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A follow-up study of 45 patients with elective mutism

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Abstract Forty five patients (23 boys and 22 girls) with elective mutism (8.7 ± 3.6 years old), who were referred to a university department and a child guidance clinic within a 15-year-period, were followed up on average 12 years later. For 41 of them, sufficient information could be obtained at follow-up, and 31 patients could be investigated personally.

At follow-up, an interview and a standardized psychopathological examination were carried out as well as two standardized biographic inventories. The main results were: 1) a high load of individual and family psychopathology was characteristic of the patients. The disorder started already at age 3 to 4 and referral age was 8 years on average. 2) In 16 out of 41 patients (39 %), a complete remission could be observed. All other patients still revealed some communication problems. 3) The formerly mute patients described themselves as less independent, less motivated with regard to school achievement, less self-confident and less mature and healthy in comparison to a normal reference group. 4) A poor outcome could be best predicted by the variable "mutism within the core family"at the time of referral.

Key words elective mutism · follow-up · family psychopathology · prediction.

Introduction and review of the literature

Elective mutism is a rare, but in most cases severe disorder of communication. The condition was first described by Kussmaul in 1877, who called it "aphasia voluntaria", and in 1934 Moritz Tramer used the term "elective mutism"for children who spoke only with a small group of people mostly coming from the children's home and who did not talk to other people.

According to DSM-IV, the essential feature of this condition is a "the persistent failure to speak in specific social situations (e. g., school, with playmates) where speaking is expected, despite speaking in other situations"(APA 1994, p. 114). Mutistic behavior can also occur as a symptom of other conditions (e. g., schizophrenia, major depression, social phobia), but in these cases the diagnosis "elective mutism"is not appropriate. The guidelines of ICD–10 require three criteria for the diagnosis of elective mutism (WHO 1992, p. 278):

- "a normal, or near normal, level of language comprehension
- a level of competence in language expression that is sufficient for social communication
- demonstrable evidence that the individual can and does speak normally or almost normally in some situations".

Epidemiology

The disorder can be found in clinical populations in about 0.2–1 % of all referred cases (Lorand 1960; Morris 1953; Reed 1963). Fundudis, Kolvin and Garside (1979) reported a prevalence of 0.8 per 1,000 children based on an epidemiological study of a total city cohort of 3,300 7-year-olds. In a questionnaire survey including all primary schools of Birmingham, the prevalence of "non-speaking at school"was 7.2 per 1,000 children. However, some five months later, 20 % of the previously non-speaking children spoke to their teachers and 40 % to other children (Brown & Lloyd 1975). Over 90 % were speaking by the 12 month follow-up. Thus, the prevalence figures of Brown & Lloyd (1975) seem to be too high and are not typical for elective mutism which is a fairly persistent disorder (Kolvin & Fundudis 1981). Recent epidemiological studies found a prevalence of 2 %

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among second graders in elementary school in Finland (Kumpulainen & Rasanen 1998) and a prevalence of 1.8 % in a school population of seven- to 15-year-olds in Sweden (Kopp & Gillberg 1997). Neither DSM-III-R nor ICD–10 do specify the duration of the condition to make the diagnosis. But this is necessary in order to avoid the inclusion of transient mutistic reactions as a symptom frequently found in children entering school (Brown & Lloyd 1975). For this reason, some authors have proposed a duration of the symptomatology of at least six months (Kolvin & Fundudis 1981; Wilkins 1985; Kaplan 1992). The DSM-IV criteria require a duration of the disturbance of at least one month.

The average *age of referral* and of the first diagnosis of the condition has been found to be between 6 to 8 years (Hayden 1980; Kolvin & Fundudis 1981; Wergeland 1979; Wright 1968). Most studies have found a preponderance of the female sex, the sex ratio of girls to boys being approximately 2:1 (Hayden 1980; Wergeland 1979; Wilkins 1985). But there are also results in the direction of an equal sex distribution (Parker et al. 1960; Andersson & Thomsen 1998; Kopp & Gillberg 1997; Kumpulainen & Rasanen 1998).

Psychopathology and comorbidity

Most authors have reported the following features of electively mute children: extreme shyness and problems of social contact, oppositional negative behavior, anxiety states, especially in public, and negativistic behavior (Hayden 1980; Kolvin & Fundudis 1981; Wergeland 1979; Wilkins 1985; Steinhausen & Juzi 1996). Several studies have found delayed speech and difficulties in articulation (Kolvin & Fundudis 1981; Wilkins 1985; Wright 1968; Andersson & Thomsen 1998; Kristensen 2000). Also a relatively high frequency of enuresis and/or encopresis has been reported (Kolvin & Fundudis 1981; Wergeland 1979; Rösler 1981; Andersson & Thomsen 1998; Kristensen 2000).

Other authors found controlling (Hayden 1980; Wilkins 1985) and obsessive-compulsive behaviors (Hayden 1980; Wergeland 1979). School refusal was observed by Elson, Pearson, Jones and Schumacher (1965) and Hayden (1980).

Recent studies found a high association of elective mutism with social phobia (Dummit & Klein 1997), anxiety disorders in up to 74 % (Kristensen 2000) and internalizing problems (Kristensen 2001).

Etiology

As far as etiology is concerned, several factors have been described that contribute to the final manifestation of elective mutism. These factors can be described as follows:

Delay of language development

A metaanalysis (Poller 1989) of seven studies (Funke et al. 1978; Kolvin & Fundudis 1981; Kurth & Schweigert 1972; Popella 1960; Rösler 1981; Wergeland 1979; Wright 1968) has demonstrated that a delay of language development can be found in nearly half of the patients (33 out of 70 patients, 47 %); speech and language disturbances, other than developmental delays, could be found in 43 out of 113 described patients (38 %). So either a delayed development of language or language and speech disturbances may be factors that contribute to the manifestation of the disorder. Recent studies confirmed the result of this metaanalysis (Andersson & Thomsen 1998; Kristensen 2000). Andersson and Thomsen found a rate of developmental language disorder of nearly 50 % in their sample.

Communication deficits and psychiatric disorders in the family

There seems to be also a high load of communication deficits and psychopathological disorders within the families of elective mute children. Some studies have found that mutism is present in other family members in approximately 50 % of the cases (Lorand 1960; Popella 1960). Taciturnity of father or mother could also be found in several studies up to 50 % (Funke et al. 1978; Misch 1952; Steinhausen & Adamek 1997). Only a few studies have looked carefully at the psychopathological disorders of the parents. Those who did so found a high rate of depression in one of the parents, a psychiatrically disturbed father (21 %, Kolvin & Fundudis 1981) or a psychiatrically disturbed mother (17 %, Kolvin & Fundudis 1981). In a recent study Steinhausen and Adamek (1997) found a significant, higher frequency of taciturnity in the first-, second- and third-degree relatives of children with elective mutism in comparison to a matched control group of children with the combination of an emotional disorder and a developmental disorder of articulation or expressive language. The family as a whole is very often described as isolated and shy (Funke *et al.* 1978; Rösler 1981; Wergeland 1979).

Maternal overprotection

Many studies found a strong and persistent bond of interdependence between the mother and the child (Hayden 1980; Wergeland 1980; Wilkins 1985; Wright et al. 1985). Hayden (1980) created the term "symbiotic mutism", and family counseling and therapy are thought crucial for intervention (Wright et al. 1985).

