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Burden, health care needs, and Quality of Life of family caregivers of adults with Autism Spectrum Disorder

Dissertation

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„Ich sag‘ auch immer, außer meinem Mann und meinem jüngeren Sohn
kann kein Mensch nachvollziehen, was wir hier machen, leisten
und – Entschuldigung – auch aushalten müssen.“

Mutter eines erwachsenen autistischen Sohnes, Fokusgruppe *BarrierefreiASS*, 2021

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Zusammenfassung

Familienangehörige unterstützen ihre autistischen Verwandten oft über die gesamte Lebensspanne hinweg. Obwohl dadurch massive Belastungen entstehen können, wurden diese bei Angehörigen von autistischen Erwachsenen kaum untersucht.

Daher verfolgte diese Dissertation vier Ziele: (1) Erstellung eines Überblicks zu vorhandener Evidenz zu Belastungen und Interventionen; (2) Vergleich der physischen und Lebensqualität (HRQoL) von Angehörigen autistischer Erwachsener mit der Allgemeinbevölkerung in Deutschland; (3) Identifizierung von Prädiktoren der HRQoL; (4) Untersuchung der aktuellen Versorgungssituation von autistischen Erwachsenen und Zusammenhängen mit Pflegebelastungen und Pflegebedarfen.

(1) In einem Scoping Review gaben *N*=33 Studien Indikationen auf multidimensionale Pflegebelastungen, wobei der Fokus auf der emotionalen Belastung lag und keine Intervention identifiziert werden konnten. (2) Zudem zeigten Querschnittsdaten von *N*=149 Angehörigen eine signifikant geringere HRQoL als die Allgemeinbevölkerung. (3) Die subjektive Pflegebelastung erklärte die meiste Varianz der psychischen HRQoL. Für die physische HRQoL konnten keine signifikanten Prädiktoren identifiziert werden. (4) In Fokusgruppen mit Angehörigen (*n*=12) und Gesundheitsdienstleistern (*n*=15) sowie Interviews mit autistischen Erwachsenen (*n*=15) zeigten sich massive Barrieren in der aktuellen Gesundheitsversorgung, eine hohe Pflegebelastung von Angehörigen und daraus resultierende Bedarfe an Unterstützungsangeboten.

Die Ergebnisse implizieren multidimensionalen Pflegebelastungen von Angehörigen autistischer Erwachsener. Es bedarf weitere Forschung und Strategien zur Reduktion der Pflegebelastung, wie der Entwicklung evidenzbasierter Angebote für Angehörige und einer Verbesserung der Gesundheitsversorgung von autistischen Erwachsenen.

Abstract

Family members often provide support for their autistic relative throughout the whole lifespan which can lead to massive burden themselves. To date, research primarily focused on the burden on parental caregivers of autistic children/adolescents, but the burden on family caregivers of autistic adults has been barely assessed.

For this purpose, this dissertation had four aims: (1) scrutinize existing evidence on burden and interventions for family caregivers of autistic adults; (2) compare the health-related Quality of Life (HRQoL) of family caregivers with the general population; (3) identify caregiver-related and care recipient-related predictors of HRQoL; (4) assess the current state of mental healthcare for autistic adults in Germany and possible associations with the burden and needs of family caregivers.

(1) Within a scoping review, *N*=33 studies indicated multidimensional burden, with most studies focusing on the emotional burden. No caregiver intervention was identified. (2) *N*=149 caregivers of autistic adults completed the Short-Form Health Survey (SF-8) in a cross-sectional online survey. Caregivers reported significantly lower physical and mental HRQoL compared to the general population. (3) Multiple linear regression analyses revealed subjective caregiver burden explaining most of variance in mental HRQoL, and no significant predictor of physical HRQoL could be identified. (4) Focus groups with caregivers (*n*=12) and healthcare providers (*n*=15), and interviews with autistic adults (*n*=15) emphasized massive barriers in the current mental healthcare, great caregiver burden, and high needs for caregiver services.

Findings indicated the existence of multidimensional caregiver burden in family caregivers of autistic adults. Therefore, strategies to reduce caregiver burden must be developed. For example, evidence-based interventions tailored for family caregivers need to be established, and the mental healthcare for autistic adults must be improved.

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List of abbreviations

ADHD	Attention Deficit-Hyperactivity Disorder
APA	American Psychological Association
ASD	Autism Spectrum Disorder
BASS	BarrierfreeASD: Mixed-methods investigation of barriers and needs in mental healthcare of adults with autism and recommendations for future care
CarerQol-7D	Care-related Quality of Life instrument
CBI	Caregiver Burden Inventory
CI	Confidence Interval
DGKJP	Deutsche Gesellschaft für Kinder- und Jugendpsychiatrie, Psychosomatik und Psychotherapie
DGPPN	Deutsche Gesellschaft für Psychiatrie und Psychotherapie, Psychosomatik und Nervenheilkunde
DSM-5	Diagnostic and Statistical Manual of Mental Disorders (5th version)
G-BA	German Federal Joint Committee
GDW	General Dominance Weight
HRQoL	Health-related Quality of Life
ID	Intellectual Disability
MCS	Mental Component Scale (SF-8)
OCD	Obsessive-Compulsive Disorder
QoL	Quality of Life
PCS	Physical Component Scale (SF-8)
SD	Standard Deviation
SE	Standard Error
SF-8/12/36	Short Form Health-Survey
UKE	University Medical Center Hamburg-Eppendorf

1. Theoretical background

Based on the most recent and increasing prevalence rate of 0.38% (Bachmann et al., 2018), it can be estimated that more than 315,000 people in Germany are diagnosed with an Autism Spectrum Disorder (ASD). ASD is a heterogeneous and complex condition, and can lead to substantial challenges in all domains of life and throughout the whole lifespan (Croen et al., 2015; Lord et al., 2022). For this reason, autistic individuals[1] require support, assistance or/and care[2] not only in the childhood, but also in the adulthood, and family caregivers (i.e., (grand)parents, partners/spouses, siblings, adult children) remain the main source of support (Cadman et al., 2012). Although caring for a loved one can be rewarding (Phelps et al., 2009), caregiving can lead to massive challenges for the family caregivers, as so-called “caregiver burden” (Marsack-Topolewski & Maragakis, 2020). However, caregiver burden on family caregivers of autistic adults has not been in the focus of research yet. For this reason, this dissertation pursues to contribute to this important but hitherto under-researched topic with the overarching goal to derive strategies for relieving caregiver burden in family caregivers of autistic adults.

1.1 Family caregiving

Family caregiving is defined as the unpaid assistance or aid provided of one or more family member(s) that is more than the assistance required as a part of regular everyday life (Walker et al., 1995). In the scientific literature, the terms “informal

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[1] The identity-first-language is used as most preferred, but the person-first-language is equally supported. Further information: https://www.liebertpub.com/doi/10.1089/aut.2020.0014

[2] For purpose of simplification, mentioned as “care” below

caregiving” and “family caregiving” are often used interchangeably since there exist no standardized definition (Bastawrous, 2013). For the purpose of this dissertation, the term “family caregiving” will be used, as it underlines the link to the family.

Due to various factors, such as an ageing population, high prevalence of chronic diseases, and lack of skilled professionals, family caregiving plays an increasingly important role in society (Broese van Groenou & De Boer, 2016). In most cases, family caregiving implies that someone is suddenly taking over caregiving tasks without prior experience or education (Fernandes & Angelo, 2016). Despite many common experiences, caregiving is highly variable. For example, the diversity of families, the caregiving tasks that need to be provided, the timing of entry into the caregiving role, the reason or condition why care must be provided, the relation to the care recipient, all shape the nature of the caregiving role (Adults, 2016). Similar heterogeneous is the impact that caregiving has on the individual caregiver. Besides enriching caregiver outcomes, such as an increased family connectedness and positive developments, caregiving can have numerous negative caregiver outcomes (Phelps et al., 2009), as described below.

1.1.1 Caregiver burden

Caregiver burden can be defined as “the level of multifaceted strain perceived by the caregiver from caring for a family member and/or loved one over time” (Liu et al., 2020). Novak and Guest (1989), who primarily focused their research on caregivers of individuals with Alzheimer’s disease, evolved five dimensions of caregiver burden. First, *time dependence burden* includes restrictions on the caregiver’s time available for personal interests and activities (M. J. Altiere & S. von Kluge, 2009; Smith et al., 2010) or privacy (Marsack & Perry, 2018). Second, *developmental burden,* refers to personal or social underdevelopment compared to peers who do not have a relative who requires care (Novak & Guest, 1989), which can lead to feelings of isolation and a perception of being disconnected or detached from the social environment (Hare et al., 2004; Monique Hines et al., 2014; Marsack & Hopp, 2018). Third, *physical burden* describes caregivers' feelings of chronic fatigue and damage to physical health (Novak & Guest, 1989). Fourth, *social burden* comprise feelings of role conflicts, as well as limited time and energy that they can invest in relationships or in occupational participation (Novak & Guest, 1989). Fifth, *emotional burden,* refers to negative feelings towards the relative with ASD (Novak & Guest, 1989), such as guilt and blame (Marsack-Topolewski & Graves, 2019; Marsack & Hopp, 2018).

Several measurements were developed to assess caregiving burden. Novak and Guest (1989) developed the Caregiver Burden Inventory (CBI), based on the five abovementioned burden dimensions. The CBI assesses the degree of agreement on 24 care-related items on a 5-point Likert scale (1=completely disagree to 5 =completely agree), ranging from 24 (no care pressure) to 120 (very severe care pressure).

In extension to this initial definition of *emotional burden* by Novak and Guest (1989), prior research reported caregiving to also worsen family caregivers' mental health, including higher levels of psychological distress (Abbeduto et al., 2004; Blacher & McIntyre, 2006), and higher prevalence's of mental disorders (Magallon-Neri et al., 2018; Schnabel et al., 2020), which can be indicated by decreased Health-related Quality of Life (HRQoL).

1.1.2 Health-related Quality of Life

HRQoL refers to “how well a person functions in their life and his or her perceived wellbeing in physical, mental, and social domains of health” (Hays & Reeve, 2010). HRQoL captures information on both physical and mental health status and its impact on Quality of Life (QoL). It is a useful indicator of overall health and is suitable for prevention and early detection of physical or mental diseases (Palermo et al., 2008; Revicki et al., 2014; Romero et al., 2013; Ware et al., 1996; Yin et al., 2016). As a so-called “patient-reported outcome”, HRQoL comprises the perspective of patients, making it suitable for validating healthcare services and interventions, and ensuring the provision of high-quality services (Churruca et al., 2021).

One of the most widely used generic HRQoL measurement is the Short-Form Health Survey (SF-8/12/36; Ware et al., 2001). Especially the SF-8 is a parsimonious and user-friendly instrument. Each of the eight single-items assess one dimension of the longer SF-36 and allow the calculation of a physical component scale (PCS) and a mental component scale (MCS; Ware & Gandek, 1998). Items are scored on a 5-point Likert scale and the PCS and MCS are derived using an algorithmic norm-based scoring procedure, with higher scores indicating better HRQoL (0-100) (Ware et al., 2001). The SF(-8/-12/-36) is used both nationally and internationally, which allows comparisons between different populations. Previous research has shown strong reliability (parallel test reliability $r = .82$) and validity of the MCS (Ware et al., 2001).

1.2 Overview of Autism Spectrum Disorder

Before heading to the distinct challenges that family caregivers of autistic adults face, an overview of ASD needs to be provided for better understanding.

1.2.1 Diagnosis and epidemiology

ASD is a neurodevelopmental disorder that summarizes previous autism subdiagnoses (i.e., childhood autism, Asperger syndrome, atypical autism, pervasive developmental disorder not otherwise specified) (American Psychiatric Association, 2013). The two core symptoms of ASD include (1) deficits in social interaction and communication, such as responding inappropriately, misinterpreting (non)verbal interactions, or difficulties in building relationships. Additionally, (2) high sensitivity to environmental changes, inflexible adherence to routines, or constant preoccupation with special interests must be present (always taking the age-typical developmental stage and sociocultural context into account) (American Psychiatric Association, 2013). The differential diagnosis is particularly relevant as these symptoms are not specific to autism and can also occur with, for example, personality disorders, anxiety disorders, obsessive-compulsive disorder (OCD), or attention deficit-hyperactivity disorder (ADHD; Lehnhardt et al., 2013). In ASD, the onset occurs during early childhood, even though some symptoms may not fully develop until a later stage, and the symptoms result in significant impairments in important areas of life, like social and educational life. According to the fifth version of the *Diagnostic and Statistical Manual of Mental Disorders* (DSM-5), ASD diagnosis requires the classification in one of three level of severity with respect to the level of required support (American Psychiatric Association, 2013).

As mentioned above, the administrative prevalence of ASD in Germany is estimated at 0.38% (Bachmann et al., 2018), with an global prevalence about approximately 1% (Rubenstein et al., 2023; Zeidan et al., 2022). Risk is 3–4-times higher in males than females (Lord et al., 2018). Despite difficulties in identifying autism especially in adults (e.g., due to recognition bias related to sex, ethnicity, socioeconomic factors), there is a considerable and growing population of autistic adults (Lord et al., 2018; Rubenstein et al., 2023).

1.2.2 Etiology and heritability

The causes of ASD are still unknown, but several environmental and genetic risk factors have been identified that, alone or in combination, are involved in the development of ASD (Sauer et al., 2021). Environmental factors mediating ASD risk include, for example, neonatal complications (Schmidt et al., 2012), advanced parental age (Grether et al., 2009), and prenatal maternal diet (Zhong et al., 2020). However, genetic factors were found to play a consistently larger role than environmental factors (Taylor et al., 2020), and a large number of heterogeneous, individual genetic variants have been associated with ASD risk (Fang et al., 2023; Lord et al., 2018). In turn, high heritability rates were found in family and twin studies with meta-analytic estimates of 64-91% (Tick et al., 2016), which also might be an important factor when examining family caregivers.

1.2.3 Comorbidities and treatment

In the clear majority, the presence of ASD is accompanied by at least one of various mental and somatic comorbidities (Croen et al., 2015; Khachadourian et al., 2023; Vohra et al., 2017) that can have a major impact on the autistic individuals and their families (Lord et al., 2018). ADHD, depressive disorders, anxiety disorders, OCD, sleep disorders, and psychosis belong to the most common mental comorbidities (Khachadourian et al., 2023; Mutluer et al., 2022). Frequent somatic comorbidities are, for example, epilepsy, allergic disorders, genetic disorders, metabolic disorders, or gastrointestinal disorders (Al-Beltagi, 2021). Furthermore, it is estimated that one-third to one-half do have an accompanying intellectual disability (ID; Maenner, 2020; Postorino et al., 2016).

The high heterogeneity in comorbid conditions in ASD, as well as the heterogeneity of ASD itself makes the elucidation on the one hand, and the treatment on the other hand a challenging task (Mutluer et al., 2022). Since the biological marker that causes ASD is still unidentified, there is no cure for ASD (Jensen et al., 2022). Given the typical impairments in autistic individuals, treatment usually focuses on the alleviation of behavioral deficits, on the development of social, education/academic/employment, and communication skills, as well as the increase of Quality of Life (QoL) (Hyman et al., 2020). Pharmacological treatment is limited to the treatment of co-occurring conditions, not to treat ASD itself (Hyman et al., 2020; Lord et al., 2018). The most common treatment for ASD is behavioral and educational interventions, such as applied behavior analysis and speech and/or occupational therapy. While these treatments can be beneficial, they require a large commitment of time and energy on the part of the autistic individual and the family (Jensen et al., 2022). For example, social skills group trainings showed improvements in related outcomes like social responsiveness, empathy, and social anxiety in both autistic children and autistic adults (McVey et al., 2016). However, most of the interventions available focus on autistic children, leading to specific challenges for autistic adults, their families, and the healthcare system.

1.2.4 ASD in adulthood

As ASD is a pervasive condition that cannot be cured, it affects individuals into adulthood, and, accordingly, the majority of autistic individuals are adults. Autistic adults typically present considerable psychosocial and healthcare needs, as indicated by increased comorbidity rated (e.g., more than 80% show a psychiatric comorbidity), reduced (HR)QoL, premature mortality or risk of suicide (Croen et al., 2015; Hirvikoski et al., 2016; Mason et al., 2019; Vohra et al., 2017). Moreover, employment can be negatively affected in autistic adults (Barnard et al., 2001; Holwerda et al., 2012; Howlin, 2013), which is demonstrated by large unemployment gaps in autistic adults without ID (factor 5 compared to the general population) despite above-average education levels (Espeloer et al., 2023). Furthermore, friendships were found to be less close, less empathic, less important, and less supportive to autistic adults compared to non-autistic adults (Baron-Cohen & Wheelwright, 2003). In addition, autistic adults are also more likely to stay unmarried or without long-term intimate relationships (Howlin & Moss, 2012). The majority of autistic adults are still living with their parents, while few adults are living independently or with their own families (Barnard et al., 2001; Roux et al., 2015). However, irrespective of the living situation, only 3% of autistic adults even at “the higher end of the autism spectrum” are living fully independently (Barnard et al., 2001, p. 7).

There is a wide range of challenges in the healthcare system that autistic adults need to deal with, starting with the transition from childhood into adulthood (Anderson et al., 2018). For those who received diagnosis in the childhood, healthcare transition is a critical time when autistic children outgrow pediatric services and enter a fragmented, insufficient, and underfunded healthcare system (Enner et al., 2020). However, although some individuals obtain ASD diagnosis in childhood, for every three known cases there are two individuals without a diagnosis and, in turn, do not receive much needed support (Baron-Cohen et al., 2009). Especially identifying so-called "high-functioning autism" as well as autistic females can be challenging, as both is typically combined with compensation or camouflaging for autistic difficulties (Lord et al., 2022; Pilling et al., 2012). In undiagnosed autistic adults, the capacity to cope with social relationships and adaptation to a non-autistic society can become increasingly challenging, and clinical presentations can occur when social demands overwhelm the capacity to compensate (World Health Organization, 2022). Accordingly, adult-diagnosed individuals are more likely to endorse mental comorbidities than their childhood-diagnosed counterparts (Jadav & Bal, 2022). In other cases, the impetus for the ASD diagnosis in adulthood comes from family members, spouses, or social or legal services (Lord et al., 2022). Nevertheless, long waiting lists (up to two years) often delay the ASD diagnosis in Germany (Vllasaliu et al., 2019).

In comparison to the longstanding clinical expertise in the diagnosis and treatment of autistic children, relatively little is known about ASD in adult medicine (Lehnhardt et al., 2013). Despite the aforementioned challenges that autistic adults face, and the resulting extensive need for psychosocial support, ASD in adulthood is still under-represented, as services and support available for autistic adults are far fewer, and very few interventions for autistic adults have been evaluated (DGKJP &

DGPPN, 2021; Lord et al., 2022). Not surprisingly, autistic adults reported significantly lower quality healthcare than non-autistic individuals (Weir et al., 2022). However, evidence on the state of healthcare for autistic adults especially in Germany is lacking, and a comprehensive picture of barriers and needs has to be drawn.

1.3 Burden on family caregivers of autistic individuals

“*A diagnosis of autism affects not just the individual, but the entire family.*”

(Lord et al., 2022, p. 45)

1.3.1 Burden on family caregivers of autistic children

The nature of ASD often goes along with the inability to take care of oneself and the dependence on family caregivers (Bal et al., 2015). In autistic children, mostly parental caregivers are the main source of support. Managing the children’s heterogeneous behavioral and communicational deficits, often combined with comorbid somatic or mental conditions, can have multiple negative implications on parental caregivers (Rasoulpoor et al., 2023). The impact of caregiving demands and challenges in raising an autistic child often take priority over the caregiver’s own needs and desires, particularly in leisure and occupational participation (Davy et al., 2022). Several scientific reviews provide evidence that parental caregivers are affected by changes in social relationships, lack of social support, changes in daily routines and financial issues due to the disorder of their child (e.g., Bonis & Sawin, 2016; Cachia et al., 2015; Davy et al., 2022; Karst & Van Hecke, 2012; Schnabel et al., 2020; Tint & Weiss, 2016). Moreover, stigmatization was found to predict parents' mental health (Farrugia, 2009).

With respect to the cut-offs of the aforementioned CBI (see Section 1.1.1; Novak & Guest, 1989), maternal caregivers of autistic children were shown to experience a severe caregiver burden (Rasoulpoor et al., 2023). Caregiver burden in ASD was significantly higher than in schizophrenia (Yildiz et al., 2021). Furthermore, parental caregivers of autistic children presented significant lower levels of HRQoL and more mental disorders than parents of a non-autistic child (Benjak et al., 2009). To release caregiver burden and increase (HR)QoL, psychosocial interventions and support programs for parental caregivers were developed and evaluated, showing promising results (Catalano et al., 2018; Ji et al., 2014).

In sum, a considerable amount of research on parental caregivers of autistic children was conducted. However, the impairments of ASD remain throughout life, and so does the caregiving demands, which results in family caregivers providing care for their autistic relative across the lifespan (Marsack & Hopp, 2018).

1.3.2 Burden on family caregivers of autistic adults

The roles of the family in provision of services changes over time, with family involvement being predominant in the early years, often decreasing during school years, and increasing again in adulthood (Shattuck et al., 2018). Evidence is lacking, but, due the above mentioned lack of services for autistic adults, many autistic adults rely on the support by their families. The demands of family caregivers of autistic children and autistic adults can be partly overlapping, but in adults, specific demand with regard to the nature of ASD in adulthood (see Section 1.2.4) can pose unique challenges for family caregivers of autistic adults.

In addition, as autistic relatives enter adulthood, the duration and ongoing demands of caregiving can accumulate and lead to increased stress for family caregivers (Matthew J. Altiere & Silvia von Kluge, 2009; Marsack & Perry, 2018), and have several negative effects on their lives, as shown by increased subjective caregiver burden (Marsack-Topolewski & Wang, 2022; Marsack & Hopp, 2019). Many family caregivers of autistic adults struggle with the care-associated responsibilities, while balancing everyday life, social interactions, and occupational responsibilities (Hare et al., 2004; M. Hines et al., 2014; Marsack & Perry, 2018; Oti-Boadi et al., 2020).

Family caregivers of autistic adults also seem to be experiencing increased physical and mental health problems (Barker et al., 2011; Rattaz et al., 2017; Tomeny et al., 2017). For example, this population reported significantly higher emotional distress compared to caregivers of patients with schizophrenia, and are at higher risk to develop health problems themselves (Grootscholten et al., 2018). Moreover, comprised levels of HRQoL was indicated (Lee et al., 2019). In attempt to identify the underlying mechanisms of this (presumed) finding, a number of potential predictors have been investigated. For instance, a systematic review by Sonido et al. (2019) scrutinized potential predictors of mental wellbeing of family caregivers of autistic adults.

In their research on parental caregiver burden in autistic adults, Marsack and Hopp (2018) were the first that added the assessment of a *financial burden*, in extension to the predefined caregiver burden dimensions by Novak and Guest (1989) (see Figure 1). The financial burden includes effects of caring on financial resources, such as direct costs related to specialized therapies, and indirect costs related to constrains on working life and productivity losses (Cidav et al., 2012; DePape & Lindsay, 2015; Marsack & Perry, 2018).

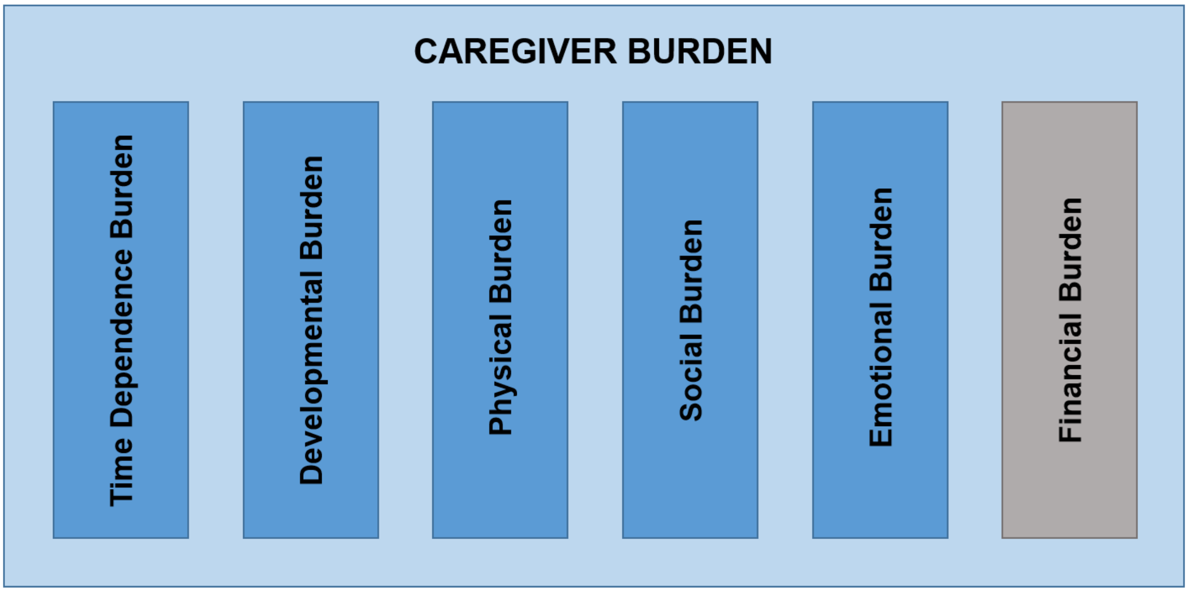


Figure 1. Dimensions of caregiver burden according to Novak and Guest (1989) (blue fields), adapted by Marsack and Hopp (2018) (grey field), own representation

These studies gain important data, but overall evidence on the burden of family caregivers of autistic adults is very limited (with no data from Germany). In addition, no comprehensive literature overview of burden on this target group was published. Moreover, most studies focused on parental caregivers, without taking into account that other relatives can also provide care. In addition, even studies reported lower HRQoL in family caregiver of autistic adults, scores have not been compared to scores of non-clinical populations. Furthermore, the picture on several potential predictors of HRQoL is less clear-cut. Discrepant or lacking evidence has been identified for various caregiver-related and care recipient-related predictors (Sonido et al., 2019). Lastly, assumed barriers and needs in the current healthcare system have never been analyzed in Germany, and it is unclear whether healthcare disparities are associated with the burden on family caregivers of autistic adults.

