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„The relationship of adaptive functioning in children with Down syndrome and parental strain“

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Abstract

Objectives The incentive of this study was to describe strain in parents of children and adolescents with a Down syndrome diagnosis and to examine the impact of children's adaptive behaviours on it. Expected parental strain is a main factor in prenatal decision making in regard to the termination of a pregnancy with a Down syndrome (DS) diagnosis. For counselling parents-to-be, we need to better understand the consequences of raising a child with DS. Implications for support interventions are discussed.

Study design Parents of 6-16-year-old children and adolescents completed a parental stress index questionnaire (PSI), a questionnaire examining adaptive functioning of their children (VABS-3), and a screening instrument for comorbid mental disorders including autism spectrum disorder (DISYPS-III) during their participation in the TriO study.

Results Family income accounted for 20.5% of variance in parental health. Child's age explained a variance of 27.3% in depression, 18.4% in parenting partner / spouse relationship, and together with the child's daily living skills and autism spectrum disorder (ASD) symptomology it accounted for 60.8% of variance in competence.

Conclusions Parents of children with DS reported more problems regarding their health, competence, parenting partner / spouse relationship quality, and depression with having older, more care-dependent children, lower family income, and their children exhibiting more ASD symptoms. Lower income families did report more health-related problems. Having an older child had a negative effect on parents' competence, their relationship quality, and depression scores. Competence did appear to be most influenced by child's age, ASD symptomology, and their child's independence.

Keywords: parental strain, children / adolescents with Down syndrome, adaptive functioning, caregiver burden, independence, ASD

The Relationship between Adaptive Functioning in Children with Down Syndrome and Parental Strain

Theoretical Background

Late parenthood

The average age in which men and women become parents has been rising since the 1950s in Europe and the United States, especially in highly educated individuals and because of societal shifts like opportunities to late child baring. Since the 1990s, the trend to late and extremely late (48+ years) first motherhood increased considerably (Beaujouan, 2020; Qu, Soriano, & Weston, 2006), partly because of the increase in people's aspirations to gain higher education and those educational programmes taking longer and longer. Late motherhood brings specific amplified risks of having a child with an intellectual or developmental disability (IDD) like Down Syndrome (Allen et al., 2009). In their large-scale investigation, Allen et al. (2009) reported that mothers of children with Down syndrome (DS) were 8.5 times as likely to be ≥40 years old than control group mothers. This difference was accounted on errors in the maternal egg rather than paternal influence factors or genome mutation. Shin et al. (2009) estimated an increase of 31.1% in prevalence of DS at birth for 10 US regions from 1979 to 2003. They also found that in the same time period the prevalence of DS was significantly higher in older mothers (1.9%) as compared to younger mothers (-0.6%). Overall, estimations by Shin et al. (2009) would add up to an incidence of 1.18 in 1000 live births in the area.

Raising a child with a disability is commonly associated with increased parental strain related to an abundance of factors especially everyday activities and challenges (Caples et al., 2018; Eisenhower, Baker, & Blacher, 2005). There is a large movement in psychological research investigating parental burden in typically developing (TD) children in contrast to children with an IDD. Findings from these studies continuously report elevated stress and burden in families caring for children with a non-typical development (Baker et al., 2003).

Parental strain and Down syndrome

Parental strain is defined as perceived stress or burden associated with caring for children. Especially in the upbringing of a child with a disability parental strain is an extensively researched field (Corrice & Glidden, 2009). Previous studies tried to predict parental stress with different types of IDDs or disorders (e.g. Down syndrome, fragile X syndrome, autism spectrum disorder, cerebral palsy, fetal alcohol spectrum disorder, …; Abbeduto et al., 2004; Blacher & McIntyre, 2006; Hayes & Watson, 2013). Another direction

of research discusses the *ambiguous loss theory* (e.g. Boss, 2007; Farkas et al., 2019) applying especially to parents of children with an IDD, explaining parental stress by having to let go of pre-birth expectations (e.g. Rafferty, Tidman, & Ekas, 2020) and hopes for the child's future while at the same time caring for their child.

DS is a disorder caused by trisomy of *Homo sapiens* chromosome 21 (HSA21) and is the most common of the IDDs (Antonarakis et al., 2020). Since lifetime prevalence has increased significantly from the 1950s to 2014 (from 3.3 to 12.8 per 10,000 persons in the United States; de Graaf, Buckley, & Skotko, 2019), the effects of raising a child with DS on parental stress has become an area of global interest to more and more researchers.

Parental strain and adaptive behaviour

Influences on parental strain can be investigated by looking at parental, societal, and / or children's factors. Previous research investigating strain in caregivers of children with DS found various dimensions with a potential negative effect like families encountering numerous obstacles and challenges with society, schools, extended family, and medical personnel (Farkas et al., 2019; Krueger et al., 2019; Takataya, Yamazaki, & Mizuno, 2016). However, Baker, Seltzer, and Greenberg (2011) suggest that parental strain or well-being should be investigated by assessing different levels of family functioning including marital quality. Additionally, Ilhan, Akhan, Baygut, Dalli, and Yildirim (2019) discuss that maternal physical and mental health acts as a predictor for better child care (less feelings of insufficiency and more perceived competence) and therefore reduced parental stress and lower scores in depression.

Parental dimensions.

Pisula and Banasiak (2019) also found that parental dimensions like sense of competence as caregiver to be one of the key predictors for polish father's well-being. Regarding competence, the randomized clinical trial performed by Iadarola et al. (2018) on behavioural strategies for parents of children with ASD, found that while their strategies did improve parental competence, these results were highly intercorrelated with parental stress and strain and could not easily be distinguished. This finding suggests sense of competence to be one of the pillars of parental stress and strain. This could be true especially for mothers of children with an IDD, since Qu et al. (2006) surprisingly found that young mothers scored higher in perceived competence levels than did older mothers, who are more likely to have a child with an IDD like DS. In fact, a main part of support interventions aimed at parents of

children with an IDD as in the study by Decroocq et al. (2020), who tested a program for parents of children with a ASD, is enhancing their parenting competences in order to facilitate their everyday lives and reduce stress.

Totsika, Hastings, Emerson, Lancaster, and Berridge, (2011) found that maternal mental health was worse with children with ASD but not an intellectual disability (ID), especially when looking at emotional problems. While they found that children's difficult behaviours negatively impact parental mental health, fathers of children with an ID have been found less likely to experience a decline in their mental health in a systematic review and meta-analysis by Dunn, Kinnear, Jahoda, and McConnachie (2019). On the other hand, in the study by Stoneman (2007), paternal depression scores were lower for fathers of children with DS while Ilhan et al. (2019) found Turkish mothers' depression scores dependent on their perceived burden and competence. Also, van Steijn, Oerlemans, van Aken, Buitelaar, and Rommelse (2014) reported both mothers' and fathers' depressive symptoms to be related to parenting stress and child's ASD status. Not only is mental health an important factor for parental strain, physical health is also considered a crucial factor predicting quality of life (Mugno, Ruta, D'Arrigo, & Mazzone, 2007) in parents generally, but specifically parents of children with an IDD. Contrary to that, Hedov, Wikblad, and Annerén (2006) did not find a significant difference in sickness absence days from work in Swedish parents of children with DS.

Part of the parenting partner / spouse relationship is working together, making decisions together, encouraging one another in child rearing, and relieving some of the stress associated with the upbringing of a child with an IDD (e.g. Marchal, Maurice-Stam, Hatzmann, van Trotsenburg, & Grootenhuis, 2013). Especially in regard to perceived stress and strain the parenting partner / spouse relationship plays a critical role (Rafferty et al., 2020). The division of labour in the household is also a crucial factor in any cohabitating relationship. Many families with children with DS continue to follow rather traditional gender roles after the diagnosis in which the mother usually stays home to care for the children and the household while the father acts as the main breadwinner to provide financial support (Rafferty et al., 2020). Better health related quality of life was reported by parents (of six to eight year old children with DS) with a stronger partner relation during the study by Marchal et al. (2013). In line with this finding, parent's marital adjustment to the rearing of a child with an IDD along with mother's well-being have been suggested to also be valid predictors for father's parenting stress (Gerstein, Crnic, Blacher, & Baker, 2009). Kózka and Przybyła-

Basista (2018) did find the quality of the marital relationship to be a predictor for parents' well-being in both mothers and fathers.

In summary, important factors of parental strain seem to be perceived competence, physical and mental health (depression), and the parenting partner / spouse relationship.

Child dimensions.

Regarding children's dimensions, researchers also identified possible predictors for parental strain including social, communicative, and daily living skills of children and youths with DS (Baker et al., 2003; Beighton & Wills, 2019; Mello, Rivard, Terroux, & Mercier, 2019). These factors taken together form the adaptive skills and behaviours of the child or adolescent, describing how well the child can adapt to his or her environment and how much help he or she needs in their daily routine.

In their systematic review, Beighton and Wills (2019) specifically reported higher daily caring demands to be significantly burdening to parents of children with an IDD. Almost two-thirds (71%) of parents in a sample of Swedish parents with children with DS reported not having enough time for themselves (Marchal et al., 2013). In this sample, 30% of parents said that they had to give up a hobby and 33% reported having lost friendships because of having to care for their children with DS. Likewise, Marchal et al. (2017) found problems with their child's independence to be an essential theme for mothers and fathers of young adolescents with DS. Independence in domains like personal hygiene, food preparation, getting dressed, and finding their own way to school and back home are regarded as facilitators for parents' quality of life because it allows them to take more time for themselves and to be able to enjoy more activities together as a family (e.g. Hedov, Annerén, & Wikblad, 2002; Marchal et al., 2013, 2017).

Language skills – especially expressive language skills – are often delayed in children with DS and these impairments persist throughout adolescents and adulthood (Grieco, Pulsifer, Seligsohn, Skotko, & Schwartz, 2015; Ilhan et al., 2019). This is only one of the factors that often make it difficult to accurately measure various levels of functioning (Antonarakis et al., 2020). Especially regarding mental health, communication difficulties hinder the administration of proper treatments (Antonarakis et al., 2020). Baker-Ericzén, Brookman-Frazee, and Stahmer (2005) discuss in their study about stress in parents of children with ASD that communication difficulties promote parent stress and depression.

Another domain extremely important in regard to parent stress is socialization of the child. As Lecavalier, Leone, and Wiltz (2006) suggested in their investigation of parental

stress predictors for raising children with ASD, the children's social behaviours or rather misbehaviours were the most prominent predictors for parental strain. They found parental outcomes to be influenced more by externalizing (problem behaviour of an acting-out nature), like conduct problems and lack of prosocial behaviour, rather than internalizing (behaviour problems of an emotional nature) children's behaviour. Social skills did in fact demonstrate to be significant predictors for maternal child related stress in the study by Baker-Ericzén et al. (2005). Interestingly, children with DS are often described as socially competent due to higher social motivation and responsiveness compared to children with other IDDs (Grieco et al., 2015).

Maladaptive, difficult behaviours are essentially behaviours depicted by a lack of social, communicative, and daily living skills. In more detail, they are described as temper tantrums, aggressive, self-abusive, obsessive, destructive, ritualistic, impulsive, and self-stimulatory behaviours and the constant need of supervision and assistance with daily living (Mugno et al., 2007; Skotko, Levine, Macklin, & Goldstein, 2016). Hauser-Cram et al. (2001) report in their longitudinal study on the cognitive and adaptive behaviour development in children with an IDD (infancy to middle school), behavioural problems to be a crucial predictor of maternal parenting stress. Mello et al. (2019) add autism spectrum disorder (ASD) symptom severity and low intellectual functioning to the children's maladaptive behaviours. Siegel and Smith (2010) add inattention, attention seeking, getting into fights, and hyperactivity to externalizing maladaptive behaviour. They also describe fears, anxiety, sadness / depression, and irritability as part of internalizing maladaptive behaviour (Siegel & Smith, 2010). Furthermore, research not targeting families with children with disabilities, but TD children, found that high internalizing and externalizing behaviour problems were most strongly associated with parental strain and stress (Vaughan, Feinn, Bernard, Brereton, & Kaufman, 2013).

Autism spectrum disorder and adaptive behaviour.

Comorbid conditions concerning physical health which are common with a Down syndrome diagnosis are congenital malformations of multiple organ systems (e.g. heart, gastrointestinal malformations), increased risk for seizures, thyroid disease, celiac disease, refractive errors, strabismus, hearing loss, eustachian tube dysfunction, occipito-atlanto-axial instability, acute leukaemia, pulmonary hypertension, and increased susceptibility to infections (cf. Bush, Galambos, & Dunbar Ivy, 2020; Siegel & Smith, 2010), while most solid tumour types less frequently occur with a DS diagnosis (Antonarakis et al., 2020).

