DET PSYKOLOGISKE FAKULTET

Receptive vocabulary, social communication and behaviour in children diagnosed with autism spectrum disorder

HOVEDOPPGAVE

Profesjonsstudiet i psykologi

Ann Heidi Båtvik

Høst 2022

Receptive vocabulary, social communication and behaviour in children diagnosed with autism spectrum disorder

Veileder:

Karsten Specht

Table of Contents

Sammendrag

Bakgrunn. Autismespekterlidelse (ASD) er en gjennomgripende utviklingsforstyrrelse karakterisert av avvikende sosial kommunikasjon og sosial interaksjon, repetitiv og stereotyp atferd og restriktive interesser. Mange med ASD har vansker med både strukturelt og pragmatisk språk og strever med sosial kommunikasjon og interaksjon. Det har blitt fremsatt hypoteser om at disse vanskene kan knyttes til forekomst av maladaptiv atferd. *Metode.* «Music for Autism» er et pågående prosjekt som sammenligner effekten av musikkterapi med leketerapi. Data om reseptivt ordforråd, sosial kommunikasjon og atferd ble innhentet ved studiens oppstart og ved regelmessige intervaller. Denne oppgaven baserer seg på data fra 15 norske og østeriske barn, innhentet ved studiens oppstart. *Resultat.* Reseptivt ordforråd korrelerer positivt med syntaks og koherens og korrelerer negativt med sosial kognisjon. Tale korrelerer negativt med sosial bevissthet og sosial kommunikasjon og korrelerer positivt med barn og ungdoms deltakelse i sosiale kontekster. *Konklusjon.* Det ble funnet begrenset bevis for et forhold mellom språk, kommunikasjon og atferd. De identifiserte korrelasjonene synes å delvis kunne forklares ved at studien bruker instrumenter som måler lignende konsept. Det er behov for flere studier for å få en dypere forståelse for forholdet mellom språk, sosial kommunikasjon og atferd. Økt kunnskap om temaet kan ha implikasjoner for intervensjoner rettet mot å forbedre livskvalitet hos barn og voksne med ASD.

Nøkkelord: Autismespekterforstyrrelse, språkforståelse, sosial kommunikasjon, adaptiv atferd, maladaptiv atferd

Receptive vocabulary, social communication and behaviour in children diagnosed with autism spectrum disorder

Abstract

Background. Autism Spectrum Disorder (ASD) is a pervasive neurodevelopmental disorder characterized by deficits in social communication and interaction, repetitive and stereotypical behaviour, and restricted interests. Many with ASD have deficits in structural and functional language and struggle with social interaction and communication. It has been hypothesised that these abnormalities in social functioning could be linked to maladaptive behaviour and reduced quality of life. *Methodology.* Music for Autism is an ongoing project comparing the effect of music therapy to a structurally matched play-therapy intervention. Data relating to receptive vocabulary, social communication and behavioural outcomes was collected at baseline and at regular follow-ups. The present paper is based on data collected at baseline from 15 Norwegian and Austrian children. *Results.* Receptive vocabulary seems to positively correlate with syntax and coherence and negatively correlate with social cognition. Further, spoken language negatively correlates with social awareness and social communication and positively correlates with child and adolescent participation in social settings. *Conclusion.* Limited evidence was found linking language and communication to behaviour, and the identified correlations seem to be partly due to the use of instruments measuring similar concepts. Further studies are needed to gain a deeper understanding of the link between language, social communication, and behaviour. This could have implications for interventions aimed at improving the quality of life of children and adults with ASD.

Keywords: Autism spectrum disorder, receptive language, social communication, adaptive behaviour, maladaptive behaviour

Abbreviations

ABA: Applied behaviour analysis.

ASD: Autism spectrum disorder.

CASP: Child and Adolescent Scale of Participation.

CCC-2: Children's Communication Checklist, version 2.

GCI: General communication index of the CCC-2.

DSM: Diagnostic and Statistical Manual of Mental Disorders.

FQoL: Family Quality of Life Scale.

HFASD: High-functioning autism spectrum disorder.

ICD: International Classification of Disease.

LFASD: Low-functioning autism spectrum disorder.

MBI: Maladaptive Behaviour Index.

MT: Music therapy.

M4A: Music for autism.

PPVT-4: Peabody Picture Vocabulary Test, version 4.

SLI: Specific language impairment.

SRS: Social responsiveness scale.

VABS: Vineland Adaptive Behaviour Scales.

Introduction

Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder characterised by repetitive and stereotypical behaviour and deficits in social communication (2017). Symptoms of the disorder are typically present from an early age, and although the symptoms' manifestation can change throughout development, it is considered a life-long condition. The severity of the symptoms varies from person to person; some are able to live independently with little to no support from others, whereas many require substantial support and help in all areas of life.

The term autism was first used by Swiss psychiatrist Eugen Bleuler in 1908 (van der Gaag, 2017). He used the term autism to refer to a sub-class of schizophrenic patients showing symptoms of withdrawal from social contact. Later, Austrian-American psychiatrist Leo Kanner published the influential article "Autistic Disturbances of Affective Contact", where he described 11 children exhibiting symptoms of stereotypical and repetitive behaviour, social communication deficits and various degrees of language deficits (Kanner, 1943). Kanner further identified common manifestations of these core symptoms and common comorbidities, such as echolalia, idiosyncratic language, unusual use of grammar, desire for sameness and routine, and sensory sensitivities. He hypothesised that autism was caused by "[…] an innate inability to form the usual, biologically provided affective contact with people […]" (p. 250). However, he also states that environmental factors, such as parenting style and child-parent attachment, could be of relevance. Hans Asperger described similar traits in a sample of Austrian boys, whom he referred to as 'autistic psychopaths' (Asperger, 1944).

Since the original observations and descriptions, our understanding of ASD has changed. It is no longer understood as a sub-type of schizophrenia, but rather as a complex neurodevelopmental disorder affecting several psychological and cognitive abilities. ASD is diagnosed using the International Classification of Disease (ICD) (World Health Organization (WHO), 1993) or the Diagnostic and Statistical Manual of Mental Disorders (DSM) (American Psychiatric Association, 1994). In previous editions of the diagnostic manuals (ICD-10 and DSM-IV), ASD was divided into several distinct diagnostic categories, such as childhood autism, atypical autism, and Asperger syndrome. In the current editions of the diagnostic manuals (ICD-11 and DSM-5) the categorical understanding of ASD has been replaced with a view of ASD as being a continuous spectrum. Although decades of research have improved our understanding of ASD, we still don't have a clear picture of the disorders' cause and why there is such variation in how the symptoms present themselves. As our knowledge about ASD increases, so does our ability to provide adequate and evidence-based support for affected persons and their caregivers.

Epidemiology

Epidemiological studies estimate the prevalence of ASD to be about 1.5% (Lyall et al., 2017). The prevalence has increased in recent years, most likely due to changes in diagnostic criteria, increased awareness in the general population and among professionals, and improved access to mental health care services. One review of epidemiological studies published between 1966 and 1998 (Fombonne, 1999) found that the reported prevalence of ASD increased with publication year. The increase was primarily among those without comorbid intellectual disability (Lyall et al., 2017). In Norway the rates of autism in children are slightly lower than those reported in other countries, where 0.7% of children receive a diagnosis before their 8th birthday (Suren, 2006). However, the lifetime prevalence is estimated to be higher, approximately between 1% and 1.5%.

The prevalence of ASD is higher in boys than in girls, with as many as 4 boys for every 1 girl (Masi et al., 2017). There are numerous theories attempting to explain this. The "extreme male brain" theory, postulated by Baron-Cohen et al. (2002) suggests that a typical male cognitive profile allows the male to be better at systemising than emphasising, and that ASD is an extreme variant of the normal male profile. One proposed mechanism for this is increased exposure to androgens during the foetal period. An alternative explanation is the "female protective model" which hypothesises that women are more resilient to genetic mutations that cause neurodevelopmental disorders, and thus more risk factors need to be present before women develop ASD (Jacquemont et al., 2014). Psychosocial theories, however, claim that the gender difference in diagnostics is due to gender bias in diagnostic criteria, and that women are under-diagnosed. Guastella (2017) claims that women with autism tend to have lower global functioning, thus having more severe deficits in social communication and interaction, poorer language abilities, and increased maladaptive behaviour. This could suggest that females require more severe symptoms in order to be diagnosed with ASD.

The aetiology of ASD is unknown, but is hypothesised to be caused by a mix between genetic predisposition and environmental influences (Masi et al., 2017). Epidemiological studies have found that in 5.5% of cases, the diagnosis of ASD could be caused by Fragile X syndrome, tuberous sclerosis, Downs syndrome, cerebral palsy, phenylketonuria, neurofibromatosis or congenital rubella (Carr, 2015). Some identified environmental risk factors are high parental age, low interpregnancy interval, maternal diet, lifestyle and use of medication, exposure to environmental chemicals and obstetric complications and traumatic birth (Carr, 2015; Lyall et al., 2017).

Core symptoms

Autism Spectrum Disorder is diagnosed using the ICD-11 (World Health Organization (WHO), 2019) or the DSM-5 (American Psychiatric Association, 2013). Both manuals present two core symptoms of ASD: 1) Persistent deficits in social communication and social interaction and 2) restricted and repetitive patterns of behaviour, interests, and activities. In addition, the symptoms must have been present from an early age and the symptoms must cause impairment in functioning. Table 1 provides the diagnostic criteria in each manual, before the subsequent section provides a brief review of available research on the core symptoms of ASD. (Abrahams & Geschwind, 2008)

Table 1

Diagnostic criteria in the DSM-5 and the ICD-11

	DSM-5: 299	**ICD-11: 6A02**
Core symptoms	A. Persistent deficits in social co-communication and social interaction across multiple contexts.	Persistent deficits in initiating and sustaining social communication and reciprocal social interactions that are outside the expected range of typical functioning given the individual's age and level of intellectual ability, and disorder severity.
	B. Restricted, repetitive patterns of behaviour, interests, or activities.	Persistent restricted, repetitive, and inflexible patterns of behaviour, interests, or activities that are clearly atypical or excessive for the individual's age and sociocultural context.

	C. Symptoms must be present in the early developmental period (but may not become fully manifest until social demands exceed limited capacities or may be masked by learned strategies in later life).	The onset of the disorder occurs during the developmental period, typically in early childhood, but characteristic symptoms may not become fully manifest until later, when social demands exceed limited capacities.
	D. Symptoms cause clinically significant impairment in social, occupational, or other important areas of current functioning.	The symptoms result in significant impairment in personal, family, social, educational, occupational, or other important areas of functioning. Some individuals with autism spectrum disorder are able to function adequately in many contexts through exceptional effort, such that their deficits may not be apparent to others. A diagnosis of autism spectrum disorder is still appropriate in such cases.
	E. These disturbances are not better explained by intellectual disability (intellectual developmental disorder) or global developmental delay. Intellectual disability and autism spectrum disorder frequently co-occur; to make comorbid diagnoses of autism spectrum disorder and intellectual disability, social communication should be below	

	that expected for general developmental level.	
Specify if	With or without accompanying intellectual impairment.	Co-occurring disorder of intellectual development. ▪ With disorder of intellectual development. ▪ Without disorder of intellectual development.
	With or without accompanying language impairment.	Degree of functional language impairment. ▪ With mild or no impairment in functional language. ▪ With impaired functional language. ▪ With complete, or almost complete, absence of functional language.
	Associated with a known medical or genetic condition or environmental factor.	Loss of previously acquired skills. ▪ With loss of previously acquired skills. ▪ Without loss of previously acquired skills.
	Associated with another neurodevelopmental, mental, or behavioural disorder.	
	With catatonia.	

Repetitive and stereotypical behaviour and interests

One core feature of ASD is repetitive and stereotypical behaviour and interests. Some examples are stimming, narrow interests, abnormal use of toys and objects, need for routine and predictability, and echolalia. One can distinguish between high-level and low-level repetitive behaviour. High-level repetitive behaviour refers to narrow interests and obsessions, rigid needs for routine, troubles adjusting to change, and unusual attachment to objects, whereas low-level repetitive behaviours refer to repetitive movements and tics (Mooney et al., 2006). Such high-level repetitive behaviours have been found to be predictive of a later diagnosis of autism. Lower-level repetitive behaviours, on the other hand, seem to be common in developmentally delayed children in general (Mooney et al., 2006). One extreme variant of repetitive behaviour is self-harm. In children with autism this can manifest as self-biting, self-hitting, or hitting one's head against the wall (Nordahl Hansen, 2015). Such self-injurious behaviour has been identified in as many as 50% of children with autism, and the prevalence seems to vary depending on IQ and receptive and expressive language abilities (McClintock et al., 2003; Richards et al., 2012).

Deficits in social communication and language development

Abnormal social communication is usually one of the first symptoms parents of autistic children notice (De Giacomo & Fombonne, 1998). Some examples of abnormal social communication are lack of response to one's own name, reduced use of communication, delays in language development and a lack of joint attention. Abnormal development of these skills can in some instances be observed already in the child's first year of life (Mitchell et al., 2006). Some studies have found correlations between social communication and language development, where joint attention and imitation seem to be important skills needed for further language development (Charman et al., 2003; Luyster et al., 2008; Smith et al., 2007).

As many as 50% do not develop functional language skills, but about 25% show normal language skills (Lord & Jones, 2012). Delays in language development are not specific to ASD, but also appear in other neurodevelopmental disorders. One issue with assessing language abilities in children with autism is that standardised tests are not sensitive enough to determine language abilities in severely delayed children. Alternative tests have been developed to meet this need, such as the Peabody Picture Vocabulary Test (PPVT), a measure of receptive vocabulary that does not require verbal language. Tests such as the PPVT has yielded useful information about the language development of children with ASD. Two studies have identified lower PPVT scores in children with ASD compared to their typically developing peers (Brignell et al., 2019; Pellicano, 2010). In a sample of 69 children with ASD, Thomas (2009) found a mean score of 70.84 on the PPVT-III, whereas Pellicano (2010) found a mean score of 97.08 in a sample of 45 children with ASD. Some studies suggest that children with ASD perform better on tests of expressive vocabulary than on tests of receptive vocabulary. In typically developing children the pattern is opposite, with children performing better on tests of receptive vocabulary compared to tests of expressive vocabulary (Brignell et al., 2019; Kover et al., 2013). However, this discrepancy between receptive and expressive vocabulary has been found to be partially explainable through variation in attention towards the speaker (McDaniel et al., 2018).

Furthermore, studies have identified both structural and pragmatic language deficits in children with ASD (Geurts & Embrechts, 2008; Rapin & Dunn, 2003; Reindal et al., 2021; Volden et al., 2009). Structural language refers to phonology, morphology, syntax, and semantics, whereas pragmatic language refers to appropriate use of language in social settings. One study by Reindal et al. (2021) found that a sample of children diagnosed with ASD showed both structural and pragmatic language deficits, and that structural language deficits were associated with reduced pragmatic language skills. The same study found that syntax seems to be the least affected structural language skill in children with ASD, whereas coherence is most affected. Among pragmatic skills, non-verbal communication seemed to be the most affected. Similarly, Boucher (2012) identified syntax and articulation as the least affected skills and semantics and morphology as most affected. However, Rapin & Dunn (2003) suggest that structural language may improve by school age, whereas pragmatic language deficits remain.

To compensate for lack of functional verbal language, variations of alternative communication have been developed to allow children with ASD to express themselves. Commonly used forms of alternative communication are sign language, simplified sign language, communication books and apps, and printed symbols and photographs that the child can point to (Mirenda, 2003). It is theorised that lack of functional language can be one cause of maladaptive behaviour in children with ASD. Miscommunication or misunderstandings are thought to trigger maladaptive behaviour, as the non-verbal child is lacking the language to express themselves. Thus, their behaviour can be understood as a way of accomplishing a goal (Walker et al., 2021). As a consequence, interventions that target maladaptive behaviour could focus on providing the child with methods of alternative communication. Furthermore, children with autism could benefit from early interventions aimed at skills such as imitation, joint attention and play, as these skills predict future development (Toth et al., 2006; Warreyn et al., 2014). Early intervention is key, as studies have shown that the majority of language development happen in the child’s early years (Sigman & McGovern, 2005). Some also propose that there is a critical period of language learning, and if language is not acquired in this time period, it cannot be learned at all (Kennison, 2014, p. 15).

Comorbidity

There is a high prevalence of comorbid neurological, somatic, and psychiatric conditions in ASD.

Epilepsy

There is a high prevalence of epilepsy in individuals with ASD (Bauman, 2010; Levisohn, 2007; Mannion et al., 2013). Levisohn et al. (2007) estimates that 5-38% of children with autism also suffer from epilepsy, and that the risk increases if the child also suffers from an intellectual disability or cerebral palsy. EEG abnormalities have also been described in autistic individuals without a history of epilepsy. Furthermore, Levisohn et al. (2007) identified one peak risk period for the development of epilepsy during childhood and a second peak in the early adolescent year. Similar peak risk periods were identified by Bauman (2010). It is however unclear if ASD and epilepsy share an underlying aetiology, if epileptic seizures early in life can cause the symptoms associated with autism, or if other factors can explain this relationship (Scott & Tuchman, 2016). One Swedish population-based cohort-study (Sundelin et al., 2016) however, found that individuals had an increased risk of developing ASD if they had epilepsy in childhood. Furthermore, they identified an increased prevalence of ASD in siblings and children of individuals diagnosed with ASD.

Intellectual disability

ASD and intellectual disabilities frequently co-occur, and one estimates that as many as 70% of individuals with ASD also have an intellectual disability (La Malfa et al., 2004; Lord & Jones, 2012; Mannion et al., 2013). Due to the high comorbidity, it has been hypothesised that ASD and intellectual disabilities could have the same genetic aetiology (Matson & Shoemaker, 2009). The proportion of children with ASD who has and IQ over 70 has increased in recent years, possibly due to changes in diagnostic criteria, changes in access to health care services, and increased knowledge of how ASD manifests in people of average intellectual ability (Lord & Jones, 2012). Furthermore, there seems to be a correlation between IQ and severity of symptoms. Lord & Jones (2012) postulates that children and adolescents with average intellect are able to compensate for deficits in social communication. Similarly, O'Brien & Pearson (2004) noted that children with ASD and an IQ over 70 have a higher chance of developing functional language. It therefore seems like IQ could mediate the relationship between ASD and language development. In the newly implemented International Classification of Disease, 11th edition (World Health Organization, 2019), one codes for whether the person also fulfil the criteria of an intellectual disability or not. One could expect this to affect the prevalence of both intellectual disability and ASD, as it has been noted that as the rate of ASD diagnoses go up, the rates of intellectual disabilities and learning disabilities goes down (Matson & Shoemaker, 2009).

Medical

Recent research has investigated the relationship between ASD and various medical conditions. Several studies have identified a correlation between ASD and symptoms of gastrointestinal illness (GI), such as diarrhoea, abdominal pain, constipation, nausea, and bloating (Chandler et al., 2013; Mannion et al., 2013; McElhanon et al., 2014). Furthermore, a literature review by Bauman (2010) identified an increased prevalence of GI, such as inflammatory bowel disease, celiac disease, and Crohn's disease in children with ASD. The results from studies investigating whether there is a correlation between ASD severity and prevalence of symptoms are mixed, with some finding such a correlation (Adams et al., 2011), while others do not find such a correlation (Chandler et al., 2013). It could be that the observed GI-symptoms are associated with avoidant-restricted food intake disorder (ARFID). ARFID-like presentations are common in children with autism, with as many as 21% being affected (Koomar et al., 2021). Some known consequences of ARFID is weight loss, vitamin and mineral deficiencies, and poor health outcome (Bourne et al., 2022). In the most severe cases, nutritional supplements and enteral feeding becomes necessary. Further studies on somatic health in children with ASD have identified correlations between ASD and mitochondrial dysfunction (Bauman, 2010; Rossignol & Frye, 2012), immune system abnormalities (Ashwood et al., 2006), cerebral palsy (Chen et al., 2009), and hormonal changes (Bauman, 2010).

