

Original Article

Discrepancies between Physician and Parent Perceptions of Psychosocial Problems of GHD Children Undergoing GH Therapy in Japan

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Abstract. This study examined discrepancies between the perceptions of physicians treating short children with GH deficiency (GHD) using GH replacement therapy (GHRT) and the perceptions of the parents of these children and identified the major causes of parental anxiety. Three attending pediatric endocrinologists and the parents of 31 GHD children participated in this study. The physicians and parents completed a specially designed questionnaire to rate the types and degrees of psychosocial problems that GHD children might experience. For 6 of the first 11 questions, the physicians rated psychological problems differently than the parents did, tending to over- or underestimate parental concerns. This discrepancy did not disappear with treatment. However, the difference in the perception of anxiety between the physicians and parents changed for issues regularly discussed between them. Physicians and nurses were ranked as the most reliable providers of information. The parents of children who had previously undergone GHRT were a highly desired source of information. Psychosocial problems remain largely unaddressed by endocrinologists. Endocrinologists treating short stature are encouraged to be more involved in understanding parents' anxieties, evaluation of misperceptions concerning parents' expectations, and addressing these issues in future communication with parents. Support by experienced psychologists may help endocrinologists with this issue.

Key words: short stature, psychosocial problem, discrepancy, perception, GH deficiency

Introduction

Short stature due to GH deficiency (GHD) has been reported to be associated with psychological maladjustment of affected children (1). Studies suggest that emotional maladjustment may result from aversive socialization, such as bullying, name-calling, or juvenilization by adults. According to Yokoya

(2), around 60% of GHD children evaluated in Japan have experienced some kind of bullying, such as verbal abuse (21.8%), mischief (15.8%), violence (12.9%), and being used as a toy (10.9%).

Many observers agree that regardless of any specific diagnosis, short children are at risk for misperception by others due to the difference between their actual chronological age versus

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their younger perceived apparent age because of their decreased stature. Furthermore, short children themselves may often feel smaller compared to their siblings, which makes them feel inferior, and they behave in a manner consistent with their height rather than their age (3). Consequently, it can be quite difficult for parents to avoid misguided overprotection of a short child. Many studies imply that these misperceptions may lead to immature and dependent behavior patterns, anxiety, a feeling of entitlement, and poor self-esteem in short children.

A recent study conducted in the U.S. revealed that many pediatric endocrinologists endorsed the belief that growth-delayed children suffer many emotional hardships (4). Furthermore, the authors hypothesized that physicians should certainly be receptive to the wishes and worries of parents and emotionally support any positive determinations of the benefits of GH replacement therapy (GHRT) for GHD children. As a physician may use his/her clinical judgment and previous experience with short stature treatment in addition to his/her emotional attitude to reflect parents' concerns, there is a possibility for differences between parents and attending physicians in their perception of the psychological impact of short stature and satisfaction with the benefits of GHRT.

Differences in perception of the effects of GHRT were evaluated by Grew *et al.* in a study that examined the parental expectations for GHRT versus physician's judgment of actual height and final height prediction in a group of GHD children (5). Using a silhouette technique (human figures drawn to the scale of 3, 25, 50, 75, and 97 percentiles), each patient and parent was asked to point to which figure best represented the current reality and future height expectation for the child. Physicians' predictions were based on actual measurements; however patients, and surprisingly their parents too, had

grossly overestimated expectations. Additional intervention through joint collaboration between endocrinologists, physicians, and parents improved the current and future expectations for effective treatment. The authors concluded that parents may have unrealistic beliefs for GHRT, and this causes disappointment for the future outcome. They suggested there is a need to check and reframe patients' perceptions in a more realistic way from the earlier stages of GHRT.

The issue of communication barriers and misunderstandings between physicians and patients/parents has recently been raised in the literature for general pediatrics (6). A high likelihood of miscommunication between parents and physicians, especially at the onset of a chronic disease, suggests a need for a deeper look at the well-being of chronically ill patients and improvement of communication to address parents worries (7). We took short stature as a model of chronic diseases with continuous stress factors including issues in social life and necessity of daily injections. The need for long-term communication makes this disease a good example for evaluation of the perceptions of well-being between physicians and parents in clinical settings (8).

Methods

Subjects

The parents of 31 GHD children aged 3–14 yr with a height $SD < -2.0$ were recruited for this study. The backgrounds of the 31 children and their parents are shown in Table 1. Height SDS was -2.95 ± 0.55 for the total group. There were no differences between the younger (≤ 9.0 yr) and older children (> 9.0 yr) (height SDS of -2.98 ± 0.55 and -2.91 ± 0.56 , respectively). At the time of study entry, children were receiving GHRT for the period of 43.2 ± 27.8 mo (mean \pm SD).

The children were treated at the Pediatric

Table 1 Patient backgrounds

Items		At the start of treatment	At study entry
Sex	Boy	23 cases (74.2%)	
	Girl	8 cases (25.8%)	
Age (yr)	Mean \pm SD	8.7 \pm 3.2	12.8 \pm 2.6
Body weight (kg)	Mean \pm SD	23.9 \pm 10.8	37.5 \pm 10.7
Height SDS	Mean \pm SD	-2.95 \pm 0.55 (for the total 31 cases) -2.98 \pm 0.55 (\leq 9 yr) -2.90 \pm 0.56 ($>$ 9 yr)	
GHRT period (mo)	Mean \pm SD	43.2 \pm 27.8	
Father's height (cm)	Mean \pm SD	166.8 \pm 6.3	
Mother's height (cm)	Mean \pm SD	153.6 \pm 4.8	

Department of Dokkyo Medical University, which is specialized in growth problems during childhood. The families were enrolled by three attending pediatric endocrinologists after informed consent was obtained from the parents. The questionnaire for the parents was answered by the mothers of the patients. The mother of only one patient consulted the child's father. The questionnaire for the physicians was answered by one physician with support from the others.