Mental retardation

In most of the patients with elective mutism, intellectual functioning is thought to be average or above average (Hayden 1980; Hesselman 1983), but some studies have reported the presence of mental retardation. Two of the

four cases observed by Reed (1963) had a nonverbal IQ below 70, and Kupietz & Schwartz (1982) described cognitive deficits in two of their three subjects. Klin & Volkmar (1993) described mental retardation in two case reports with elective mutism, and Kolvin & Fundudis (1981) found lower mean nonverbal IQ for their elective mutes in comparison to their normal and speech-disordered control group. Kumpulainen and Rasanen (1998) found that one third of the children with selective mutism was performing at school at a lower level than average.

Hospitalization or trauma before age 3

In some of the cases, mutism was precipitated by a single or a series of traumatic events (Hayden 1980) or hospitalization in early childhood (Wright et al. 1985). A traumatic experience during the development of speech was present in one third of a clinical sample of electively mute children (Andersson & Thomsen 1998).

The causative nature of the above mentioned "predisposing factors"remains altogether unclear. However, the high load of communication deficits in the families of children with elective mutism lends some evidence to the assumption that genetic factors may play a role in the etiology of this disorder.

Follow-up studies

There are not many follow-up studies in children with elective mutism. Only a few of them include more than 10 cases and only three studies compared treated with untreated patients (Kurth & Schweigert 1972; Rösler 1981; Wergeland 1979). Further on, the age at follow-up was only in a few studies above 18 (Wergeland 1979; Reed 1963). The following results can be put forward:

- With regard to the core symptomatology, complete remission or at least remarkable improvement could be found after a mean follow-up interval of five years in between 53 % (Kurth & Schweigert 1972) and 100 % (Reed 1963; Wergeland 1979). The average remission rate in our analysis of 10 follow-up studies was 74 % (100 out of 143 patients). Included were only studies with more than four cases.
- Within the three studies which differentiated between treated and untreated patients (Kurth & Schweigert 1972; Rösler 1981; Wergeland 1979), the average remission rate was 63 % (12 out of 19 patients). But only in one of the studies with the shortest follow-up interval (3.5 years) was the remission rate of the untreated patients lower (33 %) than within the treated ones (53 %) (Kurth & Schweigert 1972). In the other two studies, the results were approximately equal, one study showing a slightly higher remission rate of the untreated cases as compared to the treated (100 % vs. 94 %) (Rösler 1981).
- There seems to be a relationship between the remission rate and the follow-up interval in the direction of better results for patients who had been followed up after a longer time-span (over 10 years). This might have to do with the fact that in most of the cases the duration of the disorder is more than 5 to 7 years (Poller 1989).
- However, the remission of the main symptomatology (non-speaking to other people) does not mean that formerly mute patients are without pathological findings. Several authors report even after many years communication deficits, shy and withdrawn behavior, personality disorders and in some cases also transitions into psychotic states (Hayden 1980; Wergeland 197; Wright 1968). In a study by Kolvin (1994), 38 % of the followed-up sample of patients with elective mutism were unemployed for more than two years compared to 20 % of patients with speech disorders.
- Predictors of unfavorable outcome are poor family condition, uncooperative or psychiatrically disturbed parents (Funke et al. 1978; Kolvin & Fundudis 1981; Lowenstein 1979; Sluckin et al., 1991), intellectual impairment of the patient (Kurth & Schweigert 1972; Lorand 1960; Wright 1968), and cerebral dysfunction (Funke et al., 1978).

Because the data on course of elective mutism have only a small empirical basis and the role of predictors of outcome including the benefits of therapy are still quite unclear, we decided to perform a follow-up study on patients referred for elective mutism to our department. Additionally we compared the symptomatology of our sample at time of referral with an age-, sex- and social class-matched control group with other emotional disorders in order to give a more extensive description of symptoms that are specific for mutistic patients.

Methods

Sample

Included were all patients (n = 49) without any selection who had been referred between 1964 and 1979 to the Department of Child and Adolescent Psychiatry (inpatient and outpatient unit) and to the Child Guidance Clinic which works closely together with the department. At referral all patients suffered for at least two years from the condition which can be looked upon as an indicator of severity. All patients were re-classified according to DSM-III-R criteria. With regard to these criteria, 3 patients had to be excluded because they were mentally retarded and another patient had to be excluded who showed mutism on the basis of a hysterical reaction which lasted only a few days. The remaining 45 patients (23 boys and 22 girls) were included. Their age at time of referral was 8.7 ± 3.6 years. For 41 of the 45 patients, sufficient information could be obtained at follow-up and 31 patients could be investigated personally. For 5 of the 10 patients who could not be investigated personally, detailed information could be gathered by interview of the parents and for the other 5 cases, we received information by interviews of the family doctors, institutions and by letters from the patients themselves. All patients lived at home at the time of referral, in 2 cases there was no father at home, and one child was adopted. As far as social class is concerned, 31 of the 45 families belonged to the lower social class according to the classification of Kleining and Moore (1968). Only 10 families could be classified as belonging to the middle class, and there were only two upper class

families. For two families, the social class could not be classified correctly due to lack of information.

The average age in the sample at follow-up was 20.5 ± 6.7 years; the follow-up interval was 12.0 ± 5.2 years.

Control group

A control group matched according to age, sex, and social class comprising 46 patients with emotional disorders excluding mutistic reactions was formed out of the computerized case documentation of our department in order to compare the symptomatology of the patients with mutism at the time of referral, not at the time of follow-up. The ICD–9 diagnoses of the control group comprised disturbances of emotions specific to childhood and adolescence (ICD 313) with the subgroups 313.1 (with misery and unhappiness) and 313.3 (with relationship problems).

Instruments

Instruments at the time of referral (T_1)

All relevant information gathered at the time of referral (T_1) were filled into a standardized documentation sheet which was nearly identical with the basic documentation used at the Department of Child and Adolescent Psychiatry, Philipps-University of Marburg, since 1981. This documentation contains information about the history of the patient, the family history, pre- and perinatal data, data on individual development, school situation, general and psychopathological symptomatology at referral, treatment measures, course of treatment, and detailed information about the family (e. g., family situation, education, conflicts and problems).

Instruments at follow-up (T_2)

At follow-up, the following instruments were used:

- An individually carried out standardized psychopathological examination. This examination also included the Marburg Symptom Checklist which is an instrument for the assessment of psychiatric symptoms in children and adolescents. The Marburg Symptom Checklist is a well-constructed instrument with an interrater reliability between 0.72 and 0.83 (kappa values). The highest kappa values (> 0.80) between two independent raters in a sample of 33 patients could be obtained with regard to the scales 6, 12, 13, 16, 18, 20 and 21, the lowest kappa values (< 0.60) for scale 3. All other interrater reliability scores ranged beyond kappa values of 0.65 (Brandenbusch 1995). The Symptom Checklist consists of 22 rating scales, in which the most important symptoms respectively symptom areas are defined and specified. The names of the rating scales are:
  (1) antisocial behavior,
  (2) aggressive behavior,
  (3) social contact problems,
  (4) anxiety, fears and phobias,
  (5) mood disorders (depression),
  (6) insufficient achievement orientation,
  (7) excessive achievement orientation,
  (8) developmental delays,
  (9) psychomotor symptoms,
  (10) hyperactive symptoms,
  (11) enuresis and encopresis,
  (12) psychosomatic symptoms I (stomach ache, headache, sleeping disorders),
  (13) psychosomatic symptoms II (allergies, asthma, respiratory embarrassment, skin affections),
  (14) eating disorders,
  (15) stuttering and cluttering,
  (16) delusions, depersonalization, derealization, thought disorders,
  (17) obsessive-compulsive symptoms,
  (18) suicidal thoughts or acts,
  (19) sexual disorders,
  (20) alcohol or drug addiction,
  (21) organic diseases, handicaps or injuries,
  (22) other disorders (not coded on the preceding items).