1.4 Research gaps

“Zum psychosozialen Unterstützungsbedarf von Familien und Angehörigen – insbesondere dem von Geschwisterkindern und Lebenspartnern – soll weitere Forschung erfolgen.“ (DGKJP & DGPPN, 2021, p. 424)

In the previous sections, existing research on family caregivers in autistic individuals was described. Until now, research mainly focused on family caregivers of autistic children or adolescents, disregarding the fact that ASD-related characteristics persist into adulthood and are compensated by parents, siblings or partners/spouses over the whole lifespan. Thus, family caregivers of autistic adults remain largely uninvestigated. Consequently, the aim of this dissertation is to investigate family caregivers of autistic adults for the first time in Germany.

First, as existing reviews either focused on ASD in childhood (Tint & Weiss, 2016) or only assessed mental wellbeing of family caregivers (Sonido et al., 2019), a scoping review will be conducted in order to provide a comprehensive overview of evidence on the above mentioned dimensions of burden on family caregivers of autistic adults (i.e., time dependence, developmental, physical, social, emotional, and financial burden). Moreover, previously published interventions or supporting services designed to reduce such burden, and existing knowledge gaps should be identified.

Second, little evidence on the physical and mental HRQoL in family caregivers of autistic adults is available, and existing evidence shows many limitations. Furthermore, no data on family caregivers in Germany exist. Therefore, cross-sectional data from a German wide online survey on the family caregivers HRQoL will be analyzed and compared to data from a nonclinical, normative sample to allow proper interpretation of the data. Moreover, a comprehensive set of caregiver-related and care recipient-related predictors that might explain variance in both physical and mental HRQoL in family caregivers of autistic adults will be examined.

Lastly, the current state of mental healthcare of autistic adults in Germany should be assessed. It is expected that massive barriers impede the adequate healthcare of autistic adults in Germany, and family caregivers compensate for this. Thus, the perspective of family caregivers, healthcare providers, and autistic adults on the healthcare system will be determined in order to get a comprehensive picture. Furthermore, it should be investigated whether barriers in healthcare are associated with the burden and needs on family caregivers of autistic adults.

2. Research objectives and research questions

The main aim of this dissertation was to investigate family caregivers of autistic adults in Germany for the first time. In this section, research objectives, research questions of this dissertation, and related publications will be presented (see Table 1).

Table 1. Overview of research objectives, research questions and related publications

<table>
<tr><td>Research objective</td><td>To provide an overview of existing scientific literature on the burden on and healthcare needs/interventions for family caregivers of autistic adults.</td></tr>
<tr><td>Research question</td><td>How is the current state of evidence on different dimensions of burden and healthcare needs/interventions in/for family caregivers of autistic adults?</td></tr>
<tr><td colspan="2">Dückert, S., Gewohn, P., König, H., Schöttle, D., Konnopka, A., Rahlff, P., Vogeley, K., Schulz, H., David, N., & Peth, J. (2023a). Multidimensional Burden on Family Caregivers of Adults with Autism Spectrum Disorder: A Scoping Review. <i>Review Journal of Autism and Developmental Disorders.</i> https://doi.org/10.1007/s40489-023-00414-1</td></tr>
<tr><td>Research objective 1</td><td>To compare the physical and mental HRQoL of family caregivers of autistic adults with the general population in Germany.</td></tr>
<tr><td>Research question 1</td><td>Do differences exist between the physical and mental HRQoL in family caregivers of autistic adults and the general population in Germany?</td></tr>
<tr><td>Research objective 2</td><td>To identify caregiver-related and care recipient-related predictors explaining variance in family caregivers’ HRQoL.</td></tr>
<tr><td>Research objective 2</td><td>What caregiver-related and care recipient-related predictors can explain variance in family caregivers’ HRQoL?</td></tr>
<tr><td colspan="2">Dückert, S., Bart, S., Gewohn, P., König, H., Schöttle, D., Konnopka, A., Rahlff, P., Erik, F., Vogeley, K., Schulz, H., David, N., & Peth, J. (2023b). Health-related Quality of Life in family caregivers of autistic adults. <i>Frontiers in Psychiatry</i>, 14. https://doi.org/10.3389/fpsyt.2023.1290407</td></tr>
<tr><td>Research objective</td><td>To assess the current state of mental healthcare for autistic adults in Germany and possible associations with the burden and needs of family caregivers.</td></tr>
<tr><td>Research question</td><td>Which barriers and needs exist in the mental healthcare for autistic adults in Germany and are they associated with caregivers burden and needs?</td></tr>
<tr><td colspan="2">Dückert, S., Gewohn, P., König, H., Schöttle, D., Konnopka, A., Rahlff, P., Frank-Erik, Vogeley, K., Schulz, H., David, N., & Peth, J. (2023c). Barriers and needs in mental healthcare of adults with autism spectrum disorder in Germany: a qualitative study in autistic adults, relatives, and healthcare providers. <i>BMC Psychiatry</i>, 23(528). https://doi.org/10.1186/s12888-023-05026-x</td></tr>
</table>

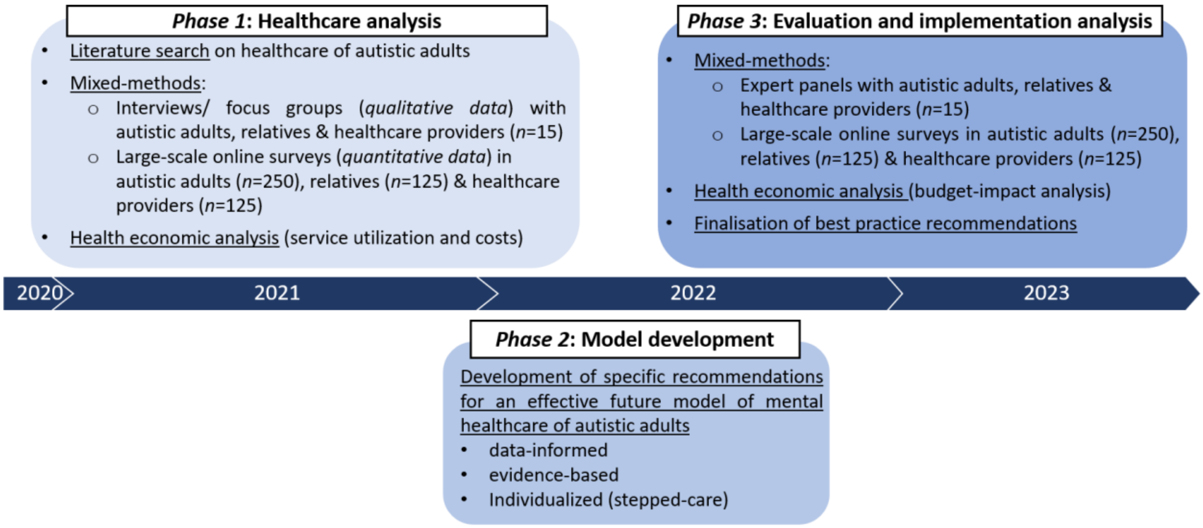
3. Context of this dissertation

The underlying dissertation was conducted within the participatory research project "*BarrierfreeASD – Mixed-methods investigation of barriers and needs in mental healthcare of adults with autism and recommendations for future care*" (German title: "*BarrierefreiASS – Versorgungs- und Bedarfsanalyse zur barrierefreien Teilhabe von Diagnostik und Therapie von Erwachsenen mit Autismus-Spektrum-Störung*", acronym "BASS"). The project is still being carried out until the end of 2023 (start: October 2020) under the lead of Dr. Judith Peth and Prof. Dr. Holger Schulz at the Department of Medical Psychology at the University Medical Center Hamburg-Eppendorf (UKE). Funding was received by the Innovation Fund of the *German Federal Joint Committee* (Gemeinsamer Bundesausschuss, G-BA, grant number: 01VSF19011). The project is carried out in accordance to the Helsinki Declaration of the World Medical Association. Furthermore, ethical approval was granted by the *Local Psychological Ethics Commission of the Center for Psychosocial Medicine* at the UKE (Lokale Psychologische Ethikkommission am Zentrum für Psychosoziale Medizin, LPEK-0227).

The project aims and project phases are threefold (see Figure 2): (1) assessment of barriers and needs as well as of utilization and related costs in the current mental healthcare for adults with autism at three levels (individual, structural, professional) and from three relevant perspectives (adults with autism, relatives, and healthcare providers). To meet this aim, a mixed-methods approach was used by conducting focus groups/interviews (qualitative data) and large-scale online surveys (quantitative data). (2) Based on the results of project phase 1 and existing evidence/guidelines, recommendations for a future healthcare model for adults with autism were derived. (3) Finally, the developed healthcare mode was evaluated with regard to future implementation and costs from the three aforementioned stakeholder perspectives using a mixed-methods design again (David et al., 2022).

In all project phases, participants were recruited throughout Germany using purposive, quota, and snowball sampling via the project's collaborating network of cooperating partners, publicly available contacts from autism-related groups (e.g., self-help groups for adults with autism and family caregivers), healthcare associations (e.g., medical chambers, therapist associations), outpatient clinics as well as social media.

This dissertation is mainly placed in the context of project phase 1.



***Phase 1*: Healthcare analysis**

- Literature search on healthcare of autistic adults
- Mixed-methods:
  - Interviews/ focus groups (*qualitative data*) with autistic adults, relatives & healthcare providers (*n*=15)
  - Large-scale online surveys (*quantitative data*) in autistic adults (*n*=250), relatives (*n*=125) & healthcare providers (*n*=125)
- Health economic analysis (service utilization and costs)

***Phase 3*: Evaluation and implementation analysis**

- Mixed-methods:
  - Expert panels with autistic adults, relatives & healthcare providers (*n*=15)
  - Large-scale online surveys in autistic adults (*n*=250), relatives (*n*=125) & healthcare providers (*n*=125)
- Health economic analysis (budget-impact analysis)
- Finalisation of best practice recommendations

***Phase 2*: Model development**

Development of specific recommendations for an effective future model of mental healthcare of autistic adults

- data-informed
- evidence-based
- Individualized (stepped-care)

Figure 2. Timeline and project phases of the BASS project (David et al., 2022, updated by the author of this dissertation)

4. Overview of publications and own contributions

Publication 1:

Dückert, S., Gewohn, P., König, H., Schöttle, D., Konnopka, A., Rahlff, P., Vogeley, K., Schulz, H., David, N., & Peth, J. (2023a). Multidimensional Burden on Family Caregivers of Adults with Autism Spectrum Disorder: A Scoping Review. *Review Journal of Autism and Developmental Disorders.* https://doi.org/10.1007/s40489-023-00414-1

Objectives: (a) Systematically map research regarding multiple dimensions of caregiver burden on family caregivers of autistic adults (i.e., time dependence, developmental, physical, social, emotional, and financial burden); and (b) identify interventions designed to prevent or reduce such burden.

Methods: A scoping review was conducted in order to gain a comprehensive overview on the current state of evidence.

Results: A total of *N* = 33 eligible studies highlighted the impact of caregiving demands for adults with ASD, mainly focusing on emotional burden of caregiving (*n* = 27), reporting decreased mental Quality of Life and mental health. Findings gave indications on all other dimensions of caregiver burden but evidence is lacking. No study was identified that provided evidence for specific interventions to reduce or to prevent caregiver burden.

Conclusion: Findings highlighted the urgent need for more research on this topic and the development of strategies to support family caregivers of adults with ASD.

Own contribution: SD made substantial contributions to the scoping review. SD developed the search strategy, screened the records in collaboration with a team member, as well as analyzed and interpreted the data. Moreover, SD wrote the first draft of the manuscripts and revised it several times according to the co-authors' feedback.

Publication 2:

Dückert, S., Bart., S., Gewohn, P., König, H., Schöttle, D., Konnopka, A., Rahlff, P., Frank-Erik, Vogeley, K., Schulz, H., David, N., & Peth, J. (2023b). Health-related Quality of Life in family caregivers of adults with Autism Spectrum Disorder. *Frontiers in Psychiatry.* https://doi.org/10.3389/fpsyt.2023.1290407

Objective: (i) Investigation on the current state of physical and mental HRQoL of family caregivers of autistic adults compared to the general population, and (ii) examination of caregiver-related (e.g., age, subjective caregiver burden) and care recipient-related variables (e.g., symptom severity, utilization of formal services) explaining variance in the caregivers' HRQoL.

Methods: *N* = 149 family caregivers completed a nationwide online survey, including the Short-Form Health Survey (SF-8; Ware et al., 2001) in order to assess the HRQoL, and the Care-related Quality of Life instrument (CarerQol-7D; Brouwer et al., 2006) to assess the subjective caregiver burden. T-tests were used to compare the HRQoL of family caregivers with the general population. Bivariate correlational and multiple linear regression analyses were conducted in order to identify predictors explaining variance in family caregivers' HRQoL.

Results: Family caregivers of autistic adults reported significantly lower physical and mental HRQoL compared to the general population. Multiple linear regression with the mental HRQoL as the outcome showed a significant model with increased subjective burden explaining most of the variance in mental HRQoL. Multiple linear regression analysis with the outcome physical HRQoL did not reveal a statistically significant model.

Discussion: Findings highlight the for further research especially on predictors of the physical HRQoL, and the need to consider HRQoL (and caregiver burden) of family caregivers of autistic adults in healthcare settings to monitor a potential comprised health status in early stages, with the long-term goal to improve family caregivers' HRQoL.

Own contributions: SD made substantial contributions to the study. She recruited participants as well as collected, analyzed, and interpreted the data. Moreover, SD wrote the first draft of the manuscripts and revised it several times according to the co-authors' feedback.

Publication 3:

Dückert, S., Gewohn, P., König, H., Schöttle, D., Konnopka, A., Rahlff, P., Frank-Erik, Vogeley, K., Schulz, H., David, N., & Peth, J. (2023c). Barriers and needs in mental healthcare of adults with autism spectrum disorder in Germany: a qualitative study in autistic adults, relatives, and healthcare providers. *BMC Psychiatry*, 23(528). https://doi.org/10.1186/s12888-023-05026-x

Objective: Investigation of barriers and needs in mental healthcare of autistic adults in Germany at the three relevant levels (individual, professional, structural) and from three relevant perspectives (autistic adults, relatives and healthcare providers), in order to get a comprehensive picture of the state of healthcare and possible associations with caregivers' burden and needs.

Methods: Semi-structured, open-ended interviews were conducted with autistic adults (*n* = 15) and focus groups with family caregivers (*n* = 12), and healthcare providers (*n* = 15). Data analysis was performed using the codebook approach of thematic analysis.

Results: Poor mental healthcare of autistic adults in Germany was characterized by six central and overarching themes: (i) lack of knowledge about autism, (ii) a need for increased participation/involvement, (iii) consideration of autism-specific needs in treatment, (iv) lack of services, (v) limited access to services, and (vi) improvement of stakeholder collaboration. Themes were similarly reported across participants, emphasizing dissatisfaction in all stakeholders. Furthermore, the burden on family caregivers and the high need for support services was emphasized.

Conclusions: We identified major barriers to mental healthcare for autistic adults in Germany that affect autistic adults, but are also of concern to relatives and healthcare providers. Our results point to specific and generic areas for improvement, independent of stakeholder perspectives, which could guide future development of needs- and evidence-based services, recommendations and guidelines of mental healthcare for autistic individuals across the lifespan. Caregiver burden has to be reduced due to the development and expansion of caregiver services.

Own contributions: SD made substantial contributions to the study. She recruited participants as well as collected, analyzed, and interpreted the data. Moreover, SD wrote the first draft of the manuscripts and revised it several times according to the co-authors' feedback.

5. Synopsis

5.1 Summary of results

From the publications included in this dissertation, the most important results relating to the questions posed in this dissertation will be presented. More detailed results can be found in the underlying publications (see Section 7). In addition, unpublished preliminary results from the BASS online survey 2 will be introduced in this dissertation.

In publication 1 (Dückert et al., 2023a), existing research regarding multiple dimensions of caregiver burden on family caregivers of autistic adults was systematically mapped, as well as interventions designed to reduce or prevent such burden identified. Therefore, a scoping review was conducted, based on the model of Novak and Guest (1989), and complemented by Marsack and Hopp (2018) on caregiver burden (i.e., time dependence, developmental, physical, social, emotional, and financial burden). A total of *N* = 33 eligible studies were included in the scoping review, consisting of 17 quantitative, eight qualitative, three mixed-methods studies, and five literature reviews. Most studies were conducted in the United States (55.9%), followed by the United Kingdom (23.5%). The family caregivers' age ranged from 18 to 87 years, caregivers were predominantly female (58.4-100%), and the majority were parents (in 58.1% of studies). Six studies investigated caregivers of adults with ASD and comorbid ID.

All dimensions of burden were detected within the included studies. However, the amount of available research data regarding the specific burdens differed (see Figure 3). The most investigated burden was the emotional burden (*n* = 27), reporting decreased mental HR(QoL) and mental health. Findings gave indications on all other dimensions of caregiver burden but evidence is lacking. No study was identified that provided evidence for specific interventions to reduce or to prevent caregiver burden.

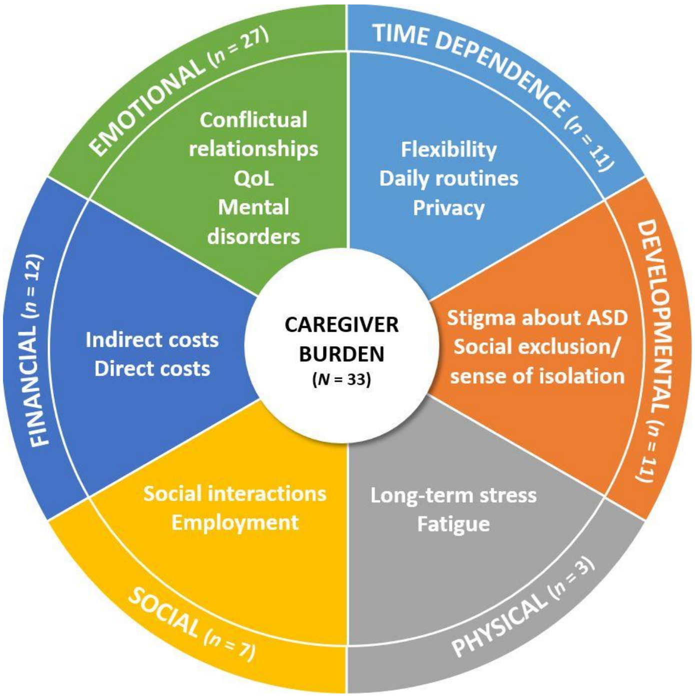


Figure 3. Summary of existing evidence of burden in family caregivers of autistic adults (Dückert et al., 2023a)

In publication 2 (Dückert et al., 2023b), cross-sectional data from a Germany wide online survey was analyzed in order to compare the physical and mental HRQoL in family caregivers of autistic adult with data from the general population. Furthermore, caregiver-related and care recipient-related variables that were assumed to explaining variance in the caregivers' HRQoL were examined. Data from *N* = 149 family caregivers of autistic adults could be analyzed. The caregivers' mean age were 51.95 years (*SD* = 9.48), and the majority were female (87.2%), in a parental relation to the care recipient (70.7%), and had an A-Level-education (74.8%). The mean score of the CarerQol-7D was 56.08 (*SD* = 14.94). The care recipients' mean age were 26.69 years (*SD* = 9.75), the majority was male (73.6%), and hat the diagnosis of an Asperger syndrome (69.1%).

Data on the family caregivers revealed a mean PCS (SF-8; Ware et al., 2001) of *M* = 46.71 (*SD* = 8.72) and a mean MCS of *M* = 40.15 (*SD* = 11.28; see Figure 4). The German normative sample (*N* = 2552) (Beierlein et al., 2012) rated a mean PCS of *M* = 50.3 (*SD* = 8.39) and a mean MCS of *M* = 53.23 (*SD* = 7.82). T-tests for independent samples indicated that the difference between the two samples was statistically significant for the PCS (*t*(129.58) = 4.42, *p* < .001, Cohen's d = 0.42), and for the MCS (*t*(124.44) = 12.56; *p* < 0.001, Cohen's d = 1.35), indicating a significantly lower physical and mental HRQoL in caregivers of autistic adults with a small effect size (PCS), respectively large effect size (MCS) (Cohen, 1988).

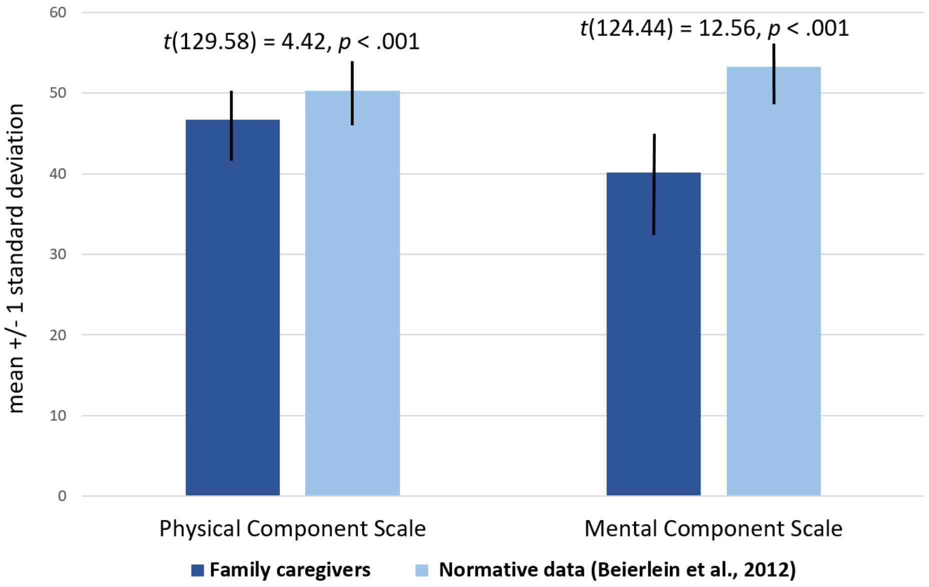


Figure 4. Health-related Quality of Life in family caregivers of autistic adults and in the general population (Dückert et al., 2023b)

With respect to the identification of caregiver-related and care recipient-related predictors explaining variance in the HRQoL of family caregivers, two multiple linear regression analyses were conducted. Multiple linear regression analysis with the outcome PCS scores did not reveal a statistically significant model (*F*(11,95) = 1.09, *p* = .38). Multiple linear regression with the mental HRQoL as the outcome showed a significant model (*F*(11, 95) = 5.53, $p < .001$), with an adjusted R^2 of .32 (see Table 2). Significant results were obtained for the following caregiver variables: school education (*ß* = -.22, GDW = .068, $p < .05$) and CarerQol-7D sum score (*ß* = .32, GDW = .141, $p < .001$). Higher school education and lower CarerQol-7D sum scores predicted lower MCS scores. For care recipient variables, the age at diagnosis (*ß* = -.20, GDW = .023, $p < .05$) and the number of formal services (*ß* = -.19, GDW = .054, $p < .05$) were significant predictors: high age at diagnosis and a higher number of formal services used by the autistic adult predicted lower mental HRQoL. Remaining predictors were not significant.

Table 2. Multiple linear regression analysis of mental HRQoL (*N* = 107), adapted from Dückert et al. (2023b)

Predictor	*b*	*SE*	*ß*	95% *CI*	GDW	VIF
Caregiver-related						
Age (years)	0.20	0.12	0.16	[-0.05, 0.44]	.018	1.53
Relationship (parental)	-3.00	2.98	-0.12	[-8.92, 2.92]	.006	2.07
School education (A-Level)	**-5.78***	**2.48**	**-0.22**	**[-10.71, -0.85]**	**.068**	1.33
Treatment-related expenses (Euro)	<0 .001	<0 .001	0.01	[<0.001, <0.001]	.003	1.14
CarerQol-7D (sum score)	**0.24*****	**0.01**	**0.32**	**[0.11, 0.37]**	**.141**	1.23
Informal care (hours)	<0.001	<0.001	-0.03	[-0.01, 0.01]	<.001	1.21
Care recipient-related						
Severity (social communication)	-0.49	1.24	-0.04	[-2.95, 1.97]	.013	1.74
Severity (behavior)	-2.05	1.21	-0.18	[-4.45, 0.35]	.039	1.65
Age at diagnosis (years)	**-0.20***	**0.10**	**-0.20**	**[-0.40, -0.01]**	**.023**	1.56
ID (yes)	4.94	3.13	0.14	[-1.28, 11.15]	.025	1.18
Formal services (sum score)	**-1.77***	**0.84**	**-0.19**	**[-3.42, -0.11]**	**.054**	1.22

Note. Outcome: Mental HRQoL (MCS of SF-8). Model fit: *F*(11, 95) = 5.53, $p < .001$, adj. R^2 = .32. ID = intellectual disability. *SE* = Standard Error. *CI* = Confidence Interval. GDW = General Dominance Weights. VIF = Variance Inflation Factor.
*$p < .05$. ** $p < .01$. *** $p < .001$.

In publication 3 (Dückert et al., 2023c), the current state of mental healthcare of autistic adults in Germany was investigated by assessing barriers and needs in focus groups with family caregivers (*n* = 12) and healthcare providers (*n* = 15), as well as interviews with autistic adults (*n* = 15). In the group of family caregivers, mean age were 54.25 years (*SD* = 9.5), and most were female (83.3%). Participants were parents (75%), or partners (33.3%) of an autistic adult. Sociodemographic information on the other stakeholders can be seen at Dückert et al. (2023c).