Individuals with a DS diagnosis are also more prone to exhibit specific cognitive conditions like intellectual disability, developmental delay, language delay, and mental disorders (one third meet criteria; Siegel & Smith, 2010) like attention deficit / hyperactivity disorder (ADHD), conduct disorder, anxiety disorder, ASD, or depression (Siegel & Smith, 2010). Approximately one in ten persons with DS is additionally diagnosed with ASD (Siegel & Smith, 2010).

Gath and Gumley (1986) described a phenomenon named *dual diagnosis* which means that individuals with an IDD often also meet the diagnostic criteria for a behaviour disorder (e.g. Eisenhower et al., 2005). In contrast to that, children with DS are often described as especially kind and good-tempered with a positive impact on their parents and other people through their joyful and positive interactions (Farkas et al., 2019; Grieco et al., 2015; Sarimski, 2020). Skotko et al. (2016) found in their study including 1,961 parents or guardians, that nearly all of them reported loving their child and being proud of her or him. Their data showed that the majority of parents felt that their outlook on life was more positive because of their child with DS.

In children with DS, maladaptive behaviours generally seem less common than in children with ASD symptomology (Eisenhower et al., 2005; Mugno et al., 2007; Pastor-Cerezuela, Fernández-Andrés, Pérez-Molina, & Tijeras-Iborra, 2020). Lower well-being for mothers of children with fragile X syndrome and autism spectrum disorder in comparison to mothers of children with DS have been found by Abbeduto et al. (2004). In a meta-analysis by Hayes and Watson (2013), examining 15 studies from 1989 to 2012, parents of children with ASD reported significantly higher levels of stress with high effect sizes compared to parents of children with other disabilities or TD children. Similarly, Blacher and McIntyre (2006) found elevated stress and lower well-being in parents of children with ASD as compared to a group of children with DS. Children with ASD were also found to exhibit more maladaptive behaviour than children with DS. Baker-Ericzén et al. (2005) also found in their quasi-experimental pre-post design before and after an inclusive toddler program that maternal stress was higher with children who exhibited more symptoms of ASD. Additionally, Rafferty et al. (2020) found in their qualitative research utilizing telephone interviews, that fathers of children with ASD reported elevated stress associated with child mood and behaviour. Consistent with these results, Pisula and Banasiak (2019) reported higher parental stress and reduced feelings of competence in fathers of children with ASD compared to other IDDs including DS, from their preliminary study. Comparing children with

DS and children with ASD, Dabrowska and Pisula (2010) found that parents of children with ASD reported higher stress than parents of children with DS.

Children with DS but not ASD showing less maladaptive behaviour is in fact representative of a controversially discussed effect named the *Down syndrome advantage.* It describes that parents of children with DS less frequently report behaviour problems in their children than parents of children with other developmental disabilities (Corrice & Glidden, 2009; Farkas et al., 2019; Skotko et al., 2016). Numerous possible explanations have been discussed including the controlling for socio-demographic factors like parent age and education level, family income, and access to resources as in the study by Stoneman (2007), which found that family income accounted for the difference in parental strain with children with DS and other IDDs, diminishing the *Down syndrome advantage*. Corrice and Glidden (2009) did explain the *Down syndrome advantage* as an effect of advanced maternal age and children's adaptive behaviour. Marquis, McGrail, and Hayes (2020) found that, rather than being influenced by children's behaviour, fathers were at elevated risk of being diagnosed with depression depending on their family income while mother's mental health depended on their child's gender. They reported mothers of girls with an IDD to be more likely to be diagnosed with depression. Similarly, Dickinson and Place (2016) did find a gender effect favouring boys. Opposed to that were the results by Senses Dinc, Cop, Tos, Sari, and Senel (2019), who reported a weak correlation of quality of life in mothers of children with DS with child gender in favour of girls. They found that having a baby girl with DS partly predicted an increase in the mother's score of World Health Organization Quality of Life (WHOQOL) physical health in comparison to having a boy. As females are less likely to meet diagnostic criteria of ASD (Baio et al., 2018), this could mean that girls exhibit less behaviour problems associated with ASD.

As child's gender has been found to be a predictor, so has child's age. Eisenhower et al. (2005) found in their study, investigating maternal well-being, that children with DS showed similarly low levels of problem behaviours as TD children, while also discussing that this might be an effect of child age rather than syndrome specificity. Child age is discussed as a relevant factor mainly because parents of younger children tend to get less sleep and generally invest more time in the physical care of their children (Hagen, Mirer, Palta, & Peppard, 2013). Nærland, Bakke, Storvik, Warner, and Howlin (2017) reported in their Norwegian study on children and adolescents with DS that their diagnosis was correlated with ASD symptoms and that these were related to age and gender of the participant. The same age-related effect was shown in the study by Dickinson and Place (2016), where mothers of

primary aged children and teenage girls with ASD exhibited more stress and dysfunctional parent-child interactions than mothers of secondary aged children with ASD.

Contrary to these findings, no effects of socio-demographic predictors have been found by Marchal et al. (2013) in their study on health related quality of life in parents of children with DS. Similarly, in their investigation of parental distress and behavioural difficulties in TD children, Broadhead, Chilton, and Crichton (2009) found that neither marital status, child's age, number of siblings, ethnicity, age of parents, nor child's gender showed a significant relationship with parent's stress levels. Pastor-Cerezuela et al. (2020) also did not find parents' socio-demographic factors like gender, educational level, economic level, occupation, residence, marital status, or number of children to be different in their group comparison of DS, ASD, and TD children. Differences in parental stress have been suggested to be accounted for by child dimensions or IDD phenotype, again favouring DS over ASD (Pastor-Cerezuela et al., 2020).

All in all, empirical results are still contradictory concerning the maladaptive behaviours of children with DS. Many studies assessing behaviour problems do not control for highly comorbid psychiatric disorders like ASD, which are in fact more frequently associated with behaviour problems (Blacher & Baker, 2019; Blacher, Baker, & Kaladjian, 2013; Corrice & Glidden, 2009; Eisenhower et al., 2005; Pastor-Cerezuela et al., 2020) and the literature is still inconsistent regarding the socio-demographic variables (e.g. Eisenhower et al., 2005; Marchal et al., 2013; Marquis et al., 2020; Senses Dinc et al., 2019). This arises the question if parental strain in parents of children with DS is rather a product of socio-demographic factors or behavioural problems that come with additional ASD symptomology, and not the DS diagnosis.

Hypotheses for analysis

Hypothesis 1(a)

Children and adolescents with a DS diagnosis and high scores in the ASD phenotype more frequently show maladaptive behaviours.

Hypothesis 1(b)

Higher adaptive behaviours are negatively related to the important subscales of parental strain.

Hypothesis 2(a)

Higher communicative skills negatively influence the important subscales of parental strain.

Hypothesis 2(b)

Higher social skills negatively influence the important subscales of parental strain.

Hypothesis 2(c)

Higher daily living skills negatively influence the important subscales of parental strain.

Hypothesis 2(d)

Higher ASD symptomology influences the important subscales of parental strain.

Hypothesis 3

Socio-demographic factors are not significantly associated with the important subscales of parental strain.

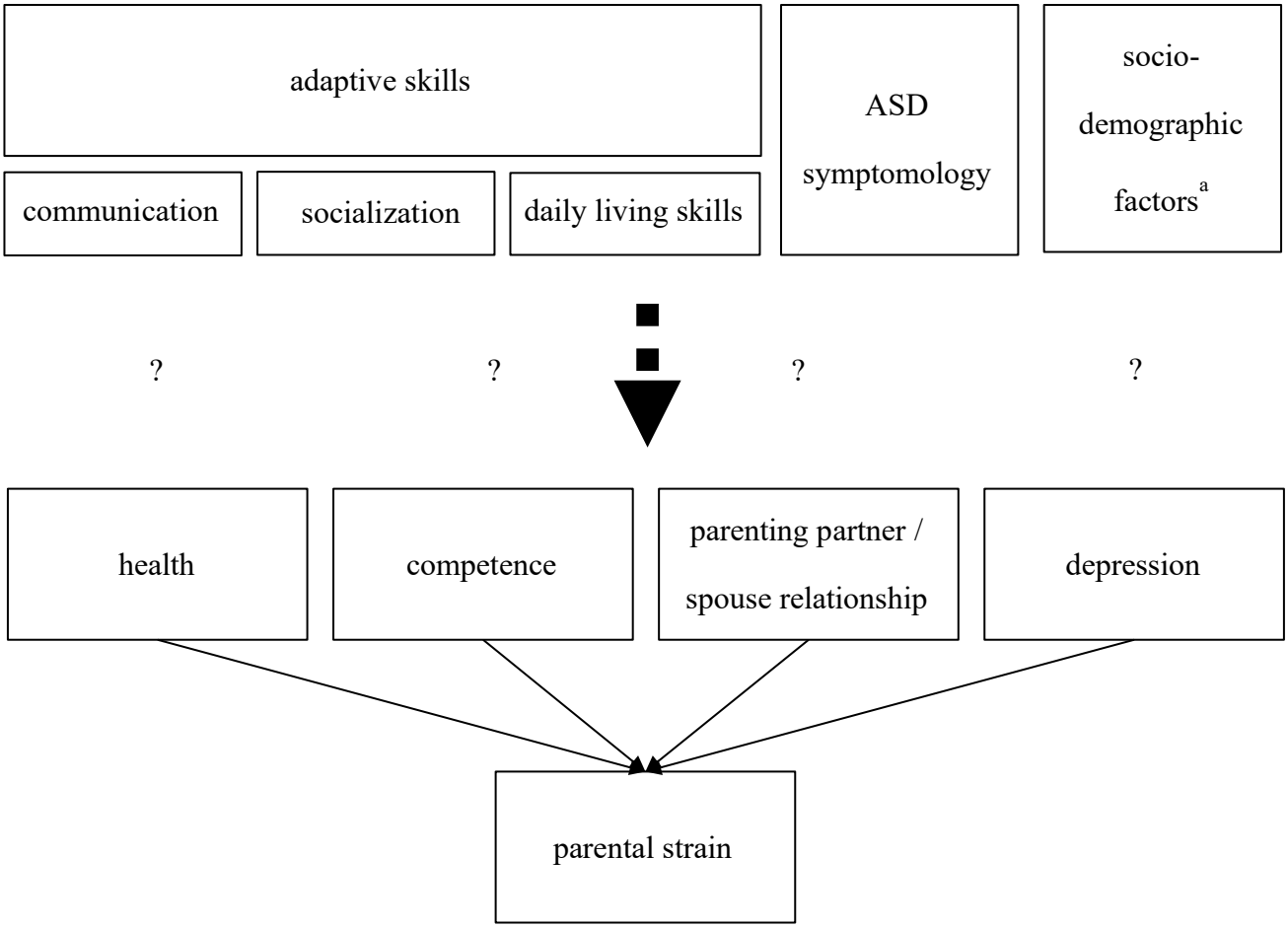

Figure 1. **Hypothetical model of predictors on parental strain.** Relationships between predictors on the main influences on parental strain are illustrated with a question mark. No hypotheses about specific associations are assumed. Main predictors on parental strain are parental health, perceived competence, parenting partner / spouse relationship, and depression. [a] socio-demographic factors include mother's age, mother's education, child's age, child's gender, number of siblings, intervention type, family income.

Relevance

Contemporary prenatal medical scanning is making it much easier for parents-to-be to early diagnose Down syndrome in their unborn child. Due to incomplete or flawed information – or the absence of it – a lot of pregnancies with a positive diagnosis to date are being terminated. Fears and preconceptions, what life raising a child with DS would be like and how stressful it would be, influence parental decision making. This is why it is essential to find out more about the factors predicting potentially increased parental strain. In this prospect, parents' consultations could benefit greatly from more up-to-date knowledge (Skotko, Levine, & Goldstein, 2011), especially concerning useful support programs or interventions.

Moreover, parental stress and burden is regarded a tremendous threat to child development and well-being (Mackler et al., 2015). Garbarski (2014) found in a longitudinal study that child's health (activity limitations) and maternal health (including mental health as in depressive symptoms) mutually influence each other. Also, research by Baker et al. (2003) suggests a reciprocal relationship between child behaviour problems and parental strain. In their study with 205 pre-school children, higher parental stress led to more behaviour problems and vice versa. Years later, van Steijn et al., (2014) found the same results in their research, addressing ASD, ADHD, and depressive symptoms in the relationship with parental stress. More recent results by Blacher and Baker (2019) were similarly highly suggestive regarding a reciprocal relationship between maternal well-being and child behaviour problems.

Taking these findings into account would mean that helping parents accomplish a healthier relationship with their children by reducing their parental strain would also greatly benefit the children's development in the long run.