Psychiatric and neurodevelopmental disorders

Both children and adults with ASD are at an increased risk of developing comorbid psychiatric and neurodevelopmental illnesses. One of the most prevalent conditions is sleep problems, estimated to affect 40-80% of children with autism (Bauman, 2010). The prevalence of depression and anxiety also seem to be elevated. As many as 15% of children with ASD fulfil the diagnostic criteria for an anxiety disorder (Mannion et al., 2013). Affective disorders seem to be prevalent, and in one sample 35% of participants fulfilled the criteria for an affective disorder. 50% of these had close relatives with an affective disorder or relatives who had committed suicide (Lainhart & Folstein, 1994). Thus, it seems like the increased prevalence of affective disorders could have a heritable component. However, it has been difficult to estimate the occurrence of psychiatric illness in ASD, as there are few standardised assessment tools designed specifically for those with low-functioning ASD (LFASD) (Magnuson & Constantino, 2011). Conducted studies suggest that children and teenagers with high-functioning ASD (HFASD) might have a higher risk of developing comorbid depression, compared to those with LFASD (Lopata et al., 2010; Vickerstaff et al., 2007). One reason for this could be that children with HFASD have a less severe social and cognitive deficiencies, which makes them aware of their lack of social competence. Further studies support this finding, showing that approximately 2% of children with ASD fulfil the criteria of a depressive disorder, compared to 30% of children with Asperger's syndrome

(Matson & Nebel-Schwalm, 2007). Recent studies have also highlighted the high comorbidity between ASD and attention deficit hyperactivity disorder (ADHD). It is estimated that 22-83% of children diagnosed with ASD also fulfil the diagnostic criteria for ADHD, whereas 30-65% of children with ADHD have clinically significant symptoms of ASD (Sokolova et al., 2017). Both ADHD and ASD are heritable neurodevelopmental disorders, and it has been hypothesised that the high comorbidity could be caused by shared genetic factors (Rommelse et al., 2010; Sokolova et al., 2017).

Maladaptive behaviour

Externalised maladaptive behaviour refers to behaviour such as aggression, impulsivity, harmful repetitive stereotypical movements, tantrums, and self-injury, whereas internalised maladaptive behaviour refers to behaviour such as fearfulness, difficulties concentrating and social withdrawal (Sparrow et al., 2005). As many as 82-94% of children with ASD present with some form of maladaptive behaviour (Jang et al., 2011; Murphy et al., 2009). Studies on maladaptive behaviour in ASD has typically focused on externalising behaviour. Anderson et al. (2011) found consistently higher levels of maladaptive behaviour in children with ASD, and the severity of the behaviour seems to relate to severity of intellectual disability. Autistic children with lower IQ were found to consistently show more externalising behaviour than autistic children with average IQ. Furthermore, level of maladaptive behaviour seems to correlate with verbal abilities, where children with poor expressive language skills show increased maladaptive behaviour (Hartley et al., 2008). The correlation between language and maladaptive behaviour is hypothesised to reflect the fact that children with poor verbal abilities are unable to accurately communicate needs, and thus their behaviour could result from miscommunication and caregiver's inability to understand the child's signals. If left untreated, maladaptive behaviour can severely impact the child's daily functioning, both in educational settings and in social settings. Other factors could also mediate the relationship between ASD and maladaptive behaviour. One study found that boys diagnosed with both ASD and ADHD showed a higher prevalence of Oppositional Defiant Disorder (ODD) than boys diagnosed with ASD only (Guttmann-Steinmetz et al., 2009).

Theories

There exist several theories trying to explain the cause and development of ASD. Earlier works on the cause of ASD focused on psychological and cognitive deficits exhibited by children with autism. More recently, ASD has been understood as a neurodevelopmental disorder heavily influenced by biological, genetic, and epigenetic factors. The psychological and cognitive theories are still hold relevance for understanding how the child's deficits in cognitive function manifests itself in real world situations.

Psychological and cognitive theories

Early works on psychological and cognitive deficits in children with autism focused on an apparent deficit in Theory of Mind (ToM). One influential study by Baron-Cohen et al. (1985) conducted predictive tasks on children with autism, where the tasks were designed to measure the child's ability to employ ToM. The study found that children with ASD had difficulties representing the mental states of others. Furthermore, the authors argued that deficits in ToM could not be due to intellectual disability, as a control group made up of children with Down's syndrome did not exhibit the same level of deficit. It is hypothesised that failure in ToM could explain why autistic children struggle with social communication. Further studies hypothesised that deficits in ToM would not only impair the child's ability to engage in social communication, but also makes the child incapable of recognising their own mental states, affecting their ability to learn from social context (Williams, 2010). However, newer studies have challenged the idea that autism cause deficits in ToM. Children with high-functioning ASD have been found to perform equally well on predictive tasks as their non-autistic peers (Bauminger & Kasari, 1999; Fisher et al., 2005; Scheeren et al., 2013). These studies identified verbal abilities and IQ as factors that predict performance on predictive tasks. Thus, deficits in ToM can be understood as a result of intellectual disability and poor verbal abilities, rather than being a symptom of autism itself. However, adequate performance on predictive tasks do not necessarily mean that this skill is generalised to the child's environment and they could still exhibit difficulties in social interactions (Scheeren et al., 2013).

Later studies by Baron-Cohen (2002) focused not only by autistic children's deficits in ToM, but also how they generally show a different cognitive profile than their non-autistic peers. He proposed the "Extreme male brain" theory of autism, which claims that men and women in general differ in ability to systemise and empathise. A typically male cognitive profile will include better abilities related to systemising, whereas a typically female cognitive profile includes better abilities related to empathising. Baron-Cohen's theory is then, that autism reflects an extreme variant of the male brain. He hypothesises that one cause of this could be exposure to androgens during the foetal period. Studies have attempted to test his hypothesis. It has been found that autistic people do outperform non-autistic people on tasks assumed to relate to systemising, but no link between autism and foetal androgen exposure have been identified (Falter et al., 2007; Kung et al., 2016). Further studies found that brain maleness, operationalised as sex differences in subcortical brain shape, is associated with autistic symptoms. However, males generally have larger brains than females. When adjusting for variation in brain size, no association between brain maleness and autistic symptoms is found (van Eijk & Zietsch, 2021).

Biological theories

Genes are considered a large contributor to the development of ASD, and some autism susceptibility genes have been identified. However, it seems like each of these genes exert a weak effect on the development of ASD (Abrahams & Geschwind, 2008; Masi et al., 2017). Heritability studies have found evidence for heritability, where a sibling of an older autistic sibling have an increased risk of receiving a diagnosis (Chaste & Leboyer, 2012; Masi et al., 2017). A similar pattern has been found in twin studies, where monozygotic twins have a higher concordant rate than dizygotic twins (Bailey et al., 1995; Chaste & Leboyer, 2012). It has been proposed that the high concordant rate in twins is not fully explained by genetic overlap, but that being a twin is itself a risk factor. Some studies have found support for this hypothesis (Betancur et al., 2002; Greenberg et al., 2001), whereas others have failed to replicate these findings (Hallmayer et al., 2002).

In addition to genetic studies, there has been conducted studies on brain abnormalities in people with autism. Brain areas of interests have been the frontal lobes, amygdala, and cerebellum, all of which show structural differences in some samples of autistic individuals (Amaral et al., 2008). Other studies have investigated patterns of functional overconnectivity and underconnectivity between relevant brain areas (Just et al., 2004; Vissers et al., 2012). There is some evidence that this underconnectivity and overconnectivity could be related to the severity of autistic symptoms (Rudie & Dapretto, 2013).

Burger & Warren (1998) observed deficiencies in the immune systems of children with autism, specifically in T-cells and some types of antibodies. They theorize that deficiencies in immune system functioning could increase the risk of autism, as it allows pathogens to damage the brain directly or indirectly through autoimmune mechanisms. Another approach to investigating the immune systems of children with autism has been to measure markers of inflammation in cerebrospinal fluid. No link between markers of central nervous system inflammation and autistic traits have been found (Zimmerman et al., 2005).

Treatment

ASD is considered a life-long disorder. Numerous treatments have been developed in an attempt to improve the affected individual's quality of life and reduce societal costs. In the United States, it has been estimated that the annual cost associated with ASD is $250 billion dollars, with a lifetime individual cost between $1.5 and $2.5 million dollars (Lyall et al., 2017). Early intervention for children with ASD is believed to be cost-effective, with between $187.000 and $208.000 USD saved per child who receives early treatment (Chasson et al., 2007; Jacobson et al., 1998).

Existing interventions are usually aimed at symptoms, such as facilitating language development and social communication, reducing the frequency of maladaptive behaviour, and treating symptoms of comorbid illness. The present section is not a comprehensive list of available treatments, but rather a brief overview of some commonly used interventions.

Pharmacological interventions

Pharmacological interventions are sometimes used to treat symptoms that decrease quality of life in children with ASD. In the USA, two antipsychotics (Risperidone and Aripiprazole) are approved for the symptomatic treatment of irritability, aggression, tantrums and self-injurious behaviour (Masi et al., 2017). However, antipsychotic medications sometimes lead to serious side effects, and is only used when the potential benefits outweigh the potential risks. Furthermore, melatonin has proven effective in the treatment of insomnia in children with autism, leading to lower sleep latency and increased duration of total sleep per night (Maras et al., 2018; Schroder et al., 2021). One meta-analysis (Rossignol & Frye, 2011) found support for melatonin abnormalities in children with ASD, including changes in circadian rhythm and low levels of melatonin. Furthermore, the meta-analysis found a positive correlation between levels of melatonin and the severity of autistic behaviour. Thus, pharmacological treatment with melatonin is hypothesised to target an underlying cause of insomnia in children with ASD. However, one study on sleep and comorbid illness in ASD found that avoidant behaviour, under-eating and GI-symptoms are predictive of the variance in sleep problems (Mannion et al., 2013). Ongoing clinical trials are currently investigating the effect of other drugs, such as Fluoxetine, Memantine, Mirtazapine and Oxytocin in treating symptoms associated with ASD (LeClerc & Easley, 2015). Such trials are still in progress, and the effectiveness of the aforementioned medications is still inconclusive.

Applied Behavioural Analysis

Applied Behaviour Analysis (ABA) is a form of therapy based on principles from operant conditioning. The core elements in ABA is shaping behaviour through the use of rewards and punishments (Wilkenfeld & McCarthy, 2020). ABA is an intensive form of treatment and requires as much as 40 hours of therapy per week. Several meta-analyses have investigated the effect of ABA. The findings generally show that ABA is highly effective, improving intellectual abilities, receptive and expressive language, adaptive behaviour, IQ, and non-verbal IQ (Eldevik et al., 2010; Makrygianni et al., 2018; Peters-Scheffer et al., 2011). However, in recent years ABA has received criticism, mainly from autistic self-advocates. Kirkham (2017) reviewed online blogposts and news articles written by adults with autism who received ABA as children. The summary draws attention to the lived experiences of adults with ASD, where several persons reject the medical model of autism, and would rather autism be considered a neuropsychological difference, rather than a deficit. Wilkenfeld & McCarthy (2017) further reviews the ethics of ABA. The authors mainly criticise ABA's exclusive use of behavioural interventions and problematises the fact that the child's autonomy is overridden. One study (Kupferstein, 2018) found an increased prevalence of posttraumatic stress-symptoms (PTSS) in adults who were subjects of ABA as children. The study found PTSS in 46% of the group treated with ABA, compared to only 28% of the ones not treated with ABA. One potential problem with studies on ABA is the fact that it is considered the most effective treatment, and thus few RCT's have been conducted as it has been considered unethical to withhold therapy (Kirkham, 2017). In addition, conducted studies often measure improvement by asking parents or teachers, rather than the child itself.

Music therapy

Music therapy (MT) aims to improve health through the use of music-based interventions (Gassner et al., 2021). In typically developing children, music therapy has been found to correlate with structural brain changes in musically relevant brain regions following 15-months of musical training (Hyde et al., 2009) and to induce structural brain changes in cortical thickness in the right and left posterior temporal gyrus and in fractional anisotropy in the corpus callosum following two years of music therapy (Habibi et al., 2018). Recent studies have evaluated the effect of MT on brain connectivity, social communication, and language abilities in children with ASD. The therapy can be delivered as individual music therapy, family centred therapy or in a group setting (Marquez-Garcia et al., 2021). There is some evidence that MT can improve social communication, brain connectivity and parent-child relationships (Gassner et al., 2021; Sharda et al., 2018). Further studies indicate that MT is superior to placebo therapy and standard care when it comes to improving social interaction and verbal- and non-verbal language skills (Geretsegger et al., 2014). However, other studies have found that MT is not significantly better at improving social affect when compared to standard care (Bieleninik et al., 2017), and that MT is equally effective as ABA when it comes to improving speech production (Lim & Draper, 2011). There is some evidence that the effect of MT and speech therapy also depend on the child's level of functioning, where low-functioning children showed greater improvement in speech production following MT, whereas high-functioning children had equal improvement following MT or speech training (Lim, 2010).

Research objective

The objective of the present paper is to investigate the relationship between receptive vocabulary, social communication skills and adaptive behaviour in children diagnosed with Autism Spectrum Disorder. The present paper will investigate three main hypotheses.

Research question 1: Receptive vocabulary and social communication

The first research question (RQ1) to be investigated is whether there is a correlation between receptive vocabulary and social communication skills in a sample of children diagnosed with ASD. The hypotheses to be tested are:

H_0: There is no significant correlation between receptive vocabulary and social communication skills.

H_1: There is a significant correlation between receptive vocabulary and social communication skills.

Research question 2: Receptive vocabulary and adaptive behaviour

The second research question (RQ2) to be investigated is whether there is a correlation between receptive vocabulary and adaptive and maladaptive behaviour in a sample of children diagnosed with ASD. The hypothesis to be tested are:

H_0: There is no significant correlation between receptive vocabulary and adaptive and maladaptive behaviour.

H_1: There is a significant correlation between receptive vocabulary and adaptive and maladaptive behaviour.

Research question 3: Social communication and adaptive behaviour

The third research question (RQ3) to be investigated is whether there is a correlation between social communication and adaptive and maladaptive behaviour in children diagnosed with ASD. The hypothesis to be tested are:

H_0: There is no significant correlation between social communication and adaptive and maladaptive behaviour.

H_1: There is a significant correlation between social communication and adaptive and maladaptive behaviour.

Method

Music for autism

Music for autism (M4A) is a collaboratory research project between the University of Bergen, Norway, the Norwegian Research Centre (NORCE) and the University of Vienna, Austria. The aim of the project is to investigate the effect of music therapy in children with ASD. The project employs an assessor-blinded crossover randomised controlled trial (RCT) design, comparing 12 weeks of music therapy (MT) to a structurally matched 12 weeks play therapy (PT) intervention. Participants are randomised to participate in MT-PT or PT-MT. Before the start of the project and after each intervention phase, the child is assessed on relevant outcome measures. The outcome measures of interest are social communication skills, brain connectivity and structural brain changes, measured through MRI-scans and various assessment forms. M4A attempts to replicate and expand a Canadian study (Sharda et al., 2018), in order to further our understanding of music therapy and the effect it could have on children with ASD. The M4A project is on-going.

I joined the project in January of 2022. My role was to conduct the PPVT-4 assessments in Norway. I was not involved in planning, recruitment, statistical analysis, interventions or writing of reports in the M4A-study. I was, however, able to access data collected at baseline in both the Norwegian and the Austrian sample, including data collected before I joined the project. Data collected between September 2021 and November 2022 is included in the present paper.

Intervention description

The children participating in the M4A study were assessed at baseline before they were randomised to participate in either 3 months of play-therapy or music-therapy. Thereafter came a 3-month wash-out period. The children who received play-therapy then received music therapy, and the children who received music-therapy received play-therapy. The present paper used data collected at baseline assessment.

Sample

Participants were recruited through advertising to relevant groups through social media, and by reaching out to various groups such as parent associations, schools, clinicians, and special educators. Children between the ages of 6 and 12 who met the diagnostic criteria for ASD as specified in DSM-5 were eligible for participation. Children of all levels of functioning were included, also those with comorbid conditions. Exclusion criteria were current or recent music therapy or music lessons, medical contraindications such as metallic or electronic implants that would prevent the child from participating in an MRI scan, and use of medication known to affect brain function. At the time of writing this paper, 41 children have been recruited for M4A. However, only 15 children had valid scores on all relevant instruments at baseline. 26 children were missing scores on one or more instruments and were thus excluded. Therefore, data from 15 children was employed in the present statistical analysis.

Table 2

Sample

Country	**N-girls**	**N-boys**	**Total**
Norway	2	2	4
Austria	1	10	11
Total	3	12	15

Instruments

All children were assessed using the Peabody Picture Vocabulary Test – 4th edition (PPVT-4), the Children's Communication Checklist – 2nd edition (CCC-2), Vineland Adaptive Behaviour Scale – 2nd Edition (VABS), Social Responsiveness Scale – 2nd edition (SRS), Family Quality of Life Scale (FqoL) and Children and Adolescent Scale of Participation (CASP). The PPVT-4 was conducted with the child. The child's parents were present during testing but were asked to not aid the child. VABS, SRS, FqoL and CASP were sent digitally to the child's parents, who filled out the form online. CCC-2 was sent digitally to the child's teacher or special educator, who filled out the form online. Thus, PPVT-4 was the only test where the child was directly involved, whereas the other tests were answered by someone who knows the child well.

Peabody Picture Vocabulary Test, version 4

The Peabody Picture Vocabulary Test 4th edition (PPVT-4) is a measure of receptive vocabulary. The test consists of 228 items, grouped into 19 sets with 12 items each (Dunn &

Dunn, 2007). The sets are arranged in order of increased difficulty. The PPVT-4 does not require the child to use verbal language. During testing, the child is shown a slide with four images, labelled 1-4. The examiner speaks a word, and the child is asked to point to or read the number of the correct image. A basal set is established when the child has zero or one mistake in a 12-item set. The ceiling set is established when the child has 8 mistakes in one set. This allows the examiner to administer only the sets appropriate for the child's vocabulary range. After testing, the child's score is converted from raw score to standard score. The PPVT-4 has an average score of 100, with a standard deviation of 15.

There is currently no Norwegian version of PPVT-4. The version used in the current study was translated from the English version. The original words were first translated from English to Norwegian by the author. The Norwegian words were then back-translated by a second collaborator who was not familiar with the original words. The translations were then compared, and the most suited word was agreed upon. There was an attempt to keep the Norwegian translation as similar to the original word as possible in terms of grammar and conjugation, while also staying true to the increasing difficulty of the sets in the PPVT-4. The translations also had to match the correct image, while also excluding the incorrect images. While attempts were made to provide a suitable translation, the version used in the Norwegian sample has not been evaluated in terms of reliability or validity. When converting raw scores to standard scores, American norms were used.

There is an official German translation of the PPVT-4 which was used to measure receptive vocabulary in the Austrian sample. The German version is standardized to German norms and has an average t-score of 50 with a standard deviation of 10. As the American and German version of the PPVT employs different standardised means and standard deviations, German t-scores were converted to American standard scores. The following formula was used for conversion:

$$(x - 50) * 1.5 + 100$$

The Children's Communication Checklist, version 2

The Children's Communication Checklist (CCC-2) screens for communication disorder and is used to identify deficits in pragmatic language and in social interaction. The test consists of 70 items, divided into ten scales. Each scale has five items that measure language deficits, and two items to identify strengths. The scales are:

- A. Spoken language.
- B. Syntax.
- C. Semantics.
- D. Coherence.
- E. Initiative.
- F. Stereotypical language.
- G. Context.
- H. Non-verbal communication.
- I. Social relationships.
- J. Interests.

The first six scales give estimations of the child's language and communication abilities, whereas the two last scales measure non-verbal behaviour often apparent in children with autism (Bishop, 2011). Raw scores are converted to standard scores using appropriate age norms. The scales have a mean of 10 and a standard deviation of 3. Lower scores indicate poor social communication skills. Standard scores on the eight first scales are used to calculate a general communication index (GCI). The GCI has a cut-off of 55, meaning that a child who scores below 55 should be further tested for language deficits. In addition, it is possible to calculate deficit in in social interaction index (IASI). The purpose of IASI is to identify those children who have a stereotypical autistic language profile. IASI is calculated by summing scale E, H, I and J, and subtracting the sum of scales A-D. A GCI below 55, in addition to a negative IASI-score indicates ASD. In contrast, a GCI below 55 in addition to an IASI-score above 9 indicates specific language impairment (SLI). Furthermore, one can calculate a structural language score by summing scale A-D and a pragmatic language score by summing scale E-H.

The Norwegian version of CCC-2 has shown satisfactory inter-rater reliability between parent and teacher reports, and between parent and professional reports. There is no standardized German version of the CCC-2. The German version was translated from Norwegian to German. Norwegian norms where thus used when calculating the GCI.

The Vineland Adaptive Behaviour Scales

The Vineland Adaptive Behaviour Scales (VABS) is an assessment tool that measures adaptive behaviour (Sparrow et al., 2005). The present paper will use data from the Maladaptive Behaviour Index (MBI), which is used to measure challenging externalizing and internalizing behaviour in children. VABS is standardized in both Norway and Austria. The child's raw scores on internalizing and externalizing behaviour were converted to v-scores using the English manual. Furthermore, the child's raw score on internalizing and externalizing behaviour was summed, to calculate a raw score for the Maladaptive Behaviour Index (MBI), which was then converted to v-scores using the English manual. V-scores have a mean of 15 and a standard deviation of 3. Scores above 18 are considered elevated, and scores above 21 are considered clinically significant.

