Social Adaptation of Children with Congenital Fecal Dysfunction: From the Viewpoint of the Mother-Child Relationship

SYUNICHI FUNAKOSI, JUNKO HAYASHI,1 TAKAMICHI KAMIYAMA,2 TAKASHI UENO,3 TOMOHIRO ISHII,2 MOTOSHI WADA,2 YUTAKA HAYASHI2 and HIROO MATSUOKA

Department of Psychiatry, Graduate School of Medicine, Tohoku University, Sendai, 1 Nagasaki University School of Medicine, Nagasaki, 2 Department of Pediatric Surgery, Graduate School of Medicine, 3 Division of Clinical Psychology, Department of Human Development and Disabilities, Graduate School of Education, Tohoku University, Sendai, Japan

FUNAKOSI, S., HAYASHI, J., KAMIYAMA, T., UENO, T., ISHII, T., WADA, M., HAYASHI, Y. and MATSUOKA, H. Social Adaptation of Children with Congenital Fecal Dysfunction: From the Viewpoint of the Mother-Child Relationship. Tohoku J. Exp. Med., 2005, 206 (2), 117-124 — Excretory dysfunction associated with congenital anal anomalies (a generic term that includes anal atresia and Hirschsprung’s disease) is presumed to greatly affect the psychology of the affected children. In this study, we conducted a survey on the psychological status of children with excretory dysfunction, and investigated the relationship between the affected children and their families in addition to their social life. Four children with fecal dysfunction due to a congenital anal anomaly and their mothers were interviewed. The results of psychological tests in the children and mothers along with our findings in the interviews with the children and mothers, were included in the data analysis. We found that regardless of the degree of excretory dysfunction, the child’s state of mind was influenced by whether the mother exhibited warmth or criticism towards her child and whether there was a support system for the mother and child. We suggest that psychiatric consultation is necessary for these children.

Liaison child psychiatry is targeted to pediatric patients with a variety of somatic disorders such as asthma, diabetes, childhood cancer, AIDS, and problems associated with kidney or liver transplantation, in which psychiatric problems in the feelings and behavior of these children are treated. Studies on the psychiatric problems of patients with somatic disorders have focused on the influence of the particular somatic disorder on the psychology of the affected children and risk factors for unfavorable psychological states (Knapp and Harris 1998a, b).

Excretory dysfunction associated with congenital anal anomalies (a generic term for imperforate anus and Hirschsprung’s disease) is presumed to greatly affect the psychology of affected
children. Anal atresia is a congenital anorectal anomaly that is diagnosed in the neonatal period and requires surgical reconstruction of the anus. After a stoma is created, except for providing instructions on how to improve sphincter function, medical institutions are rarely involved in the follow-up of these children. In children with moderate to severe anal atresia, the created anus often has poor sphincter function, resulting in problems such as fecal incontinence in school children (Diseth et al. 1998). Therefore, children with excretory dysfunction and their families are presumed to undergo a long hardship, especially during the child’s school years. A survey on the psychology of children with congenital gastrointestinal anomalies revealed that among children with anal atresia and normal intelligence, 17.9% of them had behavioral maladjustment (Tarnowski et al. 1991). A survey on psychiatric disorders using the Diagnostic and Statistical Manual of Mental Disorders III-R (DSM-III-R) (The American Psychiatric Association) revealed that 58% of children with anal atresia had some kind of psychiatric disorder (Diseth and Emblem 1996). On the other hand, another study found that there was no significant difference in social competency between children with anal atresia and healthy children (Ginn-Pease et al. 1991). Therefore, there is no general consensus on the psychosocial adjustment of children with anal atresia.

Hirschsprung’s disease is a congenital disorder in which ganglions in the wall of the colon that control its movement have not developed, and it is usually diagnosed in the neonatal period. Treatment involves surgical resection of the affected intestinal segment and reconnection of the colon. However, excision of a long segment of the intestine causes nutritional problems and excretory dysfunction. It was pointed out that severe constipation or diarrhea causes problems such as fecal incontinence in schoolchildren with Hirschsprung’s disease (Diseth et al. 1998). After the surgery, medical institutions are rarely involved in the follow-up of patients with Hirschsprung’s disease as is the case with patients with anal atresia. Psychiatric disorders as assessed by the DSM-III-R were found in 16% of children with Hirschsprung’s disease (Diseth et al. 1997). Fasten (2000) reported that treatment by not only doctors but also clinical psychologists could improve the fecal dysfunction in patients with Hirschsprung’s disease.