In conclusion, better understanding of the factors influencing parental strain in raising a child with DS will not only lead to more informed decision making about pregnancy termination or completion, but will also positively contribute to the children's development (Blacher & Baker, 2019).

For this reason, the current study is aiming to extract highly influencing factors of child adaptive behaviours and skills on parental strain. To thoroughly investigate the possible specific influences the preliminary model has been assumed (see Figure 1). It illustrates the hypothesized predictors on the main pillars of parental strain (including health, competence, parenting partner / spouse relationship, and depression). Previous research inconsistently suggests adaptive skills (communication, socialization, daily living skills) and socio-demographic influences on parental strain. Because of maladaptive behaviours being more prevalent in children with ASD symptomology (e.g. Eisenhower et al., 2005; Mugno et al., 2007; Pastor-Cerezuela et al., 2020), ASD scores are also included into the model (see Figure 1).

Study design

The data regarding parental strain outcomes and adaptive functioning in children with DS were gathered within the larger TriO project: an ongoing study with the main aim of the extraction of relevant DS subgroups with heterogeneous risk factors (including strain on the family system) in order to be better prepared for counselling of parents after the diagnosis.

Due to the heterogeneity of intellectual disability in persons with DS prenatal consultation is extremely difficult in medical practice. For this reason, the comprehensive TriO study aims to extract relevant subgroups in regard to cognitive (WISC-V, WPPSI-IV; Daseking & Petermann, 2018) and emotional development of children and adolescents (6 to 17 years).

Also of interest for this investigation are the adaptive and maladaptive behaviours, measured by the Vineland Adaptive Behavior Scales (VABS-3; Sparrow, Cicchetti, & Saulnier, 2016).

Additionally, olfactory differentiation ability (as a non-invasive marker for early onset Alzheimer's dementia) is being assessed to define subgroups at elevated risk of developing Alzheimer's dementia (for more information on the high comorbidity with Alzheimer's dementia see Antonarakis et al., 2020). Also included is a measure for differentiating parental strain between the extracted subgroups.

Recruitment techniques included distributing flyers in institutes for families with children with DS, posting advertisements on social media throughout the course of the study, and personal contacts. Interested families made appointments in the university of Vienna's lab for psychological research.

The current design

For this assessment, parents of 6-16-year-old children and adolescents taking part in the more comprehensive TriO study completed the EBI (Eltern-Belastungs-Inventar; Tröster, 2011; German version of the Parenting Stress Index, PSI; Abidin, 1995), the VABS-3 (Vineland Adaptive Behaviour Scales; Sparrow et al., 2016), and the DISYPS-III (Döpfner & Görtz-Dorten, 2017).

Participants

A power analysis using G*Power (Mayr, Erdfelder, & Buchner, 2017) concluded that for a moderate effect 20 participants (care takers / parents) were needed for the intended multiple linear regression analysis.

A total of 19 parent-child dyads participated in the study at hand over the course of 10 months from December 2019 to September 2020. The participants were recruited as part of the ongoing TriO study. Families were eligible for participation in the study group if their child was between 6 to 17 years old and had received a DS diagnosis. The majority of participating parents were mothers, although in one case the grandmother and in another case

the father accompanied the mother to the appointment. The participating parents signed an informed consent form and answered questions about the pregnancy, birth, and current living situation of the child with DS. Subsequently, the research staff conducted a semi-structured interview and three questionnaires (VABS-3, EBI, DISYPS-III) with the primary caretaker in a separate room from the child. The child was given a cognitive test (WISC-V or WPPSI-IV), an affective differentiation test (IAPS), and an olfactory discrimination test (Sniffin´ Sticks). For participating children who could not concentrate on the tests or who exhibited highly disruptive behaviours, the researchers provided alternative activities to pass the time until their care takers returned.

Predictors

From what previous literature results suggest, there are specific subscales of the parental stress index (PSI) that are especially important – and frequently reported by parents – predictors for low overall parental strain. Those are the scales competence, depression, health, and parenting partner / spouse relationship (e.g. Baker et al., 2011; Ilhan et al., 2019; Kayfitz, Gragg, & Robert Orr, 2010; Kózka & Przybyła-Basista, 2018; Marchal et al., 2017; Nomaguchi & Milkie, 2020; Pastor-Cerezuela et al., 2020; Senses Dinc et al., 2019; Shonkoff, Hauser-Cram, Wyngaarden Krauss, & Upshur, 1992; Xanthopoulos et al., 2017). To ensure highly reliable results with minimized type II errors, these subscales are included in the following multiple linear regression analysis (see Table 6) while the rest is dismissed. The final selection of predictors was clustered as parental dimensions and child dimensions.

Instruments

VABS-3.

The Vineland Adaptive Behaviour Scales (Sparrow et al., 2016) is a questionnaire assessing child dimensions via parent report. It is best administered through an interview by a trained instructor (Shonkoff et al., 1992). These interviews took place in the diagnostic facilities of the university of Vienna.

The VABS-3 includes the following domains / subdomains: communication (receptive, expressive, written), daily living skills (personal, domestic / numeric, community / school community), socialisation (interpersonal relationships, play and leisure, coping skills), and motor skills (gross motor, fine motor). The first three domains are the core domains of the VABS-3 containing the major adaptive behaviours required for the study at hand.

Communication domain.

The receptive subdomain of the VABS-3 is defined as attending, understanding, and responding appropriately to information from others. The expressive subdomain aims at the usage of words and sentences to express oneself verbally to others. An example item of the expressive subdomain is *What objects and actions does [name] say the words for, like "dog" or "eat"?*. The written subdomain contains questions about reading and writing skills (Sparrow, Cicchetti, & Saulnier, 2016).

Daily living skills domain.

In the personal subdomain self-sufficiency in such areas as eating, dressing, washing, hygiene, and health care are included. The domestic subdomain was designed in regard to performance in household tasks such as cleaning up after oneself, chores, and food preparation. The numeric subdomain includes using numeric concepts in practical ways, including time, dates, and money. The community subdomain is defined as the ability to function in the world outside the home, including topics such as safety, using money, travel, rights and responsibilities. The school community subdomain incorporates items about meeting the expectations for appropriate behaviour within the school environment. An example item of this domain is *What does [name] do to clean up after himself/herself, like with spills and dirty clothes?*.

Socialization domain.

The subdomain interpersonal relationships include questions about the child's responding and relating to others, including friendships, caring, social appropriateness, and conversation. The play and leisure subdomain is about engagement in play and fun activities with others. The coping skills subdomain is defined by demonstration of behavioural and emotional control in different situations involving others. An example item of this domain is *How do you know what feelings or emotions [name] is having, and whether he/she knows what emotions others are having?*.

Maladaptive behaviour domain.

The internalizing subdomain describes problem behaviours of an emotional nature. An example item for this subdomain is *Has he/she eating problems like overeating, refusing to eat, only eating one or two things, or hoarding food?*. The externalizing subdomain includes items about problem behaviours of an acting-out nature. An example item for this subdomain

is *Is he/she physically aggressive, like hitting, kicking, biting?*. This domain also incorporates critical items about more severe maladaptive behaviours that do not form a unified construct. An example of an item from the critical domain would be *Does [name] use strange or repetitive speech? Examples: Has he/she conversations with himself/herself in public, says things that make no sense, repeats the same thing over and over?*.

EBI.

The EBI (Eltern-Belastungs Inventar, Tröster, 2011; German version of the parental stress index, Abidin, 1995) is a self-report screening tool to systematically define two main sources of parental stress: child dimensions (behaviours and features of the child that put specific strain on the parents, like attentiveness and hyperactivity, mood, adaptability, neediness) and parental dimensions (restrictions in parental functioning that hinder them from successfully handling the challenges of child rearing, like attachment, social isolation, perceived competence, depression, health, role restriction, parenting partner / spouse relationship).

The EBI has been designed with a sample of 538 mothers (aged 20 to 53 years, M = 34.9 years, SD = 5.5 years) in Germany after the *Parenting Stress Model* (PSM, Abidin, 1995), accounting increased parental strain on a mismatch of requirements in child rearing and parental resources necessary to manage those requirements.

The EBI has been validated using samples of mothers of children with chronic disorders and disabilities.

The EBI incorporates 48 items to scan for signs of parental impairment as a result of heightened strain and to evaluate if support from outside the family is needed. On a five-point Likert scale (from *I do not agree* to *I strongly agree*), parents are asked to rate statements like *Sometimes I struggle to emphasize with my child*., *My child needs more daily attention than other children*. or *I blame it on myself when my child misbehaves*.

DISYPS-III.

The DISYPS-III (Döpfner & Görtz-Dorten, 2017) serves diagnostic purposes of psychiatric disorders including ADHD, disorders of social interaction, depressive disorders, anxiety disorders, PTSD, obsessive compulsive disorders, tic disorders, autism spectrum disorders, and attachment and relationship disorders. It is suitable for children and adolescents aged 4 to 17 years. There are parent, teacher, and self-report versions of the instrument. They are identical in terms of content although they have different item structures.

The autism spectrum diagnostic check list is based on the symptom criteria of the ICD-10 and DSM-5. There is no self-report version of this part of the instrument since self-report of autism spectrum symptomology can be challenging.

The parent report version can be divided into three scales assessing symptomology based on the DSM-5: persistent deficits in social interactions and communication, restricted and repetitive behaviours, and difficulties with using verbal and nonverbal communication in social situations. The scale contains 63 items including 16 items scanning for deficits in social interactions and communication, nine items for restricted and repetitive behaviour, interests, and activities, four items concerning difficulties with using verbal and nonverbal communication in social situations, 29 items for overall symptoms of ASD, and five items for impairment of function and psychological strain.

The item's factor loadings and internal consistencies were acceptable (internal consistencies of the overall scale were between α=.66 and α=.80) for deficits in social interactions and communication and stereotypical behaviours. The items were being rated by the interviewer on a 3-point scale ranging from *not true* (marked by a minus sign), *questionably true* (question mark), to *true* (plus sign).

Data preparation

The very few missing values (one for number of siblings, parenting partner / spouse relationship, and mothers age at birth of the child with DS) have been inserted by multiple imputation. To accomplish imputated data, 20 different data sets have been computed, inserting values that depend on the distribution in the sample.

Results

Participants

Table 1 includes the socio-demographic characteristics of the families with a child with DS included in the statistical analysis. As discussed above, late motherhood is extremely prevalent in highly educated individuals. The data suggests validity of this phenomenon by the observed distribution of mother's age and education, since the mother's mean age at birth of the child was 35.0 years (range 23-42.6 years, SD = 4.8 years) and 65% of the mothers in this sample did complete higher education like a university degree (e.g. Table 1).

Gender distribution of the children in the current sample was almost balanced (female 52.6%, male 47.4%). They were on average 9.6 years old (range 6.0-16.1 years, SD = 2.7 years) and almost all of them had at least one sibling (94.9%, range one to six siblings, SD =

1.3 siblings). The overwhelming majority of the children had received continuous interventions since birth (84.2%). The rest had merely received early interventions.

For 63.2% of children included in the current sample their parents reported behaviours beneath the cut-off of ASD-specific symptomology (within typical limits; see Table 2). Another 21.1% of children met criteria for noticeable symptoms and 15.5% for extremely noticeable symptoms. This results in a total of 36.9% children with ASD symptomology beyond the cut-off.

Table 1

Socio-demographic characteristics, n = 19

Socio-demographic	range	M (SD)	n (%)
Age mother at birth	23.0-42.6 years	35.4 years (4.8)	
Gender child (n, %)			
female			10 (52.6%)
male			9 (47.4%)
Age child	6.0-16.1 years	9.6 years (2.7)	
Education level mother			
High[a]			12 (63.2%)
Intermediate[b]			6 (31.6%)
Low[c]			1 (5.3%)
Total number of siblings	1-6 siblings	1.5 siblings (1.3)	
None, Three, Six (*respectively*)			1 (5.3%)
One			12 (63.2%)
Two			4 (21.1%)
Interventions			
only early interventions			3 (15.8%)
continuous interventions			16 (84.2%)

Note. [a] University degree or degree from an equivalent educational institution. [b] Vocational school with higher education entrance qualification. [c] Completion of secondary school

Descriptive analysis of predictors of parental strain

DISYPS-III scores: ASD symptomology.

Table 2 illustrates parent reports of slightly noticeable to noticeable symptoms of ASD in 31.6% (n = 6) of children included. Below the cut-off were 68.4% of children, exhibiting typical levels of symptomology. The data shows a gender difference in ASD symptomology

in favour of girls with only 10% of girls in this sample exhibiting ASD symptoms above the cut-off for typical behaviour phenotypes. Of the boys in this sample 55.5% showed ASD symptoms above the cut-off, with 22.2% (n = 2) exhibiting highest ASD symptomology in the sample (see Table 2).

