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# The earlier, the better? Diagnostic experiences of parents in a community-based early intervention system for preschool children with autism

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## Abstract

**Background and aims:** In many countries, early detection and diagnosis of autism spectrum disorder is largely dependent on parents' initial concern with early symptoms of autism spectrum disorder. Previous research on parental perceptions of the autism spectrum disorder diagnostic process indicates that parental satisfaction may be due to either the timing of the diagnostic notification or the provision of post-diagnostic support. The objective of this research is to study the diagnostic notification process and its impact on parents who are informed of their young child's diagnosis before they notice a problem and whose child undergoes early intervention therapy.
**Methods:** Eighty parents of preschool children diagnosed and undergoing early intervention for autism were surveyed to examine their experience of the diagnostic disclosure process.
**Results:** Of 68 respondents, 39 (58.2%) approved of the timing of diagnostic notification, while 10 of 13 dissatisfied respondents indicated that the diagnosis was communicated too late. However, there was no correlation between a higher degree of parental satisfaction with the diagnostic notification process and earlier timing of notification.
**Conclusions:** Although it is preferable to communicate a diagnosis of childhood autism as soon as possible, findings suggest that a highly individualized approach, allowing a degree of latitude in the timing of notification, may be permissible, depending on the individual case and parental readiness to receive the diagnosis.
**Implications:** Findings have clinical implications related to the concept of optimality of diagnostic disclosure as related to the diagnostic notification process, though later notification tends to lead to more dissatisfaction.

## Keywords

Autism, early diagnosis, parents, optimality in disclosure of the diagnosis

## Introduction

In recent years, new challenges have emerged as research has focused more on evidence-based practices of early intervention for children with autism spectrum disorder (hereafter ASD) who are less than three years of age (Carter et al., 2011; Dawson et al., 2010; Kasari, Gulsrud, Wong, Kwon, & Locke, 2010; Rogers, Estes, et al., 2016; Vivanti, Dissanayake, & The Victorian ASELCC Team, 2016). Although these studies differ in approach and scope, they all emphasize parental involvement in specialized therapy (Bradshaw, Steiner, Gengoux, & Koegel, 2015; Zwaigenbaum, Bauman, & Choueiri, 2015), and healthcare

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professionals must have an appropriate process for informing the parents of a child's diagnosis. With early interventions occurring at increasingly younger ages, parents must confront the reality of their child's disability more quickly than before. This means that specialists may recognise the developmental and behavioural characteristics of autism in a child even before the parents do. In accordance with National Institute for Health and Care Excellence (NICE, 2011) guidelines, specialists need to have in place a diagnostic disclosure process to convey the diagnosis to the parents without delay, while being sensitive to parental concerns.

In many countries, early detection and diagnosis of ASD is largely dependent on parents' initial concern with early symptoms of ASD. Research indicates that parental concern with a child's developmental atypicalities occurs at varying ages, but on average between one to two years of age (Brogan & Knussen, 2003; Chamak, Bonniau, Oudaya, & Ehrenberg, 2011; Crane, Chester, Goddard, Henry, & Hill, 2016; Howlin & Moore, 1997; Siklos & Kerns, 2007). Subsequently, parents will consult their general practitioner or health visitor (community health nurse), but according to degree of parental concerns, considerable variation exists between parents who take immediate action and those who do not (Ryan & Salisbury, 2012). Furthermore, even when a consultation occurs, a diagnosis is not immediately forthcoming. Parents may be referred to and consult with multiple specialists before they finally receive a diagnosis (Crane et al., 2016; Goin-Kochel, Mackintosh, & Myers, 2006). The time lag between initial diagnosis varies widely between cases and influences the treatment a child may receive; children who are diagnosed later may not have the opportunity to participate in early intervention programme.

Previous research on parental perceptions of the ASD diagnostic process indicates that both the age of the child at the time of diagnosis and length of diagnostic delay were negatively correlated with parental satisfaction with the overall diagnostic process (Brogan & Knussen, 2003; Chamak et al., 2011; Goin-Kochel et al., 2006; Howlin & Moore, 1997; Renty & Roeyers, 2006; Siklos & Kerns, 2007). Today the situation has not changed much. This is compounded by parents of children with ASD being less satisfied with the support they are offered post-diagnosis (Crane et al., 2016; Rogers, Goddard, et al., 2016). Using a qualitative methodology with three key stakeholder groups – autistic adults, parents of children on the autism spectrum, and professionals involved in autism diagnosis, Crane et al. (2018) also reported, 'it is essential to identify ways to better support them during all stages of the process – from when the early signs of autism are first noted, through the diagnostic process itself, to the provision of post-diagnostic support'.