In this study, we interviewed children with congenital anal anomalies and their mothers to examine the psychological status of children with excretory dysfunction and investigated the relationships between the affected children and their families as well as the children’s social life. Furthermore, we examined whether psychiatric consultation is necessary for children with excretory dysfunction due to congenital anal anomalies.

**METHODS**

**Participants**

The subjects consisted of four children with excretory dysfunction due to a congenital anal anomaly who were being followed in the Department of Pediatric Surgery, Tohoku University Hospital, and their mothers. The four children were between the ages of 6 and 12 years. This survey was approved by the ethical committee of Tohoku University Medical School. They were enrolled in the study after informed consent was obtained from both the child and mother.

**Instruments**

The children with anorectal malformation were assessed by the Children’s Depression Inventory (CDI). The CDI was developed by Kovacs (1981, 1985) and is an established self-assessment type of psychological test, whose reliability and validity have been demonstrated in studies conducted in Western countries. The test is composed of 27 items, and the score on the CDI ranges from 0 to 54 points. This test was initially developed to evaluate depression in children aged seven to 17 years; an individual with a higher score is regarded as having more severe depression. Kovacs set the cut-off point for depression at 18. This cut-off score is controversial, and a cut-off score of 19 (Hodges et al. 1982b) has also been proposed. We administered the Japanese version of the CDI which was provided by MHS (Multi-Health Systems, Inc., Toronto, Canada), to the four children in the present study. Regarding the Japanese version of the CDI, the average score in healthy volunteers was 9.28 according to MHS, the cut-off point for depression was

In the four mothers in the present study, anxiety and depression were evaluated by Spielberger’s State-Trait Anxiety Index (STAI) and Zung’s Self-rating Depression Scale (SDS), respectively. Spielberger developed the STAI based on the ‘characteristic-state model of anxiety’ (Spielberger 1966). Nakazato and Mizuguchi (1982) established a Japanese version of the STAI and its reliability and validity have been examined (Tonori et al. 2001). The STAI-1 is used to evaluate the state of anxiety, and the maximum score is 80. Scores of 41 or less indicate normalcy, while scores of 42-50 imply a clinically significant state of anxiety and scores of over 50 indicate an extremely high level of anxiety (neurosis level). The STAI-2 is used to evaluate characteristic anxiety, and the maximum score is 80. Scores of 44 or less represent normalcy, while scores of 45-54 indicate clinically significant characteristic anxiety and scores of 55 or higher indicate extremely severe characteristic anxiety (neurosis level). The SDS was invented by Zung (1965) and the reliability and validity of the Japanese version have been confirmed in Japanese subjects (Fukuda and Kobayashi 1973). The maximum score on the SDS is 80 points, and the average score in control subjects, subjects with neurosis, and subjects with depression was 35 (SD = 12), 49 (SD = 10), and 60 (SD = 7), respectively (Tonori et al. 2001).

Severity of dysfunction of defecation

To assess the severity of the dysfunction of defecation in the patients with anal atresia, the “Japanese Rectum Anal Malformed Meeting for the Study score” was used. In this scoring system, the four items of defecation desire, constipation, incontinence, and soiling are evaluated, and each item is rated at one of three grades. The score of the severity of the dysfunction of defecation in the four patients with anal atresia or Hirschsprung’s disease was assigned by the pediatric surgeon of each patient.

Semi-structured interviews

Psychosocial assessment of each child was performed by conducting a semistructured interview with the child based on the Child Assessment Schedule (CAS) (Hodges et al. 1982a). The reliability and validity of the CAS have been well established (Hodges et al. 1982b). The child’s answers to the questions generate scores for the following 11 content areas of the child’s life: school, friends, activities, family, fears, worries and anxieties, self image, mood, physical complaints, expression of anger, and reality testing symptomatology. Fig. 1 shows the questions that were used to guide the interviews. In the interviews, each child was also asked questions about his/her views of the present situation regarding his/her excretory dysfunction.