Hypothesis 1(a) predicted children and adolescents with a DS diagnosis and high scores in ASD symptomology to exhibit more maladaptive behaviours. Statistical analysis reported a strong negative correlative relationship between the level of ASD symptomology and child adaptive behaviour ($r = -.553$, $p = .014$). These results suggest validity of hypothesis 1(a): Children with DS and comorbid ASD symptomology exhibit more maladaptive behaviour.

Table 2

Level of ASD symptomology in children with DS in the sample. n = 19

level	total (n, %)	female (n, %)	male (n, %)
within typical limits[a]	13 (68.4)	9 (90.0)	4 (44.4)
slightly noticeable	4 (21.1)	1 (10.0)	3 (33.3)
noticeable	2 (10.5)	0	2 (22.2)
total atypical[b]	**6 (31.6)**	**1 (10.0)**	**5 (55.5)**

Note. ASD symptomology as measured by the DISYPS-III. [a] within typical limits describes the group below the cut-off for ASD-specific symptomology, [b] total atypical includes the slightly noticeable and the noticeable groups.

VABS-3 scores.

On average, the children in the current sample achieved the highest scores in the daily living skills domain (M = 85.74) while one child did achieve the highest score of 103 of all in the socialization domain (M = 83.58). The lowest scores have been achieved in the communication domain with a mean value of 74.47. High values can be interpreted as more advanced skills.

The lowest discrepancy in skills were shown in socialization domain with a standard deviation of 7.50. The most variance was shown in the communication domain with a standard deviation of 12.04 and a similar range in ability was found in the daily living skills domain with a standard deviation of 11.46.

Table 3

Means, standard deviations, and range of VABS-3 values. n = 19

VABS-3[a]	M	SD	range
COM	74.47	12.04	44 - 92
SOC	83.58	7.50	72 - 103
DLS	85.74	11.46	64 - 101

Note. [a] Child dimensions form the adaptive skills and behaviours of the child as measured by the VABS-3. M = mean, SD = standard deviation, COM = communication. SOC = socialization. DLS = daily living skills.

EBI scores.

Parental strain dimensions have been reduced to highly predictive variables according to the literature. The remaining predictors are health, competence, parenting partner / spouse relationship, and depression. Means and standard deviations have been computed (see Table 4).

Parents reported the highest scores in parenting partner / spouse relationship (M = 10.92, DS = 4.22) and depression (M = 10.37, SD = 4.09). Parents reported the lowest scores in health with a mean of 8.47 (SD = 4.50).

Table 4

Means and standard deviations of EBI values. n = 19

EBI[a]	M	SD
HEA	8.47	4.50
COMP	9.16	4.54
PPSR	10.92	4.22
DEP	10.37	4.09

Note. [a] Parent dimensions measured by the EBI are reduced to the highly predicting factors resulting from previous research. Range = 4 to 18. HEA = health, COMP = competence, PPSR = parenting partner / spouse relationship, DEP = depression, M = mean, SD = standard deviation.

Correlative relationships with parental strain subscales

Socio-demographic predictors.

For an overview of possible strong connections and to test hypothesis 3, correlative relationships between the socio-demographic variables and parental outcomes have been computed. Hypothesis 3 did predict socio-demographic predictors to not have a significant association with parental strain outcomes.

The analysis of the data did not show a significant correlative relationship (see Table 5; p-values were larger than .05) with mother's age, education, number of siblings, intervention type, or family income (except for the relationship approaching significance in the parents' health subdomain, which is why family income has not been excluded from the final analysis; see Table 5), partly confirming hypothesis 3.

Yet, significant results have been accomplished in the variables child's gender and child's age (see Table 5).

Child's gender did show high effect sizes in relation with competence (r = .473, p = .041), parenting partner / spouse relationship (r = .417, p = .085), and depression (r = .470, p = .042) with higher scores in the parental outcomes for having a male child.

The mean score in health for having a girl was 7.67 (SD = 4.56), with boys it was 9.80 (SD = 4.18). High values in the outcome health can be interpreted as an increase in health-related problems. The mean score for competence for having a girl was 7.22 (SD = 3.31), with boys it was 11.3 (SD = 4.55). High values in the outcome competence can be interpreted as a decline in perceived competence. The mean score in parenting partner / spouse relationship for having a girl was 8.94 (SD = 4.23), with boys it was 12.44 (SD = 3.84). High values in this outcome can be interpreted as a decline in the parenting partner / spouse relationship. The mean score in depression for having a girl was 8.44 (SD = 3.88), with boys it was 12.20 (SD = 3.58). High values in the outcome depression can be interpreted as an increase in depressive symptoms.

Child's age shows a large correlative relation with parental depression with an effect size of .523, which is significant (.021). An increase in child's age is related to an increase in parental depression. Having an older child was also related to a decrease in parental partner / spouse relationship quality with a large effect size of .429 approaching significance (.076).

Family income was clustered into four groups: *up to 2,500€*, *up to 3,000€*, *up to 4,000€*, and *above 4,000€* of monthly family income. This variable did show a negative relationship with parental health with an effect size of -.452 approaching significance (p = .052). A negative correlative relationship with this variable suggests increased health in higher income families and decreased health in lower income families.

Table 5

Relationship of relevant socio-demographic predictors with parental strain: Pearson's r, p

SD-P	HEA	COMP	PPSR	DEP
MA	.160; .527	.254; .309	.369; .145	.191; .448
ME	-.182; .457	.061; .804	-.132; .603	.253; .294
CG	.250, .302	**.473****, .041	**.417***, .085	**.470****, .042
CA	.270, .263	.473, .041	**.429***, .076	**.523****, .021
NS	-.210; .404	-.156; .536	-.214; .408	-.311; .209
INT	-.292; .225	-.097; .693	-.281; .258	-.099; .687
FI	**-.452***, .052	-.386, .103	-.243, .331	-.136, .578

Note. SD-P = socio-demographic predictors. Mother's age (MA), mother's education (ME), number of siblings (NS), and intervention type (INT) did not come in anywhere near a significance level of $p < .05$. CG = child's gender, CA = child's age, FI = family income, HEA = health, COMP = competence, PPSR = parenting partner / spouse relationship, DEP = depression. * approaching significance. ** $p < .05$.

Psycho-social predictors.

Table 6 describes the correlative relationships between the parental subscales and the children's psycho-social dimensions. Hypothesis 1(b) said, that higher adaptive behaviours were negatively related to the important factors of parental strain. This seems to be true for communication, socialization, daily living skills, and low ASD symptomology (see Table 6) associated with higher maladaptive behaviour.

Table 6

Relationship between parental strain and relevant psycho-social predictors: Pearson's r, p

	HEA	COMP	PPSR	DEP
COM	**-.406***, .084	**-.455***, .050	**-.347***, .090	-.180, .231
SOC	-.380, .109	**-.513******,** .025	-.343, .163	-.171, .242
DLS	**-.454***, .051	**-.618*****, .005	**-.426** *, .078	-.109, .657
ASD	**.414***, .078	**.685*****, .001	**.508****, .031	.355, .136

Note. HEA = health, COMP = competence, PPSR = parenting partner / spouse relationship, DEP = depression, COM = communication, SOC = socialization. DLS = daily living skills. * approaching significance ($p < .10$). ** significant ($p < .05$). *** highly significant ($p \leq .005$).

The adaptive skill communication did show a negative correlative relationship with medium to large effect sizes with health ($r = -.406$, $p = .084$), competence ($r = -.455$, $p =$

.050), and parenting partner / spouse relationship (r = -.347, p = .090). Higher scores for children in the communication domain meant increased parental health, competence, and parenting partner / spouse relationship quality. Highly correlated predictors have been used to illustrate the relationships with the outcomes (which have been coded in a positive way to be easier to interpret) in Figures 2.1 to 2.5.

The adaptive skill socialization showed a negative relationship with a large effect size with competence (r = -.513, p = .025), suggesting an increase in parental perceived competence with higher social skills of the child (see Figure 2.1).

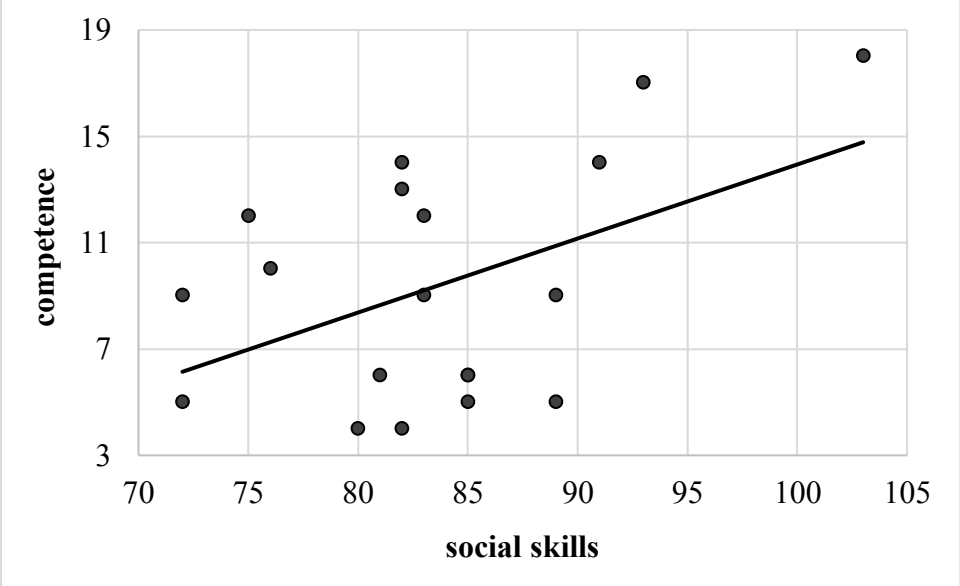

Figure 2.1. **Regression of perceived competence and child's social skills.** Parents who reported their children to exhibit better social skills also reported higher levels of competence.

The children's daily living skills have been associated with parental health with a medium to large effect size of -.454 approaching significance (p = .051), suggesting a higher score in independence from parental help in domains like personal hygiene, food preparation, and getting dressed to be related to better parental health (see Figure 2.2).

Figure 2.2. **Regression of parental health and child's daily living skills.** Parents who reported their children to exhibit better daily living skills also reported better physical health.

Also, daily living skills clearly show an extremely large and highly significant relationship with perceived competence ($r = -.618$, $p = .005$), suggesting parents' competence levels to rise with the daily living skills of their child (see Figure 2.3).

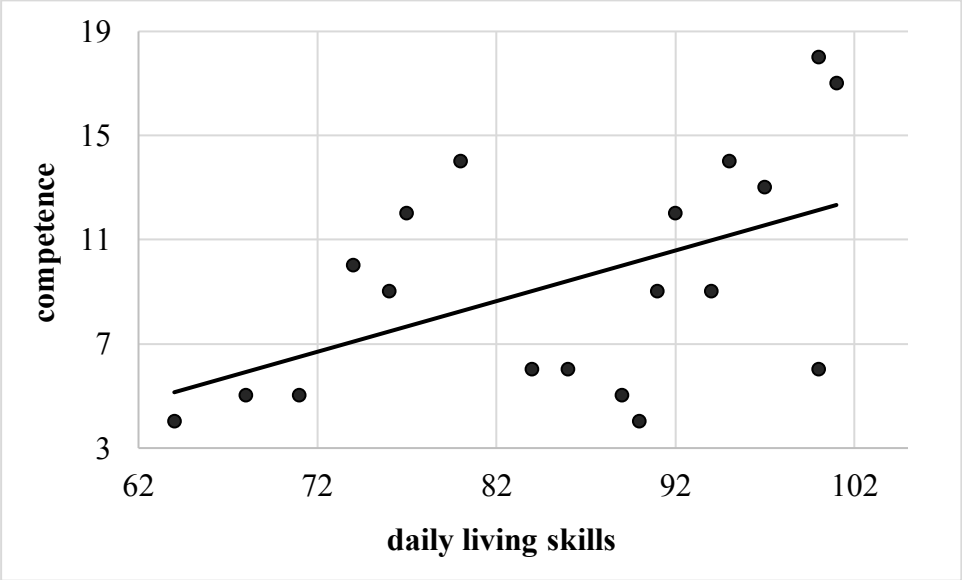

Figure 2.3. **Regression of perceived competence and child's daily living skills**. Parents who reported their children to exhibit better daily living skills also reported higher levels of competence.

Adding to that, daily living skills also show a medium to large effect size with parenting partner / spouse relationship approaching significance (r = -.426, p = .078).