Current mental healthcare of autistic adults in Germany was characterized by six central and overarching themes: (i) lack of knowledge about autism, (ii) a need for increased participation/involvement, (iii) consideration of autism-specific needs in treatment, (iv) lack of services, (v) limited access to services, and (vi) improvement of stakeholder collaboration. In addition to these overarching themes, family caregivers described to suffer from burdens related to caregiving demands. In turn, a high need for services for family caregivers was emphasized, such as the expansion of caregiver self-help groups.

With regard to the aforementioned findings, cross-sectional data from the second Germany wide BASS online survey (see Figure 2, phase 3) was analyzed that has not been published yet. Since all three publications have indicated a need for psychosocial interventions, this should now be quantitatively verified. To this end, items were developed to obtain an initial assessment of a future psychosocial intervention (e.g., “If there was a service for family caregivers of autistic adults to help reduce stress or burden, would you be interested in attending?”). Data from *N* = 63 family caregivers of autistic adults was analyzed. The family caregivers’ mean age were 54.76 years (*SD* = 9.6), the majority was female (87.3%), parental caregiver (82.3%), and had an A-level-education (67.2%). *N* = 15 (23.8%) already participated in the first BASS online survey. The mean sum score of the CarerQol-7D was 64.05

(*SD* = 18.74). The mean age of the care recipients was 26.71 years (*SD* = 9.17), most were male (77.8%), and had a formal Asperger Syndrome diagnosis (76.2%).

84.1% (*n* = 53) of the participants stated to be interested in attending a psychosocial intervention for family caregivers of autistic adults (see Table 3). The majority of the participants reported to favor physical meetings (47.2%), while there was no preference between one-to-one or group settings (54.7%). Moreover, the majority of family caregivers was willing to attend to eight appointments (73.6%) and to pay for the intervention (43.3%).

Table 3. Descriptive statistics of family caregivers' interest in a psychosocial intervention

	N	*n* (%)
Interested in intervention (yes)	63	53 (84.1)
Group vs. one-to-one	53	
One-to-one		13 (24.5)
Group		11 (20.8)
Doesn’t matter/ don’t know		29 (54.7)
Physical vs. digital meetings	53	
Physical		25 (47.2)
Digital		8 (15.1)
Doesn’t matter/ don’t know		20 (31.7)
Willingness to attend eight appointments	53	
Yes		39 (73.6)
No		3 (5.7)
Don’t know		11 (20.8)
Willingness to pay for intervention	53	
Yes		23 (43.4)
No		11 (20.8)
Don’t know		19 (35.8)

Note. *N* = Sample size.

5.2 Comprehensive discussion

In this section, main findings of all included publications as well as unpublished data incorporated in this dissertation will be summarized in a few sentences and comprehensively discussed, including associated strength and limitations as well as implications for research and practice.

This dissertation provided evidence that family caregivers of autistic adults can be exposed to several caregiving demands which can lead to the expression of multidimensional caregiver burden, especially with focus on the emotional burden. Impairments of both physical and mental HRQoL compared to the German general population have been revealed, and, in the set of investigated predictors, the perceived caregiver burden explained the most variance of mental HRQoL. With regard to the caregiver burden dimensions by Novak and Guest (1989) and Marsack and Hopp (2018), all other dimensions of caregiver burden have been touched upon in previous research, but evidence is lacking and/or conflicting. Even though the healthcare needs, and the interest in a psychosocial intervention appear to be high, no interventions could be found that were specifically developed for or utilized by caregivers in previous studies. One of a number of possible contributors to the burden on family caregivers could be current barriers to healthcare for autistic adults, which in turn create responsibilities and caregiving demands for families.

The first aspect that attracts attention is that a lot of research has been conducted on parental caregivers of autistic children but there is a huge lack of research on family caregivers of autistic adults (Khanna, 2010). Therefore, it was not surprising that the vast majority (*n* = 262) of screened studies in the scoping review were excluded because they have focused only on family caregivers of autistic children (Dückert et al., 2023a). Although these studies make important scientific contributions, they overlook the fact that ASD persists the whole lifespan, and so does the need for support.

As the findings from the scoping review revealed, family caregivers of autistic adults can suffer from a time dependence burden because as they are occupied with caregiving responsibilities around the clock and constantly on demand (Marsack & Perry, 2018). This was supported by data from the conducted online survey, as family caregivers reported to provide informal care in average of approximately 9 hours per day (Dückert et al., 2023b). Quantitative data is lacking, but it can be assumed that this time aspect can also lead to a social burden, i.e., missing capacity for social activities (Monique Hines et al., 2014; Oti-Boadi et al., 2020), and a financial burden, as family caregivers sometimes reported loss of employment as a consequence of caregiving responsibilities (Hare et al., 2004).

Besides the loss of employment, financial burden is assumed to derive from the high costs for treatment-related expenses (Oti-Boadi et al., 2020), which averaged approximately 5000€ per year in the current quantitative study (Dückert et al., 2023b). However, extensive data on the financial burden on family caregivers of autistic adults that include the costs for productivity losses, treatment-related expenses as well as costs for informal caregiving are lacking but are currently being prepared by the BASS research group (Dückert et al., in preparation).

With respect to the physical burden due to caregiving, previous evidence was very limited. However, data from the current quantitative study found the physical HRQoL of family caregivers of autistic adults to be significantly lower compared to the physical HRQoL of the general population in Germany (Dückert et al., 2023b). The attempt to identify predictors that explain variance in the physical HRQoL failed, as the multiple regression model was not statistically significant. One possible explanation could be the selection of predictors which was – as previous evidence on physical burden was lacking – mainly based on research regarding the mental HRQoL. In turn, other variables that were not included in this analysis explain variance of the physical HRQoL. Social support, caregivers' illness, lack of sleep, and lack of respite care are some of the factors that have been found to affect the physical health of parental caregivers of autistic children (Murphy et al., 2007; Myers et al., 2009; Shu, 2009).

With regard to the developmental burden, especially stigma on ASD on the one hand, and the social isolation on the other hand were assumed to cause this burden (Griffith et al., 2012). Particularly in family caregivers of autistic adults the developmental burden could be critical, as family caregivers' lives take a different path than expected (e.g., children do not move out from parents' home when they grew up).

To summarize the aspects so far, there are indications on all described caregiver burden dimensions, and this dissertation contributed to the scientific state of knowledge, but there is still a lot research to be done (see Section 5.4). However, the last caregiver burden dimension, the emotional burden, turned out to be the most researched dimension, although with some knowledge gaps (Dückert et al., 2023a). Trying to address these gaps, quantitative data of the current dissertation found significant lower mental HRQoL in family caregivers of autistic adults compared to the German general population (Dückert et al., 2023b). Moreover, based on the review of Sonido et al. (2019), predictors explaining variance in the mental HRQoL were identified. Interestingly, the aforementioned relatively high treatment-related expenses and the sum of provided informal care did not significantly explain variance, but the subjective informal care situation did. Hence, in the current sample, not the objective care provided, but the perception of caregiving explained variance in the mental HRQoL of family caregivers. This finding is contrary to a study on parents of autistic children (Khanna, 2010). Using the stress-appraisal model by Chappell and Reid (2002) as a guiding framework, the researcher captured the role of primary stressors (care recipient functional status and extent of behavioral problems), mediators (social support, coping mechanisms, and family functioning), and caregiver burden in predicting parents' HRQoL.

It can be emphasized that family caregivers of autistic adults can be affected by multidimensional caregiver burden, which, in turn, creates some healthcare needs. The vast majority of participants in the second BASS online survey expressed interest in a psychosocial intervention especially developed for family caregivers. However, according to the findings of the scoping review, no such intervention exists yet. Interventions could be effective particularly with regard to the emotional burden, as results from intervention studies on family caregivers of autistic children or relatives with Alzheimer's disease showed decreased depressive and anxiety symptoms and improvements in QoL (Beinart et al., 2012; Bekhet, 2017; Smith et al., 2010). Knowing that working on the perception of caregiving might increase the mental HRQoL of family caregivers of autistic adults provides a good basis for the development of interventions.

Obviously, the question arises as to how specific the caregiver burden and healthcare needs in this target group are? This question cannot be conclusively clarified within the scope of this dissertation, but the assumption can be made that there is some overlap with the burden experienced by family caregivers of autistic children or relatives with other chronic illnesses. This is not surprising when taking into account that symptoms in ASD are not ASD specific, but can also occur in other mental disorders. However, caregiver burden in ASD was found to be significant higher than caregiver burden in ADHD (Cadman et al., 2012) or in schizophrenia (Yildiz et al., 2021). Furthermore, the demands and challenges that ASD in adulthood entails differ to some extent from those in childhood (Lord et al., 2022), which was also recognized in the results of the scoping review. First, there are typical features that adulthood brings with it, and that can pose challenges for family caregivers, e.g., leaving school, pursuing a career, building a romantic relationship. This leads to the second aspect that in adulthood partners/spouses or own children can take on the role of a family caregiver, and be exposed to specific challenges and burden (Lewis, 2017). Third, while there are pediatric services and community programs for autistic children of school age, these are discontinued when they leave school. The healthcare system does not adequately meet the needs of autistic adults (Dückert et al., 2023c). Several barriers and unmet healthcare needs were found to entail the responsibility to the family caregivers, and might consequently lead to caregiver burden. To conclude, even if there is some overlap, specifics in the caregiver burden of family caregiver of autistic adults exist that can lead to unique healthcare needs, and have to be considered while developing tailored interventions.

5.3 Strength and limitations

The following section describes the strengths and limitations of the overarching dissertation, as all further study-specific information is described in the respective sections of the single publications.

Looking at the strength of this dissertation, this was the very first time that family caregivers of autistic adults in Germany were investigated - a highly burdened but under-researched population, as this dissertation demonstrated. The findings of this dissertation provide initial results and a groundwork for further studies.

In addition, the participatory approach of this dissertation should be underlined, which ensured the participation of family caregivers (and partly autistics adults and healthcare providers) in the surveys on the one hand, and the involvement of family caregivers, autistic adults, and healthcare providers as cooperation partners of the overarching BASS project on the other hand. The collaboration was valuable in many ways, such as the ongoing dialogue on latest study topics, the critical review of earlier versions of the surveys, knowledge transfer, and the considerable support provided on the recruitment of study participants.

Another strength of this dissertation is the recruitment of family caregivers using several strategies, such as the aforementioned support of the study's network of collaboration partners, publicly available contacts from autism-related associations (including self-help and caregiver groups), healthcare associations (medical chambers, therapist associations, etc.), local and countrywide outpatient clinics, local social media and personal contacts. In this way, an attempt was made to address the heterogeneity of the target group and also to reach caregivers who are not connected to the healthcare system.

Otherwise, looking at the limitations of this dissertation, a bias due to convenience sampling cannot be ruled out. Even all attempts were made to address the full heterogeneity of family caregivers of autistic adults, in every study predominantly highly educated mothers with a professional degree participated. Given this fact, the findings of the studies are likely to have limited generalizability. While it can be assumed that mothers might be the most prevalent group of caregivers, findings indicated that fathers, siblings and partners/spouses can also be burdened, but there is still a lack of in-depth research (Dückert et al., 2023a).

Moreover, the missing link between the results of the qualitative healthcare analysis and the possibly resulting caregiver burden has to be stated. Qualitative findings indicate that massive barriers in the current healthcare of autistic adults exist which are often compensated by family caregivers. However, there is no quantitative verification of this relationship yet.

Another limitation is the cross-sectional study design of this dissertation, as findings are limited in their ability to make causal inferences. As an example, although some predictors significantly explained variance in the mental HRQoL of family caregivers of autistic adults, it cannot be drawn the conclusion that these predictors have causal connections with the mental HRQoL. Therefore, not all of the research questions could be completely answered within this dissertation, which will be elaborated in more detail within the next section.

5.4 Implications for research and practice

Based on the results of this dissertation, the following implications can be deduced.

Looking at the implications for research first, there are several points of reference, and, as the scoping review revealed, there is missing evidence on numerous aspects. First, the dimensions of caregiver burden have to be comprehensively scrutinized in future research, e.g., influencing factors of the specific burdens (e.g., predictors of the physical HRQoL), relationships between the different dimensions of burden and potential dependencies, and associations between the presence of burden and well-being of family caregivers need to be identified. For example, previous evidence on parents of autistic children highlighted the role of social support and coping strategies in alleviating caregiver burden and improving their HRQOL (Khanna, 2010; Sarriá & Pozo, 2015), these findings could be examined in the current target group.

Furthermore, the time course of caregiver burden and the possible connection with critical live events should be investigated. For example, emotional burden, e.g., in the form of psychological exhaustion, future uncertainties, and worries about the future of the adult autistic child were found to be burdening especially in ageing parental caregivers (Oti-Boadi et al., 2020).

In addition, between-group-differences in the expression of caregiver burden and the referring healthcare needs in different groups of family caregivers (i.e., mothers, fathers, siblings, partners/spouses, children) should be further investigated. It can be assumed that caregiver group-overarching burden (e.g., time dependence burden and social burden) and group-specific burden (e.g., emotional burden due to lack of intimacy in partners/spouses, shifted relationships when children of autistic adults have to take the role of a caregiver) exist. Unfortunately, sample sizes of non-parental caregivers have been too small to conduct subgroup-analyses in the current quantitative study, but need to be analyzed in future studies to derive adequate interventions.

Moreover, as mentioned before, the association between barriers in healthcare for autistic adults and the expression of caregiver burden needs to be quantitatively analyzed. Irrespective of this, it is beyond question that the healthcare of autistic adults must be improved, which is the long-term goal of the BASS project.

Furthermore, possible effects of the high heritability of ASD on the caregiver burden needs to be determined, because there is a high probability of being more than one autistic member in a family (Khanna, 2010; Tick et al., 2016).

In order to achieve these research aims, longitudinal, large-scale quantitative studies should be designed, to allow the identification of causal relationships, with the overarching goal to shed light on the specific burden and needs of family caregivers of autistic adults on the one hand, and to use this knowledge for the development of tailored interventions on the other hand. Within the scope of this dissertation, a first draft of a psychosocial intervention was participatory developed that needs to be piloted and evaluated in the future.

In sum, more research on the psychosocial needs of family caregivers and autistic adults in general has to be conducted (DGKJP & DGPPN, 2021), with the ultimate aim to decrease caregiver burden, and improve mental healthcare in order to enhance independence and HRQoL, for both the autistic adults and their family caregivers.

Looking at the practice implications, one of the main goals is to create more awareness about ASD in the society. This dissertation indicated that stigma and misunderstanding of ASD can have negative impacts not only on autistic adults, but also on their family caregivers, as it can lead to social exclusion/isolation and developmental burden. Moreover, also healthcare providers' knowledge about ASD, and especially the awareness of the potential burden on family caregivers needs to be extended. Healthcare providers working with autistic adults and their families should form a routine to assess and monitor caregivers' (mental) health status, e.g., as part of the intake assessment or in primary care (Willet et al., 2018).

Moreover, with regard to the results of the qualitative healthcare analysis, the expansion of low-threshold services for family caregivers should be promoted (Dückert et al., 2023c). As requested in the focus groups, further self-help groups specifically for family caregivers of autistic adults should be established. Planning and implementing such groups could face lower barriers, as no healthcare professional needs to be involved. Even though self-help groups for this target group have not been empirically evaluated yet, cooperating parents' associations have reported positive effects on family caregivers, as self-help groups can increase knowledge about support services for autistic adults one the one hand, and can create a feeling of not being alone with the problems on the other hand.

Lastly, psychological, social, and economic support should be provided for families. As mentioned before, it is necessary that services must be created or expanded. Moreover, strategies must be devised to help family members find their way through the jungle of the health and social care system. Some valuable support programs already exist, but very few caregivers are aware of them such. As an example, the personal budget provides direct payments for individuals with disabilities. The personal budget can be used to ensure more professional person-centered care, and, in turn, relieve the burden on family caregivers (Turnpenny et al., 2021). Knowledge about such services must be more targeted.

5.5 Conclusion

Although concerns of family caregivers are increasingly addressed in research, there is still a lack of comprehensive, in-depth evidence regarding the underlying mechanisms of the different dimensions of caregiver burden in family caregivers of autistic adults. Using qualitative, quantitative, and review data, this dissertation makes a valuable contribution to the state of knowledge on family caregivers of autistic adults, and provides the very first data on this target group from Germany.

This dissertation provided evidence that family caregivers of autistic adults can be exposed to several caregiving demands which can lead to the expression of all previously defined dimensions of caregiver burden, i.e., time dependence, developmental, social, physical, emotional, and financial burden (Marsack & Hopp, 2018; Novak & Guest, 1989). However, all dimensions of caregiver burden have been touched upon in previous research, but evidence is lacking and/or conflicting yet, with most previous research focusing on the emotional burden.

On closer examination, impairments of both physical and mental HRQoL in family caregivers of autistic adults compared to the German general population have been revealed. In the current set of investigated predictors, the perceived caregiver burden explained the most variance of mental HRQoL, whereas predictors that capture more objective caregiver burden variables did not. Even though the healthcare needs appear to be high, no interventions could be found that were specifically developed for or utilized by caregivers in previous studies. Thus, high-quality, evidence-based interventions designed to mitigate or prevent adverse health effects tailored to the needs of this target group should be developed (Committee on Family Caregiving for Older Adults, 2016).

Not only a lack of healthcare services for family caregivers, but also for autistic adult have been identified. Massive barriers currently impede the adequate healthcare for autistic adults, and initial result indicate an association with the burden on family caregivers. Consequently, improvements in the healthcare of both autistic adults and family caregivers could foster the reduction of the caregiver burden.

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7. Publications

7.1 Publication 1

Dückert, S., Gewohn, P., König, H., Schöttle, D., Konnopka, A., Rahlff, P., Vogeley, K., Schulz, H., David, N., & Peth, J. (2023a). Multidimensional Burden on Family Caregivers of Adults with Autism Spectrum Disorder: A Scoping Review. *Review Journal of Autism and Developmental Disorders.* https://doi.org/10.1007/s40489-023-00414-1

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REVIEW PAPER

Multidimensional Burden on Family Caregivers of Adults with Autism Spectrum Disorder: a Scoping Review

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Abstract

Family caregivers often play a critical role in supporting their relative(s) with autism spectrum disorder (ASD) across the lifespan. This can lead to great burdens on family caregivers themselves. However, to date, the potential burden on family caregivers has not been in the focus of research, particularly, with respect to caregiver burden as relatives with ASD advance to adulthood. Thus, this scoping review aimed to (a) systematically map research regarding multiple dimensions of caregiver burden on family caregivers of adults with ASD (i.e., time dependence, developmental, physical, social, emotional, and financial burden) and (b) identify interventions designed to reduce such burden. A total of $N = 33$ eligible studies highlighted the impact of caregiving demands for adults with ASD, mainly focusing on emotional burden of caregiving ($n = 27$), reporting decreased mental quality of life and mental health. Findings gave indications on all other dimensions of caregiver burden, but evidence is lacking. No study was identified that provided evidence for specific interventions to reduce or to prevent caregiver burden. Findings highlighted the urgent need for more research on this topic and the development of strategies to support family caregivers of adults with ASD.

Keywords Autism spectrum disorder (ASD) · Caregiver burden · Adults · Scoping review · Mental health · Family caregivers

Abbreviations

ASD	Autism spectrum disorder
CBI	Caregiver Burden Inventory (Novak & Guest, 1989)
CRA	Caregiver Reaction Assessment (Given et al., 1992)
DASS-21	Depression, Anxiety and Stress Scale (Lovibond & Lovibond, 1995)
ID	Intellectual disability
PRISMA-ScR	Preferred Reporting Items for Systematic Reviews and Meta-Analyses Extension for Scoping Reviews
QoL	Quality of life
WHOQOL-BREF	World Health Organization Quality of Life—abbreviated version (World Health Organization, 1996)

Caregivers within families play an important role in supporting their relative(s) with autism spectrum disorder (ASD) across the lifespan. Although caring for a loved one might have positive influences on family functioning (Beighton & Wills, 2017; Phelps et al., 2009; Sarriá & Pozo, 2015), it can also be associated with numerous impacts and burdens on the lives of family caregivers of individuals with ASD (Hoefman et al., 2013; Marsack & Hopp, 2018; Sonido et al., 2022; Tint & Weiss, 2016). To date, research primarily focused on parental caregivers of children with ASD (Bonis & Sawin, 2016; Bromley et al., 2004; Davy et al., 2022; Safe et al., 2012), but the perspective of family caregivers of adults with ASD is gaining importance (Hare et al., 2004; Liao & Lin,

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2013). This is essential, as ASD is a lifelong condition and impairments are pervasive across the lifespan. However, little is known about the specific challenges of caring for adult relatives with ASD and how this might affect family caregivers (Cridland et al., 2014). Thus, a better understanding of the burden of family caregivers would "promote the well-being of families, which in turn will contribute to fostering democratic, stable and cohesive societies" (United Nations, 2012, p. 2).

Caregiving Demands of Adults with ASD

The core symptoms of ASD include persistent impairments in social communication/interactions and restrictive, repetitive, and inflexible patterns of behavior, interests, or activities (American Psychiatric Association 2013; American Psychiatric Association 2022). Additionally, challenging behaviors (e.g., self-injury, suicide attempts, and aggression) and comorbid somatic and mental disorders are common (Croen et al., 2015; Vohra et al., 2017). One-third to one-half of individuals with ASD have an accompanying intellectual disability (ID; Maenner et al., 2020; Postorino et al., 2016). The nature of these symptoms usually leads to distinct challenges in caring for an individual with ASD, such as the need for mediation in social interactions, inflexible daily routines, lack of spontaneity, or inappropriate behaviors (Cadman et al., 2012; Cridland et al., 2014). Furthermore, a lot of individuals with ASD require informal care and assistance with personal care (e.g., dressing, toileting, meals), providing transport, general housework, and/or emotional support (Järbrink et al., 2003). As there are only very few services available that support individuals with ASD in adulthood (Lord et al., 2022; Nicolaidis et al., 2015), many adults with ASD rely on support by their families (Cadman et al., 2012). For example, the majority of adults with ASD remain co-residing with their parents well into their late 20s, irrespective of the presence of a comorbid ID (Levy & Perry, 2011; Roux et al., 2015).

Caregiver Burden

Previous research defined caregiver burden as a relative's appraisal of stressors and challenges related to the provision of care (Novak & Guest, 1989). Novak and Guest (1989), who primarily focused their research on caregivers of individuals with Alzheimer's disease, defined five dimensions of caregiver burden. First, *time dependence burden* includes restrictions on the caregiver's time available for personal interests and activities (Altiere & von Kluge, 2009; Smith et al., 2010) or privacy (Marsack & Perry, 2018). Second, *developmental burden* refers to personal or social underdevelopment compared to peers who do not have a relative who requires care (Novak & Guest, 1989), which can lead to feelings of isolation and a perception of being disconnected or detached from the social environment (Hare et al., 2004; Hines et al., 2014; Marsack & Hopp, 2018). Third, *physical burden* describes caregivers' feelings of chronic fatigue and damage to physical health (Novak & Guest, 1989). Fourth, *social burden* comprise feelings of role conflicts, as well as limited time and energy that they can invest in relationships or in occupational participation (Novak & Guest, 1989). Fifth, *emotional burden* refers to negative feelings towards the relative with ASD (Novak & Guest, 1989), such as guilt and blame (Marsack & Hopp, 2018; Marsack-Topolewski & Graves, 2019). In extension to this initial definition by Novak and Guest (1989), prior research reported caregiving to also worsen family caregivers' mental health, including higher levels of psychological distress (Abbeduto et al., 2004; Blacher & McIntyre, 2006), and higher prevalence's of mental disorders (Magallon-Neri et al., 2018; Schnabel et al., 2020).

In addition to these dimensions of caregiver burden by Novak and Guest (1989), Marsack and Hopp (2018), who investigated parental caregivers of adults with ASD, added the *financial burden*. This burden includes effects of caring on financial resources, such as direct costs related to specialized therapies and indirect costs related to constrains on working life (Cidav et al., 2012; DePape & Lindsay, 2015; Marsack & Perry, 2018).

To date, no comprehensive overview of burdens on family caregivers of adults with ASD was published. Existing reviews either focused on ASD in childhood (Tint & Weiss, 2016) or only assessed mental well-being of family caregivers (Sonido et al., 2019). Therefore, this scoping review aims to provide an overview of research on the abovementioned dimensions of burden on family caregivers of adults with ASD (i.e., time dependence, developmental, physical, social, emotional, and financial burden), to identify previously published interventions or supporting services designed to reduce such burden, and to detect existing knowledge gaps for further research. Thus, the following research questions were formulated: (a) What is known about the time dependence, developmental, social, physical, emotional, and financial burden on family caregivers of adults with ASD? (b) Which interventions exist to reduce caregiver burden on family caregivers of adults with ASD?

Method

This scoping review is based on the framework for scoping reviews by Arksey and O'Malley (2005), and conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses Extension for Scoping Reviews (PRISMA-ScR; Tricco et al., 2018). A scoping review protocol was developed a priori, which was not preregistered.

Eligibility Criteria

This scoping review includes different types of peer-reviewed publications (quantitative, qualitative, mixed-method studies, and reviews). The following inclusion criteria were used: (1) publications written in English or German; (2) target population was either first- or second-degree family members (including partners/spouses); (3) of adult relatives (18+ years); (4) with secured ASD diagnosis; and (5) results included indications of caregiver burden and/or interventions to reduce caregiver burden.

Search Strategy

A comprehensive search strategy was performed in the databases PubMed and EBSCOhost between June 2022 and January 2023. Publication year was not restricted. The reference and citation lists of included studies were used to locate additional eligible studies. See Table S1 for full search terms.