The question is whether parental satisfaction with the diagnostic process is greater when the diagnosis is made at an earlier age, even when early intervention is provided in a timely manner in very early childhood. Research to date indicates statistically significant evidence showing that compared to parents who are informed earlier, parents informed later are more dissatisfied with the diagnostic notification process. The studies focused on parents, both early and late groups, whose concern with their child's problem was the stimulus for seeking consultation with a healthcare professional. Among parents who became aware of their child's problem and pursued a frustratingly circuitous path to reach a final diagnosis, there is a statistically significant difference in the degree of satisfaction with the diagnostic notification process of parents notified earlier compared to parents notified later at 10 or more years old, or in some studies, in adulthood (Brogan & Knussen, 2003; Chamak et al., 2011; Crane et al., 2016; Goin-Kochel et al., 2006; Howlin & Moore, 1997; Renty & Roeyers, 2006; Siklos & Kerns, 2007). However, when the final diagnosis occurs at a very young age, the situation is more complex. Symptoms of ASD first appear in toddlers, but the younger the child, the more the diagnostic uncertainty, and even though parents become aware of a problem, this is not equivalent to realizing and accepting that their child has a disability (Honda & Shimizu, 2002). This is often a frustrating period of uncertainty for parents (Ryan & Salisbury, 2012).

Parental satisfaction with the diagnostic process will differ when a professional discovers the problem and gently guides parents to the diagnosis compared to when, as is common in many countries, parents identify the problem first, then have difficulty finding appropriate professional help. To our knowledge, there has never been a study of parental perceptions of the diagnostic process in cases in which healthcare professionals' observations led to early detection before parents became aware of the problem. Accordingly, it is unknown whether parental satisfaction would be high in cases in which the diagnostic process begins when the healthcare professional first notices a child's problem and communicates this to the unsuspecting parents while the child is still young. To clarify this issue, research is needed focusing solely on cases in which healthcare professionals are first to identify a problem and convey the diagnosis to parents in the child's early years.

Consideration must also be given to the following methodological problems that affect previous research on parental perceptions of the ASD diagnostic process. First, the studies all rely on only parents' retrospective

memory (Brogan & Knussen, 2003; Chamak et al., 2011; Crane et al., 2016; Goin-Kochel et al., 2006; Howlin & Moore, 1997; Renty & Roeyers, 2006; Siklos & Kerns, 2007). The past is informed by current circumstances and is continually reinterpreted, and so relying solely on retrospective data limits the degree of clarity of recalled experiences. To objectively study parental perceptions regarding early detection and intervention for preschool children with ASD, a setting is required in which healthcare professionals can engage parents early, before parents are aware of a problem, and maintain a continuity of observation throughout the process. Also, because parental perceptions may be affected by their interactions with healthcare specialists, a more accurate grasp of changes in parental perceptions is made possible by inquiring in real time about parents' feelings about these interactions.

A second drawback is that survey response rates are typically low (Brogan & Knussen, 2003; Howlin & Moore, 1997; Renty & Roeyers, 2006; Siklos & Kerns, 2007) or undisclosed (Chamak et al., 2011; Crane et al., 2016; Goin-Kochel et al., 2006). Generally, when surveys inquire about customer satisfaction with services and the customer response rate is low, the results are skewed by selection bias and there is no guarantee that they reflect the entire group. To raise the validity of survey findings, it is necessary to make great efforts to ensure that a high response rate is obtained.

The Japanese city of Yokohama has an advantage with regard to facilitating the ASD diagnostic process (Figure 1). Yokohama, a large metropolis of 3.7 million people, is divided into nine regions, each of which is governed by one public habilitation centre. In the region covered by the Yokohama Rehabilitation Center (YRC), the community developmental disability Detection and Intervention System in the COmmunity for VERy Young children with developmental disorders (DISCOVERY) system (Honda & Shimizu, 2002) was created in close cooperation with community health and educational institutions. The health checkup for 18-month-olds, conducted at public health and welfare centres, serves as the basis for a highly accurate initial screening with a sensitivity rate of 81% for childhood autism (International Statistical Classification of Diseases and Related Health Problems, 10th Revision (ICD-10) Diagnostic Criteria for Research) and a specificity of 100% for all developmental disorders (Honda et al., 2005; Honda et al., 2009). The 18-month screening is conducted by public health nurses who are highly trained to provide comprehensive support for children and parents, including early identification of developmental disorders, and who, while advising on specific aspects of child-raising, also raise parental awareness of characteristics of child development (Honda et al., 2009). This community system, which identifies nearly all children suspected of childhood autism in their early childhood (by age 3), refers them promptly to YRC to be examined by a child psychiatrist specializing in developmental disabilities. In the examination, the psychiatrist makes a diagnosis based on details of the child's developmental history, and upon explaining the child's condition to the parents, recommends an early intervention programme.

The objective of this research is to study the diagnostic notification process and its impact on parents who are informed of their young child's diagnosis before they notice a problem and whose child undergoes early intervention therapy. The research questions of this investigation are as follows: (1) average age of diagnosis, (2) time at which the diagnosis was communicated, (3) parental satisfaction with time of notification, (4) how parents received the diagnosis, and (5) the gap between parents and child psychiatrists regarding

