

**SELF-UNDERSTANDING IN ADOLESCENTS WITH AUTISM SPECTRUM CONDITIONS,
DOWN'S SYNDROME AND WILLIAMS SYNDROME.**

Helena Tucker

2015

Submitted in partial fulfilment of the degree of Doctorate in Clinical Psychology
at the Division of Clinical Psychology, Liverpool University

ACKNOWLEDGEMENTS

This research would not have been possible without the help of a number of people, to only some of whom it is possible to give particular mention here.

First, I would like to express my great gratitude to Dr Laura Golding and Dr Dougal Hare, my research supervisors, for their invaluable guidance, continued encouragement and support.

My grateful thanks are extended to all the young people and parents who generously gave their time to participate in the study. Special thanks goes to the Down's Syndrome Association UK, MENCAP, the Williams Syndrome Foundation UK, and the Schools involved for their kind help with participant recruitment.

This work could have not been completed without the help of my parents. I am very thankful for their continued support on every step of the way.

TABLE OF CONTENTS

	Page
THESIS OVERVIEW	7
CHAPTER 1: LITERATURE REVIEW	
Systematic Review of Self-Concept in Adolescents with and without Intellectual Disabilities	9
Abstract	9
Introduction	10
Method	12
Search Strategy	12
Inclusion Criteria	12
Quality Assessment	12
Data Extraction	13
Results	14
Discussion	23
Limitations and Future Research Directions	24
Theoretical and Clinical Implications	26
Conclusions	28
References	31

CHAPTER 2: EMPIRICAL PAPER

Self-Understanding in Adolescents with Autism Spectrum Conditions, Down's Syndrome and Williams Syndrome

Abstract	39
Introduction	40
Method	46
Participant Characteristics	46
Power Calculations	46
Ethical Approval	47
Sampling Procedure	47
Measures	48
Data Analysis	50
Results	50
Self-as-Object/ 'Me'	51
Level of Self-Characteristics	54
Self-as-Subject/'I'	54
Discussion	59
Theoretical and Clinical Implications	62
Strengths	64
Limitations	65
Conclusions	66
References	68

LIST OF TABLES

CHAPTER 1: LITERATURE REVIEW

Table 1: Summary of Quality Assessment Results	19
Table 2: Summary of Articles Included in the Review	20

CHAPTER 2: EMPIRICAL PAPER

Table 1: Sample Characteristics	50
Table 2: Summary of Between-Group Comparison	51
Table 3: Proportion of Self-as-Object Statements	52
Table 4. Summary of Distribution of Self-as-Object Chunks	54
Table 5. Summary of Distribution of Self-as-Subject Chunks	57

LIST OF FIGURES

CHAPTER 1: LITERATURE REVIEW

Figure 1. Search Process Used in Identifying Articles for the Systematic Review	14
---	----

CHAPTER 2: EMPIRICAL PAPER

Figure 1. Development of Self-as-Subject/“I” & Self-as-Object/“Me” (Damon & Hart, 1988)	
---	--

LIST OF APPENDICES

Appendix 1 to Systematic Review:

Modified Version of the Quality Assessment Tool for Studies with Diverse Designs and Scoring

Guidance Notes 30

Main Appendices:

1. Author Guidelines	74
2. Method	76
2.1 DClinPsy Research Review Committee Approval	76
2.2 Information Pack for Parent/Carer	77
Parental/ Carer Letter	78
Information Sheet for Parent/Carer	79
Participant Information Sheet for the Young Person	82
Consent Form	84
2.3 Measures	85
2.3.1 The British Picture Vocabulary Scale-II (BPVS-II)	85
2.3.2. Wechsler Abbreviated Scale of Intelligence (WASI)	85
2.3.3 The Smarties Test	86
2.3.4. Self-Understanding Interview	87
2.3.4.1 Interview Schedule	87
2.3.4.2 Coding Instructions	88
2.3.4.3 Demonstration of Coding Process	92
References	102

Thesis word count: 24, 246

THESIS OVERVIEW

This thesis comprises a literature review, “A Systematic Review of Self-Concept in Adolescents with and without Intellectual Disabilities (ID)”; an empirical paper, “Self-understanding in adolescents with autism spectrum conditions (ASC), Down’s syndrome (DS) and Williams syndrome (WS)” and relevant appendices.

Self-concept is at the centre of a person’s psychological functioning and well-being (Elmer, 2001). It has been linked with important life outcomes in people with ID. A gap in the research was highlighted as no systematic review comparing self-concept specifically in adolescents with and without ID has been identified. The aim of the literature review, therefore, was to explore whether self-concept in adolescents with ID differs to that of adolescents without ID. Thirteen studies were reviewed and mixed findings were presented with global self-concept being mostly equivalent in both ID and non-ID groups. Although, on some domains, adolescents with ID scored lower than controls, it was concluded that adolescents with ID generally did not have lower self-concept compared to those without ID. The merits of exploring cross-syndrome differences and similarities in ID were discussed and it was proposed that more subtle patterns of performance and functioning can be revealed in cross-syndrome comparisons rather than contrasting ‘impaired’ and ‘intact’ functioning (Cebula, 2010; Karmiloff-Smith, 2010).

The empirical paper reported a study which aimed to explore self-concept in 45 adolescents with ASC, DS and WS by examining their verbal concepts, opinions and perceptions with the use of the Self-understanding Interview (Damon & Hart, 1988). The results support evidence for a specific abnormality in social self-functioning in people with ASC with implications for the development of self-concept. The distinctions in social and communication profiles in adolescents with ASC, DS and WS were discussed as well as common difficulties with more sophisticated social competences in all three groups associated with the development of self-concept. As self-concept has been linked with a range of life outcomes, including mental health problems (Harrop & Trower, 2001), the need for individualised interventions promoting positive self-concept is highlighted together with the need for further research in this area.

The systematic review and empirical paper are both planned for submission for publication in the Journal of Autism and Development Disorders. Author guidelines (Appendix 1) were followed.

References:

- Cebula, K. R., Moore, D. G., & Wishart, J. G. (2010). Social cognition in children with Down's syndrome: challenges to research and theory building. *Journal of Intellectual Disability Research*, 54(2), 113-134.
- Damon, W., & Hart, D. (1988). *Self-Understanding in Childhood and Adolescence*. Cambridge: Cambridge University Press.
- Elmer, N. (2001). *Self-esteem. The costs and causes of low self-worth*. Yorn: Joseph Rowntree Foundation.
- Harrop, C., & Trower, P. (2001). Why does schizophrenia develop at late adolescence? *Clinical Psychology Review*, 21(2), 241-65.
- Karmiloff-Smith, A. (2010). *The importance of cross-syndrome comparisons for understanding autism: a developmental approach*. Birkbeck Centre for Brain & Cognitive Development University of London, Zaragoza talk, November 2010.

CHAPTER 1: LITERATURE REVIEW

A Systematic Review of Self-Concept in Adolescents with and without Intellectual Disabilities

ABSTRACT

This review aimed to compare self-concept in adolescents with and without intellectual disabilities (ID). Using a systematic review strategy, articles were identified by searching PsychInfo, MEDLINE and Scopus electronic databases. Inclusion criteria were: (1) adolescents with ID aged between 12-19 years (2) studies comparing self-concept in adolescents with and without ID or with normative data; (3) full manuscript written in English. A total of thirteen studies were reviewed. Two reviewers independently assessed the quality of the articles using the Quality Assessment Tool for Studies with Diverse Designs. The review presented mixed findings with global self-concept being mostly equivalent in both groups. It was not possible to conclude that adolescents with ID generally have lower self-concept compared to those without ID.

Key words: Self-concept, intellectual disability, systematic review, adolescents.

A Systematic Review of Self-Concept in Adolescents with and without Intellectual Disabilities

Self-concept is a multidimensional construct defined as a set of self-attitudes that describe a person's own behaviour, characteristics and abilities (Piers, 1994). Self-concept is of central importance to psychological functioning and well-being (Taylor & Brown, 1988). The construct encompasses a person's perception of self and own identity, i.e. awareness of one's attributes and behaviour, as well as an evaluative aspect, i.e. one's own evaluation of these perceptions, often described as global self-worth or self-esteem (Belmore & Cillessen, 2006; Mruk, 2006). Examples of domains of self-concept that have been studied include physical appearance, social acceptance and perceived competencies in areas such as academia and athletics (Hattie, 2014; Marsh & Martin, 2011; Zheng, Erikson, Kingston, & Noonan, 2014).

In adolescence, self-concept is closely linked with the perception of important others (Bracken, 1996) and is associated with factors such as life-satisfaction, liking of self, self-worth and self-determination (Caruso & Gill, 1992; Zheng et al., 2014). Low self-concept in children and adolescents is linked with negative life outcomes, including reduced quality of life (Trzesniewski et al., 2006), increased risk of major mental health difficulties including depression and anxiety (Renouf & Harter, 1990), parasuicidal behaviours, suicide in adolescence (Santos, Saraiva, & De Sousa, 2009), increased antisocial and criminal behaviour (Donnellan, Trzesniewski, Robins, Moffitt, & Caspi, 2005) and lower scholastic achievement (Harper & Marshal, 1991). In comparison, positive self-concept in adolescence is associated with increased resilience and ability to cope with stress, higher academic achievements and better prospects in adult life (Diehl & Hay, 2010; Poon & Lau, 1999). Academic self-concept is closely linked with achievement (Marsh & Martin, 2010; Zheng et al., 2014) and mediates the effects of aptitudes on learning and academic performance (Chamorro-Premuzic & Furnham, 2006).

Harter (1986) described the development of self-concept through childhood to adulthood and proposed that in middle- to late-childhood [ages 8-13 years], children begin to compare themselves to their peers and become more reliable at evaluating their abilities. Consequently, their overall sense of self-worth is influenced by their self-perception of competency in important domains and feelings of inadequacy or poor competence may follow if they perceive themselves as less able than their peers (Silverman, 1983). In turn, such negative conclusions made about one's abilities compared with others may lead to low self-worth, frustration and loneliness; and negatively affect long-term personal development (Bauminger, Shulman, & Agam, 2004; Evans, 1998).

It is assumed that people with ID have a poor self-concept (Silverman & Zigmond, 1983) as the label of ID is associated with stigma and they often experience discrimination and prejudice (Finley & Lyons, 2000; Hastings & Remington, 1993). Young people with ID are often aware of their difficulties resulting in feelings of inadequacy as they do not meet the expectations of their parents and teachers (Jones, Zahl, & Huws, 2010). They face barriers to academic achievement (Johnson, Thurlow, & Stout, 2007) and may experience lower levels of self-determined behaviour linked with self-concept (Pierson, Carter, Lane, & Glaeser, 2008, Zheng et al., 2014).

Studies comparing self-concept in children and adolescents with and without ID have yielded heterogenic results over the last decades. A large body of evidence is relatively old and there is a lack of research in this area in the recent past. Some evidence suggests that young people with ID have relatively high self-concept and evaluate themselves in a positive way (Chapman, 1988; Jahoda, Markova, & Cattermole, 1988), whilst other studies report negative self-concept in young people with ID compared to typically developing controls (e.g. Harter, Whitesell, & Junkin, 1998; Renick & Harter, 1989; Valas, 1999). In their meta-analysis, Bear, Minke and Manning (2002) found that children with ID perceived their academic self-concept less favourably compared to non-ID children with a negligible effect of setting (e.g. general education class with support, resource room) but differences were less clear in other domains of self-concept.

With the equivocal evidence for lower self-concept in young people with ID, it is conjectured that to maintain positive evaluation of self, they may differentially devalue dimensions in which they perform poorly and value abilities in areas where they demonstrate a higher performance (Stanley & Murphy 1997). Therefore, young people with ID may be able to maintain a positive global self-concept despite difficulties in various domains, e.g. academic, social and behavioural (Bear et al., 2002). Similarly, protective factors in maintaining a positive global self-evaluation are provided by parents, teachers and peers (Harter, 1999; Ochoa, Lopez, & Emler, 2007) and an exaggerated self-evaluation of competence (Gresham, Lane, MacMillan, Bocian, & Ward, 2000). Furthermore, the comparison group for people with ID is of significance, with, for example, more favourable comparison with peers with ID and less favourable comparison with peers without ID in a mainstream school (Crocker & Major, 1989; Moller, Streblow, & Pohlmann, 2009). In addition, the difficulties with definition of ID and self-concept as well as numerous self-concept measures contribute to the inconsistencies in studies comparing self-concept in people with and without ID.

No systematic review comparing self-concept specifically in adolescents with and without ID has been identified. This is particularly important as due to the notable changes associated with adolescence that directly impact self-concept, e.g. increased cognitive ability contributing to the formation of abstract self-characteristics and more reliable comparison of self with peers (Harter, 1986). As an altered self-concept in adolescents with ID is linked to important life outcomes and has consequences for psychological well-being, the aim of this paper was to explore whether self-concept in adolescents with ID differs to that of adolescents without ID.

METHOD

Search Strategy

PsychInfo, MEDLINE and Scopus electronic databases were searched for relevant studies published from 1970. The search was expanded by manually searching the references of selected studies and by citation tracking of the selected studies through the Web of Science. Search terms were divided into three categories: self-concept, adolescents and ID. In each category relevant synonyms were searched including self-understanding, self-esteem, self-perception, learning disability*, mental handicap*, mental retard*, syndrome, autism*, and teenagers. The final search yielded studies that included all three categories. Papers identified had these terms in the title, abstract or keyword.

Inclusion Criteria

Studies included in the review met the following criteria: (1) participants were adolescents with ID aged 12-19 years (2) the study compared self-concept in adolescents with and without ID or with normative data; (3) the full manuscript was written in English. Titles and abstracts were initially checked to determine whether the inclusion criteria were met. The complete paper was obtained where it was uncertain whether the criteria were fulfilled.

Quality Assessment

A quality assessment of the studies selected for review was carried out using a modified version of the Quality Assessment Tool for Studies with Diverse Designs (QATSDD) (Sirriyeh, Lawton, Gardner, & Armitage, 2011). The QATSDD has been found to have good reliability and validity for use in the quality assessment of studies

with diverse designs. The QATSDD consists of 16 items. The modified version used in this study omitted three items. Two of these items focused on qualitative research methodology and were therefore irrelevant. The third item removed addresses theoretical frameworks and scores are assigned on the basis of describing broad/specific theoretical basis or model. With more than three decades between the oldest and most recent study reviewed in this paper and subsequent significant changes in theory, scores on this item were felt to be arbitrary and therefore were not included.

The quality assessment was completed independently by two reviewers with disagreements being resolved by discussion to reach a consensus. The modified quality assessment tool and scoring guidance notes are set out in (Appendix to systematic review 1).

Data Extraction

Data were extracted from the selected studies, including study aims and objectives, participant details, measures of self-concept and the main findings.

RESULTS

Searching electronic databases initially yielded N=988 studies. Following electronic and manual removal of duplicates, N=852 titles and abstracts were reviewed. After excluding irrelevant studies, N=24 articles underwent full review. A further ten articles were subsequently ineligible, whilst reference searching and citation tracking of the selected studies yielded another two eligible articles. Overall, N=13 studies were identified and included in the analysis. Figure 1 describes the search process.

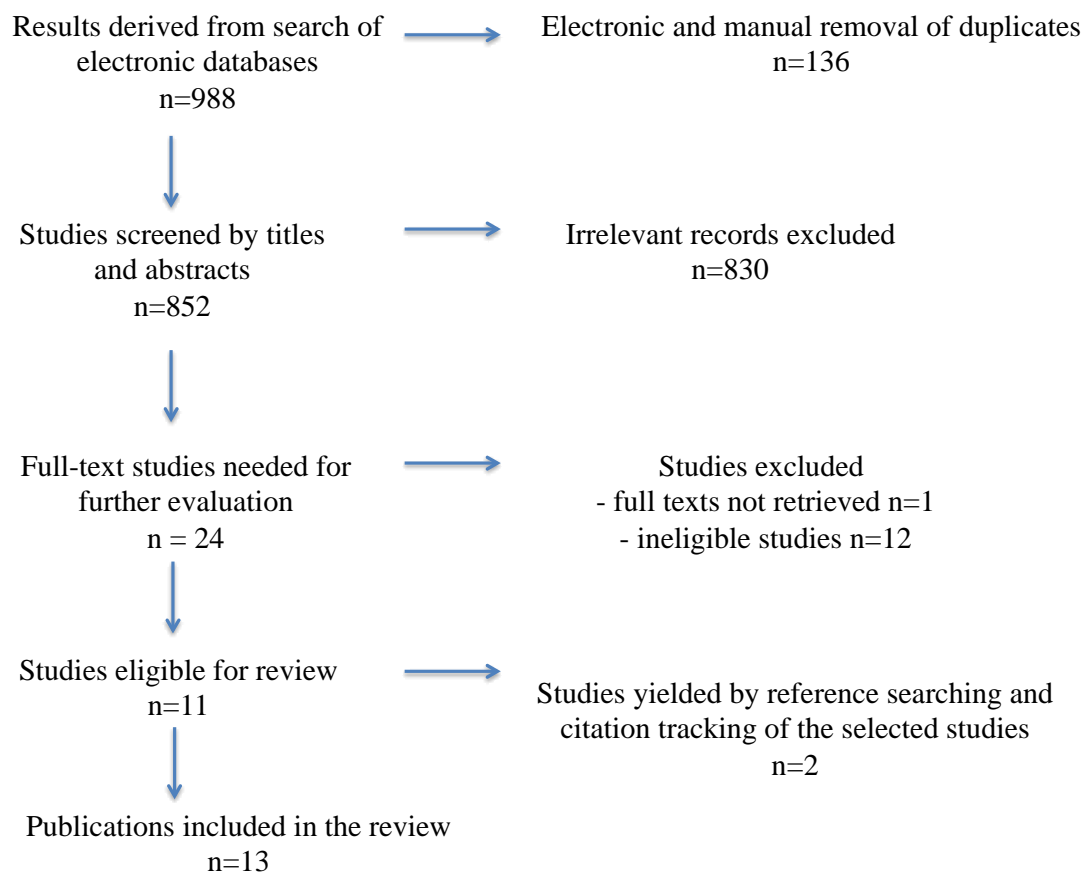


Figure 1. Search process used in identifying articles for the systematic review.

A quality assessment of the reviewed studies was completed using a modified version of the QATSDD (Sirriyeh et al., 2011). Each of the 13 items was scored on a scale of zero to three according to predetermined criteria. Summary of Quality Assessment Results is presented in Table 1. All studies were rated at 0 on two of the QATSDD indices, 'Evidence of sample size considered in terms of analysis' and 'Evidence of user involvement in design'. Total scores ranged from 14 to 23 with a maximum possible score of 39 points. Three quality

categories were assigned, i.e. low (0-13), medium (14-26) and high (27-39). All reviewed studies fell within the medium quality category.

A summary of the data from the reviewed studies is presented in Table 2. Overall areas lacking quality in the reviewed studies were the description of procedure for data collection and recruitment process, rationale for the choice of data collection tools, statistical assessment of reliability and validity of measurement tools and reporting strengths and limitations. Three studies provided very basic and brief outline of the *data collection procedure* [1, 5 & 10] and six studies provided limited detail, or described some stages and omitted others [3, 4, 8, 9, 11 & 13]. Four studies failed to provide a *rationale for the choice of measurement tools* [2, 7, 12 & 13] and three studies only provided basic explanations often omitting important information such as relevance to the study aims and assessment of tool quality. The *description of recruitment method* was also an area that required improvement in a number of the studies reviewed, i.e. six studies provided minimal recruitment data [1, 2, 4, 5, 10 & 11] and four studies provided an incomplete account of the recruitment process often without details regarding the strategy used, numbers of participants approached and recruited, and attrition data where relevant [3, 8, 9 & 13]. A significant limitation was identified in eight studies that did not report a statistical assessment of *reliability and validity of measurement tools* [5-8 & 10-13]. The *discussion of strengths and limitations* in a number of the presented studies also lacked detail and quality. Four studies failed to discuss limitations [2, 10, 11 & 13], four studies provided very limited information and omitted many key issues [1, 5, 6 & 8] and five studies discussed some of the key strengths and weaknesses but did not cover all key aspects, e.g. design, measures, procedure, sample and analysis.

All thirteen studies employed a cross-sectional design. Twelve studies compared ID and non-ID participants whilst one study [11] compared ID participants' scores with normative data. Two studies recruited ID participants from specialist schools for students with ID [2 & 7]. Eleven studies recruited participants from mainstream schools, some of which provided additional support for ID students. The studies used several measures of self-concept administered directly to the adolescents. Seven studies used multi-dimensional measures that yield global self-concept scores by summing individual subscales and these included the *Self-Perception Profile for Children (SPPC)* (Harter, 1985) [2], the *Self-Perception Profile for Adolescents (SPPA)* (Harter, 1988) [4], the *Piers-Harris Children's Self-Concept Scale (PCSCS)* (Piers, 1969) [3, 8 & 11], the *Multidimensional Self-Concept Scale for Children (MSCSC)* (Bracken, 1992), [12]; and the *Culture Free Self-*

Esteem Inventory (Battle, 2002) (CFSEI-3) [6]. One study used the *Offer Self-Image Questionnaire (OSIQ)* (Offer, Ostrov, & Howard, 1997) [9], a multidimensional measure that does not yield a global score. Four studies employed uni-dimensional measures of self-concept, namely *Coopersmith's Self-Esteem Inventory (CSEI)* (Coopersmith, 1967) [1 & 10] and *Rosenberg's Self-Esteem Inventory (RSEI)* (Rosenberg, 1965) [7 & 13]. The *Student's Perception of Ability Scale* (Boersman & Chapman, 1977) (SPAS) [5] was administered in one study to measure academic self-concept.