Study Selection and Data Extraction

After removing duplicates, titles and abstracts were screened. Studies including participants with unspecific diagnostic groups (e.g., individuals with unspecific developmental and/or intellectual disabilities) or undefined range of age were excluded to provide a homogenous evidence base. Remaining studies were screened for eligibility in full by first author (S.D.) and a trained student research assistant. Screening resulted in 93% agreement rate for a randomly selected data sample (20% of all publications). Disagreements were clarified through consented discussion.

A data extraction sheet was developed, which contained the following data items: (1) title; (2) author(s); (3) year of publication; (4) country of origin; (5) study design, method; and data analysis; (6) eligibility criteria; (7) sample characteristics of caregivers and care recipients; and (8) results and original authors' interpretation. S.D. and a trained student research assistant extracted the data from all included papers. Results were categorized into the six dimensions of caregiver burden (time dependence, developmental, physical, social, emotional, and financial burden) based on the definitions by Novak and Guest (1989) and Marsack and Hopp (2018). An overview of the burden definitions used in previous research and in the current scoping review is shown in Table S2. Some studies reported data without differentiating between the dimensions of caregiver burden, which is presented as caregiver burden composite in the "Results" section. For results with overlapping contents, burden-specific categorization was discussed with the last author (J.P.) until consensus was reached. Results over studies were synthesized by the first author and were reported according to each dimension of caregiver burden.

Results

A total of 33 articles were included in the scoping review. For details, see the flowchart of the study selection process (Fig. 1). The 33 studies consisted of 17 quantitative, eight qualitative, and three mixed methods studies, as well as five literature reviews (see Table 2 in Appendix I). Most studies were conducted in the USA (55.9%), followed by the UK (23.5%; see Table 1). The majority of family caregivers of adults with ASD were parents (in 58.1% of studies), and the most investigated burden was the emotional burden (34.6% of studies; see Fig. 2). The caregivers' age ranged from 18 to 87 years, and investigated caregivers were predominantly female (58.4–100%). The care recipients' age ranged from 18 to 96 years, and, except for one study, the majority of adults with ASD were male. Six studies investigated caregivers of adults with ASD and comorbid ID.

Dimensions of Burden on Family Caregivers of Adults with ASD

All dimensions of burden were detected within the included studies on family caregivers of adults with ASD. However, the amount of available research data differed. For an overview, see Fig. 2. In the following sections, detailed results are presented according to individual dimensions of burden. No evidence on interventions to reduce or prevent caregiver burden was identified.

Time Dependence Burden

In total, 11 studies provided evidence on time dependence burden (for details, see Table 2 in Appendix I). Except for one study, all participants were parents. All quantitative studies (*n* = 4;Marsack & Hopp, 2018; Marsack-Topolewski, 2021; Marsack-Topolewski et al., 2021; Marsack-Topolewski & Wang, 2022) measured time dependence burden with the Caregiver Burden Inventory (CBI; Novak & Guest, 1989). Studies found an increased CBI mean score for the subscale "time dependence burden," indicating that parental caregivers experienced strain

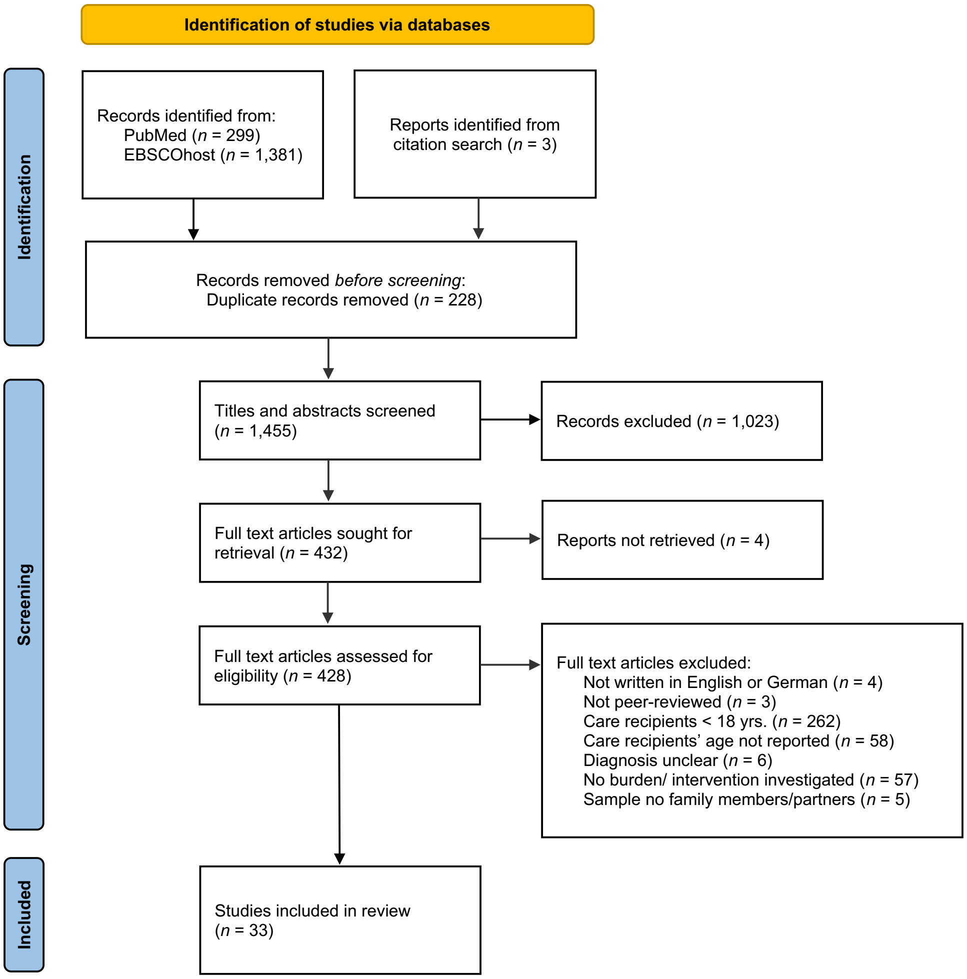

Fig. 1 PRISMA flowchart

due to caring for the adult family members with ASD on a descriptive level (Marsack-Topolewski, 2021; Marsack-Topolewski et al., 2021; Marsack-Topolewski & Wang, 2022; Marsack & Hopp, 2018). Moreover, parents who provided care to at least one adult child with ASD and another care recipient (compound caregivers) had significantly higher levels of time dependence burden than parents who only provided care for an adult child with ASD (noncompound caregivers; Marsack-Topolewski, 2021). A higher functional level or increased independence with respect to daily living skills in adult relatives with ASD was associated with less time dependence burden in parental caregivers (Marsack-Topolewski et al., 2021). However, the time dependence burden was found to be no significant predictor of parental quality of life (QoL) (Marsack-Topolewski & Church, 2019).

Qualitative studies ($n = 5$) provided deeper insights on causes and effects as well as subjective relevance of time dependence burden (Hare et al., 2004; Hines et al., 2014; Marsack & Perry, 2018; Oti-Boadi et al., 2020; Tozer & Atkin, 2015). Parents stated that they "navigate the 24/7 needs" of their adult children with ASD (Marsack & Perry, 2018, p. 545), and reported a substantial impact on their daily routines (Hines et al., 2014; Marsack & Perry, 2018) in several respects including lack of spontaneity and flexibility (Hines et al., 2014), and reduced privacy and time for themselves (Marsack & Perry, 2018; Oti-Boadi et al., 2020). Supported living for the adult child with ASD had positive effects such as more free time and freedom for the parents (Krauss et al., 2005). One study by Tozer and Atkin (2015) investigated siblings of adults with ASD and reported difficulties to spent time with their own commitments (e.g., spouses/partners, children, friends, parents, or work) and their sibling with ASD, to balance demands, and presence of a constant tension and feelings of guilt.

Developmental Burden

In total, 11 studies reported on developmental burden and all investigated parental caregivers of adult children with ASD. All quantitative studies ($n = 4$; Marsack-Topolewski, 2021;

Table 1 Identified articles

	N of included articles (%)
Country of data collection^a	
USA	19 (55.9)
UK	8 (23.5)
Australia	2 (5.9)
Other (Belgium, France, Ghana, Netherlands, New Zealand)	5 (14.7)
Relation to adult with ASD^b	
Parents (mothers and fathers)	18 (58.1)
Mixed family members	6 (19.4)
Siblings	4 (12.9)
Mothers	2 (6.5)
Spouses	1 (3.2)

N sample size, *ASD* autism spectrum disorder

[a] One study collected data from the USA and the UK

[b] Two studies did not report information on the relationship

Marsack-Topolewski et al., 2021; Marsack-Topolewski & Wang, 2022; Marsack & Hopp, 2018) detected developmental burden in parental caregivers, assessed with the CBI (Novak & Guest, 1989). Developmental burden was significantly higher when adults with ASD were more dependent in completing activities of daily living, as well as in compound caregivers (Marsack-Topolewski, 2021; Marsack-Topolewski et al., 2021; Marsack-Topolewski & Wang, 2022). It was assumed that parents who were less involved in assisting were more likely to find the time to engage in other social roles that would decrease their developmental burden (Marsack-Topolewski et al., 2021).

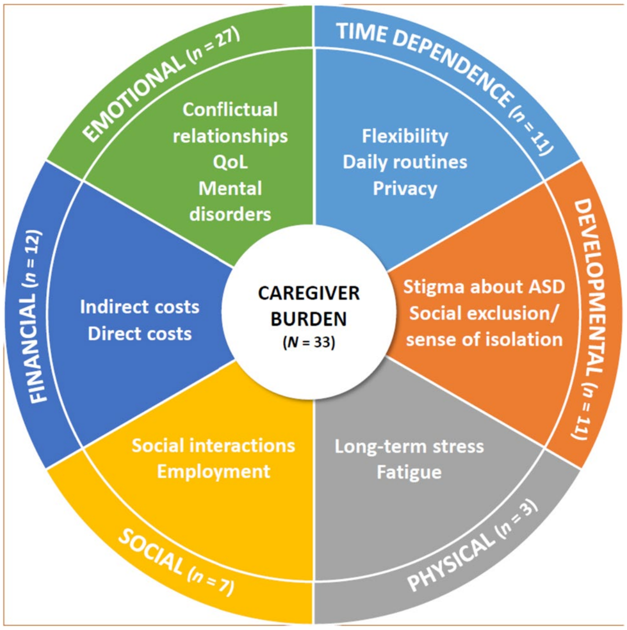

Fig. 2 Findings of burden on family caregivers of adults with ASD. ASD, autism spectrum disorder; QoL, quality of life. Some studies investigated multiple dimensions of caregiver burden

Qualitative studies ($n = 5$) discussed potential reasons for developmental burden with focus on the balancing fulfillment of own needs and providing care for the adult relative with ASD (Griffith et al., 2012; Hare et al., 2004; Hines et al., 2014; Marsack & Perry, 2018; Oti-Boadi et al., 2020). Parental caregivers reported restrictions on their lives and social exclusion, leading to feelings of isolation (Hare et al., 2004) and negative comparisons with peers (Marsack & Perry, 2018). Two main reasons for social exclusion were identified: First, caregiving demands caused the social network to shrink (Hare et al., 2004; Hines et al., 2014; Marsack & Perry, 2018; Oti-Boadi et al., 2020). Second, misunderstandings and stigma about ASD led to social exclusion (Griffith et al., 2012; Marsack & Perry, 2018; Oti-Boadi et al., 2020). The hidden nature of ASD and a lack of visible physical markers of disability may lead to the perception to be non-autistic (Griffith et al., 2012; Marsack & Perry, 2018). Parents felt that their caregiving demands were neither understood nor appreciated by the general public, leading to this sense of isolation (Griffith et al., 2012).

One mixed methods study by Krauss et al. (2005) found that one-fifth of the investigated parents reported social isolation to be a negative aspect of cohabiting with an adult child with ASD. Another mixed-method study by Marsack-Topolewski and Church (2019) found that the developmental burden (besides investigated time dependence, emotional, and financial burden) was the strongest predictor of the parental mental QoL, demonstrating an inverse relationship.

Physical Burden

Three studies (investigating siblings, parents, or especially mothers) provided indications for physical burden on family caregivers of adults with ASD. One quantitative study reported decreased general health in siblings of adults with ASD compared to siblings of adults with Down Syndrome (Hodapp & Urbano, 2007).

Qualitative data revealed that parents developed protracted physical illnesses and suggested it was caused by long-term stress and worry related to the care of their child with ASD (Oti-Boadi et al., 2020).

A mixed-method study found that living together with the adult child with ASD is related with stress and fatigue (Krauss et al., 2005).

Social Burden

The social burden was studied in one quantitative, five qualitative, and one mixed-method study, investigating siblings and parents. Quantitative data revealed that siblings of adults with ASD reported a stronger impact on their relationship with the parents compared to siblings of adults with Down Syndrome (Orsmond & Seltzer, 2007).

All qualitative studies (*n* = 5) interviewed parental caregivers and found a negative impact of continuous care on other relationships and social activities (Hare et al., 2004; Hines et al., 2014; Marsack & Perry, 2018; Oti-Boadi et al., 2020; Tozer & Atkin, 2015). Parents discussed the potential threat of a crisis in their child's life and a lack of adequate respite care impacting their own social and recreational opportunities (Hines et al., 2014). This applied to friendships as well as to relationships within the families (Marsack & Perry, 2018; Oti-Boadi et al., 2020). For example, the feeling of not knowing the own partner/spouse anymore (Hare et al., 2004) or lack of time for other children without ASD (Hare et al., 2004; Marsack & Perry, 2018) were described because the majority of the time was spend caring for the adult family member with ASD. This was assumed as possible reasons why some families took the adult child with ASD into care (Hare et al., 2004). Furthermore, one study identified disruptions in the professional careers of maternal caregivers, as they could not combine work and care for the adult child with ASD (Oti-Boadi et al., 2020).

A mixed-method study explored positive and negative aspects of adult children with ASD living at home or in care, revealing that the living situation influences the social and work life (Krauss et al., 2005).

Emotional Burden

In total, 27 studies provided evidence on the emotional burden of caregiving in family caregivers of adults with ASD. Of these studies, 11 studies provided evidence on negative feelings or relationships towards the adult with ASD according to the definition of Novak and Guest (1989) (see Table S2). In addition, 22 studies gave evidence on influences on the mental QoL or the development of mental disorders in family caregivers.

Negative Feelings/Relationships

Quantitative studies (*n* = 6; Hodapp & Urbano, 2007; Marsack-Topolewski, 2021; Marsack-Topolewski et al., 2021; Marsack-Topolewski & Wang, 2022; Marsack & Hopp, 2018; Orsmond & Seltzer, 2007) utilized either the CBI (Novak & Guest, 1989) to investigate parents (*n* = 3) or the positive affect index of relationship quality (Bengston & Black, 1973) to investigate siblings (*n* = 2). Studies in parents revealed no emotional burden (Marsack & Hopp, 2018) or only slightly increased scores on a descriptive level (Marsack-Topolewski, 2021; Marsack-Topolewski et al., 2021). Research comparing the relationship quality of siblings of adults with ASD to siblings of adults with Down Syndrome showed that the latter felt like more respectful and fair towards their sibling (Hodapp & Urbano, 2007) and reported significantly higher levels of positive affect in their relationship than siblings of adults with ASD (Orsmond & Seltzer, 2007).

Qualitative studies (*n* = 4) investigated parents (*n* = 2), spouses (*n* = 1), and siblings (*n* = 1), and indicated an often conflictual relationship towards the adult with ASD, irrespective of the relationship (Hines et al., 2014; Lewis, 2017; Marsack & Perry, 2018; Tozer & Atkin, 2015).

A mixed-method study found that emotional burden did not predict parental QoL (Marsack-Topolewski & Church, 2019).

Influences on Mental QoL/Mental Disorders

Quantitative studies (*n* = 12) investigated parents (*n* = 5; Barker et al., 2011; Lee & Shivers, 2019; Marsack-Topolewski, 2020; Marsack & Samuel, 2017; Rattaz et al., 2017), siblings (*n* = 3; Hodapp & Urbano, 2007; Orsmond & Seltzer, 2007; Tomeny et al., 2017), or mixed family caregivers of adults with ASD (*n* = 4; Grootscholten et al., 2018; Herrema et al., 2017a, 2017b; Sonido et al., 2022). Several studies found higher levels of depressive symptoms and anxiety, and lower QoL in caregivers of adult family members with ASD (e.g. Barker et al., 2011; Grootscholten et al., 2018; Lee & Shivers, 2019). Across a 10-year period, depressive symptoms in caregivers remained constant, whereas anxiety decreased, and behavior problems of the care recipient correlated positively with depressive and anxiety symptoms (Barker et al., 2011).

Studies on caregivers' mental QoL revealed a number of predictors based on caregiver variables (e.g., age, intolerance of uncertainty, unpreparedness for the future) and care-recipient variables (e.g., ID, mental comorbidities, adaptive skills, symptom severity) (Herrema et al., 2017b; Rattaz et al., 2017; Sonido et al., 2022). The care recipients' age and the utilization of formal social support revealed no significant relation to caregivers' QoL (Lee & Shivers, 2019; Marsack & Samuel, 2017). One study reported that depressive behaviors of care recipients mediated the relationship between caregiver burden and mental QoL (Sonido et al., 2022). Lower levels of QoL were reported for compound caregivers compared to noncompound caregivers (Marsack-Topolewski, 2020). Grootscholten et al. (2018) found higher emotional distress in caregivers of adults with ASD compared to caregivers of adults with schizophrenia. Furthermore, caregivers of adults with ASD frequently expressed concerns, worries, and anxiety about the future and potential support (Herrema et al., 2017a). Findings by Sonido et al. (2022) identified caregiver coping and cognitive dispositions as a predictor for caregiver mental well-being. Studies assessing siblings of adults with ASD found that parent-focused parentification was positively correlated with anxiety and stress in these siblings (Tomeny et al., 2017), and that they experienced higher levels of depressive symptoms compared to siblings of individuals with Down Syndrome (Hodapp & Urbano, 2007; Orsmond & Seltzer, 2007).

Qualitative studies (*n* = 8) investigated burden on mental health in parents (*n* = 6; e.g., Griffith et al., 2012; Hare et al., 2004; Hines et al., 2014), siblings (*n* = 1; Tozer & Atkin, 2015), and spouses of adults with ASD (*n* = 1; Lewis, 2017). In line with the quantitative results, parents reported high levels of psychological distress, anxiety, and worries about the future (e.g., Griffith et al., 2012; Marsack-Topolewski & Graves, 2019; Oti-Boadi et al., 2020). Furthermore, the strain on marriage (possibly leading to divorce), other children, and the family unit as a whole, directly attributed to having a child with ASD, were discussed as emotionally burdening (Hines et al., 2014). Parental mental health was reported to decrease due to lack of service provision, lack of knowledge in healthcare providers, maintaining emotional balance for the family, and care recipients' inappropriate behaviors (e.g., Griffith et al., 2012; Hines et al., 2014; Marsack & Perry, 2018).

One qualitative study on siblings of adults with ASD reflected on difficulties growing up with someone who has ASD, unresolved emotional issues, and resentments about the past. Most siblings expressed sadness and frustration about the limited reciprocity in their relationship, including a sense of loss because they did not have a typical reciprocal sibling relationship and rather felt the need to protect the sibling with ASD (Tozer & Atkin, 2015). In line with other caregivers, partners/spouses reported worries about the future and unmet emotional needs (Lewis, 2017). However, they reported the lack of intimacy/sex and shifted relationship roles due to the ASD of their partner/spouse as emotionally distressing (Lewis, 2017).

A mixed-method study by Krauss et al. (2005) investigated the impact of housing situations on maternal emotional burden. They reported positive aspects of living together with the child (e.g., peace of mind, shared love) and several negative aspects (stress, negative impact on siblings, worries about the future) affecting mental health. Positive aspects of children living outside the home (e.g., better marriage, benefits to other children) and negative aspects (e.g., feelings of guilt/worries/concerns, missing of child) were reported, too.

A systematic review by Sonido et al. (2019) on mental well-being of caregivers of adults with ASD differentiated contributors to carer stress and carer resources. Carer stress based on the previously mentioned care-recipient-related factors and carer-related contributors to stress. Potential carer resources included higher socioeconomic status, age, education, number of children, informal support received, marital status and support, caring relationship, perceptions of ASD, and optimism.

Financial Burden

In total, 12 studies reported evidence on financial burden in parental caregivers of adults with ASD. Quantitative studies (*n* = 5) reported financial burden (Marsack & Hopp, 2018; Marsack-Topolewski, 2021; Marsack-Topolewski et al., 2021; Marsack-Topolewski & Church, 2019; Marsack-Topolewski & Wang, 2022), always assessed with the Caregiver Reaction Assessment (CRA; Given et al., 1992). Two studies reported that financial burden was significantly higher in noncompound caregivers than in compound caregivers (Marsack-Topolewski, 2021; Marsack-Topolewski & Wang, 2022). One study found a weak significant association between the adult child's dependence level and parental financial burden (Marsack-Topolewski et al., 2021). The authors suggested that other factors (e.g., availability of medical insurance, discretionary income to pay for out-of-pocket services) are likely to have a stronger influence on the financial burden than the adult child's dependence in the activities of daily living (Marsack-Topolewski et al., 2021). Financial burden was not a predictor of parental QoL (Marsack-Topolewski & Church, 2019).

All qualitative studies (*n* = 3) reported loss of employment and high costs for care as potential sources of financial burden in parents (Hare et al., 2004;Marsack & Perry, 2018 ; Oti-Boadi et al., 2020). Two studies reported associations between difficulties to combine work with care, which led to unemployment and negative effects on the financial situation (Marsack & Perry, 2018; Oti-Boadi et al., 2020). In addition, increased need for money to finance care for their adult child was reported (Hare et al., 2004; Oti-Boadi et al., 2020).

Reviews (*n* = 4) found evidence that costs of informal care and productivity loss were substantial in caregivers and substantiated a large part of overall costs related to ASD in the USA and the UK (Buescher et al., 2014; Ganz, 2007; Knapp et al., 2009; Rogge & Janssen, 2019). Absence of comorbid ID increased costs for parents of adults with ASD (Buescher et al., 2014; Knapp et al., 2009; Rogge & Janssen, 2019).

Caregiver Burden (Composite)

Six studies (five quantitative and one mixed methods study) assessed four of the caregiver burden dimensions (time dependence, developmental, emotional, and financial burden) as a composite score (without differentiation between the four dimensions) in parental caregivers. Burden were measured with the CBI (Novak & Guest, 1989), the CRA (Given et al., 1992), or caregiver burden (Heller et al., 1994). Studies showed increased composite scores for caregiver burden in family caregivers of adults with ASD (Marsack-Topolewski, 2021). In addition, they provided evidence on factors influencing the intensity of burden of caregivers. Studies reported that care recipients' health, level of maladaptive behavior, degree of independence in activities of daily living, presence of comorbid ID, and availability of informal support were related to total caregiver burden, and partially mediated the relationship between caregiver burden and parental QoL (Burke & Heller, 2016; Marsack & Hopp, 2018; Marsack & Samuel, 2017; Marsack-Topolewski et al., 2021; Marsack-Topolewski & Maragakis, 2020). Marsack-Topolewski and Wang (2022) showed significant correlations between the four dimensions of caregiver burden, with the strongest correlations between the emotional and the developmental burden, and the time dependence and developmental burden.

Discussion

This scoping review aimed to investigate and summarize the existing literature on dimensions of burden in family caregivers of adults with ASD, thereby scrutinizing the exact nature, relevance, and potentially influencing factors of specific burdens. In total, 33 studies provided evidence on all six dimensions of burden (time dependence, developmental, social, physical, emotional, and financial) or a composite score. No study was identified that provided evidence on interventions or suggestions for services tailored for family caregivers of adults with ASD.

Emotional burden was the most prominent dimension, with a focus on the impact of caregiving on family caregivers' mental health. Several care-recipient-related and carer-related variables influencing caregivers' mental health were identified. Thus, findings suggest that the expansion of the original emotional burden definition (Novak & Guest, 1989) by including these aspects like low-threshold symptoms of mental disorders and/or QoL is justified. These aspects also play a significant role in family caregivers of individuals with other chronic diseases, such as Alzheimer's disease (Pinyopornpanish et al., 2021), schizophrenia, and bipolar disorders (Karambelas et al., 2022), and should be considered in future research.

To date, all other dimensions of caregiver burden have been less thoroughly investigated both in caregivers of adults with ASD and in caregivers of other disorders, e.g., schizophrenia (Awad & Voruganti, 2008), presumably resulting from mental health being the most extensively studied and operationalized area in research compared to the other dimensions of caregiver burden. Findings indicate the presence of all dimensions but evidence remains insufficient. Especially the presence of the social and physical burden could not be conclusively clarified as qualitative findings suggest family caregivers suffer from these burdens but quantitative evidence is lacking. However, findings of all dimensions supported the assumption that, for example, the residential situation, the level of independence of the adult with ASD, and the presence of informal and formal support might have potential to reduce the intensity of caregiver burden cross-dimensional.

Some relations between the dimensions of caregiver burden have been analyzed. One study examined influences of the time dependence, developmental, emotional, and financial burden on the parental mental QoL (Marsack-Topolewski & Church, 2019). Findings of these study found that the developmental burden strongly predicts the parental mental QoL, but the reasons for this remains unclear. While the developmental burden has been studied in other research fields (e.g., dementia), there has been less in-depth research in the area of ASD (Marsack-Topolewski et al., 2021). Therefore, developmental burden needs to become a scientific focus. However, to date, the relationship between the other dimensions of caregiver burden has not been investigated, but it can be assumed that, for example, social burden is influencing emotional burden, as it is closely related to mental QoL (Beierlein et al., 2012).