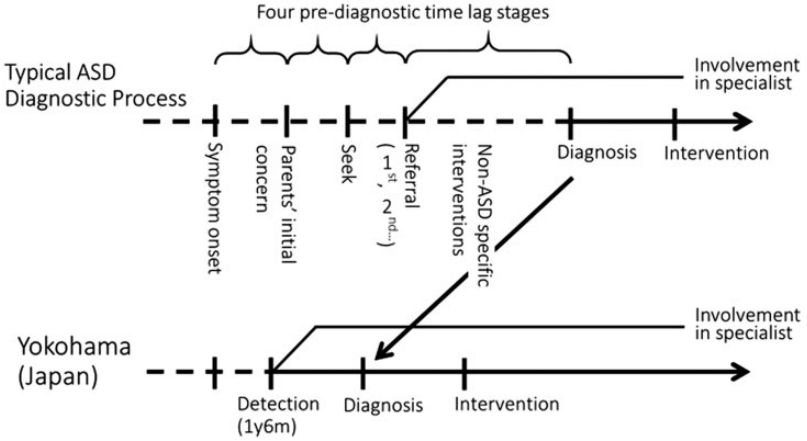


**Figure 1.** Early diagnostic process in Yokohama (Japan) compared to typical pattern.

the diagnostic notification. Although this study mainly focuses on the relationship between the timing of diagnostic notification and parental satisfaction, it also examines parental satisfaction with the notification process. We anticipate that the global trend toward increasingly earlier diagnosis and intervention with parental involvement in cases of childhood autism, aided by routine and robust community-based systems for early detection and intervention, will lead to an increase in professional – parent interactions in which specialists who have identified a child's developmental problem must inform the parents and elicit their cooperation in their child's intervention programme. The knowledge gained in this research will provide a valuable resource of basic empirical data that will shed light on this very sensitive aspect of the diagnostic process of ASD.

## Methods

### *Participants*

The subjects of this study were children with autism who were born between 2 April 1995 and 1 April 1999. They all resided in Yokohama's Kohoku Ward, which is under the jurisdiction of YRC. Ethical approval for the study was obtained by Research Ethics Committee within the Department of Developmental Psychiatry at YRC. All participants gave their informed consent to participation, prior to completing the inventory. Inclusion criteria were as below:

1. They were diagnosed with pervasive developmental disorders (PDDs) (International Statistical Classification of Disease and Related Health Problems, 10th Revision (ICD-10) Diagnostic Criteria for Research) by YRC developmental psychiatrists. This was their first clinical diagnosis.
2. They participated in an early intervention programme (4 hours a day, at least two days a week for over one year, in groups of six children to two therapists) at YRC until they entered school.

One hundred and fifty children met the inclusion criteria 1. They all underwent the introductory programme. This programme connected the first appointment with a child psychiatrist to enrol in an early intervention programme. Through the programme which offered in-depth assessment of them and discussion with their parents, 80 children met the inclusion criteria 2. Another 70 children born in the same ward during the same period were diagnosed with PDDs by YRC developmental psychiatrists, but were excluded from the study because they did not participate in the early intervention programme. Nineteen children participated in once-a-week programme for high-functioning ASD. Four participated in a speech therapy. The other children were enrolled in nursery school or kindergarten and followed about once in three months by YRC child psychiatrists and clinical psychologists.

The medical records of 80 subjects were examined to explore diagnostic process, when parents were notified of their child's diagnosis by YRC psychiatrists, if their condition had been identified at the 18-month checkup, if they had dropped out of the follow-up system between identification of the child's condition and receipt of diagnosis because of lack of need for consultation, or if the parents proactively sought out professional advice. When parents were notified of their child's diagnosis, the details were recorded in the medical record, including time of appointment, to whom notification was given, and in what manner it was communicated. A variety of the parents' status including ethnicity, age, educational status, jobs, and mental disorder were examined. With respect to children, cognitive levels based on Tanaka-Bintet intelligence test (the standardized Japanese version of Stanford-Binetintelligence test) at six-year-olds were examined. Their siblings were examined to be diagnosed with PDDs.

### *Parent Satisfaction Inventory*

At the conclusion of the early intervention programme the parents were asked to complete the Parent Satisfaction Inventory. The Satisfaction Inventory is comprised of three main components (see Appendix 1).

*Part 1 – 'Diagnostic process'.* Parents were asked about age at their child's diagnosis (requested to fill in 'X years Y months'), diagnostician's name, location, and those present at the time of diagnosis.

*Part 2 – 'Satisfaction with the diagnostic process'.* Using five-point Likert scales ('dissatisfied' to 'satisfied'), parent indicated their satisfaction with the timing and explanation of the diagnosis and adequacy of information about services. If they answered 'dissatisfied' or 'somewhat dissatisfied' to this question, they were asked to explain their reasons.

*Part 3 – 'Content of diagnostic notification and how the diagnosis was received'.* Parents were asked what the psychiatrist communicated about the child's diagnosis. In addition, parents were asked optimal open-ended questions to describe how they felt when they received their child's diagnosis.

Parents were asked to fill out the Satisfaction Inventory questionnaire at home and send it back to YRC. The intent was to gather parents' unfettered

opinions, including criticisms or reservations about provision of services, by allowing them to fill out the questionnaire outside the institutional setting where they might feel freer to give their honest opinions. Responses of the satisfaction inventory were received from 68 of 80 participants (85% response rate). Of these, 67 were from mothers and one from a father.

### Statistical analysis

The following variables were compared using a chi-square test: children's cognitive level and ASD symptomatology (childhood autism or the other ASD), and parents' nationality, age, educational status, jobs, and mental disorder. To find the proportion of parents satisfied with the time of diagnosis, satisfaction rates were calculated by ordering the children by age at diagnostic notification, from youngest to oldest, into quartiles (from Groups 1 to 4). We compared the population rate of degree of satisfaction between each group, using McNemar's test. We used the chi-square test to analyse the relationship between diagnostic process and timing of diagnostic notification. All data analyses were performed using SPSS for Windows software, version 15.0 J (IBM Japan, Tokyo, Japan), and the significance level was set at $p < .05$ for all tests.

