

Parents' Ability to Perceive Pain Experienced by Their Child with Down Syndrome

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***Aims:** To investigate parents' ability to perceive pain experienced by their offspring with Down syndrome (DS). **Methods:** Data were gathered by the use of the Oral Assessment in Down Syndrome Questionnaire in a cross-sectional survey design in France. A sample of parents of 204 children with DS and 161 of their siblings without DS was accrued. **Results:** Parental reports of difficulty discerning if their child with DS was in pain did not change with age of the child, remaining at a prevalence of 28% to 32%. Reports of difficulty discerning where that child felt pain diminished with older age from 74% to 27%. The likelihood of parents reporting difficulty discerning if and where their child with DS had pain was greater than for a sibling without DS. However, reports of pain experience for the 2 groups were the same. Moreover, different functional and dysfunctional behavioral variables were found to be predictors of these 2 pain perception variables. **Conclusion:** Parental perception of pain is less discriminant for children with DS than for their siblings without DS. J OROFAC PAIN 2003;17:347–353.*

Key words: Down syndrome, pain perception

In the last decade, the pain experience of children has been widely explored in order to improve pain control. However, studying pain in a population with cognitive impairment (CI) is difficult and few studies have focused on such groups,^{1–5} although recently published work has begun to explore the measurement of pain in this population through the observation of various pain-related behaviors.^{6,7} Pain expression depends upon basic motor competence and/or verbal capacity,⁸ but cognitive and psychomotor problems are particularly marked in persons with intellectual deficiency. In view of the fact that many persons with disability are more likely to suffer health problems and have specific health needs compared with members of the general population, any limits in the expression of pain and discomfort may put such groups at further disadvantage.⁹ Furthermore, parents of children with CI have reported that their children's pain was treated differently than that of other children and believed that care providers had difficulty assessing and treating their children's pain.¹⁰ The inability to detect or to communicate pain can lead to serious and, sometimes, life-threatening conditions.¹¹

Several case reports have described individuals with intellectual disabilities who demonstrated a decreased responsiveness to injuries that should have evoked significant pain behavior. It has been reported that descriptions of pain expression and coping behavior by parents of 145 children with CI were related to the level of CI of their child.¹⁰ Children with mild to moderate CI were more likely to be described as directly communicating their pain and exhibiting procedural coping strategies similar to those observed in children without CI. Those children who did not exhibit any sign of discomfort in potentially painful situations were assumed to be insensitive to pain both by parents and health care workers. For example, more than half of parents reported that their children experienced pain differently than did children without CI, with the majority perceiving decreased pain sensitivity and greater pain tolerance. Biersdorff¹² asked third parties (family members or care providers) to report injury or illness incidents and to describe the responses of 123 individuals with developmental disabilities in their care (22% persons with Down syndrome [DS]). Although no control group was studied, her results suggest that pain experience is altered in this population. Twenty-five percent of subjects displayed behavior suggesting that their pain thresholds were abnormally high. Based on these findings, it has been hypothesized that pain insensitivity or indifference may be related to the degree of intellectual impairment.

Down syndrome is the most common congenital cause of CI around the world, thereby making people with DS an important group to study regarding pain expression in persons with CI. Vocal responses to painful stimuli have been studied in infants. It has been found that infants without DS required less stimulation to arouse crying and had a shorter reaction time than their peers with DS.¹³ This lack of a cry response in babies with DS increased with age and those older than 1 year showed none of the visible responses, such as grimacing, limb movements, or breaks in respiration, that were seen in control infants. Like other groups of children with CI, those with DS are commonly assumed to have a defect in the processing of sensory information.^{14,15} It has recently been shown that the response to a nociceptive stimulus by people with DS is delayed in time and less precise than the response of people without DS.¹⁶ However, the responses given by people with DS were organized in the same way as those of controls and were clearly recognizable. It may be concluded that individuals with DS are not insensitive to pain; rather, they express pain or discomfort more slowly and

less precisely than the general population.¹⁶ In an investigation of postoperative pain experienced by 7- to 12-year-old children without CI, the parents' report demonstrated low levels of sensitivity in identifying when their children were experiencing clinically significant pain during 2 days following minor surgery.¹⁷ However, in their work on the development of the Non-communicating Children's Pain Checklist (NCCPC), Breau et al^{6,7} demonstrated that caregivers and health professionals are able to evaluate pain-related behaviors in cognitively impaired children.