With respect to the second research aim, no intervention study was included in the scoping review. However, it can be assumed that corresponding services for family caregivers of adults with ASD exist (e.g., self-help groups) that have not yet been empirically investigated. Clinically, most health care systems are organized to meet the needs of the individuals with ASD; the needs of family caregivers are rarely addressed (Karst & Van Hecke, 2012). Services could be potentially effective, particularly with regard to the emotional burden, as results from intervention studies on caregivers of children with ASD or Alzheimer's disease showed, for example, decreased depressive and anxiety symptoms and improvements in QoL (Beinart et al., 2012; Bekhet, 2017; Smith et al., 2010). Furthermore, some of the included studies indicated that caregivers demanded interventions to reduce caregiver burden (e.g. Grootscholten et al., 2018; Hare et al., 2004; Lewis, 2017).

In sum, the results reveal missing evidence on several aspects: (a) influencing factors of the specific burdens; (b) relationships between different dimensions of burden and potential dependencies; (c) associations between presence of burden and well-being of caregivers and other family members; (d) potential interactions between caregivers well-being and care recipients well-being; and (e) interventions/services to prevent and/or to reduce specific burden.

Limitations and Future Research

First, this review is limited in mapping all possible perspectives on caregiver burden in adults with ASD. There is a low ratio of participating fathers, reflecting the difficulty of recruiting fathers for autism research studies (Johnson & Simpson, 2013). Maternal perspectives are important but may not necessarily reflect those of other family members (Cridland et al., 2014). It is assumed that fathers either might not have time to participate or might not be involved into caregiving due to the traditional role of maternal parenting (Smith et al., 2010). Siblings and partners/spouses were barely examined so far, although the findings indicated that they also experience multiple caregiver burden. Furthermore, the identified studies were assessed in mostly high-income countries and, hence, do not account for additional challenges faced by families in low- and middle-income countries who may experience limited access to supports. It remains unclear whether the inclusion of other perspectives may have been associated with different emphasis of specific burdens on caregivers in adult ASD. Additional research is needed to illustrate multiple perspectives and address cultural impact on caregiver burden in ASD.

Second, quantitative data was often collected with the CBI (Novak & Guest, 1989), a questionnaire developed for caregivers of individuals with Alzheimer's disease. It remains unclear whether the CBI adequately applies to the burdens of caregivers in ASD. In addition, some dimensions appeared to be overlapping and should therefore be examined for discriminatory power.

Third, similarities in sample size and sociodemographic variables have been registered, giving reason to believe that data from the same sample may have been included in several publications, which must be taken into account in order to ensure the representativeness of the results of this scoping review (Marsack & Hopp, 2018; Marsack & Samuel, 2017; Marsack-Topolewski, 2020; Marsack-Topolewski, 2021; Marsack-Topolewski et al., 2021; Marsack-Topolewski & Church, 2019; Marsack-Topolewski & Maragakis, 2020; Marsack-Topolewski & Wang, 2022).

Lastly, reported findings were often secondary results, i.e., not the focus of the original research, and therefore might not provide a comprehensive picture. Future research should aim to combine longitudinal quantitative and qualitative data from heterogeneous samples to enable an increased focus on the dimensions of caregiver burden. A better understanding of the origin and relationship between dimensions of burden should be the priority of future research in this field. Based on this, the development and evaluation of services for family caregivers of adults with ASD should represent a long-term goal. The focus should not only include the treatment of manifest mental disorders, but also the prevention of perceived caregiver burden and the maintenance of mental health. Furthermore, there is a need to provide adequate healthcare for adults with ASD, which could also relieve the burdens on family caregivers. Future research should try to shed more light on these topics since they might be a key to improve the life of family caregivers of adults with ASD.

Conclusion

Based on our analysis, this is the first scoping review that gives a comprehensive overview on existing literature about different dimensions of burden (time dependence, developmental, social, physical, emotional, financial) on family caregivers of adults with ASD. Indications for all dimensions of caregiver burden were detected, highlighting the emotional burden on family caregivers with focus on family caregivers' mental well-being. Accordingly, potential factors of influence were reported (e.g., carer and care recipient related variables). However, evidence on all other dimensions of caregiver burden was partially lacking or conflicting. Despite the cross-dimensional burden and impact of caregiving, no interventions to address specific or overall burden in family caregivers of adults with ASD were identified in the literature. Although concerns of family caregivers are increasingly addressed in autism research, there is still a lack of comprehensive, in-depth evidence regarding the underlying mechanisms, interactions, and time course of the different dimensions of caregiver burden. Further research on all dimensions of caregiver burden is required to develop tailored services to reduce burden on family caregivers of adults with ASD.

Appendix 1

Table 2 Study characteristics

Author(s)	*N* (caregivers, ASD subgroup)	Caregiver sub-groups	Mean caregivers‘ age in years (*SD*/ Range)	Mean care recipients’ age in years (*SD*/ Range)	Key finding(s)	Outcome measure(s)	Dimension(s) of caregiver burden
Quantitative studies							
Barker et al. (2011)	379	Mothers	51.22 (10.52)	21.91 (9.40)	Anxiety and depressive symptoms	Center for Epidemiologic Studies Depression Scale (CES-D; Radloff, 1977); Anxiety subscale of the Profile of Mood States (POMS; McNair et al., 1981)	Emotional
Burke and Heller (2016)	130	Parents (mothers = 83.9%, fathers = 16.2%)	54.64 (9.77)	25.02 6.59)	Maladaptive behaviors and poor health of the adult with ASD related to greater caregiving burden	Caregiving satisfaction (Lawton et al., 1982); Caregiver self-efficacy (Heller et al., 1999); Caregiving burden (Heller et al., 1994)	Composite
Grootscholten et al. (2018)	NR	Spouses = 56.7%, parents = 29.8%, other (sibling, child, other family member) = 13.5%	48.2 (11.3)	39.9 (14.0)	Caregivers of adults with ASD experience overall consequences comparable to caregivers of patients with depression or schizophrenia	Involvement Evaluation Questionnaire (IEQ; van Wijngaarden et al., 2000); General Health Questionnaire (GHQ-12; Goldberg et al., 1997); Autism-Spectrum Quotient (AQ; Baron-Cohen et al., 2001)	Emotional
Herrema et al. (2017b)	109	Parents = 82%, siblings = 8%, carer = 1%, spouses = 4.5%, others = 4.5%	54 (9)	27 (9)	Co-occurring mental health difficulties of care recipient associated with higher levels of worry, depression, anxiety and stress, and poorer quality of life	Intolerance of Uncertainty Scale (IUS-12; Carleton et al., 2007); Penn State Worry Questionnaire (PSWQ; Meyer et al., 1990); Depression, Anxiety and Stress Scale (DASS-21; Lovibond & Lovibond, 1995); self-developed QoL measure	Emotional

Table 2 (continued)

Author(s)	*N* (caregivers, ASD subgroup)	Caregiver sub-groups	Mean caregivers‘ age in years (*SD*/ Range)	Mean care recipients’ age in years (*SD*/ Range)	Key finding(s)	Outcome measure(s)	Dimension(s) of caregiver burden
Herrema et al. (2017a)	120	Mothers = 72%, fathers = 9%, siblings = 9%, carer = 2%, spouses = 5%	54 (9)	28 (11)	Worries about the future care of relative with ASD	concerns about the future (derived from themes from literature review)	Emotional
Hodapp and Urbano (2007)	176	Siblings	30.6 (12.6)	26.7 (13)	Compared with siblings of adults with ASD, siblings of adults with Down syndrome showed closer, warmer sibling relationships, better health, lower levels of depressive symptoms	Adult Sibling Questionnaire (ASQ; designed for this study)	Physical, emotional
Lee and Shivers (2019)	61	Parents = 96.4%	NR	24.57 (8.93)	Lower levels of parental QoL	Caregiver Strain Questionnaire (CGSQ; Brannan et al., 1997); ASQ (Hodapp & Urbano, 2007); SF-12 (Ware et al., 1995); Family Needs Questionnaire (FNQ; Kreutzer et al., 1998); Brief COPE (Carver, 1997)	Emotional
Marsack and Hopp (2018)	320	Parents (unspecified)	NR (50–74)	NR	Parental caregivers were experiencing burden in all areas except emotional burden	Caregiver Burden Inventory (CBI; Novak & Guest, 1989), Caregiver Reaction Assessment (CRA; Given et al., 1992)	Time dependence, developmental, emotional, financial, composite
Marsack and Samuel (2017)	320	Parents (unspecified)	NR (50–70 s)	NR	Caregiver burden had a negative impact on mental QoL	World Health Organization Quality of Life—BREF (WHOQOL-BREF; World Health Organization, 1996), CBI (Novak & Guest, 1989), ENRICHD Social Support Inventory (Mitchell et al., 2003)	Emotional, composite

Table 2 (continued)

Author(s)	*N* (caregivers, ASD subgroup)	Caregiver sub-groups	Mean caregivers‘ age in years (*SD*/ Range)	Mean care recipients’ age in years (*SD*/ Range)	Key finding(s)	Outcome measure(s)	Dimension(s) of caregiver burden
Marsack-Topolewski (2020)	320	Parents (unspecified)	NR (50–70 s)	NR (18–62)	Compound caregivers had lower QoL than noncompound caregivers	ENRICHD Social Support Inventory (Mitchell et al., 2003), CBI (Novak & Guest, 1989), CRA (Given et al., 1992), WHO-QOL-BREF (World Health Organization, 1996)	Emotional
Marsack-Topolewski et al. (2021)	320	Parents (unspecified)	NR (50–70 s)	NR	Caregiver burden was negatively impacted by the lack of independence in functional ability	CBI (Novak & Guest, 1989), CRA (Given et al., 1992)	Time dependence, developmental, emotional, financial, composite
Marsack-Topolewski et al. (2021)	320	Parents (unspecified)	NR (50–70 s)	25.1 (7.09)	Caregiver burden decreased, particularly time dependence and developmental burden, when relative were less dependent in daily life	CBI (Novak & Guest, 1989), CRA (Given et al., 1992)	Time dependence, developmental, emotional, financial, composite
Marsack-Topolewski and Wang (2022)	320	Parents (unspecified)	NR (50–70 s)	24.82	Significant correlations between time dependence, developmental, emotional, and financial burden	CBI (Novak & Guest, 1989), CRA (Given et al., 1992)	Time dependence, developmental, financial, emotional
Orsmond and Seltzer (2007)	77	Siblings	38.17 (7.73)	34.88 (9.62)	Higher levels of positive affect in their relationship than siblings of adults with ASD than siblings of adults with Down Syndrome	Positive Affect Index (PAI; Bengston & Black, 1973), Revised Activities of Daily Living Index (Seltzer & Krauss, 1989); Multidimensional Coping Inventory (Carver et al., 1989); Questionnaire on Ressources and Stress (Friedrich et al., 1983)	Social, emotional

Table 2 (continued)

Author(s)	*N* (caregivers, ASD subgroup)	Caregiver sub-groups	Mean caregivers‘ age in years (*SD*/ Range)	Mean care recipients’ age in years (*SD*/ Range)	Key finding(s)	Outcome measure(s)	Dimension(s) of caregiver burden
Rattaz et al. (2017)	106	Parents (mothers = 68%, both parents = 20%, fathers = 10%, guardians = 2%)	Mothers: 51.8 (1.5); fathers: 55.1 (6.8)	20.6 (1.5)	Lower levels of parental QoL	Parental—Developmental Disorder—Quality of Life (Par-DD-QoL; Baghdadli et al., 2014)	Emotional
Sonido et al. (2022)	101	Mothers = 40.59%, spouses = 22.77%, fathers = 9.9%	56.22 (21–78)	37.50 (25–96)	Caregiver-related and care-recipient-related predictors of mental QoL	WHOQOL-BREF (World Health Organization, 1996)	Emotional
Tomeny et al. (2017)	41	Siblings	25.83 (5.36)	24.2 (4.92)	Parent-focused parentification was related with anxiety and stress	DASS-21 (Lovibond & Lovibond, 1995)	Emotional
Qualitative studies							
Griffith et al. (2012)	5	Parents (mothers = 80%, fathers = 20%)	71.2 (63–82)	39.75 (37–44)	Social isolation and future concerns of parental caregivers	N/A	Developmental, emotional
Hare et al. (2004)	26	Parents (mothers = 77%)	NR	27	Association between parental emotional distress and unmet need, social isolation, loss of employment	GHQ-12 (Goldberg et al., 1997), Family Support Scale (FSS; Dunst et al., 1988)	Time dependence, developmental, social, emotional financial
Hines et al. (2014)	16	Parents (mothers = 68.75%, fathers = 25%, stepmother = 6.25%)	66.56 (58–82)	38.85 (31–44)	Strain on marriage, daily routines, lack of flexibility	N/A	Time dependence, developmental, social, emotional
Lewis (2017)	29	Partners/spouses	43.6 (21–65)	NR	High levels of stress/ emotional burden in parners/spouses	N/A	Emotional
Marsack and Perry (2018)	51	Parents (unspecified)	NR (50–70 s)	NR (18–44)	Burden due to continuous care, social exclusion, lack of privacy, costs for care	N/A	Time dependence, developmental, social, emotional, financial
Marsack-Topolewski and Graves (2019)	51	Parents (unspecified)	NR (50–70 s)	NR	Challenges/worries to future planning for adult children with ASD	N/A	Emotional

Table 2 (continued)

Author(s)	*N* (caregivers, ASD subgroup)	Caregiver sub-groups	Mean caregivers‘ age in years (*SD*/ Range)	Mean care recipients’ age in years (*SD*/ Range)	Key finding(s)	Outcome measure(s)	Dimension(s) of caregiver burden
Oti-Boadi et al. (2020)	9	Parents (unspecified)	55.8 (50–70)	20.5 (18–23)	Reduced privacy and social interactions, social exclusion, unemployment, worries about the future	N/A	Time dependence, developmental, social, emotional, financial
Tozer and Atkin (2015)	21	Siblings	41.14 (25–67)	38.24 (25–65)	Tension and feelings of guilt, sense of loss, limited reciprocity in relationship to sibling with ASD	N/A	Time dependence, emotional, social, emotional
Mixed-method studies							
Krauss et al. (2005)	133	Mothers	Co-residing = 59.2 (8.6), living apart = 62.1 (9.4)	Co-residing = 30.2 (7.6), living apart = 32.9 (8.4)	Positive and negative effects of living situation of adult with ASD	N/A	Time dependence, developmental, social, physical, emotional
Marsack-Topolewski and Church (2019)	320	Parents (unspecified)	NR (50–74)	NR (18–44)	Developmental burden significant predictor of mental QoL	WHOQOL-BREF (World Health Organization, 1996)	Time dependence, developmental, emotional, financial
Marsack-Topolewski and Maragakis (2020)	320	Parents (unspecified)	NR (50–70 s)	NR	Behavior strongest predictor of caregiver burden	CBI (Novak & Guest, 1989)	Composite
Reviews							
Buescher et al. (2014)	NR	Parents (unspecified)	NR	NR	High costs due to productivity loss/family expenses	Productivity loss (parents) family expenses	Financial
Ganz (2007)	NR	Parents (unspecified)	NR	NR	High incremental societal indirect costs	Incremental societal costs	Financial
Knapp et al. (2009)	NR	NR	NR	NR	High costs due to productivity loss/family expenses	Average annual cost per capita for adults with ASD	Financial
Rogge and Janssen (2019)	NR	NR	NR	NR	Substantial costs of informal care and lost productivity	Costs of informal care and lost productivity for family/caregivers	Financial
Sonido et al. (2019)	NR	Multiple (parents = 69.57%)	NR	NR	Mental well-being of caregivers	N/A	Emotional

ASD autism spectrum disorder, *N* sample size, *SD* standard deviation, *NR* not reported, *N/A* not applicable

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Data Availability The datasets used and analyzed in the current study are available from the corresponding author on reasonable request.

Declarations

Conflict of Interest The authors declare no competing interests.

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Health-related quality of life in family caregivers of autistic adults

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Introduction: Family members of autistic individuals often provide support for their autistic relative throughout the lifespan which can lead to massive burden themselves. Reduced health-related Quality of Life (HRQoL) in family caregivers is assumed; however, only a handful studies on the HRQoL of family caregivers providing care to adult relatives exist as opposed to autistic children. Thus, the current study aimed to (i) investigate the current state of physical and mental HRQoL of family caregivers of autistic adults compared to the general population, and (ii) examine caregiver-related (e.g., age, subjective caregiver burden) and care recipient-related variables (e.g., symptom severity, utilization of formal services) explaining variance in the caregivers' HRQoL.

Methods: $N = 149$ family caregivers completed a nationwide online survey, including the Short-Form Health Survey (SF-8) in order to assess the HRQoL. T-tests were used to compare the HRQoL of family caregivers with the general population. Bivariate correlational and multiple linear regression analyses were conducted in order to identify predictors explaining variance in family caregivers' HRQoL.

Results: Family caregivers of autistic adults reported significantly lower physical ($M = 46.71$, SD = 8.72, Cohen's $d = 0.42$) and mental HRQoL ($M = 40.15$, $SD = 11.28$, Cohen's $d = 1.35$) compared to the general population. Multiple linear regression with the mental HRQoL as the outcome showed a significant model ($F(11, 95) = 5.53$, $p < .001$, adj. $R^2 = .32$) with increased subjective burden explaining most of the variance in mental HRQoL ($ß = .32$, GDW = .141, $p < .001$). Multiple linear regression analysis with the outcome physical HRQoL did not reveal a statistically significant model ($F(11,95) = 1.09$, $p = .38$). However, bivariate analyses also showed a positive correlation with the subjective caregiver burden ($r = .20$, $p < .05$).

Discussion: Findings highlight the need to consider HRQoL (and caregiver burden) of family caregivers of autistic adults in several healthcare settings to monitor a potential comprised health status in early stages, with the long-term goal to improve family caregivers' HRQoL.

KEYWORDS

autism spectrum disorder, adults, family caregivers, quality of life, mental wellbeing, caregiver burden

1 Introduction

Core symptoms for the diagnosis of Autism Spectrum Disorder (ASD) include persistent specifics in social communication/interaction and restrictive, repetitive, and inflexible behavior patterns (1). In addition, somatic and mental comorbidities and challenging behaviors (e.g., aggression, self-injuring, and attempting suicide) are common (2, 3). One-third to one-half of autistic individuals have an accompanying intellectual disability (ID) (4, 5). In ASD, these specifics are very heterogeneous but often result in the need for support/assistance/care (for purpose of simplification, mentioned as "care" below). Typical caregiving demands include assistance with housekeeping, transportation, personal care (e.g., dressing, eating, toileting) (6) as well as distinct challenges, such as the need for mediation in social interactions, inflexible daily routines, or inappropriate behaviors (7, 8).

Usually, family caregivers are the most important source of support for autistic individuals (7). Accordingly, "diagnosis of autism affects not just the individual, but the entire family" [p. 45, (9)]. Especially when transitioning into adulthood and massive barriers impede adequate healthcare for autistic adults (e.g., pediatric services are no longer available, lack of funding, long waiting lists for diagnostics/treatment), autistic adults rely on the support of family caregivers [e.g., (grand) parents, partners/spouses, siblings, adult children] (9–11). Although caring for a loved one with autism also include positive effects on family caregivers (12), caregiving is typically time-intensive and exhausting (13). As autistic relatives enter adulthood, the duration and ongoing demands of caregiving can accumulate and lead to increased stress for family caregivers (14, 15), and have several negative effects on their lives, as shown by increased subjective caregiver burden (16, 17). Many family caregivers of autistic adults struggle with the care-associated responsibilities, while balancing everyday life, social interactions, and occupational responsibilities (15, 18–20). Loss of employment and high costs associated with care were discussed to lead to major financial burden (15, 20). Family caregivers of autistic adults also seem to be experiencing increased physical and mental health problems (21–23). For example, this population reported significantly higher emotional distress compared to caregivers of patients with schizophrenia and are at higher risk to develop health problems themselves (24).

Furthermore, compromised levels of health-related Quality of Life (HRQoL) in autistic adults' family caregivers were reported (25, 26), which refers to "how well a person functions in their life and his or her perceived wellbeing in physical, mental, and social domains of health" (27). HRQoL captures information on both physical and mental health status and its impact on Quality of Life (QoL). It is a useful indicator of overall health and is suitable for prevention and early detection of physical or mental diseases (28–32). As a so-called "patient-reported outcome," HRQoL comprises the perspective of patients, making it suitable for validating healthcare services and interventions and ensuring the provision of high-quality services (33). However, evidence on HRQoL in family caregivers of autistic adults is lacking, with only a handful of international studies on this topic (25, 26, 34–36), and no data from Europe. Still, existing evidence indicates that HRQoL of family caregivers of autistic adults is reduced – without HRQoL scores having been compared with other populations yet.

In attempt to identify the underlying mechanisms of this (presumed) finding, a number of potential predictors have been investigated. For instance, a systematic review by Sonido et al. (37) ($N = 23$ included studies) scrutinized potential predictors of mental wellbeing of family caregivers of autistic adults. Due to the limited amount of available evidence, a broad variety of outcomes were considered (e.g., psychological stress, mental wellbeing, HRQoL, QoL), without categorizing them as discrete components to provide discriminant evidence. In addition, the picture on several potential predictors is less clear-cut. Discrepant or lacking evidence has been identified for various caregiver-related and care recipient-related predictors: Regarding caregiver-related, and especially sociodemographic predictors, two studies found older caregivers having better mental wellbeing (26, 38), while other studies could not support this finding (39, 40). Furthermore, Grootscholten et al. (24) reported that parental and spousal caregivers show higher psychological burden (i.e., stress, depression, anxiety) compared to other family caregivers (e.g., siblings, children). With respect to school education, Greenberg et al. (40) found higher educated mothers to experience better psychological wellbeing, whereas other studies did not support such finding (22, 26, 38). In addition, predictors directly related to caregiving demands were examined: The higher the subjective caregiver burden, the lower the parental HRQoL (41). However, no significant associations were found between parental financial burden and actual time spent due to caregiving demands and HRQoL (26, 35).

Besides caregiver-related predictors, different care recipient-related predictors were assumed to be associated with family caregivers' wellbeing, focusing on clinical and healthcare utilization outcomes. Rattaz et al. (22) found that more severe ASD symptoms in care recipients predicted lower caregivers' global QoL. This prediction was not confirmed in other studies (21, 26) or only between severity of behavioral symptoms and parental HRQoL (42). Contradictory results were also found with respect to the presence of care recipients' comorbid ID, with one study showing improved caregiver mental HRQoL when the care recipient has an ID (26), while other studies did not (39, 43). No significant associations were found between utilization of formal services by the care recipient and mental wellbeing outcomes in caregivers (26, 39). Moreover, in parents of autistic children, depressive symptoms increased when the child was diagnosed with ASD at a higher age (44). However, age at ASD diagnosis as a potential predictor of HRQoL and in caregivers of autistic adults has not been examined yet.

These previous published studies provide important evidence but reveal some limitations. First, the observed primary outcome differed highly over studies (e.g., psychological distress, mental-wellbeing, QoL, HRQoL). HRQoL was found to be a valuable outcome in various healthcare settings, but most predictors were investigated only in the context of the aforementioned related constructs. Moreover, when considering HRQoL, also very little is known about the distinction between mental and physical HRQoL. Second, the picture for several potential predictors is less clear. Inconsistent or lacking evidence has been identified for various caregiver-related and care recipient-related variables. Third, most studies included mainly parental caregivers and did not investigated, for example, partners/spouses and siblings (39, 42). Moreover, as comparisons to reference populations (e.g., non-clinical populations, family caregivers of relatives with other chronic diseases) are lacking, existing evidence cannot be interpreted properly.

Abbreviations: ASD, Autism Spectrum Disorder; CarerQoL-7D, Care-Related Quality of Life of Informal Caregivers; DSM-5, Diagnostic and Statistical Manual of Mental Disorders; GDW, General Dominance Weights; HRQoL, Health-related Quality of Life; ID, Intellectual Disability; MCS, Mental Component Summary Score; PCS, Physical Component Summary Score; QoL, Quality of Life; SF-8, Short-Form Health Survey; VIF, Variance Inflation Factor.

Thus, the present study aimed for the first time to (i) investigate the current state of physical and mental HRQoL in family caregivers of autistic adults compared to the general population in Germany, and (ii) analyze a comprehensive set of previously reported caregiver-related and care recipient-related predictors of both physical and mental HRQoL in family caregivers of autistic adults. To our knowledge, this is the first broad investigation of physical and mental HRQoL in family caregivers of autistic adults. Results from this study might contribute important information by identifying parameters associated with impaired HRQoL in family caregivers and, consequently, providing an indication of the need for action within professional healthcare settings, with the long-term goal of improving the HRQoL in this underserved population, as called for by the scientific community (9, 37, 45).

2 Methods

This cross-sectional observational study was conducted as part of the research project *BarrierfreeASD* (46). Ethical approval was obtained from the Local Psychological Ethics Committee at the Center for Psychosocial Medicine at the University Medical Center Hamburg-Eppendorf (#LPEK-0227; Dec. 2020), and the study was conducted in accordance with the Declaration of Helsinki. Participation was voluntary and anonymous and informed consent was obtained before participation. An inconvenience allowance was not paid. The *BarrierfreeASD* project has been preregistered with the Open Science Framework.[1] Two autistic researchers were part of the *BarrierfreeASD* project and were involved in developing the online survey, the data collection and the interpretation of the results. Furthermore, research was conducted in close collaboration with the study's collaborative network, including autism-related associations and family caregiver-related associations, which guided the research process throughout. This manuscript was conducted in accordance with the Journal Article Reporting Standards for Quantitative Research in Psychology (JARS) (47).