## Results

### Average age of diagnosis

When the diagnoses were communicated, the average age of the 68 participants who participated in the early intervention programme was 41.6 months (SD = 7.0, range 26–61). All 68 children who participated in the early intervention programme were suspected of having PDDs at their first appointment with a child psychiatrist. For the parents of 15 (22.1%) children, the psychiatrist described the child's developmental issues as autism and advised parents on managing their child-care challenges at the first appointment. All 15 children were entered without delay into the introductory programme, and at the second appointment, the psychiatrist communicated the diagnosis to the parents. Accordingly, after the second appointment, parents of all the children had received an explanation of the diagnosis. The average age of these 15 children was 41.6 months (SD = 7.2, range 30–54) at the time of diagnostic disclosure.

### Time at which the diagnosis was communicated

Sixty-eight subjects were divided into quartiles by ordering the children by the age at diagnostic notification, from youngest to oldest (Group 1; 26–37 months, Group 2; 38–40 months, Group 3; 41–46 months, Group 4; 47–61 months). There was a relationship between the age of diagnostic notification and development checkup for 18-month-olds ($\chi^2$ (3) = 9.8, $p = .020$) and drop off the follow-up list between problem detection and diagnostic notification ($\chi^2$ (3) = 8.4, $p = .038$) but not proactively seeking out professional advice ($\chi^2$ (3) = 9.3, $p = .82$) (Table 1). Other than that, there was a relationship between the age of diagnostic notification and cognitive ability ($\chi^2$ (6) = 13.7, $p = .033$), but not autism symptomatology ($\chi^2$ (3) = 3.1, $p = .37$). Among the earliest group (notified between 26 and 37 months), four of 21 children were with average or near-average cognitive abilities (IQ $\geq$ 70), conversely, among the latest group (47–61 months), four of 21 children were with moderate and severe intellectual ability (IQ < 50). Concerning the variety of parents' status, there was a relationship between the age of diagnostic notification and mother's mental disorder ($\chi^2$ (3) = 8.6, $p = .036$), but neither ethnicity ($\chi^2$ (3) = 3.9, $p = .27$), educational status (father; $\chi^2$ (9) = 13.1, $p = .16$, mother; $\chi^2$ (12) = 13.5, $p = .33$), jobs (father; $\chi^2$ (3) = 2.7, $p = .45$, mother; $\chi^2$ (3) = 2.7, $p = .44$), father's mental disorder ($\chi^2$ (3) = 4.6, $p = .21$). There was no relationship between the age of diagnostic notification and whether their siblings had already been diagnosed with PDDs or not ($\chi^2$ (3) = 6.7, $p = .082$).

### Satisfaction with time of notification

Parents were asked if they were satisfied or dissatisfied with the time of diagnostic notification. There was one invalid answer. Thirty-nine respondents (58.2%) said they were satisfied or somewhat satisfied, 13 (19.4%) were dissatisfied or somewhat dissatisfied, and 15 (22.4%) said 'Neither satisfied nor dissatisfied.' To determine the proportion of parents satisfied with the time of diagnosis, satisfaction rates were calculated by ordering 68 children by age at diagnostic notification, from youngest to oldest, into quartiles (Group 1; 26–37 months, Group 2; 38–40 months, Group 3; 41–46 months, Group 4; 47–61 months). Results show the percentage of satisfied parents in each group: parents of 12 of 18 children (66.7%) in Group 1, 12 of 18 (66.7%) in Group 2, seven of 17 (41.2%) in Group 3, and eight of 14 (57.1%) in Group 4. There was no significant difference between the population rate and intergroup rates (Figure 2; between Group 1 and Group 2: $z = 0.0$, $p = .50$, 95% CI: 30.8–30.8%; between Group 1 and Group 3: $z = 1.5$, $p = .065$, 95% CI: 6.5–57.5%; between Group 1 and Group 4: $z = 0.55$, $p = .29$, 95% CI: 24.3–43.4%; between Group 2 and Group 3: $z = 1.5$, $p = .065$, 95% CI: 6.5–57.5%; between Group 2 and Group 4: $z = 0.55$,

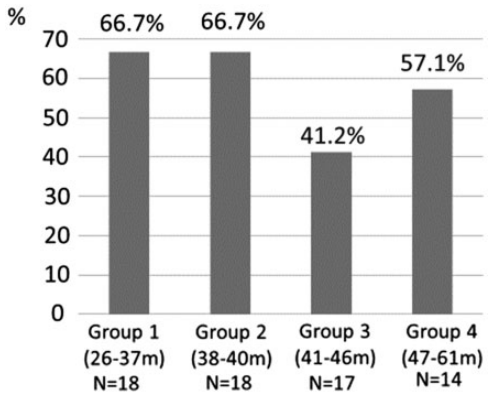


**Figure 2.** Satisfaction in the diagnostic process by percent. Between Group 1 and Group 2: $z = 0.0$, $p = .50$; between Group 1 and Group 3: $z = 1.5$, $p = .065$; between Group 1 and Group 4: $z = 0.55$, $p = .29$; between Group 2 and Group 3: $z = 1.5$, $p = .065$; between Group 2 and Group 4: $z = 0.55$, $p = .29$; between Group 3 and Group 4: $z = 0.89$, $p = .19$.