Methods

A specially designed questionnaire was used in this study that was created from interviews with parent focus groups evaluating the worries of daily life in GHD children. It consisted of two parts. The first part contained 11 questions for both parents and physicians and was aimed at comparing their perceptions (Table 2). These questions focused on psychosocial problems, expectations for GHRT, and treatment compliance; they were answered using ranked scales. Positive responders for question number 11, stating any degree of anxiety after the completion of GHRT, were asked to describe anxieties in a few words. Furthermore, the physicians were requested to answer the same questions by trying to place themselves in the

parent's shoes. Questions 12 to 14 collected more detailed information from parents regarding their major anxieties, reliable information sources about short stature, and the necessity of specific information about short stature and GHD for the future.

The second part of the questionnaire, which was not identical for parents and physicians, consisted of 4 questions. Parents were asked to describe their anxieties for GHRT and countermeasures in a more open way, while physicians were requested to describe worrisome points in a short text. These questions were rewritten for parents in easily understandable Japanese sentences.

Differences in the scores between the parents' and physicians' perceptions for the first 11 questions were analyzed by means of the Mann-Whitney Test, with $p < 0.05$ as significance level.

Parents were asked to fill out the questionnaire during a visit to endocrinologist (primary physician involved in the study). All 31 patients had already been treated with GH prior to study entry, and baseline data were received retrospectively.

Table 2 Contents of the questionnaire

Select the most appropriate answer from the following 1–5:

1. Untrue
2. Not necessarily true
3. Unsure
4. More or less true
5. True

Questionnaire	Selection
1. Your child has a normal life and is not bullied.	1, 2, 3, 4, 5
2. Your child has no inferiority complex.	1, 2, 3, 4, 5
3. Society/schools pay attention to short stature.	1, 2, 3, 4, 5
4. If GHRT is effective short stature is not handicap.	1, 2, 3, 4, 5
5. You think your child is not pitied and want him to use his short stature as a springboard for success.	1, 2, 3, 4, 5
6. Short stature has no influence on friendship.	1, 2, 3, 4, 5
7. You do not overprotect your child.	1, 2, 3, 4, 5
8. Injections are performed according to schedule.	1, 2, 3, 4, 5
9. You have strong expectations for the efficacy of GHRT.	1, 2, 3, 4, 5
10. You have no anxiety about long-term treatment.	1, 2, 3, 4, 5
11. You have no anxiety after completion of treatment.	1, 2, 3, 4, 5
12. What makes you most anxious?	
1. Nobody to consult	
2. Injections (Can I give them without making a mistake?)	
3. Adverse drug reactions	
4. Expense	
5. Other people notice that my child has a disease	
6. Daily injections are not possible	
7. Child's life at school (bullying, etc.)	
8. Nothing in particular	
9. Other ()	
13. What is the most reliable information source?	
1. Information from the internet	
2. Peer to peer communication (patients' groups) (including communication via the internet)	
3. Physicians and nurses	
4. Brochures, books, pamphlets, videotapes, etc.	
5. Other parties involved in patient care (including pharmaceutical companies)	
6. Other ()	
14. What makes you most anxious about daily injections?	
1. Pain	
2. Injections given by us	
3. The possibility of mistakes made in the procedure	
4. No visible effect of the drug	
5. Nobody to ask if I have problem	
6. Accidents with needles	
7. Injection of drugs with air bubbles	
8. Infection at the injection site	
9. Nothing in particular	
10. Other ()	
15. Desirable information in the future	
1. The most up-to-date medical information	
2. The present status of patients who have undergone GHRT previously	
3. Nothing in particular	
4. Other ()	

GHRT: GH Replacement Therapy

Table 3 Questionnaire results: Comparison of parents' and physicians' answers before the start of GHRT¹⁾

Question	Answer	Mean Score \pm SD	Difference ²⁾
1 Your child has a normal life and is not bullied.	Parents Physicians	4.5 \pm 0.7 4.9 \pm 0.2	p=0.0088
2 Your child has no inferiority complex.	Parents Physicians	3.5 \pm 1.3 4.6 \pm 0.6	p<0.0001
3. Society/schools pay attention to short stature.	Parents Physicians	2.4 \pm 1.2 1.9 \pm 0.8	p=0.1527
4. If GHRT is effective short stature is not handicap.	Parents Physicians	3.8 \pm 1.2 3.2 \pm 1.1	p=0.0216
5. You think your child is not pitied and want him to use his short stature as a springboard for success.	Parents Physicians	3.8 \pm 1.1 4.1 \pm 0.9	p=0.4196
6. Short stature has no influence on friendship.	Parents Physicians	4.2 \pm 1.3 3.5 \pm 0.8	p=0.0004
7. You do not overprotect your child.	Parents Physicians	3.0 \pm 1.5 4.6 \pm 1.0	p<0.0000
8. Injections are performed according to schedule.	Parents Physicians	4.5 \pm 0.8 4.2 \pm 0.8	p=0.0274
9. You have strong expectations for the efficacy of GHRT.	Parents Physicians	4.4 \pm 0.7 4.3 \pm 0.5	p=0.3844
10. You have no anxiety about long-term treatment.	Parents Physicians	3.5 \pm 1.3 3.7 \pm 0.7	p=0.9559
11. You have no anxiety after completion of treatment.	Parents Physicians	3.2 \pm 1.4 3.5 \pm 0.9	p=0.5073

¹⁾Answers received retrospectively. ²⁾Mann-Whitney test.

