching navigation and wayfinding skills to them. Repetition of the same journey was often mentioned as a helpful strategy to enable safe independent travel. For example, when asked about training received, respondents answered:

“[He/She] hasn’t received ‘official’ travel training only our input as parents” (caregiver of an individual with WS who travels independently)
“[She] will travel independently on a bus after my wife and I train her to use [the bus].” (caregiver of an individual with WS who travels independently)

Similarly, when asked about training needs, respondents answered:

“Mum takes her on the bus route to where she needs to go at least 10 x for her to feel safe to independently travel to a new destination” (caregiver of an individual with WS who travels independently)

**Theme 3: Learning opportunities**

Parental consensus regarding learning opportunities and training needs centered on road safety, e.g., safely crossing the road. This aligns with the categorical data above which demonstrated that standard road crossing and judging car speed were the most frequently reported difficulties for individuals with DS and individuals with WS. Parents also often cited training on how to use public transport and map reading as other important learning opportunities. Some parents also talked about opportunities to improve confidence and the usefulness of learning anxiety and panic management skills. Parents seldom mentioned navigation skills without management and training on safety and/or anxiety. For example, when asked about perceived training needs, respondents answered:

“safety on roads, strangers, where buses go and stop” (caregiver of an individual with WS who does not travel independently)

“Road safety and what to do when things go wrong” (caregiver of an individual with DS who does not travel independently)

“Map reading/orientation, road safety” (caregiver of an individual with DS who does not travel independently)

“Help to use public transport, help to identify familiar routes and plan journeys, help to have [an] action plan if lost/danger” (caregiver of an individual with WS who does not travel independently)

“relaxation techniques to stop her worrying and allow her to concentrate on where she was going” (caregiver of an individual with WS who travels independently)

**Discussion**

To-date, studies of navigation in WS and DS have investigated cognitive competence when navigating. This has been useful in determining cognitive limitations, and based on this knowledge, it is now timely to investigate other limitations to successful navigation in these groups. The current study sought to determine the complexity of independent navigation in these groups with a specific focus on the relationship between anxiety and navigation. We found a relationship between navigation anxiety and navigation competence, and for the DS group, between separation anxiety and navigation competence. Additional impacts on independent navigating also emerged: these related to specific difficulties with tasks such as road crossing, cross-syndrome differences in strategies when lost or when navigating, as well as the impact of social vulnerability, and parental confidence and anxiety regarding their son’s or daughter’s safety. Each is discussed in detail below.

Absolute levels of navigation demonstrated low scores, indicative of poor navigation skills. For the SBSOD, scores were lower than reported by Hegarty et al. (2002) for neurotypical adults. For navigation competence, the mean scores indicate agreement with negative navigation statements (e.g., “my son/daughter gets lost a lot”). However, note that there were large individual differences (Figure 1). The DS group had relatively stronger navigation abilities than the WS group. This pattern is contrary to expectation, given that behavioral studies which have included cross-syndrome comparison between DS and WS groups typically demonstrate stronger performance in WS compared to DS in both route knowledge and configural knowledge (Farran et al., 2015; Purser et al., 2015). Observation of Figure 1 demonstrates, however, that there is substantial overlap in navigation levels
between DS and WS groups and, similarly, overlap between groups in anxiety levels. Individuals with DS had lower levels of anxiety than individuals with WS. While this pattern is consistent with the literature (Vicari et al., 2016), the percentage of individuals with DS with clinically elevated anxiety was high relative to the literature (e.g., Vicari et al., 2016). This could simply reflect individual differences in samples across studies, and calls for a meta-analysis of anxiety in DS. For the WS group, the percentage with clinically elevated levels of anxiety was consistent with the literature (e.g., Royston et al., 2017).

Approximately half of all individuals with WS or DS had received some form of travel training, and although lower proportions of each group were permitted to travel independently, these two variables were associated. The causal direction of this association is unknown; it could be that travel training has a positive impact on independence or that parents are more likely to pursue some travel training for their son/daughter if they have permitted them some independence. There is some consistency with Yang et al. (2017), however, who report a positive association between parents teaching their son/daughter navigation skills, and their son’s/daughter’s navigation confidence. As observed in the qualitative analysis, parental worries about safety and social vulnerability, in addition to navigational competence, are likely to contribute to decisions regarding independent travel.