Two studies reporting self-esteem scores derived from uni-dimensional measures of self-concept found that adolescents with ID attending mainstream schools and receiving supplemental support for their specific ID did not differ on their *self-esteem* scores compared to adolescents without ID [1 & 13]. A significant difference was, however, found in pre-adolescents [1]. Self-esteem in ID adolescents in a specialist school for ID students was lower compared to typically developing controls in mainstream education [7]. Six studies using multi-dimensional self-concept scales reported no significant difference between ID and non-ID groups on global self-concept [3, 4, 6, 8, 11 & 12]. Five of these studies recruited ID participants from mainstream schools and one from a specialist school for students with moderate learning disabilities [2]. One study found lower global self-evaluation in ID adolescents in a mainstream setting [4].

Academic self-concept was reported in eight studies, four of which found no significant differences in the perceived academic/scholastic competence of ID and non-ID participants in a specialist educational setting [2] and a mainstream educational setting [6, 10 & 12]. Four studies reported lower perceived academic/scholastic competence in adolescents with ID in a mainstream educational setting [3-5 & 8].

Perceived *athletic competence* was reported in two studies. No differences were found in a specialist education setting [2], while the other study reported lower perceived athletic competence in ID participants in a mainstream educational setting [5]. Four studies reported scores on the *physical* subscale of the self-concept measures. No difference was reported in a mainstream educational setting [3, 5 & 8] while one study found that ID adolescents in a specialist educational setting scored higher compared to the mainstream control group [2].

Six studies reported scores on the *social/popularity* subscale of the self-concept measures. Four studies found that adolescents with ID scored lower compared to their non-ID peers [2, 4, 8 & 10]. Two studies found no

difference in ID and non-ID participants on social self-concept [3 & 6]. Out of four studies reporting scores on the *behaviour* subscale, three found that ID participants scored lower than their non-ID peers [2-4]. One study found no difference on the behavioural subscale [8].

In addition, two studies reported scores on the *anxiety and happiness* subscales and found no differences in ID and non-ID participants [2 & 8]. One study reported lower scores on *job competence* in ID participants and found no difference on scores on *close friendships* and *romantic appeal* subscales [4].

No differences were reported on the *general* and *parental/home* subscales [6 & 10]. Furthermore, no difference was found on the *personal* subscale [6]. Scores on the OSIQ presented lower scores in the ID group on the *Coping Self* scale consisting of Mastery of External World, Psychopathology, and Superior Adjustment subscales [9]. No differences were found on Family Self and Social Self scales. With regard to *Psychological Self* scale, participants with ID reported lower Body and Self-Image subscale scores and no difference was found on Impulse Control and Emotional Tone subscales [9].

A number of the studies reviewed provided detailed descriptions of participants including their maths and reading abilities, assessment of cognitive ability and academic achievement [3, 5 & 12]. In contrast, no measurement of cognitive or verbal ability of participants was a limitation in other studies [2 & 6]. Study [4] included a number of ethnic groups from six schools increasing the generalizability of the results.

Generalizability of a number of the reviewed studies was affected by factors such as small sample size and inclusion of male students only [1, 6, 7 & 10], recruitment of participants from schools in affluent areas only [1, 5, 6, 9, & 12], and an ethnic minority sample, i.e. primarily Hispanic sample [3]. Other factors include cultural issues in a Christian Arab sample [7] e.g. expectations by parents to achieve in order to improve social status and unusual academic challenges likely to contribute to the formation of academic self-concept, i.e. having to learn three languages. This was also the case with variability in educational provision, e.g. adolescents attending large classes (35 – 40 pupils), and special educational assistance was provided for 90 minutes per day in a separate classroom [7]. Further cultural issues are highlighted as ten of the reviewed studies were conducted in the USA. In addition, participants in one study were described as non-typical of those enrolled in the majority of

public schools' special education programmes due to their relatively high cognitive functioning and academic achievement although given a diagnosis of ID [12].

A discrepancy in the reported conclusions and actual results was seen in study [2]. The conclusions stated a lack of major differences in self-evaluation in line with predictions, however this was not congruent with the results, i.e. although no differences were reported in the over all global self-concept, there were significant differences on three out of six measured subscales. Study [10] reported a trend towards a more negative self-concept based on lower mean scores for participants with ID on a number of domains, however no SD scores were provided and, therefore, it was impossible to determine the significance of these differences. Study [11] did not report cluster score results for their groups, which would be beneficial to enable a more detailed exploration of the results. Study [8] reported a gender effect on self-concept, however there was a small number of female participants in this study and so these results are tenuous.

Table 1: Summary of Quality Assessment Results

	Aims	Research setting	Procedure for data collection	Rationale for choice of data collection tools	Recruitment data	Strengths and limitations	Representative sample	Reliability and validity of measures	Fit between research question and method of data analysis	Total
Bingham, 1980	2	2	1	2	1	1	2	1	3	15
Crabtree & Rutland, 2001	3	2	3	1	1	0	2	3	3	18
Gans, Kenny, & Ghanny, 2003	3	3	2	3	2	2	2	3	3	23
Harter, Whitesell, Junkin, 1998	3	3	2	3	1	2	3	3	3	23
Heibert, Wong, & Hunter 1982	3	2	1	2	1	1	2	0	3	15
Ntshangase, 2008	3	3	3	3	3	1	1	0	3	20
Peleg, 2009	3	3	3	1	3	2	2	0	2	19
Pickar & Tori, 1986	2	2	2	2	2	1	2	0	3	16
Raviv & Stone, 1991	3	2	2	3	2	2	2	3	3	22
Rosenberg & Gaiger, 1977	3	3	1	3	1	0	2	0	2	14
Silverman & Zigmond, 1983	1	2	2	3	1	0	3	0	3	15
Stone & May, 2002	3	2	3	1	3	2	2	0	2	18
Tollefson et al., 1982	2	2	2	1	2	0	2	0	3	14

Table 2: Summary of Articles Included in the Review

	Citation/ Author	Sample/ Participants	Study aims	Measures	Results/ Findings	Quality Rating
1	Bingham, 1980 USA	N=60 30 ID 30 non-ID Mainstream school	Explore differences in self-esteem in boys with specific ID and without ID in 2 age levels, 6th grade and 9th–10th grades.	Coopersmith's Self-Esteem Inventory (Coopersmith, 1967)	Significantly lower self-esteem scores in the ID group.	15
2	Crabtree & Rutland, 2001 UK	N=290 145 ID 145 non-ID School for students with moderate learning disabilities	Explore how adolescents with and without mild to moderate ID differ in the importance they assign to dimensions of comparison.	Self Perception Profile for Children (Harter, 1985)	No difference in the academic/scholastic, athletic, and global self-perception sub-scales. Significantly lower scores on social and behavioural subscales in the ID group. Significantly higher scores on physical sub-scale in the ID group.	18
3	Gans et al., 2003 USA	N=124 50 ID 74 non-ID Mainstream school	Compare adolescents with and without ID on self-concept, using primarily Hispanic sample and examining gender differences.	The Piers-Harris Children's Self-Concept Scale (Piers, 1969)	Significantly lower scores on intellectual/school status and behaviour subscales in the ID group. No difference in global-self concept and in the physical, anxiety and happiness subscales.	23
4	Harter et al., 1998 USA	N=423 118 ID 235 non-ID 70 'behaviourally disordered' Mainstream school	Explore differences in domain specific and global self-evaluations of ID and non-ID adolescents.	The Self-Perception Profile for Adolescents (Harter, 1988)	No difference in the physical, close friendship and romantic appeal subscales. Significantly lower scores on global self-concept, academic/cognitive, athletic, social/peer likability, behaviour and job competence subscales in the ID group.	23

Table 2 Continued

	Citation/ Author	Sample/ Participants	Study aims	Measures	Results/ Findings	Quality Rating
5	Heibert, 1982 USA	N=82 39 ID 43 non-ID Mainstream school	Investigates differences between ID and non-ID adolescents regarding academic self-concept and academic expectations; teacher and parent perception of academic expectation and parental stress.	Student's Perception of Ability Scale (Boersman & Chapman, 1977)	Lower academic self-concept and academic expectations in the ID group.	15
6	Ntshangase, 2008 South Africa	N = 29 14 ID 15 non-ID Mainstream school	Investigates levels of self-esteem in adolescent boys without ID in mainstream school setting and those with ID who moved from a special school setting into an inclusive school setting.	Culture Free Self-Esteem Inventory (Battle, 2002)	No differences in global self-concept and academic, social, general, parental and personal subscales.	20
7	Peleg, 2009 Israel	N=102 52 ID 50 non-ID Special school	Explores differences in test anxiety and self-esteem between ID and non-ID adolescents.	Self-esteem inventory (Rosenberg, 1965).	Lower self-esteem in ID group.	19
8	Picar & Tori, 1985 USA	N=86 39 ID 47 non-ID Mainstream school	Investigate differences in ID and non-ID adolescents in psychological development, self-concept and delinquent behaviour.	The Piers-Harris Children's Self-Concept Scale (Piers, 1969).	No difference in physical, behavioural, anxiety and happiness and global self-perception subscales. Significantly lower scores on popularity/social and academic subscales in the ID group.	16
9	Raviv & Stone, 1991 USA	N=98 49 ID 49 non-ID Mainstream school	Explores differences in self-esteem in ID and non-ID.	The Offer Self Image Questionnaire (Offer, Ostrov, & Howard, 1997)	Lower scores in the ID group on the Coping Self scale consisting of Mastery of external world, Psychopathology, and Superior Adjustment subscales. No differences were found on Family Self and Social Self scales. On the Psychological Self scale, participants with ID reported lower Body and self-image subscale scores and no difference was found on Impulse Control and Emotional Tone subscales.	22

Table 2 Continued

	Citation/ Author	Sample/ Participants	Study aims	Measures	Results/ Findings	Quality Rating
10	Rosenberg & Gaiger, 1977 USA	N=70 (all male) 23 ID 47 non-ID Mainstream school	Explores differences in self-esteem in ID and non-ID adolescents; relationship between self-concept and the number of years in special classroom.	The Coopersmith Self Esteem Inventory (Coopersmith, 1967)	No differences found on the academic subscale. Significantly lower scores on the social subscale for ID participants. Number of years in special class not associated with self-concept.	14
11	Silverman & Zigmond, 1983 USA	N=159 159 ID Normative sample= 1,183 Mainstream school	Explores differences in self-concept in ID and non-ID children and adolescents.	The Piers-Harris Children's Self-Concept Scale (Piers, 1969).	No differences found in global self-concept.	15
12	Stone & May, 2002 USA	N=101 52 ID 49 non-ID Mainstream school	Explores the degree of overestimation of academic skills in students with and without ID.	The Multidimensional Self-Concept Scale for Children (Bracken, 1992)	No significant difference found in general self-concept. Significantly lower academic self-concept in students with ID.	18
13	Tollefson et al., 1982 USA	N= 134 35 ID 99 non-ID Mainstream school	Compares general self-esteem and attributions of ID and non-ID students; and the general attributions and task-specific attribution of ID students.	Self-esteem inventory (Rosenberg, 1965).	No significant difference in self-esteem.	14

DISCUSSION

This paper reviewed thirteen studies with the aim to explore whether self-concept in adolescents with ID differs to that of adolescents without ID. All of the reviewed studies scored within the medium quality category with the total quality scores ranging from 14 to 23 with a maximum possible score of 39. Overall, the main areas lacking quality in the reviewed studies included the description of the procedure for data collection and the recruitment process, providing evidence of considering the sample size in terms of analysis, statistical assessment of reliability and validity of measurement tools, reporting strengths and limitations and using a service user in the research design.

This systematic review of self-concept in adolescents with and without ID identified mixed findings across the studies reviewed. Global self-concept was largely equal for both ID and non-ID participants with the exception of one study [4] reporting lower scores for ID adolescents. These findings appear to be congruent with the limited differences in ID and non-ID students reported in Bear et al.'s (2002) meta-analysis. The lower self-concept in adolescents attending mainstream school [4] may be in line with the hypothesis that comparison with more able peers may lead to negative self-evaluations in ID adolescents (Harter, 1986, 1998). However, this finding was not replicated in other studies with participants from mainstream settings reviewed in the current paper and one study [7] found that ID adolescents in a specialist school had lower self-esteem compared to controls. It is proposed that the number of years in special class is not associated with self-concept [10], which is in line with Bear et al.'s (2002) findings highlighting the negligible effect of educational setting on self-concept.

This review found that adolescents with ID had lower scores on behavioural, job competence and coping-self domains of self-concept when compared to non-ID adolescents. No overall differences were found in this review in the domains of general, parents/home/family self, personal, close friendships, romantic appeal, happiness and anxiety. Mixed findings were presented on academic, athletic, physical and social domains.

It is often assumed that ID adolescents have lower academic self-concept (Chapman, 1988, Gans et al., 2003) but these findings were not supported by the current review. Stone and May (2002) and Strein and Signor-Buhl (2005) conjectured that, although there is some evidence for lower academic self-concept in young people with ID, this group over-predicts their achievements, i.e. their academic self-concept is higher than expected based on their measured skills. The implication here is that students with ID are less accurate in their academic self-evaluation compared to their non-ID peers. However, these comparisons were made with no control for level of achievement and, therefore, need to be interpreted with caution.

Overall, although on some domains adolescents with ID scored lower than non-ID participants, it is not possible from this review to conclude that adolescents with ID, in general, have a lower self-concept compared to those without ID. The lack of conclusive evidence of lower self-concept in ID adolescents leads to hypotheses about strategies young people with ID may use to maintain a positive self-concept. It is proposed that they may devalue domains in which they experience poor competence and ascribe importance to domains in which they excel (Harter, 1999; Stanley et al., 1997). It may be the case that adolescents with ID selectively compare themselves with those of similar ability rather than with more able peers (Moller et al., 2009). Also, they may attribute negative feedback to discrimination or other external factors (Harter 1986, Crocker & Major, 1989). In addition, protective factors that maintain a positive global self-evaluation may be provided by parents, teachers and peers (Ochoa et al., 2007) and an exaggerated self-evaluation of competence (Gresham et al., 2000).

Limitations and Future Research Directions

The studies reviewed are relatively old and there is more than three decades between the oldest and most recent study reviewed. This raises a number of difficulties including changes in classification of ID over the time period. The much-debated lack of definitional clarity has impacted selection of people for research studies in the area of ID as well as identification of people in need of provision of educational and other services (Fletcher, Lyon, Fuchs, & Barnes, 2007). Significant changes have taken place in public and educational policy, and in the protocols and procedures used to identify people with ID, not only in the USA where the majority of the reviewed studies were based, but also in other Anglophone countries. Changes in criteria for classification have implications on the internal and external validity of the ID label used in the reviewed studies. Also, as the educational provision for people with ID has changed significantly the validity of making comparisons of adolescents with ID in mainstream and specialist education is questioned.

The difficulty with the measurement of self-concept is a significant consideration. The variable self-concept scales used in the reviewed studies, and the number of different domains measured, contributed to this review's inconclusive results. Although the majority of tools employed are considered to be the most frequently used and standardised norm referenced measures of self-concept (i.e. PCSCS; RSEI, MSCS; CSEI; SPPC) (Butler & Gasson, 2005), a number of conceptual and methodological issues are raised. The various scales reflect different dimensions of self-concept and self-functioning, e.g. a broad overview of self-characteristics (MSCS), a notion of self-worth (RSEI, MSCS and a domain of SPPC), and self-esteem inferred from PCSCS and CSEI. However, there is an absence of a theoretical stance and framework for many of the scales and variations in how each scale

addresses the multidimensional nature of self-concept (Butler & Gasson, 2005). Some measures have been developed for the use for both children and adolescents; however, with the increase of multidimensionality and more complex organisation of hierarchy through development (Harter, 1999; Butler, 2001) it should not be assumed that measures are suitable for children and adolescents alike. Furthermore, young people may have difficulties with understanding the language used in some measures, (e.g. RSEI and MSCS using '*strongly agree – strongly disagree*'; CSEI '*like me – not like me*'). Many high school students found difficulties with understanding the language used in the CSEI (Byrne, 1983), which is even more pertinent for adolescents with ID.

A number of the scales in the reviewed studies consist of items that have been generated by the authors and often little rationale is provided for the choice of these items (e.g. RSEI, SPPC, MSCS). Concerns regarding the suitability and validity of self-concept measures are illustrated by researchers such as Brinthaup and Erwin (1992), who demonstrated that only 4% of self-descriptions provided by students fall into the pre-determined categories of the widely used PCSCS. To address this issue, Coppersmith sought the advice of psychologists for the selection of the CSEI categories. Still, it is proposed that the young people themselves should provide their own descriptions of their self-concept (Hughes, 1984; Pestana, 2015). In line with these suggestions, Butler (2001) developed a self-concept scale where items were generated by young people themselves and selected based on the frequency with which each item was elicited. Such measures provide increased validity and address the relevance of assessing self-concept by researchers as well as clinicians and educational professionals. Further research to determine the suitability and validity of self-concept measures is necessary. In line with these suggestions, Pestana (2015) asserts that using qualitative methods helps elucidate a more specific and detailed self-understanding of participants, which is more accurate and grounded in real life experiences rather than fitting into questionnaires and predetermined categories. The use of qualitative methodology would be beneficial in exploring areas of difficulty that can then be addressed by clinicians or educational professionals.

It is important to note that there are a number of variables linked with self-concept that were not measured in the reviewed studies. These include socio-economic status, family circumstances, relationship with teachers (Hagborg, 1996; Wiest, Wong & Kreil, 1998), family and school expectations and experiences, self-determination (Zheng et al., 2014), physical attractiveness (Gresham & McMillan, 1997) the adolescents' reference group, (i.e. comparing self to other ID students, typically developing peers, siblings or friends) (Bear et al., 1991), (Zheng et al., 2014), time of diagnosis and emotional adjustment (Punshon, Skirrow, & Murhpy,

2009). Although these variables are not easy to control for, future research should incorporate such variables that are known to play a significant role in the formation of self-concept.

Previous reviews of self-concept (Chapman, 1988; Bear et al., 2002) included studies comparing both children and adolescents with and without ID. The merits of including studies of participants across such a wide age range are questioned as self-concept differs significantly in childhood and adolescence. For this review, due to the notable changes associated with adolescence that directly impact self-concept, specific focus was given to this stage of development. Such changes include an increased cognitive ability contributing to the formation of abstract self-attributes in early adolescence, links emerging between self-concept and perceived validation of self by important others, and increasing ability to make realistic comparisons with others accounting for a more realistic view of self (Harter, 1999). Although only adolescents were included in the studies reviewed, still a relatively wide age range was covered and, therefore, potential differences in self-concept in early and late adolescence may have been missed. A suggestion for future research is to focus on the development of self-concept in adolescence over the three developmental stages, i.e. early, middle and late adolescence, as described by Harter (1999) exploring the developmental trajectory of self-concept in adolescents with ID.

The quality of some of the reviewed studies was modest and strengthening methodological rigour in future studies is important. For example, increasing generalizability by a more representative sample, assessing reliability and validity of measures used, providing data on cognitive and verbal abilities, academic achievement, SES and other variables and controlling for these. Similarly, increasing the quality of reporting in future studies would be of great value, e.g. detailed description of recruitment method, data collection procedure and rationale for the choice of measurement tools.

Although the QATSDD has been found to have good reliability and validity for use in the quality assessment of studies with diverse designs (Sirriyeh et al., 2011), it is important to note that the minor modifications made to the tool in this study may have impacted on its reliability and validity.

Theoretical and Clinical Implications

The premise of comparing ID and non-ID adolescents has been questioned. It is proposed that making the distinction between 'impaired' and 'intact' functioning is limited and recent research has focused on the importance of comparison of people with ID, i.e. across a number of syndromes. Karmiloff-Smith (2010) asserts that exploring cross-syndrome dissociation and associations will help identify similar outcomes in the conditions

studied. In line with this view, a number of studies have compared diverse areas of functioning across various syndromes, e.g. self-concept in people with Williams syndrome and Prader-Willi syndrome (Plesa-Skwerer, Sullivan, Joffe, & Tager-Flusberg, 2004); sociability (Jones et al., 2000), pragmatic aspects of language and social relationships (Laws & Bishop, 2003) and the development of holistic face recognition (Annaz, Karmiloff-Smith, Johnson, & Thomas, 2008) in people with autism, Down's syndrome and Williams Syndrome. Cross-syndrome comparison in the area of self-concept should be considered in future research in order to provide a thorough understating of people with ID with regard to their genotype and allowing clinicians to tailor individualised interventions accordingly.

As discussed, poor self-concept is linked with negative life outcomes and major mental health difficulties. Feelings of inferiority, failure and low-self esteem are linked with higher levels of psychopathology in people with ID (Trzesniewski et al., 2006; Santos et al., 2009). Prospective cohort studies exploring self-concept in people with ID would provide a valuable insight regarding the changes in self-evaluation through development and elucidate causal mechanisms and relationships between self-concept, life outcomes and mental health. Better understanding of this area of research would be beneficial with a view to identifying potential risk factors and needs for early interventions to enhance the quality of life of people with ID.

It is not possible from the findings of this review to conclude that adolescents with ID, in general, have lower self-concept compared to those without ID. Even though no significant difference was reported, it is proposed that when considering interventions aimed at promoting positive self-concept, it is equally important to focus on adolescents with negative self-concept as well as those who may present to conceptualise themselves in a more positive light. Portraying a high self-esteem, amongst other reasons, can be a compensatory measure for difficulties with self-concept demonstrating a desire to be socially accepted (Ntshangase et al., 2008; Elksnin & Elksnin, 2004).