2.1 Participants

This study analyzed data from $N = 149$ family caregivers of autistic adults. Participants were recruited throughout Germany using purposive, quota and snowball sampling methods via the study's network of collaborating partners, publicly available contacts from autism-related associations (including self-help and caregiver groups), and social media. Therefore, response rates could not be calculated. Participants were included if they were at least 18 years old, a first- or second-degree relative or partner/spouse of an autistic adult (care-recipient), and had sufficient language skills. The nationwide online survey was distributed using LimeSurvey (48). The data collected included questions on sociodemographic and clinical information about the family caregiver, details about the autist adult, and details about the informal care provided (see measurement section for details). Sample characteristics of caregivers and care recipients are presented in Table 1.

TABLE 1 Sample characteristics ($N = 149$).

	N	*n* (%) / *M* (*SD*)
Family caregiver-related characteristics		
Age (in years)	149	51.95 (9.48) Min-Max: 20–65
Sex	149	
Female		130 (87.2)
Male		18 (12.1)
Diverse		1 (0.7)
Relationship to autistic adult	140	
Parent		99 (70.7)
Partner/spouse		19 (13.6)
Child		14 (10)
Sibling		4 (2.9)
Other		4 (2.9)
Marital Status	149	
Married/Relationship		116 (77.9)
Divorced		17 (11.4)
Single		9 (6)
Widowed		4 (2.7)
Married (in separation)		3 (2)
Highest school education	147	
University entrance qualification (A-Levels)		110 (74.8)
Secondary school certificate		30 (20.4)
First school certificate		7 (4.8)
Highest professional degree	145	
University degree		82 (56.6)
Vocational school		36 (24.8)
Technician/master school		18 (12.4)
No professional degree		4 (2.8)
Engineering school		2 (1.4)
Other		3 (2.1)
Employment status[a]	145	
Part-time employed		64 (44.1)
Full-time employed		40 (27.6)
Not employed		20 (13.8)
Minor employment (Minijob)		8 (5.5)
In vocational training		2 (1.4)
Not applicable		12 (8.3)
Care recipient-related characteristics		
Age (in years)	140	26.69 (9.75) Min-Max: 18–60
Sex	140	
Female		32 (22.9)
Male		103 (73.6)
Diverse		5 (3.6)
Diagnosis	139	

(Continued)

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[1] https://osf.io/5x8pg

TABLE 1 (Continued)

	N	*n* (%) / *M* (*SD*)
Asperger Syndrome		96 (69.1)
Childhood Autism		19 (13.7)
Atypical Autism		18 (12.9)
Other		6 (4.3)
Co-occuring ID (yes)	140	13 (9.3)

N, sample size; SD, standard deviation; M, mean; ID, intellectual disability. ^{a} multiple responses were possible.

2.2 Measurement

2.2.1 Caregiver-related measurements

Sociodemographic data about family caregivers included age, sex, marital status, relationship to an autistic relative, school education, and employment status. *Treatment-related expenses* were measured using the questionnaire of Mory et al. (49) which assesses treatment-related expenses (e.g., medication, co-payments for therapies), practical living support (e.g., rent, help in the household), and extraordinary expenses (e.g., debt repayments, special purchases) for the autistic relative in the past year (sum in Euro). *Subjective caregiver burden* was measured using the CarerQol-7D as a feasible, valid and reliable instrument (50, 51). Seven dimensions were included in the CarerQol-7D: fulfillment, relational, mental health, social, financial, perceived support, and physical dimension. Each item measured one dimension and was rated with "no," "some," or "a lot." Tariff-based sum scores were calculated, ranging from 0 (worst informal care situation) to 100 (best informal care situation) (52). *Objective caregiver burden.* Self-developed items were used to measure the capacity of informal care. Participants were asked to indicate whether they had provided informal care for the adult relative with autism in the past 6 months. If they agreed, the number (in days) and the average duration per appointment (in hours) of each type of informal care (household, personal care, intake of medication, visits to administrative authorities, doctor's appointments, finances, other) were estimated. For the purposes of this study, the total time for informal care (in hours) was calculated.

2.2.2 Care recipient-related measurements

Clinical information on the care recipient comprised the age at ASD diagnosis and the presence of an ID. As there was no standardized proxy measurement to assess the care-recipients' symptom severity of ASD, family caregivers rated their relatives' symptom severity based on two self-developed items analogous to the "Diagnostic and Statistical Manual of Mental Disorders" (DSM-5) (1) classification of severity/required support for the two main diagnostic criteria (A. Persistent deficits in social communication and social interaction, B. Restricted, repetitive patterns of behavior, interests, or activities). Family caregivers rated the following two items on a 5-point Likert scale ("1 = none" to "5 = severe"): "How much does your autistic relative require support due to difficulties in interpersonal communication and social interactions?" and "How much does your autistic relative require support because she/he is holding on to behavioral habits, routines, or interests (e.g., difficulties in self-organization or dealing with change)?" Ratings for the two domains were used as separate independent variables for the analyses. Number of received professional healthcare and support services in the past 6 month were collected using modified versions of the German Questionnaire for the Assessment of Health Services in Old Age (FIMA) (53) and the Questionnaire on the Utilization of Medical and Nonmedical Care Services in Mental Disorders (FIMPsy) (54).

2.2.3 Health-related quality of life (outcome)

Health-related Quality of Life (HRQoL) was measured using the Short-Form Health Survey (SF-8) because it is a generic instrument to assess HRQoL in physically and mentally health as well as in burdened populations (55). As a parsimonious and user-friendly instrument, each of the eight single-items assessed one dimension of the longer SF-36 health survey and allowed the calculation of a physical component scale (PCS) and a mental component scale (MCS) (56). Items were scored on a 5-point Likert scale and the PCS and MCS were derived using an algorithmic norm-based scoring procedure, with higher scores indicating better HRQoL (0–100) (55). The SF(−8/−12/−36) is used both nationally and internationally, which allows comparisons between different populations. Previous research has shown strong reliability (parallel test reliability $r = 0.82$) and validity of the MCS (55).

2.3 Data analysis

Data were analyzed using IBM SPSS version 27 (57). Missing data were not imputed. All decisions regarding the statistical significance of findings were made using a criterion alpha level of 0.05.

Three distinct analyses were performed. First, both physical and mental HRQoL scores (PCS and MCS of the SF-8) were compared with those of the general population in Germany ($N = 2{,}552$) using two two-tailed t-tests for independent samples (58). Normative data were taken from a study by Beierlein et al. (59). Interpretation of effect size (Cohen's d) were based on the conventions of Cohen (60) (small effect size: $d = 0.2$, medium effect size: $d = 0.5$, large effect size: $d = 0.8$).

Second, bivariate correlational analyses (Pearson's correlation for metric variables, point-biserial correlations for one dichotomous and one metric variable, and Phi coefficients for both dichotomous variables) were performed in order to examine correlations between physical and mental HRQoL and variables. Interpretation of effect sizes (r) were based on the conventions of Cohen (60) (small effect size: $r = 0.10$, medium effect size: $r = 0.30$, large effect size: $r = 0.50$).

Third, two multiple linear regression models (PCS and the MCS of the SF-8 as dependent variables) were calculated to examine potential predictors of the family caregivers' physical and mental HRQoL as a comprehensive set of predictors. An *a priori* power calculation revealed a required sample size of $N = 87$, assuming a moderate to high effect of $R^2 = 0.18$, based on effect sizes of previous studies, with a statistical power of 0.80 and an alpha level of 0.05 for 11 predictors (61). Multicollinearity between predictors was assessed using the variance inflation factor [VIF; critical VIF > 2.5 (62)]. All other assumptions of multiple regression analysis could also be verified (linearity, normality, homoscedasticity, independence of errors). Predictors were entered into regression analyses in continuous or binary categorical data format in one block. To be included in regression analyses, categorical variables with more than two values/categories were recoded into dichotomous format, e.g., relationship to care recipient (1 = parental caregiver, 0 = non-parental caregiver), education (1 = A-levels/tertiary school education, 0 = no A-levels), ID

(1 = ID, 0 = no ID, see Table 2). Interpretations of effects sizes (R^2) were based on the recommendations of Cohen (60) (small effect size: $R^2 = 0.01$, medium effect size: $R^2 = 0.09$, large effect size: $R^2 = 0.25$). In order to compare the relative importance of predictors, dominance analysis was performed using R version 3.6.2 (63) package "yhat" (64). The General Dominance Weights (GDW) of predictors were calculated by averaging the squared semipartial correlations across all of the possible subset models. This measure indexes a variable's contribution to the prediction of the dependent variable, by itself and in combination with the other predictors (65).

3 Results

Descriptive statistics of the dependent and independent variables are shown in Table 2.

3.1 Comparison of HRQoL with general population

Physical HRQoL. Data of the study ($N = 120$) showed a mean PCS score of $M = 46.71$ ($SD = 8.72$; see Figure 1). The German normative sample ($N = 2{,}552$) (59) rated a mean PCS score of $M = 50.3$ ($SD = 8.39$,). T-test for independent samples indicated that the difference between the two samples was statistically significant for the PCS [$t(129.58) = 4.42$, $p < 0.001$, Cohen's $d = 0.42$], indicating a significantly lower physical HRQoL for caregivers of autistic adults compared to the general population with a small effect (60).

Mental HRQoL. Data of the study ($N = 120$) showed a mean MCS score of $M = 40.15$ ($SD = 11.28$; see Figure 1). Normative data from Germany (59) reported for the general population ($N = 2{,}552$) a mean MCS score of $M = 53.23$ ($SD = 7.82$). T-test for independent samples revealed statistically significant difference for MCS scores [$t(124.44) = 12.56$; $p < 0.001$, Cohen's $d = 1.35$], indicating a significantly lower mental HRQoL for caregivers of autistic adults compared to the general population with a large effect (60).

3.2 Correlation analyses

Bivariate analyses found several significant weak to high correlations between included HRQoL and predictors (see Table 3 for details) (60). *Physical HRQoL* was significantly positively correlated with the subjective caregiver burden ($r = 0.20$, $p = 0.031$) and negatively correlated with the symptom severity regarding behavior symptoms ($r = -0.21$, $p = 0.024$). *Mental HRQoL* was positively correlated with caregiver's age ($r = 0.19$, $p = 0.035$), subjective caregiver burden ($r = 0.44$, $p < 0.001$), care recipient's comorbid ID ($r = 0.18$, $p = 0.048$), and the amount of received formal services ($r = 0.21$, $p = 0.005$). Inverse correlations were found with the school education ($r = -0.27$, $p = 0.003$) and the symptom severity regarding behavior symptoms ($r = -0.27$, $p = 0.005$). Remaining variables did not show significant correlations with the physical and mental HRQoL.

3.3 Predictors of HRQoL

Multiple linear regression analysis with the outcome PCS scores did not reveal a statistically significant model [$F(11, 95) = 1.09$, $p = 0.38$]. For details, see Table 4.

Multiple linear regression with the mental HRQoL as the outcome showed a significant model [$F(11, 95) = 5.53$, $p < 0.001$], with an adjusted R^2 of 0.32 (see Table 4). Significant results were obtained for

TABLE 2 Descriptive statistics of variables.

Variable	*N*	*M (SD)*	Min. – Max.	*n* (%)
Dependent variables				
Physical HRQoL (PCS of SF-8)	120	46.71 (8.72)	25.9–61.8	
Mental HRQoL (MCS of SF-8)	120	40.15 (11.28)	15.08–62.86	
Independent variables				
Caregiver-related variables				
Age (years)	149	51.95 (9.48)	20–65	
Relationship (parental)	149			99 (70.7)
School education (A-Levels)	147			110 (74.8)
Treatment related expenses (Euro)	125	4968.82 (7490.66)	0–49,900	
CarerQol-7D (sum score)	114	56.08 (14.94)	11–89	
Informal care (hours)	125	1540.54 (3163.73)	0–23,280	
Care recipient-related variables				
Severity (Communication)	129	3.74 (0.99)	1–5	
Severity (Behavior)	129	3.77 (1.01)	1–5	
Age at diagnosis (years)	140	17.44 (12.02)	3–57	
ID (yes)	140			13 (9.3)
Formal services (sum score)	139	1.21 (1.16)	0–5	

N, sample size; M, mean; SD, Standard Deviation; HRQoL, health-related Quality of Life; SF-8, Short Form Health-Survey; PCS, Physical Component Scale; MCS, Mental Component Scale; ID, Intellectual Disability.

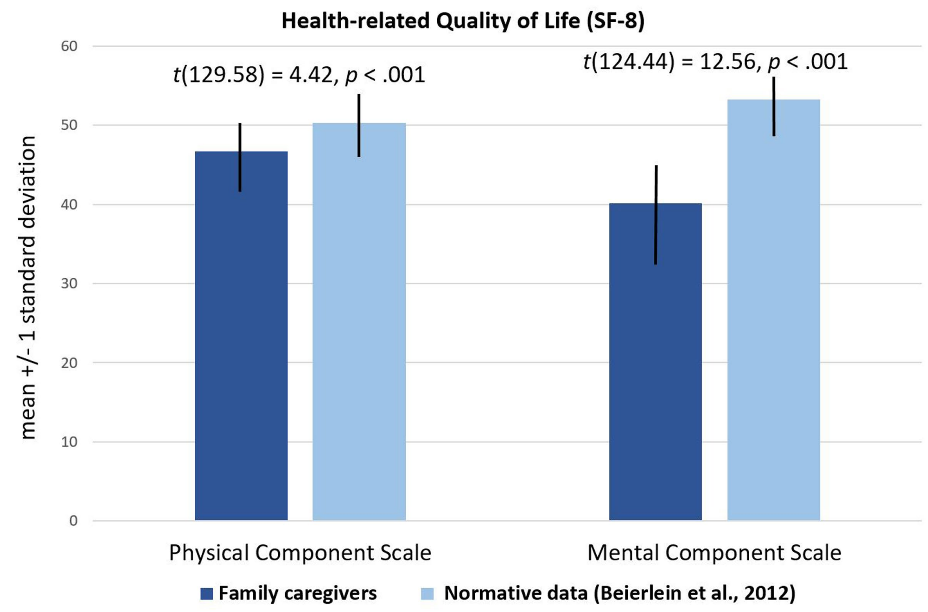

FIGURE 1
Comparison of HRQoL in family caregivers of autistic adults and in the general population.

TABLE 3 Correlation matrix ($N = 120$).

	1	2	3	4	5	6	7	8	9	10	11	12
1. Caregiver's age (years)												
2. Relationship (parental)	**0.45****											
3. School education (A-Level)	−0.16	**−0.22***										
4. Treatment-related expenses (Euro)	0.04	−0.02	0.16									
5. CarerQol-7D (sum score)	0.05	0.00	**−0.27****	−0.10								
6. Informal care (hours)	−0.03	0.11	−0.12	0.18	0.01							
7. Severity (social communication)	0.06	0.11	−0.05	0.09	−0.05	**0.33****						
8. Severity (behavior)	0.02	0.12	−0.16	0.02	**−0.20***	0.18	**0.59****					
9. Age at ASD diagnosis (years)	**−0.17***	**−0.48****	0.06	−0.03	0.08	**−0.21***	−0.11	−0.12				
10. ID (yes)	−0.11	0.10	0.01	−0.05	0.08	**0.20***	**0.22***	0.15	−0.15			
11. Formal services (sum score)	0.03	−0.04	0.10	**0.22***	−0.11	0.01	**0.27****	0.14	0.07	−0.16		
12. PCS (SF-8)	−0.12	−0.12	0.04	−0.05	**0.20***	−0.11	−0.13	**−0.21***	0.10	0.04	−0.08	
13. MCS (SF-8)	**0.19***	0.14	**−0.27****	−0.08	**0.44****	0.01	−0.13	**−0.27****	−0.13	**0.18***	**−0.26****	**0.21***

PCS, Physical Component Scale; MCS, Mental Component Scale; SF-8, Short Form Health Survey; ID, Intellectual Disability. *$p < 0.05$; **$p < 0.01$. Bold indicates significant values.

the following caregiver variables: school education ($\beta = -0.22$, GDW = 0.068, $p < 0.05$) and CarerQol-7D sum score ($\beta = 0.32$, GDW = 0.141, $p < 0.001$). Higher school education and lower CarerQol-7D sum scores predicted lower MCS scores. For care recipient variables, the age at diagnosis ($\beta = -0.20$, GDW = 0.023, $p < 0.05$) and the number of formal services ($\beta = -0.19$, GDW = 0.054, $p < 0.05$) were significant predictors: high age at diagnosis and a higher

TABLE 4 Multiple linear regression analyses (*N* = 107).

	Physical HRQoL				Mental HRQoL					
Predictor	*b*	*SE*	*ß*	*95% CI*	*b*	*SE*	*ß*	*95% CI*	GDW	VIF
Caregiver										
Age (years)	−0.17	0.11	−0.84	[−0.40, 0.05]	0.20	0.12	0.16	[−0.05, 0.44]	0.018	1.53
Relationship (parental)	0.40	2.74	0.02	[−5.05, 5.85]	−3.00	2.98	−0.12	[−8.92, 2.92]	0.006	2.07
School education (A-Level)	−0.02	2.29	−0.01	[−4.56, 4.52]	**−5.78***	**2.48**	**−0.22**	**[−10.71, −0.85]**	**0.068**	1.33
Treatment-related expenses (Euro)	<0 0.001	<0 0.001	0.02	[0.00, 0.00]	<0 0.001	<0 0.001	0.01	[< 0.001, <0.001]	0.003	1.14
CarerQol-7D (sum score)	0.11	0.06	0.19	[−0.01, 0.23]	**0.24*****	**0.01**	**0.32**	**[0.11, 0.37]**	**0.141**	1.23
Informal care (hours)	0.00	0.00	−0.08	[−0.01, 0.00]	<0.001	<0.001	−0.03	[−0.01, 0.01]	<0.001	1.21
Care recipient										
Severity (social communication)	−0.57	1.14	−0.06	[−2.83, 1.70]	−0.49	1.24	−0.04	[−2.95, 1.97]	0.013	1.74
Severity (behavior)	−0.52	1.11	−0.06	[−2.73, 1.69]	−2.05	1.21	−0.18	[−4.45, 0.35]	0.039	1.65
Age at diagnosis (years)	0.07	0.09	0.09	[−0.12, 0.25]	**−0.20***	**0.10**	**−0.20**	**[−0.40, −0.01]**	**0.023**	1.56
ID (yes)	1.41	2.88	0.05	[−4.31, 7.13]	4.94	3.13	0.14	[−1.28, 11.15]	0.025	1.18
Formal services (sum score)	−0.29	0.77	−0.04	[−1.81, 1.24]	**−1.77***	**0.84**	**−0.19**	**[−3.42, −0.11]**	**0.054**	1.22

Outcomes: physical and mental HRQoL (PCS and MCS of SF-8). Model fit (physical HRQoL): F(11, 95) = 1.09, $p = 0.38$. Model fit (mental HRQoL): F(11, 95) = 5.53, $p < 0.001$, $R^2 = 0.32$. ID, intellectual disability; SE, Standard Error; CI, Confidence Interval; GDW, General Dominance Weights; VIF, Variance Inflation Factor. *$p < 0.05$; **$p < 0.01$; ***$p < 0.001$. Bold indicates significant values.

number of formal services used by the autistic adult predicted lower mental HRQoL. Remaining predictors were not significant.

4 Discussion

To our knowledge, the present study is the first to provide an assessment of physical and mental HRQoL in family caregivers of autistic adults in Germany. The main results show that both mental and physical HRQoL were significantly reduced compared to the German normative population. Especially family caregivers' mental HRQoL was considerably lower than that of the normative population. To shed light on the reduced HRQoL, bivariate analyses was conducted in order to detect correlations between the physical and mental HRQoL and the variables: Subjective caregiver burden and ASD severity in terms of repetitive and restrictive behaviors, interests, and activities correlated with both physical and mental HRQoL. Furthermore, a comprehensive set of potential predictors of HRQoL was examined: The set of variables investigated in this study showed that the strongest predictor of mental HRQoL was the subjective caregiver burden. Understanding the underlying causes of reduced HRQoL in family caregivers of autistic adults might help to inform professionals in the healthcare system about especially vulnerable individuals and to develop and decide about adequate support strategies.

4.1 Reduced HRQoL in family caregivers of autistic adults

In contrast to previous studies, the reported HRQoL scores were compared to the general population to provide a reference. Here, family caregivers of autistic adults reported significantly lower physical and mental HRQoL compared to the German general population. The HRQoL scores found in the current study are comparable to a study from the United States that investigated HRQoL in parental caregivers of young autistic adults (25), suggesting a ubiquitously reduced HRQoL in caregivers of autistic adults regardless of country of origin. However, the mental HRQoL scores in our study were slightly lower than the findings of Lee and Shivers (25). In addition, compared to family caregivers of patients with severe mental illness (66) or Down Syndrome (67), participants in the current study reported the lowest mental HRQoL scores. It is questionable whether, for example, autism-specific care needs or differences in healthcare structures due to regional disparities or inequalities in the healthcare of autistic adults and their family caregivers compared to other mental or physical disorders lead to these findings. As poor HRQoL scores are associated with several negative outcomes in the general population such as multimorbidity (68) or higher mortality risk (69), these findings are a matter of great concern and should motivate the development of support systems. Furthermore, the findings emphasize the importance of HRQoL as a relevant outcome not only in autistic adults but also in their family caregivers in order to obtain early indications of the family caregivers' health status (70). Professionals working with autistic adults and their families should be aware that provided care may have an impact on the physical and mental health status (41). In addition, the findings highlight the need to address the lack of evidence on physical HRQoL in future research.

4.2 Bivariate and multiple regression analyses of family caregivers' HRQoL

The results of this study revealed caregiver-related and care recipient-related predictors explaining variance of HRQoL in family caregivers of autistic adults, which were predominantly differing between the two outcomes physical and mental HRQoL. However, two variables showed significant correlations with both physical and mental HRQoL in the bivariate analyses: Subjective caregiver burden and ASD severity regarding repetitive and restrictive behaviors, interests, and activities. Also in the multiple regression model with the *mental HRQoL* as dependent variable, subjective caregiver burden explained most variance in family caregivers' mental HRQoL. That is, the lower the perceived caregiver burden, the higher the mental HRQoL [in line with (26, 41)]. As described before, caregiver burden entails the demands, challenges, and stressors experienced by those who are providing care (41) on several dimensions, i.e., negative feelings resulting from informal caregiving, lack of support from family and friends, relationship problems, mental and physical health problems, problems with activities of daily living, and financial problems due to caregiving responsibilities (50). The present results indicate that the sum of these dimensions of caregiver burden explain variance in the mental (and in the bivariate analyses also in the physical) HRQoL, but it remains unclear whether certain dimensions clarify more/less variance in HRQoL compared to others, so that precise support approaches to reduce perceived caregiver burden cannot yet be defined. Interestingly, more objective indicators of caregiver burden (i.e., capacity of informal care) did not significantly predict the HRQoL (26), suggesting that it is not the actual care provided, but the individual's perception of the care situation that has an impact on the HRQoL. This provides an initial benchmark for developing appropriate services, such as psychosocial interventions. Evidence is lacking, but first data on interventions for parents of autistic youth and adults seem promising, as mindfulness-based group interventions significantly reduced parental stress (71, 72). In addition to formal services, informal social support (i.e., support from unpaid sources such as family members, friends, or acquaintances) was found to reduce both subjective caregiver burden and mental HRQoL (41).

Better school education has previously been reported to increase maternal mental well-being (40). Interestingly, our data revealed a significant inverse association, indicating that higher school education predicted lower mental HRQoL. It is possible that family caregivers with higher school education tend to have, in turn, higher ambitions for their own lives and for the life of their care recipients, but are constrained by the demands of caregiving. Well-educated people were found to have higher levels of dissatisfaction, and mental distress was largely reduced by paid work (73), but employment is often negatively affected by caregiving demands among caregivers of autistic adults. Such associations need to be investigated in future research.

To our knowledge, this is the first study to show an association between care recipient's age at ASD diagnosis and family caregiver's mental HRQoL with an inverse relationship: Higher age at diagnosis predicted lower mental HRQoL. As mentioned in the Introduction, autistic adults face massive healthcare barriers, such as long waiting lists for diagnostics (3, 9, 10, 74). Delayed diagnosis might lead to delayed receipt of appropriate formal support. Such associations need further investigation, but it can be assumed that inadequate healthcare structures for autistic adults affect the whole family.

As expected by Sonido et al. (37), a higher amount of received formal services predicted lower mental HRQoL among family caregivers. The number of formal services could be a proxy for care recipients' symptom severity and/or treatment needs. However, there was only a weak correlation between formal services and the severity of social interaction and communication symptoms. Recent studies have shown that the presence of care recipient's depression (26) and general health deterioration (75) are associated with reduced caregiver well-being, but associations with HRQoL need to be clarified in future research. Furthermore, many family caregivers have had negative experiences with formal services (18), which may account for the negative association. In addition, in line with previous research, the presence of an ID (26) and higher caregiver's age (26, 38) correlated with better mental HRQoL (only) in the bivariate analyses. Previous evidence confirmed that autistic adults without ID often report higher healthcare needs and barriers to accessing appropriate healthcare (76–78).

The remaining predictors were not found to significantly explain the variance of mental HRQoL in the current study, as was the relationship with the autistic adult. In contrast to Grootscholten et al. (24), the current study mainly included parental caregivers, which may result in a lack of variance to detect potential differences between different relationship groups (i.e., parents, siblings, spouses). Moreover, differences between maternal and paternal caregivers were not examined in the current study. Research on parents has shown that mothers often focus on caring for the autistic child and thus have a more intense bonding (79, 80). Nonetheless, in a recent meta-analysis, parental gender did not emerge as a significant moderator of proportions of levels of parental psychopathology, but the authors called for further investigation (81). Moreover, treatment-related costs were not associated with the family caregivers' mental HRQoL even when family caregivers reported high amounts of care-related costs (35). This finding differed from qualitative findings (20), as almost all participating parental caregivers complained about the financial constraints they faced. Buescher et al. (82) also reported substantial costs for caregivers of autistic adults, such as medical services, employment support or accommodations, which can have a tremendous impact on families. Due to the high proportion of highly educated caregivers in the current study, the income of the sample is expected to be relatively high. In addition, the positive correlation between school education and treatment-related expenses showed that family caregivers with higher education reported more treatment-related expenses. Therefore, further research should examine the subjective financial burden.