With these observations in mind, the aim of this study was to investigate the hypothesis that parents experience more difficulty perceiving pain in their child with DS compared to the sibling closest in age without DS. A secondary aim was to investigate what child-related variables predict parents' ability to perceive if their child is in pain and where the pain is experienced.

Materials and Methods

Study Design and Sample

The study, which was undertaken in November 1997, was a cross-sectional survey of a sample of parents of persons with DS from throughout France. The sample consisted of parents attending a national conference of the Fédération des Associations pour l'Insertion Sociale des Personnes Porteuses d'une Trisomie 21 (FAIT 21), a French national organization for parents of, and health care professionals working with, individuals with DS. Following a presentation concerning the nature and purpose of the study at the conference, parents of persons with DS were given 2 differently colored copies of a questionnaire. The first was to be completed with respect to their child with DS and the second with respect to the sibling without DS who was closest in age to the DS child. The second questionnaire was to enable comparison between children with and without DS, and the same parents were used to evaluate both groups. Parents were asked to return the 2 questionnaires in the stamped, addressed envelopes provided. In France, epidemiologic investigations of this nature do not need specific ethical committee approval. The parents were at liberty to complete the questionnaires (or not complete the questionnaires) as they wished, and all respondents were anonymous.

Of the 350 sets of questionnaires given out at the conference, 204 (58.3%) returned completed questionnaires concerning individuals with DS and

Table 1 Study Sample Descriptive Statistics—Prepared Data Only

Group	n	Mean age (y)	Median age (y)	Age range (y)	Gender distribution	
					Male	Female
Persons with DS	161	9.23	8	1–39	83	78
Siblings without DS	161	11.83	11	1–38	80	81

DS = Down syndrome.

161 returned completed questionnaires concerning siblings without DS. It is not possible to evaluate the response rate for the latter group, because some of the persons with DS in our study may have been single children. The descriptive statistics for the study sample are shown in Table 1. There was a significant difference in age between the 2 groups; the group with DS has a younger mean age than the sibling group.

Data Collected

The dependent variables were from the pain domain of the Oral Assessment in Down Syndrome (OADS) questionnaire, a validated, French, parental assessment of oral health problems in persons with DS.¹⁸ Questions in the pain domain were (1) Is it difficult to judge if your child is in pain? (2) Is it difficult to judge where your child has pain? and (3) In as much as you can judge, has your child ever suffered from painful dental problems? The responses to all of these items were a dichotomous “yes/no.”

While the matched nature of this data set considerably reduces the number of possible correlates of differences in dependent variable responses, it is conceivable that age, dental and other health status indicators are different within the pairs and that they are related to the dependent variables. Clinical dental status data were not collected, but data concerning dental symptoms other than pain, parental perception of childrens’ dental status, and the receipt of dental treatment were collected, as were age, gender, and indicators of disability and health status. Indicators of disability were taken from the OADS questionnaire, while those concerning health status were data gathered on frequency of consultations with specialists concerning heart, immunologic, otolaryngologic, and speech problems. Those visiting a specialist twice a year or more were categorized as having a health problem. The exact degree of CI was not measured by the questionnaire as this data is often not available to parents in France. Moreover, it is now accepted that intellectual level cannot be described by a single index.