Each mother was interviewed separately by a child psychiatrist and by a medical student using a slightly modified version of the Parental Account of Children’s Symptoms (PACS) (Taylor 1986). The interviews were tape-recorded, and each interview by the child psychiatrist and by the medical student lasted 20 to 75 minutes. Fig. 1 shows the questions that were used to guide the interviews. The PACS is a standardized, semistructured, investigator-based interview. The PACS score includes the interviewer’s assessment of whether the mother displays warmth or criticism towards her child. The PACS had been adapted from interviews of parents that were

| Questions regarding the life of the child: |
| School life |
| 1. How many friends do you have? |
| 2. What do you do when you play with your friends at school? |

| Home life |
| 1. Who takes care of you? How does that person take care of you? |
| 2. How do you spend your time after school? |

| Questions regarding the mother’s life and environment: |
| Mother’s life |
| 1. When did you learn that your child has fecal dysfunction due to a congenital anal anomaly? How did you feel when you heard that your child has fecal dysfunction? |
| 2. What do you do to take care of your child? If you do not take care of your child, who takes care of your child? |

| Environment around the mother |
| 1. Who has helped you when you have to take care of your child? How has that person(s) helped you? |
| 2. What feelings do you have towards families having a child with the same disease? |

Fig. 1. Guidelines for the unstructured interviews with children with fecal dysfunction due to a congenital anal anomaly and their mothers.
conducted in the expressed emotion research tradition in relation to adult psychopathology (Brown et al. 1972; Vaughn and Leff 1976).

In the interview, the mother was also asked semi-structured questions about her experiences and concerns regarding her child with congenital anal anomaly and her child’s siblings, the emotional and practical aspects of the treatment procedures, and the medical follow-up of her affected child. Whether the mother accepted the dysfunction in her child, which was presumed to influence the psychological status of her child, was assessed by the first author in view of the published paper by Hassink et al. (1998).

The interviews with each mother and child were transcribed verbatim and analyzed. The central theme that emerged was whether the mother was able to adapt to her child’s disorder. Factors that contributed to a mother’s successful adaptation were identified. The findings of this study were compared with the findings of previous studies on children with congenital fecal dysfunction and their mothers. The concepts on parents’ adaptation to a child with congenital fecal dysfunction that emerged were discussed with the families and were further modified until the investigators and a colleague found them credible.

The interview data were analyzed by a psychiatrist (S. F.) who has seven years’ experience, a pediatric surgeon (T. K.) who has fifteen years’ experience, and a medical student (J. H.).

### RESULTS

The interview data of the four children and their mothers were analyzed by a psychiatrist, a pediatric surgeon and a medical student. Table 1. shows the scores of the children and their mothers on the psychological tests.

**Table 1.** Results of psychological tests in the children with fecal dysfunction due to a congenital anal anomaly and their mothers

<table>
<thead>
<tr>
<th>Age (year)</th>
<th>Diagnosis, Surgical treatment</th>
<th>Psychological status of child (CDI score)</th>
<th>Psychological status of the mother</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
<td>STAI-I</td>
</tr>
<tr>
<td>A 6</td>
<td>High anal atresia, colostomied</td>
<td>9</td>
<td>26 31 28</td>
</tr>
<tr>
<td>B 7</td>
<td>High anal atresia, anal reconstruction</td>
<td>19</td>
<td>43 43 29</td>
</tr>
<tr>
<td>C 8</td>
<td>Intermediate anal atresia, anal reconstruction</td>
<td>8</td>
<td>29 30 33</td>
</tr>
<tr>
<td>D 12</td>
<td>Hirschsprung’s disease, Total colostomied</td>
<td>20</td>
<td>69 63 56</td>
</tr>
</tbody>
</table>
Although Child A sometimes complains about her excretory dysfunction, she fully obeys what her mother says. Child A has a sister who is three years older whom she likes, and often does things with her. On her father’s days off from work, her family often does something together. Her mother said that Child A is talkative at home and at school.