  All rating scales (except the item "sexual disorders") have five rating-points: 0 = symptoms not present, 1 = minimal level, 2 = slight/mild level, 3 = remarkable/significant level, 4 = extreme level. The rating points are explained for each rating scale.

  The development of the Marburg Symptom Checklist was originally based on a comprehensive checklist with 98 single symptoms. This checklist was administered in a clinical population of 1591 children and adolescents (inpatients and outpatients). Factor analyses of these data showed that the list could be reduced to 22 independent factors. These results were taken as basis for the definition of the items in the Marburg Symptom Checklist so that each item represents an independent symptom factor (Mattejat & Remschmidt 1993).
- The standardized basic documentation of the Department of Child and Adolescent Psychiatry at the Philipps-University Marburg.
- The Mannheim Biographic Inventory (MBI) (Jäger et al. 1973) for patients younger than 18 years. The MBI is a well-constructed questionnaire for children and adolescents from 11 to 18 years with 11 subtests comprising the following problem areas: family situation, parental support (S1), independence (S2), social contact (S3), school situation (S4), working/learning behavior (S5), creativity (S6), assertiveness/endurance (S7), achievement motivation/ambition (S8), stress behavior/frustration tolerance (S9), self esteem/ego representation (S10), health/physical maturity (S11) and a total score: positive stimulation vs. deprivation as experienced by the proband. The questionnaire has a sufficient discriminant and factorial validity; the re-test reliability after a time-interval between 8 days and 6 weeks varies between 0.63 and 0.88 in relation to age.
- The Biographic Inventory (BI) (Jäger et al. 1976) for patients older than 18 years. The Biographic Inventory for the diagnosis of behavior disturbances (BI) is a multidimensional questionnaire for the use in the clinical and non-clinical field to diagnose behavior disorders and personality disturbances. It can be used in adult age from 18 years onwards. The inventory contains 8 scales, addressing the following problem areas: family situation in childhood and adolescence (S1), ego strength/assertiveness (S2), current social situation (S3), parental education style (S4), neuroticism/emotional lability (S5), social activity/contact (S6), psychological robustness/stress tolerance (S7), extraversion/social open-mindedness (S8). Also this method has a sufficient validity; the re-test reliability quotients after a time-span of 14 days varying between 0.61 and 0.85.

Data analysis

When analyzing the relationship between dichotomized variables in 2x2-tables, besides Fisher's exact test, Stuart's τ_c measure of association was used in Tables 6, 7 a and 7b. τ_c measures the relative excess of the number of positive concordant observations over the number of negative concordant observations and is adjusted for ties and table size (Brown & Benedetti 1977).

For the purpose of prediction of the variable 'need for therapeutic intervention at follow-up' from the state at first referral time we used the CART (classification and regression trees) program of Breiman, Friedman, Olsen and Stone (1984). CART is a nonparametric method for discrimination (if the predicted variable is discrete, as in our case) and regression (if the predicted variable is continuous). It provides decision-trees with nodes built up from the predicting variables and a threshold.

Results

Approximately half of the patients were referred for investigation by the family physicians (21 of 45 cases 47 %). In three cases the referral was carried out by other psychiatric hospitals (3 of 45,7 %), and in 17 cases (38 %), the parents came spontaneously by themselves to our department or child guidance clinic in order to seek help. Two patients were referred by the public health agency, and in two cases no information about the kind of referral were available.

Results at the time of referral (T_1)

Family psychopathology

The family psychopathology was rated by one of the investigators (MP) according to the detailed information about the family history and the family behavior at referral and during the phase of treatment. For this global rating, three levels were used: 0 = no relevant psychopathology, 1 = minor psychopathological disturbances which do not alter remarkably every day life, 2 = severe psychopathological disorders with a clear negative impact on every day life and family functioning.

According to this rating, only 18 of the 45 *mothers* (40 %) could be rated as psychiatrically healthy. All other mothers revealed some kind of psychopathology; 18 mothers showed minor psychopathological disturbances as frequent mood changes, irritability, lack of drive, disturbances of social contact. Nine mothers (20 %) showed severe psychopathological symptoms such as chronic depression (n = 2), psychopathological problems in relation with intellectual impairment (n = 3), chronic neurotic disorders (n = 1) and personality disorders (n = 1). One mother suffered from brain damage and another one from chronic bronchial asthma.

As far as the *fathers* were concerned, only 4 showed no relevant psychopathology, 12 revealed minor psychopathological symptoms, and 16 remarkable psychopathological disorders. Among them were 3 alcoholics, 2 with severe personality disorders, one father, suffering from chronic depression, had committed suicide. Thirteen fathers revealed a remarkable strange behavior characterized by withdrawal, shyness, irritability, or aggressive behavior.

Family conflicts

In line with the frequency of psychopathology, there was also a high rate of conflicts within the families. In 19 out of 45 families (42 %), abnormal psychosocial conditions could be observed. Three families (7 %) lived in extreme social isolation and far away from communities, but the 16 other families were also isolated in terms of poor social contacts, withdrawal, conflicts with the neighbors and a position as outsiders in their community. Thirteen of the 45 families had an insufficient income. In more than half of the families (28 out of 45, 62 %), remarkable intrafamilial conflicts could be detected. The majority of them dealt with marriage problems of the parents (n = 21, 47 %), the rest with quarrels with the grandparents or with rivalry between the siblings.

The parenting style was also very problematic. In the majority of the families (38 out of 45 families, 84 %), it could be rated as deviant or insufficient. Whereas the behavior management of the fathers was more often characterized by poor control, overstrain and punishment, the mothers tended more to overprotection and close bonding.

A comparison of the group of mutistic patients in terms of a standardized symptom list at the time of referral with a control group matched according to age, sex and social class of patients with emotional disorders, revealed only two significant differences concerning soft neurological signs and attempts of suicide. The first condition was more frequent in the mutistic group. Twenty-four of the 45 patients (53 %) had suffered from some kind of brain dysfunction until referral or during the treatment period, whereas only 6 out of 46 patients (13 %) of the control group showed the same condition.

Attempts of suicide, however, were by far more frequent in the control group of patients with emotional disorders (14 out of 46, 30 %) and did not occur at all in the mutistic group.

Intragroup comparison

A further comparison of the subgroup of mutistic patients who were younger than 10 years at time of referral with those who were older, also showed two significant differences: obsessive-compulsive symptoms only occurred in the older group (4 out of 7 patients, 57 %) but in none of the younger group (n = 38) ($X^2 = 17.30$ $df = 1$ $p < 0.0001$), and all patients of the older group had been treated as inpatients which applied only to 16 out of 38 patients (42 %) of the younger age group ($X^2 = 5.78$ $df = 1$ $p < 0.016$).

Language development and age of manifestation

In 15 out of 45 patients (33 %), developmental speech and language disorders could be diagnosed. These disorders were mild in 7 cases and severe in 8 cases (18 %). Stammering and other problems of articulation were diagnosed in 10 patients (22 %), a dysgrammatical language in 6 patients (13 %). Stuttering was found only in two patients (4.4 %) who suffered from multiple developmental speech and language disorders.