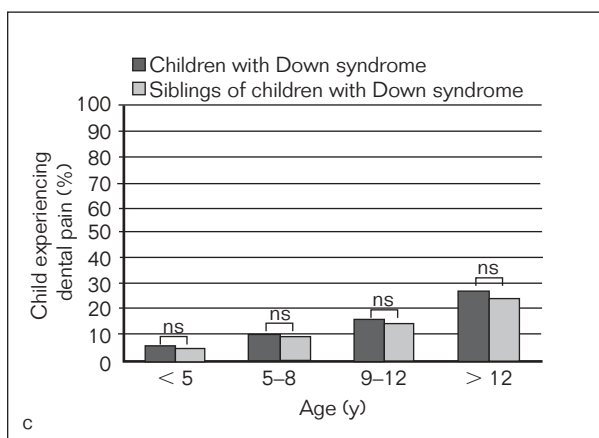
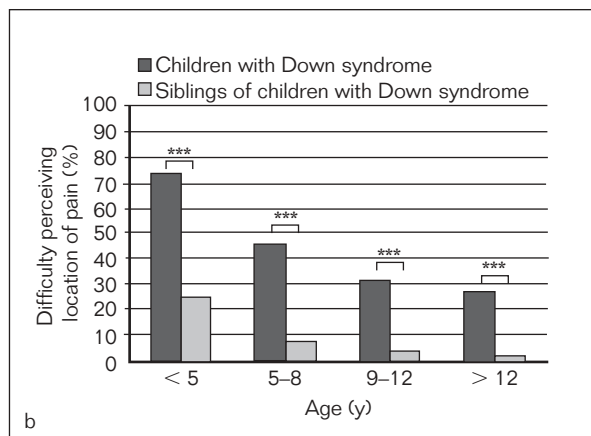
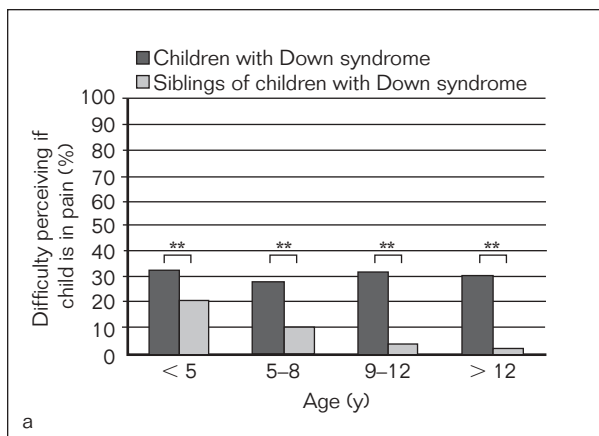
Statistical Analyses

Analyses were performed on the paired data set of 161 siblings only. Following descriptive statistics, analyses of differences in responses to dependent variables were performed using a McNemar test because of the paired nature of the data.¹⁹ The analysis focused on the discordant pairs since only those situations where the behavior of the family pairs was different were of interest. The McNemar test compared the number of discordant pairs in 2×2 tables; for example, in this data set it tested the hypothesis that the number of family pairs in which the parent reported that the child with DS has experienced dental pain while the sibling has not is the same as the reverse (ie, the number of pairs in which only the sibling has experienced dental pain). A P value $< .05$ was considered to indicate statistical significance.

Following the matched bivariate analysis investigating the effect of DS status on dependent variables responses within families, conditional multiple logistic regression analyses were performed for the 3 dependent variables to control for age, dental status, and indicators of disability and health status. All variables with the probability of no association ($P < .1$) were entered in the multivariate models. In addition, the 2 perceptual difficulty variables were entered into the model for predictors of dental pain experience.

Results

The results of the age-stratified paired analysis of the effect of DS status on the 3 dependent variables are demonstrated in Fig 1. For all age groups, the proportion of parents reporting difficulty perceiving whether their DS child is in pain differed significantly from the proportion of parents reporting difficulty perceiving whether a sibling is in pain ($P < .01$ for all age groups). Furthermore, the proportion of parents reporting difficulties perceiving whether their child is in pain was approximately the same for all age groups in the children with DS (27.6% to 32.3%), while the same proportion for



Figs 1a to 1c Comparison of the effect of Down syndrome status on the parental reports of difficulty perceiving if their child is in pain (a), of difficulty perceiving where their child's pain is located (b), and of their child having experienced dental pain (c). ** $P < .01$; *** $P < .0001$; ns = nonsignificant; McNemar test.

their siblings decreased with increasing age from 20.0% to 1.5% (Fig 1a). The pattern for parents' ability to perceive where pain is experienced by the child was slightly different. Again, the paired analyses showed significant differences in parental reports of perceptual difficulties between children with DS and siblings ($P < .0001$) for all age groups. However, across age groups, the pattern was the same in both groups with respect to reported ability to discern where pain is experienced, which improved with age (from 73.9% reporting difficulty to 26.8% in the DS group and from 25.0% reporting difficulty to 1.5% in the sibling group) (Fig 1b). Finally, Fig 1c shows an altogether different pattern in reported dental pain experience. The paired analyses showed no differences in reported dental pain experience between the children with DS and their siblings, across all agegroups. In addition, there was a consistent increase in the proportion of both groups reported as having experienced dental pain with older age.

