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Original article

Screening for risk factors of relational withdrawal behaviour in infants aged 14–18 months

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Abstract

Objectives. – The objectives of this study were (1) to evaluate the prevalence of relational withdrawal behaviour in infants aged 14–18 months attending a public health centre in Paris, (2) to check some identified risk factors for relational withdrawal behaviour in this population.

Methods. – A cross-sectional study was conducted in infants aged 14–18 months attending a child health screening centre during the year 2005.

Results. – A total of 640 children were included in the study. Thirteen percent of the 640 infants ($n = 83$, 95% CI [10.4%; 15.6%]) had an ADBB score at 5 and over 5 on the ADBB. There was a clear relationship between withdrawal behavior and having psychological difficulties as reported by parents, and between withdrawal and developmental delay. Withdrawal was also significantly associated with being a boy, with living in risk conditions (e.g. child being in joint custody, or with living in a foster family), with being adopted, or with being a twin. More withdrawn infants were taken care of at home.

Conclusion. – Sustained relational withdrawal behaviour was linked with developmental disorders and psychopathology and not with SES, ethnical origin or rank of birth. The scale could be used in screening early psychopathology in infants aged 2–24 months of age.

Keywords: Screening; Infancy withdrawal behaviour; Assessment of relational abilities; Alarm Distress Baby Scale (ADBB)

1. Introduction

Infants are born with both biologically determined abilities and an urge to participate in human interaction, the early parent-infant relationship providing the scaffolding necessary for the infant to develop [3,8,38]. The interactional skills of the infant include the ability to initiate and maintain eye contact with another person, to vocalize, and to use facial expressions and body and head movements to engage the caregiver or other persons in interaction. All normally developing infants display these skills during the first two months after birth, and even though infants may differ in their style and degree of responses to various stimuli – i.e. have different temperaments – they still are responsive to social interaction with an adult [11]. Withdrawn social behavior in infants is indicated by diminished or lacking positive behaviours (e.g. eye contact, smiling, cooing) or negative behaviours (e.g. crying).

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Withdrawal is to a certain degree a normal feature of infant behaviour in parent-infant interactions, and a way for the infant to regulate the flow of interaction [3]. However, increased or sustained withdrawal reaction in infants can be observed in inadequate parent-infant interactions, for example between a depressed mother and her infant or with a substance-dependent mother. The infant's "depressed" style of interacting may be carried over to other relationships as well, and be apparent even when the infant interacts with a non-depressed adult [10]. Observations of young children separated from their caregivers led Robertson and Bowlby [31] to describe a three-stage emotional reaction in young children, comprising protest, despair, and eventually detachment in the face of prolonged separation. Along these lines, sustained withdrawal behavior may be viewed as a chronic diminution of the attachment system, which is gradually generalized into a diminished engagement and lowered reactivity to the environment as a whole. Withdrawn social behavior from as early as 2 months of age, indicated by a lack of either positive (e.g., smiling, eye contact) or negative (e.g., vocal protestations) behavior, is more akin to a state of learned helplessness [34] and should alert the clinician to the possibility that the infant is not displaying age-appropriate emotional/social behavior. Infants may also appear socially withdrawn in several clinical conditions, for example in autism, chronic or severe pain, failure to thrive, or posttraumatic stress disorder [13,33,38]. Withdrawal behaviour is also a key symptom of infant depression [36]. That social withdrawal could be a sign of infant depression is supported by several studies. For instance, frontal electroencephalographic (EEG) asymmetry and high basal and reactive cortisol levels found in depressed adults have also been found in infants presenting withdrawn social behaviour, and sadness [4,5,17]. The findings are similar to those for depressed adults [22] and for preschoolers [23,24]. Withdrawal behavior is also a feature of most attachment disorders, particularly disorganized attachment [2].

In light of data gathered from various studies, withdrawal from social interaction is a sign of infant distress regardless of the cause, and can reflect the problems of the infant but also those of the caregivers [18,20,25,26]. There is an increasing awareness that assessing the level of an infant's social behavior is important: while diminished social behavior in the infant may not necessarily be an indicator of pathology, it should alert the clinician to undertake further assessment of both the infant and the environment. In some cases, it may reveal that the mother is experiencing psychosocial difficulties (e.g., postnatal depression, anxiety, or bonding difficulties) which she may have been reluctant to disclose [35]. Since development is an interactive process, withdrawal behavior is a developmental risk in itself [32,37], and may lead to further social withdrawal, anxiety or conduct disorders [12].