When asked what individuals find hard when navigating, a need for support when crossing the road (either via special crossings or a caregiver) was strongly emphasized. Difficulties with car speed and crossing the road were experienced by the majority of individuals. Even in situations where traffic support was available such as traffic lights or special crossings (e.g., pelican crossings or zebra crossings) over 20% of individuals had difficulty. These difficulties were mirrored in the qualitative analysis, where a common training need reflected safety when crossing a road. Difficulties stepping down from the pavement were also experienced by both groups, emphasizing the negative impact of motor and perceptual difficulties (Farran & Karmiloff-Smith, 2012; Lanfranchi, 2019) on navigation.

When asked about planning and strategy use, the majority of individuals were not able to plan alone. Planning is a subdomain of executive function, and this finding aligns with evidence that both individuals with WS and individuals with DS have poor planning performance (Costanzo et al., 2013; Lanfranchi, Jerman, Dal Pont, Alberti, & Vianello, 2010). When asked about the strategies used when their son/daughter was lost or needed to find their way, group differences emerged due to the WS group having a stronger tendency to ask someone to help them, than the DS group. When lost, asking a safe person, as well as texting/phoning were the main strategies for individuals with WS, and for individuals with DS the main strategy was to walk around, closely followed by texting/phoning. Similarly, navigation strategies demonstrated a stronger tendency in WS to ask for help than in DS. Nevertheless, for both groups, asking someone to help them to navigate was the dominant strategy, along with remembering things (objects or buildings) that they see along the way (i.e., use of landmarks to remember a route). Group differences in asking for help could reflect the WS characteristic of hypersociability (Jones et al., 2000) or could relate to known differences in communication between these groups, in which individuals with DS are relatively poorer (Del Cole, Caetano, Ribeiro, Kümmers, & Jackowski, 2017). Asking for help is a useful strategy, but the high level of sociability in WS can make these individuals socially vulnerable. This was strongly reported in the qualitative analysis, where for both groups, safety when navigating was related to social vulnerability and knowing who is a safe person. Social vulnerability is high for both individuals with WS and individuals with DS, and has some syndrome-specific patterns, associated with responding appropriately to strangers and detecting risk, in WS and in DS, respectively (Fisher et al., 2013), both of which are relevant to successful navigation. Furthermore, qualitative analysis indicated that parent knowledge of their son’s or daughter’s social vulnerability impacts the restrictions that they place on their navigation experiences. This might have a downstream impact on the level of navigation anxiety of the individual. This is supported by evidence that parents’ behaviors and emotions influence a child’s level of worry and navigation confidence in
DS (Yang et al., 2017). This is particularly relevant in the development and maintenance of anxiety disorders and the ability to travel independently, where a parent can become a safe person (i.e., a trusted person who provides safety/security in an anxiety provoking situation) (Rachman, 1984).

Parents reported that their son/daughter uses landmarks as a strategy for navigating. This is aligned with previous research, which suggests that, with sufficient practice, individuals with WS and individuals with DS can gain both landmark knowledge and route knowledge (Farran et al., 2015). The relative inflexibility of a fixed route and limitations in the ability to choose appropriate landmarks can, however, impede successful navigation (Courbois et al., 2013; Farran et al., 2016; Nardini, Atkinson, Braddick, Burgess, 2008), and likely contributes to the overarching need to ask someone to help.

Associations between anxiety and navigation demonstrated that navigation anxiety was a significant predictor of SBSOD scores, but not navigation competence scores, for both groups. Thus, the pattern of relatively low anxiety and relatively high navigation skills in DS, compared to the opposite pattern in WS, might be partly accounted for by this association. Specifically, higher navigation anxiety was associated with poorer sense of direction as measured by the SBSOD. Furthermore, higher separation anxiety (a subdomain of the SCAS) predicted poorer navigation as measured by both the SBSOD and navigation competence scores, in DS only. The relationship with navigation anxiety reflects an influence of situation-specific anxiety, a pattern that was also reflected in the qualitative analyses. At first look, however, it is surprising that this pattern was not consistent across our two measures of navigation, SBSOD score and navigation competence score. This likely reflects the nature of the navigation questions asked; the SBSOD measures sense of direction, which can be considered a trait (e.g., Cornell et al., 2003), and the navigation score measures wayfinding skills, i.e., an aspect of spatial cognition. It has been suggested that sense of direction is poor if one has less experience, and thus one way to positively manipulate sense of direction is to increase engagement with the environment (Condon et al., 2015). Thus, we propose that anxiety might relate to sense of direction here on account of reduced experience and avoidance of navigation situations. Viaud-Delmon et al. (2002) reported an association between general anxiety and configural knowledge, but not route knowledge. Given that individuals with WS and individuals with DS rarely develop configural knowledge, the lack of association between general anxiety and navigation competence is consistent with Viaud-Delmon et al. (2002). Furthermore, we know that navigation competence is also limited by cognitive factors such as age, IQ, long-term memory, and executive functions (e.g., Purser et al., 2012), factors not measured here. Our analyses demonstrated that both sense of direction and navigation competence were stronger with increasing age. It is also possible that, for wayfinding, cognitive limitations dominate over any relationship with anxiety. Future research should investigate the relationship between IQ, long-term memory, executive function, and anxiety, with navigation within the same model.