The relationship of selected variables, other than age and DS status, with the 3 dependent variables is demonstrated in Table 2. Within the group with DS, parental ratings of their child's dental health

was related to all 3 variables. Parents of children with DS and poor dental health were more likely to report perceptual difficulties and dental pain experience. The receipt of some form of dental treatment was associated with parents being less likely to report difficulties perceiving where the pain was felt for their child with DS but more likely to report experience of dental pain. The latter association was the only observation to be repeated in the sibling group, in whom there were no other significant relationships. Again, within the DS group, those with speech problems, eating problems, and tooth grinding were all more likely to report difficulties perceiving where their child was in pain.

The bivariate associations demonstrated in Fig 1 and Table 2 were largely retained in the multivariate models shown in Table 3. Difficulty judging whether their child is in pain was associated with DS status and tooth grinding; difficulty judging where their child is in pain was associated with DS status, age, and eating and speech problems; and parental reporting of dental pain experience was associated with age, dental status, and receipt of some form of dental treatment.

Table 2 Frequency Distributions for the 3 Dependent Variables (Difficulty Judging if Child is in Pain, Difficulty Judging Where Child's Pain is Located, and Has Child Ever Experienced Dental Pain) by Selected Independent Variables, Within the Group with DS and Their Siblings

Variable	"If pain" [†] (% yes)		"Where pain" [‡] (% yes)		Dental pain experience [§] (% yes)	
	DS	Sibling	DS	Sibling	DS	Sibling
Dental health						
Poor	47.8*	0	56.5*	0	52.2*	20.0
Good	27.1	6.4	38.1	9.0	0.5	16.0
Dental treatment						
Yes	27.7	2.0	30.1*	4.0	26.5*	22.0*
No	30.6	13.1	47.1	16.4	7.4	6.6
Speech problems						
Yes	33.8	16.7	51.2*	16.7	13.5	0
No	26.9	5.8	33.9	8.4	16.2	16.8
Eating problems						
Yes	31.1	8.3	46.4*	16.7	16.6	0
No	24.5	6.0	22.6	8.1	11.3	17.5
Tooth grinding						
Yes	36.3*	10.0	46.1*	15.0	15.7	15.0
No	22.6	5.7	34.3	7.8	14.7	16.3
"If pain" [†]						
Yes					18.3	20.0
No					13.9	15.9
"Where pain" [‡]						
Yes					14.6	14.3
No					15.6	16.3

DS: Child with Down syndrome, Sibling: sibling without Down syndrome.

*Significant differences in proportions of parents responding yes to the dependent variables concerned (chi-square test; $P < .05$).

[†]Parents have difficulty perceiving if their child is in pain; [‡]Parents have difficulty perceiving where their child's pain is located; [§]Child reported as having experienced dental pain.

Table 3 Multiple Logistic Regression Models for the 3 Dependent Variables (Difficulty Judging if Child is in Pain, Difficulty Judging Where Child's Pain is Located, Has Child Ever Experienced Dental Pain)

Dependent variable	Independent predictor variables	OR	95% CI
Difficulty judging if the child is in pain	DS status (DS = 1; sibling = 0)	4.7	2.2–9.8
	Child grinds teeth (no = 0; yes = 1)	1.9	1.0–3.3
Difficulty judging where child's pain is located	DS status (DS = 1; sibling = 0)	2.5	1.1–5.6
	Age (continuous variable)	0.9	0.8–0.9
	Eating problem (no = 0; yes = 1)	2.9	1.5–5.6
	Speech problems (no = 0; yes = 1)	2.0	1.1–3.7
Has child ever experienced dental pain	Age (continuous variable)	1.1	1.0–1.1
	Dental status (poor = 1; good = 0)	6.2	2.6–14.7
	Dental treatment (received treatment = 1; no treatment experience = 0)	2.7	1.3–6.0

DS: Child with Down syndrome, Sibling = sibling without Down syndrome.

OR = odds ratio; 95% CI = 95% confidence interval.

Discussion

The aim of this study was to investigate the hypothesis that parents experience more difficulty perceiving pain in their child with DS compared to the sibling closest in age without DS. The study findings supported this hypothesis. Furthermore, in

an exploration of predictors of parents' perceptual difficulties, it was found that these variables were associated with certain indicators of disability (tooth grinding, difficulties in eating and speech). Finally, it was observed that dental pain experience was not associated with DS status or any indicators of disability, nor was it associated with perceptual

difficulty reported by the parent; rather it was associated with age, parental rating of dental status, and the receipt of dental treatment.