In nearly all of the cases, the first age of manifestation was three years. We call this primary manifestation. Only in two cases take, the first manifestation of the disorder did place later, in one child, as a secondary reaction on a decline in performance, in another case as a reaction to

separation from the mother. However, most of the children showed a remanifestation of mutistic reactions when admitted to the kindergarten (17 out of 45) or when they were sent to school or preschool (35 out of 45).

The duration of mutism from the beginning (3rd year of life) until diagnosis varied remarkably between 2 and 14 years with an average duration of 5.5 ± 3.3 years.

All cases could be diagnosed as elective mutism; only two patients revealed very short phases of total mutism which means that during these phases they did not speak to anybody at all. All of the children had at least a selective mutistic behavior outside the family. Thirty-seven of the 45 children (82 %) did not speak at all with adult persons outside the family; 8 of 45 (18 %) refused to talk only to special key people, mostly teachers.

Speech and language problems within the family

A high frequency of speech and language problems could be found in the families. In many of them, mutistic reactions, poor language production, extreme shyness, speech and language disorders or a remarkable dialect could be found. This was the case in 35 out of 45 families (78 %). Table 1 gives a short summary of speech and language problems within the families.

As Table 1 demonstrates, 4 fathers and 8 mothers had a history of mutism which was not present when the child developed this symptomatology, and in 8 families one of the siblings had also a mutistic symptomatology.

Intelligence

Because of the refusal to speak, a language-free test had to be administered. In most of the cases, the Wechsler-Scales were used. In some cases, the Culture-Fair Test by WEISS and the RAVEN-Test were administered. In 39 of the 45 children (87 %), the IQ varied between 85 and 114, in 6 children (13 %) an IQ between 70 and 84 was found, and there was no child with an IQ above 114.

The average performance-IQ of those children who had been tested by the Wechsler-Scales was 98 ± 6, range: 79–118. In 15 of the 45 patients (33 %), a specific developmental dyslexia (n = 14) and/or a specific arithmetical retardation was found.

Developmental data

In 21 of the children (47 %), complications during pregnancy, at birth or in the neonatal period could be documented. In 7 cases, these complications were severe. The mean age of the mothers at delivery was 27 ± 5 years (range: 17–39 years).

In 13 of the 45 children (29 %), a separation experience (separation of more than 4 weeks from the mother) was reported. In 22 of the patients (49 %), the communication behavior within the family was reported as being deviant in the sense of poor speech motivation, withdrawal and poor communication in general. When first attending school, in all patients communication deficits were present, 21 (64 %) revealed achievement disorders or attention deficit disorders, and 8 of these patients repeated a school class once or several times. One child was transferred to a special school.

At the time of referral, 30 patients (67 %) went to school, 21 of them to an elementary school, 4 to a preschool, 4 to an advanced elementary school, and one patient to a special school for learning disabled children. From the remaining 15 patients, 8 were preschool children, 4 were expelled from school (due to their communication problems and oppositional behavior), and 3 had already left school.

Symptomatology at the time of referral (T_1)

According to a symptom list which included somatic and psychopathological symptoms, a very careful examination of child's symptomatology was carried out.

Table 2 gives an overview about the somatic symptoms at referral and Table 3 an analogous one about the psychopathological symptoms at referral and at follow-up.

Therapy

In 17 of the patients, an inpatient therapy was carried out lasting between 21 and 399 days; the average dura-

Table 1 Speech and communication problems in the families of 45 mute children

History and symptoms	n	%
History of mutism or mutistic reactions in the parents	12	27 %
Mother	8	18 %
Father	4	9 %
Mutistic reactions in the siblings	8	18 %
Other language and communication problems in the family		
Taciturnity of the father	23	51 %
Taciturnity of the mother	20	44 %
Taciturnity of the siblings	8	18 %
Speech disorders (stuttering, cluttering, articulative problems)	4	9 %
Remarkable dialect (i. e., inability to talk standard)	4	9 %
Remarkable differences in speech/language functions between family members	19	42 %

Table 2 Somatic symptoms of 45 mute children at referral. Symptomatology was rated according to a standardized symptom list

Somatic symptoms	n	%
Deformations of the face (Ichthyosis, vitiligo, eczema)	3	7 %
Epilepsy	1	2 %
Functional motor disorders (thumb sucking, nail biting, tics, motor stereotypies)	8	18 %
Enuresis/encopresis	15	33 %
Eating disorders	21	47 %
Psychosomatic symptoms I (stomach ache, headache, sleeping disorders)	20	45 %
Psychosomatic symptoms II (allergies, skin affections, asthma)	9	20 %
Vegetative symptoms	5	11 %

Table 3 Psychopathological symptoms at referral (n = 45) and at follow-up (n = 31). Symptomatology was rated according to a standardized symptom list

Symptoms	T_1, referral (n=45)		T_2, follow-up (n=31)	
Lack of contact	34	76%	3	10%
Separation anxiety	12	27%		
Psychomotor disturbances	34	76%	11	35%
Insecurity/low self esteem	35	78%	4	13%
Poor concentration	7	15%	9	29%
Pronounced anxiety	36	80%	2	6%
Irritability	12	27%	3	10%
Dysphoric mood	6	13%	3	10%
Depression	2	4%	6	19%
Impulsivity	8	18%	15	48%
Permanent mutistic	28	62%	5	16%
Intermittent mutistic behavior	17	38%	8	26%
Global rating: severe psychopathological disturbances	26	58%	13	42%

tion of the inpatient treatment was 131 ± 126 days. The inpatient therapeutic regime was based on a behavioral approach including a stepwise training program to facilitate speech production starting with non-verbal activities in a single patient-therapist session which was extended later to sessions with other children who were well-known to the patient. In the younger patients also an individual play therapy was carried out.

In addition, the therapeutic program contained the following elements:

- Systematic counseling of the parents including clear information about the nature of the disorder and the kind how to communicate with the child,
- School attendance within the hospital as soon as this was possible,
- Facilitating speech production during a wide range of recreation activities.

In 22 of the cases, only family counseling was carried out, not a systematic therapy, and in 6 cases a long-lasting outpatient play therapy was carried out.

Taking into account the long recruitment period (1964–1979), it is self-evident that there were many changes concerning the therapeutic regime as well as the attitudes to inpatient and outpatient or daypatient treatment. For the whole period, a daypatient treatment was not yet available. It is however interesting that the number of children with elective mutism referred to our department did not change over time. In the first time period of our study (1964–1971), 23 children were presented, while between 1972 and 1979, 21 children were referred. But during the first period, approximately half of the patients were referred to inpatient treatment (n = 11, 48 %), whereas during the second period, only a quarter of them was hospitalized (n = 6, 27 %).

Results at follow-up

As already mentioned, 31 of the patients could be investigated personally, in 5 further patients, sufficient information could be obtained by the parents, and in 5 other cases, reliable information could be gathered by other persons who knew the patients very well. So the results at follow-up vary in relation to the information that could be obtained.

General aspects

School. Thirty-seven patients were still attending school at the time of follow-up. From these, 22 attended the upper classes of the elementary school (59 %), 8 attended the high school (22 %), 2 after having finished the elementary school attended a special high school (5 %) and 5 (14 %) a special school for children with learning disabilities. Nineteen patients had already left school and started a professional training which had already been finished by 9 of them.

