

Diagnosis is based exclusively on information of behavioural features, at the moment, which must come from various sources, ages and situations. There exist sophisticated standardised diagnostic and screening tools (e.g. Le Couteur et al., 2003; Lord et al., 2003), by the aid of which diagnosis is more reliable and – especially valuably for research – data from various research sites becoming more comparable. Possible reasons for the increasing prevalence of autism are the rise to autism. Data from various methodologies make the conclusion that autism comes from dominantly genetic causes quite a firm one. It is also evident that in some cases (foenotypic) exogenous factors play some role, too. Heterogeneous etiological factors seem to lead to a complex neurological impairment – so autism is most probably not a case of a single focal neural injury. One of the key open issues about autism is the precise causal-temporal pattern of neurodevelopmental anomalies, and their relation to symptoms (for a review, see Vollenmar et al., 2004).

Until the etiological background becomes clear, biological treatments may play just a minor role in the treatment of autism. At the moment, the most efficient way of treatment is an early, complex, and individualised educational and educational-developmental approach, which comes in various forms (Iowdin, 2002).

2. COGNITIVE/PSYCHOLOGICAL EXPLANATIONS OF AUTISM

The second chapter of the dissertation is a discussion of the cognitive background of autism, with a special focus set onto the naive theory of mind impairment and its possible early manifestations, serving as a specific starting point for my empirical studies. In the present summary I shall focus on the aspects of the issue which have a direct relevance to the empirical work, so executive and central coherence impairments are ignored here (but not in the dissertation).

Current cognitive explanations of autism

By the beginning of the 1990's, three cognitive hypotheses have been delineated in order to explain the symptoms of autism, and have been supported by empirical data: the *naive theory of mind hypothesis* (Baron-Cohen, Leslie, 1985), the *executive dysfunctioning hypothesis* (Ozonoff, et al., 1991), and the *weak central coherence hypothesis* (Frith, 1989). Though originally these hypotheses were interpreted as rival ones, in the more recent years findings show the cognitive background of autism more complex than once believed, and this calls for a re-evaluation of the once dominating single-factor model of the cognitive basis of autism.

The naive theory of mind hypothesis

It's an essential basis of our social competence that we are able to attribute mental states to others, and to explain, interpret and predict behaviour as causal consequence of these mental states. The mechanism underlying this ability seems to have an innate

basis. Following Premack & Woodruff (1978) we call this ability "naive theory of mind". Leslie (1987) argued that pretend play is an early behavioural manifestation of this ability. Retrospectively, therefore, it may seem quite plausible to attempt to explain autism in terms of a developmental impairment in theory of mind – as social and communicative impairments dominate the symptomatology, and impoverished and compromised pretend play is observed in autistic children.

This theory has been tested and supported in hundreds of studies applying various experimental and other techniques (for a review, see Gyi'et, 2004). These results have had an important impact on practice, but they have raised new questions as well.

(1) Does the impairment in naive theory of mind explain the entire behavioural pattern of autism?

The naive theory of mind impairment hypothesis seems to give a quite efficient explanation of the symptoms in the fields of reciprocal socialisation and reciprocal communication. Baron-Cohen and his colleagues (1985), first formulating this hypothesis, also pointed out that autistic children show a specific impairment in behavioural rigidity and stereotypic repetitive tendencies as well. The few studies aimed at evaluating this specific causal hypothesis, however, has brought quite ambiguous results, mostly failing to find a significant relationship between the depth of theory of mind impairment and severity of stereotypic and repetitive symptoms (Lunner, 1997; Frith & Illappé, 1994).

(2) Is theory of mind impairment universal in autism?

It is an open issue if all individuals with autism spectrum disorders show an impairment in theory of mind. It is possible that there are individuals with the so-called *high-functioning autism* (e.g., Baron-Cohen, Gyi'et, 1994; Gillberg, 2004) in which the fact that there exist individuals with autism who pass – sometimes even quite complex – theory of mind tasks (see Dahlgren & Trillingsgaard, 1996; Gyi'et, 2004). It is also possible, however, that theory of mind is non-impaired in a subgroup of individuals with autism spectrum disorders.

(3) Is impairment in naive theory of mind a specific characteristic of autism?

The answer to this question is a decisive no. Now it is well-documented that there is theory of mind impairment in schizophrenia (e.g., Corcoran et al., 1993); Williams syndrome (e.g., Yirmiya et al., 1998). Although these findings clearly suggest that the impairment in naive theory of mind as a single causal factor is insufficient to explain autism specifically, it is also true that this impairment arises more dramatically in the population with autism, suggesting a kind of "relative specificity".