The Social Responsiveness Scale, version 2 (SRS)

The Social Responsiveness Scale (SRS) is a measure of the severity of social symptoms in children with ASD. The form consists of 65 items, divided into five subscales. These subscales are social awareness, social cognition, social communication, social motivation, and restricted interests and repetitive behaviour. The summed raw score is converted to T-scores, with a mean of 50 and a standard deviation of 10. The minimum possible score is 20 and the highest possible score is 80. A t-score below 59 is within normal limits, a score between 60-65 is considered to indicate mild deficiency, a score between 66 and 75 is considered to indicate moderate deficiency, and a score above 76 indicate severe deficiencies in social interaction (Constantino & Gruber, 2005).

In the Norwegian sample, the official Norwegian adaption was used. The SRS-II is not adapted to German, thus the officially adapted SRS-I was used.

The Child and Adolescent Scale of Participation (CASP)

The Child and Adolescent Scale of Participation (CASP) is a measure of children and adolescent's participation in activities at home, at school and in society (Bedell, 2011). In addition, the form investigates strategies and aids the child uses to aid participation. The form consists of 20 items divided into four subscales. The four subscales are Home, Community, School and Home and Community Living Activities. The items are rated on a four-point scale, where the child's parents are asked to rate their child's participation by comparing the child to their peers:

- Age expected.
- Somewhat restricted.
- Very restricted.
- Unable.

CASP has shown satisfactory test-retest reliability, construct and discriminant validity and internal consistency. The child's score is reported on a 100-point scale, where higher scores indicate higher participation in society (Bedell, 2011).

The Family Quality of Life Scale (FqoL)

The Family Quality of Life Scale (FqoL) assesses parent's degree of satisfaction with the quality of life in their family. The scale consists of five subscales: Family Interaction, Parenting, Emotional Well-Being, Physical/Material Well-Being, and Disability Related Support. Degree of satisfaction is measured on a five-point scale, where 1 equal very dissatisfied, 3 equals neither satisfied nor dissatisfied, and 5 equals very satisfied (Beach Center of Disability, 2012). FqoL has a range of 25-125, where higher scores indicate higher satisfaction with family life.

Statistical analysis

Statistical analysis was conducted in R, version 4.2.1 For the R-script, see Appendix 2. R-packages used in analysis are in the R-script. Prior to analysis, the significance level was set to $\alpha = 0.05$. Thus, a p-value below 0.05 will be considered statistically significant, and the null hypothesis will be rejected. When interpreting correlational coefficients, 0.1 will be considered a small effect, 0.3 will be considered a medium effect, and 0.5 will be considered a large effect (Cozby & Bates, 2020, p. 240; Cumming & Calin-Jageman, 2017, pp. 318-319). P-values will also be reported.

Preliminary analysis

First, descriptive statistics will be calculated for the data material as a whole, and the Norwegian and Austrian samples will also be investigated separately. Potential outliers in the data material will be identified using histograms and boxplots.

Due to the small sample size (N=15) it is unknown if the two samples are normally distributed and if they have equal variance. Heterogeneity of variance will be checked for using Levene's test and normal distribution will be checked using a Shapiro-Wilks test. If heterogeneity of variance and a normal distribution can be assumed, an independent t-test will be conducted for each instrument used to assess whether there are significant differences between the Norwegian sample and the Austrian sample. If there is homogeneity of variance or one cannot assume normal distribution, the non-parametric Mann-Whitney-Wilcoxon u-test will be conducted instead of a t-test.

Research question 1

To investigate whether there is a relationship between receptive vocabulary and social communication, a correlational analysis will be conducted. The instrument used to measure receptive vocabulary is the PPVT-4 and the instrument used to measure social communication skills is the CCC-2. As the CCC-2 consists of 10 subdomains, in addition to a general communication index (GCI), a deficit in social interaction index (IASI), a structural language score and a pragmatic language score, a correlational matrix will be reviewed to investigate the correlation between the PPVT-4 and each domain. If preliminary analysis suggest that the sample does not approximate a normal distribution and/or if the sample contain significant outliers, Spearman's non-parametric correlation coefficient (ρ) will be calculated. If not, Pearson's correlational coefficient I will be used. In addition, a regression analysis and regression diagnostic tests will be conducted. The purpose of the regression diagnostic tests is to assess the fit of the model and linear regression assumptions.

Research question 2

To investigate whether there is a relationship between receptive vocabulary and adaptive and maladaptive behaviour, several correlational tests will be conducted. The instrument used to measure receptive vocabulary is the PPVT-4, the instruments used to measure maladaptive behaviour is VABS-MBI and SRS, and the instruments used to measure adaptive behaviour is the FqoL and CASP.

The score on VABS-MBI provides an estimation of the prevalence and severity of maladaptive behaviour, whereas SRS measures the severity of social symptoms. Thus, the VABS-MBI gives a more general estimate of a wide range of maladaptive traits, whereas the SRS is limited to measuring maladaptive behaviour in the form of deficits in social interaction and social behaviour. In addition, the SRS consists of five subdomains that measure various aspects of deficit in social interaction and social behaviour. To investigate whether there is an overlap in the constructs measured by the two forms, a correlational test will be conducted. If the preliminary analysis shows no significant outliers and a satisfactory normal distribution, Pearson's correlational coefficient I will be calculated. If these conditions are not met, the non-parametric Spearman's rho (ρ) will be used instead.

The score on FqoL provides an estimate of family satisfaction, whereas CASP provides an estimate of the child's ability to participate in society. These measures will be used to estimate adaptive behaviour. It is hypothesised that FqoL estimates adaptive behaviour, as children's behaviour will affect the family dynamic as a whole. If the child exhibits adaptive behaviour, the child will have the necessary skills to participate positively at home. CASP is believed to measure adaptive behaviour, as children with satisfactory social skills and reduced need of support, will find it easier to participate in activities at home, at school, and in society. To investigate whether there is an overlap in the constructs measured by the two forms, a correlational test will be conducted. If preliminary analysis shows no significant outliers and a satisfactory normal distribution, Pearson's correlational coefficient I will be calculated. If these conditions are not met, the non-parametric Spearman's rho (ρ) will be used instead.

To investigate the relationship between receptive vocabulary and adaptive and maladaptive behaviour, several correlational analyses will be conducted. The PPVT-4 will be compared to each of the four instruments used to estimate maladaptive and adaptive behaviour. If preliminary analysis suggest that the sample does not approximate a normal distribution and/or if the sample contain significant outliers, Spearman's non-parametric correlation coefficient (ρ) will be calculated. If not, Pearson's correlational coefficient I will be used. Second, regression analyses and model-fit tests will be conducted. Simple linear regression will be used to investigate the potential linear relationship between PPVT-4 and each of the four instruments used to estimate adaptive and maladaptive behaviour. In addition, a multiple regression analysis will be conducted to investigate the relationship between receptive vocabulary and maladaptive behaviour (PPVT-4 and VABS-MBI + SRS), and the relationship between receptive vocabulary and adaptive behaviour (PPVT-4 and FqoL + CASP).

Research question 3

The approach taken to investigate RQ3 will mirror the approach taken in investigating RQ2. To investigate whether there is a correlation between social communication and adaptive and maladaptive behaviour, correlational and regression analysis between CCC-2 and VABS-MBI, SRS, FqoL and CASP will be completed. The statistical analysis will follow the same steps at RQ2, except the variable PPVT-4 will be replaced with CCC-2 and its subdomains.

Ethics

Music for Autism was approved by the ethics committee of Western Norway (REK) and the institutional ethics committee of the University of Vienna (See: Appendix 1).

Results

Sample

The final sample consists of 15 children (M-age = 8.5, SD = 1.96, range = 6 – 12). The sample consists of 3 girls (20%) and 12 boys (80%). 4 children were recruited in Norway and the remaining 11 were recruited in Austria. Table 3 shows the mean score on all relevant instruments.

Table 3

Average scores

Mean score	Spread	M	SD
PPVT-4	71.5 – 145	92.83	21.22
VABS-MBI	16 – 21	18.60	1.40
CCC-GCI	13 – 68	45.13	16.41
CCC-IASI	-32 – 11	-7.40	13.46
SRS	59 – 87	72.40	7.44
FqoL	30 – 116	89.27	24.00
CASP	55 – 87.5	72.58	11.07

Notes: PPVT = Peabody Picture Vocabulary Test, VABS = Vineland Adaptive Behaviour Scale – Maladaptive Behaviour Index, CCC-2 = Children's Communication Checklist, General Communication Index, SRS = Social Responsiveness Scale, FqoL = Family Quality of Life Scale, CASP = Child and Adolescent Scale of Participation. M = Mean, SD = Standard deviation.

PPVT-4

The mean score on the PPVT-4 was 92.83 (SD = 21.22), meaning slightly below the standardised mean (M = 100). 4 children scored above average (26.7%), and 11 children scored below average (73.3%). However, 8 children (53.3%) scored within 1 SD of the mean and 14 children (93.3%) scored within 2 SD of the mean.

CCC-2

Scores on the GCI of the CCC-2 showed a mean of 45.13 (SD = 16.41). 10 children (66.7%) scored below 55 on the GCI, which is the cut-off for when further assessment of language deficiencies is recommended. Scores on the IASI of the CCC-2 showed a mean of -7.40 (SD = 13.46). Of the total sample, 7 children (46.7%) had a language profile characteristic for children with ASD (GCI < 55 and IASI < 0). No children had a language profile characteristic for SLI (GCI < 55 and IASI > 9). The sum of subdomain A-D makes up the child's structural language score. The sample showed a mean structural language score of 22.67 (SD = 12.12). The sum of subdomain E-H makes up the child's pragmatic language score. The sample showed a mean pragmatic language score of 15.13 (SD = 5.89).

Table 4 shows spread, mean and SD for each subdomain of the CCC-2.

Table 4

Average scores

	Spread	M	SD
CCC-GCI	13 – 68	45.13	16.41
CCC-IASI	-32 – 11	-7.40	13.46
A. Spoken language	0 – 12	7.00	4.14
B. Syntax	0 – 12	5.20	3.73
C. Semantics	0 – 10	5.07	2.71

D. Coherence	2 – 14	5.40	3.72
E. Initiation	0 – 8	4.33	2.29
F. Stereotyped language	1 – 10	3.73	2.37
G. Context	1 – 7	3.47	1.64
H. Non-verbal communication	0 – 6	3.60	1.84
I. Social relations	0 – 10	3.87	2.75
J. Interests	1 – 9	3.47	2.07

Notes: GCI = General communication index, IASI = Deficit in social interaction index, M = Mean, SD = Standard deviation.

VABS

The mean score of VABS-MBI was 18.60 (SD = 1.40). According to the VABS scoring manual a score above 18 (1 SD above the mean) is considered elevated. 11 children (73.3%) scored above 18. No children scored below the mean. These findings suggest elevated presence of maladaptive behaviour in the sample.

SRS

On the SRS the average score was 72.40 (SD = 7.44). According to the SRS scoring manual, a t-score below 59 is within normal limits, a score between 60-65 indicate mild severity of social symptoms, a score between 66-75 indicate moderate severity of social symptoms, and a score above 76 indicate severe deficiency of social symptoms. 13 children (86.6%) of the sample fell within the moderate to severe social symptoms categories.

Table 5

Scores on the SRS by subdomain

	Spread	Mean	SD
SRS t-score	59 – 87	72.40	7.44
Social awareness	51 – 86	69.33	9.82
Social cognition	59 – 90	71.20	8.25
Social communication	52 – 84	71.07	8.18
Social motivation	46 – 87	65.80	10.51
RRB	53 – 90	70.40	9.98

Notes: RRB = Restricted interests and repetitive behaviour.

CASP

On the CASP, the mean score was 72.58 (SD = 11.07). CASP uses a 100-point scale, where higher scores indicate higher participation in activities at home, at school and in society. Thus, the sample score is relatively high, indicating a high degree of participation.

FqoL

On the FqoL, the mean score was 89.27 (SD = 24). The FqoL is scored between 25 and 125, so the results indicate relatively high family satisfaction.

Outliers

Boxplots were used to identify potential outlier scores in the data material. No outliers were identified on the VABS-MBI, CCC-GCI, CCC-IASI, SRS or CASP. 1 outlier was identified on the PPVT-4; one child received a score of 145, meaning 3 SD above the mean. 2 outliers were identified on the FQoL. Z-scores were calculated to identify how far they deviated from the mean score. For the raw scores 30 and 47, $z = -2.47$ and -1.76, respectively. Although the scores deviate significantly from the mean, low scores are still valid as the range of possible scores on the FqoL is 25-125. Thus, no scores were removed or replaced.

Preliminary analysis

In the preliminary analysis, Levene's test was used to investigate whether the Austrian and the Norwegian sample have equal variance. For PPVT-4 the null hypothesis of equal variance between the two groups is rejected, $F(1, 13) = 5.18$, $p = 0.04$. However, an independent-samples Wilcoxon-Mann-Whitney U-test revealed that the two groups do not significantly differ from each other ($W = 22$, $p = 1$). For VABS-MBI, CCC-GCI, CCC-IASI, SRS, FqoL and CASP, Levene's test revealed no significant p-values, and thus equal variance can be assumed. Independent samples t-tests revealed no significant differences between the Norwegian and the Austrian samples on the VABS-MBI, CCC-GCI, CCC-IASI, SRS, FqoL and CASP.

Shapiro-Wilk test of normality revealed no significant deviation from the assumption of a normal distribution when investigating the Norwegian and Austrian sample separately. However, the null hypothesis of normal distribution of scores was rejected for the PPVT-4 and the FqoL when analysing the Norwegian and Austrian sample combined. A Shapiro-Wilk test on the PPVT-4 scores showed $W = 0.86$, $p = 0.02$, and a Shapiro-Wilk test on the FqoL showed $W = 0.79$, $p = 0.002$.

As a consequence of the preliminary analysis, non-parametric statistical tests will be used when analysing the correlations between the PPVT-4 and the FqoL and the other instruments. Otherwise, parametric tests will be used.

Research question 1

As the null hypothesis of normal distribution of PPVT-4 scores was rejected, the non-parametric Spearman's rho (ρ) correlation coefficient was calculated. A positive correlation between the PPVT-4 and the CCC-GCI was found, $\rho(13) = .54$, 95% CI [.02, .83], $p = .037$. No significant correlation between the PPVT-4 and the IASI was found ($\rho(13) = -.49$, 95% CI [-.81, .04], $p = .061$). Furthermore, the CCC-2 allows for computing a structural language score and a pragmatic language score. No significant correlation between structural language score and PPVT-4 ($\rho(13) = .49$, 95% CI [-.03, .80], $p = .062$), nor between pragmatic language score and PPVT-4 ($\rho(13) = .35$, 95% CI [-.19, .73], $p = .197$) was found.

A further analysis revealed that the PPVT-4 significantly correlates with two subdomains on the CCC-2: B. Syntax ($\rho(13) = .70$, 95% CI [.27, .89], $p = .004$) and D. Coherence ($\rho(13) = .52$, 95% CI [-.01, .82], $p = .048$). Table 6 shows Spearman's rho correlation coefficient between PPVT-4 score and each index and subdomain of the CCC-2.

Table 6

Correlations by subdomain

	PPVT
CCC-GCC	.54*
CCC-IASI	-.49
Structural language	.49
Pragmatic language	.35
A. Spoken language	.41

B. Syntax	.70**
C. Semantics	.50
D. Coherence	.52*
E. Initiation	.00
F. Stereotyped language	.07
G. Context	.32
H. Non-verbal communication	.47
I. Social relations	.18
J. Interests	-.10

Notes: CCC-GCC: General communication index. CCC-IASI: Deficit in social interaction index. Structural language: Sum of scales A-D. Pragmatic language: Sum of scales E-H. Spearman's correlation coefficient (ρ). * = *p*<0.05*,* **=*p*<0.01.

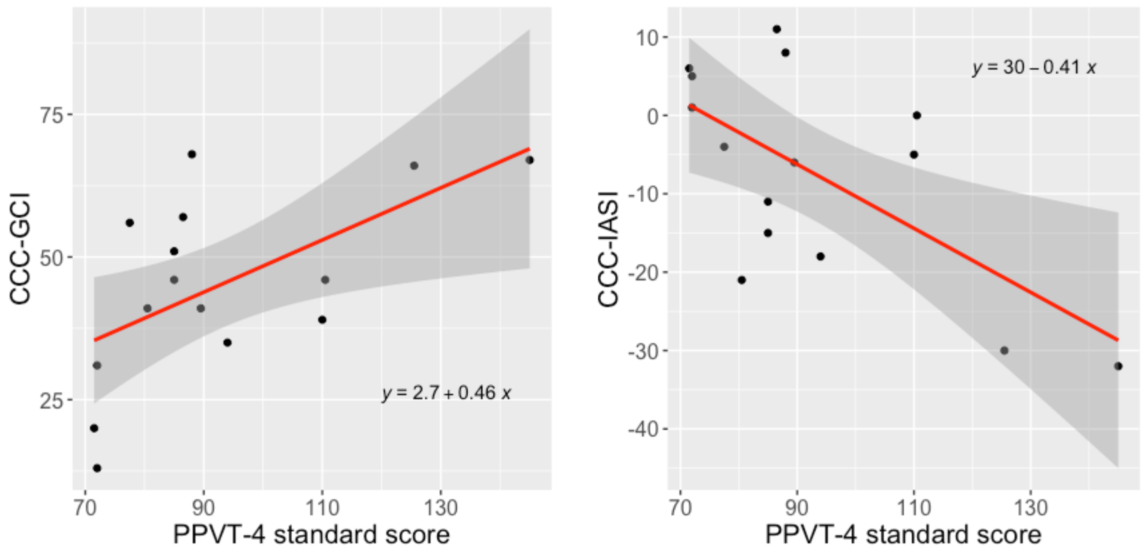
A linear regression analysis was conducted to determine the effect of receptive vocabulary (PPVT-4) on social communication skills (CCC-GCI). The model was statistically significant (F(1, 13) = 6.97, p = .023). Adjusted R^2 indicates that 29.90% of variance in CCC-GCI score can be explained by variance in PPVT-4 score. The regression coefficient (B = 0.46, 95% CI [0.08, 0.83]) indicate that CCC-GCC score increase by 0.46 for each additional point on the PPVT-4. A test of linear model assumptions identified no significant problems with the equation. The scatterplot and regression line are visualised in figure 1a.

A second linear regression analysis was conducted to determine the effect of PPVT-4 score on CCC-IASI score. The model was statistically significant (F(1, 13) = 9.18, p =

0.010). Adjusted R^2 indicates 36.89% of variance in PPVT-4 score can be explained by variance in CCC-IASI score. The regression coefficient (B = -0.41, 95% CI [-0.70, -0.12] indicate that CCC-IASI score decreases by 0.41 for each additional point on the PPVT-4. A test of linear regression assumptions identified no significant problems with the equation. The scatterplot and regression line are visualised in figure 1b.

Figure 1

Linear regression predicting CCC-GCI and CCC-IASI score from PPVT-4 score



However, it is important to note that both GCI and IASI are made up of a combination of subdomains on the CCC-2. Thus, certain subdomains of the CCC-2 are included when calculating both the GCI and the IASI.

As a correlation between the PPVT-4 and the syntax and coherence subdomains was identified, a multiple linear regression analysis was conducted to determine the effect of syntax and coherence on the PPVT-4. The model was statistically significant ($F(2, 12)$ = 10.87, p = .002). Adjusted R^2 indicates that 58.50% of variance in PPVT-4 score can be explained by variance in syntax and coherence. However, a large significant correlation was found between the syntax and coherence subdomains (r(13) = .72, 95% CI [.33, .90], p = .002) and a Ramsey Regression Equation Specification Error Test (RESET) revealed a significant problem with the model's functional form (p =.021).

Research question 2

To investigate whether instruments of maladaptive behaviour (VABS-MBI and SRS) and instruments of adaptive behaviour (CASP and FQoL) measure similar concepts, correlation tests were conducted. Neither the correlation between VABS-MBI and SRS (r(13) = .34, 95% CI [-.20, .73], p = .209) nor the correlation between CASP and FQoL (ρ(13) = -.09, 95% CI [-.59, .45], p = .737) were statistically significant.

Maladaptive behaviour

The correlation between PPVT-4 and VABS-MBI was not statistically significant (ρ(13) = -.16, 95% CI [-.63, .40], p = .569). No significant correlation between PPVT-4 and SRS t-score was found (ρ(13) = -.40, 95% CI [-.76, .16], p = .141). However, a significant correlation was found between PPVT-4 and the social cognition subdomain of SRS (ρ(13) = -.54, p = .036).