Results

Table 3 summarizes the answers by parents and physicians for the first 11 questions. In regard to the 3 questions concerning the patients' lives, i.e. no bullying (Q1), no inferiority complex (Q2), and the parents' normal attitude toward their children without being overprotective (Q7), the affirmative ratings for the scores by the physicians were significantly higher than those of the parents (p<0.01); however, the parents' affirmative ratings were significantly higher (p<0.01) when answering the question regarding their children's friendships (Q6). This implied that there may be some important events in the daily lives of GHD children

that can be perceived only by their parents. Physicians might address this issue with parents during a follow-up visit. Significant differences (p<0.05) were recognized between the answers for the 2 questions concerning the GHRT, i.e. short stature is not a handicap as long as GHRT is effective (Q4) and compliance with daily GH injections (Q8). Predictably, the scores of the parents' answers to the former question were higher than those of the physicians, which may imply parental overestimation of GHRT. On the other hand, the physicians have some degree of distrust of patient compliance.

The raw data for the scores before and after treatment are shown in Fig. 1.

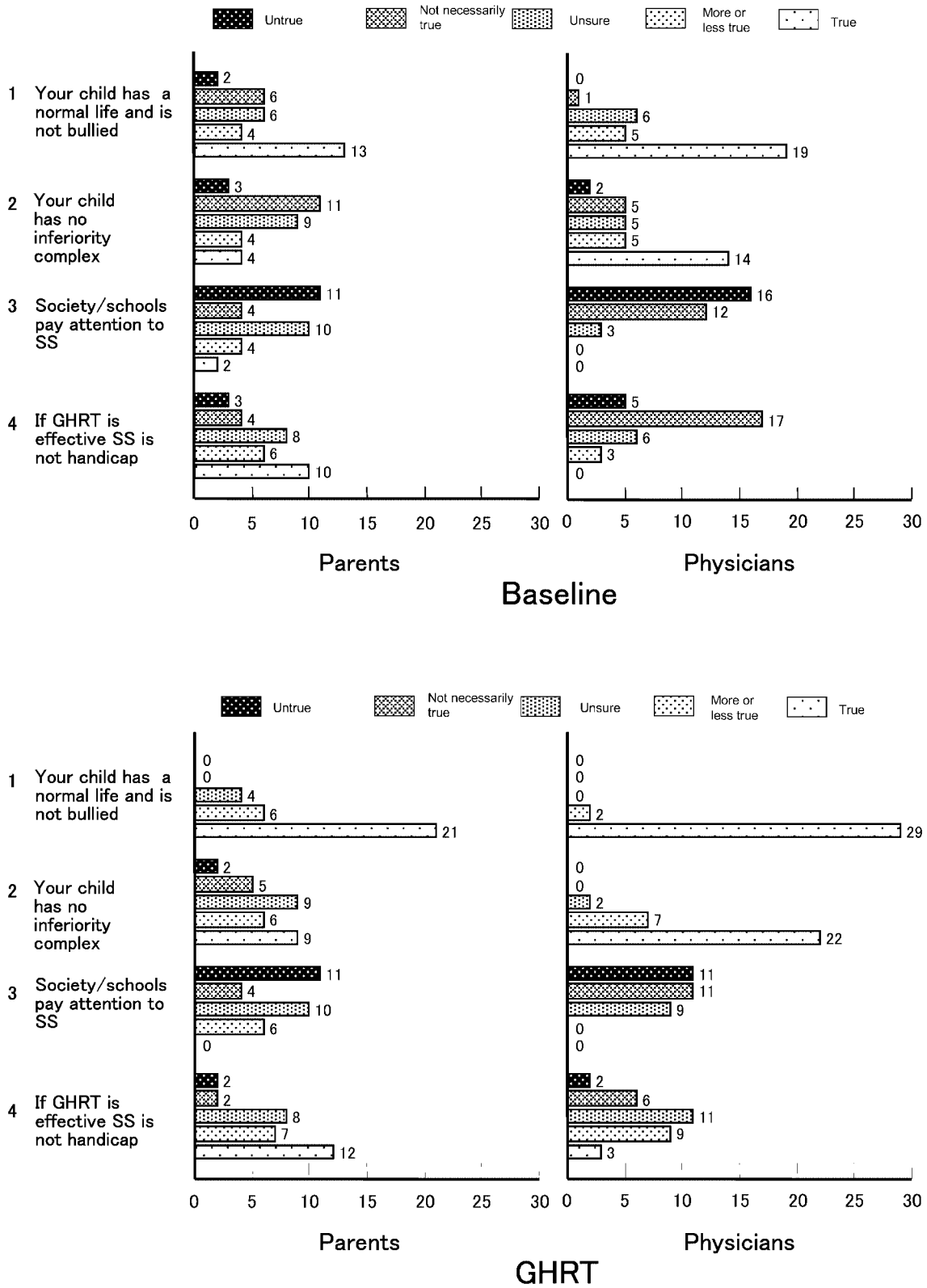


Fig. 1 Raw score items at baseline and after GHRT. SS: Short stature.