(4) Is theory of mind impairment the primary deficit in autism?

Focusing first on the sceptical hypotheses, suggesting that it is not the theory of mind impairment which is the primary psychosocial deficit in autism, one may conclude that the 'no alternative hypothesis' has been supported by strong empirical data so far. On this basis, it is likely that the 'no alternative hypothesis' is the correct one. It appears that it is only joint attention behaviours that show significant correlation to naive theory of mind (Charman et al., 2000). The causal background of this relationship, however, has not yet been clarified. So various causal patterns are still empirically possible. Although pretend play is intuitively a strong candidate for being an early manifestation of naive theory of mind, no strong empirical data support this specific connection. Imitation, in turn, shows a relationship to language development but not to theory of mind development. Though some authors take predictive relationships as strong evidence for a causal link between assumed precursors and naive theory of mind (e.g., Charman et al., 2000), I believe that decisive conclusions should be only drawn on the basis of direct relationships.

Summary

There is little doubt today that in order to explain autism, one has to take into account naive theory of mind impairment, executive dysfunctioning, and weak central coherence as well. It is highly implausible today that one single cognitive deficit could explain all the behavioural manifestations of the syndrome. It is possible that various subgroups of the autism spectrum are to be explained by various patterns of cognitive impairments, but no such subgroups have been convincingly identified so far. The causal background between these three core cognitive impairments is a central issue in the current research. It is not clear whether they support any reduction either within them, or to a fourth, non-yet-known cognitive factor.

3. AUTISM AT VARIOUS AGES – THE ISSUE OF PROGNOSIS

The first part of the *third chapter* shortly summarises the symptoms of autism at various ages. The second, more detailed part of the chapter I review the direct precedents of my studies in the literature – that is, those studies which were aimed at developmental changes and their prognostic factors in autism, with an eye on follow-up methodology.

Autism at various ages – a cross-sectional view

The behavioural patterns specific to autism change a lot with development. While in pre-school years impairments in reciprocal social interactions and reciprocal communication dominate the symptoms, later significant transitions take place, and in adulthood obsessive and ritual tendencies may become more dominant, additional

medical problems may arise, and also the individual may become aware of his/her atypical personality, that leading to further difficulties.

Attempts to predict developmental changes and their outcome on the basis of cross-sectional data have brought a quite limited success. Beyond neural maturation, intellectual abilities and language-use have proven to have prognostic power.

Prognostic factors

Focusing on long-term follow-up studies we find that the relatively consistent variables that predict adulthood prognosis, general outcome, and adaptive behaviours are early non-verbal IQ and the level of receptive language. These factors, however, allow only rough estimations:

(1) Early IQ below 70 predicts worse outcome, but early IQ above 70 does not necessarily bring about good outcome in adulthood (Rutter, 1970; Lotter, 1974; Gillberg & Steffenberg, 1987; Howlin et al., 2004).

(2) level of early receptive language predicts later level of linguistic skills (Mawhood et al., 2000);

(3) early linguistic skills (functional speech arising before age 5 and the level of receptive language) predict later overall outcome (Rutter, 1970; Lotter, 1974; Gillberg & Steffenberg, 1987; Mawhood et al., 2000); at the same time, however, adults with Asperger syndrome with no childhood language delay show no advantage in overall outcome above subject with autism showing early delay in language development (Howlin, 2003).

It is important to note that in most studies on the predictive relationship between childhood assessment results and overall adult outcome there were no other measures available beyond non-verbal IQ and receptive vocabulary. The relatively strong relationship between these and adult outcome is of course important, but one may expect that more detailed data on early functioning may serve with more refined prognostic relationships.

More recent and more short-term clinical follow-up studies (Freeman et al., 1999; Szatmari et al., 2003; Szatmari et al., 2004; Starr et al., 2003) early assessments was applied to predict later outcome using a methodology and methodology by some standardised measures. Beyond focusing the above-mentioned predictive factors, some further relationships have been revealed:

(1) school-age communication skills are best predicted by early non-verbal IQ and early linguistic skills, while daily self-supporting skills are best predicted by full IQ (Freeman et al., 1999; Szatmari et al., 2003; Szatmari et al., 2004; Starr et al., 2003);

(2) school-age adaptive skills show no relationship to early IQ scores (Freeman et al., 1999), but predicted by linguistic skills and non-verbal IQ (Szatmari et al., 2003; Szatmari et al., 2000; Starr et al., 2003);

(3) none of the early-measured factors proved predictive of changes in overall patterns of symptoms (Szatmari et al., 2003; Szatmari et al., 2006; Starr et al., 2003).