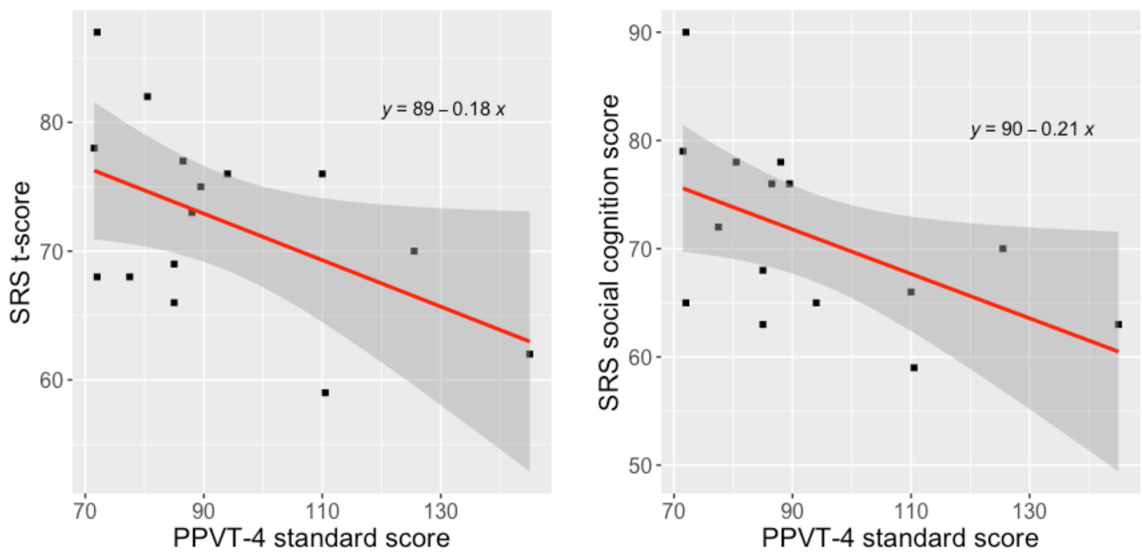
A linear regression model attempting to predict SRS t-score on the PPVT-4 was statistically significant (F(1, 13) = 4.69, p = .050). Adjusted R^2 suggests that 20.86% of variance in SRS t-score can be explained by variance in PPVT-4 score. The regression coefficient (B = -0.18, 95% CI [-0.36, -4.24]) indicate that SRS t-score decreases by 0.18 for each additional point on the PPVT-4. A test of linear model assumptions revealed no significant problems with the equation. The linear equation model predicting SRS t-score from PPVT-4 score is visualised in figure 2a.

As previous tests indicated a significant correlation between PPVT-4 and the social cognition subdomain of SRS, a linear regression model was calculated. The model predicting

SRS social cognition score from PPVT-4 score was statistically significant, (F(1, 13) = 5.03, p = .043). Adjusted R^2 suggest that 22.34% of variance in social cognition score can be explained by variance in PPVT-4 score. The regression coefficient (B = -0.25, 95% CI [-0.40, -0.007]) indicate that SRS social cognition score decreases by 0.25 for each additional point on the PPVT-4. A test of linear model assumptions revealed no significant problems with the equation. The linear equation model predicting SRS social cognition score from PPVT-4 score is visualised in figure 2b.

Figure 2

Linear regression predicting SRS t-score and SRS social cognition score from PPVT-4 score



The present analysis did not identify a significant linear regression model when attempting to predict VABS-MBI score from PPVT-4 score.

Adaptive behaviour

No significant correlation between PPVT-4 and CASP (ρ(13) = .34, 95% CI [-.23, .73], p = .220), nor between PPVT-4 and FQoL (ρ(13) = .17, 95% CI [-.39, .64], p = .546) was identified. Linear regression was conducted to investigate whether PPVT-4 score can be used to predict CASP and FQoL score. Neither the model attempting to predict CASP-score from the PPVT-4 ($F(1, 13) = 2.22$, $R^2 = .08$, $p = .160$), nor the model attempting to predict FQoL score from the PPVT-4 ($F(1, 13) = 0.55$, $R^2 = -0.03$, $p = .470$) were significant.

Research question 3

The present analysis attempts to answer RQ3. The approach taken mirrors the approach used in the previous section; however, PPVT-4 has been replaced by CCC-2 as the independent variable.

Maladaptive behaviour

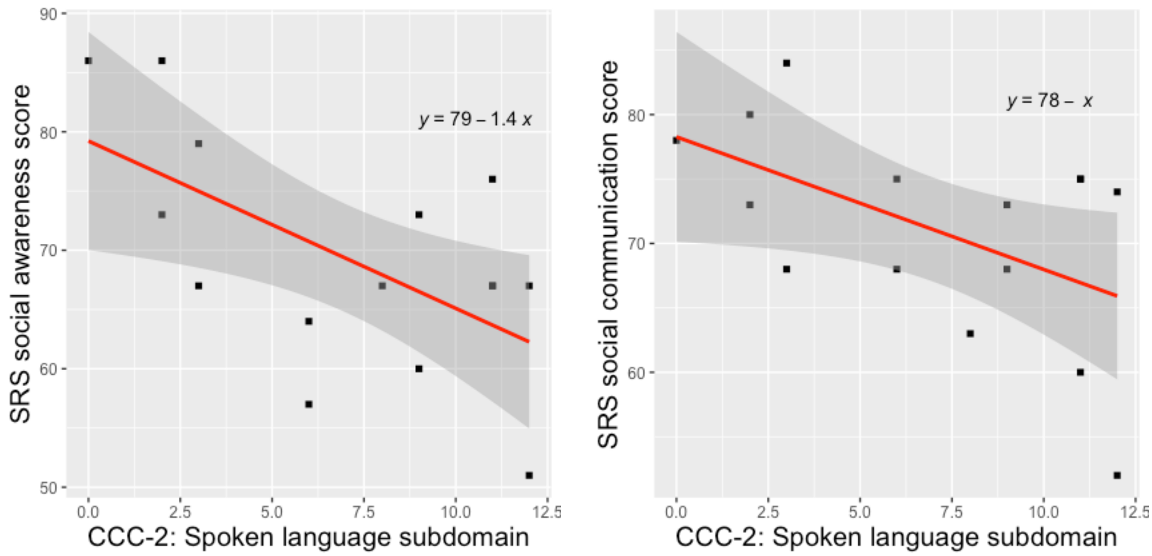
No significant correlation between CCC-GCI and VABS-MBI or SRS t-score was identified. However, a correlation matrix revealed a correlation between subdomain A of the CCC-2 (Spoken language) and the social awareness and social communication subdomains of the SRS. A correlation test showed a large correlation between social awareness and spoken language, $t(13) = -2.67$, $r = -.60$, 95% CI[-.85, -.12], $p = .019$. Another large correlation was identified between social communication and spoken language, $t(13) = -2.20$, $r = -.52$, 95% CI[-.82, -.01], $p = .046$.

A linear regression model attempting to predict SRS t-score from CCC-GCI score was not statistically significant. As significant correlations were identified between the spoken language subdomain of CCC-2 and the social awareness and social communication subdomains of SRS, linear regression models were estimated for these variables. A linear regression equation predicting social awareness from spoken language was statistically significant ($F(1, 13) = 7.14$, $p = .019$. Adjusted R^2 indicates that 35% of variance in social awareness can be explained by variance in speech. The regression coefficient ($B = -1.41$, 95% CI [-2.55, -0.27]) indicates that SRS social awareness score decreases by 1.41 points for each additional point on the CCC-2 spoken language subdomain. No significant problems with the equation were identified by regression diagnostic tests. The regression line is illustrated visually in figure 3a.

A linear regression equation predicting social communication from spoken language was statistically significant (F(1, 13) = 4.84, p = .047. Adjusted R^2 indicates that 21.53% of variance in social communication can be explained by variance in speech. The regression coefficient (B = -1.03, 95% CI [-2.04, 0.02]) indicates that SRS social communication score decreases by 1.03 points for each additional point on the CCC-2 spoken language subdomain. No significant problems with the equation were identified by regression diagnostic tests. The regression line is illustrated visually in figure 3b.

Figure 3

Linear regression model predicting SRS social awareness score and SRS social communication score from CCC-2 spoken language score



Adaptive behaviour

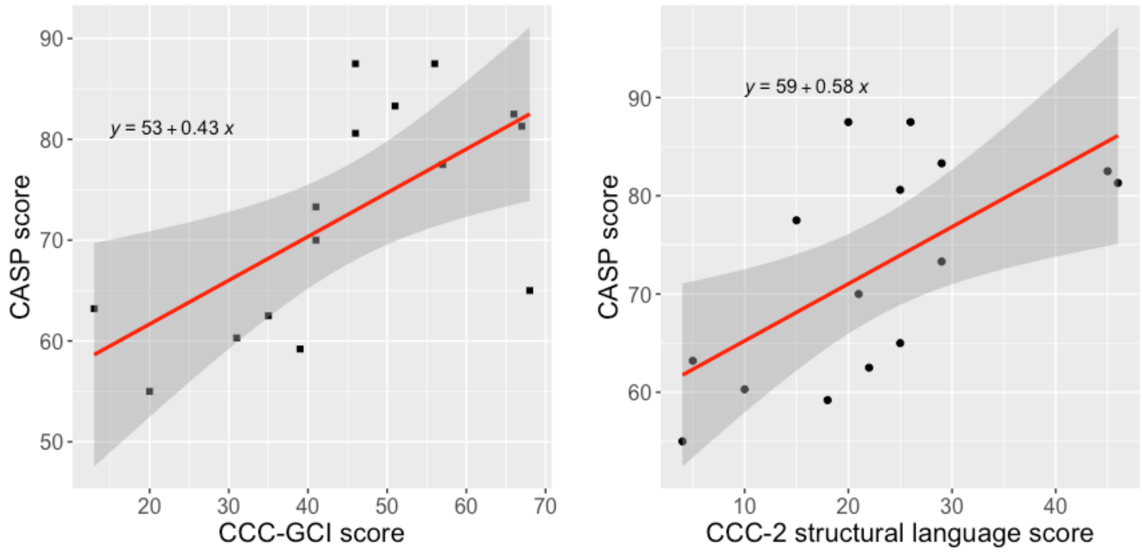
A correlation test revealed no significant correlation between CCC-GCI and FQoL (ρ(13) = .14, 95% CI [-.62, .41], p = .612). A significant correlation between CCC-GCI and CASP was identified, ρ(13) = .64, 95% CI [.20, .87], p = .010. Further analysis revealed that CASP correlates with CCC-2 structural language score ($r(13) = .64$, 95% CI [.18, .86], $p = .011$), but not with CCC-2 pragmatic language score ($r(13) = .32$, 95% CI [-.26, .72], $p = .239$). On a subdomain level, CASP correlates with spoken language ($r(13) = .67$, 95% CI [.24, .88], $p = .007$) and semantics ($r(13) = .56$, 95% CI [.07, .83], $p = .029$).

No significant linear regression model was identified when trying to predict FQoL score from CCC-GCI. A significant linear regression model predicting CASP score from CCC-GCI score was found, $F(1, 13) = 9.18$, $p = .010$. Adjusted R^2 indicates that 36.88% of variance in CASP score can be explained by variance in CCC-GCI score. The regression coefficient ($B = 0.43$, 95% CI [0.12, 0.74]) indicates that CASP score increases by 0.43 points for each additional point on the CCC-GCI. No significant problems with the equation were identified by regression diagnostic tests. The regression line is illustrated visually in figure 4, image 1.

As previous tests revealed a significant correlation between CASP and structural language, but not pragmatic language, further regression analyses were conducted. A significant linear regression model was identified when attempting to predict CASP score from CCC-2 structural language score ($F(1, 13) = 8.79$, $p = .011$). Adjusted R^2 indicate that 35.76% of variance in CASP score could be explained by structural language score. No significant problems with the equation were identified by regression diagnostic tests. The regression coefficient ($B = 0.58$, 95% CI [0.16, 1.00]) indicates that CASP score increases by 0.58 points per additional point on the CCC-2 structural language index. The regression line is illustrated visually in figure 4, image 2.

Figure 4

Linear regression model predicting CASP score from CCC-GCI score and CASP score from CCC-2 structural language score



A linear regression model attempting to predict CASP score from CCC-2 pragmatic language score was non-significant ($F(1, 13) = 1.53$, $R^2 = 0.04$, $p = .239$).

Discussion

Summary of key findings

The objective of the present paper is to investigate the relationship between receptive vocabulary, social communication skills and adaptive behaviour in a sample of children diagnosed with ASD. To investigate these relationships correlational and regression analysis was used.

Statistical analysis indicates a large positive correlation between PPVT-4 and CCC-GCI. However, further analysis indicates that the positive correlation mainly appears to be between PPVT-4 score and two subdomains of the CCC-GCI: Syntax and coherence. Syntax and coherence also significantly correlate with each other. A near-significant negative correlation was found between PPVT-4 and CCC-IASI ($p = .061$). This could possibly be due to both CCC-GCI and CCC-IASI including the syntax and coherence subdomains when calculating the index scores. A second near-significant correlation was identified between CCC-2 structural language score and PPVT-4 score ($p = .062$).

VABS-MBI and SRS t-score were used as measures of maladaptive behaviour, whereas CASP and FQoL were used as measures of adaptive behaviour. Statistical analysis did, however, not find a correlation between the instruments thought to measure similar concepts. PPVT-4 score did not correlate with measures of adaptive behaviour, nor with measures of maladaptive behaviour. However, a large negative correlation was found between PPVT-4 score and score on the social cognition subdomain of the SRS.

No significant correlation between CCC-GCI and measures of maladaptive behaviour was identified. However, a significant correlation was identified between the spoken language subdomain of the CCC-2 and the social awareness and social communication subdomains of the SRS. A positive correlation between CCC-GCI and CASP was identified. Furthermore, CASP correlated with the structural language index of the CCC-2, but not with the pragmatic language index. On a subdomain level, CASP correlates with syntax and spoken language.

Research question 1

The purpose of RQ1 was to investigate whether there is a correlation between receptive vocabulary and social communication skills. Previous studies have found that abnormalities in social communication are one of the first symptoms of ASD noticed by parents of autistic children (De Giacomo & Fombonne, 1998) and that as many as 50% of people with ASD do not develop functional language skills (Lord & Jones, 2012).

Previous studies have investigated receptive vocabulary and social communication skills in children with ASD. The present study found a mean PPVT-4 score of 92.83, whereas comparable studies using the PPVT-III have found means ranging from 68.69 to 97.08 (Kover et al., 2013; Pellicano, 2010; Thomas, 2009). In regard to social communication, the present study found that spoken language was the least affected structural language subdomain, whereas semantics was the most affected. Among pragmatic language skills, initiation was the least affected and context was most affected. A comparable study by Reindal et al. (2021) found similar results. Both the present study and the study by Reindal et al. (2021) found greater impairment of pragmatic language skills compared to structural language skills. Mean score on the general communication index was seven points lower in the Reindal et al. (2021) sample, but both sample means were below cut-off for when further language assessment is recommended. Although minor differences in mean scores, the results from the present study seem comparable to studies with larger sample sizes.

A correlation between receptive vocabulary and social communication skills was found. When analysing the results in detail, receptive vocabulary did not significantly correlate with pragmatic language, whereas a near significant correlation was identified between structural language and receptive vocabulary ($p = .062$). As the correlation was near significant, further studies could expand on this knowledge by employing a larger sample size to identify whether the correlation is actually significant or a result of sample size and sampling bias. Although CCC-2 was used as a measure of social communication skills in children with ASD, it seems like receptive vocabulary is more closely tied to structural language skills, rather than pragmatic language. In addition, when breaking down the structural language index, and analysing correlations on a subdomain level, it seems like the correlation between the general communication index and receptive vocabulary could mainly be explained by coherence and syntax. Syntax refers to the arrangement of words to create sentences, whereas coherence refers to logical and semantical consistency within a text. The results could be interpreted as receptive vocabulary constituting an important feature of structural language, and that a decent vocabulary size could be necessary in order to form sentences with logical and semantic consistency. However, a significant correlation was also identified between the syntax and coherence subdomains; thus, it is difficult to differentiate which of the subdomains exert influence on receptive vocabulary. Although the present study finds tentative support for the idea that receptive vocabulary is tied to structural language skills, future studies could employ stricter methodological approaches to investigate the nature and direction of the relationship. The present results must be interpreted with care, as the present sample size is small. A potential third variable that could be explored in future research is whether the child's learning environment and the child's participation in social contexts could facilitate growth in receptive vocabulary, structural language skills and social communication. This hypothesis is consistent with previous literature, which has found that therapy based on ABA principles is effective in improving receptive and expressive vocabulary (Eldevik et al., 2010; Makrygianni et al., 2018; Peters-Scheffer et al., 2011) and that MT can improve social communication (Gassner et al., 2021; Sharda et al., 2018). Furthermore, it would be useful for future studies to expand on current findings and available research to provide a more comprehensive understanding of how children with ASD develop language. Available research supports the notion that language development is often delayed or abnormal in children with ASD, whilst there is also some evidence that there is a critical period for language acquisition. A more comprehensive understanding of language acquisition could have practical implications in terms of early intervention to facilitate language development.

Although no direct measure of expressive language was employed, the CCC-2 contains a spoken language subdomain, which measures the child's speech. The spoken language subdomain did not significantly correlate with PPVT-4. The results must be interpreted with caution, as spoken word makes up a relatively small part of the CCC-2 questionnaire, but it is still interesting that no relationship between receptive vocabulary and spoken word was identified. The results are consistent with findings from McDaniel et al. (2018) who identified a discrepancy between receptive and expressive vocabulary in children with ASD.

Research question 2

The purpose of RQ2 was to investigate whether there is a correlation between receptive vocabulary and adaptive and maladaptive behaviour.

One study by Sharda et al. (2018) employed several of the same instruments as the present study. Although the present study had a smaller sample size (15 versus 51), the mean scores on instruments were largely comparable on the VABS-MBI and SRS. Both the present study and Sharda et al. (2018) found that mean baseline scores on VABS-MBI are elevated (>18) and mean SRS scores indicate moderate to severe deficiencies in social symptoms. Other studies on maladaptive behaviour in children with ASD have used other instruments in assessment, but regardless of instrument, sampled children with ASD tend to exhibit elevated levels of challenging behaviour (Jang et al., 2011; Murphy et al., 2009). On the FQoL the present study had a lower mean than the Sharda et al. (2018) study. One cause of this could be two extreme low scores in the present sample. No comparable studies examining child and adolescent participation in society, as measured by CASP, was identified. Overall, the results seem consistent with previous literature.

Although VABS-MBI and SRS t-scores were both used as measures of maladaptive behaviour, they did not significantly correlate with each other. One cause of this could be that the instruments measure different aspects of maladaptive behaviour. SRS is specifically developed to measure symptoms associated with ASD, whereas VABS-MBI is part of a broader assessment tool also used to assess typically developing children. Thus, the forms of maladaptive behaviour measured by the two does not necessarily need to correlate. Rather, they could both be of use to give a broad understanding of the child's behaviour. Similarly, CASP and FQoL, used as measures of adaptive behaviour did not correlate. The two instruments both provide an indirect way of estimating adaptive behaviour. CASP could be considered an indicator of adaptive behaviour, as it provides information about the child's participation in relevant activities. FQoL could be considered an indicator of adaptive behaviour, as it provides information about the family's self-rated quality of life. Other factors than the child's behaviour, such as access to support, economy, and living situation, could affect the child's opportunities to participate in social activities and/or the family's quality of life.

The present study did not find a correlation between receptive vocabulary and adaptive behaviour. As previously mentioned, other factors not measured in the present study could exert an influence on whether the child participates in social situations and the family's quality of life.

Receptive vocabulary did not correlate with maladaptive behaviour, as measured by VABS-MBI and SRS t-score. Past studies have found that poor expressive language skills are related to maladaptive behaviour (Hartley et al., 2008). It is hypothesised that this is due to misunderstandings caused by poor communication skills. The influence of receptive vocabulary on maladaptive behaviour, however, is not well understood, and it could be that the child can use alternative modes of communication, even in instances where receptive vocabulary is poor.

A significant negative correlation was found between receptive vocabulary and the social cognition subdomain of the SRS. Social cognition refers to the ability to interpret social behaviour, thus the results indicate that a higher receptive vocabulary correlates with a better ability to interpret social behaviour in others. The relationship between social cognition and language in general is not well studied. Existing papers have hypothesised that deficiencies in social cognition and language in individuals with ASD could be explained by abnormalities in functional connectivity and activity in brain areas related to social and linguistic processing (Kana et al., 2017) or that vocabulary size and having words for mental states can affect social cognition (Dove, 2019). However, no studies have found conclusive evidence for either hypothesis being correct. There is a need for further studies in order to gain a deeper understanding of the relationship between social cognition and language, as both tend to be affected in individuals with ASD.