Certain limitations of the research design need to be recognized before these results may be discussed. The most obvious limitation is the source of the sample for the study. The data analyzed were from 46% of the questionnaires distributed to a group of parents attending a congress. Respondents are likely to represent the most motivated of parents of persons with DS and may be assumed to be more aware of their children's health care problems and needs than the population of parents of individuals with DS as a whole. However, in comparison with other investigations, this study produced similar results in terms of the prevalence of dental pain experience in children, which lends credibility to the study sample. The proportion of parents reporting that their 5-year-old child had ever experienced dental pain in a UK dental health survey ranged between 17% and 9%, varying with water fluoridation.²⁰ In Australia, Slade et al found that 12% of 5-year-olds and 32% of 12-year-olds were reported by their parents to have a history of toothache.²¹ These figures are similar to those found in our study. Another limitation of the study relates to the nature of the variables. The variables concerning pain, functional and developmental problems, disabilities, and signs of oral disease have been validated as part of the OADS questionnaire,¹⁸ but they remain somewhat crude indicators of oral health-related problems for the DS population. In view of the study design and the variables, the results of the present investigation should be interpreted with caution but may be useful for generating hypotheses that could shed further light on the issue of pain perception for patients with DS.

The results of this study suggest an interesting interrelationship between the 3 pain perception variables used. Predictably, there was a large difference in the proportion of parents reporting difficulty discerning whether and where their child was in pain between the DS and sibling groups. However, it was more intriguing that, controlling for age, the proportion of parents reporting dental pain experience for the 2 groups was similar and that neither of the pain perception variables was associated with reports of dental pain experience. Together, these findings suggest that the parents of children with DS overcome the problems of pain perception in their offspring. In childhood, the most common causes of dental pain are probably tooth eruption, tooth exfoliation, and caries.²¹⁻²³ Theoretically, the only reason to expect that chil-

dren with DS should experience a higher incidence of dental pain than their peers could be due to temporomandibular and other chronic facial pain disorders,²⁴⁻²⁶ or possibly to the higher incidence of periodontal disease. The limited evidence suggests that, when age and eruption dates are controlled for, children with DS have the same dental pain incidence as their siblings without DS.

Another intriguing finding of this study was the relationship, within the DS group, between age and parental report of difficulty discerning whether their child was in pain and where that pain was experienced. Compared to mothers of children with a motor impairment, mothers of children with DS are more concerned about their child's development in the area of communication.²⁷ Communication between children and parents improves over the years. However, the results of this study showed that the proportion of parents reporting difficulty judging the location of their child's pain decreased with increasing age, while the proportion of parents reporting difficulty judging whether their child was in pain remained the same. Interestingly, the proportion reporting difficulty locating the pain fell from 74% to 27%; the latter value is very similar to the proportion of parents reporting difficulty judging whether their child is in pain. These observations suggest that there is a core group of children with DS, comprising approximately a quarter of the population, who present their parents with a long-standing problem in pain perception, which does not improve over time. In other words there are 2 groups: the first for whom an improvement is seen in parental pain perception with age and the second for whom there is no such improvement.

Different functional and dysfunctional behavioral variables were also found to be predictors of the 2 pain perception variables. These 2 sets of findings suggest that the inability to judge whether a child with DS is in pain could be a sign of more severe neuromotor expression of the underlying DS and that there may be little improvement with age or time for this subgroup. It would be interesting to investigate whether members of this subgroup had undertaken specific neuromotor education programs, or whether lack of emotional feedback had led to discontinuation of active stimulation. This inference remains to be confirmed through hypothesis-driven research.

In conclusion, this study underlines the need for the development and dissemination of more objective tools of measurement for pain and discomfort specifically designed for populations with cognitive impairment, such as the NCCPC.^{6,7} These tools need to be sensitive to language and culture and be

validated in their different countries of use. Such tools may be useful in both clinical and domestic settings, because the inability to detect pain raises concerns over access to health care and rapidity of management for these groups (for example, the prevention of bacterial endocarditis). On average, persons with DS experience greater health problems than their peers and the detection and management of disease may depend upon expression of pain. The present investigation suggests parents are able to compensate to a certain extent for difficulties in perceiving pain in their offspring with DS, but that their discrimination is lower than for children without DS.

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