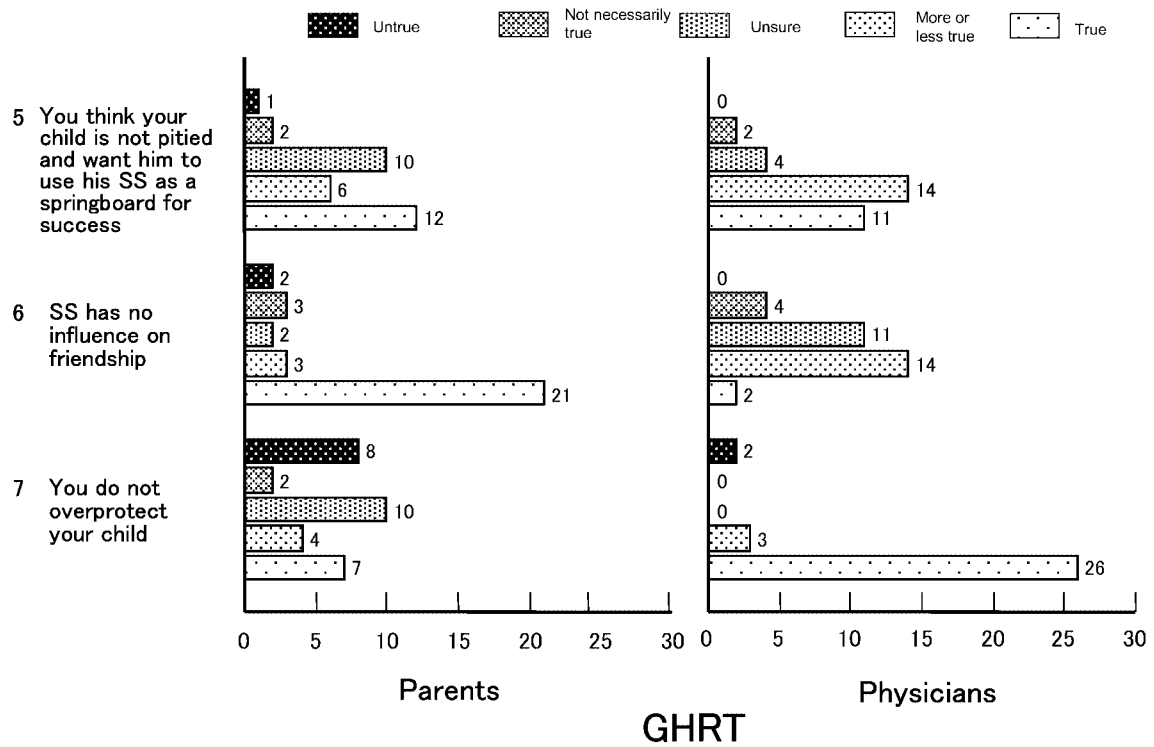
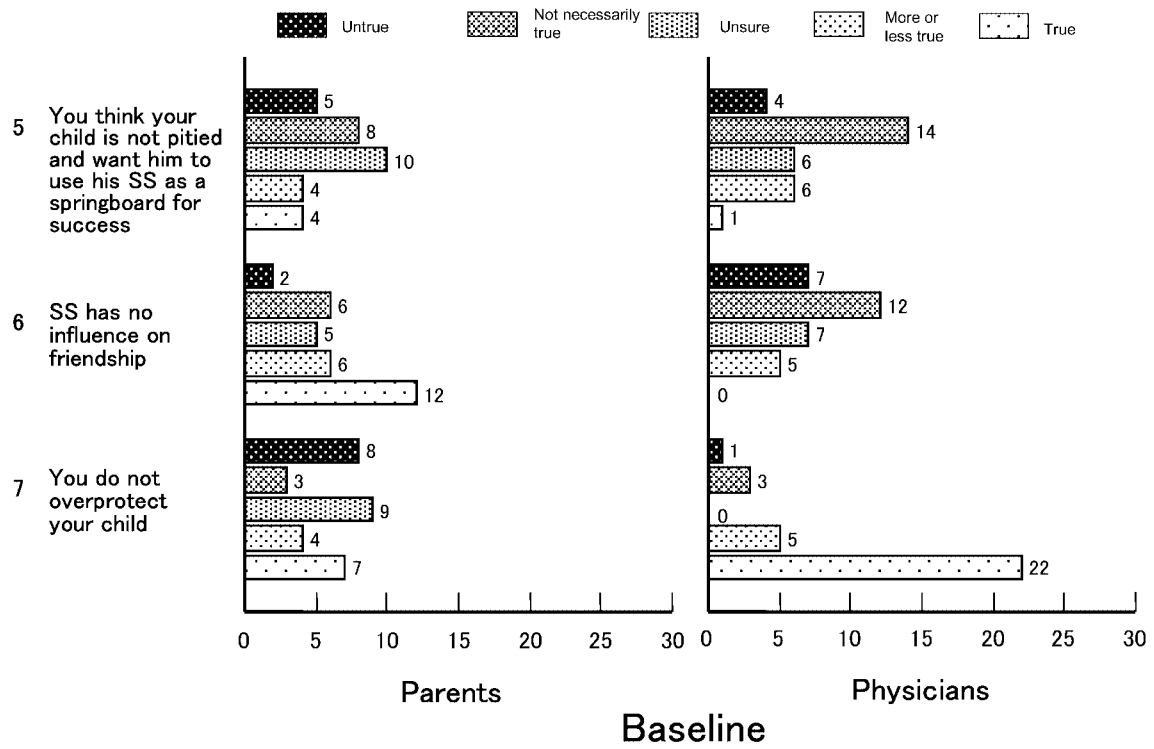


Fig. 1 Continued-1.