Research question 3

The purpose of RQ3 was to investigate whether there is a correlation between social communication and adaptive and maladaptive behaviour in children diagnosed with ASD.

No significant correlation between CCC-GCI and instruments measuring maladaptive behaviour was identified. However, significant negative correlations were identified between the spoken language subdomains of the CCC-2 and the social awareness and social communication subdomains of the SRS. Social awareness refers to the child's ability to identify and understand social cues from others, whereas social communication refers to ability to participate in reciprocal communication in social settings. The correlation between spoken language and social communication is perhaps not too surprising, as it seems like the two subdomains measure similar concepts. It is however interesting that spoken language correlate with improved ability to interpret social cues. One possible explanation for this is that higher verbal abilities enable the child to participate in social relationships and thus they receive practice in interpreting social cues. A second possible interpretation is that participation in social contexts could facilitate both development of expressive language, and also teach the child to recognise social cues in other. As the present paper cannot identify the nature of the correlation, these findings raise intriguing questions for further research. Although spoken language and social awareness could reflect two interrelated skills, further understanding of this relationship could have implications for early intervention and treatment to facilitate social and language development. Furthermore, this could relate to maladaptive behaviour, as previous studies have found that level of maladaptive behaviour correlates with verbal abilities; specifically expressive language (Hartley et al., 2008). One possible explanation for this is that children with poor expressive language are unable to accurately communicate needs, which could then lead to miscommunication and maladaptive behaviour due to not being understood. If spoken language, social communication and social awareness are interrelated, it could be that children who generally perform well on these skills necessary for social participation show reduced levels of maladaptive behaviour. More research is needed before a full understanding of the relationship between language, social communication and maladaptive behaviour is established.

A positive correlation between social communication skills and participation in activities at home, at school and in society was identified. The correlation seems to be mainly between CASP and the spoken language and syntax subdomains of the CCC-2; two subdomains which makes up part of the structural language index. Interestingly, CASP did not correlate with pragmatic language skills, which include language skills important for social interaction. One possible explanation for these findings is that children with good spoken language and ability to form sentences can participate in social settings despite lacking pragmatic skills. Perhaps strong structural language skills can compensate for pragmatic language deficiencies. In addition, previous studies have identified syntax as the least affected language skill in children with ASD (Boucher, 2012; Reindal et al., 2021), and Rapin & Dunn (2003) found that structural language skills improve at a greater rate than pragmatic language skills in school-aged children. It could also be that family life, school and social settings constitute environments that facilitate structural language development. For example, structural ABA therapy – which often focuses on teaching socially appropriate behaviour and communication skills – has been found to improve intellectual abilities, language skills and adaptive behaviour (Eldevik et al., 2010; Makrygianni et al., 2018; Peters-Scheffer et al., 2011).

Limitations

Lack of standardized tests

The PPVT-4 was not standardized to Norwegian norms. Thus, the tests validity and reliability are unknown. Although the test was translated using a blind back-translation method, the Norwegian translation might not be accurate in estimating children's vocabulary. There are two main issues with the Norwegian PPVT translation. First, the Norwegian language consists of many compound nouns. Thus, if the child understood one word in the compound noun, they could use reasoning to identify the correct image. Secondly, word frequency varies between Norwegian and English. Some words that were included in the first sets of the PPVT are frequently used in colloquial English are rarely used in colloquial Norwegian, and opposite. One example is "pedagogue" which is the last word in the most difficult set of the English PPVT. The Norwegian word, "pedagog", is commonly used in compound nouns referring to educational professions and might thus be known to Norwegian children.

There is some evidence that the Norwegian PPVT-4 does not accurately reflect the child's vocabulary. First, PPVT-4 employs a stop criterion of eight, meaning the test is complete once the child makes eight mistakes within one twelve-word set. During assessment, several children received six or seven mistakes, multiple sets in a row. Thus, it seems like the limit of the children's s receptive vocabulary was reached, but the test could not be stopped, and the children had to complete the full test. A second line of evidence for the tests lacking validity and reliability stems from the fact that each assessment typically lasted much longer than 10-15 minutes, which is the average assessment time for the English PPVT.

Small sample size

By November 2022, 41 children had been recruited for the M4A project. However, 26 children were missing data on one or more instruments. Thus, these children had to be excluded from statistical analyses. The total sample size included in statistical analyses consists of 15 children. The main reason why children were excluded was because they were missing data on the VABS-MBI (N=21). It is unknown why this form in particular was not returned by the child's parents. In general, most children had valid scores on tests the children performed themselves (PPVT-4) and on data provided by the child's teacher or special educator (CCC-2). As the parent-reported forms were the main forms missing, it could be that the present sample suffer from a sampling bias, and that there are quantitative differences in maladaptive behaviour between the children included and the children excluded.

Tests of normality and equal variance revealed that in some instances, the data did not have a normal distribution. Thus, non-parametric tests were used. The small sample size and the partial use of non-parametric tests makes it difficult to generalize the results to the population as a whole. Results should consequently be interpreted with caution. However, the trend seems to be that results from the present study are consistent with comparable studies employing the same instruments.

Correlation

The present study cannot identify causal relationships between variables. Although the results showed significant correlations between certain instruments, there is uncertainty about the nature of these correlations. In addition, the present study did not control for other variables that could affect language, social communication, and behaviour.

Conclusion

Much work remains before a full understanding of the relationship between language skills, social communication and behaviour is fully understood. Language is a broad term, which refers to several interrelated skills necessary for communication. Similarly, social communication could refer to a variety of ways these language skills are actually practiced in daily life. Language and social communication are fundamental in forming social relationships and bonds with others. Therefore, these skills arguably have consequences for behaviour, both adaptive and maladaptive. However, as this paper demonstrates, the relationship between language, social communication and behaviour is complex and not fully understood, especially in children with ASD where deficits in social interaction and abnormal behavioural patterns are common. In addition, the present paper investigates only a few measures of language, communication, and behaviour. Other factors of importance, such as socioeconomic status, available support systems, IQ, and past treatment, is not controlled for. Thus, there is a possibility that other factors mediate the relationship between variables explored.

The hypotheses being tested cannot be conclusively confirmed, nor rejected. This is partly due to the hypotheses using general terms. The present study indicates that both social communication and behaviour are broad terms referring to several interrelated skills and actions. Thus, whether the null hypotheses are rejected or not, depends on which subset of social communication and behaviour one measures. Further research within this area could have important implications for interventions aimed at improving quality of life for children and adults with ASD. Early assessment and intervention could then use this knowledge in order to target those skills necessary for positive development.

References

Abrahams, B. S., & Geschwind, D. H. (2008). Advances in autism genetics: on the threshold of a new neurobiology. *Nat Rev Genet*, *9*(5), 341-355. https://doi.org/10.1038/nrg2346

Adams, J. B., Johansen, L. J., Powell, L. D., Quig, D., & Rubin, R. A. (2011). Gastrointestinal flora and gastrointestinal status in children with autism -- comparisons to typical children and correlation with autism severity. *BMC Gastroenterology*, *11*, 22. https://doi.org/https://doi.org/10.1186/1471-230X-11-22

Amaral, D. G., Schumann, C. M., & Nordahl, C. W. (2008). Neuroanatomy of autism. *Trends Neurosci*, *31*(3), 137-145. https://doi.org/10.1016/j.tins.2007.12.005

American Psychiatric Association. (1994). *Diagnostic and Statistical Manual of Mental Disorders* (4 ed.).

American Psychiatric Association. (2013). *Diagnostic and Statistical Manual of Mental Disorders* (5 ed.). https://doi.org/https://doi.org/10.1176/appi.books.9780890425596

Ashwood, P., Wills, S., & Van de Water, J. (2006). The immune response in autism: a new frontier for autism research. *J Leukoc Biol*, *80*(1), 1-15. https://doi.org/10.1189/jlb.1205707

Bailey, A., Le Couteur, A., Gottesman, I., Bolton, P., Simonoff, E., Yuzda, E., & Rutter, M. (1995). Autism as a strongly genetic disorder: evidence from a British twin study. *Psychol Med*, *25*(1), 63-77. https://doi.org/10.1017/s0033291700028099

Bauman, M. L. (2010). Medical Comorbidities in Autism: Challenges to Diagnosis and Treatment. *Neurotherapeutics*, *7*(3), 320-327. https://doi.org/10.1016/j.nurt.2010.06.001

Bauminger, N., & Kasari, C. (1999). Brief report: Theory of mind in high-functioning children with autism. *J Autism Dev Disord*, *29*(1), 81-86. https://doi.org/10.1023/A:1025974701090

Beach Center of Disability. (2012). *The Family Quality of Life Scale (FQOL).* https://doi.org/https://dx.doi.org/10.13072/midss.327

Bedell, G. (2011). *The Child and Adolescent Scale of Participation (CASP): Administration and Scoring Guidelines*

Betancur, C., Leboyer, M., & Gillberg, C. (2002). Increased Rate of Twins among Affected Sibling Pairs with Autism. *Am J Hum Genet*, *70*(5), 1381-1383. https://doi.org/10.1086/340364

Bieleninik, L., Geretsegger, M., Mössler, K., Assmus, J., Thompson, G., Gattino, G., Elefant, C., Gottfried, T., Igliozzi, R., Muratori, F., Suvini, F., Kim, J., Crawford, M. J., Odell-Miller, H., Oldfield, A., Casey, Ó., Finnemann, J., Carpente, J., Park, A. L., . . . Gold, C. (2017). Effects of Improvisational Music Therapy vs Enhanced Standard Care on Symptom Severity Among Children With Autism Spectrum Disorder: The TIME-A Randomized Clinical Trial. *JAMA*, *318*(6), 525-535. https://doi.org/10.1001/jama.2017.9478

Bishop, D. V. M. (2011). *The Children's Communication Checklist, Second edition*. Pearson Education.

Boucher, J. (2012). Research Review: Structural language in autistic spectrum disorder - characteristics and causes. *J Child Psychol Psychiatry*, *53*(3), 219-233. https://doi.org/10.1111/j.1469-7610.2011.02508.x

Bourne, L., Mandy, W., & Bryant-Waugh, R. (2022). Avoidant/restrictive food intake disorder and severe food selectivity in children and young people with autism: A scoping review. *Dev Med Child Neurol*, *64*(6), 691-700. https://doi.org/10.1111/dmcn.15139

Brignell, A., May, T., Morgan, A. T., & Williams, K. (2019). Predictors and growth in receptive vocabulary from 4 to 8 years in children with and without autism spectrum disorder: A population-based study. *Autism*, *23*(5), 1322-1334. https://doi.org/10.1177/1362361318801617

Carr, A. (2015). *The Handbook of Child and Adolescent Clinical Psychology: A contextual approach* (3rd Edition ed.). Routledge. https://doi.org/https://doi.org/10.4324/9781315744230

Chandler, S., Carcani-rathwell, I., Charman, T., Pickles, A., Loucas, T., Meldrum, D., Simonoff, E., Sullivan, P., & Baird, G. (2013). Parent-Reported Gastro-intestinal Symptoms in Children with Autism Spectrum Disorders. *Journal of Autism and Developmental Disorders*, *43*(12), 2737-2747. https://doi.org/https://doi.org/10.1007/s10803-013-1768-0

Charman, T., Drew, A., Baird, C., & Baird, G. (2003). Measuring early language development in preschool children with autism spectrum disorder using the MacArthur Communicative Development Inventory (Infant Form). *J. Child Lang*, *30*(1), 213-236. https://doi.org/10.1017/S0305000902005482

Chasson, G. S., Harris, G. E., & Neely, W. J. (2007). Cost Comparison of Early Intensive Behavioral Intervention and Special Education for Children with Autism. *Journal of child and family studies*, *16*(3), 401-413. https://doi.org/10.1007/s10826-006-9094-1

Chaste, P., & Leboyer, M. (2012). Autism risk factors: genes, environment, and gene-environment interactions. *Dialogues Clin Neurosci*, *14*(3), 281-292. https://doi.org/10.31887/DCNS.2012.14.3/pchaste

Chen, C.-Y. P., Chen, K.-H. M., Liu, C.-Y. P., Huang, S.-L. M., & Lin, K.-M. M. D. M. P. H. (2009). Increased Risks of Congenital, Neurologic, and Endocrine Disorders Associated with Autism in Preschool Children: Cognitive Ability Differences. *J Pediatr*, *154*(3), 345-350.e341. https://doi.org/10.1016/j.jpeds.2008.09.043

Constantino, J. N., & Gruber, C. P. (2005). *Social Responsiveness Scale (SRS)*. Western Psychological Services.

Cozby, P. C., & Bates, S. C. (2020). *Methods in behavioral research* (Fourteenth edition, [International student edition]. ed.). McGraw-Hill Education.

Cumming, G., & Calin-Jageman, R. (2017). *Introduction to the new statistics : estimation, open science, and beyond*. Routledge.

De Giacomo, A., & Fombonne, E. (1998). Parental recognition of developmental abnormalities in autism. *Eur Child Adolesc Psychiatry*, *7*(3), 131-136. https://doi.org/10.1007/s007870050058

Dove, G. (2019). Language influences social cognition: Comment on “Words as social tools: Language, sociality and inner grounding in abstract concepts” by Anna M. Borghi et al. *Phys Life Rev*, *29*, 169-171. https://doi.org/10.1016/j.plrev.2019.03.007

Dunn, L. M., & Dunn, D. M. (2007). *The Peabody Picture Vocabulary Test, Fourth Edition*. NCS Pearson.

Eldevik, S., Hastings, R. P., Hughes, J. C., Jahr, E., Eikeseth, S., & Cross, S. (2010). Using Participant Data to Extend the Evidence Base for Intensive Behavioral Intervention for Children With Autism. *Am J Intellect Dev Disabil*, *115*(5), 381-405. https://doi.org/10.1352/1944-7558-115.5.381

Falter, C. M., Plaisted, K. C., & Davis, G. (2007). Visuo-spatial Processing in Autism—Testing the Predictions of Extreme Male Brain Theory. *J Autism Dev Disord*, *38*(3), 507-515. https://doi.org/10.1007/s10803-007-0419-8

Fisher, N., Happé, F., & Dunn, J. (2005). The relationship between vocabulary, grammar, and false belief task performance in children with autistic spectrum disorders and children with moderate learning difficulties. *J Child Psychol Psychiatry*, *46*(4), 409-419. https://doi.org/10.1111/j.1469-7610.2004.00371.x

Fombonne, E. (1999). The epidemiology of autism: a review. *Psychol. Med*, *29*(4), 769-786. https://doi.org/10.1017/S0033291799008508

Gassner, L., Geretsegger, M., & Mayer-Ferbas, J. (2021). Effectiveness of music therapy for autism spectrum disorder, dementia, depression, insomnia and schizophrenia: Update of systematic reviews. https://doi.org/https://doi.org/10.1093/eurpub/ckab042

Geretsegger, M., Elefant, C., Mössler, K. A., Gold, C., & Gold, C. (2014). Music therapy for people with autism spectrum disorder. *Cochrane Database Syst Rev*, *2016*(3), CD004381-CD004381. https://doi.org/10.1002/14651858.CD004381.pub3

Geurts, H. M., & Embrechts, M. (2008). Language profiles in ASD, SLI, and ADHD. *J Autism Dev Disord*, *38*(10), 1931-1943. https://doi.org/10.1007/s10803-008-0587-1

Greenberg, D. A., Hodge, S. E., Sowinski, J., & Nicoll, D. (2001). Excess of Twins among Affected Sibling Pairs with Autism: Implications for the Etiology of Autism. *Am J Hum Genet*, *69*(5), 1062-1067. https://doi.org/10.1086/324191

Guttmann-Steinmetz, S., Gadow, K. D., & DeVincent, C. J. (2009). Oppositional Defiant and Conduct Disorder Behaviors in Boys With Autism Spectrum Disorder With and Without Attention-Deficit Hyperactivity Disorder Versus Several Comparison Samples. *J Autism Dev Disord*, *39*(7), 976-985. https://doi.org/10.1007/s10803-009-0706-7

Habibi, A., Damasio, A., Ilari, B., Veiga, R., Joshi, A. A., Leahy, R. M., Haldar, J. P., Varadarajan, D., Bhushan, C., & Damasio, H. (2018). Childhood Music Training Induces Change in Micro and Macroscopic Brain Structure: Results from a

Longitudinal Study. *Cereb Cortex*, *28*(12), 4336-4347. https://doi.org/10.1093/cercor/bhx286

Hallmayer, J., Glasson, E. J., Bower, C., Petterson, B., Croen, L., Grether, J., & Risch, N. (2002). On the Twin Risk in Autism. *Am J Hum Genet*, *71*(4), 941-946. https://doi.org/10.1086/342990

Hartley, S. L., Sikora, D. M., & McCoy, R. (2008). *Prevalence and risk factors of maladaptive behaviour in young children with Autistic Disorder* [819-829]. [Oxford, England] :.

Hyde, K. L., Lerch, J., Norton, A., Forgeard, M., Winner, E., Evans, A. C., & Schlaug, G. (2009). Musical Training Shapes Structural Brain Development. *J Neurosci*, *29*(10), 3019-3025. https://doi.org/10.1523/JNEUROSCI.5118-08.2009

Jacobson, J. W., Mulick, J. A., & Green, G. (1998). Cost-benefit estimates for early intensive behavioral intervention for young children with autism-general model and single state case. *Behavioral Interventions*, *13*(4), 201-226. https://doi.org/10.1002/(SICI)1099-078X(199811)13:4<201::AID-BIN17>3.0.CO2-R

Jacquemont, S., Coe, Bradley P., Hersch, M., Duyzend, Michael H., Krumm, N., Bergmann, S., Beckmann, Jacques S., Rosenfeld, Jill A., & Eichler, Evan E. (2014). A Higher Mutational Burden in Females Supports a "Female Protective Model" in Neurodevelopmental Disorders. *Am J Hum Genet*, *94*(3), 415-425. https://doi.org/10.1016/j.ajhg.2014.02.001

Jang, J., Dixon, D. R., Tarbox, J., & Granpeesheh, D. (2011). *Symptom severity and challenging behavior in children with ASD* [1028-1032]. [Amsterdam] :.

Just, M. A., Cherkassky, V. L., Keller, T. A., & Minshew, N. J. (2004). Cortical activation and synchronization during sentence comprehension in high-functioning autism:

evidence of underconnectivity. *Brain*, *127*(8), 1811-1821. https://doi.org/10.1093/brain/awh199

Kana, R. K., Sartin, E. B., Stevens, C., Deshpande, H. D., Klein, C., Klinger, M. R., & Klinger, L. G. (2017). Neural networks underlying language and social cognition during self-other processing in Autism spectrum disorders. *Neuropsychologia*, *102*, 116-123. https://doi.org/10.1016/j.neuropsychologia.2017.06.008

Kanner, L. (1943). Autistic disturbances of affective contact. *Nervous child*, *2*(3), 217-250.

Kennison, S. M. (2014). *Introduction to language development*. SAGE Publications, Ltd.

Kirkham, P. (2017). 'The line between intervention and abuse' – autism and applied behaviour analysis. *History of the human sciences*, *30*(2), 107-126. https://doi.org/10.1177/0952695117702571

Koomar, T., Thomas, T. R., Pottschmidt, N. R., Lutter, M., & Michaelson, J. J. (2021). Estimating the Prevalence and Genetic Risk Mechanisms of ARFID in a Large Autism Cohort. *Frontiers in psychiatry*, *12*, 668297-668297. https://doi.org/10.3389/fpsyt.2021.668297

Kover, S. T., McDuffie, A. S., Hagerman, R. J., & Abbeduto, L. (2013). Receptive Vocabulary in Boys with Autism Spectrum Disorder: Cross-Sectional Developmental Trajectories. *J Autism Dev Disord*, *43*(11), 2696-2709. https://doi.org/10.1007/s10803-013-1823-x

Kung, K. T. F., Spencer, D., Pasterski, V., Neufeld, S., Glover, V., O'Connor, T. G., Hindmarsh, P. C., Hughes, I. A., Acerini, C. L., & Hines, M. (2016). No relationship between prenatal androgen exposure and autistic traits: convergent evidence from studies of children with congenital adrenal hyperplasia and of amniotic testosterone concentrations in typically developing children. *J Child Psychol Psychiatr*, *57*(12), 1455-1462. https://doi.org/10.1111/jcpp.12602

Kupferstein, H. (2018). Evidence of increased PTSD symptoms in autistics exposed to applied behavior analysis. *Advances in Autism*, *4*(1), 19-29. https://doi.org/10.1108/AIA-08-2017-0016

La Malfa, G., Lassi, S., Bertelli, M., Salvini, R., & Placidi, G. F. (2004). Autism and intellectual disability: a study of prevalence on a sample of the Italian population. *J Intellect Disabil Res*, *48*(3), 262-267. https://doi.org/10.1111/j.1365-2788.2003.00567.x

Lainhart, J. E., & Folstein, S. E. (1994). AFFECTIVE-DISORDERS IN PEOPLE WITH AUTISM - A REVIEW OF PUBLISHED CASES. *J Autism Dev Disord*, *24*(5), 587-601. https://doi.org/10.1007/BF02172140

LeClerc, S., & Easley, D. (2015). Pharmacological therapies for autism spectrum disorder: a review. *P T*, *40*(6), 389-397.