ASD symptomology has unmistakeably been shown to be related to parental competence with an extremely high effect size (r= .685, p = .001) in a way that suggests that the more ASD symptoms a child exhibits, the lower the perceived competence score of the parents becomes (see Figure 2.4).

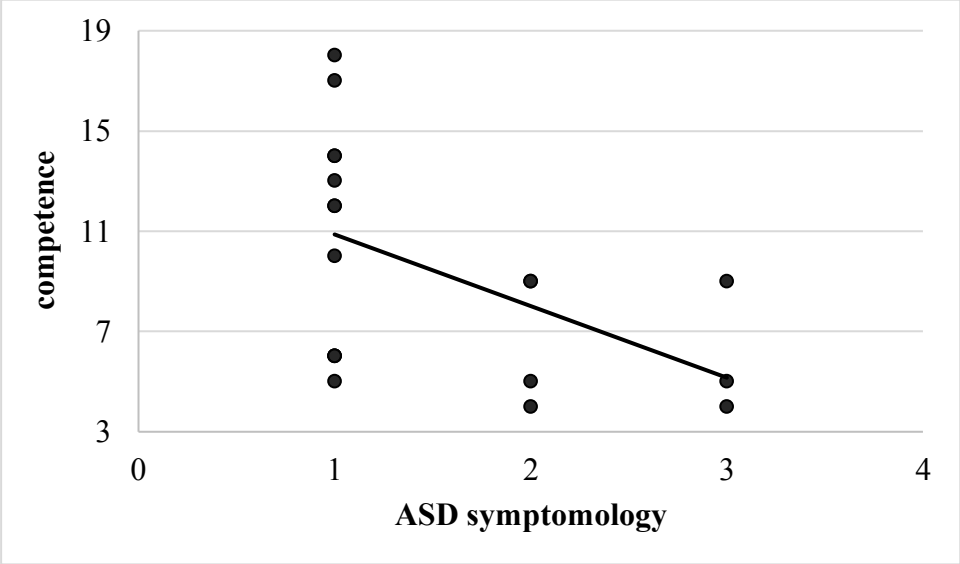

Figure 2.4. **Regression of perceived competence and child's ASD symptomology.** Parents who reported their children to exhibit more ASD-specific symptoms also reported a decline in perceived competence. ASD symptomology: group 1 = within typical limits, group 2 = slightly noticeable symptomology, group 3 = noticeable symptomology.

Furthermore, ASD symptomology has been significantly associated with parenting partner / spouse relationship with a large effect size (.508, p = .031). This result illustrates the parenting partner relationship quality to be worse with an increase of child's ASD symptomology (see Figure 2.5).

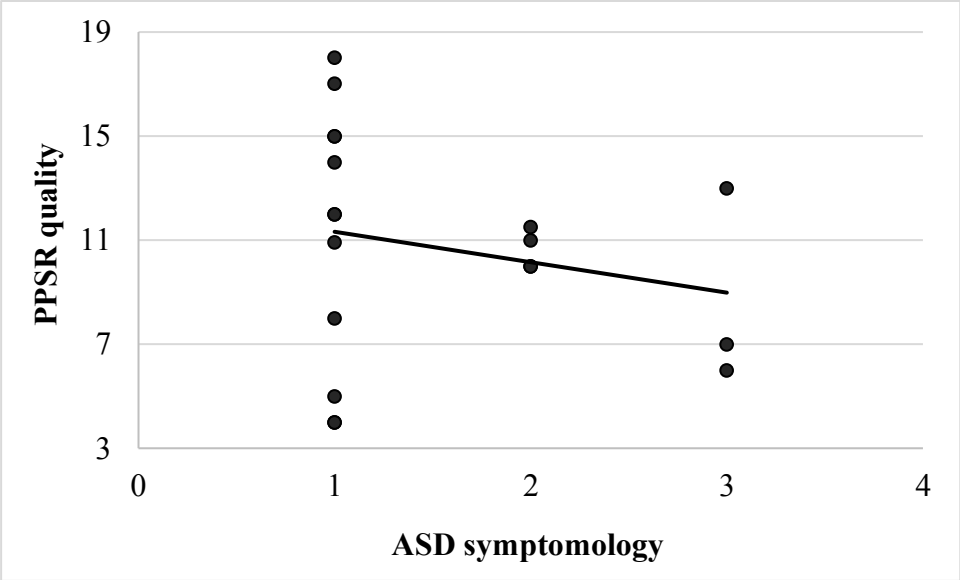

Figure 2.5. **Regression of PPSR quality and child's ASD symptomology.** Parents who reported their children to exhibit more ASD-specific symptoms also reported a decline in parenting partner / spouse relationship quality. ASD symptomology: group 1 = within typical limits, group 2 = slightly noticeable symptomology, group 3 = noticeable symptomology.

Approaching significance was the result of the relationship between ASD symptomology and parental health with a medium to large effect size ($r = .414$, $p = .078$). This relationship suggests increased ASD symptomology exhibited by the child to be associated with a decrease in parental health.

Main analysis

Multiple linear regression analysis: Health.

Hypotheses 2(a), 2(b), 2(c), and 2(d) predicted higher communicative, social, and daily living skills to negatively, and for higher ASD symptomology to positively impact the relevant subscales for parental strain.

For the main analysis, backwards stepwise regression has been used to create the models predicting parental strain outcomes health, competence, parenting partner / spouse relationship, and depression. Included predictors have been selected by correlative relationships previously established. Those include communication, socialization, daily living skills, ASD symptomology, child's gender, child's age, and family income.

The value of R^2 at step 1 of the regression for the outcome health was .345, which means that 34.5% of variability in health is accounted for by all seven predictors. At step 2, child's gender has been excluded, which did alter the result to an explained variance of

34.4%. At step 3, social skills have been excluded, leaving 33.5% (R^2 = .335) of variance accounted for. At step 4, ASD symptomology has been excluded, leaving 32.5% variance in parental health. At step 5, daily living skills have been excluded, leaving 30.5%. At step 6, child's age has been removed from the model, leaving 27.4% of variance accounted for by communicative skills and family income. At the last step, communicative skills have been excluded, leaving 20.5% variance accounted for by family income (see Table 7). Leaving communicative skills in the model would improve it by 7.0%.

Adjusted R^2 has been calculated to estimate the amount of explained variance for the population. For this model the adjusted R^2 is .158. In the population, 15.8% would be accounted for by family income and ASD symptomology.

The change in the amount of variance causes an F-ratio of 1.536 which is not significant (p = .233).

A Durbin Watson test was requested to control if the assumptions of the independent errors have been plausible. As a conservative rule, values less than 1 and over 3 should be alarming. For these data the value is 2.597, which is within the limits, so the assumptions have likely been met.

Finally, an ANOVA has been produced to test whether the model is significantly better at predicting the outcome than using the mean. None of the steps came in under the cut-off of .05 significance, although step 6 did approach significance (p = .052).

Multiple linear regression analysis: Competence.

The value of R^2 at step 1 of the regression for the outcome competence was .646, which means that 64.6% of variability in competence is accounted for by all seven predictors. At step 2, family income has been excluded, which did not alter the result by much (R^2 = .645). At step 3, child's gender has been excluded, leaving 64.1% of variance in perceived competence accounted for. At fourth step, communicative skills have been removed from the model, leaving 63.4% or variance accounted for. At the final step, social skills have been excluded, leaving 60.8% of variance in competence accounted for by child's age, ASD symptomology, and daily living skills. All three predictors have been accepted into the final model (see Table 7).

Adjusted R^2 for this model is .529. In the population, 52.9% would be accounted for by child's age, ASD symptomology, and daily living skills. The change in the amount of variance causes an F-ratio of .989, which is not significant (p = .337).

The Durbin Watson test revealed the value of 2.188, which is within the limits, so the assumptions have likely been met.

The ANOVA clearly reported significance from step 2 ($p = .027$) to step 5 ($p = .002$). This suggests that using the model predicts the outcome competence significantly better than using the mean.

Multiple linear regression analysis: Parenting partner / spouse relationship.

The value of R^2 at step 1 of the regression for the outcome parenting partner / spouse relationship was .409, which means that 40.9% of variability in competence is accounted for by all seven predictors. At step 2, communicative skills have been excluded, which did not alter the result by much ($R^2 = .406$). At step 3, ASD symptomology has been excluded, leaving 40.6% of variance in parenting partner / spouse relationship accounted for. At the fourth step, family income been excluded, leaving 38.7% variance. At the fifth step, social skills have been removed from the model ($R^2 = .357$). At step 6, child's gender has been removed, leaving 22.5% of variance in parenting partner / spouse relationship explained by child's age and daily living skills. At step 6, daily living skills have been removed, leaving 18.4% of variance explained by child's age (see Table 7). Including the child's daily living skills would improve the model by 4.1%.

Adjusted R^2 for this model is .133. In the population 13.3% of the variance in parenting partner / spouse relationship would be accounted for by ASD symptomology. The change in the amount of variance from seven to one predictor causes an F-ratio of 2.915 which is not significant ($p = .108$).

The Durbin Watson test revealed the value of 1.576, which is within the limits, so the assumptions have likely been met.

In the ANOVA, the fifth step, including daily living skills and ASD symptomology approached significance ($p = .058$) even better than the sixth step, including only ASD symptomology ($p = .076$).

Multiple linear regression analysis: Depression.

The value of R^2 at step 1 of the regression for the outcome health was .468 (see Table 7), which means that 46.8% of variability in competence is accounted for by all seven predictors. At step 2, daily living skills have been excluded, which did not alter the result. At step 3, gender of the child has been excluded, leaving 45.7% ($R^2 = .457$) of variance accounted for. At step 4, family income has been removed from the model, leaving 42.6%

explained variance. At step 5, communicative skills have been removed, leaving 38.8% variance. At step 6, social skills have been excluded, leaving 36.5% of variance. In the last step, all predictors except child's age have been removed for not meeting the inclusion criterion (Probability of F-to-remove >= .100). Child's age accounted for 27.3% of variance in parental depression. Leaving social skills in the model would improve the value of the explained variance by 9.2% (R^2 change = .092).

Adjusted R^2 for this model is .230. In the population, 23.0% would be accounted for by child's age.

The change in the amount of variance causes an F-ratio of 2.311 which is not significant (p = .148).

The Durbin Watson test calculated a value of 2.410 for these data, which is within the limits, so the assumptions have likely been met.

Significant results have been produced for the ANOVA at step 6 (p = .026), in which social skills and child's age have been included in the model. Without social skills, the p-value was further reduced to .022.

To test if multicollinearity between the predictors did influence the regression of my model in a noteworthy way, variance inflation factor (VIF) values have been tested. For all steps and predictors, the VIF was smaller than 10 (range 1.000 – 2.531). Multicollinearity between the predictors did not likely influence the model fit.

The final model did include child's age, family income, daily living skills, and ASD symptomology as predictors meeting inclusion criteria for the explained variance in parental strain dimensions health, competence, parenting partner / spouse relationship, and depression (see Table 7, Figure 3).

The adaptive behaviours communication and socialization have been excluded from the model. Therefore, hypotheses 2(a) and 2(b) could not be confirmed, while hypotheses 2(c) and 2(d) were partly confirmed.

Table 7

Final multiple linear regression model for parental strain: R^2

	HEA	COMP	PPSR	DEP
CA			.184	.273
FI	.205			
DLS + ASD + CA		.608		
all predictors[a]	.345	.646	.409	.468

Note. Predictors left in the final model are presented with their explained variance (R^2) in the outcome. HEA = health, COMP = competence, PPSR = parenting partner / spouse relationship, DEP = depression, DLS = daily living skills, ASD = autism spectrum symptomology, CA = child's age, FI = family income. [a] all predictors include communication, socialization, daily living skills, ASD symptomology, child's age, child's gender, and family income.