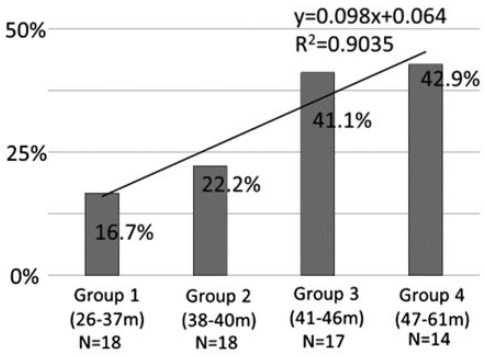


**Figure 3.** Percentage of parents answering 'The diagnosis was too late'. Between Group 1 and Group 2, $z = 0.42$, $p = .33$; between Group 1 and Group 3, $z = 1.6$, $p = .054$; between Group 1 and Group 4, $z = 1.6$, $p = .051$; between Group 2 and Group 3, $z = 1.2$, $p = .11$; between Group 2 and Group 4, $z = 1.2$, $p = .11$; between Group 3 and Group 4, $z = 0.09$, $p = .46$.

$p = .29$, 95% CI: 24.3–43.4%; between Group 3 and Group 4: $z = 0.89$, $p = .19$, 95% CI: 19.0–50.9%).

In the comments column, parents were asked to describe why they were dissatisfied with time notification. Of the 13 dissatisfied respondents, 11 provided a reason in the comments column. Ten of the 11 said they wished the diagnosis had been communicated earlier. The average age of the children of these 10 respondents was 44.0 months at time of notification (SD = 6.9, range 33–55), yet there was no significant difference ($t(75) = 0.99$, $p = .16$) compared to the average age of all 68 children (41.6 months). Only one respondent (whose child was 31 months at time of notification) expressed dissatisfaction, saying she was, 'taken aback by receiving the diagnosis of autism at the first appointment.'

Twenty respondents wrote, 'The timing of diagnosis was too late.' A quartile breakdown showed this view was expressed by three of 18 parents (16.7%) in Group 1 (26–37 months), four of 18 (22.2%) in Group 2 (38–40 months), seven of 17 (41.2%) in Group 3 (41–46 months), and six of 14 (42.9%) in Group 4 (47–61 months). The linear approximation of the coefficient of determination ($R^2 = 0.9035$) shows that the older the child in a group, the higher the percentage of answers indicating that time of diagnosis was too late (Figure 3). However, intergroup differences did not differ significantly from the population rate (between Group 1 and Group 2, $z = 0.42$, $p = .33$, 95% CI: 20.2–31.3%; between Group 1 and Group 3, $z = 1.6$, $p = .054$, 95% CI: 4.5–53.6%; between Group 1 and Group 4, $z = 1.6$, $p = .051$, 95% CI: 11.3–49.2%; between Group 2 and Group 3, $z = 1.2$, $p = .11$, 95% CI: 11.3–49.2%; between Group 2 and Group 4, $z = 1.2$, $p = .11$, 95% CI: 11.6–52.9%; between Group 3 and Group 4, $z = 0.09$, $p = .46$, 95% CI: 33.2–36.6%). In contrast, parents of two children (diagnosed at 31 and 37 months) wrote in the comments column, 'The diagnosis was communicated too soon.'

Satisfaction with the time of diagnosis was associated with the satisfaction with the manner in which the diagnosis was communicated ($\chi^2$ (6) = 30.2, $p < .001$), cognitive level ($\chi^2$ (6) = 10.4, $p = .033$), and mother's jobs ($\chi^2$ (3) = 3.0, $p = .041$). On the other hand, it was not associated with ASD symptomatology ($\chi^2$ (3) = 1.8, $p = .40$) nor diagnostic process such as 18-month checkup ($\chi^2$ (3) = 0.05, $p = .97$), drop off the follow-up list between problem detection and diagnostic notification ($\chi^2$ (3) = 2.1, $p = .36$), proactively seeking out professional advice ($\chi^2$ (3) = 4.5, $p = .11$).

## *How parent's received the diagnosis*

Sixty-eight parents gave at least one answer. In total 88 answers were coded, as some parents gave more than one reason in their answer (Table 2). Twenty-seven (39.7% of total) said they were shocked and emotionally upset. Among these, 16 (59.3%) were satisfied or relatively satisfied with the timing of the diagnosis, four indicated dissatisfaction or relative dissatisfaction (2 of these said they wished the diagnosis had been earlier), six said they were neither satisfied nor dissatisfied, and there was one invalid response. In contrast, 24 respondents (35.3%) acknowledged that it was what they thought or expected. Seven respondents (10.3%) expressed relief, saying, 'When I found out, it was a weight off my mind' or 'I was relieved to find out.' Seven respondents (10.3%) wanted information on the intervention programme and coping strategies. Another seven (10.3%) said they had expected a different diagnosis.