Self-concept can be enhanced in a number of ways and creating an intervention strategy that is unique and tailored specifically to the individual is essential (Dagnan, 2007). A detailed understanding of domains of self-concept and how important these are to the individual is needed in order to identify appropriate strategies. For example, one assigns high importance to cognitive ability while relevant competence does not match expectations leading to a discrepancy and negative effect on self-concept. The intervention here would focus on increasing competence and reducing importance assigned to the dimension if one has unrealistic ambitions (Harter, 1999). Thorough assessment and individualised formulation are necessary and the use of qualitative

measures is encouraged to gain a more detailed understanding of the adolescent's self-concept based on their experiences.

Elbaum and Vaughn (2001) propose that counselling and mediated interventions are effective in positively affecting general self-concept. Clinicians have a significant part to play in promoting self-concept in individual work with people with ID as well in systemic interventions. For example, support can be given to parents to increase their knowledge about ID and promote openness about and acceptance of their child's difficulties, which is associated with increasing positive self-concept in adolescents with ID (Raviv & Stone, 2001). This highlights the role of diagnostic services in supporting parents and the child with ID in providing post diagnostic support, psychoeducation, encouraging parents to be open about the diagnosis with their child, modelling and promoting positive talk and interaction (Reese, Bird, & Tripp, 2007). A link between talk about positive past events and self-esteem (Reese et al., 2007) can inform interventions such as narrative therapy and family therapy and can be easily implemented with families as well as in other settings such as school and residential and day services for people with ID.

Conclusions

This review compared self-concept in adolescents with and without intellectual disabilities. Mixed findings were presented with global self-concept being mostly equivalent in both ID and non-ID groups. Although on some domains, adolescents with ID scored lower than controls, it was concluded that adolescents with ID generally do not have lower self-concept compared to those without ID. The quality of the reviewed studies was assessed and discussed. Issues relating to quality of the studies reviewed included limits of generalizability, lack of definition clarity of ID and its implications, limitations of self-concept measurement tools and the need for developing measures encompassing dimensions and items generated by the young people themselves and using qualitative measures to explore their experiences. Other variables associated with self-concept were considered such as SES and family and school expectations and experiences highlighting the complex interplay of various individual, social and environmental factors. The merits of exploring cross-syndrome differences in ID were also discussed.

Although the results of this review conclude that adolescents with ID do not have lower self-concept compared to those without ID, it is argued that focus on self-concept in both of these groups is fundamental given the significant links with life outcomes. This developmental stage is associated with changes directly impacting self-concept, e.g. increased cognitive ability contributing to more reliable comparison of self with others (Harter, 1986), and therefore enhancing self-concept in these groups is of great value. Creating an intervention strategy

that is unique and tailored specifically to an individual who may experience difficulties with their self-concept is essential. Having an ID is an important consideration with implications for the process of assessing, formulating and providing an intervention. Therefore, adolescents with ID and their families should be offered a thorough assessment and detailed conceptualisation of their difficulties. Clinicians can provide support in a number of settings, e.g. providing timely diagnosis and post-diagnostic support, promoting self-concept in individual work with people with ID as well in systemic interventions with families, schools and other systems around the adolescent.

Appendix 1 to Systematic Review 1: Modified Version of the Quality Assessment Tool for Studies with Diverse Designs and Scoring Guidance Notes

Criteria	0 = Not at all	1 = Very slightly	2 = Moderately	3 = Complete
Statement of aims/objectives in main body of report	No mention at all.	General reference to aim/objective at some point in the report including abstract.	Reference to broad aims/objectives in main body of report.	Explicit statement of aims/objectives in main body of report.
Clear description of research setting	No mention at all.	General description of research area and background, e.g. 'in primary care'.	General description of research problem in the target population, e.g. 'among GPs in primary care'.	Specific description of the research problem and target population in the context of the study, e.g. nurses and doctors from GP practices in the east midlands.
Description of procedure for data collection	No mention at all.	Very basic and brief outline of data collection procedure, e.g. 'using a questionnaire distributed to staff'.	States each stage of data collection procedure but with limited detail, or states some stages in details but omits others.	Detailed description of each stage of the data collection procedure, including when, where and how data were gathered.
Rationale for choice of data collection tool	No mention at all.	Very limited explanation for choice of data collection tool.	Basic explanation of rationale for choice of data collection tool, e.g. based on use in a prior similar study.	Detailed explanation of rationale for choice of data collection tool, e.g. relevance to the study aims and assessments of tool quality either statistically, e.g. for reliability & validity, or relevant qualitative assessment.
Detailed recruitment data	No mention at all.	Minimal recruitment data, e.g. no. of questionnaire sent and no. returned.	Some recruitment information but not complete account of the recruitment process, e.g. recruitment figures but no information on strategy used.	Complete data regarding no. approached, no. recruited, attrition data where relevant, method of recruitment.
Strengths and limitations critically discussed	No mention at all.	Very limited mention of strengths and limitations with omissions of many key issues.	Discussion of some of the key strengths and weaknesses of the study but not complete.	Discussion of strengths and limitations of all aspects of study including design, measures, procedure, sample & analysis.
Representative sample of target group of a reasonable size	No statement of target group.	Sample is limited but represents some of the target group or representative but very small.	Sample is somewhat diverse but not entirely representative, e.g. inclusive of all age groups, experience but only one workplace. Requires discussion of target population to determine what sample is required to be representative.	Sample includes individuals to represent a cross section of the target population, considering factors such as experience, age and workplace.
Statistical assessment of reliability and validity of measurement tool(s)	No mention at all.	Reliability and validity of measurement tool(s) discussed, but not statistically assessed.	Some attempt to assess reliability and validity of measurement tool(s) but insufficient, e.g. attempt to establish test-retest reliability is unsuccessful but no action is taken.	Suitable and thorough statistical assessment of reliability and validity of measurement tool(s) with reference to the quality of evidence as a result of the measures used.
Fit between research question and method of analysis	No mention at all.	Method of analysis can only address the research question basically or broadly.	Method of analysis can address the research question but there is a more suitable alternative that could have been used or used in addition to offer greater detail.	Method of analysis selected is the most suitable approach to attempt answer the research question in detail.
Evidence of sample size considered in terms of analysis	No mention at all.	Basic explanation for choice of sample size.	Evidence that size of the sample has been considered in study design.	Evidence of consideration of sample size in terms of saturation/information redundancy or to fit generic analytical requirements.
Evidence of user involvement in design	No mention at all.	Use of pilot study but no involvement in planning stages of study design.	Pilot study with feedback from users informing changes to the design.	Explicit consultation with steering group or statement or formal consultation with users in planning of study design.

References:

- Annaz, D., Karmiloff-Smith, A., Johnson, M. H., & Thomas, M. S. C. (2009). A cross-syndrome study of the development of holistic face recognition in children with autism, Down syndrome and Williams syndrome. *Journal of Experimental Child Psychology*, 102, 456-486.
- Battle, J. (2002). *Culture-Free Self-Esteem Inventories Examiner's Manual*. (3rd Ed.). Austin, TX: Pro-Ed.
- Bauminger, N., Shulman, C., & Agam, G. (2004). The Link Between Perceptions of Self and of Social Relationships in High-Functioning Children with Autism. *Journal of Developmental and Physical Disabilities*, 16(2), 193-214.
- Bear, G. G., Minke, K. M., & Manning, M. A. (2002). The self-concepts of students with learning disabilities: A meta-analysis. *School Psychology Review*, 31, 405-427.
- Bellmore, A. D., & Cillessen, A. R. N. (2006). Reciprocal influences of victimization, perceived social preferences, and self-concept in adolescence. *Self and Identity*, 5, 209-229.
- Bingham, G. (1980). Self esteem among boys with and without learning disabilities. *Child Study Journal*, 10, 41-47.
- Boersma, F. J., & Chapman, J. W. (1977). *Student's Perception of Ability Scale*. Edmonton: PsiCan Consulting.
- Bracken, B. (1992). *Examiner's manual for the Multidimensional Self-Concept Scale*. Austin, TX; Pro-Ed.
- Bracken, B., A. (Ed. 1996). *Handbook of self-concept: Development, social and clinical considerations*. New York, NY: John Wiley & Sons Inc.
- Brinthaup, T., & Erwin, J. (1992). *The self: Definitional and methodological issues*. SUNY Series, Studying the Self.
- Butler, R. J. (2001). *The Self Image Profiles*. London: The Psychological Corporation.
- Butler, R. J., & Gasson, S. L. (2005). Self esteem/self concept scales for children and adolescents: A review. *Child and Adolescent Mental Health*, 10(4), 190-201.

- Byrne, B. M. (1983). Investigating measures of self concept. *Measurement and Evaluation in Guidance*, 16, 115–126.
- Caruso, C., & Gill, D. L. (1992). Strengthening physical self-perception through exercise. *Journal of Sport Medicine and Physical Fitness*, 32, 416-427.
- Chamorro-Premuzic, T., & Furnham, A. (2008). Personality, intelligence and approaches to learning as predictors of academic performance. *Personality and Individual Differences*, 44(7), 1596-1603.
- Chapman, J. W. (1988). Learning disabled children's self-concepts. *Review of Educational Research*, 58, 347-371.
- Coopersmith, S. (1967). *The antecedents of self-esteem*. San Francisco: W. H. Freeman.
- Crabtree, J. & Rutland, A. (2001). Self-evaluation and social comparison amongst adolescents with learning disabilities. *Journal of Community and Applied Psychology*, 11(5), 347-359.
- Crocker, J., & Major, B. (1989). Social stigma and self-esteem: The self-protective properties of stigma. *Psychological Review*, 96, 608-630.
- Crocker, J., & Major, B. (1994). Reactions to stigma: The moderating role of justifications. In M.P. Zanna, P. Herman (Eds.), *The Ontario Symposium: The psychology of prejudice*, 7, 289-314. Hillsdale, NJ: Erlbaum.
- Dagnan, D. (2007). Psychosocial intervention for people with learning disabilities. *Advances in Mental Health and Learning Disabilities*, 1(2), 3-7.
- Diehl, M., & Hay, E. L. (2010). Risk and resilience factors in coping with daily stress in adulthood: The role of age, self-concept incoherence, and personal control. *Developmental Psychology*, 46(5), 1132–1146.
- Donnellan, M. B., Trzesniewski, K. H., Robins, R. W., Moffitt, T. E., Caspi, A. (2005). Low self-esteem is related to aggression, antisocial behavior, and delinquency. *Psychological Science*, 16:328–335.
- Elbaum, B., & Vaughn, S. (2001). School-based interventions to enhance the self-concept of students with learning disabilities: A meta-analysis. *The Elementary School Journal*, 303-329.
- Elksnin, L. K., & Elksnin, N. (2004). The social-emotional side of learning disabilities. *Learning Disability Quarterly*, 27(1), 3-8.

- Evans, D. W. (1998). Development of the self-concept in children with mental retardation: Organismic and contextual factors. In J. A. Burack, R. M. Hodapp, & E. Zigler (Eds.), *Handbook of mental retardation and development* (pp. 462–489). New York: Cambridge University Press.
- Finlay, W. M. L., & Lyons, E. (2000). Social categorizations, social comparisons and stigma: Presentations of self in people with learning difficulties. *British Journal of Social Psychology*, 39, 129-146.
- Fletcher, J. M., Lyon, G. R., Fuchs, L. S., & Barnes, M. A. (2006). *Learning disabilities: From identification to intervention*. Guilford press.
- Gans, A. M., Kenny, M. C., & Ghany, D. L. (2003). Comparing the self-concept of students with and without learning disabilities. *Journal of Learning Disabilities*, 36, 287–295.
- Gresham, F. M., & MacMillan, D. L. (1997). Social competence and affective characteristics of students with mild disabilities. *Review of Educational Research*, 67, 377– 415.
- Gresham, F. M., Lane, K. L., MacMillan, D. L., Bocian, K. M., & Ward, S. L. (2000). Effects of positive and negative illusory biases: Comparisons across social and academic self-concept domains. *Journal of School Psychology*, 38 , 151-175.
- Hagborg, W. J. (1996). Self-concept and middle school students with learning disabilities: A comparison of scholastic competence subgroups. *Learning Disability Quarterly*, 19, 117 – 126.
- Harper, J. & Marshall, E. (1991). Adolescent's problems and their relationship to self-esteem. *Adolescence*, 26, 799-808.
- Harter S. (1985). *Manual for the Self-Perception Profile for Children*. Denver, CO: University of Denver.
- Harter, S. (1986). Processes underlying the construction, maintenance, and enhancement of the self- concept in children. In J. Suls & A. G. Greenwald (Eds.), *Psychological Perspectives on the Self*, 3, (pp. 137–181). Hillsdale, NJ: Erlbaum.
- Harter S. (1988). *Manual for the Self-Perception Profile for Adolescents*. Denver, CO: University of Denver.
- Harter, S. (1999). *The construction of the self: A developmental perspective*. New York: Guilford Press.

- Harter, S., Whitesell, N. R., & Junkin, L. J. (1998). Similarities and differences in domain-specific and global self-evaluations of learning-disabled, behaviourally disordered, and normally achieving adolescents. *American Educational Research Journal*, 35(4), 653-680.
- Hastings, R. & Remington, B. (1993). Connotations of labels for mental handicap and challenging behaviour: a review and research evaluation. *Mental Handicap Research*, 6, 237-249.
- Hattie, J. (2014). Self-concept. Abingdon, Oxon: Psychology Press.
- Hiebert, B., Wong, B., & Hunter, M. (1982). Affective influences on learning disabled adolescents. *Learning Disabilities Quarterly*, 4: 334-343.
- Hughes, H. M. (1984). Measures of self concept and self esteem for children ages 3–12 years: A review and recommendations. *Clinical Psychology Review*, 4, 657–692.
- Jahoda, A., Markova, I., & Cattermole, M. (1988). Stigma and the self-concept of people with a mild mental handicap. *Journal of Mental Deficiency Research*, 32, 103-115.
- Johnson, D. R., Thurlow, M. L., & Stout, K. E. (2007). *Revisiting graduation requirements and diploma options for youth with disabilities: A national study (Tech. Rep. 49)*. Minneapolis: University of Minnesota, National Center on Educational Outcomes.
- Jones, W., Bellugi, U., Zona, L., Chiles, M., Reilley, J., Lincoln, A., & Al., E. (2000). Hypersociability in Williams syndrome. *Journal of Cognitive Neuroscience*, 12, 30–46.
- Jones, R. S. P., Zahl, A. & Huws, J. C. (2001). First-hand Accounts of Emotional Experience in Autism: a qualitative analysis. *Disability & Society*, 16(3), 393-401.
- Karmiloff-Smith, A. (2010). *The importance of cross-syndrome comparisons for understanding autism: a developmental approach*. Birkbeck Centre for Brain & Cognitive Development University of London, Zaragoza talk, November 2010.
- Laws, G., & Bishop, D. (2003). Pragmatic language impairment and social deficits in Williams syndrome: a comparison with Down's syndrome and specific language impairment. *International Journal of Language Disorders*, 39:1, 45-64.

- Marsh, H. W., & Martin, A. J. (2011). Academic self-concept and academic achievement: Relations and causal ordering. *British Journal of Educational Psychology*, 81(1), 59-77.
- Möller, J., Streblow, L., & Pohlmann, B. (2009). Achievement and self-concept of students with learning disabilities. *Social Psychology of Education*, 12(1), 113-122.
- Mruk, C. J. (2006). *Self-esteem research, theory, and practice: Toward a positive psychology of self-esteem*. New York: Springer.
- Ntshangase, S. (2008). A Comparative Study of Self-Esteem of Adolescent Boys with and without Learning Disabilities in an Inclusive School. *International Journal of Special Education*, 23:2, 75-84.
- Ochoa, G. M., Lopez, E. E., & Emler, N. P. (2007). Adjustment problems in the family and school contexts, attitude towards authority, and violent behavior at school in adolescence. *Adolescence - San Diego*, 42(168), 779.
- Offer, D., Ostrov, E., & Howard, K. I. (1977). The Self-Image of Adolescents: A Study of Four Cultures. *Journal of Youth and Adolescence*, 6, 265-280.
- Peleg, O. (2009). Test anxiety, academic achievement, and self-esteem among Arab adolescents with and without learning disabilities. *Learning Disability Quarterly*, 32(1), 11-20.
- Pestana, C. (2015). Exploring the self-concept of adults with mild learning disabilities. *British Journal of Learning Disabilities*, 43(1), 16-23.
- Pickar, D. B., & Tori, C. D. (1986). The learning disabled adolescent: Eriksonian psychosocial development, self-concept, and delinquent behavior. *Journal of Youth and Adolescence*, 15, 429-440.
- Piers E. V. (1994). *Revised Manual for the Piers-Harris Children's Self-Concept Scale*. Western Psychological Services, Los Angeles, CA.
- Piers, E. V., & Harris, D. B. (1969). *Manual for the Piers-Harris Children's Self-Concept Scale*. Nashville, Tenn.: Counselor Recordings and Tests.

- Pierson, M. R., Carter, E. W., Lane, K. L., & Glaeser, B. C. (2008). Factors influencing the self-determination of transition-age youth with high-incidence disabilities. *Career Development for Exceptional Individuals*, 31, 115–125.
- Plesa-Skwerer, D., Sullivan, K., Joffe, K., & Tager-Flusberg, H. (2004). Self-concept in people with Williams syndrome and Prader-Willi syndrome. *Research in developmental disabilities*, 25(2), 119–38.
- Poon, W. T., & Lau, S. (1999). Coping with failure: Relationship with self-concept discrepancy and attribution style. *Journal of Social Psychology*, 139:639-53.
- Punshon, C., Skirrow, P., & Murhpy, G. (2009). The not guilty verdict: psychological reactions to a diagnosis of Asperger syndrome in adulthood. *Autism*, 13(3), 265–83.
- Raviv, D., & Stone, C. A. (1991). Individual differences in the self-image of adolescents with learning disabilities: The role of severity, time of diagnosis, and parental perceptions. *Journal of Learning Disabilities*, 24, 602-612.
- Reese, E., Bird, A., & Tripp, G. (2007). Children's Self-esteem and Moral Self: Links to Parent–Child Conversations Regarding Emotion. *Social Development*, 16(3), 460-478.
- Renick, M. J., & Harter, S. (1989). Impact of social comparisons on the developing self-perceptions of learning disabled students. *Journal of Educational Psychology*, 81(4), 631.
- Renouf, A. G., & Harter, S. (1990). Low self-worth and anger as components of the depressive experience in young adolescents. *Development and Psychopathology*, 2, 293-310.
- Rosenberg, M. (1965). *Society and the adolescent self-image*. Princeton, NJ: Princeton University Press.
- Rosenberg, B. S. & Gaier, E. L. (1977). The Self-Concept of the Adolescents with Learning Disabilities. *Periodicals Archive Online*, 12(48) 489-500.
- Samuels, S. (1977). *Enhancing Self-Concept in Early Childhood*. New York: Human Science Press.
- Santos, J. C., Saraiva, C. B., & De Sousa, L. (2009). The role of expressed emotion, self-concept, coping, and depression in parasuicidal behavior: A follow-up study. *Archives of Suicide Research*, 13, 358-367.

- Silverman, L. K. (1983). Personality development: The pursuit of excellence. *Journal for the Education of the Gifted*, 6(1), 5-19.
- Silverman, R., & Zigmond, N. (1983). Self-concept in LD adolescents. *Journal of Learning Disabilities*, 16, 478-482.
- Sirriyeh, R., Lawton, R., Gardner, P. & Armitage, G. (2011). Reviewing studies with diverse designs: the development and evaluation of a new tool. *Journal of Evaluation in Clinical Practice*, 18(4), 746-752.
- Stanley, K. D., & Murphy, M. R. (1997). A comparison of general self-efficacy with self-esteem. *Genetic, Social, and General Psychology Monographs*, 123, 79-100.
- Stone, C. A., & May, A. L. (2002). The accuracy of academic self-evaluations in adolescents with learning disabilities. *Journal of Learning Disabilities*, 35, 370-383.
- Strein, W., & Signor-Buhl, S. (2005). *Academic self-concepts of adolescents with learning disabilities: A large-scale database study*. Paper (poster session) presented at the Annual Convention of the American Psychological Association, Washington, August 20, 2005.
- Taylor, S. E., & Brown, J. D. (1988). Illusion and well-being: A social psychological perspective on mental health. *Psychological Bulletin*, 103, 193-210.
- Tollefson, N., Tracy, D. B., Johnsen, E. P., Buenning, M., & Farmer, A. (1981). Implementing goal setting activities with LD adolescents. *Psychology in the Schools*, 21(2), 224 – 233.
- Trzesniewski, K. H., Donnellan, M. B., Moffitt, T. E., Robins, R. W., Poulton, R., & Caspi, A. (2006). Low self-esteem during adolescence predicts poor health, criminal behavior, and limited economic prospects during adulthood. *Developmental Psychology*, 42, 381-390.
- Valas, H. (1999). Students with learning disabilities and low-achieving students: Peer acceptance, loneliness, self-esteem, and depression. *Social Psychology of Education*, 3, 173-192.
- Wiest, D. J., Wong, E. H., & Kreil, D. A. (1998). Predictors of global self-worth and academic performance among regular education, learning disabled, and continuation high school students. *Adolescence*, 33, 601-618.

Zheng, C., Erickson, A. G., Kingston, N. M., & Noonan, P. M. (2014). The relationship among self-determination, self-concept, and academic achievement for students with learning disabilities. *Journal of Learning Disabilities*, 47(5), 462-474.