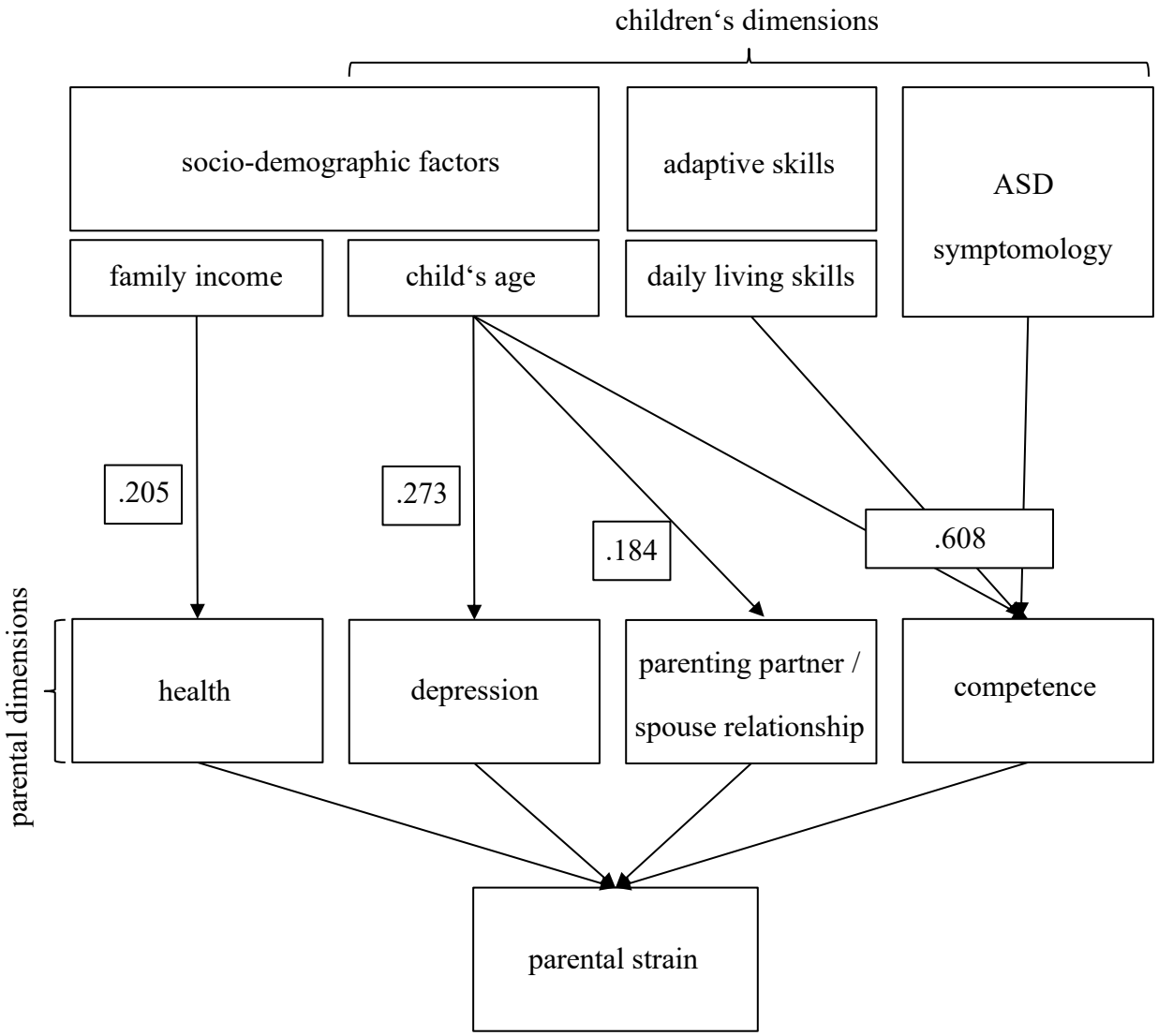

Figure 3. **Final multiple linear regression model with predictors for parental strain.** Socio-demographic factors include family income and child's age. Children's dimensions include child's age, adaptive skills, and ASD symptomology. Parental dimensions include health, depression, parenting partner / spouse relationship, and competence. Connections between socio-demographic factors, adaptive skills, and ASD symptomology and the parental dimensions are illustrated by their respective R^2-values (amount of variance accounted for by the predictor).

Limitations and implications for future research

A possible limitation of this study is the sample size which may be too small to find small effects. Through the reduction of the parental dimensions to only highly salient factors (health, competence, parenting partner / spouse relationship, depression) and the reduction of predictors through the analysis of the correlative relationships beforehand (leaving child's age, family income, communication, socialization, daily living skills, and ASD

symptomology for the main analysis), medium to large effects have been expected; ensuring a sufficient sample size.

In the current sample I only included families with children with DS, so generalizability for other IDDs is not guaranteed. Nevertheless, other studies frequently compare psycho-social effects of having a child with DS with other IDDs, as a result of factors outside the family like discrimination and stigmatization (e.g. Farkas et al., 2019; Krueger et al., 2019).

The lack of a TD control group could be perceived by some readers as a limitation. Through the inclusion of ASD symptomology as a predictor the influence of high and low maladaptive behaviours should have been accounted for. For this reason and since Eisenhower et al. (2005) reported children with DS to show similarly low levels of maladaptive behaviour as TD children, I refrained from the use of a TD control group but it is definitely recommended for future research to gain more valid results.

The author is sensitive to the fact that another limitation of this study is the possibility of a systematic discrepancy between the participants in this sample and families who chose not to participate or have not been reached by the team's recruiting endeavours.

Also, the fact that the current sample consists of mainly highly educated mothers speaks of a particularly selective group. The team's recruiting technique was not directed at especially high socio-economic groups but recruitment in centres for families who seek counselling could be viewed as selective for people who already have the resources to know where to get help. Therefore, future research should investigate if persons who completed higher education are more likely to take part in these kinds of studies.

Recruitment for this study did also not specify if the mother, the father, or both should fill out the questionnaires. In fact, in the majority of cases only the mother accompanied their child to the appointment at the research centre. Previous research suggests that mothers and fathers differ in their experience of parental strain with a child with IDD and their need for support interventions (e.g. Dabrowska & Pisula, 2010; Hedov et al., 2002; Kózka & Przybyła-Basista, 2018; Marchal et al., 2017; Marsh, Brown, & McCann, 2020; Rafferty et al., 2020). As the bulk of studies before this, my data at hand relies on mothers reports only. Future research should take into account possible differences between the parents and include both parents' experiences.

Some researchers discussed further unique dimensions influencing parental strain with children with DS like complex medical treatments because of comorbid physical conditions (e.g. Bush et al., 2020; Siegel & Smith, 2010), interactions with medical personnel, and the

ambiguous loss theory (c.f. Boss, 2007; Farkas et al., 2019), which are not accounted for in this investigation since this research concentrates on children's factors.

Also, as in most research, this study largely focusses on the negative outcomes on the parental dimensions. To gain a more comprehensive understanding of the situations for parents, future research should take into account positive effects of raising a child with DS or more generally IDD; possibly building on positive psychology theories.

Discussion

To the best of my knowledge to date, there is no study investigating the relationship of adaptive behaviour in children with DS and parental strain while also including ASD symptomology. The aim of this investigation was to isolate strong predictors in children's dimensions related to parental strain; mainly parental physical and mental health, parenting partner / spouse relationship, and competence. In brief, the results of the main analysis suggest a couple of socio-demographic and psycho-social predictors of child behaviour to be relevant to parental strain.

The remaining socio-demographic predictors in the final model were child's age and family income. The psycho-social predictors of child behaviours were daily living skills and ASD symptomology. Socio-demographic factors like mother's age, mother's education, or number of siblings did not show significant relationships with parental strain, which is in line with literature on children with DS and / or TD children, that did not find any socio-demographic effect on parental strain (e.g. Broadhead et al., 2009; Marchal et al., 2013; Pastor-Cerezuela et al., 2020). Contrary to larger studies that reported a lack of involvement of these variables, the current investigation found socio-demographic variables like child's age, child's gender, and family income to be related to some degree to parental strain.

Correlative relationships and results of the main multiple linear regression analysis between the socio-demographic and psycho-social predictors and parental strain subscales are discussed.

Family income did play a role in parental strain as parents with more financial resources reported better physical health. The final regression model did report family income to be the best predictor of variance in physical health with 20.5% accounted for. A possible explanation for this result would be that families with higher income also have more and better access to health promoting resources. These resources seem to be more important to the parents' physical health than any child-related factors. Taking this into account would mean that improving the access to health promoting resources for lower-income families would

consequently also positively impact their perceived parenting strain (e.g. Mugno et al., 2007). No other predictors having a noteworthy relationship with parental health is in line with research by Hedov, Wikblad, and Annerén (2006), who did not find any effect of having a child with DS on parental physical health as compared with a control group of TD children. In light of the results by Stoneman (2007) that did find family income to be related to parental strain, the current result of family income makes sense.

With more independence, meaning higher daily living skills, seem to come an enhancement in parental physical health during the correlative analysis, suggesting that if parents have to invest less time in the daily physical care of their children, they benefit by being healthier in the long run. A possible explanation could be that parents have more time to take care of themselves if their children are more independent. An additional possible positive effect with increasing the child's independence from their parents' daily care would be an improvement in parents' perceived competence. These results from the correlative and main analysis are in line with previous research reporting increased strain in parents who did have to invest more hours per day in the care of their child (e.g. Hedov et al., 2002; Marchal et al., 2013, 2017).

Child's gender was found to be related to parental perceived competence, their marital / partner relationship quality, and depression in the correlative analysis. Having a boy with DS was associated with lower competence, lower relationship quality, and more depressive symptoms during the analysis of correlative values. This result seems to contradict research finding a gender effect on parental strain in favour of having a boy (Dickinson & Place, 2016; Marquis et al., 2020). The result of this study is rather comparable to the study by Senses Dinc et al. (2019) on infants and toddlers, reporting a positive effect on mother's health related quality of life with having a girl. Possible explanations of the current result in favour of girls could be that girls generally are four times less likely to meet diagnostic criteria for ASD (Baio et al., 2018) and therefore show less maladaptive behaviours related to ASD symptomology. This finding has – on a much lower scale – seemingly been replicated in the current study, with 55.5% of boys exhibiting symptoms of ASD above the cut-off opposed to only 10% of girls showing slightly noticeable ASD-specific behaviours. Nevertheless, the sample in this study was too small to be interpreted as a serious replication of the results by Baio et al. (2018).

Also, a lack of social skills has been found to be related to a decline in perceived competence in the correlative analysis. As children with DS have often been described as socially competent (e.g. Corrice & Glidden, 2009; Farkas et al., 2019; Skotko et al., 2016) and

children with ASD were described as having problems with social interactions (e.g. Hayes & Watson, 2013; Lecavalier et al., 2006), a possible ASD-specific effect on parental strain should hence forth be more thoroughly investigated. According to the model derived from the current data, enhancing children's social skills in a support intervention program suitable for the specific IDD should increase parent's competence and therefore relieve their experienced strain.

Child's age was very clearly related to parents' depression outcomes. Discrepancies in the parent-reported depressive symptoms have been best explained by child's age in the final multiple linear regression model. With having an older child also came an increase of depressive symptomology reported by parents. Also, having an older child with DS was associated with lower parenting partner / spouse relationship quality. In the final model, parenting partner / spouse relationship was best predicted by child's age, as it did account for 18.4% of the variance in this parental strain outcome. Having an older child seems to be partly responsible for a decline in the parental relationship quality. This finding is in line with previous research suggesting that having an older child increases parental strain because of disruptions in the parent-child interactions when children enter into puberty (e.g. Dickinson & Place, 2016). Dickinson and Place (2016) and Nærland et al. (2017) found the observed age-related effects to be associated with ASD symptomology. Future research should therefore control for ASD symptomology or incorporate a non-DS, ASD control group.

ASD symptomology was found to be related to perceived competence in the main analysis and to some degree also to parenting partner / spouse relationship in the correlative analysis. If the children with DS displayed more symptoms of ASD, the parents reported a lack in perceived competence and a decline in their parenting partner / spouse relationship. Competence was found to be one of the pillars of parental strain during this investigation, being negatively related to having a boy and the final multiple linear regression model reporting that 60.8% of variance in competence can be explained with child's age, their daily living skills, and ASD symptomology. These results suggest that improving parental competence through IDD-specific training and education at the right time, parental stress and burden could perhaps be reduced considerably. Especially for perceived competence, previous research has suggested that intervention programmes fostering parental understanding and skill set on how to handle and communicate with their children with ASD can improve not only their parent-child relationship quality but in turn also parental quality of life (e.g. Decroocq et al., 2020).

In summary, the findings in this investigation question the *Down syndrome advantage* hypothesis (c.f. Corrice & Glidden, 2009; Farkas et al., 2019; Skotko et al., 2016). If children in this sample exhibited less ASD symptomology they also less likely showed maladaptive behaviours. The *Down syndrome advantage* could therefore be an effect of a lack in ASD symptomology rather than an effect of the DS phenotype. The field begs for more thorough research investigating ASD symptomology as a mediating factor in strain of parents with children with DS. Behaviour problems related to a highly comorbid ASD have to be thoroughly distinguished from possible behaviour problems related to DS.

Parental strain is an extremely complex topic with heterogeneous influence factors. Since parents and their children reciprocally influence each other's behaviour and development (e.g. Baker et al., 2003; Blacher & Baker, 2019; Garbarski, 2014; Mackler et al., 2015; van Steijn et al., 2014), the question of *what was first,* parental strain or child's misbehaviour (e.g. Broadhead et al., 2009), may perhaps never be fully answered. Nevertheless, interventions targeting either one angle should positively alter the other, ultimately improving the parent-child relationship quality and children's development.