Clinically motivated follow-up studies so demonstrated that early assessment results show a significant relationships to later outcome. One must note, however, that our predictive-prognostic potential has increased just slightly since the first follow-up studies were published (Rutter, 1976; Lotter, 1974). It is still unclear what predicts – if anything – developmental changes in the fields of the autism triad, as well as the equally important expressive and receptive language skills.

It seems quite plausible to turn to findings from theoretically motivated follow-up and longitudinal studies, especially because early theory of mind related behaviours may play a future role in predicting later clinical picture, especially on social and communicative reciprocity.

(1) level of linguistic skills appears to be predicted by early joint attention behaviours (Mundy et al., 1990; Rosenthal Rollins & Snow, 1998; Sigman & Ruskin, 1999; Charman et al., 2003; Charman, 2003), and imitation (Charman et al., 2000 2003; Charman, 2003; Stone & Yoder, 2001);

(2) the relationship between linguistic measures and quality of early play activity is still not clear (Sigman & Ruskin., 1999; Charman et al., 2000; 2003; Charman, 2003);

(3) at least two missing from among imitation, pretend-declarative positioning and pretend play at the age of 18 months predict a diagnosis of autism spectrum disorder beyond 3 years of age (Baron-Cohen et al., 1992; Baron-Cohen et al., 1996);

(4) imitation and declarative joint attention at the age of 20 months predict milder symptoms for the age of 42 months (Charman et al., 2003, Charman, 2003);

(5) performance on tests of naive theory of mind seems to be predicted by early receptive and expressive linguistic skills and early joint attention behaviours (Stel et al., 2003; Charman et al., 2006).

Summarising the somewhat confusing results of these studies, it is striking that only a few of them was aimed at testing the relationship between early naive theory of mind abilities, and their precursors, and clinically crucial later adaptive skills and symptomatology. Several aspects of this relationship have not been investigated, and on important aspects – like the relationship between joint attention and naive theory of mind – there are findings only from one study (in this case, Charman et al., 2000).

4. INVESTIGATING THE ROLE OF EARLY NAIVE THEORY OF MIND RELATED BEHAVIOURS IN DETERMINING THE SCHOOL-AGE PROGNOSIS IN CHILDREN WITH AUTISM

In the *North Ayrshire* I present my follow-up studies aimed at identifying further factors that may predict communicative and social skills, and the severity of symptoms. It may be possible to identify further factors that may predict the severity of symptoms, development, and to plan intervention more accurately. Thus, our starting point is clinical, but our methodology is theoretically based.

Hypotheses

- (1) It is generally assumed that there is a significant relationship between the results of early measurements and later performance.
- (2) Early IQ level predicts later IQ level.
- (3) Early non-verbal IQ predicts later level of behavioural adaptation.
- (4) Early level of receptive language predicts later level of linguistic skills, adaptive skills, and performance on tests of naive theory of mind.
- (5) Better performance on attention behaviours at the time of early assessment predicts better linguistic skills.
- (6) Presence of naive theory of mind skills at the time of early assessment predicts better performance in later testing of naive theory of mind.
- (7) Early-present naive theory of mind related behaviours predicts milder symptoms and better adaptive levels in the fields of social reciprocity and communicative reciprocity.

Study 1: naive theory of mind related early behaviours in autism

Synoptic hypothesis in Study 1

- (1) children with autism underperform controls in joint attention behaviour and pretend play;
- (2) there's no difference between the two groups in mirror self-recognition;
- (3) children with autism show less "self-admiring" behaviours in front of mirror than controls do;
- (4) pretend play shows significant relationship to joint attention behaviours in both groups;
- (5) "self-admiring" behaviours show significant relationship to theory of mind related behaviours (pretend play and joint attention behaviours) in both groups.

Method

Subjects

22 children with autism (20 boys, 2 girls), selected from a pool of 202 children of the adequate age.

	means	range
age	67.45 months	36–86 months
non-verbal IQ	91.86	62–122
non-verbal mental age	61.68 months	37–90 months

Table 4. 1. A few summary data of the total autism group from Study 1.

Control subjects

15 control subjects – children (10 girls and 5 boys); 13 with typical development, 2 showing IQ scores in the mildly impaired range – selected from a pool of 50 children, on the basis of best fit to the subjects with autism.