Levisohn, P. M. (2007). The autism-epilepsy connection. *Epilepsia*, *48*(s9), 33-35. https://doi.org/10.1111/j.1528-1167.2007.01399.x

Lim, H. A. (2010). Effect of "Developmental Speech and Language Training Through Music" on Speech Production in Children with Autism Spectrum Disorders. *J Music Ther*, *47*(1), 2-26. https://doi.org/10.1093/jmt/47.1.2

Lim, H. A., & Draper, E. (2011). The Effects of Music Therapy Incorporated with Applied Behavior Analysis Verbal Behavior Approach for Children with Autism Spectrum Disorders. *J Music Ther*, *48*(4), 532-550. https://doi.org/10.1093/jmt/48.4.532

Lopata, C., Toomey, J. A., Fox, J. D., Volker, M. A., Chow, S. Y., Thomeer, M. L., Lee, G. K., Rodgers, J. D., McDonald, C. A., & Smerbeck, A. M. (2010). Anxiety and Depression in Children with HFASDs: Symptom Levels and Source Differences. *J Abnorm Child Psychol*, *38*(6), 765-776. https://doi.org/10.1007/s10802-010-9406-1

Lord, C., & Jones, R. M. (2012). Annual Research Review: Re-thinking the classification of autism spectrum disorders. *Journal of Child Psychology and Psychiatry*, *53*(5), 490-509. https://doi.org/10.1111/j.1469-7610.2012.02547.x

Luyster, R. J., Kadlec, M. B., Carter, A., & Tager-Flusberg, H. (2008). Language Assessment and Development in Toddlers with Autism Spectrum Disorders. *J Autism Dev Disord*, *38*(8), 1426-1438. https://doi.org/10.1007/s10803-007-0510-1

Lyall, K., Croen, L., Daniels, J., Fallin, M. D., Ladd-Acosta, C., Lee, B. K., Park, B. Y., Snyder, N. W., Schendel, D., Volk, H., Windham, G. C., & Newschaffer, C. (2017). The Changing Epidemiology of Autism Spectrum Disorders. *Annu Rev Public Health*, *38*(1), 81-102. https://doi.org/10.1146/annurev-publhealth-031816-044318 (Annual Review of Public Health)

Magnuson, K. M., & Constantino, J. N. (2011). Characterization of Depression in Children With Autism Spectrum Disorders. *J Dev Behav Pediatr*, *32*(4), 332-340. https://doi.org/10.1097/DBP.0b013e318213f56c

Makrygianni, M. K., Gena, A., Katoudi, S., & Galanis, P. (2018). The effectiveness of applied behavior analytic interventions for children with Autism Spectrum Disorder: A meta-analytic study. *Research in autism spectrum disorders*, *51*, 18-31. https://doi.org/10.1016/j.rasd.2018.03.006

Mannion, A., Leader, G., & Healy, O. (2013). An investigation of comorbid psychological disorders, sleep problems, gastrointestinal symptoms and epilepsy in children and adolescents with Autism Spectrum Disorder. *Research in autism spectrum disorders*, *7*(1), 35-42. https://doi.org/10.1016/j.rasd.2012.05.002

Maras, A., Schroder, C. M., Malow, B. A., Findling, R. L., Breddy, J., Nir, T., Shahmoon, S., Zisapel, N., & Gringras, P. (2018). Long-Term Efficacy and Safety of Pediatric Prolonged-Release Melatonin for Insomnia in Children with Autism Spectrum

Disorder. *J Child Adolesc Psychopharmacol*, *28*(10), 699-710. https://doi.org/10.1089/cap.2018.0020

Marquez-Garcia, A. V., Magnuson, J., Morris, J., Iarocci, G., Doesburg, S., & Moreno, S. (2021). Music Therapy in Autism Spectrum Disorder: a Systematic Review. *Review journal of autism and developmental disorders*, *9*(1), 91-107. https://doi.org/10.1007/s40489-021-00246-x

Masi, A., DeMayo, M. M., Glozier, N., & Guastella, A. J. (2017). An Overview of Autism Spectrum Disorder, Heterogeneity and Treatment Options. *Neurosci Bull*, *33*(2), 183-193. https://doi.org/10.1007/s12264-017-0100-y

Matson, J. L., & Nebel-Schwalm, M. S. (2007). Comorbid psychopathology with autism spectrum disorder in children: An overview. *Res Dev Disabil*, *28*(4), 341-352. https://doi.org/10.1016/j.ridd.2005.12.004

Matson, J. L., & Shoemaker, M. (2009). Intellectual disability and its relationship to autism spectrum disorders. *Res Dev Disabil*, *30*(6), 1107-1114. https://doi.org/10.1016/j.ridd.2009.06.003

McClintock, K., Hall, S., & Oliver, C. (2003). Risk markers associated with challenging behaviours in people with intellectual disabilities: a meta-analytic study. *J Intellect Disabil Res*, *47*(6), 405-416. https://doi.org/10.1046/j.1365-2788.2003.00517.x

McDaniel, J., Yoder, P., Woynaroski, T., & Watson, L. R. (2018). Predicting Receptive-Expressive Vocabulary Discrepancies in Preschool Children With Autism Spectrum Disorder. *J Speech Lang Hear Res*, *61*(6), 1426-1439. https://doi.org/10.1044/2018_JSLHR-L-17-0101

McElhanon, B. O., McCracken, C., Karpen, S., & Sharp, W. G. (2014). Gastrointestinal Symptoms in Autism Spectrum Disorder: A Meta-analysis. *Pediatrics*, *133*(5), 872-883. https://doi.org/10.1542/peds.2013-3995

Mirenda, P. (2003). Toward Functional Augmentative and Alternative Communication for Students with Autism: Manual Signs, Graphic Symbols, and Voice Output Communication Aids. In (Vol. 34, pp. 203-216).

Mitchell, S., Brian, J., Zwaigenbaum, L., Roberts, W., Szatmari, P., Smith, I., & Bryson, S. (2006). Early language and communication development of infants later diagnosed with autism spectrum disorder. *J Dev Behav Pediatr*, *27*(2), S69-S78. https://doi.org/10.1097/00004703-200604002-00004

Mooney, E. L., Gray, K. M., & Tonge, B. J. (2006). Early features of autism - Repetitive behaviours in young children. *Eur Child Adolesc Psychiatry*, *15*(1), 12-18. https://doi.org/10.1007/s00787-006-0499-6

Murphy, O., Healy, O., & Leader, G. (2009). Risk factors for challenging behaviors among 157 children with autism spectrum disorder in Ireland. *Research in autism spectrum disorders*, *3*(2), 474-482. https://doi.org/10.1016/j.rasd.2008.09.008

Nordahl Hansen, A. J. (2015). *Assessment of language and social communication in children with autism : measurement issues* [PhD, University of Oslo]. Oslo.

Pellicano, E. (2010). The Development of Core Cognitive Skills in Autism: A 3-Year Prospective Study. *Child Dev*, *81*(5), 1400-1416. https://doi.org/10.1111/j.1467-8624.2010.01481.x

Peters-Scheffer, N., Didden, R., Korzilius, H., & Sturmey, P. (2011). A meta-analytic study on the effectiveness of comprehensive ABA-based early intervention programs for children with Autism Spectrum Disorders. *Research in autism spectrum disorders*, *5*(1), 60-69. https://doi.org/10.1016/j.rasd.2010.03.011

Rapin, I., & Dunn, M. (2003). Update on the language disorders of individuals on the autistic spectrum. *Brain Dev*, *25*(3), 166-172. https://doi.org/10.1016/S0387-7604(02)00191-2

R Core Team (2022). R: A language and environment for statistical computing. R Foundation for Statistical Computing, Vienna, Austria. URL https://www.R-project.org/.

Reindal, L., Nærland, T., Weidle, B., Lydersen, S., Andreassen, O. A., & Sund, A. M. (2021). Structural and Pragmatic Language Impairments in Children Evaluated for Autism Spectrum Disorder (ASD).

Richards, C., Oliver, C., Nelson, L., & Moss, J. (2012). Self‐injurious behaviour in individuals with autism spectrum disorder and intellectual disability: Self-injury in autism spectrum disorder. *Journal of intellectual disability research*, *56*(5), 476-489. https://doi.org/10.1111/j.1365-2788.2012.01537.x

Rommelse, N. N. J., Franke, B., Geurts, H. M., Hartman, C. A., & Buitelaar, J. K. (2010). Shared heritability of attention-deficit/hyperactivity disorder and autism spectrum disorder. *Eur Child Adolesc Psychiatry*, *19*(3), 281-295. https://doi.org/10.1007/s00787-010-0092-x

Rossignol, D. A., & Frye, R. E. (2011). Melatonin in autism spectrum disorders: a systematic review and meta-analysis. *Dev Med Child Neurol*, *53*(9), 783-792. https://doi.org/10.1111/j.1469-8749.2011.03980.x

Rossignol, D. A., & Frye, R. E. (2012). Mitochondrial dysfunction in autism spectrum disorders: a systematic review and meta-analysis. *Mol Psychiatry*, *17*(3), 290-314. https://doi.org/10.1038/mp.2010.136

Rudie, Jeffrey D., & Dapretto, M. (2013). Convergent Evidence of Brain Overconnectivity in Children with Autism? *Cell Rep*, *5*(3), 565-566. https://doi.org/10.1016/j.celrep.2013.10.043

Scheeren, A. M., de Rosnay, M., Koot, H. M., & Begeer, S. (2013). Rethinking theory of mind in high‐functioning autism spectrum disorder: Advanced theory of mind in autism. *Journal of Child Psychology and Psychiatry*, *54*(6), 628-635. https://doi.org/10.1111/jcpp.12007

Schroder, C. M., Banaschewski, T., Fuentes, J., Hill, C. M., Hvolby, A., Posserud, M.-B. R., & Bruni, O. (2021). Pediatric prolonged-release melatonin for insomnia in children and adolescents with autism spectrum disorders. https://doi.org/https://doi.org/10.1080/14656566.2021.1959549

Scott, R. C., & Tuchman, R. (2016). Epilepsy and autism spectrum disorders: Relatively related. *Neurology*, *87*(2), 130-131. https://doi.org/10.1212/WNL.0000000000002852

Sharda, M., Tuerk, C., Chowdhury, R., Jamey, K., Foster, N., Custo-Blanch, M., Tan, M., Nadig, A., & Hyde, K. (2018). Music improves social communication and auditory-motor connectivity in children with autism. *Transl Psychiatry*, *8*(1), 231-213. https://doi.org/10.1038/s41398-018-0287-3

Sigman, M., & McGovern, C. W. (2005). Improvement in cognitive and language skills from preschool to adolescence in autism. *J Autism Dev Disord*, *35*(1), 15-23. https://doi.org/10.1007/s10803-004-1027-5

Smith, V., Mirenda, P., & Zaidman-Zait, A. (2007). Predictors of Expressive Vocabulary Growth in Children With Autism. *J Speech Lang Hear Res*, *50*(1), 149-160. https://doi.org/10.1044/1092-4388(2007/013)

Sokolova, E. S., Sluiter-Oerlemans, A. M., Rommelse, N. N. J., Groot, P., Hartman, C. A., Glennon, J. C., Claassen, T., Heskes, T., & Buitelaar, J. K. (2017). A Causal and Mediation Analysis of the Comorbidity Between Attention Deficit Hyperactivity Disorder (ADHD) and Autism Spectrum Disorder (ASD). *J Autism Dev Disord*, *47*(6), 1595-1604. https://doi.org/10.1007/s10803-017-3083-7

Sparrow, S. S., Cicchetti, D., & Balla, D. A. (2005). *Vineland Adaptive Behavior Scales, Second Edition (Vineland-II)*.

Sundelin, H. E. K., Larsson, H., Lichtenstein, P., Almqvist, C., Hultman, C. M., Tomson, T., & Ludvigsson, J. F. (2016). Autism and epilepsy: A population-based nationwide cohort study. *Neurology*, *87*(2), 192-197. https://doi.org/10.1212/WNL.0000000000002836

Suren, P. (2006, 21.09.2020). *Autisme*. Folkehelseinstitutet Retrieved 03.10.2022 from https://www.fhi.no/fp/psykiskhelse/psykisk-helse-barn-unge/autisme---faktaark/

Thomas, C. J. (2009). *Analyses of five years of results from Project DATA (Developmentally Appropriate Treatment for Autism), a high -quality early childhood special education program for students with Autism Spectrum Disorders* ProQuest Dissertations Publishing].

Toth, K., Munson, J., Meltzoff, A. N., & Dawson, G. (2006). Early predictors of communication development in young children with autism spectrum disorder: Joint attention, imitation, and toy play. *J Autism Dev Disord*, *36*(8), 993-1005. https://doi.org/10.1007/s10803-006-0137-7

van der Gaag, R.-J. (2017). Autism Spectrum Disorders: Developmental History of a Concept. In (pp. 1-27). Cham: Springer International Publishing. https://doi.org/10.1007/978-3-319-42713-3_1

van Eijk, L., & Zietsch, B. P. (2021). Testing the extreme male brain hypothesis: Is autism spectrum disorder associated with a more male-typical brain? *Autism Res*, *14*(8), 1597-1608. https://doi.org/10.1002/aur.2537

Vickerstaff, S., Heriot, S., Wong, M., Lopes, A., & Dossetor, D. (2007). Intellectual ability, self-perceived social competence, and depressive symptomatology in children with high-functioning autistic spectrum disorders. *J Autism Dev Disord*, *37*(9), 1647-1664. https://doi.org/10.1007/s10803-006-0292-x

Vissers, M. E. P., Cohen, M. X., & Geurts, H. M. (2012). Brain connectivity and high functioning autism: a promising path of research that needs refined models, methodological convergence, and stronger behavioral links. *Neurosci Biobehav Rev*, *36*(1), 604-625. https://doi.org/10.1016/j.neubiorev.2011.09.003

Volden, J., Coolican, J., Garon, N., White, J., & Bryson, S. (2009). Brief Report: Pragmatic Language in Autism Spectrum Disorder: Relationships to Measures of Ability and Disability. *J Autism Dev Disord*, *39*(2), 388-393. https://doi.org/10.1007/s10803-008-0618-y

Walker, V. L., Carpenter, M. E., Lyon, K. J., Garcia, M., & Johnson, H. (2021). Coaching paraeducators to implement functional communication training involving augmentative and alternative communication for students with autism spectrum disorder. *Augment Altern Commun*, *37*(2), 129-140. https://doi.org/10.1080/07434618.2021.1909650

Warreyn, P., Paelt, S., & Roeyers, H. (2014). Social-communicative abilities as treatment goals for preschool children with autism spectrum disorder: the importance of imitation, joint attention, and play. *Dev Med Child Neurol*, *56*(8), 712-716. https://doi.org/10.1111/dmcn.12455

Wilkenfeld, D. A., & McCarthy, A. M. (2020). Ethical Concerns with Applied Behavior Analysis for Autism Spectrum "Disorder". *Kennedy Inst Ethics J*, *30*(1), 31-69. https://doi.org/10.1353/ken.2020.0000

Williams, D. (2010). Theory of own mind in autism: Evidence of a specific deficit in self-awareness? *Autism*, *14*(5), 474-494. https://doi.org/10.1177/1362361310366314

World Health Organization (WHO). (1993). *The ICD-10 classification of mental and behavioural disorders*. World Health Organization.

World Health Organization (WHO). (2019). *International statistical classification of diseases and related health problems* (11 ed.). https://doi.org/https://icd.who.int/

Zimmerman, A. W., Jyonouchi, H., Comi, A. M., Connors, S. L., Milstien, S., Varsou, A., & Heyes, M. P. (2005). Cerebrospinal Fluid and Serum Markers of Inflammation in Autism. *Pediatr Neurol*, *33*(3), 195-201. https://doi.org/10.1016/j.pediatrneurol.2005.03.014

Appendixes

Appendix 1: Ethical approval

REK
REGIONALE KOMITEER FOR MEDISINSK OG HELSEFAGLIG FORSKNINGSETIKK

Region:	Saksbehandler:	Telefon:	Vår dato:	Vår referanse:
REK sør-øst D	Silje U. Lauvrak	22845520	07.05.2021	246145

Christian Gold

Prosjektsøknad: M4A
Søknadsnummer: 246145
Forskningsansvarlig institusjon: NORCE Norwegian Research Centre AS
Samarbeidende forskningsansvarlige institusjoner: Universitetet i Bergen

Prosjektsøknad godkjennes med vilkår av REK

Søkers beskrivelse

Bakgrunn: Musikk har blitt brukt i flere tiår til å hjelpe mennesker med autismespektrumforstyrrelse (ASD). Musikkterapi lover å bli en effektiv form for intervensjon for ASD. Likevel, systematisk forskning på musikkterapiens effektivitet og biologisk virkemodus har kun nylig begynt.
Formål: Music for autism (M4A) er det første europeiske forskningsprosjekt som undersøker musikkterapiens effekt på sosial kommunikasjon; deltakelse; familiens livskvalitet; og psykisk helse; samt effekter på funksjonell hjernekonnektivitet og hjernestruktur, i barn med ASD.
Metode: Kartlegger-blindet, randomisert kontrollert kryssover-forsøk. Alle deltakere vil få klinisk kartlegging og hjerneskanninger (magnetresonanstomografi) hver tredje måned over en periode av ni måneder. I de første og siste 3 måneder vil deltakerne få ukentlige sesjoner med musikk eller lek (rekkefølge bestemt tilfeldig). På de midterste 3 månedene vil deltakerne få ingen terapi (washout-periode).
Populasjoner: M4A vil inkludere totalt 80 barn, alder 6-12 år, på alle kognitive funksjonsnivå, på to europeiske land (Østerrike og Norge).
Forventet utfall: Vi forventer musikkterapi å forbedre sosial kommunikasjon, deltakelse, psykisk helse, hjernekonnektivitet, og hjernemasse i utvalgte arealer. Vitenskapelig og populærvitenskapelig formidling skal skje gjennom publikasjoner, nasjonale og internasjonale konferanser, og flere kommunikasjonskanaler som TV, radio og sosiale medier, gjennom prosjektets forløp og etter prosjektets avslutning.
Nytteverdi: M4A vil levere verdifulle data som vil forbedre betydelig det teoretiske og empiriske grunnlag for anvendelse av musikkterapi. Prosjektet vil hjelpe å lukke viktige nåværende kunnskapshull på feltet og vil bidra til videre utvikling av personalisert klinisk praksis. Det vil også bidra til videre etablering av effektive, evidensbaserte intervensjoner for ASD.

Vi viser til søknad om forhåndsgodkjenning mottatt 17.03.2021.

REKs vurdering

Vi viser til søknad om forhandsgodkjenning av ovennevnte forskningsprosjekt. Søknaden ble behandlet av Regional komite for medisinsk og helsefaglig forskningsetikk (REK sør-øst D) i møtet 14.04.2021. Vurderingen er gjort med hjemmel i helseforskningsloven § 10.

Prosjektets formål er å undersøke musikkterapiens effekt på sosial kommunikasjon, deltakelse, familiens livskvalitet, psykisk helse, samt effekter på funksjonell hjernekonnektivitet og hjernestruktur, hos barn med autismespektrumforstyrrelse. Prosjektet gjøres i samarbeid med Østerrike.

Det skal inkluderes 80 barn i alderen 6-12 år med diagnose på autismespekteret, hvorav 40 barn i Norge. Informasjon om prosjektet vil bli gjort tilgjengelig på barnepsykiatriske poliklinikker i Bergen og omegn og ved autismeteamet ved spesialpoliklinikken PBU i Helse Bergen. Foresatte og unge vil der bli gitt informasjon for å kunne henvende seg ved interesse for deltakelse til studien.

Deltagelse innebærer at det gjøres klinisk kartlegging av barnet og tas bilder av hjernen (strukturell og funksjonell MRI) hver tredje måned i en periode på 9 måneder. I de første 3 månedene og de 3 siste månedene vil barna motta ukentlige sesjoner med enten musikkterapi eller terapi med lek (tilfeldig rekkefølge). Det vil tas videoopptak av terapisesjonene for å kvalitetssikre behandlingen. I de tre midterste månedene får barna ingen terapi. Det kan være aktuelt å overføre opplysninger og videoopptak til Østerrike som ledd i forskningssamarbeidet.