Conclusion

Parents of children with DS did report more problems regarding their physical health, perceived competence, parenting partner / spouse relationship quality, and depression with having older and more physical care demanding children, lower family income, and their children exhibiting more ASD symptoms. Lower income families did report more physical health related problems. Having an older child was negatively related with parents' perceived competence, the parents' relationship quality, and the parent's depression scores. The parents' perceived competence did appear to be most influenced by child's age, ASD symptomology, and their child's independence.

Disclosure statement

The author declares no potential conflict of interest with respect to the research or authorship of this article.

References

Abbeduto, L., Seltzer, M. M., Shattuck, P., Krauss, M. W., Orsmond, G., & Murphy, M. M. (2004). Psychological well-being and coping in mothers of youths with autism, Down syndrome, or fragile X syndrome. *American Journal on Mental Retardation*, *109*(3), 237-254+271. https://doi.org/10.1352/0895-8017(2004)109<237:PWACIM>2.0.CO;2

Abidin, R. R. (1995). *Parenting Stress Index (PSI) Professional Manual* (3rd ed.). Odessa, FL: Psychological Assessment Resources.

Allen, E. G., Freeman, S. B., Druschel, C., Hobbs, C. A., O'Leary, L. A., Romitti, P. A., … Sherman, S. L. (2009). Maternal age and risk for trisomy 21 assessed by the origin of chromosome nondisjunction: A report from the Atlanta and National Down Syndrome Projects. *Human Genetics*, *125*(1), 41–52. https://doi.org/10.1007/s00439-008-0603-8

Antonarakis, S. E., Skotko, B. G., Rafii, M. S., Strydom, A., Pape, S. E., Bianchi, D. W., … Reeves, R. H. (2020). Down syndrome. *Nature Reviews Disease Primers*, *6*(1), 1–20. https://doi.org/10.1038/s41572-019-0143-7

Baio, J., Wiggins, L., Christensen, D. L., Maenner, M. J., Daniels, J., Warren, Z., … Dowling, N. F. (2018). Prevalence of Autism Spectrum Disorder Among Children Aged 8 Years - Autism and Developmental Disabilities Monitoring Network, 11 Sites, United States, 2014. *MMWR Surveill Summ.*, *67*(6). Retrieved from https://www.cdc.gov/mmwr/volumes/67/ss/pdfs/ss6706a1-H.pdf

Baker-Ericzén, M. J., Brookman-Frazee, L., & Stahmer, A. (2005). Stress levels and adaptability in parents of toddlers with and without autism spectrum disorders. *Research and Practice for Persons with Severe Disabilities*, *30*(4), 194–204. https://doi.org/10.2511/rpsd.30.4.194

Baker, B. L., McIntyre, L. L., Blacher, J., Crnic, K., Edelbrock, C., & Low, C. (2003). Pre-school children with and without developmental delay: Behaviour problems and parenting stress over time. *Journal of Intellectual Disability Research*, *47*, 217–230. https://doi.org/10.1111/j.1365-2788.2005.00691.x

Baker, J. K., Seltzer, M. M., & Greenberg, J. S. (2011). Longitudinal effects of adaptability on behavior problems and maternal depression in families of adolescents with autism. *Journal of Family Psychology*, *25*(4), 601–609. https://doi.org/10.1037/a0024409

Beaujouan, E. (2020). Latest-late fertility? Decline and resurgence of late parenthood across the low-fertility countries. *Population and Development Review*, *6*, 219–247. https://doi.org/10.1111/padr.12334

Beighton, C., & Wills, J. (2019). How parents describe the positive aspects of parenting their

child who has intellectual disabilities: A systematic review and narrative synthesis. *Journal of Applied Research in Intellectual Disabilities*, *32*(5), 1255–1279. https://doi.org/10.1111/jar.12617

Blacher, J., & Baker, B. L. (2019). Collateral effects of youth disruptive behavior disorders on mothers' psychological distress: Adolescents with autism spectrum disorder, intellectual disability, or typical development. *Journal of Autism and Developmental Disorders*, *49*(7), 2810–2821. https://doi.org/10.1007/s10803-017-3347-2

Blacher, J., Baker, B. L., & Kaladjian, A. (2013). Syndrome specificity and mother-child interactions: Examining positive and negative parenting across contexts and time. *Journal of Autism and Developmental Disorders*, *43*(4), 761–774. https://doi.org/10.1007/s10803-012-1605-x

Blacher, J., & McIntyre, L. L. (2006). Syndrome specificity and behavioural disorders in young adults with intellectual disability: Cultural differences in family impact. *Journal of Intellectual Disability Research*, *50*(3), 184–198. https://doi.org/10.1111/j.1365-2788.2005.00768.x

Boss, P. (2007). *Ambiguous loss theory: Challenges for scholars and practitioners*. *56*(2), 105–110.

Broadhead, M., Chilton, R., & Crichton, C. (2009). Understanding parental stress within the Scallywags service for children with emotional and behavioural difficulties. *Emotional and Behavioural Difficulties*, *14*(2), 101–115. https://doi.org/10.1080/13632750902921880

Bush, D., Galambos, C., & Dunbar Ivy, D. (2020). Pulmonary hypertension in children with Down syndrome. *Pediatric Pulmonology*, (July 2019), 1–9. https://doi.org/10.1002/ppul.24687

Caples, M., Martin, A. M., Dalton, C., Marsh, L., Savage, E., Knafl, G., & Van Riper, M. (2018). Adaptation and resilience in families of individuals with Down syndrome living in Ireland. *British Journal of Learning Disabilities*, *46*(3), 146–154. https://doi.org/10.1111/bld.12231

Corrice, A. M., & Glidden, L. M. (2009). The Down syndrome advantage: Fact or fiction? *American Journal of Intellectual and Developmental Disabilities*, *114*(4), 254–268. https://doi.org/10.1352/1944-7558-114.4.254

Dabrowska, A., & Pisula, E. (2010). Parenting stress and coping styles in mothers and fathers of pre-school children with autism and Down syndrome. *Journal of Intellectual Disability Research*, *54*(3), 266–280. https://doi.org/10.1111/j.1365-2788.2010.01258.x

Daseking, M., & Petermann, F. (2018). Die neuen Wechsler: WPPSI-IV und WISC-V. *Kindheit & Entwicklung*, *27*(3), 127–132. https://doi.org/10.1026/0942-5403/a000252

de Graaf, G., Buckley, F., & Skotko, B. (2019). *People living with Down syndrome in the USA: Births and population*. Retrieved from https://go.dselink.net/us-population-factsheet 1

Decroocq, C., Soulas, T., Lichtlé, J., Sankey, C., Engelberg, A., & Cappe, E. (2020). Facilitators' perspectives on a psychoeducational program for parents of an autistic child. *Autism*. https://doi.org/10.1177/1362361319899766

Dickinson, K., & Place, M. (2016). Living with children with autistic spectrum condition: Parental stress and the impact upon family functioning. *Advances in Social Sciences Research Journal*, *3*(3). https://doi.org/10.14738/assrj.33.1900

Döpfner, M., & Görtz-Dorten, A. (2017). *Diagnostik-System für psychische Störungen nach ICD-10 und DSM-5 für Kinder- und Jugendliche (DISYPS-III). [Diagnostic System for Mental Health Disorders According to ICD-10 and DSM-5 for Children and Adolescents (DISYPS-III)]*. Retrieved from Google Scholar

Dunn, K., Kinnear, D., Jahoda, A., & McConnachie, A. (2019). Mental health and well-being of fathers of children with intellectual disabilities: Systematic review and meta-analysis. *BJPsych Open*, *5*(6), 1–10. https://doi.org/10.1192/bjo.2019.75

Eisenhower, A. S., Baker, B. L., & Blacher, J. (2005). Preschool children with intellectual disability: Syndrome specificity, behaviour problems, and maternal well-being. *Journal of Intellectual Disability Research*, *49*(9), 657–671. https://doi.org/10.1111/j.1365-2788.2005.00699.x

Farkas, L., Cless, J. D., Cless, A. W., Nelson Goff, B. S., Bodine, E., & Edelman, A. (2019). The ups and downs of Down syndrome: A qualitative study of positive and negative parenting experiences. *Journal of Family Issues*, *40*(4), 518–539. https://doi.org/10.1177/0192513X18812192

Garbarski, D. (2014). The interplay between child and maternal health: Reciprocal relationships and cumulative disadvantage during childhood and adolescence. *Journal of Health and Social Behavior*, *55*(9), 91–106. https://doi.org/10.1017/CBO9781107415324.004

Gath, A., & Gumley, D. (1986). Behavior problems in retarded children with special reference to Down's syndrome. *British Journal of Psychiatry*, *149*(AUG.), 156–161. https://doi.org/10.1192/bjp.149.2.156

Gerstein, E. D., Crnic, K. A., Blacher, J., & Baker, B. L. (2009). Resilience and the course of daily parenting stress in families of young children with intellectual disabilities. *Journal of Intellectual Disability Research*, *53*(12), 981–997. https://doi.org/10.1111/j.1365-

2788.2009.01220.x

Grieco, J., Pulsifer, M., Seligsohn, K., Skotko, B., & Schwartz, A. (2015). Down syndrome: Cognitive and behavioral functioning across the lifespan. *American Journal of Medical Genetics, Part C: Seminars in Medical Genetics*, *169*(2), 135–149. https://doi.org/10.1002/ajmg.c.31439

Hagen, E. W., Mirer, A. G., Palta, M., & Peppard, P. E. (2013). The sleep-time cost of parenting: Sleep duration and sleepiness among employed parents in the Wisconsin Sleep Cohort Study. *American Journal of Epidemiology*, *177*(5), 394–401. https://doi.org/10.1093/aje/kws246

Hauser-Cram, P., Warfield, M. E., Shonkoff, J. P., Krauss, M. W., Sayer, A., & Upshur, C. C. (2001). Children with disabilities: A longitudinal study of child development and parent well-being. *Monographs of the Society for Research in Child Development*, *66*(3), i–viii, 1–114; discussion 115-26. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/11677873

Hayes, S. A., & Watson, S. L. (2013). The impact of parenting stress: A meta-analysis of studies comparing the experience of parenting stress in parents of children with and without autism spectrum disorder. *Journal of Autism and Developmental Disorders*, *43*(3), 629–642. https://doi.org/10.1007/s10803-012-1604-y

Hedov, G., Annerén, G., & Wikblad, K. (2002). Swedish parents of children with Down's syndrome. *Scandinavian Journal of Caring Sciences*, *16*(4), 424–430. https://doi.org/10.1046/j.1471-6712.2002.00109.x

Hedov, G., Wikblad, K., & Annerén, G. (2006). Sickness absence in Swedish parents of children with Down's syndrome: Relation to self-perceived health, stress and sense of coherence. *Journal of Intellectual Disability Research*, *50*(7), 546–552. https://doi.org/10.1111/j.1365-2788.2006.00810.x

Iadarola, S., Levato, L., Harrison, B., Smith, T., Lecavalier, L., Johnson, C., … Scahill, L. (2018). Teaching parents behavioral strategies for autism spectrum disorder (ASD): Effects on stress, strain, and competence. *Journal of Autism and Developmental Disorders*, *48*(4), 1031–1040. https://doi.org/10.1007/s10803-017-3339-2

Ilhan, N., Akhan, L. U., Baygut, Ş., Dalli, G. N., & Yildirim, C. (2019). The factors affecting the family burden and depression status of families of children with Down syndrome in Turkey. *Journal of Mental Health Research in Intellectual Disabilities*, *12*(3–4), 176–195. https://doi.org/10.1080/19315864.2019.1654573

Kayfitz, A. D., Gragg, M. N., & Robert Orr, R. (2010). Positive experiences of mothers and

fathers of children with autism. *Journal of Applied Research in Intellectual Disabilities*, *23*(4), 337–343. https://doi.org/10.1111/j.1468-3148.2009.00539.x

Kózka, A., & Przybyła-Basista, H. (2018). Perceived stress, ego-resiliency, and relational resources as predictors of psychological well-being in parents of children with down syndrome. *Health Psychological Report*, *6*(1), 50–59. https://doi.org/10.5114/hpr.2018.71212

Krueger, K., Cless, J. D., Dyster, M., Reves, M., Steele, R., & Goff, B. S. N. (2019). Understanding the systems, contexts, behaviors, and strategies of parents advocating for their children with down syndrome. *Intellectual and Developmental Disabilities*, *57*(2), 146–157. https://doi.org/10.1352/1934-9556-57.2.146