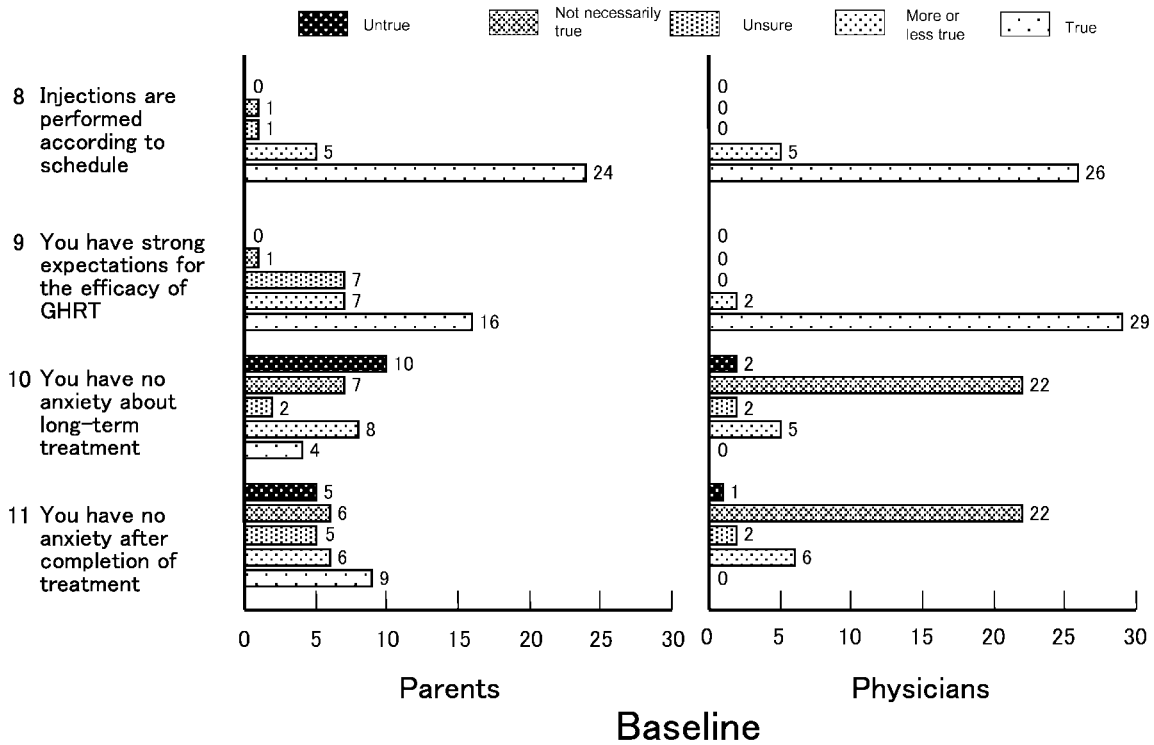
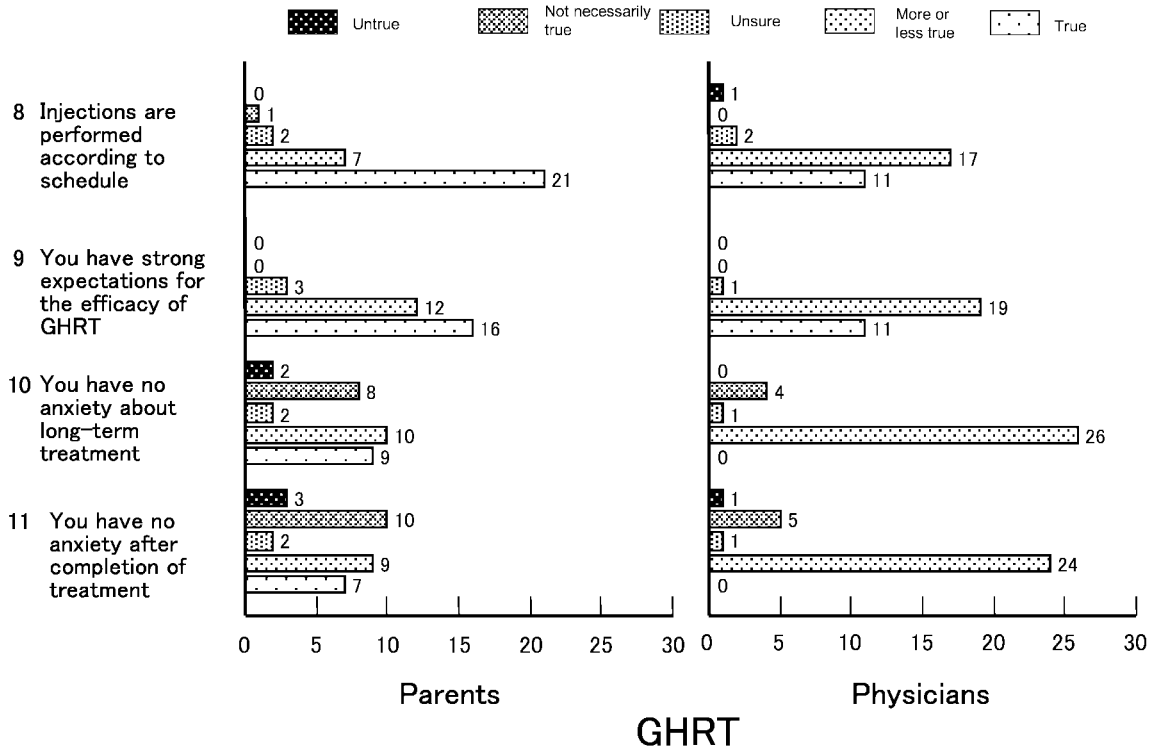


Fig. 1 Continued-2.