Of the patients 27 (68 %) out of 40 for whom sufficient information was available still lived with their parents. Twenty-three patients who were older than 18 lived outside the family, 10 of them together with a partner.

Speech and language problems (course of the disorder). With regard to mutism, at the time of follow-up, the following results could be obtained. In 16 out of 41 cases (39 %), a complete remission could be stated. In 12 cases the patients or the relatives described a remarkable improvement (29 %), in 8 cases only a mild improvement was reported (20 %) and in 5 cases the symptomatology was unchanged.

For those patients who showed a complete remission or remarkable improvement of the symptomatology (n = 28), the duration of mutism was on average 9 ± 4 years (range 3 to 15 years). For those with an unchanged symptomatology, the duration of the disorder was between 20 and 30 years.

In patients with a partial or complete remission (n = 36), 7 cases (19 %) showed an abrupt normalization of their disorder. In the majority of the cases (29 out of 36 patients, 81 %), however, improvement occurred step by step with relapses in 7 cases (19 %).

In addition, it was interesting to note that except of the 16 patients with a complete remission (39 %), all other patients had still some communication problems. Eleven of them (27 %) were afraid of unknown situations and of talking to strangers; 6 patients (15 %) had general problems of verbal communication. They were, e. g., afraid of talking on the telephone or talking in shops and offices and 8 patients (20 %) revealed mutistic reactions from time to time. Among them were also the 5 patients in whom the symptomatology was unchanged, i. e., they were still mute.

Subjective experiences of the patients with their disorder

In 25 patients, reliable information about their subjective experiences with their disorder could be obtained. All of them were willing and able to talk with the investigators but the majority were still shy and somewhat withdrawn. Nevertheless it was possible to obtain verbal answers from them on the questions of the standardized interview concerning their subjective experiences. This was a very interesting aspect. All of these patients looked upon their disorder as a very serious problem from which they suffered intensively. Only 3 of these patients did not report intensive suffering.

For many patients, mutism was associated with remarkable anxiety states which were reported by 14 patients and with feelings of shame and insufficiency (n = 18). Fourteen patients also reported a very sceptic attitude and mistrust of nearly all people in their environment. The majority of the patients (n = 17) also reported a connection of their symptomatology with conflict situations inside and outside the family. Only 6 out of the 25 patients looked upon their mutistic reaction as a strange and unexplainable phenomenon.

Psychopathological symptoms. Psychopathological symptoms at follow-up of the patients who could be investigated personally were rated according to the same standardized symptom list that was used already at the time of referral. Table 3 describes the results. In comparison with the time of referral, a remarkable decline of symptomatology can be observed. However, there are still several psychopathological symptoms present, especially psychomotor disturbances, poor concentration, impulsivity and also intermittent mutistic behavior. In addition, 13 of the 31 patients (42 %) are rated as severely disturbed.

Results of psychometric measurements

Results of the Mannheim Biographic Inventory (MBI) for patients younger than 18 years. For the 13 patients who were younger than 18 years at follow-up, the results with the Mannheim Biographic Inventory (MBI) are listed up in Table 4. With regard to multiple testing, Bonferroni corrections were used in order to avoid significant results by chance.

After adjustment, Table 4 demonstrates four significant differences between the group of the younger mutistic patients and the reference group of the Mannheim Biographic Inventory (n = 2832). These differences are all pointing into the same direction: the results are unfavorable for the mute patients as compared with the normal reference group. The patients of the mutistic group (n = 13) describe themselves in the Mannheim Biographic Interview as being less independent, less motivated with regard to school and efficiency, less self-confident and less mature, respectively healthy.

Results of the Biographic Inventory (BI) for patients older than 18 years. Table 5 demonstrates the results of the comparison of the patients who were older than 18 years at follow-up with the reference group of the Biographic Inventory (n = 751). There are three significant differences between the groups, one in a positive direction (social activity/sociability) and two in a negative direction, concerning psychological robustness/stress tolerance and extraversion/social open-mindedness.

Results with regard to prognosis

At the end of the investigation at follow-up, a global rating of psychosocial and speech disturbances was carried out by one of the investigators (M. P.) using a three-point scale with 1 = complete recovery, 2 = mild disturbances, and 3 = severe disturbances. The patients received the global rating 1 when completely recovered and free of marked psychosocial or speech disturbances. Rating 2 was given if mild psychosocial or speech disturbances could be detected which did not interfere intensively with everyday functioning at home, at school or at work, and rating 3 was given if there were severe disturbances

Table 4 Comparison at follow-up of the mute patients younger than 18 (n = 13) with regard to the 11 scales (S) of the Mannheim Biographic Inventory (MBI) with the reference group of this inventory (n = 2832). α-adjustment for multiple testing of 11 scales according to Bonferroni-Holm, global $\alpha = 0.05$, adjusted α's: $\alpha/11, \alpha/10, \ldots, \alpha$

Scales of MBI	Follow-up group (n=13) $x_1 \pm s_1$	Reference group (n=2832) $x_2 \pm s_2$	t	p
Family situation/parental support (S1)	6.85 ± 1.82	5.91 ± 1.16	1.87	0.0861
Independence (S2)	4.69 ± 2.14	7.03 ± 1.23	3.95	0.0019*
Social contact (S3)	5.08 ± 2.69	5.06 ± 2.05	0.03	0.9761
School situation/achievement motivation (S4)	5.77 ± 1.54	7.54 ± 1.07	5.94	0.0000*
Learning behavior (S5)	4.69 ± 2.36	5.88 ± 1.59	2.70	0.0070
Creativity (S6)	4.77 ± 2.01	6.36 ± 1.20	2.85	0.0146
Assertiveness/endurance (S7)	5.00 ± 2.04	5.38 ± 1.26	0.67	0.5155
Achievement motivation/ambition (S8)	3.85 ± 2.58	5.69 ± 1.60	2.57	0.0245
Stress behavior/frustration tolerance (S9)	6.46 ± 1.71	7.97 ± 1.09	3.21	0.0075
Self esteem/ego representation (S10)	5.23 ± 2.13	7.58 ± 1.23	3.98	0.0018*
Physical maturity/health (S11)	6.62 ± 1.61	8.15 ± 1.30	4.23	0.0006*
Total score:				
Positive stimulation vs. deprivation	59.20 ± 15.6	72.50 ± 6.59	3.08	0.0095

Table 5 Comparison at follow-up of the mute patients older than 18 (n = 18) with regard to the scales of the Biographic Inventory (BI) with the reference group of this inventory (n = 751). α-adjustment according to Bonferroni-Holm procedure, adjusted α's: 0.05/8, 0.05/7, . . . , 0.05

Scales of the Biographic Inventory (BI)	Follow-up group (n=18) $x_1 \pm S_1$	Reference group (n=751) $x_2 \pm S_2$	t	p
Family situation in childhood and adolescence	4.11±3.68	5.28±4.04	1.22	0.2228
Ego strength/assertiveness	3.56±2.53	4.02±2.50	0.77	0.4415
Actual social situation	1.28±1.49	1.44±1.63	0.41	0.6819
Parenting style	5.44±5.12	7.08±5.23	1.31	0.1906
Neuroticism/emotional lability	3.11±2.35	4.19±2.78	1.64	0.1014
Social activity/sociability	5.39±2.62	3.38±2.57	3.28	0.0011*
Psychological robustness/stress tolerance	2.28±1.64	3.57±2.80	3.22	0.0050*
Extraversion/social open-mindedness	3.06±1.63	4.88±2.61	5.68	0.0000*

in the psychosocial and/or the language field that caused remarkable problems in everyday life.