**Table 1.** Time at which the diagnosis was communicated: from the parental reports.

| | Group 1 (26–37 m) | Group 2 (38–40 m) | Group 3 (41–45 m) | Group 4 (46–61 m) | Significance |
|---|---|---|---|---|---|
| *N* | 18 | 18 | 18 | 14 | |
| Nationality | | | | | |
| Japanese | 18 | 18 | 18 | 13 | $p = .27$ |
| Other | 0 | 0 | 0 | 1 | |
| Education status | | | | | |
| Mother | | | | | |
| Junior high school | 0 | 1 | 0 | 1 | $p = .33$ |
| High school | 4 | 2 | 4 | 4 | |
| Vocational college | 4 | 5 | 1 | 0 | |
| Women's junior college | 7 | 6 | 9 | 2 | |
| University | 3 | 1 | 3 | 3 | |
| Father | | | | | |
| Junior high school | 0 | 0 | 0 | 1 | $p = .16$ |
| High school | 5 | 2 | 1 | 2 | |
| Vocational college | 1 | 3 | 4 | 0 | |
| University | 12 | 12 | 12 | 5 | |
| Jobs | | | | | |
| Mother | | | | | |
| Yes | 3 | 1 | 4 | 1 | $p = .54$ |
| No | 15 | 17 | 14 | 12 | |
| Father | | | | | |
| Yes | 18 | 17 | 18 | 11 | $p = .45$ |
| No | 0 | 1 | 0 | 1 | |
| Mental disorder | | | | | |
| Mother | | | | | |
| Yes | 0 | 0 | 0 | 2 | $p = .036$# |
| No | 18 | 18 | 18 | 11 | |
| Father | | | | | |
| Yes | 0 | 0 | 0 | 1 | $p = .21$ |
| No | 18 | 18 | 18 | 11 | |
| Divorced | | | | | |
| Yes | 0 | 0 | 0 | 1 | $p = .27$ |
| No | 18 | 18 | 18 | 13 | |
| Siblings | | | | | |
| Elder | 8 | 10 | 10 | 3 | $p = .35$ |
| Younger | 3 | 3 | 5 | 4 | |
| None | 7 | 5 | 3 | 7 | |
| Elder siblings had been already diagnosed with ASD | | | | | |
| Yes | 3 | 5 | 0 | 1 | $p = .082$ |
| No | 15 | 13 | 18 | 13 | |
| Diagnostic process | | | | | |
| Identified at the 18-month checkup | | | | | |
| Yes | 16 | 15 | 13 | 6 | $p = .020$# |
| No | 2 | 3 | 5 | 8 | |
| Dropped out of the follow-up system | | | | | |
| Yes | 1 | 6 | 7 | 7 | $p = .038$# |
| No | 17 | 12 | 11 | 7 | |

*(continued)*

**Table 1.** (continued)

| | Group 1 (26–37 m) | Group 2 (38–40 m) | Group 3 (41–45 m) | Group 4 (46–61 m) | Significance |
|---|---|---|---|---|---|
| Proactively sought out professional advice | | | | | |
| Yes | 2 | 3 | 2 | 3 | $p = .82$ |
| No | 16 | 15 | 16 | 11 | |
| Child | | | | | |
| ASD category | | | | | |
| Childhood autism | 16 | 17 | 17 | 10 | $p = .37$ |
| Other ASD | 2 | 1 | 1 | 3 | |
| Cognitive ability | | | | | |
| Normal/borderline intelligence | 2 | 4 | 9 | 8 | $p = .033$# |
| Mild intellectual disability | 9 | 4 | 4 | 3 | |
| Moderate/severe intellectual disability | 7 | 10 | 5 | 3 | |

#$p < .05$.

**Table 2.** How diagnosis was received.

| Comments | Sample quotes | Response rate (%) |
|---|---|---|
| Was shocked; Was upset ($n = 27$) | I had received a great deal of shock.<br>I was in a flurry because of being very surprised.<br>After all, my mind went blank, although I had been determined to undergo a diagnosis. | 39.7 |
| Thought so; It was expected ($n = 24$) | Just as I expected.<br>'Ah, just as I suspected (with a smile)', because I became fully conscious that my son had autistic characteristics. | 35.3 |
| Felt better after receiving diagnosis; Was relieved ($n = 7$) | I felt fine in receiving diagnosis, because I had been gloomy in not understanding the reason why my son's behaviour was different than other children.<br>Feelings mixed 'Just as I expected' and 'I can move forward in being refreshed'.<br>I got comfortable rather than was shocked. | 10.3 |
| Wanted information: On how to cope; On treatment programme ($n = 7$) | What's the best thing to do hereafter?<br>I want information on how to cope.<br>I want information on treatment programme. | 10.3 |
| Doubted diagnosis; Was unexpected ($n = 7$) | This is different than what I was expecting (I had only seen autism on TV).<br>I was expecting LD, so I was doubted diagnosis it was not expected at all. | 10.3 |
| Did not want to believe it; Did not want to accept it ($n = 4$) | I didn't want to believe it.<br>Feelings I didn't want to accept it. | 5.9 |

Note: Multiple answers possible.