CHAPTER 2: EMPIRICAL PAPER

Self-understanding in adolescents with autism spectrum conditions, Down's syndrome and Williams syndrome.

ABSTRACT

Self-understanding is a fundamental aspect of psychological functioning. This study aimed to explore self-understanding in adolescents with autism spectrum conditions (ASC), adolescents with Down syndrome (DS) and adolescents with Williams syndrome (WS). The Self-understanding Interview was used to examine the verbal concepts and opinions of self of 45 adolescents (15 ASC, 15 DS and 15 WS). Adolescents with ASC generated significantly fewer social statements compared with adolescents with WS. There was a close comparability between adolescents with ASC, DS and WS on the number of self-as-object self-statements indicating similarities in the overall development of self-concept. The development of social self-understanding and its implications were discussed. The need for individually tailored interventions enhancing social skills promoting positive self-concept is highlighted.

Key words: Autism, Down's syndrome, Williams syndrome, Self-understanding, Self-concept

Self-understanding in adolescents with autism spectrum conditions, Down's syndrome and Williams syndrome.

Self-understanding is a conceptual system of thoughts and attitudes about and towards oneself that is formed in early childhood, the development of which continues throughout life (Damon & Hart, 1988; Piers, 1994). This fundamental concept is at the centre of one's psychological functioning and well-being (Elmer, 2001; Glick, Bybee & Zigler, 1997). Self-understanding, also referred to in the literature as self-concept, encompasses the considerations one uses to define self and to distinguish self from others. It includes perceptions of physical and materialistic qualities, activities and abilities, social and psychological characteristics, and philosophical views and values. Further conceptions include past and future directions, beliefs about how one changes over time, and reflections on autonomy and own consciousness (Damon & Hart, 1988). It also includes the evaluative aspect of self-understanding, i.e. one's own evaluation of these perceptions, which is often described as global self-worth or self-esteem (Harter, 1985).

James (1892/1961) distinguished two components of the self. 'Me' is the sum of all personal attributes, with its primary elements including material (body, possessions), social (relations, roles, personality) and spiritual (consciousness, thoughts, psychological mechanisms) constituents. 'I' is the abstract component of self, categorised into four types of experience: agency (autonomy of self), distinctness (how one is different to others), continuity (stability of the 'I') and reflection (self-consciousness). Building on the work of James (1892/1961), Damon and Hart (1988) described the multi-faceted developmental model of self-understanding. They organised the self-as-object, into four constituents of physical, active, social and psychological self and described a development trajectory the child moves through, i.e. 'categorical identifications', 'comparative assessment', 'inter-personal implications' and 'systematic beliefs and plans' (Hart & Damon, 1988, p.10). The 'I', the self-as-subject, is divided into three concepts of continuity, distinctiveness and agency and also follows the above stages. See Figure 1 reproduced from Jackson, Skirrow, & Hare (2012) for details.

Developmental level	Early childhood	Middle and late childhood	Early adolescents—interpersonal	Late adolescents
Organising principle	Categorical identifications	Comparative assessments	Implications	Systematic beliefs and plans
Physical self	Bodily properties, descriptions or material possessions	Capability related physical attributes	Physical attributes that influence social appeal and social interactions	Physical attributes reflecting volitional choices or personal and moral standards
Active self	Typical behaviour	Abilities relative to others, self or normative standards	Active attributes that influence social appeal and social interactions	Active attributes that reflect choices, personal or moral standards
Social self	Fact of membership in particular social relations and groups	Abilities and acts considered in light of the reaction of others	Social-personality characteristics	Moral or personal choices concerning social relations or social characteristics
Psychological self	Momentary moods, feelings, preferences and aversions	Knowledge, cognitive abilities or ability related emotions	Social sensitivity, communicative competence and other psychologically related social skills	Belief systems, personal philosophy, self's own thought processes
"Agency"	External, uncontrollable factors determine self	Efforts, wishes and talents influence self	Communication and reciprocal interaction influence self	Personal and moral evaluations influence self
"Continuity"	Categorical identifications	Permanent cognitive and active capabilities and immutable self characteristics	Ongoing recognition of self by others	Relations between past, present and future selves
"Distinctness"	Categorical identifications	Comparisons between self and other along isolated dimensions	Unique combination of psychological and physical attributes	Unique subjective experience and interpretations of events

Figure 1. Development of self-as-subject/“I” & self-as-object/“me” (Damon and Hart 1988) reproduced from Jackson et al. (2011)

Self-concept has been associated with a range of life outcomes in both cross-sectional and prospective studies. Low self-concept and low self-esteem are linked with reduced quality of life (Bos, Huijding, Muris, Vogel, & Biesheuvel, 2010), lower academic achievement and risk of mental health difficulties, such as depression and anxiety (Coudeville, Gernigon, & Ginis, 2011; Fathi-Ashtiani, Ejei, Khodapanahi, & Tarkhorani, 2007), eating difficulties (Courtney, Gamboz, & Johnson, 2008), para-suicidal behaviours, increased risk taking, self-harm and suicide attempts, suicide in adolescence (Santos, Saraiva, & De Sousa, 2009), and increased antisocial and criminal behaviour (Donnellan, Trzesniewski, Robins, Moffitt, & Caspi, 2005). Conversely, positive self-concept in children and adolescents is linked with increased resilience and ability to cope with stress (Diehl & Hay, 2010), higher academic achievements (Booth & Gerard, 2011; Mohammad, 2010; Rahmani, 2011), improved health and behaviour (Dalgas-Pelish, 2006), peer acceptance (Thomaes et al., 2010) and better prospects in adult life (Paradise & Kernis, 2002).

Young people with intellectual disabilities (ID) are at risk of developing a negative self-concept due to the stigma associated with ID, the common experiences of discrimination and prejudice and their awareness of not meeting the expectations of other people such as parents, teachers or peers (Finlay & Lyons, 2000). Also, a

number of studies have compared self-concept in children and adolescents with and without ID and have reported varying results (see Bear, Minke, & Manning, 2002 and Chapman, 1988 for reviews). More recently, it has been proposed that making the distinction between ‘impaired’ and ‘intact’ functioning in developmental conditions is inadequate, highlighting the importance of cross-syndrome comparison studies (Cebula, Moore, & Wishart, 2010; Karmiloff-Smith, 2010). Karmiloff-Smith (2010) proposes that making cross-syndrome comparisons, particularly cross-syndrome dissociation and associations, will help distinguish similar outcomes in the conditions under investigation. Cross-syndrome comparisons including ASC, DS and WS have helped to elucidate areas such as pragmatic aspects of language and social relationships (Laws & Bishop, 2003) and the development of holistic face recognition (Annaz, Karmiloff-Smith, Johnson, & Thomas, 2008).

The current study focused on self-concept in adolescents with ASC, DS and WS. ASC, DS and WS are neurodevelopmental conditions with known (DS, WS) and evident (ASC) genetic aetiologies that have distinct social phenotypes. Individuals with these conditions experience difficulties with social understanding, e.g. discerning others’ emotional expressions, developing social relationships, understanding social cues and attributing others’ mental states (Gagliardi, Frigerio, Burt, Cazzaniga, Perrett, & Borgattim 2003; Tager-Flusberg, Skwerer, & Joseph, 2006). Yet, there are some clear distinctions in their social and communication profiles.

Social behaviour in WS is defined by three discrete dimensions: enhanced motivational social drive, atypical emotional sensitivity, and increased salience of social stimuli (Järvinen, Korenberg, & Bellugi, 2013; Järvinen-Pasley, Vines, Hill, Yam, Grichanik, & Mills, 2010). In comparison to ASC, WS is associated with sociability and high empathy in response to others (Meyer-Lindenberg, Mervis, & Berman, 2006; Jones, Zahl, & Huws, 2000). Jones et al., (2000) compared sociability in ASC, WS, DS and typically developing controls and reported that participants with ASC were the least sociable while participants with WS were most sociable. Plesa-Skwerer Sullivan, Joffe, & Tager-Flusberg (2004) suggest that due to the emphasis people with WS place on social aspects of their environment, they perceive themselves in the context of their relationships with others and, therefore, their self-understanding is qualitatively different compared to other people with developmental disabilities. Studies exploring sociable traits in WS reported a high frequency of eye contact, lower frequency of displaying negative facial expression (e.g. Jones et al., 2000), and readiness to initiate interaction with others including strangers (Dodd, Porter, Peters, & Rapee, 2010). While enhanced motivational social drive is present, research suggests that people with WS lack skills in sustaining conversations and developing friendships (Davies Udwin, & Howlin, 1998). WS is also associated with attentional problems, social maladjustment and

diagnostically significant non-social anxiety (Davies et al., 1998; Leyfer, Woodruff-Borden, Klein-Tasman, Fricke, & Mervis 2006; Stinton, Tomlinson, & Estes, 2012). Non-social anxiety is a significant concern for adults with WS as it is proposed that it increases with age and leads to isolation (Udwin, 1990). Riby et al. (2014) reported a link between severity of anxiety and social functioning in people with WS including children and adults, which is an important concern when considering one's self-understanding.

People with ASC present with unique cognitive, social, sensory and behavioural impairments (Mesibov & Shea, 1996; Greene, Colich, Iacoboni, Zaidel, Bookheimer, & Dapretto, 2011; Rosset, Santos, Da Fonseca, Rondon, Poinso, & Deruelle, 2011). They have difficulties with the concept of inter-personal self (Capps, Sigman, & Yirmiya, 1995; Lee & Hobson, 1998), perception of relationships between self and others in social cognition (Baron-Cohen, Wheelwright, Hill, Raste, & Plumb, 2001) linked with deficits in communication abilities (Tager-Flusberg & Sullivan, 2000) and there is evidence to support difficulties in social attention and processing of social cues (Dalton et al., 2005; Greene et al., 2011). An overlap is proposed between the socio-communicative functioning of children with ASC and WS (Herguner & Motavalli, 2006) with similarities including social isolation, distractibility, pragmatic deficits, indiscriminately approaching of strangers, hyperacusis, and other types of social impairment (Gillberg & Rasmussen, 1994). Although ASC and WS may appear to be on the opposing sides of the spectrum of sociability, both people with ASC and WS display significant difficulties that affect their ability to interact with others (Philofsky, Fidler, & Hepburn, 2007). It is proposed that relational impairment can lead to difficulties such as identifying with attitudes of others and conceptualizing interpersonal relationships (Hobson, 1993) and that these limitations in self-other experiences in ASC are likely to be linked with social understating of the self (Knott, Dunlop, & Mackay, 2006; Lee & Hobson, 1998).

Young people with DS have been described as having particular strengths in social functioning, communication and social engagement behaviours despite their difficulties with the structural aspects of language (Franco & Wishart 1995; Singer-Harris, Bellugi, Bates, Jones, & Rossen, 1997; Fidler, Most, Booth-LaForce, & Kelly, 2008). Kasari, Freeman and Bass (2003) report strengths in empathic functioning, including prosocial responses. While strengths in the area of social functioning in people with DS are evident in early childhood, more sophisticated social competences and cognitions in later childhood and adulthood are lacking (Wishart, 2007). Unlike in WS, the social strength of people with DS does not seem to involve the social disinhibition and hyper-sociability, for example when communicating with strangers (Laws & Bishop, 2003). Children with DS show higher social competency (Rosner, Hodapp, Fidler, Sagun, & Dykens, 2004) and pragmatic language abilities (Law & Bishop, 2003) compared to children with WS.

An area of research in social cognition, particularly in ASC but also other conditions, focuses on Theory of Mind (ToM), i.e. the ability to attribute mental states to self and others (Premack & Woodruff, 1978) with implications for self-understanding. It is proposed that there are two distinct components of theory of mind, - social-cognitive and social-perceptual (i.e. less related to cognitive abilities and language, involved in making mental and evaluative judgments about others) (Baron-Cohen, 2000; Tager-Flushberg & Sullivan, 2000). There is much evidence for the impairment of ToM in people with autism, both in social-cognitive and social-perceptual components (e.g. Baron-Cohen, Joliffe, Mortimore, & Rebotson, 1997), associated with attempts to explain the psychological origins and the nature of ASC (Baron-Cohen, Leslie, & Frith, 1985). Later studies however concluded that impairment in ToM is not unique in autism (e.g. Yirmiya, Erel, Shaked, & Solomonica-Levi, 1998). It is proposed that although people with WS acquire a sophisticated ToM, there is a developmental delay (Brock, Einav, & Riby, 2008) and specific deficits have been reported on ToM measures such as the false belief task (Porter, Coltheart, & Langdon, 2008). Tager-Flusberg and Sullivan (2000) reported that social-perceptual component of ToM is relatively intact in WS, i.e. in comparison with other groups of similar level of cognitive ability and that ToM is constrained by more general reasoning abilities resulting in problems with complex social reasoning which may account for difficulties in maintaining friendships (Brock et al., 2008). Children with DS also present difficulties on ToM tasks compared to those with non-specific ID and typically developing children (Giaouria, Alevriadoua, & Tsakiridoub, 2009; Yirmiya, et al., 1998) and it is proposed that they perform similarly to people with ASC and other ID (Losh, Klusek, Martin, Sideris, Parlier, & Piven, 2012).

The development of self-understanding is also closely linked with factors that are not condition-specific including environment, parenting style and life experiences. There is evidence for close links with self-concept and variables such as socio-economic status, family circumstances, family and school expectations, experiences and relationships (Wiest, Wong & Kreil, 1998; Zheng et al., 2014), the group of peers the young people compares themselves with (e.g. other people with ID, typically developing peers) (Bear et al., 1991), self-determination (Zheng, Erickson, Kingston, & Noonan, 2014), and time of diagnosis and emotional adjustment (Punshon, Skirrow, & Murhpy, 2009). It is therefore important to acknowledge a degree of variation in the studied groups based on these factors.

Research on self-understanding in adolescents with ASC, DS and WS is limited. There is a need for thorough investigation in order to explore how these young people, at this pivotal stage of development, construe themselves in various domains. This will have implications for existing psychological theory and clinical practice. Difficulties with the development of self-concept have been associated with mental health problems

(Harrop & Trower, 2001) and are, therefore, of clinical importance. For example, disturbances in continuity and distinctness have been linked with a negative effect on personal identity resulting in poor outcomes such as negatively affecting commitment to social relationships and occupations (Marcia, 1980). In addition, examination of self-descriptions of adolescents with these conditions may advance the understanding of maladaptive behaviours and have implications with regard to practical support and interventions. For example, recent research by Skirrow, Jackson, Perry and Hare (2014) proposes that people with AS experience difficulties in developing a functional self-concept linked with behaviours such as collecting and hoarding. In this case, their self-understanding of continuity, particularly difficulties with awareness of own existence in time, may lead to collecting for the purpose of maintaining factors of their sense of self.

The present study aims to provide a cross-syndrome perspective in exploring the nature of self-understanding in adolescents with ASC, DS and WS. The Self-understanding Interview (Damon & Hart, 1988) was used to examine participants' verbal concepts, opinions and self-perceptions. The interview is based on Damon and Hart's (1988) developmental model of self-understanding and has been used in research to explore self-understanding in typically developing children (Malti, 2006), adolescents with eating disorders (Levitt & Hart, 1991) and young people with ID (Jackson et al. 2012; Lee & Hobson, 1998; Plesa-Skwerer et al., 2004).

The present study aimed to explore how adolescents with ASC, DS and WS understand themselves through examining verbally articulated concepts. A degree of heterogeneity in these disorders was acknowledged and the aim was to investigate the following hypotheses based on previous research (Jackson et al. 2012; Lee & Hobson, 1998; Plesa-Skwerer et al., 2004) :

1. Adolescents with ASC will be least likely to understand themselves in a social way, as indicated by the fewest social self-statements, followed by adolescents with DS and then adolescents with WS, who will be most likely to understand themselves in a social way, indicated by providing the most social self-statements.
2. Adolescents with ASC, DS and WS will not differ on the number of *self-as-object* self-statements.
3. Adolescents with ASC will produce the fewest references to systematic beliefs and life plans in the Self-understanding Interview, followed by adolescents with DS and then adolescents with WS, who will produce the most references to systematic beliefs and life plans.

In addition, as stated above, ToM is an area of social cognition previously explored in these conditions. One's ability to attribute mental states to self and others has implications for self-concept and therefore this study

sought to examine ToM in the three groups. Similarities in ToM in people with ASC, DS and other ID have been reported in the literature (e.g. Losh et al., 2012). Also, although Tager-Flusberg and Sullivan (2000) suggested that social-perceptual component of ToM is relatively intact in WS, developmental delay in acquiring sophisticated ToM (Brock et al., 2008) and specific deficits on ToM measures such as the false belief task (Porter et al., 2008) have been observed in WS. Therefore, it was hypothesized that no difference would be found in the three groups on a measure of ToM (the Smarties Test).

METHOD

A cross-syndrome comparison was carried out to explore differences in self-understanding in adolescents with ASC, DS and WS.

Participant Characteristics

Participants were 45 adolescents with a formal diagnosis of ASC, DS or WS (15 ASC, 15 DS and 15 WS). Participants were included if they were aged between 12 and 20 years, fluent in English and scored on the British Picture Vocabulary Scale II above 5 years range. Exclusion criteria were a diagnosis of another intellectual disability syndrome or a diagnosis of both ASC and DS. Participants were recruited with the help of the Down's Syndrome Association UK (DSA), MENCAP, the Williams Syndrome Foundation UK (WSF) and one specialist school in the North West of England. The geographical area of recruitment included the Midlands, North East and North West of England.

Power Calculations

Past research used the *Self-understanding Interview* to compare self-concepts in adolescents with ASC and non-autistic individuals with intellectual disability matched for age and verbal ability (Lee & Hobson, 1998). The two groups are relevant to the current study of adolescents with ASC and non-autistic participants with intellectual disability, applicable to WS and DS. In Lee and Hobson's study (1998) large effect sizes were detected. These studies informed the power calculations for the study:

Hypothesis 1: The effect size calculated from social self-statements comparisons ($M = 1.5$ in ASC and $M = 5.6$ in non-ASC) was 2.04. It was calculated that for 80% power, effect sizes of 2.04 could be detected using ANOVA with 5% significance level, and assuming equal group sizes, for total sample size of 9.

Hypothesis 2: The effect size calculated from *self-as-object* comparisons ($M = 12.8$ in ASC and $M = 14.4$ in non-ASC) was 0.80. It was calculated that for 80% power, effect sizes of 0.8 could be detected using ANOVA with 5% significance level, and assuming equal group sizes, for total sample size of 21.

Hypothesis 3: The effect size calculated from level 4 self-statements comparisons ($M = .167$ in ASC and $M = 1.40$ in non-ASC) was 0.61. It was calculated that for 80% power, effect sizes of 0.61 could be detected using ANOVA with 5% significance level, and assuming equal group sizes, for total sample size of 30.

No differences in *self-as-subject* statements were found in ASC and non-ASC groups in previous research (Lee & Hobson, 1998) and, therefore, no specific prediction was made. Even though previous research has shown a large effect size with a relatively small sample size (12 ASC and 10 non-ASC), in order to maximise generalizability, the sample size for the current study was increased to 15 participants in each group.

Ethical Approval

Ethical approval was sought from and granted by the University of Liverpool's Research Ethics Committee (see Appendix 2.1).

Sampling Procedure

Two specialist schools in the North West of England, selected on the basis of their Specialist School Status and their provision for pupils with ASC, agreed to contact potential participants and their parents and inform them of the project. Eight participants were recruited through one of these school and no responses were obtained from the other. MENCAP and the DSA advertised the project through their social media and their websites. Local MENCAP and DSA groups in the Midlands, North East and North West of England were provided with information about the project to forward to their members. In response to this advertisement, eight ASC and 15 DS participants were recruited. WS participants were recruited through the WSF. Due to the nature of the project advertisement and local groups contacting their members independently of the researcher, the actual numbers of potential participants contacted are unknown.

Particular emphasis was placed on ensuring that each potential participant made an informed choice about their participation (Dye, Hare, & Hendy, 2005). Consent to participate was sought from the young person and their parent/carer. The first author made initial contact with potential participants' parent/carer via a letter or email including a parent/carer information sheet, information sheet for the young person and a consent form (see appendix 2.2). Parents were asked to complete and sign a consent form and return this to the researcher. The

consent to participate from the young people was sought by means appropriate to their age and competence level. The participant information sheet was discussed with the young person and verbal consent sought. This was in the presence of an appropriate guardian. Participants were given ample opportunity to understand the nature, purpose and anticipated outcomes of the research participation, so that they were able to consent to the extent that their capabilities allowed. Participants were seen in their school or their family home depending on their and their parents' preference. In schools, sessions took place in a quiet area easily accessible by staff. Parents were usually present in sessions carried out in the family homes. The time spent conducting the research procedure was approximately 60 minutes. In appreciation for their time, participants were each given a £5 store voucher. Following consent from parents, one adolescent (ASC) declined to participate and four adolescents (three with DS and one with WS) were unable to participate due to their limited verbal communication.

Measures

The British Picture Vocabulary Scale-II (BPVS), a widely used, normed-referenced measure with evidence of good validity and reliability (Dunn, Dunn, Styles, & Sewell, 2009), was used to measure receptive vocabulary level. The BPVS is commonly used in comparison studies with people with ID (Annaz et al., 2009; Wilson, Pascalis, & Blades, 2007). (See appendix 2.3.1).

Wechsler Abbreviated Scale of Intelligence (WASI) (Wechsler, 1999) is a reliable measure of cognitive functioning that is normed against the Wechsler Adult Intelligence Scale–Third Edition (WAIS-III) (Wechsler, 1997). The two-subtest format of the WASI was used to provide a brief measurement of participants' general level of cognitive functioning. (See appendix 2.3.2).