With regard to these criteria, 41 patients could be rated, 31 of them by the use of personal information, and 10 based on information from the family and other persons who knew the patients very well.

In the light of this subdivision of the patients into two groups (group A comprising the recovered or mildly disturbed patients (n = 30) and group B comprising the severely disturbed patients (n = 11)), all relevant items at referral were tested with regard to group differences in order to find out a rank order of symptoms or problems that might be of prognostic value. The results of this testing are demonstrated in Table 6. It shows that the most pronounced prognostic item is mutistic behavior within the core family, followed by deviant parenting style, psychiatric disorders in the family, depression or dysphoric mood of the patient and psychiatrically disturbed father. Mutism within the core family means that the child did not speak with family members, either.

Table 6 includes all the patients of whom we received relevant information. So the question was whether these prognostic factors might be age-dependent. For this reason, the same procedure was applied to the subgroup of patients who were 16 and older at the time of follow-up. Again, this group (n = 31) was subdivided into a group C comprising recovered and mildly disturbed patients (n = 22) and a group D of severely impaired patients (n = 9). The comparison of groups C and D showed the same rank order of the prognostic items as demonstrated in Table 6; the most pronounced prognostic item differentiating between the two groups was mutism within the core family ($X^2 = 12.68$ df = 1, $p < 0.0004$).

Finally, two other statistical methods were applied in order to predict the results at follow-up by variables defined at the time of referral: logistic regression and the Classification and Regression Trees (CART) by Breiman et al.(1984). A case is placed left at a node, if it has a value of that item which is less than or equal to the threshold; otherwise it is placed on the right branch. At the end of the classification process, each case belongs to exactly one terminal node. The terminal nodes are assigned to the values of the predicted variable according to a weighted majority role, depending on the given a-priori distribution of that variable and a given loss function.

As usual for scientific purposes, we assigned equal a-priori probabilities to the two values of the predicted variable and equal loss associated with false positive and false negative decision since this choice usually yields well differentiated trees.

As part of the CART algorithm, first a tentative large tree based on the whole derivation sample is created, then from this tree, unstable branches are cut off (pruned) by an internal cross-validation process. In our case, after this pruning process, only the variable 109 ('mutism of the child present also in the core family') remained in the tree which therefore reduces to a simple 2x2 table. This result is interpreted in such a way that, according to CART, only this one variable contains enough predictive information to be extracted when sample size is as ours about 50.

The subdivision of the entire sample of formerly elective mute patients (n = 41) is the same as in Table 6. Group A (recovered or mildly disturbed) patients (n = 30) were distinguished from Group B (severely disturbed patients at follow-up, n = 11). Both methods led

Table 6 Prognostic value of symptoms and problems at referral with regard to outcome after an average follow-up interval of 12.0 ± 5.2 years

Problems at referral (T_1)	Absolute frequency and percentage of psychosocial and speech disturbances at follow-up (T_2)				One-sided p-value (Fisher's exact test)
	Group A Recovered or mildly disturbed (n=30)		Group B Severely disturbed (n=11)		
Mutism within the core family	5	17%	9	82%	0.0002
Deviant parenting style	10	33%	9	82%	0.0074
Psychiatric disorders in the family	11	37%	9	82%	0.0120
Depressive/dysphoric mood	7	23%	8	73%	0.0058
Psychiatrically disturbed father	8	27%	7	64%	0.0360

to the same result, the variable with the highest predictive power being the item "mutism within the core family". Table 7 a shows a rank order of variables with high association to the most predictive variable "mutism within the core family". All variables are restricted to the time of referral.

Table 7 b demonstrates the association of several variables gathered at follow-up with the most powerful predictive variable "mutism within the core family". A high association exists with severity of psychopathology at follow-up, global rating of the severity of speech and language behavior, with psychiatric disturbances of the patient during the last five years and with his or her social adaptation at follow-up. All associations express the same general tendency: the higher the degree of pathology the higher the association with the predictive variable.

Finally, a further CART analysis was carried out including 48 variables defined at referral (T_1) and categorizing only those 29 patients who, at the time of follow-up, could doubtlessly be classified as needing (n = 9) or not needing therapeutic intervention (n = 20). The results of this analysis are demonstrated in Table 8.

Table 8 demonstrates that from the 9 patients who were classified as needing therapeutic intervention 7 could be predicted correctly, whereas from those who were classified as not needing therapeutic intervention, 18 out of 20 could be predicted correctly.

Again, the variable with the highest predictive power was mutism in the core family. If this item was positive, the prognosis at follow-up was poor; if it was negative, the prognosis at follow-up was good. Despite the fact that in the CART procedure a cross-validation is integrated these results should be considered as exploratory due the small sample size in relation to the number of dependant variables.

Table 7 a Association with key variable 'mutism in the core family'. Variables associated at first referral. The top 8 variables from a list of 40 variables, sorted by their p-values. α-adjusted according to Bonferroni-Holm procedure, global $\alpha = 0.05$, adjusted α's: $\alpha/40$, $\alpha/39$, …, α

Variable	p-value Fisher's test	Stuart's τ_c
Deviant parenting style	0.00004*	0.591
Psychiatric disorders in the family	0.00156⁻	0.371
Psychopathology disturbances of the patient at referral	0.00407⁻	0.423
Psychiatrically disturbed father	0.01100⁻	0.298
Abnormal family relationship success of therapy	0.01400⁻	–0.308
Anxiety, fears, phobias	0.01900⁻	0.000
Eating disorders	0.03400⁻	0.290
Antisocial behavior	0.04700⁻	0.200

p-value of Fisher's exact test is used as descriptive tool indicating dependence of variables. Stuart's τ_c measures monotone association.

Table 7 b Association with key variable 'mutism within the core family'. Variables associated with follow-up. The top 6 variables from a list of variables, sorted by their p-value. α-adjustment according 17 to Bonferroni-Holm-procedure, global $\alpha = 0.05$, adjusted α's: $\alpha/17$, $\alpha/16$, …, α

Variable	p-value Fisher's test	Stuart's τ_c
Psychopathology at follow-up	0.00004*	0.585
Global rating of speech and language behavior	0.00033*	0.531
Psychiatric disturbances of the patient during the last 5 years	0.00057*	0.535
Global rating of social adaptation	0.0024*	0.364
Adverse social influences	0.00442⁻	0.231
Global rating of the course of mutism	0.04600⁻	0.366

p-value of Fisher's exact test is used as descriptive tool indicating dependence of variables. Stuart's τ_c measures monotone association.