### *The gap between parents and child psychiatrists regarding the diagnostic notification*

At the time of diagnostic notification, the child psychiatrist enters into the medical record information such as when and to whom the notification was given. The questionnaire asks parents this same information. A comparison shows that (1) discrepancy between the diagnostic notification date entered in the medical record and the date cited by parents was over six months, the typical interval between medical appointments, (2) in some cases it was difficult to determine whether the discrepancy in time of notification was

six or more months (for example, parents gave the child's age in only '3 years old', while his/her medical record described '3 years 3 months'), there were also inaccuracies in other information such as the place, people present, and the psychiatrist's name (for example, mother stated 'she was notified at the tatami's room and the father is not with her', while medical record described 'at the room of western-style with both the mother and father') and (3) even when a parent identified the date of notification within six months of the actual date, the medical record revealed that the parent was referring to a different appointment that took place within the six-month period. These three criteria illustrate the gap between parental perceptions and the medical record.

Parents of two children did not identify the time of notification. The perceived gap was seen in parents of 14 (20.6%) of 66 children.

Ultimately, parents of all 68 children were told that their child was diagnosed with autism. All 68 questionnaire respondents confirmed that their child psychiatrist had told them about their child's diagnosis of autism or developmental disorder.

## Discussion

In addition to implementing early interventions for preschool children with autism in the community, an extremely critical clinical issue is when and how to convey to parents their child's diagnosis of autism. Previous research on parental perceptions of the ASD diagnostic process has investigated the delay between parental concerns and confirmation of diagnosis. The information obtained by this means shows that parents' dissatisfaction with the diagnostic process increases the longer they wait for a diagnosis. This is essential information autism specialists must bear in mind. In today's environment in which early intervention programmes have been developed and can be implemented in early childhood (<3 years), it will become increasingly necessary for autism specialists to identify the characteristics of autism before the parents do and, as stated in the NICE (2011) guidelines, initiate the diagnostic process nurturing parental understanding and informing parents at the earliest possible stage. How will parents respond to the diagnosis? This question cannot be addressed by research methods used thus far.

This critical question was explored in this research in three ways that are missing in prior research methods. First, the study was based in a community with an ongoing early detection and intervention programme in which all research subjects were diagnosed before entering school, most when they were toddlers. After being identified with characteristics of autism at the 18-month infant health checkup and referred to YRC, the diagnosis was conveyed to parents by a YRC child psychiatrist. This diagnostic process enables healthcare professionals to become aware of a child's autism before the parent, making it possible to discover how the parent feels about the diagnosis. Second, information about parents' feelings about the process leading to and the manner in which the diagnosis was communicated was not derived only from parental recall, but was supplemented by the medical record. This made it possible to obtain clear and objective observations rather than simply parents' subjective recollections of the diagnostic process. Third, by devising a parent-specific survey instrument, a high response rate was obtained that elicited parents' candid opinions about the diagnostic process.

At the root, public health and welfare policy in Japan generally stems from ideology of paternalism and the early detection and intervention system has been created by means of applying this to the field of ASD. Also, Japanese-style informed consent has been the importation and absorption of Western concepts into Japanese culture by transforming them into forms more compatible with existing social and political arrangements (Leflar, 1996). Therefore, the situation of Japan about the ASD diagnostic process could be different from the one of Western countries, anyway by using an innovative research design with these distinctive elements, it was possible to examine parents' reception of their child's autism diagnosis with a different procedure that had previously reported about the diagnostic process.

This study is divided broadly into two categories, the perspective of the review of records and parental reports.

At first, in relation to the review of records, it is suggested that a variety of factors influence earlier diagnostic notification. Cognitive ability and early detection are considered important variables that promote earlier diagnostic notification. Nevertheless, there are a few parents who are notified earlier though their children have been detected later, on the other hand a few parents who are notified tardily though their children have been detected earlier.

Subsequently, findings from parent report distinguished three aspects of parental perceptions of the timing of diagnostic notification. First, the majority of parents (58.2%) who were notified of their child's diagnosis in early childhood were satisfied with the time of notification. This is a relatively high figure compared to that of prior research studies that included cases in which notification occurred after the child entered school. Approximately 40% of parents wrote that they felt shocked or upset when they received the diagnosis. It can be assumed that when the diagnosis of autism was communicated when their children were

toddlers, some parents were not psychologically prepared to hear it. However, 60% of the parents who were shocked or upset indicated satisfaction with the time of diagnosis. This suggests that, even when parents are taken aback by the diagnosis, most would hope to be notified at an early stage if it ensures appropriate early intervention.

Second, the research also showed that a considerable number of parents wished that diagnostic notification had been even sooner, even within the stage of early childhood. Of the 13 respondents (19.4%) who were dissatisfied or somewhat dissatisfied with the time of notification, 10 were parents of children whose average age was 44.0 months at time of notification and although they had been notified by the time their child was midway through early childhood, they still expressed dissatisfaction with the late receipt of the diagnosis. These 10 respondents included four who said they felt shocked or upset when they received the diagnosis. Only one parent expressed dissatisfaction at receiving the diagnosis when the child was very young (31 months).

Contrary to expectation, this research shows that, even if a problem is discovered early, some parents may desire over a period of time before notification of the diagnosis. Broadly speaking, all previous research, including Howlin and Moore (1997), whose research subjects ranged widely from childhood to adulthood, show a linear relationship between parental satisfaction and younger age at time of diagnostic notification. However, as shown in our survey, when the focus is limited to toddlerhood, the degree of satisfaction is roughly the same whether parents were notified earlier or later. Also, although a somewhat higher percentage of parents whose children were diagnosed in later childhood felt that the diagnosis came too late, there was no statistically significant difference compared to the early group. All our findings suggest that it is not possible to simply state that earlier notification led to higher parental satisfaction, though later notification tend to lead to more dissatisfaction.