Measure of Theory of Mind – The Smarties Test (Perner, Frith, Leslie, & Leekam, 1989), a widely used measure of theory of mind (ToM), was used to assess participants' ability to attribute mental states to both themselves and others (Frith, 1989; Frith & Happé; 1994; Premack & Woodruff, 1978). (See appendix 2.3.3).

The Self-Understanding Interview (Damon & Hart, 1988) is based on Damon and Hart's developmental model of self-concept (1988). The measure has been used in previous research with people with ID (Jackson et al., 2011; Plesa-Skwerer et al., 2004). In studies with children with autism compared to typically developing children, the interview demonstrated theory-consistent differences (Lee & Hobson, 1998; Yoshii & Yoshimatsu, 2003). The measure has a high level of inter-rater agreement (84 per cent, $k = .75$), and good internal consistency ($\alpha = .70-.83$). Also, 1-month test-retest reliability was satisfactory for a developmental measure ($r = .49$).

Participants were interviewed using an interview schedule (see appendix 2.3.4.1). The procedure requires, when the participant provides a self-characteristic (*"I am tall"*), the interviewer to ask further probe questions to elicit the meaning or importance of the characteristic (*"Why is that important for me to know?"*). A scoring unit is described as a "chunk" of reasoning, i.e. a self-characteristic mentioned by the participants (e.g. *"What kind of person are you."* *"I am a boy."*) and responses to probe questions explicating the meaning or importance (e.g. *"Why is being a boy important?"* *"Because it makes me strong"*). Scoring proceeded initially by identifying one of seven categories of self-concept for each chunk:

Four self-as-object/ 'Me':

- *Physical* - statements concerned with physical body or material possessions.
- *Active* - statements concerned with activities or abilities.
- *Social* - statements concerned with social personality characteristics.
- *Psychological* - statements concerned with thoughts, emotions or cognitive processes.

Three self-as-subject/ 'I':

- *Agency* - statements concerned with the formation, existence, or control of self.
- *Continuity* - statements concerned with the sense of self-continuity over time.
- *Distinctness* - statements concerned with the sense of distinctness from others.

Each chunk was then rated according to Damon & Hart's (1988) levels of self-development:

- Level 1 - self being understood in terms of simple categorical identifications.
- Level 2 - comparative assessment between the self and others or normative standards.
- Level 3 - understanding focuses on the characteristics of self that determines the nature of one's interactions with others.
- Level 4 - understanding organises self-characteristics in terms of systematic beliefs and life plans.

Although it is possible to score some chunks at more than one level within a single category (e.g. physical self), in line with Damon and Hart (1988), chunks were only scored for the highest applicable level. It was possible to score chunks in two or more categories at the same level (See appendix 2.3.4.2 for detailed scoring criteria). For inter-rater reliability purposes, transcribed chunks were edited to remove identifying information. Thirty randomly chosen chunks were scored by an independent researcher blinded to the study and inter-rater reliability was assessed using the kappa statistic ($\kappa = .807$, $p = .001$) as very good. (See appendix 2.3.4.3 for demonstration of coding process).

Data analysis

The Self-understanding Interview was scored according to the predetermined categories and a data analysis was carried out to explore between-group differences using a Kruskal-Wallis test after assumptions of normality were not met.

RESULTS

Forty-five adolescents participated in the study (27 male, 18 female). There were two participants in the WS group who were Asian British and one participant in the ASC group was Afro-Caribbean British. The rest of the sample were White British. The majority of participants were attending specialist schools/accessing special educational needs provision, 5 participants (1 ASC, 2 DS and 2 WS) were attending a mainstream school. Sample characteristics are summarised in Table 1.

Table 1: Sample Characteristics

Group	N	Gender F:M	WASI	WASI confidence levels	Chronological age years : months (SD)	BPVS verbal comprehension level (SD)	BPVS confidence levels
Whole sample	45	18:27	65	61 – 77	16:2 (2:4)	9:2 (2:11)	8:5 – 9:11
ASC	15	2:13	75	70 – 83	16:10 (1:5)	11:5 (2:8)	10:6 – 12:3
DS	15	9:6	57	53 – 66	14:10 (2:4)	6:9 (1:9)	6:2 – 7:5
WS	15	7:8	64	60 – 73	16:10 (2:9)	9:4 (2:4)	8:7 – 10:1

A Kolmogorov-Smirnov test indicated that some data did not meet the assumptions of normality. In the ASC group, these were total number of ‘active’ chunks ($p = .002$), ‘social’ chunks ($p = .016$), ‘agency’ chunks ($p = .001$), ‘continuity’ chunks ($p = .001$), and ‘distinctness’ chunks ($p = .001$). In the DS group, these were the WASI scores ($p = .004$) and BPVS scores ($p = .03$), ‘agency’ chunks ($p = .001$), ‘continuity’ chunks ($p = .001$), and ‘distinctness’ chunks ($p = .001$). In the WS group, these were the total number of ‘physical’ chunks ($p = .016$), ‘psychological’ chunks ($p = .003$), ‘agency’ chunks ($p = .001$), and ‘distinctness’ chunks ($p = 0.19$). Levene’s test for homogeneity of variances indicated unequal variances in the WASI scores, ($F(2,42) = 11.26, p = .001$). Non-parametric tests were therefore indicated.

A Kruskal-Wallis test revealed a statistically significant difference in WASI scores across the three groups [χ^2 (2, $n= 45$) = 20.28, $p= .001$]. The ASC group recorded the highest median score (Md= 76), followed by WS (Md= 63) and DS group (Md= 55). BPVS age equivalent scores were statistically different across the groups [χ^2 2 (2, $n= 45$) = 19.47, $p= .001$]. The ASC group recorded the highest median score (Md= 12 years 5 months), followed by WS (Md= 8 years 8 months) and DS group (Md= 6 years 1 month).

Post-hoc Mann-Whitney U tests revealed differences between pairs of the groups after a Bonferroni adjustment was applied ($p= .017$). There was a difference between the ASC and DS groups in their WASI scores ($U= 18.5$, $p= .001$) and BPVS scores ($U= 18$, $p= .001$). Between the DS and WS groups, there was a difference in their WASI scores ($U= 37$, $p= .001$) and BPVS scores ($U= 37.5$, $p= .001$). Table 2 provides a summary of between-group differences.

Table 2: Summary of Between-Group Comparison

	ASC WASI	DS WASI	WS WASI	ASC BPVS	DS BPVS	WS BPVS
ASC WASI	-	$U= 18.5$ $p= .001$	$U= 56.5$ $p= .020$	-	-	-
DS WASI	$U= 18.5$ $p= .001$	-	$U= 37$ $p= .001$	-	-	-
WS WASI	$U= 56.5$ $p= .020$	$U= 37$ $p= .001$	-	-	-	-
ASC BPVS	-	-	-	-	$U= 18$ $p= .001$	$U= 62$ $p= .036$
DS BPVS	-	-	-	$U= 18$ $p= .001$	-	$U= 37.5$ $p= .001$
WS BPVS	-	-	-	$U= 62$ $p= .036$	$U= 37.5$ $p= .001$	-

Self-as-Object/ 'Me'

The self-as-object data were examined to explore group differences in (a) total number of self-as-object 'chunks', which is indicative of general self-understanding, (b) number of self-as-object chunks in physical, active, social and psychological domains and (c) level of self-understanding implicit in self-statements.

There was no significant difference in the number of self-as-object statements in the three groups [$\chi^2 (2, n= 45) = 5.56, p= .062$]; ASC (Md= 17.8), DS (Md= 22.2), WS (Md= 29). No significant difference in the number of self-as-object statements was indicative of similar productivity of self-statements in the three groups. The proportion of self-as-object statements was calculated based on the overall number of statements generated in each group, i.e. self-as-object statements in the ASC group / total number of statements in the ASC group. In the ASC group, self-as-object statements accounted for 78.4% of the overall number of statements (200 self-as-object statements / 255 overall statements). Self-as-object statements accounted for 93.2 % (259/378) and 82.5% (278/337) in the DS and WS groups respectively.

There was no significant difference in the number of '*physical*' chunks in the three groups [$\chi^2 (2, n= 45) = 2.42, p= .298$], ASC (Md= 19.67), WS (Md= 22.37), DS (Md= 26.97). There was no significant difference in the number of '*active*' chunks in the three groups [$\chi^2 (2, n= 45) = 3.78, p= .151$], ASC (Md= 17.73), DS (Md= 24.97), WS (Md= 26.30). There was a significant difference in the total number of '*social*' chunks [$\chi^2 (2, n= 45) = 7.56, p= .023$]; ASC (Md= 17.13), DS (Md= 21.83), WS (Md= 30.03). A post-hoc Mann-Whitney U tests revealed a significant differences between ASC and WS group (U= 47, p= .006). There was no significant difference in the number of '*psychological*' chunks in the three groups [$\chi^2 (2, n= 45) = 2.35, p= .307$]; DS (Md= 19.33), ASC (Md= 23.07), WS (Md= 26.60).

The proportion of self-as-object statements in each of the categories was calculated based on the overall number of statements in each group. These findings are presented in Table 3.

Table 3: Proportion of Self-as-Object Statements

Condition	Categories			
	Physical	Active	Social	Psychological
ASD	19.6% (50/255)	25.1% (64/255)	15.7% (40/255)	18% (46/255)
DS	28.1% (78/278)	30.6% (85/278)	22.7% (63/278)	11.9% (33/278)
WS	17.8% (60/337)	26.7% (90/337)	23.1% (78/337)	14.8% (50/337)

No correlation was found between BPVS and WASI scores and total number of self-as-object statements ($r_s = .118$, $p = .440$) and ($r_s = .141$, $p = .357$) respectively. No correlation was found between BPVS and WASI scores and total number of statements across the four categories.

Table 4 provides a summary of the distribution of self-as-object chunks.

Table 4. Summary of Distribution of Self-as-Object Chunks

Level	Categories																Sum
	Physical				Active				Social				Psychological				
	1	2	3	4	1	2	3	4	1	2	3	4	1	2	3	4	
A1	1	1	1	0	3	0	1	0	0	2	1	0	0	0	1	0	11
A2	1	0	0	0	1	0	0	0	1	0	2	0	0	6	3	0	14
A3	2	0	0	0	5	2	0	0	0	2	1	0	0	0	0	0	12
A4	5	0	0	0	3	0	0	0	0	0	1	0	3	1	0	0	13
A5	3	0	0	0	4	0	0	0	1	0	0	0	1	0	0	0	9
A6	0	0	0	0	1	0	0	2	0	0	2	0	0	0	2	1	8
A7	7	0	0	0	4	0	0	0	1	2	1	0	1	5	0	0	21
A8	0	0	0	0	5	0	0	1	0	0	1	1	3	0	1	0	12
A9	6	1	0	0	3	1	0	0	0	0	0	0	0	0	0	0	11
A10	3	2	0	0	8	1	0	0	0	1	0	0	0	1	0	0	16
A11	4	0	0	0	2	0	0	0	2	3	3	0	3	2	0	0	19
A12	1	0	0	0	4	0	1	0	0	0	5	3	1	0	1	0	16
A13	4	0	0	0	4	0	0	0	1	0	0	0	6	0	3	0	18
A14	3	0	1	0	3	1	0	0	0	0	2	0	0	0	1	0	11
A15	4	0	0	0	4	0	0	0	1	0	0	0	0	0	0	0	9
Totals	44	4	2	0	54	5	2	3	7	10	19	4	18	15	12	1	200
Sum	50				64				40				46				
D1	5	0	0	0	7	0	0	0	8	0	0	0	0	0	0	0	15
D2	5	0	1	0	7	1	0	0	4	0	2	0	2	0	0	0	22
D3	5	0	0	0	4	1	0	0	1	0	0	0	1	0	0	0	12
D4	8	0	0	0	0	0	0	0	3	0	0	0	3	0	0	0	14
D5	2	0	0	0	8	0	0	0	3	1	0	0	0	1	0	0	15
D6	5	0	1	3	1	0	0	0	1	2	0	0	0	0	0	0	13
D7	1	0	0	0	4	0	0	0	9	8	0	1	8	1	0	0	32
D8	9	1	0	0	14	0	1	0	1	3	2	0	4	1	0	0	36
D9	10	0	0	0	4	1	0	0	0	0	0	0	0	0	0	0	15
D10	1	0	0	0	6	0	0	0	1	0	0	0	4	1	0	0	13
D11	6	0	0	0	3	0	0	0	2	0	0	0	1	0	0	0	12
D12	6	0	1	0	5	0	0	0	1	0	0	0	3	0	0	0	16
D13	2	0	0	0	4	0	0	0	2	0	0	0	1	1	0	0	10
D14	2	0	0	0	6	0	2	0	4	0	2	0	1	0	0	0	17
D15	3	1	0	0	6	0	0	0	2	0	0	0	0	0	0	0	12
Totals	70	2	3	3	79	3	3	0	42	14	6	1	28	5	0	0	259
Sum	78				85				63				33				
W1	1	0	0	0	3	0	0	0	2	0	0	0	1	1	0	0	8
W2	2	0	0	0	3	0	0	0	0	1	1	0	1	0	2	0	10
W3	5	0	0	0	5	1	0	0	0	0	1	2	2	0	0	0	16
W4	4	1	0	0	3	3	0	2	3	0	1	2	0	1	0	1	21
W5	3	0	1	0	3	1	5	0	0	0	5	0	1	0	4	0	23
W6	4	0	0	0	4	0	0	0	4	3	0	0	0	1	0	0	16
W7	11	0	0	0	5	0	0	0	0	3	6	0	0	1	0	1	27
W8	4	0	0	0	3	2	2	1	1	0	3	1	0	0	1	0	18
W9	3	0	0	0	5	0	0	0	1	1	4	3	1	0	0	0	18
W10	2	0	0	0	12	0	1	0	4	0	3	0	1	1	0	0	24
W11	4	0	0	0	6	0	0	0	2	0	1	0	3	1	0	0	17
W12	4	0	0	0	4	0	0	0	2	0	4	0	2	2	2	0	20
W13	7	0	1	1	6	0	0	1	2	1	1	0	4	0	1	0	25
W14	0	0	0	0	3	0	0	0	2	0	0	0	2	0	0	0	7
W15	2	0	0	0	2	0	0	4	0	5	2	1	1	0	7	4	28
Totals	56	1	2	1	67	7	8	8	23	14	32	9	19	8	17	6	278
Sum	60				90				78				50				

Level of Self-Characteristics

Self-characteristics understood at level 4 (i.e. self-characteristics involving systematic beliefs, personal philosophy and life plans) of the self-as object statements did not differ significantly in the three groups [χ^2 (2, n= 45) = 4.74, p= .093]; DS (Md= 19.80), ASC (Md= 21.63), WS (Md= 27.57). A statistically significant difference was observed for level 1 responses [χ^2 (2, n= 45) = 6.37, p= .041]; ASC (Md= 16.80), WS (Md= 23.40), DS group (Md= 28.80). Also, a significant difference was also found for level 3 responses [χ^2 (2, n= 45) = 9.28, p= .010], DS (Md= 15.17), ASC (Md= 24.93), WS (Md= 28.90). There was a significant difference between ASC and DS group in their level 1 responses (U= 54.5, p= .015) and DS and WS group in their level 3 responses (U= 47, p= .004). A significant negative correlation was found between BPVS scores and total number of statements on level 1 (r_s = -.302, p = .044). A significant positive correlation was found between BPVS scores and total number of statements on levels 2, 3 and 4 (r_s = .337, p = .023); (r_s = .426, p = .004); (r_s = .317, p = .034) respectively. There was a significant positive correlation between WASI scores and the total number of statements on level 2 and level 3 (r_s = .305, p = .042) and (r_s = .408, p = .005) respectively.

In the ASC group, level 4 of self-as-object statements accounted for 3.1% of the overall number of statements (8 level 4 self-as-object statements / 255 overall statements). Level 4 self-as-object statements accounted for 1.4% (4/278) and 7.1% (24/337) in the DS and WS groups respectively.

Self-as-Subject/'I'

Participants were asked the following self- as- subject/'I' questions:

Agency: How did you get to be the way you are?

There was a significant difference in the number of 'agency' chunks in the three groups [χ^2 (2, n= 45)= 8.19, p= .017]; DS (Md= 16.03), ASC (Md= 25.73), WS (Md= 27.23). There was a significant difference between the WS and DS group (U= 56.50, p= .007). The difference between the ASC and DS group (U= 64, p= .018) was not significant after a Bonferroni adjustment was applied. In the ASC group, agency statements accounted for 5.5 % of the overall number of statements (14 agency statements / 255 overall statements). Agency statements accounted for 0.1% (4/278) and 3.6% (12/337) in the DS and WS groups respectively.

Continuity: Do you change at all from year to year? If you do change from year-to-year how do you know it is always still you?

There was a significant difference in the number of 'continuity' chunks in the three groups [χ^2 (2, n= 45) = 15.33, p= .001]; DS (Md= 12.77), ASC (Md= 26.13) and WS (Md= 30.10). There was a significant difference between the ASC and DS group (U= 41, p= .002) and the DS and WS group (U= 30.5, p= .001). A significant difference was observed for level 1 responses [χ^2 (2, n= 45) = 8.86, p= .012]; DS (Md= 15.77), ASC (Md= 24.27), WS (Md= 28.97). There was a significant difference between the DS and WS group (U= 50.00, p= .005). A significant difference was observed for level 2 responses [χ^2 (2, n= 45) = 7.19, p= .027]; DS (Md= 17.50), WS (Md= 25.03), ASC (Md= 26.47). There was a significant difference between ASC and DS group (U= 67.50, p= .007) and DS and WS group (U= 75.00, p= .016).

In the ASC group, continuity statements accounted for 10.2 % of the overall number of statements (26 continuity statements / 255 overall statements). Continuity statements accounted for 2.5% (7/278) and 9.5% (32/337) in the DS and WS groups respectively.

Distinctness: Do you think there is anyone who is exactly like you? What makes you different from anyone you know?

There was no significant difference in the number of 'distinctness' chunks in the three groups [χ^2 (2, n= 45)= 4.21, p= .122]; DS (Md= 17.77), WS (Md= 24.83), ASC (Md= 26.40). A significant difference was observed for level 2 responses [χ^2 (2, n= 45) = 10.09, p= .006]; DS (Md= 16.50), WS (Md= 24.10), ASC (Md= 28.40). There was a significant difference between the ASC and DS group (U= 52.50, p= .001). Table 4 provides a summary of the distribution of self-as-subject chunks. A significant positive correlation was found between BPVS scores and total number of agency, continuity and distinctness statements (r_s = .421, p = .004); (r_s = .551, p = .001); (r_s = .504, p = .001) respectively. A similar correlation was also found between WASI scores and total number of agency, continuity and distinctness statements (r_s = .557, p = .001); (r_s = .454, p = .002); (r_s = .406, p = .006) respectively.

In the ASC group, distinctness statements accounted for 5.6% of the overall number of statements (15 distinctness statements / 255 overall statements). Distinctness statements accounted for 2.9% (8/278) and 4.2% (15/337) in the DS and WS groups respectively.

Table 5 provides a summary of the distribution of self-as-subject chunks.

Table 5. Summary of Distribution of Self-as-Subject Chunks

Level	Agency				Continuity				Distinctness			
	1	2	3	4	1	2	3	4	1	2	3	4
A1	0	0	1	0	0	0	0	0	0	1	0	0
A2	0	0	1	0	1	0	0	0	0	1	0	0
A3	0	0	1	0	1	1	0	0	0	1	0	0
A4	0	0	0	0	1	0	0	0	0	0	0	0
A5	0	0	0	0	1	0	0	0	0	0	0	0
A6	0	0	1	0	0	1	0	1	1	0	0	0
A7	0	0	1	0	1	1	0	0	1	1	0	0
A8	0	0	0	2	0	1	0	1	0	1	0	0
A9	0	0	0	0	1	0	0	0	1	0	0	0
A10	0	0	0	0	3	1	0	0	1	0	0	0
A11	0	0	0	0	0	0	0	1	2	0	0	0
A12	0	0	1	0	2	2	2	0	0	2	0	0
A13	1	0	0	0	2	0	0	0	0	1	0	0
A14	0	0	1	0	1	0	0	0	0	1	0	0
A15	0	0	0	0	0	0	0	0	0	0	0	0
Totals	1	0	7	6	14	7	2	3	6	9	0	0
Sum	14				26				15			
D1	0	0	0	0	0	0	0	0	1	0	0	0
D2	0	0	0	0	0	0	0	0	1	0	0	0
D3	0	0	0	0	0	0	0	0	2	0	0	0
D4	0	0	0	0	0	0	0	0	0	0	0	0
D5	0	0	0	0	4	0	0	0	1	0	0	0
D6	0	0	0	0	0	0	0	0	0	0	0	0
D7	0	0	1	0	2	0	0	0	0	0	0	0
D8	0	0	0	0	1	0	0	0	0	0	0	0
D9	0	0	0	0	0	0	0	0	0	0	0	0
D10	0	0	0	0	0	0	0	0	0	0	0	0
D11	0	0	0	0	0	0	0	0	0	0	0	0
D12	0	0	0	0	0	0	0	0	0	0	0	0
D13	0	0	3	0	0	0	0	0	3	0	0	0
D14	0	0	0	0	0	0	0	0	0	0	0	0
D15	0	0	0	0	0	0	0	0	0	0	0	0
Totals	0	0	4	0	7	0	0	0	8	0	0	0
Sum	4				7				8			
W1	0	0	0	0	1	0	0	0	2	0	0	0
W2	1	0	0	0	1	0	0	0	1	0	0	0
W3	0	0	1	0	0	2	0	1	0	1	0	0
W4	0	0	1	0	1	0	0	0	0	0	0	1
W5	0	0	1	0	3	0	1	0	2	1	0	0
W6	1	0	0	0	3	0	0	0	0	0	0	0
W7	0	0	1	0	2	0	1	0	2	0	0	0
W8	0	1	0	0	1	1	0	0	0	2	0	0
W9	0	1	0	0	0	1	0	0	0	0	0	0
W10	0	0	0	0	2	0	0	0	0	0	0	0
W11	0	0	0	0	1	0	0	0	0	0	0	0
W12	0	0	1	0	2	1	0	0	0	1	0	0
W13	1	2	0	0	2	0	0	0	1	1	0	0
W14	0	0	0	0	0	0	0	0	0	0	0	0
W15	0	0	0	0	1	1	0	3	0	0	0	0
Totals	3	4	5	0	20	6	2	4	8	6	0	1
Sum	12				32				15			

Total number of self-as subject and self-as-object chunks

The ASC group generated 255 chunks, the DS group generated 278 chunks and the WS group generated 337 chunks. There was no significant difference in the total number of all statements between the three groups [χ^2 (2, $n = 45$) = 4.08, $p = .13$]; DS (Md = 19.63), ASC (Md = 20.83), WS (Md = 28.53). The total number of all statements did not correlate with BPVS and WASI scores ($r_s = .228$, $p = .133$); ($r_s = .234$, $p = .122$) respectively.