Child B. Seven-year-old girl (high anal atresia, anal reconstruction, CDI score 19).

School life of Child B: Child B is a second-grade elementary school student. She goes to school nearly every day with occasional hesitation. She sometimes has loss of fecal control and incontinence in the classroom (severity of the dysfunction of defecation, 8), and she is sometimes bullied for it. She said that she has several friends, but she often plays alone by drawing pictures during the time between classes. She is unable to talk about her disability when asked by her classmates. Her test scores at school are ranked as high.

Home life of Child B: Child B’s mother has taken care of her excretory dysfunction. Her family is fatherless and her mother alone takes care of her. She can talk about anything with her mother including bullying at school. She has been able to continue going to school with her mother’s encouragement. Child B has a much older sister whom she likes, but her sister does not take care of her very much. Her mother said that since she started going to school, Child B had become introverted and becomes depressed more easily.

Child C. Eight-year-old boy (intermediate anal atresia, anal reconstruction, CDI score 8).

School life of Child C: Child C is a second-grade elementary school student. He goes to school every day without hesitation. He sometimes has loss of fecal control (severity of the dysfunction of defecation, 6) and incontinence in the classroom. However, his classmates understand his disability to some extent and he has never been bullied. When he has incontinence at school, he disposes it in the restroom without being noticed by his classmates, and he brings extra underwear to school. After school, he often plays with his friends at their house. When asked about his disability by his classmates, he can clearly explain what he had been told by his mother. His test scores at school are ranked as slightly below average.

Home life of Child C: His mother has taken care of his excretory dysfunction. He can talk with his mother about anything that happened at school and tries to obey whatever his mother says. On his father’s days off from work, his family goes out together. His mother said that since he became eight years of age, he started talking about feeling inferior to his classmates.

Child D. 12-year-old girl (Hirschsprung’s disease, total colectomy, CDI score 20).

School life of Child D: Child D is a sixth-grade elementary school student. She goes to school every day without hesitation. She suffers from Hirschsprung’s disease and feels annoyed by severe constipation or diarrhea. She sometimes has fecal incontinence (severity of the dysfunction of defecation, 8). Because she had undergone total colectomy for treatment of the disease, her growth has been disturbed and she is shorter than her classmates. Several friends help take care of her when she needs help. In addition, she receives treatment with drip infusion eight hours every day at home due to her shortened intestine, and therefore she has to go home immediately after school. It is physically difficult for her to go to school every day. Since she has forced herself to do so, she recently became debilitated and had to be hospitalized. Her test scores at school are ranked as slightly below average.

Home life of Child D: Her excretory dysfunction is taken care of partly by her mother and partly by herself. When she does not feel well, her mother has mostly taken care of the management of drip infusion, etc. Child D rarely talks to her mother about what happened at school. Her mother is busy taking care of a much younger brother and does not have enough time to spend with her. Her father is also busy at work and is hardly involved in taking care of her excretory dysfunction. Child D is quiet and sometimes be-
comes so melancholic that even her mother becomes depressed. Child D also frequently suffers from acute abdominal cramps of unknown etiology at night.

**Interviews with the mothers**

Through the interviews, we found that each mother in this study had trained her child to defecate at home and bore a heavy burden. However, the interviews showed that each mother accepted the excretory dysfunction differently. The interviews with the mothers are described below.

**Interview with the mother of Child A.**

(STAI-I: 26, STAI-II: 31 and SDS: 28)

She accepts her child’s disability as it is. She said that since her child was born in a way that could not be fixed, she tries to solve problems together with her child at the child’s pace. However, since she is occupied with taking care of her child, she feels sorry that she does not have enough time to take care of her child’s older sister.

**Environment around the mother of Child A.**

After delivering Child A, the mother was extremely shocked by her child’s disability. Her husband and parents-in-law sympathized with her and earnestly supported her. She felt that she was blessed with good physicians and nurses as well as the relationships with families having a child with the same disease. Above all, the support of her husband who lived together with her enabled her to take good care of the child.