Matching

The pairwise matching was done on the basis of biological age (± 4 months) and non-verbal IQ (± 5 points).

Procedure

Both pre-testing (40-60 min.) and data-gathering (30-60 min.) took place in individual sessions. Target behaviours were video-recorded and coded by an independent rater blind to diagnosis. See Table 4.2. For details of procedure.

task	reference	goal
<i>Letter non-verbal performance code</i>	Letter, 1979	non-verbal IQ (also for matching)
<i>Picture PVT</i>	Dunn, 1959 Orlitzky, 1974	receptive language (vocabulary)
<i>Observing spontaneous play</i>	Hobson et al., 1996; Chapman & Baron-Cohen, 1997	identifying forms of spontaneous play
<i>Elicited functional play</i>	Hobson et al., 1996; Chapman & Baron-Cohen, 1997	assessing ability of functional play if prompted
<i>Elicited pretend play</i>	Hobson et al., 1996; Chapman & Baron-Cohen, 1997	assessing ability of pretend play (object substitution) if prompted
<i>Spontaneous joint attention</i>	-	amount of spontaneous initiation of joint attention
<i>Elicited joint attention</i>	on the basis of Baron-Cohen et al., 1992	assessing co-ordination of gaze and joint attention behaviours if prompted
<i>Mirror self recognition</i>	Gallup, 1976; Amsterdam, 1972; on the basis of Baron-Cohen & Golan, 1978	manifestations of mirror self-recognition
<i>Behaviours in form of mirror</i>	on the basis of Amsterdam, 1972	identifying patterns of behaviours in form of mirror

Table 4.2. Tools from Study 1

Statistical analysis

Robust tests (Kendall's tau, chi-square test, Pearson's χ^2 , t-test) were used in statistical analysis. In group comparisons only the matched sub-groups were involved. In within-group analyses the entire samples were involved.

Most important findings from Study 1

Group comparisons – spontaneous and elicited play

There was not significant difference between the two groups in spontaneous play activity in its any form, and in elicited functional play. There was a significant difference in elicited pretend play ($\chi^2 = 14.221$; $p = 0.001$).

Group comparisons – spontaneous and elicited joint attention

Spontaneous joint attention arose very rarely in both groups, yielding no significant difference between groups. In elicited joint attention, however, the two groups showed significant differences in various aspects, including coordination with gaze, the aggregated measure of elicited joint attention being also significantly higher in the control group ($\chi^2 = 20$; $p < 0.001$).

Group comparisons – behaviours in form of mirror

None of the behavioural patterns shown in this situation distinguishes the two groups.

Group comparisons – complex, aggregated variables

The aggregate variable composed of measures of naive theory of mind related behaviours (spontaneous and elicited pretend play and joint attention) indicates that controls outperformed subjects with autism ($\chi^2 = 5.879$; $p < 0.043$). There was not such difference along an aggregated variable composed of functional and pretend play measures.

Within-group relationships between variables

There were significant correlations found between receptive vocabulary and mental age in both groups ($r = 0.751$; $p < 0.01$), and between spontaneous pretend play and chronological age in the autism group ($r = 0.491$; $p = 0.02$). No further important significant correlations were found.

Brief summary and discussion

- (1) As a methodological conclusion it is important that observing spontaneous behaviours, a longer warm-up phase is necessary.
- (2) The fact that I found no significant difference between the two groups in spontaneous pretend play but the autism group underperformed controls in elicited play raises the possibility that children with autism manifested previously learnt play

pattern in the spontaneous situation. This implies that the elicited play situation may better mirror their competence to play pretend play.

(3) My findings raise the question to what extent the spontaneous and elicited play situations involve the same competences, and whether there is a difference between typically developing children and children with autism in this respect.

(4) My findings suggest that the social skills of children with autism are best distinguished by the relative lack of gaze monitoring during joint attention behaviours.

(5) My findings confirm the hypothesis that children with autism are not impaired in mirror self-recognition, and patterns of spontaneous behaviours shown in front of a mirror do not distinguish them from typically developing / non-autistic controls.

Study II: follow-up study on the development of social and communicative competences

The aim of my second study was to reassess children with autism who took part in Study I, in order to reveal predictive relations between earlier measures and later adaptive social and communicative behaviours, symptomatology, language level, and naive theory of mind performance. I formed no particular hypotheses for Study II, as the goal was more descriptive, and not hypothesis-driven. The dissertation offers a detailed analysis of relationships between early and later measures as well as relationships between variables in Study II – here, however, I present only a summary of predictive relationships between results from Study I and Study II.