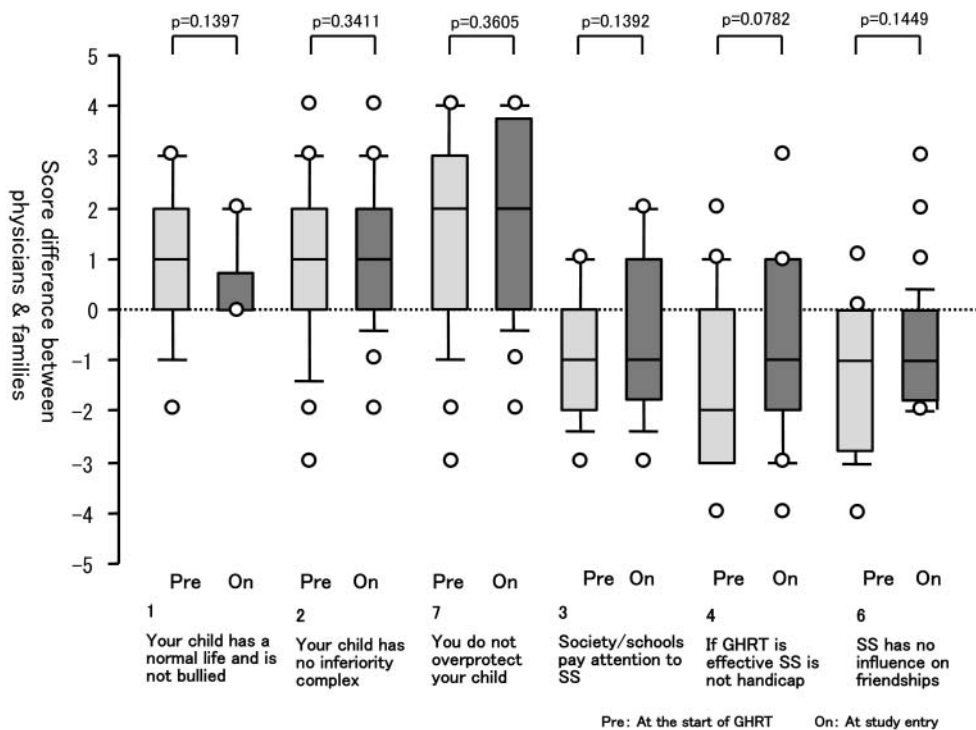


Fig. 2a Physicians perceive psychological problems differently than parents and tend to overestimate (questions 1–3) and underestimate (questions 4–6) parents’ worries. This discrepancy did not disappear on treatment. The top and bottom lines of the boxes show the 25th and 75th percentiles, and the middle line shows 50th percentile. Each end of the vertical bars shows the 10th (top) and 90th percentiles (bottom) respectively. ○ shows distribution of scores that were not between the 10th and 90th percentiles. SS: Short stature.

As the primary purpose of this paper was to investigate the similarities and differences in perception between physicians and parents in the psychosocial and treatment outcomes of GHD children, we used a delta score for each question in part 1 and compared the data from before GHRT (baseline) to the current GHRT data (on GH).

The pattern of change for the median delta scores allowed us to split the questions into three groups as follows:

- 1) Presence of over- or underestimations (positive or negative delta scores) without statistically significant changes in treatment, including questions related to the social lives

of the patients (Fig. 2a).

- 2) Absence of over- or underestimations (median delta scores equal to zero) without statistically significant changes in treatment, including questions related to anxieties for GHD itself and GHRT (Fig. 2b).
- 3) Delta scores with significant ($p < 0.05$) changes in treatment, including questions related to compliance and expectations for GHRT (Fig. 2c).

The similar opinions were confirmed for the physicians and parents for 2 of the first 11 questions, i.e. anxiety for long term treatment and the wish to use short stature as a springboard to success (Fig. 2b). For 6 of the first 11