**Interview with the mother of Child B.**

(STAI-I: 43, STAI-II: 43 and SDS: 29)

The mother accepts her child’s disability as unchangeable. She loves her child and wants to let her child live a regular life. To that end, she wants her child to do her best so that she does not fall behind her classmates. She was overjoyed when her child won a sprint race. She wants her child to be better than other children in many things. The mother often takes a critical attitude when Child B hesitates about going to school. Her mother’s critical attitude puts Child B in a depressive mood.

**Environment around the mother of Child B.**

It took a very long time for the mother to accept her child’s disability after feeling shock at the time of delivery. Since the family is fatherless, the mother does not have a good relationship with her own mother, and she and her child have undergone the hardship together without the help of extended family members. She recollected that she did not have good relationships with the physicians and nurses at the hospital. The other families with an infant with the same disease at the hospital did not accept support from other families.

**Interview with the mother of Child C.**

(STAI-I: 29, STAI-II: 30 and SDS: 33)

The mother recently has come to accept the disability of her child as it is. Despite poor fecal control, the child has been cheerful and tries to fulfill the expectations of his mother, and the mother was able to overcome her depression. The mother recently felt that she is prepared to overcome many problems together with her child.

**Environment around the mother of Child C.**

After the delivery, she was in shock for many days and could not focus on doing anything. Her husband took a one-month leave of absence from work to support her, and she interacted with physicians, nurses, and other families having a child with the same disease. The mother was able to recover from the shock and resume her life. Her husband continues to support her, and the family often goes out together on holidays.

**Interview with the mother of Child D.**

(STAI-I: 69, STAI-II: 63 and SDS: 86)

The mother accepts the disability of her child as unchangeable. However, she wishes that the child could take care of herself. Since she has had to take care of Child D’s younger brother, she could not spend all of her time taking care of Child D. She wishes that Child D could be more independent. The mother sometimes becomes enraged with her child’s uncommitted way of excr-
Environment around the mother of Child D.

She was in shock after the delivery, but was able to recover with support from the people around her (family members, physicians, nurses, families of a child with the same disease). However, the mother has taken care of the excretory management of her child and she could not rely on her husband. Her parents lived far from her, and she struggled to endure the hardship alone.

DISCUSSION

Three of the four children with excretory dysfunction in the present study did not have good fecal control, except for Child A with a stoma, and the three children could not avoid fecal incontinence at school. Child A has to take care of the stoma at school and therefore does not have a normal school life. The CDI scores of Child A and C were in the normal range, and the CDI scores of Child B and D were just above the cut-off point for depression. The mothers of Child A and C had normal STAI and SDS scores, while the mother of Child B had significant anxiety and the mother of Child D had depression and extremely severe anxiety. Child B is occasionally truant from school, and Child D was diagnosed by the psychiatrist as having psychosomatic disease.

The mother of Child D has not been able to stop comparing her child with other healthy children and urges her child not to fall behind her classmates. She has tended to ask her child to meet impractical demands and, consequently, her child often feels depressed. The mother of Child D has tried to push her child to ‘catch up’ with other children. The mother of Child D developed a psychosomatic disease, and Child D showed symptoms of depression. Ditesheim and Templeton (1987) studied a population with high quality-of-life (QOL) among children aged 17 years or older with severe dysfunction of defecation, and their results suggested that a high level of support from the child’s family and other people around the child was associated with high QOL. Child D, who did not have much support from either parent, developed depression that required treatment by a psychiatrist.

After children with anal atresia undergo surgical treatment during the neonatal period, the level of medical care for their fecal dysfunction at a medical institution generally decreases as the child becomes older. It was thought that children with anorectal anomalies come to accept their disability little by little during their school years (Diseth and Emblem 1996). The present investigation showed a patient who did not adjust to her fecal dysfunction and developed depression that required treatment by a psychiatrist. The interview data suggested that this case may not be an isolated case, and that a particular proportion of patients with fecal dysfunction may have depression. Further studies on a much larger number of patients are needed.

References