Det skal også tas hårprøver av barna for kortisolmåling. Prøvene skal overføres til Østerrike for analyse og skal deretter destrueres senest to måneder etter prøvetaking.

I tillegg skal barnas foreldre og lærere besvare spørreskjemaer som tar for seg kommunikasjon, deltakelse, livskvalitet, psykisk helse, uønskede effekter og preferanse.

Etter komiteens syn er dette et nyttig prosjekt som skal evaluere om musikkterapi har positiv effekt for barn med autismespekterforstyrrelser. Deltagelse vil dermed kunne være til direkte nytte for barna som deltar. Videre er prosjektet nytenkende ved at det også søker å forstå vikningsmekanismene bak musikkens betydning for helse, noe som er viktig for å forstå hvorfor noen har effekt av musikkterapi og andre ikke, slik at behandling i fremtiden bedre kan persontilpasses.

Komiteen har spesielt vurdert om vilkårene i helseforskningsloven § 18 for forskning som inkluderer mindreårige, er oppfylt:

a. eventuell risiko eller ulempe for personen er ubetydelig,

b. personen selv ikke motsetter seg det, og

c. det er grunn til å anta at resultatene av forskningen kan være til nytte for den aktuelle personen eller for andre personer med samme aldersspesifikke lidelse, sykdom, skade eller tilstand.

Loven stiller i tillegg krav om at tilsvarende forskning ikke kan gjennomføres på personer som ikke er mindreårige.

Intervensjonen med musikkterapi er etter komiteens vurdering forbundet med liten risiko, så fremt barna selv er innstilt på å gjennomføre denne form for terapi, jf. vilkår b.

Det foreligger per i dag ikke dokumenterte, uønskede bivirkninger ved en MR-undersøkelse. Selve undersøkelsen kan imidlertid oppleves som ubehagelig for mange. Det er i søknaden grundig redegjort for hvordan undersøkelsen skal gjennomføres, og det er lagt opp til at barna kan trene seg på MR-undersøkelsen i forkant ved å bli introdusert for omgivelsene til skanneren, selve skanneren og støyen fra den. Under selve skanningsøktene vil foreldrene til barna også få hørselsvern og få mulighet til å være med inne i skannerrommet hvis det er nødvendig. Skanningstiden vil holdes på et minimum som kreves for å innhente nødvendige data.

Med denne gjennomføringsplanen vurderer komiteen at MR-undersøkelsene ikke vil medfører spesiell påkjenning for barna, og at eventuell risiko eller ulempe for barna er ubetydelig i henhold til vilkår a. Komiteen forutsetter at undersøkelsen ikke gjøres dersom barna viser tegn til at de ikke vil, jamfør vilkår b, og også i tråd med det som er oppgitt i søknaden. Vilkår c om at prosjektet skal være nyttig for barna og denne gruppen barn i fremtiden anses også oppfylt, som redegjort for over.

Komiteen vurderer også at tilleggskravet om at tilsvarende forskning ikke kan gjennomføres på personer som ikke er mindreårige, er oppfylt ved at musikkterapi har vist å ha helsefremmende effekt for andre grupper, og at man nå skal undersøke om dette også gjelder for gruppen barn med autismeforstyrrelser.

I tillegg til god gjennomføringsplan for selve MR-undersøkelsene, finner komiteen at beredskapen i prosjektet for øvrig også er vel ivaretatt. Det skal opprettes en uavhengig data- og sikkerhetsmonitoreringskomité (DSMC) på tre personer med sterk metodisk og klinisk ekspertise. DSMC skal få ublindet tilgang til studiedata og skal også motta jevnlige oppdateringer om rekruttering, opptak av intervensjoner, uforutsette hendelser, uønskede hendelser og umiddelbar informasjon om alvorlige bivirkninger. Prosjektgruppen skal videre ha halvårlige møter med DSCM.

Etter en helhetlig vurdering har komiteen kommet til at nytten for barna ved å delta i prosjektet overstiger ulempene. Beredskapen er godt ivaretatt og deltagerne får god informasjon om hva deltagelse innebærer. Komiteen finner det dermed forsvarlig å gjennomføre prosjektet slik det er beskrevet i søknad og protokoll. Komiteen forutsetter at samtykke innhentes fra begge foresatte.

Komiteen har noen merknader til informasjonsskrivet:

- For å gjøre skrivet mer nøytralt bes det om at «Kjære foreldre/verge» tas bort. Det bes også om at setningene "Vi inviterer deg og ditt barn til å delta i studien ovenfor. Informasjonen om dette foregår i en detaljert diskusjon med studiekoordinator, Marianna Ruiz (xxxx@norceresearch.no)." endres til «Dette er et spørsmål til deg og ditt barn om å delta i studien ovenfor. Du kan få muntlig informasjon, med mulighet til å stille spørsmål, fra studiekoordinator, Marianna Ruiz (xxxx@norceresearch.no)"».

- Andre avsnitt om muligheten til å trekke seg kan tas bort siden det kommer informasjon om dette senere i skivet.

- Det står i skrivet at "Spørreskjemaene brukes til å registrere visse atferdsrelaterte variabler, og med ett unntak blir de fylt ut av lærerne til utdanningsinstitusjonen barnet ditt går på». Komiteen ber om at det fremkommer hva det ene unntaket er. I tillegg bes det om at 'utdanningsinstitusjonen' erstattes med 'skolen'.

- Da også barnas lærere skal delta i prosjektet ved å besvare spørreskjemaer, må det også utarbeides et eget informasjonsskriv til denne gruppen der det fremkommer hva prosjektet går ut på og hva deltagelse i prosjektet innebærer for dem.

- Det oppgis at: «Ved å delta i denne studien har barnet ditt mulighet, på grunn av den individuelt tilpassede støtten, til å takle vansker forårsaket av en autismespektrumforstyrrelse, for eksempel å utvide sine sosiale og kommunikasjonsevner, å øke selvtilliten og å utvikle kreativiteten og fleksibilitet». Etter komiteens syn er dette å forskuttere resultatet av prosjektet, da det er nettopp hvorvidt musikkterapi har effekt på denne gruppen, som skal undersøkes. Komiteen ber om at setningen omformuleres og nøytraliseres.

- Det står i skrivet at opplysningene som registreres planlegges brukt til 2034. Komiteen ber om at dette rettes til 2024, i henhold til prosjektperioden.

- Siste setning i skrivet til barna er «Dersom du ikke deltar vil du ikke møte terapeuten eller psykologen.» Komiteen kan ikke se nødvendigheten av å ha med dette, og ber om at setningen tas bort.

Komiteen legger også merke til at spørreskjemaene som skal benyttes i prosjektet ikke er vedlagt søknaden, og ber om at disse ettersendes.

På bakgrunn av ovennevnte merknader godkjennes prosjektet på følgende vilkår:

- Informasjonsskrivene må revideres i henhold til komiteens merknader.

- Spørreskjemaene som skal benyttes i prosjektet må ettersendes komiteen.

Reviderte informasjonsskriv og spørreskjemaer ettersendes ved å benytte skjemaet 'Endring og/eller henvendelse' (som ligger under fanen 'Andre valg' når innlogget i REK-portalen og i det aktuelle prosjektet).

Vedtak

REK har gjort en helhetlig forskningsetisk vurdering av alle prosjektets sider. Prosjektet godkjennes med hjemmel i helseforskningsloven § 10, under forutsetning av at ovennevnte vilkår er oppfylt.

Vi gjør samtidig oppmerksom på at etter ny personopplysningslov må det også foreligge et behandlingsgrunnlag etter personvernforordningen. Det må forankres i egen institusjon.

I tillegg til vilkår som fremgår av dette vedtaket, er godkjenningen gitt under forutsetning av at prosjektet gjennomføres slik det er beskrevet i søknad og protokoll, og de bestemmelser som følger av helseforskningsloven med forskrifter.

Med hjemmel i helseforskningsloven § 29 tillater komiteen at humant biologisk materiale utføres til utlandet.

Tillatelsen gjelder til 30.04.2024. Av dokumentasjonshensyn skal opplysningene likevel bevares inntil 30.04.2029. Forskningsfilen skal oppbevares atskilt i en nøkkel- og en opplysningsfil. Opplysningene skal deretter slettes eller anonymiseres, senest innen et halvt år fra denne dato.

Forskningsprosjektets data skal oppbevares forsvarlig, se personopplysningsforskriften kapittel 2, og Helsedirektoratets veileder for «Personvern og informasjonssikkerhet i forskningsprosjekter innenfor helse og omsorgssektoren».

Komiteens avgjørelse var enstemmig.

Med vennlig hilsen

Finn Wisløff
Professor em. dr. med.
Leder

Silje U. Lauvrak
Seniorrådgiver

Kopi til: NORCE Norwegian Research Centre AS; Universitetet i Berge

Sluttmelding
Prosjektleder skal sende sluttmelding til REK på eget skjema via REK-portalen senest senest 6 måneder etter sluttdato 30.04.2024, jf. helseforskningsloven § 12. Dersom prosjektet ikke starter opp eller gjennomføres meldes dette også via skjemaet for sluttmelding.

Søknad om endring
Dersom man ønsker å foreta vesentlige endringer i formål, metode, tidsløp eller organisering må prosjektleder sende søknad om endring via portalen på eget skjema til REK, jf. helseforskningsloven § 11.

Klageadgang
Du kan klage på REKs vedtak, jf. forvaltningsloven § 28 flg. Klagen sendes på eget skjema via REK portalen. Klagefristen er tre uker fra du mottar av dette brevet. Dersom REK opprettholder vedtaket, sender REK klagen videre til Den nasjonale forskningsetiske komité for medisin og helsefag (NEM) for endelig vurdering, jf. forskningsetikkloven § 10 og helseforskningsloven § 10.

Kopi til:

Receptive vocabulary, social communication and behaviour in children diagnosed with autism spectrum disorder

NORCE Norwegian Research Centre AS
Universitetet i Bergen
Karsten Specht

Beschluss der Ethikkommission Decision of the Ethics Committee	universität wien Ethikkommission

Antragsteller*in / Applicant: **Assoz. Prof. Giorgia Silani, Privatdoz. PhD**
Bearbeitungsnummer / Reference Number: **00634**
Projekttitel / Title of Project: **Music for Autism (M4A) – A randomized controlled trial examining clinical effects and biological basis of the effects of Music therapy on children with Autism Spectrum disorder.**

Die Stellungnahme der Ethikkommission erfolgt aufgrund folgender eingereichter Unterlagen / The decision of the Ethics Committee is based on the following documents:

20.02.2021

- CASP-Administration-Scoring-Guidelines-8-19-11
- CASP-Youth-Version-Revised-12-29-11
- Family Quality of Life Psychometric Characteristics and Scoring Key
- Jan_2021_Antragsformular_Ethikkommission2
- M4A full proposal submitted version
- M4A_TeilnehmerInneninformation_und_Einverständniserklärung_V2
- Questionnaires List

16.04.2021

- Cover letter_M4A
- M4A_Teilnehmerinformation_Kinder
- M4A_TeilnehmerInneninformation_und_Einverständniserklärung_V4

Die Kommission fasst folgenden Beschluss (mit X markiert) / The Ethics Committee has made the following decision (marked with an X):

☒ Zustimmung: Es besteht kein ethischer Einwand gegen die Durchführung der Studien. / Consent: There is no ethical objection to conduct the study as proposed.

☐ Negative Beurteilung: Der Antrag wird von der Ethikkommission abgelehnt. / Negative evaluation: The proposal is rejected by the Ethics Committee.

Inhaltliche Abänderungen müssen der Ethikkommission vorgelegt werden. / Amendments to the content must be presented to the Ethics Committee.

Unterschrift / Signature	Datum / Date
eigenhändig: Martin Voracek	*21.04.2021*

Vorsitzender der Ethikkommission / Chair of the Ethics Committee
Univ.-Prof. MMag. DDDr. Martin Voracek