The effect of age

Further analyses explored the extent to which age determined differences in performance across and within the groups. Initially, the whole sample was split at the median for chronological age (CA) (16 years and 1 month) and Mann-Whitney U analyses were performed to explore the effect of age on performance. Participants with higher CA performed better on the BPVS ($U = 129.5$, $p = .005$) and on the ToM measure ($U = 167.0$; $p = .019$). No significant differences were found on WASI scores, number of physical, active, social, psychological, agency, continuity and distinctness statements, total number of self-as-object and self-as subject statements, the overall number statements and all 4 level of statements.

In the participant group with lower CA across the three groups, there was a significant difference in the BPVS score [χ^2 (2, $n = 23$) = 8.48, $p = .014$]. A post-hoc Mann-Whitney U tests revealed significantly higher scored in the DS group compared to the WS group ($U = 14$, $p = .017$). The difference between the ASC and DS group ($U = 6$, $p = .030$) was not significant after a Bonferroni adjustment was applied. No significant differences were found in ToM scores in the participant group with lower CA across the three groups. These variables were also explored in the participant group with higher CA across the three groups and no significant differences were found.

The effect of age within each of the diagnostic groups separately was explored using the above median split approach. No significant differences were found on WASI and BPVS scores, ToM scores, number of physical, active, social, psychological, agency, continuity and distinctness statements, total number of self-as-object and self-as subject statements, the overall number statements and all 4 level of statements.

Theory of Mind

As hypothesised, there was no significant difference in the ToM scores in the three groups [χ^2 (2, $n = 45$) = 4.74, $p = .093$]. A correlation was found between ToM scores and CA ($r_s = .383$, $p = .009$), BPVS scores ($r_s =$

.473, $p = .001$), WASI scores ($r_s = .314$, $p = 0.36$), total number of agency statements ($r_s = .259$; $p = 0.49$), total number of distinctness statements ($r_s = .398$, $p = .007$) and social level 1 statements ($r_s = -.312$, $p = .037$). Table 6 demonstrates proportion of correct and incorrect answers on the measure.

Table 6: Summary of ToM Scores

	ASC	DS	WS
Correct	13 (13.3%)	8 (53.3%)	8 (53.3%)
Incorrect	2 (86.7%)	7 (46.7%)	7 (46.7%)

DISCUSSION

The present study examined inter-syndrome differences in self-understanding in adolescents with ASC, DS and WS. Forty-five adolescents (15 in each group) between the ages of 12 and 20 participated in the study. The participants completed the BPVS, WASI and the Smarties test, the latter being a measure of ToM. Self-understanding was examined with the use of the Self-understanding Interview exploring verbally articulated concepts, opinions and perceptions. In the following discussion of the study's findings, it is important to note that the DS group had significantly lower WASI scores and BPVS scores compared to the other two groups. The DS group's significantly lower cognitive functioning and receptive vocabulary scores, therefore, need to be taken into consideration when interpreting the results of this study.

The results are discussed, initially, under the headings of the study's hypotheses:-

1. Adolescents with ASC will be least likely to understand themselves in a social way, indicated by the fewest social self-statements, followed by adolescents with DS and then adolescent with WS, who will be most likely to understand themselves in a social way, indicated by the most social self-statements.

The hypothesis was supported as participants with ASC generated significantly fewer social statements compared with the WS group, demonstrating their ability to understand themselves in a social way. No significant differences between groups were found in generating physical, active and psychological statements. No correlation was found between BPVS and WASI scores and total number of statements across the four categories. It is proposed that the findings of adolescents with WS producing most social self-statements reflect

their greater sociability (Jones et al., 2000) and understanding of themselves in the context of relationships with others (Plesa-Skwerer et al., 2004).

It could be argued that lower language productivity in adolescents with ASC could account for these results (Jackson et al., 2011), but categories of response were recorded on the basis of the very first part of the chunk regardless of how brief or elaborate it was and there was no significant difference in the number of chunks produced by participants across the three groups. It could also be suggested that people with ASC have difficulties with abstract concepts such as social self-understanding, yet this is incongruent with the limited difference in other abstract concepts such as psychological and active categories of self-understanding. It is unlikely that the low number of social statements in the ASC group follows typical development with no significant differences observed in physical, social and psychological statements (Lee & Hobson, 1998); and lack of language productivity and difficulties with abstract concepts also do not provide explanation for the differences in social self-statements. These findings are congruent with those of Lee and Hobson (1998) who reported a significantly lower number and quality of social statements in adolescents with ASC compared with non-ASC adolescents with ID. This supports the hypothesis of a specific abnormality in social self-understanding in adolescents with ASC. This may also be linked with the difficulties people with ASC experience in autobiographical memory and autonoetic consciousness, which provide a framework to reflect on the self in the past, present and future (Klein, German, Cosmides, & Gabriel, 2004; Skirrow et al., 2014). It is important to consider other factors that are not condition-specific and are closely linked with self-concept. For example, limited social contact due to other causes may play a role in the development of social self-understanding in people with ASC. The person's environment, life experiences, family and school expectations, experiences and relationships are closely linked with self-concept (Wiest et al., 1998; Zheng et al., 2014) and, therefore, a degree of heterogeneity should be expected and factors that are not condition-specific should be considered.

2. Adolescents with ASC, DS and WS will not differ on the number of self-as-object self-statements.

No significant differences were found in the number of self-as-object self-statements in the three groups and therefore the hypothesis was supported. No correlation was found between BPVS and WASI scores and the total number of self-as-object statements. This is in line with Lee & Hobson's study (1998) where ASC adolescents compared to non-autistic adolescents with ID did not differ on the amount of self-as-object statements. Also, in Plesa-Skwerer et al.'s study (2004) comparing self-concepts in people with WS and Prader-Willi syndrome, no

significant difference in the overall number of self-as-object statements was reported and significant syndrome-group effects were found only for social and psychological categories supporting the hypothesis of comparability in the current cross-syndrome comparison. This was not the case when comparing people with AS to a neurotypical control group, as Jackson et al. (2011) found that the AS group generated significantly fewer self-as-object statements demonstrating a relatively under-developed self-concept compared to adolescents without ID.

Table 2 demonstrated within subject variability in the number of self-as-subject statements. A number of factors may have played a role in the variability across subjects including age, the measured receptive vocabulary, cognitive ability, ToM, as well as a number of other factors such as pragmatic language ability, educational setting, family circumstance and the participant's reference group.

The effect of age was examined within each of the three groups and no significant differences were found between younger and older participants based on the median split approach. Furthermore, no correlations were found between age, BPVS, WASI and ToM scores on the total number of self-as-object statements and total number of statements across the four categories. It may be the case that other variables not measured in the study affected the within subject variability and assessing and controlling for these factors in future research would be beneficial.

3. Adolescents with ASC will produce fewest references to systematic beliefs and life plans in the Self-understanding Interview, followed by adolescents with DS and then adolescents with WS, who will produce the most references to systematic beliefs and life plans in the interview.

The DS group generated the lowest number of level 4 statements (i.e. references to systematic beliefs and life plans) followed by the ASC group and then the WS group, however these differences were not significant. Participants with DS also produced more self-as-subject level 1 statements and fewer level 3 statements. This demonstrated this group's focus on physically based descriptions and a relative lack of self-characteristics of social quality and interpersonal relations. The participants with DS had significantly lower cognitive functioning compared to the ASC and WS groups. Their mean receptive vocabulary level, as measured by the BPVS, was 6 years and 9 months. It can be hypothesized, therefore, that these findings may reflect a general developmental delay and that these adolescents followed the developmental trajectory, described by Damon & Hart (1988). They produced more categorical identifications, such as descriptions of their appearance, material possessions,

typical behaviour, fact of membership in particular social group and momentary moods, feelings and preferences, rather than higher level descriptions relating to implications for social interactions and social appeal. These findings are in line with the correlations found between BPVS scores and total number of statements on all four levels and correlation between WASI scores and the total number of statements on level 2 and level 3.

No significant difference was found in the ASC and WS group. In previous research, Lee and Hobson (1998) found no differences in level 4 statements between ASC adolescents compared to non-autistic adolescents with ID. In Plesa-Skwerer et al.'s study (2004) comparing self-concepts in adolescents and adults with WS and Prader-Willi syndrome matched on cognitive or linguistic levels, participants with WS provided more level 3 and 4 statements. The authors concluded that more 'sophisticated' responses in WS may be attributed to these individuals' sociability and, consequently, a developmentally more advanced self-understanding. The hypothesis that adolescents with WS would produce the most references to systematic beliefs and life plans in the interview based on their suggested advanced social self-understanding was not supported. Current research in the developmentally 'higher level' of self-understanding in the studied conditions is limited and further research is needed.

Agency, continuity and distinctness

Lee and Hobson (1998) found no differences in self-descriptions across agency, continuity and distinctness between ASC adolescents compared to non-autistic adolescents with ID. In the current study, only the DS group had a lower number of self-characteristics in the agency and continuity category, and this again may be explained by the lower cognitive functioning in these participants and potential developmental delay. There were no differences found between the ASC and WS groups. This is congruent with correlations between BPVS and WASI scores and total number of agency, continuity and distinctness statements across the three groups.

Theoretical and Clinical Implications

Deficits in social cognition, impairment in communication (Dalton et al., 2005; Greene et al., 2011; Rosset et al., 2011) and difficulties with seeing oneself as a part of a social context (Lombardo & Baron-Cohen, 2011) in ASC are well described in the literature. The current study supports the evidence for a specific abnormality in social self-functioning in people with ASC linked with restrictions for their social experience and social understanding with implications for the development of social self-understanding (Lee & Hobson, 1998). Lee & Hobson (1998) found that adolescents with ASC made fewer social self-statements compared to non-autistic adolescents with ID. The current study extends the literature and provides a systematic and rigorous cross-syndrome comparison

with DS and WS adolescents that has not been examined before. Evidence for restricted ways of thinking about self in a relational dimension in adolescents with ASC was found and it is proposed that these difficulties may contribute to the relatively poor social adjustment in these young people (Szatmari, Bartolucci, & Bremner, 1989). Difficulties with the development of self-concept have been linked with mental health problems (Harrop & Trower, 2001) and maladaptive behaviours (Jackson et al., 2012; Skirrow et al., 2014) and, therefore, should be focus of clinical interventions.

The significantly higher number of social self-statements in WS compared to ASC, found by this study, extends previous research linking a developed social self-understanding and the adolescents' greater sociability (Jones et al., 2001). Plesa-Skwerer et al. (2004) compared self-concepts in people with WS and Prader-Willi syndrome and found that people with WS produced more social statements. They concluded that, based on a unique component of their phenotype, i.e. their sociability, people with WS have more advanced self-understanding in comparison to other people with ID. However, although people with WS construe their self-understanding in a social context and there are arguments for a more advanced self-understanding, they experience social difficulties that affect their ability to interact with others (Philofsky et al., 2007) and lack skills in sustaining conversations and developing friendships leading to isolation (Davies et al., 1998). High sociability and well-developed social self-understanding in people with WS may be misleading in concluding that people with WS do not require support in promoting social skills and developing and maintaining friendships. With the evidence for increasing anxiety as people with WS reach adulthood, there is a growing risk of inter-personal difficulties and isolation with implications for psychological well-being and mental health.

There is a need for individually tailored interventions to enhance communication and social skills, promoting the formation and maintenance of friendships and, in turn, promoting positive self-concept in adolescents with ID. Various social skills interventions have been used with people with ASC (see Reichow, Steiner, & Volkmar, 2009 for review) delivered by clinicians, teachers, parents and peers. There is evidence for the effectiveness of various methods including applied behavioural analysis (e.g. Jung et al. 2008; Loftin et al. 2008); naturalistic techniques used to provide structure to parent child interactions (e.g., Ingersoll & Gergans, 2007), parent training (Lord & McGee, 2001) and social skills groups (White et al., 2007). As discussed previously, there are significant distinctions in the social and communication profiles in adolescents with ASC, DS and WS, however common difficulties with social understanding and more sophisticated social competences in all three groups were highlighted. The need for enhancing communication and social skills and promoting positive self-understanding is relevant to all young people with ID. Research in the area of social skills interventions and

methods to promote positive self-concept in non-autistic individuals with ID is limited and more research is needed.

In the current study, adolescents with DS had a significantly lower WASI and BPVS scores. Yet, no differences were identified in the number of self-as-subject statements across the groups indicating similarities in the overall development of self-concept. More self-as-subject level 1 statements and fewer level 3 statements in DS adolescents demonstrated a focus on physically based descriptions and a relative lack of self-characteristics of social quality and interpersonal relations. Fewer level 4 statements, representing systematic beliefs and life plans, provide support for the developmental trajectory of self-understanding described by Damon & Hart (1988). These findings highlight the importance of adapting approaches for people ID. Some of the abstract concepts related to level 3 and 4 statements (i.e. reflections on the self's social appeal, social interactions, social relations, or group membership; and reflections on the self's personal philosophy, moral standards, or lifestyle) were not simple topics to articulate. In therapeutic interventions, similar abstract concepts are often addressed and the importance for augmentation of therapy and approaches for people with ID is essential to make therapeutic interventions more accessible and effective (Taylor, Lindsay, & Willner, 2008).

Strengths

The current study extends the literature and provides a systematic and rigorous cross-syndrome comparison of adolescents with ASC, DS and WS that has not been examined before. The merits of comparison studies including children and adolescents (Chapman, 1988; Bear et al., 2002) were questioned due to the significant changes in self-concept through development (Harter, 1999). Therefore, a specific focus was given to the adolescent developmental phase associated with increased cognitive abilities directly impacting self-concept, such as increasing ability to make more realistic comparisons with others (Harter, 1999).

Participants' receptive vocabulary level, cognitive ability and ToM were measured with the use of reliable a normed-referenced measures commonly used in comparison studies with people with ID (e.g. Wilson et al. 2007). The Self-Understanding Interview (Damon & Hart, 1988) has been used in previous research with people with ID (Jackson et al., 2011; Plesa-Skwerer et al., 2004) and the measure has a high level of inter-rater agreement and good internal consistency. An independent researcher blinded to the study scored 30 randomly chosen chunks and inter-rater reliability was assessed as very good. The total number of participants for this study was established *a priori* on the basis of power calculations (30 participants) and in order to maximise

generalizability, the sample size was increased to 45, 15 participants in each group. A wide geographical area was covered in participant recruitment with favourable implications for generalizability.

Limitations

The current study has a number of limitations. As discussed, there are a number of factors such as SES, family and school expectations, experiences and relationships, the adolescents' reference group, time of diagnosis and emotional adjustment all of which are associated with self-concept. These factors were not examined and controlled for in the present study and future research would benefit from assessment of these variables and their effect on self-understanding.

Although the total number of participants for this study was as established *a priori* on the basis of power calculations, the sample size was relatively small with an unequal ratio of males and females in each group. This has implications for the generalizability of the findings. In addition, a specific diagnostic tool was not used to re-diagnose the participants for this study in the ASC group, therefore participants may have not met the same diagnostic criteria, potentially leading to heterogeneity of the group. Participants across the three groups were not matched on cognitive and verbal ability and the DS group's significantly lower cognitive functioning and receptive vocabulary scores have implications for the findings. As the parametric assumptions were not met, the ability to detect a significant difference across the groups was limited due to the use of the less sensitive non-parametric analyses.

Some of the participants in this study chose to complete the research procedure while a parent or a teaching assistant was present, which may have had implications for the content of the adolescents' responses to some questions on the Self-understanding Interview. For example, when a parent left the room one participant with DS began to talk about her wish to move away from the family home and live independently and expressed that this is a difficult topic to raise with her parents.

Interviews were coded according to the predetermined criteria (Damon & Hart, 1988) and further qualitative analysis was not within the scope of this study. Plesa-Skwerer et al. (2004) used the Self-understanding Interview and coded the presence or absence of recurring themes in participants' responses to questions providing opportunities for more detailed discussion reflecting specific future choices, self-evaluations and reflections on development and agency. Such analysis would provide more detail about the nature of self-understanding in the studied groups in line with calls for the use of qualitative methods to assess self-concept

(Pestana, 2015). The use of qualitative methodology would be beneficial in order to elucidate more specific and detailed self-understanding of participants based on real life experiences. For example, a thematic analysis of the interviews may highlight themes such as roles one perceives one-self to have (e.g. role in their family, social circle, employment); hopes and expectations for the future; understanding of one's continuity and how one has changed over time; understanding of one's autonomy; how one compares self to others; understanding of one's condition and attitude towards diagnosis; as well as areas of one's difficulty that can be addressed by parents, clinicians or educational professionals.

As participants represented a relatively wide age range, differences in self-concept in early and late adolescence may have been missed. It is proposed that distinguishing between developmental stages, i.e. early, middle and late adolescence (Harter, 1999) would be beneficial in exploring the developmental trajectory of self-concept in adolescents with ID in more detail. Furthermore, there would be great value in carrying out prospective cohort studies to explore self-concept in people with ID and examine changes in self-evaluation through development and elucidate causal mechanisms and relationships between self-concept, life outcomes and mental health. Such information would help to identify potential risk factors and needs for early interventions to enhance the quality of life of people with ID.

The measure of ToM used in this study is a brief false-belief task, which allowed for low participant demand alongside other measures used in the study. However, in the light of previous research reporting a relatively intact social-perceptual component of ToM in WS, it is possible that if more sophisticated ToM paradigms were used, such as jokes, metaphor, irony, and faux pas (Brüne & Brüne-Cohrs, 2006; Frith & Corcoran, 1996) some differences may have been detected between the groups. Furthermore, the Smarties test relies heavily upon the participant's language ability. Use of another measure, e.g. employing drawings or cartoons (Corcoran & Frith, 2003) would have been beneficial to reduce such potential bias. ToM is a complex construct and a variety of tools used in the literature do not necessarily measure the same aspects of ToM. There is also a lack of normative data and validity and reliability of measures (Bora, Yucel, & Pantelis, 2009). Further research is needed to explore various aspects of ToM in these conditions.

Conclusions

The current research provides a cross-syndrome comparison in ASC, DS and WS across the various categories and levels of development in self-concept. As proposed by Karmiloff-Smith (2010), more subtle patterns of

performance and functioning can be revealed in such comparisons unlike contrasting ‘impaired’ and ‘intact’ functioning (Cebula et al., 2010). The evidence for a specific abnormality in social self-functioning in people with ASC was discussed with implications for self-understanding. The distinctions in social and communication profiles in adolescents with ASC, DS and WS were discussed as well as common difficulties with social understanding and more sophisticated social competences in all three groups. As difficulties with the development of self-concept have been linked with important life outcomes, including mental health problems (Harrop & Trower, 2001) and maladaptive behaviours, (Jackson et al., 2012; Skirrow et al., 2014) this is an area requiring further research and highlighting a need for individualised interventions promoting positive self-understanding.

References:

- Annaz, D., Karmiloff-Smith, A., Johnson, M. H., & Thomas, M. S. C. (2009). A cross-syndrome study of the development of holistic face recognition in children with autism, Down syndrome and Williams syndrome. *Journal of Experimental Child Psychology*, 102, 456-486.
- Baron-Cohen, S. (2000). Theory of mind and autism: A fifteen year review. In S. Baron-Cohen, H. Tager-Flusberg, & D. J. Cohen (Eds.), *Understanding other minds: Perspectives from developmental cognitive neuroscience* (pp. 3-20). Oxford: Oxford University Press.
- Baron-Cohen, S., Jolliffe, T., Mortimore, C., & Robertson, M. (1997). Another advanced test of theory of mind: Evidence from very high functioning adults with autism or Asperger syndrome. *Journal of Child Psychology and Psychiatry*, 38(7), 813-822.
- Baron-Cohen, S., Leslie, A. M., & Frith, U. (1985). Does the autistic child have a "theory of mind"? *Cognition*, 21(1), 37-46.
- Baron-Cohen, S., Wheelwright, S., Hill, J., Raste, Y., & Plumb, I. (2001). The "Reading the Mind in the Eyes" Test Revised Version: A Study with Normal Adults and Adults with Asperger Syndrome or High-functioning Autism. *Journal of Child Psychology and Psychiatry*, 24(2), 241-251.
- Bear, G. G., Minke, K. M., & Manning, M. A. (2002). The self-concepts of students with learning disabilities: A meta-analysis. *School Psychology Review*, 31, 405-427.
- Booth, M., & Gerard, J. (2011). Self-Esteem and Academic Achievement: A Comparative Study of Adolescent Students in England and the United States. *Journal of Comparative and International Education*, 41(5), 629-648.
- Bora, E., Yucel, M., & Pantelis, C. (2009). Theory of mind impairment in schizophrenia: meta-analysis. *Schizophrenia research*, 109(1), 1-9.