We would like to propose, therefore, the concept of optimality of diagnostic disclosure as related to the diagnostic disclosure process, in the community-based system of early detection and intervention. This is a comprehensive concept composed of elements such as the time of diagnostic notification and manner of disclosure. A fundamental principle is that disclosure of an autism diagnosis is made, preferably without delay, but numerous factors may allow for some variation in timing. By 'numerous factors,' one can presume such factors as the characteristics and severity of autism symptom expression, cognitive levels, capacity to make an accurate diagnosis, whether an appropriate intervention is ensured, and characteristics of the caregiver. Maybe the provision of post-diagnostic support, such as Crane et al. (2016) state, is also one of the major candidate of optimality of diagnostic disclosure. It is noted that 'current best practices for interventions for children $<3$ years with suspected or confirmed ASD should include a combination of developmental and behavioural approaches and begin as early as possible (Zwaigenbaum et al., 2015).' However, if a diagnosis is communicated before optimal conditions are in place, the danger is that parents may experience extreme cognitive dissonance, be burdened with overwhelming stress, and become unable to engage with healthcare professionals in mutual decision-making related to early intervention, especially if the diagnosis is communicated when the child is very young. For example, according to Chamak et al. (2011) 'while parents often felt relieved (43%) when they obtained the diagnosis for their children with ASD at the age of four or more, the majority of parents (75%) had difficulties accepting it when the child was younger than 4'. Our research found that, one in five parents did not want to be informed about their child's diagnosis at the first psychiatrist's appointment, and accordingly, the child psychiatrists determined that they took precedence over describing the child's developmental issues from the perspective of autism and advising the parents on managing their childcare challenges. However, this did not delay the provision of essential early intervention. In the survey, even children whose parents were not informed at the first examination were entered without delay into the intervention programme and were informed of the diagnosis at the second examination. Before the second appointment, a clinical psychologist would have met with the parents to discuss their child's development and offer child-raising advice in response to parents' questions, thereby gradually nurturing parental awareness of their child's condition. Because this research prioritized this issue, this may account for the large number of respondents who said they had already anticipated their child's diagnosis of autism before the child psychiatrist informed them. This group roughly equalled the number of respondents who said they were shocked when they heard the diagnosis.

In approximately one in five parents there was a discrepancy between the time of diagnostic notification as noted by the child psychiatrist in the medical record and the time respondents claimed to have been notified. To our knowledge, there are no studies that explore the gap between the psychiatrist and parental perceptions regarding notification of an ASD diagnosis. In our questionnaire, all the parents acknowledged that the psychiatrists informed them about autism and developmental disabilities, and ultimately and most importantly, their child's diagnosis was communicated

appropriately. However, although a psychiatrist communicates the diagnosis clearly, some parents may interpret the explanation as not be communicated clearly (perhaps due to conscious as well as subconscious factors), and so this research suggests that psychiatrists may exercise discretion by adopting an intermediate step, if warranted, based on the mental state of such parents. In an optimal diagnostic disclosure process, a most important characteristic is two-way communication between the psychiatrist and parents.

It is major limitation that the prompt timing of the diagnosis and the provision of post-diagnostic support are conflated. In this research, it is assumed that both of these have been linked to parental satisfaction with the ASD diagnostic process. For example, Crane et al. (2016) stated that they analysed factors affecting overall satisfaction with the diagnostic process and the support offered post diagnosis was one of the predictor variables of overall satisfaction with the diagnostic process. Next time, we need to try to reveal which aspects drive parental satisfaction or whether this is a cumulative effect.

Also, in this research the focus was narrowed to the diagnosis of childhood autism. In some cases, a child presented typical symptoms of autism when diagnosed, but later developed atypical symptoms, and because they could not participate in the early intervention programme, they were excluded from the study. Although one of the limitations of this research is the small sample size, most of the children with severe ASD symptoms who were difficult to manage at community kindergartens and nursery schools were admitted to the early intervention programme, so it was possible to narrow the research focus to cases with typical ASD symptoms. Future research must be conducted on a broader range of subjects with all types of ASD. Given the diversity of ASD symptoms, optimal diagnostic disclosure strategies will surely require an even more individualized approach than required for childhood autism.

## Authors' note

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## Appendix 1: The Parent Satisfaction Inventory

1. We ask you about situation when you first received your child's diagnosis.

   How old was your child at that time? ( ) years ( ) months
   Where did you received your child's diagnosis?
   Who were there with you?
   Who did you notify your child's diagnosis? (fill in Dr's name)
   Are you firstly notified with your child's diagnosis in your family? (Yes /No)

2. How satisfied are you with the timing and explanation of the diagnosis and adequacy of information about services.

   five-point Likert scales ('dissatisfied' to 'satisfied').
   If you answered 'dissatisfied' or 'somewhat dissatisfied' to this question, please explain your frank reasons (in an open-text box).

3. We ask you about content of diagnostic notification.

   What were you notified about their child's diagnosis?
   How did you feel when you received their child's diagnosis?