The association between situation-specific anxiety and navigation is consistent with evidence from the general population that anxiety has a negative impact on navigation (Lawton & Kallai, 2002; Pazzaglia et al., 2018; Walkowiak et al., 2015). We did not measure neural association here, but our findings are also consistent with a hippocampal association between anxiety and navigation as suggested by Mueller et al. (2009).

For the DS group, separation anxiety predicted navigation performance for both measures. This syndrome-specific association is not accompanied by an increased incidence of separation anxiety in DS. The association is unlikely to reflect an awareness in DS of their own social vulnerability because Fisher et al. (2013) report that individuals with DS are less aware of their disability than individuals with WS. Furthermore, it is unlikely to be a knock-on effect of parental confidence (Rachman, 1984) because parental confidence was given as a reason for not permitting independent travel to a similar extent for the WS and DS groups. It could reflect differences in sociability in WS and DS (Jones et al., 2000) given that the characteristic hypersociability in WS extends to approaching unfamiliar people. This is a tentative suggestion, however, and other factors not measured here, such as attachment, should also be taken into account. To our knowledge, there are no studies of attachment in WS.
Mothers of toddlers with DS use more intrusive and directive strategies than mothers of typically developing toddlers, which may lead to insecure attachment patterns in their children (a risk factor for separation anxiety) (Al-Yagon & Margolit, 2012). However, evidence for similar patterns in mother–child interactions for children with non-specified intellectual disability (Feniger-Schall & Joels, 2018) suggests that this is not syndrome specific and would not account for the syndrome-specific finding here. Clearly further research is needed to truly explain this group difference.

The qualitative data emphasize the intricate nature of independent travel for WS and DS individuals and the role that parents have in facilitating that process. This is consistent with Yang et al. (2017) who also report an association between parental concerns and both navigation competence and navigation confidence in DS. In the current study, parents reported that safety concerns played an important role in stopping individuals from traveling independently. Parents’ behaviors and emotions reflected their concerns and anxieties about safety and how unexpected events may place their son or daughter at a higher risk of harm.

This study is a first step to understanding the complex set of mechanisms which contribute to independent navigation in WS and DS. While we concentrated on the relationship between anxiety and navigation, qualitative analysis emphasized the importance of parental anxiety and of social vulnerability of the individual (both of which are related to one another) as also being a large part of the picture of the relationship between anxiety and navigation. These factors could be a target for future research. A questionnaire measure was an ideal way to start to address our current research questions, but we also note that parent-report is not without limitations with respect to reliability. Fisher et al. (2014), however, compared parent report and actual behavior in adults with WS with respect to social approach behavior, and found a high level of agreement. Similarly, in the memory domain, a high level of agreement between parent report and behavior is reported in DS (Spanò & Edgin, 2017). This, along with broad agreement with the literature on the measures of navigation and anxiety in our questionnaire, provides reassurance that our data are reliable. The lack of demographic information about the parents (e.g., gender, age, and socioeconomic status) or the environment (e.g., urban and rural) in this study is also a limitation; this did not allow us to take these variables into account in our analyses.

To move beyond parent-report, future research could extend this line of questioning by including physiological measures (e.g., galvanic skin response and heart rate) during navigation tasks for individuals with WS and individuals with DS. A clearer understanding of the role of anxiety in navigation in WS and DS, at both behavioral and neural levels, has the potential to lead to the development of more effective training schemes by addressing navigation difficulties that are compounded by anxiety. This knowledge will also inform professional services of the complexities of the limitations to independent living in these groups. For example, our data suggest that road safety and navigation training are paramount to empowering individuals to travel independently. However, due to difficulties in dealing with unforeseen changes and heightened anxiety, individuals with WS and DS would also benefit from anxiety management training and training on how to stay safe when dealing with unexpected events.

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