Table 8 Classification table (cross-validated by CART via leaving-10%-out-method) showing need for therapeutic intervention at follow-up. Prediction of 'need for therapeutic intervention at follow up', using CART with 48 variables describing patients state at time T_1, including into the analyses only those 29 patients which at T_2 could be clearly classified as needing or not needing therapeutic intervention at follow up.
After cross-validation, only variable 109 'Mutism in the family' was built into the classification tree by CART

		predicted no	predicted yes	Total
true	no	18	2	20
	yes	2	7	9
	Total	20	9	29

Specificity:	90%	Sensitivity:	78% (row percent)
pos.pred.value:	78%	neg.pred.value:	90% (column percent)
Youden Index:	68%	(spec. (%) + sens. (%) –100)	
Predictive gain:	68%	(pos.pred.value (%) + neg.pred.value (%) –100	

Discussion

Limitation of the study

As all studies based on retrospectively recruited samples, our study has some methodological shortcomings. First of all, we had to rely on the data collected at time of referral. These data were collected by different investigators, but along standardized procedures (standardized history taking interview, standardized documentation system). Insofar, we do not assume a remarkable distortion of our data set. Secondly, we tried to control our data in two different ways: 1) for the comparison of symptoms at time of referral (and only for that purpose), we formed a control group of 46 patients with emotional disorders excluding mutism, matched according to age, sex, and social class with the mutism group. This control group was retrieved out of the computerized case documentation of our department. 2) in order to compare some of our results at follow-up, we used the norms of two well-constructed and standardized instruments, the Mannheim Biographic Inventory (MBI) and the Biographic Inventory (BI). The application of these instruments occurred in order to compensate to some extent the data collection at follow-up, though done in a standardized way, only by one investigator. Nevertheless, data collection by one investigator

was done in many follow-up studies of the same type as our study.

Results at referral

Our sample of 45 ICD–9 and DSM-III-R diagnosed children with elective mutism comprises only patients with severe disorders. This can be derived from the fact that all of them had been sick for at least two years before they were referred. The average age at time of referral was 8.7 ± 3.6 years and the follow-up interval was 12.0 ± 5.2 years. A thorough examination of history and all relevant data revealed that in all cases, the disorder began even earlier (around the age of 3 to 4 years). The average duration of the disorder until diagnosis was 5.5 ± 3.3 years. Thus the results of our study are based on the data of patients who fulfill a crucial criterion of elective mutism, "the persistent refusal to talk". Many of the previous studies either do not specify the duration of speech refusal (Cunningham 1983; Goll 1979; Lowenstein 1979) or include patients with a minimum length of refusal to talk of merely 8 weeks (Hayden 1980). Hence these studies are probably biased by comprising patients with transient mutistic syndromes which should be distinguished from protracted elective mutism.

The observed equal sex ratio differs from other studies (Hayden 1980; Wergeland 1979; Wilkins 1985; Parker et al. 1960; Kopp & Gillberg 1997; Kumpulainen & Rasanen 1998), which reported at least a small female preponderance. However, our sample is not a population-based sample and therefore a "selection bias"may be present in the way that the more handicapped children with additional psychiatric and/or developmental disorders are more likely to be referred and it is known that psychiatric disorders and developmental disorders in this age group are more likely to be present in boys than in girls.

With regard to intelligence, the IQ varied between 85 and 114 in 39 of the patients (87 %) and was in six children (13 %) between 70 and 84. In the sample of Kolvin & Fundudis (1981), 19 % had a performance IQ between 70 and 84, another 19 % proved to be even of lower intelligence. We did not find mute children with IQs above 114 as sometimes reported in the literature (Funke et al. 1978; Hayden 1980; Lowenstein 1979; Rösler 1981; Wright 1968).

As far as psychopathological symptoms at referral are concerned, a comparison with a control group of children with emotional disorders, exactly matched with regard to age, sex, intelligence and social class, revealed only two statistically significant differences. Symptoms of formerly called "minimal brain damage"(neurological soft signs, motor developmental disorders, immaturity signs in the EEG, etc.) were more frequent in the mutistic group whereas attempts of suicide were more frequent in the control group.

There was a high load of psychopathology in the families of the elective mute children. A high proportion of the mothers and fathers as well revealed severe psychopathological symptoms. In line with these results was also a high rate of conflicts within the families and also a high frequency of abnormal psychosocial conditions (in more than 60 % of the families). In 35 of 45 families (78 %), speech and language problems could be detected. One third of the patients themselves had developed speech and language disorders in comparison to 50 % of the Kolvin & Fundudis sample (1981) and to Andersson and Thomsen (1998) who also found a rate of approximately 50 % in their sample. Another third of our subjects had developmental dyslexia and/or specific arithmetical retardation. Also other signs of developmental delay were found in a substantial proportion of the children (see Table 2).

These results are in line with several reports in the literature (Kolvin & Fundudis1981; Goll 1979; Steinhausen & Adamek 1997; Kristensen 2000) and demonstrated that in the etiology of elective mutism two components play an important role: developmental delays and family psychopathology.

Results at follow-up

At follow-up 16 (39 %) out of 41 patients for whom reliable information could be gathered showed a complete remission. In 12 cases (29 %) a remarkable improvement could be observed. In all other cases, only a mild improvement and in five cases no improvement could be stated. It is remarkable that except of the 16 patients (39 %) who showed a complete remission, all other patients had still communication problems (being afraid of unknown situations, of talking to strangers, being afraid of using the telephone, etc.) which was a handicap for them at school, at work and during leisure activities. This result confirmed the observation of Kolvin & Fundudis (1981) that elective mutism is a very persistent condition with a general tendency of poor outcome.

Our results contradict to a certain extent the results of other authors who found a favorable outcome and after some years a complete remission (Reed 1963; Wergeland 1979) it can be assumed that these authors have described cases of minor severity.

With regard to other psychopathological symptoms, the formerly mute patients were not free of psychopathological findings. A substantial group of them revealed psychomotor symptoms (35 %), attention deficit problems (29 %) and emotional problems as depression (19 %), dysphoric mood states (10 %) and impulsivity (48 %).

Also the results of psychometric measures demonstrate that the group of formerly elective mute patients has more problems than normal reference groups. They describe themselves as more dependent, less motivated for school and work, less mature and less healthy (MBI). In addition, they are less stress-tolerant and less open-minded than a reference group (BI). Finally, the results

of the Social Communicative Inventory (SCI) reveal that the formerly mute patients are characterized by a cluster of symptoms including embarrassment and shame, mistrust, phobic avoidance of interpersonal contacts and blocking of speech. In the follow-up report by Kolvin (1994), they also describe worse relationships with their parents – including criticism and physical abuse – than a speech disorder control group.

In summary, the formerly mute patients can be characterized by a high load of psychopathological symptoms mainly in the area of psychosocial relationships and communication. If we put these results together with family pathology investigated at the time of referral, it becomes evident that individual psychopathology and family psychopathology correspond with each other and may form the background for the high persistence and poor outcome of this disorder. Our results are in line with the 2–10 year follow-up study by Sluckin *et al.* (1991) who also found a poor outcome associated with marked family psychopathology.

Finally, we tried to predict the long-term outcome by a set of variables gathered at the time of referral (T_1). Both statistical methods, logistic regression and the classification and regression trees (CART), arrived at the same result. The variable with the highest predictive power for poor outcome was the existence of mutistic behavior of the child within the core family, i. e., the child did not talk to the members of the core family (mother, father, siblings). This result underlines once more the importance of family psychopathology.