Similar to mental HRQoL findings, subjective caregiver burden also correlated with the *physical HRQoL* with a small effect size, as caregiving may have negative long-term physical effects (e.g., fatigue, muscular tension, physiological exhaustion). Another correlation was found between the physical HRQoL and the severity of care-recipients' ASD symptoms of repetitive, restrictive, and stereotype behaviors with an expected inverse relationship: The more severe the symptoms, the lower the physical HRQoL (22, 42). Managing the behavioral characteristics of care recipients may be a stressor for family caregivers, which is in accordance with research on caregivers of Alzheimer's patients (83). In contrast, ASD symptom severity regarding specifics of social interactions and communication did not explain variance in this study [*cf.* (42)].

Nonetheless, the multiple linear regression model with *physical HRQoL* as the outcome was not statistically significant. The selection of predictors in this study was based on previous evidence that mainly focused on mental HRQoL without considering physical HRQoL, which could explain the lack of significance. Other variables that were not included in our analyses could explain more variance in family caregivers' physical HRQoL, such as family caregivers' mental and physical illness, social support, or coping strategies (26, 66).

4.3 Limitations and future directions

There are limitations that need to be considered in future research. First, because this was a cross-sectional study, no conclusions about causal relationships between variables and HRQoL can be drawn. Longitudinal studies would help to further describe the HRQoL of family caregivers of autistic adults over time in order to identify possible well-suited time points for different types of interventions to support caregivers. Second, a post-hoc analysis to attempt to estimate the power of the regression coefficients showed insufficient power (84). A sample size of approximately 250 relatives would have been required to achieve adequate power. Third, as no more recent SF-8 normative data from Germany exists, normative data collected in 2004 were used (59). Recent longitudinal data from the German general population showed that the overall HRQoL increased in the meantime (85). Taking this finding into account, the difference between the normative sample and the caregiver sample might be even larger, but further investigations are needed. In addition, study population and normative population were not stratified, for example, by age or gender, because these data were not available from the normative sample. Fourth, the generalizability of the data was limited by the characteristics of the sample. For example, the majority of the current sample were mothers with high levels of education and professional degree. Maternal perspectives are important but may not necessarily reflect those of other family members (8). Equal proportions of family members are needed, to examine possible differences in terms of different relationships with the autistic adult (e.g., spouses, siblings, children). Finally, family caregivers rated their relative's symptom severity using self-developed items without prior psychometric validation.

5 Conclusion and implications

Family caregivers of autistic adults represent a highly under-recognized population in clinical research, healthcare, society, and policy. To our knowledge, this is the first study to comprehensively assess the physical and mental HRQoL of this population in Germany, not only by comparing the HRQoL of family caregivers with the general population, but also by identifying potential predictors. The results outlined considerably lower physical and mental HRQoL compared to the general population, emphasizing the need for action to improve family caregivers' HRQoL, as well as certain implications for future research and healthcare. Especially the subjective burden of care was found to have impact on both dimensions of HRQoL in this study and seems to be a promising candidate for interventions. In addition to the few emerging studies on family caregivers' mental HRQoL, the current findings reveal that physical HRQoL might depend on other predictors as mental HRQoL. There is a lack of research investigating this aspect: What causes the lower physical HRQoL? Moreover, healthcare providers working with autistic adults and their families should be aware of the potential impairment of caregivers' health status and form a routine to assess and monitor caregivers' HRQoL, for example as part of the intake assessment or in primary care (86). Further longitudinal research assessing diverse samples of family caregivers of autistic adults is needed, to elucidate specific underlying mechanisms and potential risk factors for HRQoL in this heterogeneous, complex population (9, 26, 45).

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by Local Psychological Ethics Committee at the Center for Psychosocial Medicine at the University Medical Center Hamburg-Eppendorf. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

SD: Conceptualization, Data curation, Formal analysis, Investigation, Supervision, Visualization, Writing – original draft, Writing – review & editing. SB: Formal analysis, Writing – original draft, Writing – review & editing. PG: Investigation, Writing – original draft, Writing – review & editing. HK: Conceptualization, Investigation, Writing – original draft, Writing – review & editing. DS: Conceptualization, Funding acquisition, Supervision, Writing – original draft, Writing – review & editing. AK: Conceptualization, Funding acquisition, Supervision, Writing – original draft, Writing – review & editing. PR: Investigation, Writing – original draft, Writing – review & editing. FE: Writing – original draft, Writing – review & editing, Investigation. KV: Conceptualization, Funding acquisition, Supervision, Writing – original draft, Writing – review & editing. HS: Conceptualization, Data curation, Funding acquisition, Investigation, Project administration, Writing – original draft, Writing – review & editing, Supervision, Validation. ND: Conceptualization, Funding acquisition, Investigation, Project administration, Writing – original draft, Writing – review & editing. JP: Conceptualization, Formal analysis, Funding acquisition, Investigation, Project administration, Supervision, Visualization, Writing – original draft, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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7.3 Publication 3

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RESEARCH

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Barriers and needs in mental healthcare of adults with autism spectrum disorder in Germany: a qualitative study in autistic adults, relatives, and healthcare providers

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Abstract

Background Autism refers to a neurodevelopmental condition with characteristic impairments in social interaction and communication, restrictive and repetitive behaviors, as well as difficulties in sensory information processing and daily living skills. Even though symptoms persist from early childhood throughout the lifespan and often require long-term support, there is a lack of mental health services that sufficiently meet the needs of autistic adults. Previous evidence suggested individual, professional and structural barriers to healthcare for autistic adults. Here, using a peer research approach, we sought to systematically investigate barriers and needs in mental healthcare of autistic adults in Germany at the three relevant levels (individual, professional, structural) and from three relevant perspectives (autistic adults, relatives and healthcare providers), in order to obtain specific recommendations for optimized healthcare.

Methods Maximum variation sampling was used to account for the complexity of the research field. Semi-structured, open-ended interviews were conducted with autistic adults ($n = 15$) and focus groups with relatives/partners ($n = 12$), and healthcare providers of several professions ($n = 15$). Data analysis was performed using the codebook approach of thematic analysis.

Results Poor mental healthcare of autistic adults in Germany was characterized by six central and overarching themes: (i) lack of knowledge about autism, (ii) a need for increased participation/involvement, (iii) consideration of autism-specific needs in treatment, (iv) lack of services, (v) limited access to services, and (vi) improvement of stakeholder collaboration. Themes were similarly reported across participants, emphasizing dissatisfaction in all stakeholders.

Conclusions We identified major barriers to mental healthcare for autistic adults in Germany that affect autistic adults, but are also of concern to relatives and healthcare providers. Our results point to specific and generic areas

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for improvement, independent of stakeholder perspectives, which could guide future development of needs- and evidence-based services, recommendations and guidelines of mental healthcare for people with autism across the lifespan.

Trial registration This study protocol was preregistered at the Open Science Framework (https://osf.io/5x8pg).

Keywords Autism spectrum disorder (ASD), Adults, Mental healthcare, Qualitative research, Participatory research

Background

Autism spectrum disorder (ASD) is a pervasive neurodevelopmental disorder with a global prevalence of approximately 1% [1]. Manifestations of autism include characteristic impairments in social interaction and communication, as well as restrictive and repetitive behaviors, including abnormalities in sensory information processing, and difficulties in critical domains of daily living [2]. Previous evidence showed increased rates of comorbid somatic diseases, such as epilepsy, diabetes, gastrointestinal, cardiovascular and respiratory diseases [3–5], and psychiatric conditions [4, 5]. Psychiatric comorbidities affect 75–80% of autistic adults[1], with the most common including anxiety and mood disorders, attention-deficit/hyperactivity disorder (ADHD), and schizophrenia [3, 5]. Furthermore, increased rates of premature mortality [4] and risk of committing suicide and dying by suicide [6, 7] have been reported. As such, autistic adults often require a broad variety of medical as well as mental healthcare services, including social services like supported employment [8]. As autism persists across the lifespan, the need for support also persists across the transition from childhood to adult services. Nevertheless, the majority of specialized autism services have focused on autistic children [9] or autistic adults with an accompanying intellectual disability (ID; IQ<70), with little attention paid to the needs of autistic adults without ID. Nevertheless, half to two-thirds of autistic adults do not have an ID [10, 11], with prevalence rates increasing over the last few years [12]. However, affected adults often report multiple health conditions, severe limitations in practical living skills and a substantial need for support [13], while –at the same time—having difficulties to accessing or finding appropriate services [5, 14] compared to the general population [15]. As a result, autistic adults reported higher odds of unmet healthcare needs, showed a lower general healthcare and ineffective utilization of services [16, 17]. These, in turn, feed into a vicious circle associated with declines in psychosocial functioning or health-related quality of life and higher rates of comorbidity, chronicity, and suicidality [5, 18–20]. Furthermore, the unmet healthcare needs of autistic adults lead to increased economic costs for the healthcare system, autistic adults and their families [8]. In addition to the financial burden, families and partners of autistic adults typically compensate for the lack of support, often facing high levels of responsibility and stress as reflected in, for example, increased time and emotional burden [21, 22], anxiety and depression [23]. Thus, relatives and partners of autistic adults also face healthcare needs that are currently massively underserved [24, 25].

This evidence emphasizes the need to develop more effective healthcare structures that make best use of scarce societal resources and to investigate the mechanisms underlying ineffective healthcare of autistics adults. Internationally, some barriers have already been suggested, which hinder adequate and effective healthcare for autistic adults. For the U.S., for example, Nicolaidis and colleagues [26] suggested barriers to general healthcare for autistic patients at three different levels: the individual level, the professional level, and the structural or systemic level (for purpose of simplification, mentioned as "structural level" below). Contributing factors at an individual level include impairments due to autism-related difficulties, e.g. hypersensitivity in physical examinations [27, 28] or difficulties in verbalizing medical conditions or pain [29]. The professional level, or provider level, refers to, for example, healthcare providers' (lack of) knowledge about autism [26] or providers' degree of flexibility in contact with autistic patients [30], and structural level factors generally refer to the lack of availability or accessibility of healthcare services [28, 29]. Similar barriers have been suggested for the United Kingdom [15] or Canada [29]. However, evidence on mental healthcare of autistic adults is limited, especially in Germany. The German healthcare is defined as "social insurance system" [31] and is based on the welfare state, where a statutory or private health insurance can be opted for. Both insurance systems are separated along different organizational, regulative, and financial sectors. Due to separate legislation within public health services (i.e. inpatient, outpatient, and long-term care), there is a fragmentation of service provision [32], which could, for example, lead to healthcare services not being funded. According to data of the World Health Organization (WHO), Germany's health expenditure in 2018 ranked second among European countries (11.4% of Gross Domestic Product (GDP)) [32]. Despite this resource and

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[1] We use the identity-first language, but equally support the person-first language. For further information, see https://autisticadvocacy.org/about-asan/identity-first-language/.

an established system of supply, the German Guidelines Report "Autism spectrum disorders in childhood, adolescence and adulthood Part 2: Therapy" critically remarked the lack of epidemiological or healthcare research in Germany, and the importance of analyzing care pathways, barriers and accessibility of facilities in order to make suggestions for improvement [33]. This is in line with the recently published Lancet Commission on the future of care and clinical research in autism, which emphasized the need for developing and implementing a high-quality, evidence-based, and cost-effective healthcare model [34, 35], tailored to meet the individual needs of this heterogeneous population [1, 26].

Within the realms of the bigger research project *BarrierfreeASD* [36], the present study aimed at assessing the current state of mental healthcare for autistic adults in Germany. Specifically, in a three-by-three fashion, we sought to systematically investigate barriers and needs in mental healthcare of autistic adults at the three relevant levels (individual, professional, structural) and from three relevant perspectives involving all relevant stakeholders into research (autistic adults, relatives/partners, healthcare provider) in order to gather a comprehensive picture. For this purpose, one-to-one interviews with autistic adults and focus groups with relatives/partners and healthcare providers from different professional groups were conducted. In line with recommendations for participatory research on autism [37, 38], the entire study was planned, conducted, and analyzed by a neurodivergent team involving autistic adults, as well as in cooperation with autism-, relative-, and service-related associations.

Methods

This qualitative study was conducted as part of the project *BarrierfreeASD* [36]. Ethical approval was received by the Local Psychological Ethics Commission at the Center for Psychosocial Medicine of the University Medical Center Hamburg-Eppendorf (#LPEK-0227; Dec. 2020) and the study was carried out in accordance with the Declaration of Helsinki. All participants provided consent prior to participating. Participants did not receive compensation for expenses. The *BarrierfreeASD* project was preregistered at Open Science Framework (https://osf.io/5x8pg).

Participants

Three groups of stakeholders were interviewed: (i) adults (18+years) with a diagnosis of childhood autism, Asperger syndrome, atypical autism, or autism spectrum disorder, without ID (IQ>70), (ii) first or second-degree relatives or partners (18+years) of autistic adults (not necessarily related to the autistic adult sample), (iii) healthcare providers from varying professional backgrounds and healthcare sectors (see below). A total sample size of $N=45$ (each stakeholder group $n=15$) was aimed for in order to adequately address the research questions [39].

In order to account for the heterogeneity and complexity of the research field, maximum variation sampling was conducted, a purposive sampling strategy that obtains fundamental understanding through diverse groups of individuals and perspectives [40, 41]. Furthermore, exponential discriminative snowball sampling was applied to access susceptible populations [42]. Within the group of autistic adults, a purposive sample was drawn to achieve the broadest possible diversity in terms of age at diagnosis, living situation, and support needs. The same criteria were applied in the group of relatives of autistic adults. In the sample of healthcare providers, the maximum variation was achieved in terms of their experience in the treatment of adult autistic persons and their professional background (e.g. primary care, pediatrics, adult and child psychiatry or psychotherapy, occupational therapy, speech therapy). Nonspecific recruitment criteria for all three groups were age, sex, and place of residence, respectively place of work within the group of healthcare providers.

Participants were recruited throughout Germany via the study's collaborating network of cooperating partners, publicly available contacts from autism-related associations (including self-help and caregiver groups), healthcare associations (medical chambers, therapist associations, etc.), outpatient clinics, as well as social media.

Autistic adults ($n=15$) averaged 38.9 years of age (Range=22–58; see Table 1) and 60% were male ($n=9$). Participants reported to have Asperger's syndrome (86.7%; $n=13$) or atypical autism (13.3%; $n=2$) and most of them received their formal diagnosis in adulthood (80%; $n=12$). The average time until correct ASD diagnosis was 8.1 years (Range=0.5–45). Every participant reported at least one psychiatric or neurological co-morbidity, while 40% ($n=6$) reported two co-morbidities, 13.3% ($n=2$) reported three, and one autistic adult reported five co-morbidities (6.7%). The most frequently reported psychiatric co-morbidities were depression (64.3%; $n=9$), ADHD (42.9%; $n=6$), and anxiety disorders (28.6%; $n=4$).

In the group of relatives/ partners ($n=12$) the mean age were 54.25 years (Range=32–65), and most were female (83.3%; $n=10$). The majority of the participants were parents or people under legal custody (75%; $n=9$) and the others were partners (33.3%; $n=4$). In the group of healthcare provider, the mean age was 49.8 years (Range=33–71) and most of them were female (86.7%; $n=13$). Healthcare providers reported different professions: nine were psychotherapists (60%), two were

Table 1 Demographic characteristics

	Autistic adults ($N=15$)	**Relatives/ partners ($N=12$)**	**Healthcare providers ($N=15$)**
	N (%)/ *M* (*SD*)	*N* (%)/ *M* (*SD*)	*N* (%)/ *M* (*SD*)
Demographic characteristics			
Age at participation (years)	38.9 (12.5)	54.3 (9.5)	49.8 (10)
Gender (male)	9 (60)	2 (16.7)	2 (13.3)
Marital status			
single	11 (73.3)		
relationship	4 (26.7)	12 (100)	
Employment *[1]			
full-time	3 (20)	6 (50)	
part-time	4 (26.7)	3 (25)	
minor	1 (6.7)	1 (8.3)	
not employed	2 (13.3)	2 (16.7)	
Region of Germany *[2]			
North	7 (46.7)	7 (58.3)	6 (40)
East	1 (6.7)	0	5 (33.3)
South	5 (33.3)	2 (16.7)	1 (6.7)
West	2 (13.3)	2 (16.7)	3 (20)

Note. N: sample size; *M*: mean, *SD*: standard deviation; *[1]Missing data of five autistic adults; *[2]Missing data of one relative/partner; One healthcare provider specified two regions

psychiatrists (13.3%), and one each were psychologist, pediatrician, occupational therapist, and art therapist (each 6.7%). The average experience of working with autistic individuals were 16.93 years (Range=5–43). 80% reported working with autistic adults in their daily business ($n=12$). The majority of the healthcare providers reported providing counselling (93.3%; $n=14$), diagnostics (80%; $n=12$), and/or psychotherapy (individual therapy=73.3%; $n=11$; group therapy=60%; $n=9$).

Data collection

First, a systematic literature search of national and international evidence on mental healthcare in autistic adults was conducted to identify previously published barriers, facilitators, and needs in order to develop guideline questions. Because of the limited evidence on mental healthcare for autistic adults, we employed an exploratory qualitative approach with broad guideline questions to obtain a comprehensive and deep understanding of participants' mental healthcare experiences [43]. Three interview guides were developed, tailored to each stakeholder group. All interview guides contained identical questions about barriers, facilitators, and needs in healthcare, especially with focus on mental healthcare, of autistic adults on an individual, professional, and structural level in general and while transitioning into adulthood, and recommendations for improving mental healthcare for autistic adults. The interview guide for autistic adults was developed in collaboration with peer researchers of the *BarrierfreeASD* study and included additional questions about their diagnosis and possible psychiatric or psychotherapeutic treatment. Relatives' interview guide also included questions about the use of services especially for relatives (such as self-help groups for relatives). Healthcare providers were also asked about severity-based measures in the mental healthcare of autistic adults. Interview guides where piloted twice, once internally with study staff and once with an external psychologist.

In previous studies, participants with autism preferred one-to-one interviews [44]. Therefore, data collection in autistic adults ($n=15$) was conducted via semi-structured open-ended interviews, whereas relatives/ partners of autistic adults ($n=12$) and healthcare providers ($n=15$) participated in semi-structured focus groups. Data collection was conducted by two trained researchers (SD & PG, psychologists with expertise in interviewing autistic adults). An external psychologist maintained an observational protocol during the focus groups.

Because of the Covid-19 pandemic, interviews and focus groups were performed online enabled by an online video communication provider. To accommodate individual specifics in communication, autistic participants were also given the opportunity to answer interview questions in writing ($n=3$), via chat or phone ($n=3$) or to include a supporting person. Interviews lasted approximately 45–60 min and focus groups about 100–120 min, both were audio-recorded and transcribed orthographically. Identifying features in the transcription were anonymized prior to data analysis.

Data analysis

A thematic analysis was conducted, a six-phase approach to find repeated patterns of meaning across a set of data [45], following the common procedure: familiarization with data, generating initial codes, developing themes, reviewing themes, defining and naming themes, and producing the report. This approach was used independently for each data source (interviews or focus groups). MAXQDA 2020 [46] was used for data management and analysis. A mixed inductive-deductive codebook approach at a semantic level was applied. This approach is suitable for describing and summarizing qualitative data and participants' views on a specific topic [47]. For this purpose, codes were generated both deductively (developed from the existing literature on the mental healthcare of autistic adults) and inductively (derived from the qualitative data). All codes were combined into a codebook and had clinical or policy implications for the mental healthcare of autistic adults. Finally, these codes were used to identify themes (i.e. main issues, complex concepts). Two authors as well as two student research assistants completed a training program for thematic analysis and performed the coding procedure. To verify interpretation of the data and ensure inter-rater reliability, codes were discussed with the multidisciplinary study team, and the themes were developed iteratively.

Using a participatory approach, two autistic peer workers as well as study's collaboration network, including autism-related and family caregiver-related associations, were involved in developing the interview guides, the recruitment of participants, and the data analysis.

Results

Data analysis for the three stakeholder groups revealed a variety of barriers, facilitators, and needs in the healthcare of autistic adults in Germany, focusing on medical and psychotherapeutic care. In addition, recommendations for improving healthcare were identified. This resulted into six group-overarching themes with several subthemes that are outlined below: (1) lack of knowledge about autism, (2) a need for increased participation/ involvement, (3) consideration of autism-specific needs in treatment, (4) lack of services, (5) limited access to services, (6) improvement of stakeholder collaboration (see Table 2). The majority of themes and subthemes were reported equally across the three stakeholder groups. Differences are reported within the description of the themes.

Although interviews and focus groups were structured to assess barriers and needs per level (individual, professional, structural) across groups, the resulting themes were not selective to a single level but overarching in nature and were not perfectly equal across groups. Figure 1 provides a simplified visualization of the main findings, clustering the themes (shown in circles) by relevant stakeholder group (x-axis) and respective level (y-axis).

The following sections give a detailed overview about the separate themes. Participant identification numbers are used below to indicate direct quotations in accordance with the consolidated criteria for reporting qualitative research (COREQ) [48].

Lack of knowledge about autism

The theme "lack of knowledge about autism" in adulthood was mentioned by all participants as an important barrier in healthcare of autistic adults at all three levels (individual, professional and structural; see Fig. 1). For this theme, several subthemes were identified in the data: (i) stigma about ASD in society, (ii) lack of knowledge among healthcare providers, (iii) research about ASD in adulthood and in women.

First, *(i) stigma about ASD in the society* and "that public and media often have a very distorted picture of what autism actually is, and especially the area of high-functioning[2] is extremely difficult" (autistic adult; ibew6). According to participants, the portrayal of autism in the media has a negative impact on the healthcare of autistic adults because of misconceptions developed by stakeholders in the healthcare system.

Second, all stakeholder groups saw *(ii) a lack of expertise among healthcare providers* in adult autism as another major barrier. This "cluelessness" (autistic adult; ibew10) about autism would affect all professions in the healthcare system, e.g., physicians, psychotherapists, occupational therapists, and assistance providers. For example, autistic adults reported to hesitate sharing their diagnosis with healthcare providers because they feared discrimination. Participants considered the lack of education and training programs about autism to be the primary reason for this. It was "not easy at all" (healthcare provider; B1) to gain expertise in this field because healthcare providers "do not come in contact with any autism spectrum disorder treatment or even diagnostic theoretical idea, such as with anxiety disorder, personality disorder, etc." (healthcare provider; B1). A psychotherapist described:

> From my experience in advanced training in behavioral therapy, I can say that I certainly did not gain my knowledge about autism from advanced training in psychotherapy, so after six hundred hours of theory or something, I have already stopped counting and I have not had a single autism seminar, not a single one. […] I haven't had a single teaching unit in my whole education on this, which is from my current perspective, absolutely devastating […]. (B4)

Participants described that healthcare providers frequently refused treatment based on lack of knowledge. For example, responses included "oh, you are autistic,

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[2] The authors state that this is a verbatim quote, although they take a critical view of the use of the term "high-functioning". For further information, see https://pubmed.ncbi.nlm.nih.gov/31215791/.