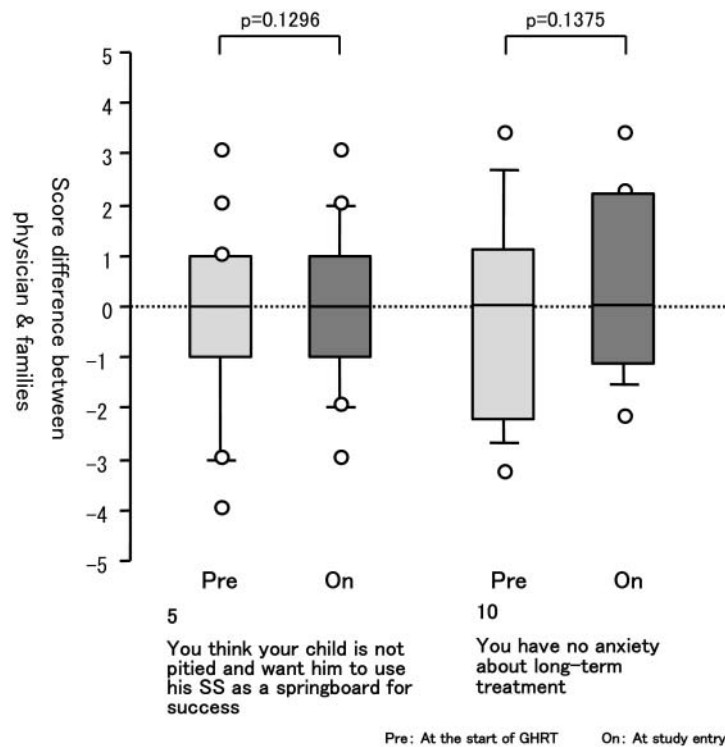


Fig. 2b Patients and physicians perceive similar anxiety for long term treatment and express similar encouragement for lifelong success for GHD children. The top and bottom lines of the boxes show the 25th and 75th percentiles, and the middle line shows 50th percentile. Each end of the vertical bars shows the 10th (top) and 90th percentiles (bottom) respectively. ○ shows the distribution of scores that were not between the 10th and 90th percentiles. SS: Short stature.

questions, physicians either overestimated or underestimated parents' worries. All questions were related mainly to psychosocial problems (Fig. 2a) those were not improved by treatment. The perceptions for 3 questions changed at the start of GH therapy (Fig. 2c). There were no discrepancies between the physicians' and parents' expectations for the question concerning injections being performed every day before the start of GH treatment (median=0). However, during therapy, physicians slightly, but significantly, underestimated compliance with daily injections ($p=0.0102$) Prior to GH therapy, physicians slightly overestimated parental

response to the question asking them to rate their expectations for GH therapy, but this difference decreased to zero ($p=0.0003$) at the start of therapy. Physicians underestimated parents' anxiety after stopping GH treatment at the starting point of GH therapy. However, communication with parents during GHRT improved the physicians' understanding of the parents' anxiety (mean change for delta score increased from -1 to 0).

Analysis of the 2nd part of questionnaire is shown in Table 4a. While 7 parents (22.6%) selected "Nothing-particular". Nine parents (29.1%) pointed out "Adverse drug reactions" as

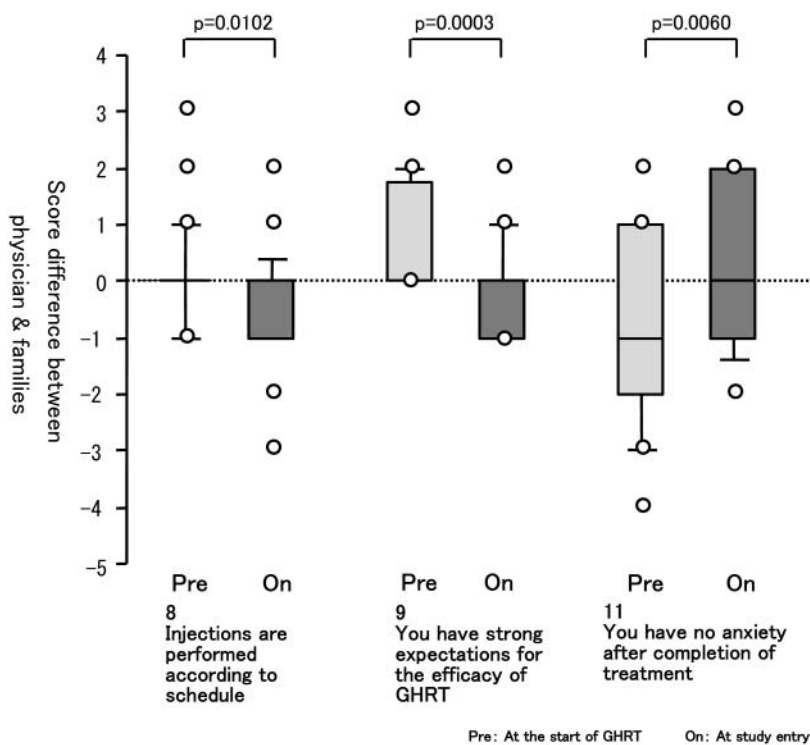


Fig. 2c Observation during GHRT changed perception of compliance, expectations for the drug, and anxiety after GH discontinuation between physicians and parents. The top and bottom lines of the boxes show the 25th and 75th percentiles, and the middle line shows 50th percentile. Each end of the vertical bars shows the 10th (top) and 90th percentiles (bottom) respectively. ○ shows the distribution of scores that were not between the 10th and 90th percentiles. SS: Short stature.

a major source of anxiety. This was followed by “Child’s life at school”, including bullying (4 parents, 12.9%), “Nobody to consult”, and “Daily injections are not possible” (3 parents, 9.7%, each).

For the answers to the latter question regarding anxieties due to daily GH injections, 9 parents (29.1%) indicated “Pain” and 4 parents (12.9%) pointed out “Infection at the injection site”. Although 12 parents (38.7%) answered “Nothing particular” before the start of GH therapy, the same answer was only obtained from 2 parents (6.5%) in the same cohort when GHRT

was started.

For question number 13, which asked “What is the most reliable information source?”, 27 parents (87.1%) and 4 parents (12.9%) indicated “Physicians and nurses” and “Information from the internet”, respectively (Table 4b).