- Bos, A. E. R., Huijding, J., Muris, P., Vogel, L. R. R., & Biesheuvel, J. (2010). Global, contingent and implicit self-esteem and psychopathological symptoms in adolescents. *Personality and Individual Differences*, 48(3), 311–316.
- Brock, J., Einav, S., & Riby, D. (2008). The other end of the spectrum? Social cognition in Williams syndrome. In T. Striano & V. Reid (Eds.), *Social Cognition: Development, Neuroscience and Autism*. Oxford: Blackwell.
- Capps, L., Sigman, M., & Yirmiya, N. (1995). Self-competence and emotional understanding in high-functioning children with autism. *Development and Psychopathology*, 7(1), 137–149.
- Chapman, J. W. (1988). Learning disabled children's self-concepts. *Review of Educational Research*, 58, 347–371.
- Cebula, K. R., Moore, D. G., & Wishart, J. G. (2010). Social cognition in children with Down's syndrome: challenges to research and theory building. *Journal of Intellectual Disability Research*, 54(2), 113–134.
- Coudevylle, G. R., Gernigon, C., & Martin Ginis, K. A. (2011). Self-esteem, self-confidence, anxiety and claimed self-handicapping: A mediational analysis. *Psychology of Sport and Exercise*, 12(6), 670–675.
- Courtney, E., Gamboz, J., & Johnson, J. (2008). Problematic eating behaviors in adolescents with low self-esteem and elevated depressive symptoms. *Eating Behaviors*, 9(4), 408–414.
- Dalgas-Pelish, P. (2006). Effects of a self-esteem intervention program on school age children. *Paediatric Nursing*, 32, 341–348.
- Dalton, K. M., Nacewicz, B. M., Johnstone, T., Schaefer, H. S., Gernsbacher, M. A., Goldsmith, H. H., & Alexander, A. L., (2005). Gaze fixation and the neural circuitry of face processing in autism. *Nature Neuroscience*, 8, 519–526.
- Damon, W., & Hart, D. (1988). *Self-understanding in Childhood and Adolescence*. Cambridge: Cambridge University Press.
- Davies, M., Udwin, O., & Howlin, P. (1998). Adults with Williams syndrome. Preliminary study of social, emotional and behavioural difficulties. *The British Journal of Psychiatry*, 172(3), 273–276.

- Donnellan, M. B., Trzesniewski, K. H., Robins, R. W., Moffitt, T. E., & Caspi, A. (2005). Low self-esteem is related to aggression, antisocial behavior, and delinquency. *Psychological science*, 16(4), 328-335.
- Diehl, M., & Hay, E. L. (2010). Risk and resilience factors in coping with daily stress in adulthood: The role of age, self-concept incoherence, and personal control. *Developmental Psychology*, 46, 1132-1146.
- Dodd, H. F., Porter, M., Peters, G. L., & Rapee, R. M. (2010). Social approach in pre-school children with Williams syndrome: the role of the face. *Journal of intellectual disability research*, 54, 194–203.
- Dunn, L. M., Dunn, D. M., Styles, B., & Sewell, J. (2009). *The British Picture Vocabulary Scale III – 3rd Edition*. London: GL Assessment.
- Dye, L., Hare, D. J., & Hendy, S. (2005). Capacity of people with learning disabilities to consent to take part in a research study. *Autism*, 9(5), 515–31.
- Elmer, N. (2001). *Self-esteem. The costs and causes of low self-worth*. Yorn: Joseph Rowntree Foundation.
- Fathi-Ashtiani, A., Ejei, J., Khodapanahi, M., & Tarkhorani, H. (2007). Relationship Between Self-Concept, Self-esteem, Anxiety, Depression and Academic Achievement in Adolescents. *Journal of Applied Sciences*, 7(7), 995.
- Fidler, D. J., Most, D. E., Booth-LaForce, C., & Kelly, J. F. (2008). Emerging social strengths in young children with Down syndrome. *Infants & Young Children*, 21(3), 207-220.
- Finlay, W. M. L., & Lyons, E. (2000). Social categorizations, social comparisons and stigma: Presentations of self in people with learning difficulties. *British Journal of Social Psychology*, 39, 129-146.
- Franco, F. & Wishart, J. G. (1995). Use of pointing and other gestures by young-children with Down syndrome. *American Journal on Mental Retardation*, 100, 160-182.
- Frith, U. (1989). Autism and "Theory of Mind". In C. Gillberg (Ed.), *Diagnosis and Treatment of Autism*. (pp. 33-52). New York: Plenum Press.
- Frith, U., & Happé, F. (1994). Autism: beyond "theory of mind". *Cognition*, 50; 115-132.

- Gagliardi, C., Frigerio, E., Burt, D. M., Cazzaniga, I., Perrett, D. I., & Borgatti, R. (2003). Facial expression recognition in Williams syndrome. *Neuropsychologia*, 41(6), 733–738.
- Giaouria, S., Alevriadou, A., Tsakiridou, E. (2010). Theory of mind abilities in children with Down syndrome and non-specific intellectual disabilities: An empirical study with some educational implications. *Procedia - Social and Behavioral Sciences*, 2(2), 3883–3887.
- Gillberg, C., & Rasmussen, P. (1994). Brief report: four case histories and a literature review of Williams syndrome and autistic behavior. *Journal of Autism and Developmental Disorders*, 24(3), 381-393.
- Glick, M., Bybee, J., & Zigler, E. (1997). Self-Image a Cognitive- Developmental Approach. In R. Leahy (Ed.), *The development of self*. New York: Academic Press.
- Greene, D. J., Colich, N., Iaconi, M., Zaidel, E., Bookheimer, S. Y., & Dapretto, M. (2011). Atypical neural networks for social orienting in autism spectrum disorders. *NeuroImage*, 56(1), 354–62.
- Harrop, C., & Trower, P. (2001). Why does schizophrenia develop at late adolescence? *Clinical Psychology Review*, 21(2), 241–65.
- Harter S. (1985). *Manual for the Self-Perception Profile for Children*. Denver, CO: University of Denver.
- Harter, S. (1999). *The construction of the self: A developmental perspective*. New York: Guilford Press.
- Herguner, S., & Motavalli Mukaddes, N. (2006). Autism and Williams syndrome: a case report. *The World Journal of Biological Psychiatry*, 7(3), 186-188.
- Hobson, R. P. (1993). *Autism and the development of mind*. Hove, UK: Erlbaum.
- Ingersoll, B., & Gergans, S. (2007). The effect of a parent-implemented imitation intervention on spontaneous imitation skills in young children with autism. *Research in Developmental Disabilities*, 28(2), 163-175.
- Jackson, P., Skirrow, P., & Hare, D. J. (2011). Asperger through the looking glass: an exploratory study of self-understanding in people with Asperger's syndrome. *Journal of Autism and Developmental Disorders*, 42(5), 697–706.
- James, W. (1892/1961). *Psychology: The Briefer Course* (p. 44). New York: Harper.

- Järvinen, A., Korenberg, J. R., Bellugi, U. (2013). The social phenotype of Williams syndrome. *Current Opinion in Neurobiology*, 23, 414–422.
- Järvinen-Pasley, A., Vines, B. W., Hill, K. J., Yam, A., Grichanik, M., Mills, D., Reiss, A., Korenberg, J., & Bellugi, U. (2010). Cross-modal influences of affect across social and non-social domains in individuals with Williams syndrome. *Neuropsychologia*, 48(2), 456-466.
- Jones, W., Bellugi, U., Zona, L., Chiles, M., Reilley, J., Lincoln, A., & Al., E. (2000). Hypersociability in Williams syndrome. *Journal of Cognitive Neuroscience*, 12, 30–46.
- Jung, S., Sainato, D. M., & Davis, C. A. (2008). Using high-probability request sequences to increase social interactions in young children with autism. *Journal of Early Intervention*, 30(3), 163-187.
- Karmiloff-Smith, A. (2010). *The importance of cross-syndrome comparisons for understanding autism: a developmental approach*. Birkbeck Centre for Brain & Cognitive Development University of London, Zaragoza talk, November 2010.
- Kasari, C., Freeman, S. F., & Bass, W. (2003). Empathy and response to distress in children with Down syndrome. *Journal of Child Psychology and Psychiatry*, 44(3), 424-431.
- Klein, S. B., German, T. P., Cosmides, L., & Gabriel, R. (2004). A theory of autobiographical memory: Necessary components and disorders resulting from their loss. *Social Cognition*, 22(5), 460–490.
- Knott, F., Dunlop, A-W., & Mackay, T. (2006). Living with ASD: How do children and their parents assess their difficulties with social interaction and understanding? *Autism International Journal*, 10(6), 603-611.
- Laws, G., & Bishop, D. (2003). Pragmatic language impairment and social deficits in Williams syndrome: a comparison with Down's syndrome and specific language impairment. *International Journal of Language Disorders*, 39:1, 45-64.
- Lee, A., & Hobson, R. P. (1998). On developing self-concepts: a controlled study of children and adolescents with autism. *Journal of Child Psychology and Psychiatry, and Allied Disciplines*, 39(8), 1131–44.
- Levitt, M. Z., & Hart, D. (1991). Development of self-understanding in anorectic and nonanorectic adolescent girls. *Journal of Applied Developmental Psychology*, 12(3), 269–288.

- Leyfer, O. T., Woodruff-Borden, J., Klein-Tasman, B. P., Fricke, J. S., & Mervis, C. B. (2006). Prevalence of psychiatric disorders in 4 to 16-year-olds with Williams syndrome. *American Journal of Medical Genetics Part B: Neuropsychiatric Genetics*, 141(6), 615-622.
- Loftin, R. L., Odom, S. L., & Lantz, J. F. (2008). Social interaction and repetitive motor behaviors. *Journal of Autism and Developmental Disorders*, 38(6), 1124-1135.
- Lombardo, M. V., & Baron-Cohen, S. (2011). The role of the self in mindblindness in autism. *Consciousness and Cognition*, 20(1), 130-140.
- Lord, C., Bristol-Power, M., Cafiero, J. M., Filipek, P. A., Gallagher, J. J., Harris, S. L., et al. (2001). *Educating children with autism*. Washington, DC: National Academy Press.
- Lord, C., & McGee, J. P. (2001). *Educating children with autism*. Washington, DC: National Academy Press.
- Losh, M., Klusek, J., Martin, G.E., Sideris, J., Parlier, M., & Piven, J. (2012). Defining genetically meaningful language and personality traits in relatives of individuals with fragile X syndrome and autism. *American Journal of Medical Genetics: Neuropsychiatric Genetics*, 159B, 660-668.
- Malti, T. (2006). Aggression, Self-Understanding, and Social Competence in Swiss Elementary-School Children. *Swiss Journal of Psychology*, 65(2), 81-91.
- Marcia, J. E. (1980). Identity in adolescence. In J. Adelson (Ed.), *Handbook of adolescent psychology* (pp. 159-187). New York: Wiley.
- Mesibov, G. B., & Shea, V. (1996). Full Inclusion of Students with Autism. *Journal of Autism & Developmental Disorders*, 26(3), 337-46.
- Meyer-Lindenberg, A., Mervis, C. B., & Berman, K. F. (2006). Neural mechanisms in Williams syndrome: a unique window to genetic influences on cognition and behaviour. *Nature reviews. Neuroscience*, 7(5), 380-93.
- Mohammad, A. (2010). Relationship Between Self-esteem and Academic Achievement Amongst Pre-University Students. *Journal of Applied Sciences*, 10(20), 2474.
- Paradise, A. W., & Kernis, M. H. (2002). Self-esteem and Psychological Well-being: Implications of Fragile Self-esteem. *Journal of Social and Clinical Psychology*, 21(4), 345-361.

- Perner, J., Frith, U., Leslie, A. M., & Leekam, S. R. (1989). Exploration of the autistic child's theory of mind: knowledge, belief, and communication. *Child Development*, 60, 688–700.
- Pestana, C. (2015). Exploring the self-concept of adults with mild learning disabilities. *British Journal of Learning Disabilities*, 43(1), 16-23.
- Philofsky, A., Fidler, D. J., & Hepburn, S. (2007). Pragmatic language profiles of school-age children with autism spectrum disorders and Williams syndrome. *American Journal of Speech-Language Pathology*, 16(4), 368-380.
- Piers E. V. (1994). *Revised Manual for the Piers-Harris Children's Self-Concept Scale*. Western Psychological Services, Los Angeles, CA.
- Plesa-Skwerer, D., Sullivan, K., Joffe, K., & Tager-Flusberg, H. (2004). Self-concept in people with Williams syndrome and Prader-Willi syndrome. *Research in Developmental Disabilities*, 25(2), 119–38.
- Porter, M. A., Coltheart, M., & Langdon, R. (2008). Theory of mind in Williams syndrome assessed using a nonverbal task. *Journal of Autism and Developmental Disorders*, 38(5):806-14.
- Premack, D. G., & Woodruff, G. (1978). Does the chimpanzee have a theory of mind? *Behavioral and Brain Sciences*, 1(4), 515–526.
- Punshon, C., Skirrow, P., & Murhpy, G. (2009). The not guilty verdict: psychological reactions to a diagnosis of Asperger syndrome in adulthood. *Autism*, 13(3), 265–83.
- Rahmani, P. (2011). The relationship between self-esteem, achievement goals and academic achievement among the primary school students. *Social and Behavioral Sciences*, 29, 803–808.
- Reichow, B., Steiner, A. M., & Volkmar, F. (2013). Cochrane review: social skills groups for people aged 6 to 21 with autism spectrum disorders (ASD). *Evidence- Based Child Health: A Cochrane Review Journal*, 8(2), 266-315.
- Riby, D. M., Hanley, M., Kirk, H., Clark, F., Little, K., Fleck, R., ... & Rodgers, J. (2014). The interplay between anxiety and social functioning in Williams syndrome. *Journal of Autism and Developmental Disorders*, 44(5), 1220-1229.

- Rosner, B. A., Hodapp, R. M., Fidler, D. J., Sagun, J. N., & Dykens, E. M. (2004). Social competence in persons with Prader- Willi, Williams and Down's syndromes. *Journal of Applied Research in Intellectual Disabilities*, 17(3), 209-217.
- Rosset, D., Santos, A., Da Fonseca, D., Rondan, C., Poinso, F., & Deruelle, C. (2011). More than just another face in the crowd: Evidence for an angry superiority effect in children with and without autism. *Research in Autism Spectrum Disorders*, 5(2), 949–956.
- Santos, J. C., Saraiva, C. B., & De Sousa, L. (2009). The role of expressed emotion, self-concept, coping, and depression in parasuicidal behavior: A follow-up study. *Archives of Suicide Research*, 13(4), 358-367.
- Singer Harris, N. G., Bellugi, U., Bates, E., Jones, W., & Rossen, M.L. (1997). Contrasting profiles of language development in children with Williams and Down syndromes. *Developmental Neuropsychology*, 13, 345–370.
- Skirrow, P., Jackson, P., Perry, E., & Hare, D. J. (2014). I collect therefore I am—autonoetic consciousness and hoarding in asperger syndrome. *Clinical Psychology & Psychotherapy*. 22, 3: 278–284.
- Stinton, C., Tomlinson, K., & Estes, Z. (2012). Examining reports of mental health in adults with Williams syndrome. *Research in Developmental Disabilities*, 33(1), 144-152.
- Szatmari, P., Bartolucci, G., & Bremner, R.. (1989). Asperger's syndrome and autism. *Developmental Medicine and Child Neurology*, 31, 709-720.
- Tager-Flusberg, H., & Sullivan, K. (2000). A componential view of theory of mind: evidence from Williams syndrome. *Cognition*, 76(1), 59–90.
- Tager-Flusberg, H., Skwerer, D. P., & Joseph, R. M. (2006). Model syndromes for investigating social cognitive and affective neuroscience: a comparison of Autism and Williams syndrome. *Social Cognitive and Affective Neuroscience*, 1(3), 175–82.
- Taylor, J. L., Lindsay, W. R., & Willner, P. (2008). CBT for people with intellectual disabilities: emerging evidence, cognitive ability and IQ effects. *Behavioural and Cognitive Psychotherapy*, 36(06), 723-733.

- Thomaes, S., Reijntjes, A., Orobio De Castro, B., Bushman, B. J., Poorthuis, A., & Telch, M. J. (2010). I like me if you like me: on the interpersonal modulation and regulation of preadolescents' state self-esteem. *Child Development, 81*(3), 811–825.
- Udwin, O. (1990). A survey of adults with Williams syndrome and idiopathic infantile hypercalcaemia. *Developmental Medicine and Child Neurology, 32*(2):129–141.
- Wechsler, D. (1997). *Wechsler Adult Intelligence Scale—3rd Edition*. San Antonio, TX: Harcourt Assessment.
- Wechsler, D. (1999). *Wechsler Abbreviated Scale of Intelligence (WASI)*. San Antonio, TX: Harcourt Assessment.
- Wiest, D. J., Wong, E. H., & Kreil, D. A. (1998). Predictors of global self-worth and academic performance among regular education, learning disabled, and continuation high school students. *Adolescence, 33*(131), 601.
- White, S. W., Keonig, K., & Scahill, L. (2007). Social skills development in children with autism spectrum disorders: A review of the intervention research. *Journal of Autism and Developmental Disorders, 37*(10), 1858-1868.
- Wilson, R., Pascalis, O., & Blades, M. (2007). Familiar face recognition in children with autism: The differential use of inner and outer face parts. *Journal of Autism and Developmental Disorders, 37*(2), 314-320.
- Wishart, J. G. (2007). Socio- cognitive understanding: a strength or weakness in Down's syndrome? *Journal of Intellectual Disability Research, 51*(12), 996-1005.
- Yirmiya, N., Erel, O., Shaked, M., & Solomonica-Levi, D. (1998). Meta-analyses comparing theory of mind abilities of individuals with autism, individuals with mental retardation, and normally developing individuals. *Psychological Bulletin, 124*(3), 283.
- Yoshii, H., & Yoshimatsu, Y. (2003). Self-Understanding, Understanding of Others, and Affective Understanding in Adolescents with Autism. *Japanese Journal of Special Education, 41*, 217–226.

Zheng, C., Erickson, A. G., Kingston, N. M., & Noonan, P. M. (2014). The relationship among self-determination, self-concept, and academic achievement for students with learning disabilities. *Journal of Learning Disabilities*, 47(5), 462-474.

APPENDICES

1. AUTHOR GUIDELINES

Journal of Autism and Developmental Disorders

The preferred article length is 20-23 double-spaced manuscript pages long (not including title page, abstract, tables, figures, addendums, etc.) Manuscripts of 40 double-spaced pages (references, tables and figures counted as pages) have been published. The reviewers or the editor for your review will advise you if a longer submission must be shortened.

Text must be double-spaced; APA Publication Manual standards must be followed.

ABSTRACT

Please provide an abstract of 120 words or less. The abstract should not contain any undefined abbreviations or unspecified references.

KEYWORDS

Please provide 4 to 6 keywords which can be used for indexing purposes.

TEXT

Text Formatting

Manuscripts should be submitted in Word.

Use a normal, plain font (e.g., 10-point Times Roman) for text.

Use italics for emphasis.

Use the automatic page numbering function to number the pages.

Do not use field functions.

Use tab stops or other commands for indents, not the space bar.

Use the table function, not spreadsheets, to make tables.

Use the equation editor or MathType for equations.

Save your file in docx format (Word 2007 or higher) or doc format (older Word versions).

Headings

Please use no more than three levels of displayed headings.

Abbreviations

Abbreviations should be defined at first mention and used consistently thereafter.

Footnotes

Footnotes can be used to give additional information, which may include the citation of a reference included in the reference list. They should not consist solely of a reference citation, and they should never include the bibliographic details of a reference. They should also not contain any figures or tables.

Footnotes to the text are numbered consecutively; those to tables should be indicated by superscript lower-case letters (or asterisks for significance values and other statistical data). Footnotes to the title or the authors of the article are not given reference symbols.

Always use footnotes instead of endnotes.

Acknowledgments

Acknowledgments of people, grants, funds, etc. should be placed in a separate section on the title page. The names of funding organizations should be written in full.

Proposed journal for Chapter 1 & 2.

BODY

The body of the manuscript should begin on a separate page. The manuscript page header (if used) and page number should appear in the upper right corner. Type the title of the paper centered at the top of the page, add a hard return, and then begin the text using the format noted above. The body should contain:

Introduction (The introduction has no label.)

Methods (Center the heading. Use un-centered subheadings such as: Participants, Materials, Procedure.)

Results (Center the heading.)

Discussion (Center the heading.)

HEADINGS

Please use no more than three levels of displayed headings.

Level 1: Centered

Level 2: Centered Italicized

Level 3: Flush left, Italicized

FOOTNOTES

Center the label "Footnotes" at the top of a separate page. Footnotes can be used to give additional information, which may include the citation of a reference included in the reference list. They should not consist solely of a reference citation, and they should never include the bibliographic details of a reference. They should also not contain any figures or tables.

Footnotes to the text are numbered consecutively; those to tables should be indicated by superscript lower-case letters (or asterisks for significance values and other statistical data). Footnotes to the title or the authors of the article are not given reference symbols.