Table 2 Overview of themes and subthemes

Theme	Subtheme	Example quotes
Lack of knowledge about autism		
	Stigma about ASD in society	"[…] public and media often have a very distorted picture of what autism actually is, and especially the area of high-functioning is extremely difficult" (autistic adult; ibew6)
	Lack of knowledge among healthcare providers	"Well, most people have heard of autism […] but very few people have any idea about autism, even the doctors, even the psychiatrists." (autistic adult; ibew10)
	Research about ASD in adulthood and women	"[…] there are no good screening tools at all." (healthcare provider; B2)
A need for increased participation/ involvement		"It is important trying to have a stronger exchange with family members" (autistic adult; ibew8)
Consideration of autism-specific needs in treatment		
	Difficulties in executive functions and self-management	"[…] estimating how long it will take me to get there, even with public transportation, because I don't drive a car. […] Or, in general, not to take on too much for one day and to estimate how long I have to plan for this appointment." (Autistic adult; ibew2)
	Need for consistency and transparency	"The biggest challenge is when I go to a new physician where I don't know the environment, the practice, everything." (autistic adult, ibew5)
	Autism-specific sensory sensitivities	"Noises are very unpleasant and hardly bearable […]" (autistic adult; ibew2)
	Difficulties in verbal and nonverbal communication	"[…] they like to be on the matter level and tend to communicate information and the emotional stuff doesn't get carried along" (autistic adult; ibew3)
Lack of services		
	Transitioning	"What would have been great is having someone by your side through transition from adolescence to adulthood, who has been by their side for a long time" (relative; I3)
	Diagnostic services	"The waiting time for a diagnosis has grown over two years." (autistic adult; ibew10)
	Psychotherapeutic services	"There are not enough diagnostic and therapeutic centers at all." (healthcare provider; T5)
	Inpatient services	"In inpatient healthcare of people with autism spectrum disorders, […] there are very, very few offers" (healthcare provider; B2)
	Low-threshold services	"There is only one telephone emergency service for autistic people in Germany and that is a voluntary one." (healthcare provider; T3)
	Assistance services	"In my opinion, even a high-functioning autistic person like me needs some kind of lifelong assistance service" (autistic adult; ibew 11).
Limited access of services		
	Lack of transparency about healthcare services	"Access to services is so massively divided among the various social systems by law that it is incredibly difficult for me to even find out who is responsible for me and from whom I have to apply for what." (autistic adult; ibew4)
	Funding	"[…] an autism therapy is usually not funded by the health insurance, because with health insurance financing there is always this healing aspect involved" (autistic adult; ibew2)
Improvement of stakeholder collaboration		"The dream of an interdisciplinary team, a place where physicians and therapists work together in a practice, accompanied by people who are well informed and well connected and can give information, kind of an autism support center […]" (relative; I7)

Note. ASD: Autism Spectrum Disorder

no, I don't have any experience with that, better contact someone else" (autistic adult; ibew11). Another autistic adult reported that "with Asperger's and Attention deficit hyperactivity disorder [ADHD] in particular, I found that I had to make a lot of phone calls to find a therapist. I think nineteen out of twenty said, 'we have no idea about that'" (ibew15). Autistic adults reported experiences like this and the subsequent burden. In addition, knowledge gaps were suggested as a reason for the high rate of misdiagnosis and resulting mistreatment of autistic adults. This was a prominent theme across all three stakeholder groups, but autistic adults and family/partners in particular reported stories of misdiagnosis, years of mistreatment, and resulting negative impact on a personal and emotional level. A man described his autistic wife's diagnostic path:

[…] my wife was also diagnosed with all kinds of things in life: schizophrenia, depression, and so on. So almost every diagnosis, but no one had the idea that it could

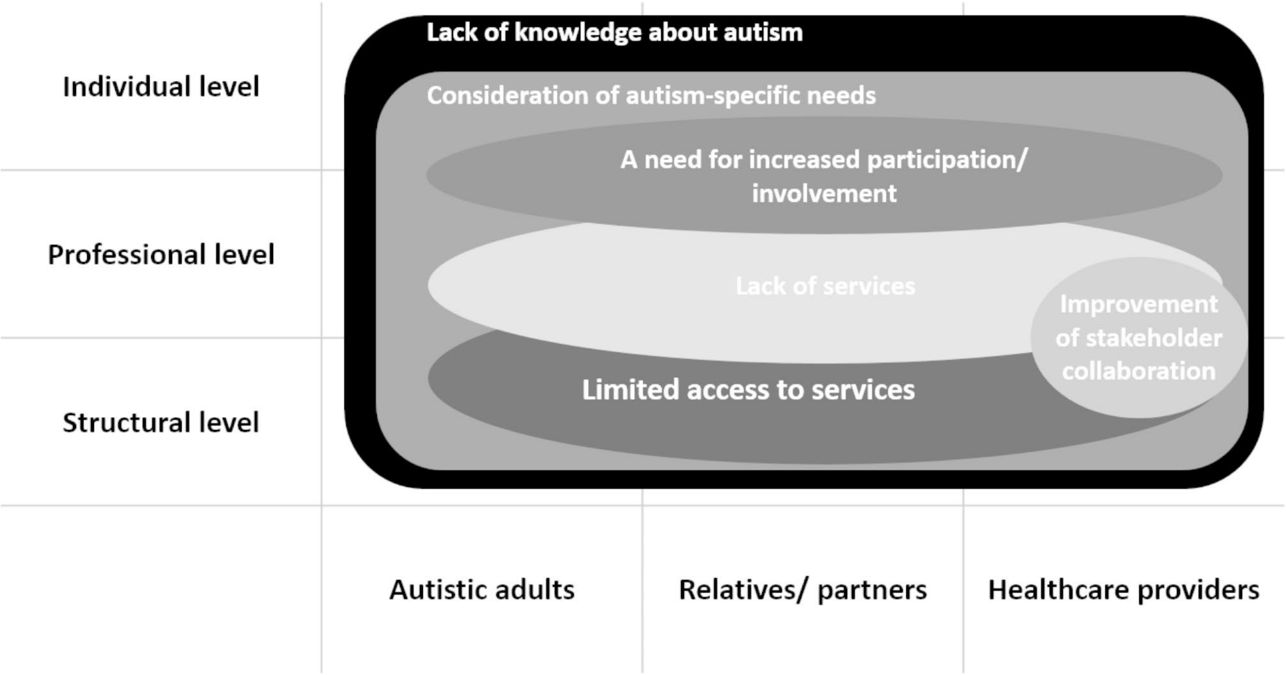

Fig. 1 Simplified visualization of themes related to level and group of stakeholders

be autism and that's just a pity that someone wastes her youth and her childhood. (Relative; I1)

Third, all three groups emphasized the need of further *(iii) research on ASD in adulthood and in women*. Participants demanded better screening and diagnostic tools for adults in general and especially for women, as it would allow sooner diagnoses and "filtering of waiting lists" (healthcare provider; B2). Furthermore, "girls *fall through the cracks*" (relative, I5) in mental healthcare because characterization and manifestations of symptoms often differ in women and they tend to be more adapted than male autistic persons:

[…] I think this is very regrettable. And I know many young women who were diagnosed in adulthood. Before that, they had been diagnosed with borderline or something else. I think that's very bad for the young women. And I would like to see that, in general, neurodiversity […] is taken into account when it comes to burnout and depression." (relative, I5).

In addition, healthcare providers identified the lack of valid measurements for severity assessment to be another field to be researched: "severity is poorly operationalized and therefore it is not possible to say in a very general way what the individual person needs in terms of support" (healthcare provider, B2).

A need for increased participation/ involvement

The theme "a need for increased participation/ involvement" refers to both the autistic adults and the relatives which should become more involved and participate in different areas of the healthcare system, according to most participants. This theme is classified at both the individual and professional level (see Fig. 1).

On the one hand, autistic adults and relatives postulated to become better involved in examinations and treatment in general. They demanded to be directly involved in decisions and to be informed about fundamental issues, such as medication, diagnoses and interventions and recommended treatment to be as transparent as possible.

Moreover, autistic adults recommended to systematically involve autistic adults into knowledge dissemination and research on ASD:

What is very important to me is bringing education about autism into the broad field. […] This is about medicine, care, and that must be brought into the broad area, and definitely in participation with autistic people. Only from the view inside it can be explained what autism is. (Autistic adult; ibew11)

In addition, all groups highlighted that "it is important trying to have a stronger exchange with family members" (autistic adult; ibew8). Support of relatives and partners, e.g., in planning and/ or accompanying appointments or activities of daily living was essential according to the participants. Without this support, many autistic adults would not be able to participate in the mental healthcare system. Nevertheless, they acknowledged barriers in involvement of relatives and demanded to "take a supporting person with you without being ridiculed" (autistic adult; ibew 2). Healthcare providers described that relatives also have a great impact on diagnostics and therapy, but are rarely involved at the moment:

And the family is not involved, it is not even implemented by the insurance company. There are always a few sessions with relatives, but the family is not really involved. And I also find that totally difficult when adults from the spectrum then end up in such an ordinary behavioral therapy. (Healthcare provider; T4)

Consideration of autism-specific needs in treatment

This theme describes challenges in accessing or participating healthcare services due to specific characteristics of autism itself. It addresses all relevant levels and was mentioned by all of the three stakeholder groups but was predominantly represented in the group of autistic adults. Following subthemes were identified in the data: (i) difficulties in executive functions and self-management, (ii) need for consistency and transparency, (iii) autism-specific sensory sensitivities, (iv) difficulties in verbal and nonverbal communication.

Participants reported *(i) difficulties in executive functions and self-management* on healthcare seeking behaviors affecting access to healthcare services, for example, when "completing very simple paperwork" (healthcare provider; B4), adhering to medication, navigating in the healthcare system, or planning/ attending appointments:

So, something like making an appointment, but also attending the appointment if I don't know the place yet. That means estimating how long it will take me to get there, even with public transportation, because I don't drive a car. So to organize something like that. Or, in general, not to take on too much for one day and to estimate how long I have to plan for this appointment. (Autistic adult; ibew2)

Furthermore, the *(ii) need for consistency and transparency* in treatment and healthcare settings was emphasized. "New situations are difficult" (autistic adult; ibew 14), including changes in staff, appointments or environment. An autistic adult described:

The biggest challenge is when I go to a new physician where I don't know the environment, the practice, everything. To get used to it is a big challenge for me, for which there is not really a solution apart from doing it. (Autistic adult, ibew5)

Autistic adults said that it would be difficult not to be involved in treatment and not to obtain "background information", for example about physical examinations (autistic adult; ibew11). Some said it was helpful if providers "explained [treatment] step by step", if they involved in medication adjustments, and informed about diagnoses.

Most participants highlighted the impact of *(iii) autism-specific sensory sensitivities* on seeking or receiving mental healthcare for autistic adults. Healthcare facilities would cause stress and overload because they are not tailored to autistic peoples' needs. For example, it was described that "physical contact is unpleasant" (autistic adult; ibew8), "smells can be very disturbing and irritating" (autistic adult; ibew4), "noises are very unpleasant and hardly bearable" (autistic adult; ibew2), "sensitivity to light leads to distraction" (autistic adult; ibew4), and overall "chaos reigns in the practices" (autistic adult; ibew 14). In addition, difficulties in body awareness and pain perception were mentioned:

On the one hand, being very sensitive during examinations and being unable to handle and endure a lot of situations, but on the other hand, not always noticing when there is pain or symptoms. In any case, these are also major barriers. (Autistic adult; ibew2)

Autism-related *(iv) difficulties in verbal and nonverbal communication* with medical staff also were a prominent topic, such as difficulties in making appointments by phone or miscommunications with healthcare providers during treatment or examinations. Autistic adults reported to struggle in following spoken instructions or answering open-ended questions. For example, an autistic participant described the following interaction with a physician:

"'Where exactly does it hurt?'" - I always feel very stupid when I can't answer that. It would be better to ask, "Is this a pain in the whole abdomen or specifically in one spot?" Then it would be easier for me to understand that it may just hurt in the whole abdomen and that this is okay as a description. (Autistic adult; ibew12)

In addition, autistic participants described problems in communicating symptoms because "they like to be on the matter level and tend to communicate information and the emotional stuff doesn't get carried along" (autistic adult; ibew3). This would cause that "many symptoms are not even recognized properly or are dismissed as incidental [...]" (autistic adult; ibew4). Autistic adults also described difficulties with typically very brief clinical appointments: "I feel rushed under time pressure and then I forget a lot of what I actually wanted to discuss. The appointments are often too short" (autistic adult, ibew9).

As mentioned before, this theme was predominately represented in the group of autistic adults. In general, they emphasized individualization of treatment of autistic adults as a facilitating factor to cross barriers in these autism-related difficulties.

Lack of services

All stakeholder groups commonly agreed upon a lack of mental healthcare services for adults with autism: "The whole system is not intended for autistic adults. It seems as autism is seen as a children's disease [...]" (autistic adult; ibew6). Especially "the high-functioning are not acknowledged or are not seen as a relevant group" (autistic adult; ibew6). Following subthemes were identified, referring to the professional and structural level: (i) transition, (ii) diagnostic services, (iii) psychotherapeutic services, (iv) inpatient services, (v) low-threshold services, (vi) assistance services.

Participants criticized the lack of adequate support systems for *(i) transition* from youth into adulthood. Mental healthcare would be provided during childhood and adolescence, but collapses once autistic individuals reach adulthood. An autistic adult reported:

The main problem is that the areas are separated from each other. The area of child and adolescent psychiatry and the area of adult psychiatry, not only in psychiatry, but also in medical care. Then you have to go from one to the other and are and treated completely differently. (Autistic adult, ibew2)

Participants demanded transitioning being adjusted in regard to the development of the individual rather than to the age and to "install a support system at least temporarily" (healthcare provider; B4).

Another prominent topic, particularly discussed by healthcare providers, was limited *(ii) diagnostic services* for adults seeking an ASD diagnosis as expressed by long waiting times for obtaining diagnostic assessment:

In [city name], healthcare is, I would say horrible, waiting times at the [name of hospital] for the autism consultation two and a half years, only a handful psychiatrists in practice, completely overloaded and waiting times about three years. (Healthcare provider; B2)

This was explained by a limited amount and limited capacity of specialized services: "there are not enough diagnostic and therapeutic centers at all" (healthcare provider; T5), especially in rural areas. Furthermore, it was expressed that psychometrically valid and specific screening and diagnostic instruments for autistic adults, also suitable for use in primary care, need to be developed which could shorten or optimize the diagnostic process.

After ASD diagnosis, participants reported "being left completely alone" (autistic adult; ibew13) with insufficient availability of subsequent *(iii) psychotherapeutic services*. Participants also described limited and scattered specialized services ("in the country side, very very few therapy options for older autistics or Asperger's autistics […]. I would have to drive 100 kilometers to get anything, any help at all." (relative; I4)). Autistic adults and relatives remarked to put up with long-distance therapy because there were no services close to their residence or waiting lists were at maximum capacity. Reported waiting times for psychotherapy ranged from nine months to one and a half years. Even when successfully accessed therapy, a few autistic adults highlighted their need for individually tailored support and personalized treatment: "The needs are very individual. And even in the high-functioning area, if you compare two people, then the needs or the level of suffering, etc. and the living situation might be very different (healthcare provider; B3)."

With respect to *(iv) inpatient services*, including psychiatric or rehabilitation services, all stakeholder groups agreed upon the need for increased specialized and personalized services. "In inpatient healthcare of people with autism spectrum disorders, […] there are very, very few offers" (healthcare provider; B2), and "shared rooms in hospital are just not acceptable. I think if you are autistic, you should have the opportunity, at least if it is possible, to get a single room" (relative; I2).

Furthermore, all groups of stakeholders criticized a lack of *(v) low-threshold services*, suggesting, for example, to expand self-help groups for autistic adults as well as for relatives:

Very little attention is paid to the fact that not only the affected person is impacted, but also a large number of people around them. I think it would be very, very good and very important if more counselling were created, for example get relatives informed or they can also experience support for themselves. (Autistic adult; ibew15)

Relatives/ partners described stress and burden due to the support of the autistic relative. Therefore, they "would really like to have support also as a relative. That you are taken by the hand in order to understand conflicts better, to accept peculiarities better and to spend a good and stress-free life together with the affected person" (relative; I2).

Autistic adults highlighted the need for further low-threshold services, such as music, sports, or occupational therapy, specialized for autistic adults or low-threshold crisis counselling: "there is only one telephone emergency service for autistic people in Germany and that is a voluntary one. And that's also relatively small given such a high suicide prevalence" (healthcare provider; T3). Counselling services for different purposes were mentioned. Healthcare providers described the need especially for social therapeutic services in particular: "because often it's not about psychotherapy or processing the experiences from the past, but supporting, in order to somehow cope with the daily challenges, for Asperger's, […] with the social challenges" (healthcare provider; B2). The need for peer counseling and information about existing services was also emphasized. Lastly, the lack of services for those who are not able to get manifest therapy or are waiting for it was expressed:

I think a larger network of counselling centers would be helpful, because I think that many affected people, many relatives experience greater barriers finding information at a clinic, a therapy center, than going to a counselling center or to a regulars' table and seeking advice and exchange there. So, I think that would be a practical way to facilitate initial contact. (Autistic adult; ibew5)

Furthermore, participants across all groups highlighted the lack of *(vi) assistance services* for autistic adults without ID "because autism doesn't mean therapy will be done and then everything will be okay again" (relative; T6). It was emphasized that case management (i.e. services to provide adequate healthcare for autistic adults according to their individual needs) needs to be implemented to relieve the burden on autistic adults, but also on relatives who often provide care and assistance. An autistic adult responded to the question how relatives and partners could be supported: "others who provide required assistance. In other words, assistance for autistic people across the whole lifespan. In my opinion, even a high-functioning autistic person like me needs some kind of lifelong assistance service" (autistic adult; ibew 11).

Limited access to services

Not only the lack of services was described, but also barriers in accessing the available services were highlighted in most interviews and focus groups. Two subthemes were identified in the data, which are loading on the professional and structural level: (i) lack of transparency about healthcare services, (ii) funding.

On the one hand, the *(i) lack of transparency about healthcare services* was described. Neither autistic adults nor relatives or healthcare providers were fully informed about available services. A healthcare provider described: "That's the problem, caregivers don't have an overview of available services, including myself. It's not easy to find one's way in this network, but that's actually the most important thing" (healthcare provider; B1). For example, patients often do not receive adequate care after diagnosis and have to obtain information on their own or with the support of relatives. Those who found services reported to have problems with the complex application process that could not be managed without external support. Healthcare providers reported regularly assisting autistic people with application forms, even though they are not responsible to provide assistance services. Furthermore, autistic adults reported that, based on the division of healthcare services among different social systems, navigating the healthcare system would be difficult:

Access to services is so massively divided among the various social systems by law that it is incredibly difficult for me to even find out who is responsible for me and from whom I have to apply for what. Yes, the fact that the individual social systems are always trying to pass the buck to each other is also not conducive. (Autistic adult; ibew4)

In relation to this, it was criticized that *(ii) funding* of autism therapy is not provided by health insurance. An autistic adult shared her opinion on this topic:

Because at the moment autism therapy and psychotherapy are separated from each other. And a classic autism therapy is also not funded by the health insurance, because with health insurance financing, this healing aspect is always included, which is a wrong approach, in my opinion. (Autistic adult; ibew2)

Regular psychotherapy, on the other hand, is not funded by health insurance when only ASD is diagnosed. To be approved for psychotherapy, a mental disorder such as depression or anxiety disorder must be present. A healthcare provider mentioned "almost three-quarters have a relevant co-morbidity, so the largest group finds access" (healthcare provider; T5) to psychotherapeutic services, but it would be a problem anyway. In addition, it was criticized that statutory health insurance companies do not enable rapid support in case of crisis situations.

Improvement of stakeholder collaboration

This theme includes the collaboration of relevant stakeholders in the healthcare of autistic adults on a professional and structural level, which were mentioned to be healthcare providers from various professions. Primarily healthcare providers, but also few autistic adults emphasized the need for collaboration between different groups of professions and "multi-professional teams work[ing] with affected people" (autistic adult, ibew7). Many participants described that this could allow for simplification of healthcare processes, as all providers can be on the same level of knowledge about the patient - as long as the autistic adult consents. Some participants mentioned that collaboration would also facilitate the transition between pediatric and adult healthcare providers:

That the new doctor can also call the old doctor if there are any questions until the autistic person is really back at home with the new one, where he or she is again in safe hands during this transition phase. (Autistic adult; ibew11)

Few healthcare providers also highlighted that exchange with other providers also contributes to increase knowledge about autism: "there should actually be more networking and more exchange of experiences" (healthcare provider; B2).

Discussion

To our knowledge, this is the first comprehensive investigation including the perspectives of autistic adults, relatives and healthcare providers on barriers and needs at the individual, professional and structural levels of mental healthcare for autistic adults without ID. We identified six major barriers to mental healthcare for autistic adults in Germany, which primarily affect autistic adults but are also of concern for relatives and healthcare providers: (i) lack of knowledge about autism, (ii) a need for increased participation/involvement, (iii) consideration of autism-specific needs in treatment, (iv) lack of services, (v) limited access to services, and (vi) improvement of stakeholder collaboration. The majority of the themes are in line with existing literature, focusing on general healthcare in the U.S [26]., thus, previously reported barriers similarly apply to mental healthcare.

The results of this study partially replicated the three previously proposed levels of barriers (see Fig. 1). In addition to these three levels, we included the stakeholder group that is mainly affected by the existing barriers with the goal to give an overview about potential adjustments. Some themes were not presented in previous models, therefore we tried to integrate them based on existing literature and the qualitative study results. In line with previous research, "lack of knowledge about autism" and "consideration of autism-specific needs in treatment" were present on all levels [26]. These barriers have implications for all requested stakeholder groups. "Lack of services" and "limited access to services" were previously defined as barriers on the structural level. Based on our data, the professional level also seems to be important. Over the last years, participation and empowerment became more important in the area of mental healthcare including autism [37, 38] and was not considered as barrier in mental healthcare so far. Based on our data, the theme "a need for increased participation/involvement" was interpreted as the relation between autistic individuals including their relatives/partners (individual level) and healthcare providers (professional level) and has implications for all stakeholder groups. Likewise, "stakeholder collaboration" was not considered as specific barrier [26]. Previous research on interprofessional collaboration in the treatment of autistic individuals showed that cross-disciplinary collaboration can lead to improved patient care and maximal outcomes by capitalizing on the varying expertise, as professionals from a broad range of disciplines are needed to address the heterogeneous core symptoms and co-morbidities [49]. This has mainly implications for the healthcare providers themselves.

The identified barriers and stakeholder groups may apply for both national and international mental healthcare of autistic adults, since ubiquitous barriers seem to exist for adults with autism across countries and healthcare systems. This is supported by the fact that our findings are in line with previous evidence from various countries, e.g. United States [26, 30, 50], Canada [29], and United Kingdom [15, 51]. A common feature of all countries is the high level of healthcare expenditures by international standards, ranging from 17.7% of the GDP in die United States to 10% in the United Kingdom [32, 52, 53]. Thus, barriers to healthcare are stable across these countries, despite comparatively large amounts of funding being invested into healthcare. Therefore, the results of the current study could be of global appeal and point to specific areas for improvement, which could guide future development of needs- and evidence-based services and guidelines of mental healthcare for people with autism across the lifespan.

Policy-makers need to increase the availability of formal healthcare services and, in turn reduce healthcare inequalities by providing the necessary resources. For example, diagnostic and psychotherapeutic services need to be expanded in both urban and rural areas. Transitional periods (e.g., transitioning from adolescence to adulthood) need improved coordination. In general, healthcare facilities and processes should be more accessible for autistic adults, e.g. communication pathways. Most important, transfer of knowledge about autism needs to take place in the community to reduce stigma of autistic individuals, to train healthcare providers and inform about skills and behaviors necessary to provide respectful and adequate mental healthcare for autistic adults. In light of the massive burden autism represents for autistic individuals, relatives and society, research on ASD (e.g., symptomatology of autistic girls and women, screening and diagnostics of autistic adults, burden and support of relatives and partners) and its funding needs improvement. Education, research, and treatment should be addressed through the participation and collaboration of relevant stakeholders. In autistic individuals, symptom expression and symptom severity vary both inter- and intrapersonally, so mental healthcare has to be flexible and tailored to the state of the condition. Findings indicate that individualized and personalized healthcare would provide best practice for autistic individuals [34, 54].

Limitations and future directions

A major strength of the current study is the participatory approach, which ensured involvement of autistic peer researchers and relatives as well as service providers at all stages. Participation, which we had identified as important theme, in the current study was further maximized by offering several methodological participation modes to autistic adults (i.e., by e-mail or chat). The use of a purposeful, maximum variation sampling strategy allowed to obtain rich, comprehensive data from a wide range of participants. However, as with most qualitative studies, the sample does not represent a random selected sample of population and the sample size is limited. Moreover, it was not possible to recruit participants from some federal states (e.g. Brandenburg, Saarland; see Table S1). In addition, generalization of results across the entire autism spectrum is limited due to the lack of participants with ID, which may have reported different support needs. Nonetheless, their perspective was brought into focus groups by professionals, who also worked with autistic adults with higher levels of support needs. Furthermore, due to the COVID-19 pandemic, interviews and focus groups were conducted via video communication. Hence, it is possible that there might be a loss of subtle information that would have been presented in personal face-to-face settings. Lastly, stakeholder groups were interviewed using different settings (one-by-one interviews with autistic adults and focus groups with relatives and healthcare providers). Even though data collection and data analysis were carried out in a standardized manner independent of the setting, it cannot be entirely excluded that results were affected by this.

As manifestation of symptoms, severity, and secondary impairments vary widely between autistic individuals, further research should focus on the development of an individualized, needs- and evidence-based, severity-adjusted healthcare model. To allow an appropriate scaling of this services reliable epidemiological data from Germany will be needed [33]. To this end, within the scope of the *BarrierfreeASD* project, a follow-up large-scale online survey will be conducted to collect quantitative data in larger samples and provide a representative overview of the healthcare situation in Germany [36]. This mixed-methods approach aims to obtain a complete set of information to develop specific recommendations for a future healthcare model of autistic adults. Ultimate aim will be to develop recommendations for reforms and changes at all levels to improve mental healthcare of autistic adults in order to enhance independence and quality of life, for both the autistic adults and their relatives.

Abbreviations

ASD	Autism Spectrum Disorder
ADHD	Attention Deficit Hyperactivity Disorder
COREQ	Consolidated criteria for reporting qualitative research
ID	Intellectual Disability
GDP	Gross Domestic Product
N	Sample size
SD	Standard deviation
WHO	World Health Organization

Supplementary Information

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Supplementary Material 1 Table S1 Sample distribution by region and federal state

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Authors' contributions

Holger Schulz is the main PI of the study; Judith Peth, Nicole David, Daniel Schöttle, Alexander Konnopka, and Kai Vogeley are co-PIs. They all conceptualized and designed the study. Petia Gewohn, Hannah König, Pascal Rahlff, Frank-Erik, and Sophia Dückert are research staff. Pascal Rahlff and Frank-Erik are also peer researchers. Petia Gewohn and Sophia Dückert performed data collection and data analysis. Sophia Dückert mainly drafted this manuscript. All authors participated, reviewed, edited and approved the manuscript.

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Data availability

The datasets used and analyzed in the current study are available from the corresponding author on reasonable request.

Declarations

Competing interests

DS received honoraria for lectures from or has been an advisor to Janssen GmbH, ROVI, Lundbeck GmbH, Otsuka Pharma GmbH and Takeda. This has no influence on the present research project. The remaining authors declare that there is no conflict of interest.

Ethics approval and consent to participate

The study has been approved of by the Local Psychological Ethics Commission at the Center for Psychosocial Medicine of the University Medical Center Hamburg-Eppendorf (#LPEK-0227; Dec. 2020) and was carried out in accordance with the WMA Declaration of Helsinki. Study participation was voluntary and there were no foreseeable risks for participants due to participation. Participants were fully informed about the aims of the study, data collection, handling and storage. Written informed consent was obtained before participation. Principles of data sensitivity, data protection and confidentiality requirements were observed.

Consent for publication

Not applicable.

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Affidavit

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