Lecavalier, L., Leone, S., & Wiltz, J. (2006). The impact of behaviour problems on caregiver stress in young people with autism spectrum disorders. *Journal of Intellectual Disability Research*, *50*(3), 172–183. https://doi.org/10.1111/j.1365-2788.2005.00732.x

Mackler, J. S., Kelleher, R. T., Shanahan, L., Calkins, S. D., Keane, S. P., & O'Brien, M. (2015). Parenting stress, parental reactions, and externalizing behavior from ages 4 to 10. *Journal of Marriage and Family*, *77*(2), 388–406. https://doi.org/10.1111/jomf.12163

Marchal, J. P., Maurice-Stam, H., Hatzmann, J., van Trotsenburg, A. S. P., & Grootenhuis, M. A. (2013). Health related quality of life in parents of six to eight year old children with Down syndrome. *Research in Developmental Disabilities*, *34*(11), 4239–4247. https://doi.org/10.1016/j.ridd.2013.09.011

Marchal, J. P., van Oers, H. A., Maurice-Stam, H., Grootenhuis, M. A., van Trotsenburg, A. S. P., & Haverman, L. (2017). Distress and everyday problems in Dutch mothers and fathers of young adolescents with Down syndrome. *Research in Developmental Disabilities*, *67*(May), 19–27. https://doi.org/10.1016/j.ridd.2017.05.005

Marquis, S. M., McGrail, K., & Hayes, M. V. (2020). Mental health outcomes among parents of a child who has a developmental disability: Comparing different types of developmental disability. *Disability and Health Journal*, *13*(2), 100874. https://doi.org/10.1016/j.dhjo.2019.100874

Marsh, L., Brown, M., & McCann, E. (2020). The views and experiences of fathers of children with intellectual disabilities: A systematic review of the international evidence. *Journal of Policy and Practice in Intellectual Disabilities*, *17*(1), 79–90. https://doi.org/10.1111/jppi.12328

Mayr, S., Erdfelder, E., & Buchner, A. (2017). G Power 3.1 manual. *Tutorials in Quantitative Methods for Psychology*, *76*, 1–80. https://doi.org/10.1037/0096-1523.32.4.932

Mello, C., Rivard, M., Terroux, A., & Mercier, C. (2019). Quality of life in families of young children with autism spectrum disorder. *American Journal on Intellectual and Developmental Disabilities*, *124*(6), 535–548. https://doi.org/10.1352/1944-7558-124.6.535

Mugno, D., Ruta, L., D'Arrigo, V. G., & Mazzone, L. (2007). Impairment of quality of life in parents of children and adolescents with pervasive developmental disorder. *Health and Quality of Life Outcomes*, *5*, 1–9. https://doi.org/10.1186/1477-7525-5-22

Nærland, T., Bakke, K. A., Storvik, S., Warner, G., & Howlin, P. (2017). Age and gender-related differences in emotional and behavioural problems and autistic features in children and adolescents with Down syndrome: A survey-based study of 674 individuals. *Journal of Intellectual Disability Research*, *61*(6), 594–603. https://doi.org/10.1111/jir.12342

Nomaguchi, K., & Milkie, M. A. (2020). Parenthood and well-being: A decade in review. *Journal of Marriage and Family*, *82*(1), 198–223. https://doi.org/10.1111/jomf.12646

Pastor-Cerezuela, G., Fernández-Andrés, M. I., Pérez-Molina, D., & Tijeras-Iborra, A. (2020). Parental stress and resilience in autism spectrum disorder and Down syndrome. *Journal of Family Issues*. https://doi.org/10.1177/0192513X20910192

Pisula, E., & Banasiak, A. (2019). Empowerment in Polish fathers of children with autism and Down syndrome: The role of social support and coping with stress – a preliminary report. *Journal of Intellectual Disability Research*, 1–8. https://doi.org/10.1111/jir.12681

Qu, L., Soriano, G., & Weston, R. (2006). Starting early, starting late: The health and wellbeing of mother and child. *Family Matters*, (74), 4–11.

Rafferty, D., Tidman, L., & Ekas, N. V. (2020). Parenting experiences of fathers of children with autism spectrum disorder with or without intellectual disability. *Journal of Intellectual Disability Research*, *64*(6), 463–474. https://doi.org/10.1111/jir.12728

Sarimski, K. (2020). Down-Syndrom: Auswirkungen auf die Familie aus Sicht von Müttern und Vätern. *Praxis Der Kinderpsychologie Und Kinderpsychiatrie*, *69*(3), 236–251. https://doi.org/10.13109/prkk.2020.69.3.236

Senses Dinc, G., Cop, E., Tos, T., Sari, E., & Senel, S. (2019). Mothers of 0–3-year-old children with Down syndrome: Effects on quality of life. *Pediatrics International*, *61*(9), 865–871. https://doi.org/10.1111/ped.13936

Shin, M., Besser, L. M., Kucik, J. E., Lu, C., Siffel, C., & Correa, A. (2009). Prevalence of Down syndrome among children and adolescents in 10 regions of the United States. *Pediatrics*, *124*(6), 1565–1571. https://doi.org/10.1542/peds.2009-0745

Shonkoff, J. P., Hauser-Cram, P., Wyngaarden Krauss, M., & Upshur, C. C. (1992).

Development of infants with disabilities and their families: Implications for theory and service delivery. In *Monographs of the Society for Research in Child Development* (Vol. 57). https://doi.org/10.4135/9781452286143.n496

Siegel, M. S., & Smith, W. E. (2010). Psychiatric features in children with genetic syndromes: Toward functional phenotypes. *Child and Adolescent Psychiatric Clinics of North America*, *19*(2), 229–261. https://doi.org/10.1016/j.chc.2010.02.001

Skotko, B. G., Levine, S. P., & Goldstein, R. (2011). Having a son or daughter with Down syndrome: Perspectives from mothers and fathers. *American Journal of Medical Genetics, Part A*, *155*(10), 2335–2347. https://doi.org/10.1002/ajmg.a.34293

Skotko, B. G., Levine, S. P., Macklin, E. A., & Goldstein, R. D. (2016). Family perspectives about Down syndrome. *American Journal of Medical Genetics, Part A*, *170*(4), 930–941. https://doi.org/10.1002/ajmg.a.37520

Sparrow, S. S., Cicchetti, D. V., & Saulnier, C. A. (2016). *Vineland adaptive behavior scales (Vineland-3)*. Antonio: Psychological Corporation.

Stoneman, Z. (2007). Examining the Down syndrome advantage: Mothers and fathers of young children with disabilities. *Journal of Intellectual Disability Research*, *51*(12), 1006–1017. https://doi.org/10.1111/j.1365-2788.2007.01012.x

Takataya, K., Yamazaki, Y., & Mizuno, E. (2016). Perceptions and feelings of fathers of children with Down syndrome. *Archives of Psychiatric Nursing*, *30*(5), 544–551. https://doi.org/10.1016/j.apnu.2016.04.006

Totsika, V., Hastings, R. P., Emerson, E., Lancaster, G. A., & Berridge, D. M. (2011). A population-based investigation of behavioural and emotional problems and maternal mental health: Associations with autism spectrum disorder and intellectual disability. *Journal of Child Psychology and Psychiatry and Allied Disciplines*, *52*(1), 91–99. https://doi.org/10.1111/j.1469-7610.2010.02295.x

Tröster, H. (2011). *Eltern-Belastungs-Inventar (EBI)*. Göttingen: Hogrefe.

van Steijn, D. J., Oerlemans, A. M., van Aken, M. A. G., Buitelaar, J. K., & Rommelse, N. N. J. (2014). The reciprocal relationship of ASD, ADHD, depressive symptoms and stress in parents of children with ASD and/or ADHD. *Journal of Autism and Developmental Disorders*, *44*(5), 1064–1076. https://doi.org/10.1007/s10803-013-1958-9

Vaughan, E. L., Feinn, R., Bernard, S., Brereton, M., & Kaufman, J. S. (2013). Relationships between child emotional and behavioral symptoms and caregiver strain and parenting stress. *Journal of Family Issues*, *34*(4), 534–556. https://doi.org/10.1177/0192513X12440949

Xanthopoulos, M. S., Walega, R., Xiao, R., Prasad, D., Pipan, M. M., Zemel, B. S., … Kelly, A. (2017). Caregiver-reported quality of life in youth with Down syndrome. *Journal of Pediatrics*, *189*, 98-104.e1. https://doi.org/10.1016/j.jpeds.2017.06.073

Appendix

Abstract

Objectives The incentive of this study was to describe strain in parents of children and adolescents with a Down syndrome diagnosis and to examine the impact of children's adaptive behaviours on it. Expected parental strain is a main factor in prenatal decision making in regard to the termination of a pregnancy with a Down syndrome (DS) diagnosis. For counselling parents-to-be, we need to better understand the consequences of raising a child with DS. Implications for support interventions are discussed.

Study design Parents of 6-16-year-old children and adolescents completed a parental stress index questionnaire (PSI), a questionnaire examining adaptive functioning of their children (VABS-3), and a screening instrument for comorbid mental disorders including autism spectrum disorder (DISYPS-III) during their participation in the TriO study.

Results Family income accounted for 20.5% of variance in parental health. Child's age explained a variance of 27.3% in depression, 18.4% in parenting partner / spouse relationship, and together with the child's daily living skills and autism spectrum disorder (ASD) symptomology it accounted for 60.8% of variance in competence.

Conclusions Parents of children with DS reported more problems regarding their health, competence, parenting partner / spouse relationship quality, and depression with having older, more care-dependent children, lower family income, and their children exhibiting more ASD symptoms. Lower income families did report more health-related problems. Having an older child had a negative effect on parents' competence, their relationship quality, and depression scores. Competence did appear to be most influenced by child's age, ASD symptomology, and their child's independence.

Zusammenfassung

Ziele Ziel war die Beschreibung elterlicher Belastung mit Kindern und Jugendlichen mit einer Down Syndrom (DS) Diagnose zu erforschen und den Einfluss der adaptiven Fähigkeiten und Verhaltensweisen der Kinder darauf zu erfassen. Im Hinblick auf einen möglichen Schwangerschaftsabbruch sind Befürchtungen über die zu erwartende familiäre Belastung eine der wichtigsten Einflussgrößen. Für die Schwangerschaftsberatung ist es daher essentiell, besser zu verstehen, was auf Eltern von Kindern mit DS zukommen kann. Schlussfolgerungen für Interventionen werden diskutiert.

Studiendesign Eltern von Kindern zwischen 6 und 16 Jahren bearbeiteten das Eltern-Belastungs-Inventar (EBI), einen Fragebogen zu den adaptiven Fähigkeiten und Verhaltensweisen ihrer Kinder (VABS-3) und einen Screening-Fragebogen zu komorbiden psychischen Störungen wie der Autismus Spektrum Störung (ASS) im Rahmen der umfangreicheren TriO-Studie.

Ergebnisse Das familiäre Einkommen war verantwortlich für 20.5% der Varianz in der physischen Gesundheit der Eltern. Das Alter des Kindes erklärte 27.3% der Varianz in depressiven Symptomen, 18.4% in der Qualität der Partnerbeziehung und gemeinsam mit den praktischen Fähigkeiten und dem Ausmaß der ASS, erklärte Kindesalter 60.8% der Varianz in der Domäne Kompetenz.

Schlussfolgerungen Eltern von Kindern mit DS berichteten über mehr Probleme mit ihrer physischen Gesundheit, Kompetenz, Partnerbeziehung und depressiven Symptomen, wenn sie ältere Kinder mit mehr ASS-Symptomen und weniger praktischen Fähigkeiten hatten und ihr Familieneinkommen geringer war. Familien mit geringerem Einkommen berichteten über mehr gesundheitliche Probleme. Ältere Kinder zu haben hatte einen negativen Effekt auf die elterliche Kompetenz, die Partnerbeziehung und auf depressive Symptome. Unterschiede in der elterlichen Kompetenz konnten am besten durch Kindesalter, ASS-Symptome und den praktischen Fähigkeiten des Kindes erklärt werden.

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ADHD	Attention deficit / hyperactivity disorder
ASD	Autism spectrum disorder
ASS	Autismus-Spektrum-Störung
DISYPS-III	Diagnostik-System für Psychische Störungen nach ICD-10 und DSM-5 für Kinder und Jugendliche – III (diagnostic system for mental disorders corresponding to ICD-10 and DSM-5 for children and adolescents – III)
DS	Down syndrome
EBI	Eltern-Belastungs-Inventar (German version of the Parental Stress Index, PSI)
ID	Intellectual disability
IDD	Intellectual or developmental disorder
PSI	Parental stress index
TD	Typically developed
VABS-3	Vineland Adaptive Behavior Scales 3