Method

Subjects

16 children with autism (15 boys and 1 girl) from the original sample of 21 took part in Study II.

Procedure

The three sessions of Study II took place 40-55 months after the sessions of Study I. Tools applied in Study II are shown below in Table 4.3.

tools	references	goals
Merge-17 Adaptive Behavioural Scales (17-ABS)	Spitzer, 1993 Sattelmaher, Volkmar & Cohen, 1994	Q: to assess general adaptive behaviour, especially on the social and communicative domains
TROGF II	Kauf, Beitchman, Young, & Bell, 1999 Rombay, 1998 (translated by: A. Laksic)	Q: to assess general linguistic development by assessing naive theory of mind performance
verbal first order false belief non-verbal false belief joint attention task	Brown-Gibbs et al., 1983; on the basis of Golan et al., 2007 Golan et al., 2007 on the basis of Golan et al., 2007	Q: to assess naive theory of mind performance
theory of mind theory of mind	on the basis of Baron-Cohen, 1989 on the basis of Golan, 2014	Q: to assess naive theory of mind performance on more advanced level

Table 4.3. Tools used in the follow-up study (Study II)

Most important results from contrasting early and late measurements

Factors predicting severity of school-age IQ
School-age MAWG-FHR IQ showed the strongest relationship to early non-verbal IQ (by Leiter) ($r=0.604$, $p<0.005$).

Factors predicting school-age level of adaptive behaviours

There was no strong relationship between this variable and any of the early measures.

Factors predicting school-age level of language skills

It was only the early test of receptive vocabulary that showed a – actually weak – relationship to later grammatical development (as measured by TROG); ($r=0.496$; $p=0.072$).

Factors predicting severity of social and communicative symptoms of school-age I
The early appearance of spontaneous functional play ($r=-0.471$, $p=0.05$). Factors emphasised in the literature – joint attention behaviours, pretend play and receptive vocabulary – showed no significant relationships to ADOS scores.

Factors predicting school-age performance in naive theory of mind tasks

Neither verbal nor non-verbal false belief attribution test measures showed significant correlations with early naive theory of mind related measures or receptive vocabulary. There was a correlation between later understanding of simple irony and early elicited joint attention ($r=0.575$, $p=0.015$), as well as between later understanding of simple irony and early co-ordination in joint attention ($r=0.685$, $p=0.008$). Later understanding of simple irony showed correlation with early receptive vocabulary ($r=0.631$; $p=0.021$).

Brief summary and discussion

(1) In line with other findings, our data indicate the stability of IQ scores in autism; school-age IQ is predicted by early non-verbal IQ.

(2) Early receptive vocabulary does not show particular relationship to later measures of language skills – earlier findings are also ambiguous in this respect. Development in this respect is likely to be more strongly related to early joint attention. Predictors of linguistic measures to later language level – at least in the case of a long-term follow-up of a relatively homogeneous sample like this.

(3) I found no significant relationship between early joint attention behaviours and later linguistic measures. It is possible – as suggested by Charman and his colleagues (2000) – that this is so because naive theory of mind, joint attention related to it, and

language show a joint development only in an early phase of development, and later their trajectories gradually become separate. As the time-range of follow-up was longer than in most studies, the early association among these skills may have faded by separation of developmental pathways.

(4) The tendency-level correlation between early spontaneous pretend play and school-age daily adaptive behaviours may suggest that those children who are capable of observational-imitative learning in early childhood manifested in play situations may show better performance in simple, routine-based daily social situations.

(5) In line with the results of Sigman et al (1999) I found that the frequency of early spontaneous functional play showed a tendency-level correlation with the later ADOS score in the social domain. This may indicate, on the one hand, that those children with autism who are able to manipulate objects according to their functions have a bigger chance to be involved in situations where social learning and practicing social skills are crucial. On the other hand, the higher level of spontaneous functional play in early functional play is just an indicator of less impaired social reciprocity, without a direct causal link between early play skills and later social symptoms.

(6) In line with findings of Charman and his colleagues (2000) I found that early joint attention skills predict later complex understanding of mental states, as measured by simple irony task. This finding confirms the key role of joint attention behaviours in the developing understanding of mental states.

(7) I found a tendency-level correlation between early receptive vocabulary and later performance in the irony tasks. This is in line with the widespread view that language skills are crucial for the development of irony tasks. It is, however, interesting to note that not in contradiction with the previous conclusion – that a relatively high level of early language opens a compensatory pathway for a non-typical way of solving tasks that involve mental states.