References

1. American Psychiatric Association (1987) Diagnostic and Statistical Manual of Mental Disorders, 3rd edition, revised (DSM-III-R). APA, Washington
2. American Psychiatric Association (1994) Diagnostic and Statistical Manual of Mental Disorders. (DSM-IV). APA, Washington
3. Andersson CB, Thomsen PH (1998) Electively mute children: an analysis of 37 Danish cases. Nordic Journal of Psychiatry 52:231–238
4. Brandenbusch A (1995) Die Marburger Symptomliste. Eine Untersuchung zur Beurteiler-Übereinstimmung. Diplom-Arbeit am FB Psychologie, Marburg
5. Breiman L, Friedman F, Olsen, A, Stone CJ (1984) CART: Classification and Regression Trees. Wadsworth, Belmont, CA
6. Brown JB, Lloyd H (1975) A controlled study of children not speaking at school. Journal of the Association of Workers with Maladjusted Children 3:49–63
7. Brown MB, Benedetti JK (1977) On the mean and variance of the tetrachoric correlation coefficient. Psychometrika 42:347–355
8. Cunningham C, Cataldo H, Mellion C (1983) A review and controlled single case evaluation of behavioral approaches to the management of elective mutism. Child & Family Behavior Therapy 5:25–49
9. Dummit ES, Klein RG, Tancer NK, Asche B, Martin J, Fairbanks JA (1997) A systematic assessment of 50 children with selective mutism. Journal of the American Academy of Child and Adolescent Psychiatry 36:653–660
10. Elson A, Pearson C, Jones D, Schumacher E (1965) Follow-up study of childhood elective mutism. Archives of General Psychiatry 13:182–187
11. Fundudis T, Kolvin I, Garside RF (1979) Speech Retarded and Deaf Children: Their Psychological Development. Academic Press, London
12. Funke P, Schlange H, Ulrich I (1978) Klinische Untersuchungen und Therapie bei Kindern mit selektivem Mutismus. Acta Paedopsychiatrica 43:47–55
13. Goll K (1979) Role structure and subculture in the families of elective mutists. Advances in Family Psychiatry 2:141–161
14. Hayden TL (1980) The classification of elective mutism. Journal of the American Academy of Child Psychiatry 19:118–133
15. Jäger R, Berbig E, Geisel B, Gosslar H, Hagen J, Liebich W, Schafheutle R (1973) Mannheimer Biographisches Inventar (M-B-I). Hogrefe, Göttingen
16. Jäger R, Lischer S, Münster B, Ritz B (1976) Biographisches Inventar zur Diagnose von Verhaltensstörungen (B-I-V). Hogrefe, Göttingen
17. Kaplan TN (1992) Elective mutism. A review of the literature. Advances in Clinical Child Psychology 14:265–288
18. Kleining G, Moore H (1968) Soziale Selbsteinstufung (SSE). Ein Instrument zur Messung sozialer Schichten. Kölner Zeitschrift für Soziologie und Sozialpsychologie 20:502–552
19. Klin A, Volkmar FR (1993) Elective mutism and mental retardation. Journal of the American Academy of Child Psychiatry 32:860–864
20. Kolvin I (1994) The origin of elective mutism. 13th International Congress of the International Association for Child and Adolescent Psychiatry and Allied Professions, San Francisco, USA, July 24–28, 1994, Abstracts, 181
21. Kolvin I, Fundudis T (1981) Elective mute children: psychological development and background factors. Journal of Child Psychology and Psychiatry 22: 219–232
22. Kopp S, Gillberg C (1997) Selective mutism: a population-based study: a research note. Journal of Child Psychology and Psychiatry 38:257–262
23. Kristensen A (2000) Selective mutism and comorbidity with developmental disorder/delay, anxiety disorder, and elimination disorder. Journal of the American Academy of Child and Adolescent Psychiatry 39:249–256
24. Kristensen A (2001) A multiple informants' report of emotional and behavioural problems in a nation-wide sample of selective mute children and controls. European Child and Adolescent Psychiatry 10:135–142
25. Kumpulainen KE, Rasanen E, Raaska H, Somppi V (1998) Selective mutism among second graders in elementary school. European Child and Adolescent Psychiatry 7:24–29
26. Kupietz S, Schwartz I (1982) Elective mutism. Evaluation and behavioral treatment of three cases. NYS Journal of Medicine 82:1073–1076
27. Kurth E, Schweigert K (1972) Ursachen und Entwicklungsverläufe des Mutismus bei Kindern. Psychiatrie, Neurologie und medizinische Psychologie 24:741–749
28. Kussmaul A (1877) Die Störungen der Sprache. Schwabe, Basel: Chapter 33 and 31, p. 211 (cit. Misch 1952)
29. Lóránd B (1960) Katamnese elektiv mutistischer Kinder. Acta paedopsychiatrica 27:273–289
30. Lowenstein LF (1979) The result of twenty-one elective mute cases. Acta paedopsychiatrica 45:17–23
31. Mattejat F, Remschmidt H (1993) Marburger Symptomliste: Interviewanleitung und Glossar. Klinik für Kinder- und Jugendpsychiatrie der Philipps-Universität Marburg, Marburg
32. Misch A (1952) Elektiver Mutismus im Kindesalter. Zeitschrift für Kinderpsychiatrie 19:49–87
33. Morris JV (1953) Cases of elective mutism. American Journal of Mental Deficiency 57:661–668 (cit. Reed 1963)
34. Parker EB, Olsen TF, Throckmorton MC (1960) Social casework with elementary school-children who do not talk in school. Social Work 5:64–70 (cit. Kolvin & Fundudis, 1981)
35. Poller M (1989) Mutismus bei Kindern und Jugendlichen. Eine katamnestische Untersuchung. Medical Dissertation, University of Marburg
36. Popella E (1960) Psychogener Mutismus bei Kindern. Nervenarzt 31:257–263

37. Raven JC, Court J, Raven J (1979) Raven-Matrizen-Test. Standard Progressive Matrices. Deutsche Bearbeitung von H. Kratzmeier unter Mitarbeit von R. Horn. Beltz, Weinheim
38. Reed GF (1963) Elective mutism in children: a re-appraisal. Journal of Child Psychology and Psychiatry 4:99–107
39. Rösler M (1981) Befunde beim neurotischen Mutismus der Kinder. Eine Untersuchung an 32 mutistischen Kindern. Praxis der Kinderpsychologie und Kinderpsychiatrie 30:187–194
40. Sluckin A, Foreman N, Herbert M (1991) Behavioural treatment programs and selectivity of speaking at follow-up in a sample of 25 selective mutes. Australian Psychologist 26:132–137
41. Steinhausen HC, Juzi C (1996) Elective mutism: an analysis of 100 cases. Journal of the American Academy of Child and Adolescent Psychiatry 35:606–614
42. Steinhausen HC, Adamek R (1997) The family history of children with elective mutism: A research report. European Child and Adolescent Psychiatry 6:107–111
43. Tramer M (1934) Elektiver Mutismus bei Kindern. Zeitschrift für Kinderpsychiatrie 1:30–35
44. Weiß RH (1980) Grundintelligenztest Skala 2 (CFT 20). 2. edition. Deutsche Adaptation von Cattell RB. Culture Fair Intelligence Test – Scale 2. Westermann, Braunschweig
45. Wergeland H (1979) Elective mutism. Acta Psychiatrica Scandinavica 59:218–228
46. Wilkins R (1985) A comparison of elective mutism and emotional disorders in children. British Journal of Psychiatry 146:198–203
47. World Health Organization (1992) The ICD-10 Classification of Mental and Behavioural Disorders. Clinical Descriptions and Diagnostic Guidelines. WHO, Geneva
48. Wright HL (1968) A clinical study of children who refuse to talk in school. Journal of the American Academy of Child Psychiatry 7:603–617
49. Wright HH, Miller MD, Cook MA, Littmann JR (1985) Early identification and intervention with children who refuse to speak. Journal of the American Academy of Child Psychiatry 24:739–746