Appendix 2: R-code

```
#1. Load excel, set working directory and import excel data

install.packages("easypackages")

library(easypackages)

packages("readxl", "datasets", "ggplot2", "ggpubr", "ggsci", "ggsignif",
         "graphics", "grDevices", "labeling", "methods", "openxlsx",
"psych",
         "qgraph", "moments", "summarytools", "tidyverse", "outliers",
"car",
         "astatur", "GGally", "Hmisc", "DescTools", "skimr",
"correlation",
         "rstatix", "dplyr")


setwd("~/Desktop/R_datasets")
data <- read_excel("data.xlsx")
dataf <- tibble(data)
view(dataf)

#2. Sample characteristics
dataf %>% select("Age") %>% descr
dataf %>% select("Gender", "City") %>% freq
dataf %>% select("ppvt", "vineland_v", "srs_t", "ccc_gcc", "ccc_iasi",
                 "fqol", "casp") %>% descr

#PPVT
dataf %>% select("ppvt") %>% count(ppvt >= 85 & ppvt <= 115)
dataf %>% select("ppvt") %>% count(ppvt >= 70 & ppvt <= 130)
dataf %>% select("ppvt") %>% count(ppvt <= 85)

#CCC-2
dataf %>% select(starts_with("ccc")) %>% descr
dataf %>% select("ccc_gcc") %>% count(ccc_gcc <= 55)
dataf %>% select("ccc_iasi", "ccc_gcc") %>% count(ccc_iasi <= 0 & ccc_gcc
<= 55)
dataf %>% select("ccc_gcc", "ccc_iasi") %>% count(ccc_iasi >= 9 & ccc_gcc
<= 55)

#CCC-2 structural language score
dataf %>% select("ccc_spo", "ccc_synt", "ccc_sem", "ccc_coh") %>%
  rowSums() %>%
    mean()
dataf %>% select("ccc_spo", "ccc_synt", "ccc_sem", "ccc_coh") %>%
  rowSums() %>%
    sd()

#CCC-2 pragmatic language score
dataf %>% select("ccc_ini", "ccc_ster", "ccc_cont", "ccc_nonv") %>%
  rowSums() %>%
    mean()
dataf %>% select("ccc_ini", "ccc_ster", "ccc_cont", "ccc_nonv") %>%
  rowSums() %>%
    sd()

#VABS-MBI
dataf %>% select("vineland_v") %>%  count(vineland_v >= 18)
```

```
#SRS
dataf %>% select(starts_with("srs")) %>% descr

#Relevant data about FQoL and CASP is already covered by sample
characteristics

#Boxplots to identify outliers

#PPVT
dataf %>% select("ppvt") %>%
  ggplot(aes(y=ppvt)) +
  labs(x="Combined", y="PPVT standard score") +
  geom_boxplot()
#One outlier was identified - identify its z-value
dataf %>% select("ppvt") %>% scale()

#VABS
dataf %>% select("vineland_v") %>%
  ggplot(aes(y=vineland_v)) +
  labs(x="Combined", y="VABS score") +
  geom_boxplot()

#CCC-GCC
dataf %>% select("ccc_gcc") %>%
  ggplot(aes(y=ccc_gcc)) +
  labs(x="Combined", y="GCC score") +
  geom_boxplot()

#CCC-IASI
dataf %>% select("ccc_iasi") %>%
  ggplot(aes(y=ccc_iasi)) +
  labs(x="Combined", y="IASI score") +
  geom_boxplot()

#SRS
dataf %>% select("srs_t") %>%
  ggplot(aes(y=srs_t)) +
  labs(x="Combined", y="SRS score") +
  geom_boxplot()

#CASP
dataf %>% select("casp") %>%
  ggplot(aes(y=casp)) +
  labs(x="Combined", y="CASP score") +
  geom_boxplot()

#FQoL
dataf %>% select("fqol") %>%
  ggplot(aes(y=fqol)) +
  labs(x="Combined", y="FQoL score") +
  geom_boxplot()
#Two outliers were identified - identify their z-value
dataf %>% select("fqol") %>% scale()

#Preliminary analysis

#Equal variance and normal distribution
```

```
#PPVT
ppvt_b <- c(dataf$ppvt[dataf$City=="b"])
ppvt_v <- c(dataf$ppvt[dataf$City=="v"])

ppvt_vector <- c(ppvt_b, ppvt_v)
ppvt_group_vector <- as.factor(c(rep(1, length(ppvt_b)),
                                 rep(2, length(ppvt_v))))
ppvt_variance <- leveneTest(ppvt_vector, ppvt_group_vector)
print(ppvt_variance)
ppvt_variance_pvalue <- ppvt_variance$`Pr(>F)`[1]

ppvt_b_sw <-shapiro.test(ppvt_b)
print(ppvt_b_sw)
ppvt_b_sw_pvalue <- ppvt_b_sw$`p.value`
ppvt_v_sw <-shapiro.test(ppvt_v)
print(ppvt_v_sw)
ppvt_v_sw_pvalue <- ppvt_v_sw$`p.value`

if(ppvt_variance_pvalue > 0.05 &
   ppvt_b_sw_pvalue > 0.05 &
   ppvt_v_sw_pvalue > 0.05) {
  ppvt_ttest <- t.test(ppvt_b, ppvt_v, alternative="two.sided",
var.equal=TRUE)
} else {
  ppvt_wilcox <- wilcox.test(ppvt_b, ppvt_v, exact=FALSE)
}

if(ppvt_variance_pvalue > 0.05 &
   ppvt_b_sw_pvalue > 0.05 &
   ppvt_v_sw_pvalue > 0.05) {
  print(ppvt_ttest)
} else {
  print(ppvt_wilcox)
}

#VABS-MBI
vabs_b <- c(data$vineland_v[data$City=="b"])
vabs_v <- c(data$vineland_v[data$City=="v"])

vabs_vector <- c(vabs_b, vabs_v)
vabs_group_vector <- as.factor(c(rep(1, length(vabs_b)),
                                 rep(2, length(vabs_v))))
vabs_variance <- leveneTest(vabs_vector, vabs_group_vector)
print(vabs_variance)
vabs_variance_pvalue <- vabs_variance$`Pr(>F)`[1]

vabs_b_sw <-shapiro.test(vabs_b)
print(vabs_b_sw)
vabs_b_sw_pvalue <- vabs_b_sw$`p.value`
vabs_v_sw <-shapiro.test(vabs_v)
print(vabs_v_sw)
vabs_v_sw_pvalue <- vabs_v_sw$`p.value`

if(vabs_variance_pvalue > 0.05 &
   vabs_b_sw_pvalue > 0.05 &
   vabs_v_sw_pvalue > 0.05) {
```

```
  vabs_ttest <- t.test(vabs_b, vabs_v, alternative="two.sided",
var.equal=TRUE)
} else {
  vabs_wilcox <- wilcox.test(vabs_b, vabs_v, exact=FALSE)
}

if(vabs_variance_pvalue > 0.05 &
   vabs_b_sw_pvalue > 0.05 &
   vabs_v_sw_pvalue > 0.05) {
  print(vabs_ttest)
} else {
  print(vabs_wilcox)
}

#CCC-GCC
ccc2_b <- c(data$ccc_gcc[data$City=="b"])
ccc2_v <- c(data$ccc_gcc[data$City=="v"])

ccc2_vector <- c(ccc2_b, ccc2_v)
ccc2_group_vector <- as.factor(c(rep(1, length(ccc2_b)),
                                 rep(2, length(ccc2_v))))
ccc2_variance <- leveneTest(ccc2_vector, ccc2_group_vector)
print(ccc2_variance)
ccc2_variance_pvalue <- ccc2_variance$`Pr(>F)`[1]

ccc2_b_sw <-shapiro.test(ccc2_b)
print(ccc2_b_sw)
ccc2_b_sw_pvalue <- ccc2_b_sw$`p.value`
ccc2_v_sw <-shapiro.test(ccc2_v)
print(ccc2_v_sw)
ccc2_v_sw_pvalue <- ccc2_v_sw$`p.value`

if(ccc2_variance_pvalue > 0.05 &
   ccc2_b_sw_pvalue > 0.05 &
   ccc2_v_sw_pvalue > 0.05) {
  ccc2_ttest <- t.test(ccc2_b, ccc2_v, alternative="two.sided",
var.equal=TRUE)
} else {
  ccc2_wilcox <- wilcox.test(ccc2_b, ccc2_v, exact=FALSE)
}

if(ccc2_variance_pvalue > 0.05 &
   ccc2_b_sw_pvalue > 0.05 &
   ccc2_v_sw_pvalue > 0.05) {
  print(ccc2_ttest)
} else {
  print(ccc2_wilcox)
}

#CCC-IASI
ccc_iasi_b <- c(data$ccc_iasi[data$City=="b"])
ccc_iasi_v <- c(data$ccc_iasi[data$City=="v"])

ccc_iasi_vector <- c(ccc_iasi_b, ccc_iasi_v)
ccc_iasi_group_vector <- as.factor(c(rep(1, length(ccc_iasi_b)),
                                     rep(2, length(ccc_iasi_v))))
ccc_iasi_variance <- leveneTest(ccc_iasi_vector, ccc_iasi_group_vector)
print(ccc_iasi_variance)
```

```
ccc_iasi_variance_pvalue <- ccc_iasi_variance$`Pr(>F)`[1]

ccc_iasi_b_sw <-shapiro.test(ccc_iasi_b)
print(ccc_iasi_b_sw)
ccc_iasi_b_sw_pvalue <- ccc_iasi_b_sw$`p.value`
ccc_iasi_v_sw <-shapiro.test(ccc_iasi_v)
print(ccc_iasi_v_sw)
ccc_iasi_v_sw_pvalue <- ccc_iasi_v_sw$`p.value`

if(ccc_iasi_variance_pvalue > 0.05 &
   ccc_iasi_b_sw_pvalue > 0.05 &
   ccc_iasi_v_sw_pvalue > 0.05) {
  ccc_iasi_ttest <- t.test(ccc_iasi_b, ccc_iasi_v,
alternative="two.sided", var.equal=TRUE)
} else {
  ccc_iasi_wilcox <- wilcox.test(ccc_iasi_b, ccc_iasi_v, exact=FALSE)
}

if(ccc_iasi_variance_pvalue > 0.05 &
   ccc_iasi_b_sw_pvalue > 0.05 &
   ccc_iasi_v_sw_pvalue > 0.05) {
  print(ccc_iasi_ttest)
} else {
  print(ccc_iasi_wilcox)
}

#SRS-t
srs_b <- c(data$srs_t[data$City=="b"])
srs_v <- c(data$srs_t[data$City=="v"])

srs_vector <- c(srs_b, srs_v)
srs_group_vector <- as.factor(c(rep(1, length(srs_b)),
                                rep(2, length(srs_v))))
srs_variance <- leveneTest(srs_vector, srs_group_vector)
print(srs_variance)
srs_variance_pvalue <- srs_variance$`Pr(>F)`[1]

srs_b_sw <-shapiro.test(srs_b)
print(srs_b_sw)
srs_b_sw_pvalue <- srs_b_sw$`p.value`
srs_v_sw <-shapiro.test(srs_v)
print(srs_v_sw)
srs_v_sw_pvalue <- srs_v_sw$`p.value`

if(srs_variance_pvalue > 0.05 &
   srs_b_sw_pvalue > 0.05 &
   srs_v_sw_pvalue > 0.05) {
  srs_ttest <- t.test(srs_b, srs_v, alternative="two.sided",
var.equal=TRUE)
} else {
  srs_wilcox <- wilcox.test(srs_b, srs_v, exact=FALSE)
}

if(srs_variance_pvalue > 0.05 &
   srs_b_sw_pvalue > 0.05 &
   srs_v_sw_pvalue > 0.05) {
  print(srs_ttest)
} else {
```

```
  print(srs_wilcox)
}

#CASP
casp_b <- c(data$casp[data$City=="b"])
casp_v <- c(data$casp[data$City=="v"])

casp_vector <- c(casp_b, casp_v)
casp_group_vector <- as.factor(c(rep(1, length(casp_b)),
                                 rep(2, length(casp_v))))
casp_variance <- leveneTest(casp_vector, casp_group_vector)
print(casp_variance)
casp_variance_pvalue <- casp_variance$`Pr(>F)`[1]

casp_b_sw <-shapiro.test(casp_b)
print(casp_b_sw)
casp_b_sw_pvalue <- casp_b_sw$`p.value`
casp_v_sw <-shapiro.test(casp_v)
print(casp_v_sw)
casp_v_sw_pvalue <- casp_v_sw$`p.value`

if(casp_variance_pvalue > 0.05 &
   casp_b_sw_pvalue > 0.05 &
   casp_v_sw_pvalue > 0.05) {
   casp_ttest <- t.test(casp_b, casp_v, alternative="two.sided",
var.equal=TRUE)
} else {
  casp_wilcox <- wilcox.test(casp_b, casp_v, exact=FALSE)
}

if(casp_variance_pvalue > 0.05 &
   casp_b_sw_pvalue > 0.05 &
   casp_v_sw_pvalue > 0.05) {
  print(casp_ttest)
} else {
  print(casp_wilcox)
}

#FQoL
fqol_b <- c(data$fqol[data$City=="b"])
fqol_v <- c(data$fqol[data$City=="v"])

fqol_vector <- c(fqol_b, fqol_v)
fqol_group_vector <- as.factor(c(rep(1, length(fqol_b)),
                                 rep(2, length(fqol_v))))
fqol_variance <- leveneTest(fqol_vector, fqol_group_vector)
print(fqol_variance)
fqol_variance_pvalue <- fqol_variance$`Pr(>F)`[1]

fqol_b_sw <-shapiro.test(fqol_b)
print(fqol_b_sw)
fqol_b_sw_pvalue <- fqol_b_sw$`p.value`
fqol_v_sw <-shapiro.test(fqol_v)
print(fqol_v_sw)
fqol_v_sw_pvalue <- fqol_v_sw$`p.value`

if(fqol_variance_pvalue > 0.05 &
   fqol_b_sw_pvalue > 0.05 &
```

```
   fqol_v_sw_pvalue > 0.05) {
  fqol_ttest <- t.test(fqol_b, fqol_v, alternative="two.sided",
var.equal=TRUE)
} else {
  fqol_wilcox <- wilcox.test(fqol_b, fqol_v, exact=FALSE)
}

if(fqol_variance_pvalue > 0.05 &
   fqol_b_sw_pvalue > 0.05 &
   fqol_v_sw_pvalue > 0.05) {
  print(fqol_ttest)
} else {
  print(fqol_wilcox)
}

#Shapiro-Wilk-test of normal distribution on the complete sample
dataf %>% select("ppvt", "vineland_v", "srs_t", "ccc_gcc", "ccc_iasi",
                 "fqol", "casp") %>% lapply(shapiro.test)


#Research question 1

#Correlation between PPVT and CCC-GCC
dataf %>% select("ppvt", "ccc_gcc") %>%
  correlation::cor_test(x="ppvt", y="ccc_gcc", method='spearman', ci=0.95)

#Correlation between PPVT and CCC-IASI
dataf %>% select("ppvt", "ccc_iasi") %>%
  correlation::cor_test(x="ppvt", y="ccc_iasi", method='spearman',
ci=0.95)

#Correlation by CCC-subdomain
dataf %>% select("ppvt", starts_with("ccc")) %>%
  cor_mat(method="spearman", conf.level=0.95) %>%
  cor_get_pval

#Correlation between CCC-synt and CCC-coh
dataf %>% select("ccc_synt", "ccc_coh") %>%
  correlation::cor_test(x="ccc_synt", y="ccc_coh", method='pearson', ci=.
95)

#Correlation between PPVT-4 and structural language
struc_lang <- dataf %>% select("ccc_spo", "ccc_synt", "ccc_sem",
"ccc_coh") %>%
  rowSums()
cor.test(struc_lang, data$ppvt, method='spearman')
SpearmanRho(struc_lang, data$ppvt, conf.level=0.95)

#Correlation between PPVT-4 and pragmatic language
prag_lang <- dataf %>% select("ccc_ini", "ccc_ster", "ccc_cont",
"ccc_nonv") %>%
  rowSums()
cor.test(prag_lang, data$ppvt, method='spearman')
SpearmanRho(prag_lang, data$ppvt, conf.level=0.95)

#Get CI's for significant correlations
dataf %>% select("ppvt", "ccc_synt") %>%
```

```
  correlation::cor_test(x="ppvt", y="ccc_synt", method='spearman',
ci=0.95)
dataf %>% select("ppvt", "ccc_coh") %>%
  correlation::cor_test(x="ppvt", y="ccc_coh", method='spearman', ci=0.95)

#Linear regression - CCC-GCI and PPVT
dataf %>% select("ppvt", "ccc_gcc") %>%
  lm(ccc_gcc ~ ppvt, data= .) %>%
  summary()
dataf %>% select("ppvt", "ccc_gcc") %>%
  lm(ccc_gcc ~ ppvt, data= .) %>%
  confint( ,level=0.95)
dataf %>% select("ppvt", "ccc_gcc") %>%
  lm(ccc_gcc ~ ppvt, data= .) %>%
  regression.diagnostics()
dataf %>% select("ppvt", "ccc_gcc") %>%
  ggplot(aes(x=ppvt, y=ccc_gcc)) +
  geom_point() +
  stat_smooth(method="lm", col="red") +
  labs(x="PPVT-4 standard score", y="CCC-GCI") +
    theme(axis.text=element_text(size=12)) +
    theme(axis.title=element_text(size=15)) +
  stat_regline_equation(label.x=120, label.y=26)

#Linear regression - CCC-IASI and PPVT
dataf %>% select("ppvt", "ccc_iasi") %>%
  lm(ccc_iasi ~ ppvt, data= .) %>%
  summary
dataf %>% select("ppvt", "ccc_iasi") %>%
  lm(ccc_iasi ~ ppvt, data= .) %>%
  confint( ,level=0.95)
dataf %>% select("ppvt", "ccc_iasi") %>%
  lm(ccc_iasi ~ ppvt, data= .) %>%
  regression.diagnostics()
dataf %>% select("ppvt", "ccc_iasi") %>%
  ggplot(aes(x=ppvt, y=ccc_iasi)) +
  geom_point() +
  stat_smooth(method="lm", col="red") +
  labs(x="PPVT-4 standard score", y="CCC-IASI") +
    theme(axis.text=element_text(size=12)) +
    theme(axis.title=element_text(size=15)) +
  stat_regline_equation(label.x=120, label.y=6)

#Multiple regression - Syntax and coherence subdomains
dataf %>% select("ppvt", "ccc_synt", "ccc_coh") %>%
  lm(ppvt ~ ccc_synt + ccc_coh, data= .) %>%
  summary()
dataf %>% select("ppvt", "ccc_synt", "ccc_coh") %>%
  lm(ppvt ~ ccc_synt + ccc_coh, data= .) %>%
  regression.diagnostics()

#Research question 2

#Correlation between SRS and VABS-MBI
dataf %>% select("vineland_v", "srs_t") %>%
  correlation::cor_test(x="vineland_v", y="srs_t", method='pearson', ci=.
95)
```

```
#Correlation between FQoL and CASP
dataf %>% select("fqol", "casp") %>%
  correlation::cor_test(x="fqol", y="casp", method='spearman', ci=.95)

#Correlation between PPVT and measures of adaptive/maladaptive behaviour

dataf %>% select("ppvt", "vineland_v") %>%
  correlation::cor_test(x="ppvt", y="vineland_v", method='spearman', ci=.
95)
dataf %>% select("ppvt", "casp") %>%
  correlation::cor_test(x="ppvt", y="casp", method='spearman', ci=.95)
dataf %>% select("ppvt", "fqol") %>%
  correlation::cor_test(x="ppvt", y="fqol", method='spearman', ci=.95)
dataf %>% select("ppvt", "srs_t") %>%
  correlation::cor_test(x="ppvt", y="srs_t", method='spearman', ci=.95)
dataf %>% select("ppvt", starts_with("srs")) %>%
  cor_mat(method="spearman", conf.level=0.95) %>%
  cor_get_pval
#Correlation between PPVT-4 and social cognition identified
dataf %>% select("ppvt", "srs_cog") %>%
  correlation::cor_test(x="ppvt", y="srs_cog", method='spearman', ci=.95)


#Linear regression

#PPVT and VABS-MBI
dataf %>% select("ppvt", "vineland_v") %>%
  lm(vineland_v ~ ppvt, data= .) %>%
  summary()

#PPVT and FQoL
dataf %>% select("ppvt", "fqol") %>%
  lm(fqol ~ ppvt, data= .) %>%
  summary()

#PPVT and CASP
dataf %>% select("ppvt", "casp") %>%
  lm(casp ~ ppvt, data= .) %>%
  summary()

#PPVT and SRS
dataf %>% select("ppvt", "srs_t") %>%
  lm(srs_t ~ ppvt, data= .) %>%
  summary()
dataf %>% select("ppvt", "srs_t") %>%
  lm(srs_t ~ ppvt, data= .) %>%
  regression.diagnostics()
dataf %>% select("ppvt", "srs_t") %>%
  lm(srs_t ~ ppvt, data= .) %>%
  confint( ,level=0.95)

dataf %>% select("ppvt", "srs_t") %>%
  ggplot(aes(x=ppvt, y=srs_t)) +
  geom_point(colour="black", shape=15) +
  geom_smooth(method="lm", colour="red") +
  labs(x="PPVT-4 standard score",
       y="SRS t-score") +
  theme(axis.text=element_text(size=12)) +
```

```
  theme(axis.title=element_text(size=15)) +
  stat_regline_equation(label.x=120, label.y=81)

#PPVT and SRS social cognition
dataf %>% select("ppvt", "srs_cog") %>%
  lm(srs_cog ~ ppvt, data= .) %>%
  summary()
dataf %>% select("ppvt", "srs_cog") %>%
  lm(srs_cog ~ ppvt, data= .) %>%
  confint( , level=0.95)
dataf %>% select("ppvt", "srs_cog") %>%
  lm(srs_cog ~ ppvt, data= .) %>%
  regression.diagnostics()

dataf %>% select("ppvt", "srs_cog") %>%
  ggplot(aes(x=ppvt, y=srs_cog)) +
  geom_point(colour="black", shape=15) +
  geom_smooth(method="lm", colour="red") +
  labs(x="PPVT-4 standard score",
       y="SRS social cognition score") +
  theme(axis.text=element_text(size=12)) +
  theme(axis.title=element_text(size=15)) +
  stat_regline_equation(label.x=120, label.y=81)

#Research question 3

#Correlation between CCC-GCI and VABS
dataf %>% select("ccc_gcc", "vineland_v") %>%
  correlation::cor_test(x="ccc_gcc", y="vineland_v", method='pearson',
ci=.95)

cor.test(struc_lang, data$vineland_v, method='pearson', ci=.95)
cor.test(prag_lang, data$vineland_v, method='pearson', ci=.95)

#Correlation between CCC-GCI and FQoL
dataf %>% select("ccc_gcc", "fqol") %>%
  correlation::cor_test(x="ccc_gcc", y="fqol", method='spearman', ci=.95)

cor.test(struc_lang, data$fqol, method='spearman', ci=.95)
cor.test(prag_lang, data$fqol, method='spearman', ci=.95)

#Correlation between CCC-GCI and CASP
dataf %>% select("ccc_gcc", "casp") %>%
  correlation::cor_test(x="ccc_gcc", y="casp", method='pearson', ci=.95)

dataf %>% select("ccc_iasi", "casp") %>%
  correlation::cor_test(x="ccc_iasi", y="casp", method='pearson', ci=.95)

cor.test(struc_lang, data$casp, method='pearson', ci=.95)
cor.test(prag_lang, data$casp, method='pearson', ci=.95)

dataf %>% select("casp", starts_with("ccc")) %>%
  cor_mat(method='pearson', conf.level=0.95) %>%
  cor_get_pval

dataf %>% select("casp", "ccc_spo") %>%
  correlation::cor_test(x="casp", y="ccc_spo", method='pearson', ci=.95)
dataf %>% select("casp", "ccc_sem") %>%
```

```
  correlation::cor_test(x="casp", y="ccc_sem", method='pearson', ci=.95)

#Correlation between CCC-GCI and SRS
dataf %>% select("ccc_gcc", "srs_t") %>%
  correlation::cor_test(x="ccc_gcc", y="srs_t", method='pearson', ci=.95)

cor.test(struc_lang, data$srs_t, method='pearson', ci=.95)
cor.test(prag_lang, data$srs_t, method='pearson', ci=.95)

#Regression between SRS and CCC structural and pragmatic language
lm(prag_lang ~ data$srs_t) %>% summary()
lm(prag_lang ~ data$srs_t) %>% regression.diagnostics()

lm(struc_lang ~ data$srs_t) %>% summary()
lm(struc_lang ~ data$srs_t) %>% regression.diagnostics()

#Correlation matrix by subdomain of the SRS and CCC

#CCC speech and SRS social awareness/SRS social communication
dataf %>% select("ccc_spo", "srs_awr") %>%
  correlation::cor_test(x="ccc_spo", y="srs_awr", method='pearson', ci =.
95)

dataf %>% select("ccc_spo", "srs_com") %>%
  correlation::cor_test(x="ccc_spo", "srs_com", method='pearson', ci=.95)

#Linear regression

#CCC-GCI and VABS-MBI
dataf %>% select("ccc_gcc", "vineland_v") %>%
  lm(vineland_v ~ ccc_gcc, data= .) %>%
  summary()

#CCC-GCI and FQoL
dataf %>% select("ccc_gcc", "fqol") %>%
  lm(fqol ~ ccc_gcc, data= .) %>%
  summary()

#CCC-GCI and CASP
dataf %>% select("ccc_gcc", "casp") %>%
  lm(casp ~ ccc_gcc, data= .) %>%
  summary()
dataf %>% select ("ccc_gcc", "casp") %>%
  lm(ccc_gcc ~ casp, data = .) %>%
  regression.diagnostics
dataf %>% select("ccc_gcc", "casp") %>%
  ggplot(aes(x=ccc_gcc, y=casp)) +
  geom_point(colour="black", shape=15) +
  geom_smooth(method="lm", colour="red") +
  labs(x="CCC-GCI score",
       y="CASP score") +
  theme(axis.text=element_text(size=12)) +
  theme(axis.title=element_text(size=15)) +
  stat_regline_equation(label.x=15, label.y=81)

#CASP and structural and pragmatic language
lm(data$casp ~ prag_lang) %>% summary()
lm(data$casp ~ prag_lang) %>% regression.diagnostics()
```

```
lm(data$casp ~ struc_lang) %>%
  summary()
lm(data$casp ~ struc_lang) %>%
  confint( , level=0.95)
lm(data$casp ~ struc_lang) %>%
  regression.diagnostics()

ggplot(dataf, aes(x=struc_lang, y=casp)) +
  geom_point() +
  stat_smooth(method='lm', col="red") +
  labs(x="CCC-2 structural language score",
       y="CASP score") +
  theme(axis.text=element_text(size=12)) +
  theme(axis.title=element_text(size=15)) +
  stat_regline_equation(label.x=10, label.y=91)

#CCC-GCI and SRS
dataf %>% select("ccc_gcc", "srs_t") %>%
  lm(srs_t ~ ccc_gcc, data= .) %>%
  summary()
dataf %>% select("ccc_gcc", "srs_t") %>%
  lm(srs_t ~ ccc_gcc, data= .) %>%
  confint( , level=0.95)

#CCC-Spo and SRS-Awr
dataf %>% select("ccc_spo", "srs_awr") %>%
  lm(srs_awr ~ ccc_spo, data= .) %>%
  summary()
dataf %>% select("ccc_spo", "srs_awr") %>%
  lm(srs_awr ~ ccc_spo, data= .) %>%
  confint( , level=0.95)
dataf %>% select("ccc_spo", "srs_awr") %>%
  lm(ccc_spo ~ srs_awr, data= .) %>%
  regression.diagnostics()
dataf %>% select("srs_awr", "ccc_spo") %>%
  ggplot(aes(x=ccc_spo, y=srs_awr)) +
  geom_point(colour="black", shape=15) +
  geom_smooth(method="lm", colour="red") +
  labs(x="CCC-2: Spoken language subdomain",
       y="SRS social awareness score") +
  theme(axis.text=element_text(size=9)) +
  theme(axis.title=element_text(size=15)) +
  stat_regline_equation(label.x=9, label.y=81)

#CCC-Spo and SRS-Com
dataf %>% select("ccc_spo", "srs_com") %>%
  lm(srs_com ~ ccc_spo, data= .) %>%
  summary()
dataf %>% select("ccc_spo", "srs_com") %>%
  lm(srs_com ~ ccc_spo, data= .) %>%
  confint( , level=0.95)
dataf %>% select("ccc_spo", "srs_com") %>%
  lm(ccc_spo ~ srs_com, data= .) %>%
  regression.diagnostics()
dataf %>% select("srs_com", "ccc_spo") %>%
  ggplot(aes(x=ccc_spo, y=srs_com)) +
  geom_point(colour="black", shape=15) +
```

```
  geom_smooth(method="lm", colour="red") +
  labs(x="CCC-2: Spoken language subdomain",
       y="SRS social communication score") +
  theme(axis.text=element_text(size=9)) +
  theme(axis.title=element_text(size=15)) +
  stat_regline_equation(label.x=9, label.y=81)

#SRS and structural and pragmatic language
lm(prag_lang ~ data$srs_t) %>% summary()
lm(prag_lang ~ data$srs_t) %>% regression.diagnostics()

lm(struc_lang ~ data$srs_t) %>% summary()
lm(struc_lang ~ data$srs_t) %>% regression.diagnostics()
```