Always use footnotes instead of endnotes. Type all content footnotes and copyright permission footnotes together, double-spaced, and numbered consecutively in the order they appear in the article. Indent the first line of each footnote 5-7 spaces. The number of the footnote should correspond to the number in the text. Superscript arabic numerals are used to indicate the text material being footnoted.

2. METHOD

2.1 DClinPsy Research Review Committee Approval

14 July 2014 12:53

Dear Dr Golding and Mrs Tucker

I am pleased to inform you that the Sub-Committee has approved your application for ethical approval for your study. Details and conditions of the approval can be found below.

Ref:	RETH000705
Sub-Committee:	Physical Interventions
Review type:	Full committee review
PI:	Dr Laura Golding
School:	Division of Clinical Psychology
Title:	Self-understanding in adolescents with autism spectrum conditions, Down syndrome and Williams syndrome.
First Reviewer:	Dr Ana Alfirevic
Date of initial review:	27/02/14
Date of Approval:	14/07/14

The application was APPROVED subject to the following conditions:

Conditions

All serious adverse events must be reported to the Sub-Committee within 24 hours of their occurrence, via the Research Integrity and Governance Officer (ethics@liv.ac.uk).

This approval applies for the duration of the research. If it is proposed to extend the duration of the study as specified in the application form, the Sub-Committee should be notified. If it is proposed to make an amendment to the research, you should notify the Sub-Committee by following the Notice of Amendment procedure outlined at <http://www.liv.ac.uk/media/livacuk/researchethics/notice%20of%20amendment.doc>. If the named PI / Supervisor leaves the employment of the University during the course of this approval, the approval will lapse. Therefore please contact the Research Integrity and Governance Officer at ethics@liverpool.ac.uk in order to notify them of a change in PI / Supervisor.

Kind regards

Matthew

Matthew Billington
Research Integrity and Governance Officer

Research Support Office
University of Liverpool
Waterhouse Building (2nd Floor, Block C)
3 Brownlow Street
Liverpool
L69 3GL

Email: ethics@liverpool.ac.uk
Telephone: 0151 794 8290
Website: [Research Support Office](#)

2.2 Information Pack for Parent/Carer Including the Following:

- **Parental/ Carer Letter**
- **Information Sheet for Parent/Carer**
- **Participant Information Sheet for the Young Person**
- **Consent Form**



Helena Tucker
Doctorate of Clinical Psychology
University of Liverpool
Institute of Psychology, Health and Society
Whelan Building, Brownlow Hill
Liverpool
L69 3GB
Telephone: 0151 794 5530
Email: Helena.tucker@liv.ac.uk

Date

Dear Parent/ Carer,

I am writing to you about an opportunity for adolescents with autism, adolescents with Down's syndrome and adolescents with Williams syndrome to be involved in doctoral research on self-understanding. Knowledge in this area is limited and it is proposed that further research will be beneficial for the young people themselves, their parents, carers, teachers and other professionals, e.g. improving the provision of focused and individually tailored methods of education and psychological therapy. This research has been approved by the University of Liverpool's Research Ethics Committee.

Taking part in this will involve the young person spending approximately 40-60 minutes with a researcher completing an interview which includes questions relating to their self-understanding, for example: "What are you especially proud of about yourself?", "What do you like most about yourself?"; and some simple tasks, for example matching spoken words to pictures to assesses verbal comprehension. This can take place in your home or if you prefer at the young person's school.

In appreciation for their time participants will be given a £5 store voucher.

We would like to invite your child to participate in this project. Please find further information enclosed in this letter and do not hesitate to contact me with any queries.

To participate, please contact me on my email address or the telephone number above. Alternatively, please complete the attached consent form and return it in the prepaid envelope and I will in turn contact you.

Thank you for taking the time to read the attached information and considering taking part.

Kind regards,

Helena Tucker



INFORMATION SHEET FOR PARENT/CARER

Title of Project: Self-understanding in adolescents with autism spectrum conditions, Down syndrome and Williams syndrome.

Name of researcher: Helena Tucker

Name of project supervisors: Dr Laura Golding; Dr Dougal Julian Hare

You are being invited to take part in a research study. Before you decide it is important that you understand why the research is being done and what it involves. Please take time to read the following information carefully and feel free to ask us if you would like more information or if there is anything that you do not understand. Please also feel free to discuss this with your friends, relatives and your child's school if you wish.

Thank you for reading this.

What is the purpose of the study?

This research aims to explore self-understanding in adolescents with autism spectrum conditions (ASC), adolescents with Down syndrome (DS) and adolescents with Williams syndrome (WS).

Self-understanding is of central importance in a young person's psychological functioning and well being as well as an important factor in a one's development. The knowledge in this area, especially with adolescents with the above conditions, is very incomplete and it is hoped further research and better understanding of the issue will be beneficial for the young people themselves, their parents, carers, teachers and other professionals.

Why has my child been chosen to take part?

We have chosen to invite your child to participate on the basis of his/her condition. We were able to contact you with the help of the Williams Syndrome Foundation/ Down's Syndrome Association/ MENCAP. The aim is that 45 young people will take part in this study.

What will happen if my child takes part?

Your child's participation is voluntary. It is up to you and them to decide whether or not to take part and you are free to withdraw at anytime without explanation and without incurring any disadvantages.

If you agree for your child to participate in this research the process will be as follows:

We would ask you to discuss this with your child first. If he/she would like to take part, the researcher (Helena Tucker) will contact you to arrange an appointment. This can take place at your home or the school/educational facility the child is attending. You may be present if you wish.

When the researcher meets with your child, he/she will be informed about the study and asked if he/she wants to take part. He/she will be made aware that taking part is voluntary and that he/she can withdraw at any point. If he/she wishes to participate, he/she will complete the following measures:

- The British Picture Vocabulary Scale-II – a simple task where words are matched to pictures.
- Wechsler Abbreviated Scale of Intelligence – two simple tasks assessing vocabulary and non verbal problem solving.
- The Smarties test – a brief task assessing understanding of other people’s knowledge and belief.
- The Self-Understanding Interview – this interview will include questions relating to the young person’s understanding of him/herself e.g. “What are you especially proud of about yourself? What do you like most about yourself?”

We will ask your and your child’s consent to audio record this interview so that it can be transcribed. The audio recording will be stored securely at the university and destroyed after transcription.

All of the above will take around 40-60 minutes to complete in total. This can be done in one or multiple sessions depending on your and your child’s preference.

In appreciation for the time your child has given, he/she will be given a £5 store voucher.

Are there any risks in taking part?

There are no perceived disadvantages or risks involved. If the participant experiences any discomfort or disadvantage as part of the research that this should be made known to the researcher immediately.

CRB Disclosure

The researcher involved has obtained a Criminal Records Bureau CRB Disclosure. If you wish you may request evidence of this.

What if I am unhappy or if there is a problem?

If you are unhappy, or if there is a problem, please feel free to let us know by contacting Helena Tucker and we will try to help. If you remain unhappy or have a complaint which you feel you cannot come to us with then you should contact the Research Governance Officer at ethics@liv.ac.uk. When contacting the Research Governance Officer, please provide details of the name or description of the study (so that it can be identified), the researcher involved, and the details of the complaint you wish to make.

Will my participation be kept confidential?

All data will be treated with full confidentiality. The materials will be kept safe and destroyed when the results have been analysed. Results from this study will be included a Doctoral thesis and published in a peer review journal. Participants will not be identifiable from the results.

The results will be made available to the participants via post or email at the end of the research project. When the results are published, details of how and where they will be accessible will be provided.

What will happen if I want to stop taking part?

You can withdraw at anytime, without explanation. Results up to the period of withdrawal may be used, if you are happy for this to be done. Otherwise you may request that they are destroyed and no further use is made of them.

Who can I contact if I have further questions?

You can contact Helena Tucker. Address: Doctorate of Clinical Psychology; University of Liverpool; Institute of Psychology, Health and Society; Whelan Building; Brownlow Hill; Liverpool; L69 3GB; Telephone: 0151 794 5530; Email: Helena.tucker@liv.ac.uk



PARTICIPANT INFORMATION SHEET FOR THE YOUNG PERSON

Title of Project: Self-understanding in adolescents with autism, Down syndrome and Williams syndrome.

Name of researcher: Helena Tucker

Name of project supervisors: Dr Laura Golding; Dr Dougal Julian Hare

You are invited to take part in a research study. Before you decide, it is important that you understand what it is about, why the research is being done and what it involves. You can contact me to ask any questions if you would like more information or if there is anything that you do not understand. Please feel free to talk about it to your parents/carers, friends and teachers in your school.

What is the purpose of the study? / Why is it being done?

This research will look at how young people with autism, young people with Down syndrome and young people with Williams syndrome understand themselves.

The way young people understand themselves is very important and plays a big part in how they feel and think about themselves as they get older.

We do not know very much about this area and hope that understanding this better will help young people with these conditions, their parents, carers, teachers and others.

What will happen if you take part?

Taking part in the study is voluntary, which means it is up to you if you want to take part. If you decide to take part and then change your mind, that is OK. If we have started and you do not like it or do not want to carry on you can stop at anytime without explaining why.

If you agree to take part, then I will ask you to complete some tasks with me such as looking at and naming pictures and doing puzzles. I will also ask you some questions about yourself, for example about the things you are good at. It is OK if you don't know answers to any questions or if you do not want to answer at all. If you do not enjoy it and want to finish, you can just tell me. We can stop or take breaks when you want to.

If it is OK with you, when I am asking you these questions, I will record your voice so I can later write down what you said. I will keep the recording safe at the university and delete it after I write it down.

All together, this will take around 40-60 minutes. This can be done in one or more sessions depending on what you want.

To thank you for taking part you will be given a £5 store voucher.

What if you are unhappy or if there is a problem?

If you are unhappy, or if there is a problem, please tell me and I will try my best to help. You can also talk to your parent/carer or your teacher. They can then contact me and I will try to help.

Will information about you be kept confidential?

The information about you will be kept safe and confidential, that means that other people will not be able to see information about you. Results/what we found out in the study will be published in a journal. People reading these results will not be able to recognise information about you.

If you want to take part please talk to your parent or carer who can then contact me on this address: Helena.tucker@liv.ac.uk or on this number 0151 794 5530.



PARTICIPANT CONSENT FORM

Title of Research Project: Self-understanding in adolescents with autism spectrum conditions, Down syndrome and Williams syndrome.
Researcher(s): Helena Tucker

Project supervisors: Dr Laura Golding; Dr Dougal Julian Hare

PLEASE MAKE SURE TO COMPLETE YOUR DETAILS SO WE CAN CONTACT YOU

Parent/carer's name:

The young person's name:

Diagnosis (e.g. autism, DS, WS):

Age:

D.O.B:

Contact details (email or telephone number, home address)

**Please
initial box**

1. I confirm that I have read and have understood the information sheet for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.
2. I understand that my child's participation is voluntary and that he/she is free to withdraw at any time without giving any reason, without his/her or my rights being affected. In addition, should he/she not wish to answer any particular question or questions, he/she is free to decline.
3. I understand and agree that the 30-minute interview with my child will be audio recorded and I am aware of and consent to your use of these recordings for the purposes of transcription. The recording will then be deleted.
4. I understand that, under the Data Protection Act, my child and myself can at any time ask for access to the information my child provides and can also request the destruction of that information if I and my child wishes.
5. I agree for my child to take part in the above study.

Parent, Carer's name

Date

Signature

Researcher

Date

Signature

2.3 Measures

2.3.1 The British Picture Vocabulary Scale-II (BPVS-II) (Dunn, Dunn, Styles, & Sewell, 2009)

BPVS – II is a widely used, normed-referenced measure with evidence of good validity and reliability (Dunn, Whetton, & Burley, 1997). It assesses receptive vocabulary by presenting participant with four black and white illustrations. The participant is asked to select a picture that illustrates the word spoken by the researcher. The BPVS-II contains 168 items in 14 sets and only items that correspond with the participants' range are administered and a discontinuation rule is applied. Items presented cover a range of content e.g. animals, toys, emotions, actions, parts of speech such as nouns, verbs and attributes across all levels of difficulty. The BPVS is commonly used in comparison studies with people with ID (Annaz, Karmiloff-Smith, Johnson & Thomas, 2009; Wilson, Pascalis & Blades, 2007).

2.3.2. Wechsler Abbreviated Scale of Intelligence (WASI) (Wechsler, 1999)

WASI is a reliable, nationally standardized measure of cognitive functioning normed against the Wechsler Adult Intelligence Scale–Third Edition (WAIS-III) (Wechsler, 1997). The WASI has a four-subtest and a two-subtest versions. Estimated full IQ scores on both of these versions correlate strongly with WAIS-III IQ scores (Strauss, Sherman & Spreen, 2006). The two-subtest format was used to provide a brief measurement of participants' general level of cognitive ability. The two-subtest format comprises of Vocabulary and Matrix Reasoning subtests.

2.3.3 The Smarties Test (Perner, Frith, Leslie, & Leekam, 1989).

1. Produce a Smarties box from your bag and ask the child: *"What's in here?"*

Participants answer

2. Open the box, and to the subject's surprise, a pencil emerges. Say: *"No, it's a pencil."*

3. Put the pencil back into the box, close the box, and asked two Prompt Questions:

Reality Prompt: *"What's in here?"*

Own-Response Prompt: *"When I first asked you, what did you say?"*

4. Now let's say we are going to get your friend/mum and ask them the same question

"Who should we get?" (Subject names next person.)

"S/he hasn't seen this box. When s/he comes in, I'll show her/him this box just like this and ask:

[Name] what's in here?"

Prediction Test: *"What will [Name] say?"*

Reality Check: *"Is that what's really in the box?"* (if answer is "No"):

"What is really in the box?"

5. Own-Response Check: *"Do you remember, when I took the box out of my bag [experimenter reenacts that episode] and asked you what was in it, what did you say?"*

.....

Scoring:

1...Pass - Self correct response & correct attribution

Answers correctly question number 3: reality prompt – pencil, own response prompt – smarties &

Answers correctly question number 4: prediction test – smarties, reality check – pencil

0... Fail – Answers one or both of the questions incorrectly.

If subject spontaneously corrects their wrong answer and do no dither on any other questions, accept their spontaneous corrections.

2.3.4. Self-Understanding Interview (Damon & Hart 1988)

2.3.4.1 Interview Schedule

Item 1: *Self-definition*: What are you like? What kind of person are you? What are you not like? How would you describe yourself?

Probes: What does that say about you? Why is that important? What difference does that (characteristic) make? What would be different if you were/were not like that?

Item 2: *Self-evaluation*: What are you especially proud of about yourself? What do you like most about yourself? What are you not proud of? What do you like least about self?

Probes: What does that say about you? Why is that important?

Item 3: *Self in the past and future*: Do you think you'll be the same or different 5 years from now? How about when you're an adult? How about 5 years ago? How about during your childhood?

Probes: What will be the same? What will be different? Why is that important?

Item 4: *Self-interest*: What do you want to be like? What kind of person do you want to be? What do you hope for in life? If you could have three wishes, what would they be? What do you think is good for you?

Probes: Why do you want to... be that way?...wish for that?...believe that is good for you? What else do you...hope for?...wish for?...believe is good for you? Why is that good for you?

Item 5: *Continuity*: Do you change at all from year to year? How (how not)? If you do change from year to year, how do you know it's still always you?

Probes: In what ways do you stay the same? Is that an important thing to say about you? Why?

Item 6: *Agency*: How did you get to be the way you are?

How did that make you the kind of person you are? How could you become different?

Probes: What difference did that make? Is that the only reason you turned out like you did? What else could make you different? How would that work?

Item 7: *Distinctness*: Do you think there is anyone who is exactly like you? What makes you different from anyone you know?

Probes: Why is that important? What difference does that make? In what other ways are you different? Are you completely different or just partly different? How do you know? Are you different from everybody or just from some people? How can you be sure you're different from everybody else when there are many people in world you do not know?

2.3.4.2 Coding instructions

The complete scoring manual (44 pages) was kindly provided by the authors (See disc attached). The transcript is divided into scoring units called chunks. A chunk is composed of a) a self-characteristic mentioned by the participants (e.g. "What kind of person are you." "I am a boy."), and b) all the participant's statements, whether spontaneous or in response to the interviewer's probe questions, that explicate the meaning or importance of that characteristic. (e.g. "Why is being a boy important?" "Because it makes me strong".).

Each chunk is matched with a one of the seven categories below and one of the four levels.

Although some chunks may be scorable at more than one levels within a single category, chunks are only scored at the highest applicable level in the category. Some chunks may be scorable in difference categories. A) If the chunk is scorable in two or more categories at the same level, the chunk is given all the applicable scores. B) Chunks may contain elements that can be construed at different levels in difference categories, e.g. social level 1 and psychological level 4. In such cases the scorer is to assume that the higher level of one category incorporates the lower level of another category, unless the interviewer probed in a way demonstrating that the lower level category has independent meaning.

A. Physical Self Category

- Level 1. Physical and material attributes of self have significance in and of themselves.
- Level 2. Physical and material attributes of self influence or reflect the nature of the self's activities or capabilities.
- Level 3. Physical and material attributes of self influence or reflect the self's social appeal, social interactions, social relations, or group membership.
- Level 4. Physical and material attributes of self influence or reflect the self's personal philosophy, moral standards, or lifestyle.

B. Active Self Category

- Level 1. Active attributes of self reflect the activities that the self performs, or the activities that are allowed, forbidden, or demanded of the self.
- Level 2. Active attributes of self are abilities considered relative to either other abilities of the self or to the abilities of other people.
- Level 3. Active attributes of self influence the self's social appeal, social interactions, social relations, or group membership.
- Level 4. Active attributes reflect or influence the self's personal philosophy, moral standards, or lifestyle.

C. Social Self Category

- Level 1. Social attributes of self reflect the fact of self's membership in social groups.
- Level 2. Self's activities or abilities are considered with reference to the reactions of other people.
- Level 3. Social attributes of self reflect self's personality characteristics or group membership that influences the nature of self's social interactions.
- Level 4. Social attributes of self reflect or influence the self's personal philosophy, moral standards, or lifestyle.

D. Psychological Self Category

- Level 1. Psychological attributes of self are moods and feelings that have significance in and of themselves.
- Level 2. Psychological attributes of self reflect one's cognitive capabilities, acquired knowledge, or activity-related emotional states.
- Level 3. Psychological attributes of self reflect or influence social skills or social interactions.
- Level 4. Psychological attributes of the self are personal philosophy, moral standards, or lifestyle.

E. Agency Category

- Level 1. Supernatural, biological, or social forces influence the existence or formation of the self.
- Level 2. Talents, abilities, wishes, motivations, or efforts of the self influence the existence or formation of the self.
- Level 3. Communication with others influences the existence or formation of the self.
- Level 4. Personal or moral evaluations of life possibilities influence the existence or formation of the self.

F. Continuity Category

- Level 1. Reference to one's continuity is relations to stable physical properties, possessions, and behaviours. The self's continuity over time is defended by reference to externally observable physical or behavioural characteristics of self.
- Level 2. Reference to one's continuity in relation to one's cognitive and active capabilities. At level 2 the child moves from a simple physicalistic assertion of self-sameness to an explanation that includes psychological characteristics. These psychological qualities are considered as immutable and permanent.
- Level 3. Reference to one's continuity in relation to recognition from others. Self-continuity is no longer defended by reference to static physical or psychological characteristics. At

level 3 self-continuity is related to the social context that extends beyond the self. One's sense of self-continuity is in part dependent upon others' continuing recognition of the self over time.

- Level 4. Reference to one's continuity in terms of the relationship between one's earlier and present characteristics of the self.

G. Distinctness Category

- Level 1. Reference to one's distinctness as base upon one's physicalistic features. The self's distinctness is asserted on the basis of observable physical properties and social group membership, which may include the self's appearance, name, typical activities, or social groups.
- Level 2. Reference to one's distinctness as stemming from differences between self and other along one character, personality, or cognitive dimension.
- Level 3. Reference to one's distinctness as deriving from a unique combination of psychological and physical characteristics.
- Level 4. Reference to one's distinctness as a consequence of the self's own unique subjective experience and subjective interpretations of the world.

References:

- Annaz, D., Karmiloff-Smith, A., Johnson, M. H., & Thomas, M. S. C. (2009). A cross-syndrome study of the development of holistic face recognition in children with autism, Down syndrome and Williams syndrome. *Journal of Experimental Child Psychology*, 102, 456-486.
- Dunn, L. M., Dunn, D. M., Styles, B., & Sewell, J. (2009). *The British Picture Vocabulary Scale III – 3rd Edition*. London: GL Assessment.
- Dunn, L. M., Dunn, L. M., Whetton, C., & Burley, J. (1997). *British Picture Vocabulary Scale*. GL Assessment.
- Perner, J., Frith, U., Leslie, A. M., & Leekam, S. R. (1989). Exploration of the autistic child's theory of mind: knowledge, belief, and communication. *Child Development*, 60, 688–700.
- Strauss, E., Sherman, E. M., & Spreen, O. (2006). *A compendium of neuropsychological tests: Administration, norms, and commentary*. Oxford University Press, USA.
- Wechsler, D. (1997). *Wechsler Adult Intelligence Scale—3rd Edition*. San Antonio, TX: Harcourt Assessment.
- Wechsler, D. (1999). *Wechsler Abbreviated Scale of Intelligence (WASI)*. San Antonio, TX: Harcourt Assessment.
- Wilson, R., Pascalis, O., & Blades, M. (2007). Familiar face recognition in children with autism: The differential use of inner and outer face parts. *Journal of Autism and Developmental Disorders*, 37(2), 314-320.