As for desirable sources of information (question 15), 23 parents (74.2%) and 7 parents (22.6%) quoted “The present status of patients who have undergone GHRT previously” and “The most up-to-date medical information”, respectively (Table 4c).

Table 4 Parents' anxieties and needs for information at present and for the future

a. Major anxieties

Anxieties	n (%)
1. Adverse drug reactions	9 (29.1%)
2. Injections (Can they be performed without any mistakes?)	1 (3.2%)
3. Nobody to consult	3 (9.7%)
4. Expense	1 (3.2%)
5. Disclosure of disease to other people	1 (3.2%)
6. Injection everyday without fail is difficult	3 (9.7%)
7. Child's life at school	4 (12.9%)
8. Real efficacy of GHRT	0
9. Influence on the next generation	1 (3.2%)
10. When GHRT can be terminated	1 (3.2%)
11. Nothing in particular	7 (22.6%)

b. Reliable information sources

Sources	n (%)
1. Physicians and nurses	27 (87.1%)
2. Information from the internet	4 (12.9%)
3. Acquaintances and children with the same disease	0
4. Other medical staff	0

c. Desirable information in the future

Information	n (%)
1. The most up-to-date medical information	7 (22.6%)
2. The present status of patients who have undergone GHRT previously	23 (74.2%)
3. Nothing in particular	1 (3.2%)

Discussion

Communication between physicians and patients is an important component of medical care. The value of effective communication increases in lifelong treatment of rare diseases. In this case, parental collaboration and understanding have a major impact on treatment compliance and long-term outcome in affected children. GHD short stature is a good example of a disease requiring special attention from both parents and physicians. We evaluated the similarities and discrepancies in physicians' and parents' perceptions of anxieties for GHD short

stature and GHRT. According to the baseline data, more parents than physicians considered short stature not to be a handicap as long as GHRT is effective, which may imply parental overestimation towards a final positive effect of treatment. Contrarily, the physicians' ratings were significantly lower than those of the parents regarding compliance with daily injections, which implies that the physicians had some degree of distrust of patient compliance.

Despite our findings are limited due to retrospective design, there was a significant rate of misperceptions (6/11) between endocrinologists and parents that did not improve

during GHRT. Physicians tended to over- and underestimate parents' worries related to different aspects of the patients' social lives. The results of this survey showed us that patient and parent well-being is basically out of the scope of endocrinologist's attention during examinations and talking with parents. Further prospective research using similar easy to understand questionnaires answered by parents and evaluated by endocrinologists will be helpful for doctors to better address parental anxiety.

Diagnosis of chronic diseases in children leads to dramatic changes in family life. Initial parents' worries are usually not specific and may not be well understood by health care professionals. Structured disease-specific assessments of parents' worries will greatly contribute to better understanding between pediatricians and parents, and enhance treatment compliance and satisfaction (8). Detection of a significant misperception for specific items will be a signal for a follow-up talk with parents. Pediatric psychologists trained to understand the problems of GHD children or trained nurses can also be involved at this point (9).

If we apply the results of other studies (7, 10) showing improvement of compliance with treatment after addressing misunderstandings between parents and physicians to our findings, we should advise endocrinologists to restructure their communication with parents in order to address major parents' worries effectively. Despite the fact that endocrinologists may not be as well trained as psychologists in addressing parental anxiety, general communication skills (being precise, asking open questions rather than giving orders, showing a less paternal attitude and not acting bossy, expressing general empathy) can make a significant difference in patient compliance and satisfaction (6, 11).

Close communication with a psychologist would be a desirable solution to correct communication between endocrinologists and parents. However, this type of care specialist is

not clearly defined in Japan at present. There is a very limited number of trained psychologists who have a history of long collaboration with endocrinologists in taking care of patients with different endocrine disorders, including short stature. Experience overseas has shown that clinical psychologists working closely with endocrinologists caring for patients with short stature can help to address patients'/parents' concerns properly and cover the gap of misunderstandings between parents and endocrinologists.

Daily GH injection is a disease-specific factor affecting the well-being of GHD children and their families. The necessity to maintain compliance with daily GH injections is a well-known barrier between endocrinologists' and patients' understanding because the job of the former is to insist on 100% compliance and the dilemma of the latter is to take drug holidays. Daily injections were a significant source of anxiety that was not recognized by parents before the start of GHRT in our study. Anxiety for daily injections during treatment was expressed by 10 of 12 patients in the group without any worries being expressed before treatment.

The desire for high compliance was detected before the start of GHRT; however, this parameter decreased after three years of treatment (Fig. 2c, question 1). The raw data (Fig. 1) shows that patients did not confirm incompliance (no change from baseline), but the physician's opinion for patient compliance decreased (26/31 cases were judged as noncompliant at the beginning, but this number decreased to 11/31 during treatment).

The physicians' perception of decreased compliance can be explained by the possibility of no reported drug holidays and decreased prediction of the GHRT effect due to the start of puberty. The issue of taking drug holidays in relation to GHRT is well known. Several authors have reported that the level of incompliance may be quite high (41–91%) (12) and refer to the

fear of injections as the major cause.

Development of an alternative GH delivery system (e.g. inhalation or oral) that does not require daily injections or long acting formulations may improve compliance with GHRT in the future. However, one should not underestimate the simple need for better communication with patients, clarifying and reframing patients' expectations and concerns. The results of this study show that there is a clear need for further development of collaboration between physicians and parents for treatment of GHD children in Japan.

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