However, detection of infant withdrawal may be difficult, particularly for front line workers who may not have much training in infant mental health issues. A simple screening scale to enhance structured observation of infant social behaviour, the Alarm Distress Baby Scale or ADBB [14] has been developed and used in several studies, in several countries, and in different settings. The ADBB scale was designed to assess infant withdrawal behavior in the course of routine physical examinations carried out by a range of health professionals [1,15,31]. Recently, high scores on the ADBB (indicative of sustained withdrawn behavior) have been shown to be associated with sub-optimal interactive behaviors in both the mother and her infant in a Finnish study of 127 two-month-old infants [30], and with increased psychopathology in an Israeli study (the level of sustained withdrawal behavior as assessed with the scale was compared with the level of developmental risk in the same population (High/Low), and the cut-off score of 5 (or 4/5, i.e. 5 and above) yielded a sensitivity of 0.82 and a specificity of 0.78 [7]. In another Finnish study [29], scores with the ADBB were compared with scores with Murray & Fiori's Global Rating Scale in a face to face procedure. The cut off score of 5 (or 4/5) yielded gain the best specificity and sensitivity. A recent Brazilian study [9] compared ADBB scores with clinical diagnostic, and found the cut off score of 5 to give a sensitivity of 0.79 and a specificity of 0.81. The scale has demonstrated good metrological properties in a Well Baby clinic in the 14th *arrondissement* in Paris [13], and in several other countries [1,7,9,27,28,30]. The use of the ADBB scale in Well baby clinics in France has helped to screen for various conditions [14].

The aim of the present study was (1) to evaluate at 14–18 months the prevalence of relational withdrawal behaviour in children seen at the *Département des Examens Périodiques de l'Enfant* (DEPSE), and (2) to identify and verify the weight of some risk factors for relational withdrawal using a multivariate analysis. We performed a cross-sectional study, exploring links between the level of withdrawal behaviour, as assessed with the ADBB scale during a nursing assessment, and biological, biometrical, social, medical and psychological data gathered the same day in the centre.

2. Methods

2.1. Study design

The DEPSE of the *Caisse Primaire d'Assurance Maladie de Paris* (CPAM) sees infants from families on social welfare living in Paris and the *Ile de France* Region. The centre provides half-day free check-ups for infants aged 14–18 months [28,39]. Families receive a letter with the information about the centre and the check up, or baby welfare clinics may directly inform families about the centre.

We performed a cross-sectional study in one of the three DEPSE between September 2005 and July 2006. All infants aged between 14 and 18 months, plus or minus 2 weeks, were eligible for the study if their parent understood and signed written informed consent for the study and an agreement that the data could be computed. Thus on any day of the week during which one of the specifically trained nurses was present, all eligible infants whose parents could understand and sign the written consent were included in the study. The study was approved by the French Data Protection Authority.

2.2. Data collection

In half-day consultations at the DEPSE, parents and infants are first seen by an assistant who asks for socio-economic (SES) information, checks addresses and country of origin, languages spoken at home, number of rooms in the home, occupation and level of education of parents, and number of pregnancies and children. Infants and parents are then seen by a pediatric nurse, who records perinatal history, developmental and medical history, sleep and feeding routines, behavioural difficulties, types of day care, and familial environment. The nurse measures weight, stature, and cranial perimeter. Following this, blood tests (cell count, iron level, lead level if necessary) are performed. There is also a complete physical examination by a pediatrician, Ear and Nose Tractus disorders (ENT), auditory and ophthalmological testing by specialists, psychological examination, and testing by a psychologist using the French-validated Brunet-Lézine developmental test. The ADBB scale assessment was made by a specifically trained nurse, immediately after examining the infant. All participants in the assessment were blind to the ADBB scores as assessed by the nurses.

The complete procedure takes about 3 h, with rests and snacks in a large waiting room provided with games and toys. All information is gathered in a file and entered into the DEPSE database. In this database, medical symptoms and disorders are classified according to wide medical categories: Ear and Nose Tractus disorders (ENT) disorders, Nervous system disorders, Pneumological disorders, Digestive disorders, Dermatological disorders, Relational & Developmental disorders, Thriving and Endocrine disorders. At the end of the procedure, the pediatrician in charge gathers the information and drafts a written report with recommendations which will be sent to the parents, to the family general practitioner or to the family pediatrician.

2.3. Variables studied

2.3.1. The Alarm Distress Baby Scale (ADBB)

The ADBB scale has been designed to assess withdrawal behaviour in infants. The scale has 8 items, rated 0–4: facial expression; Eye Contact; General Level of Activity; Self-stimulating Gestures; Vocalizations; Response to stimulation, Ability to engage in a relationship. Withdrawal behaviour was assessed using the ADBB by 3 pediatric nurses from DEPSE who volunteered for the study, thus forming a team who could assess all eligible infants, coming to the DEPSE on any week day. Each infant was assessed by one of the trained nurses. The three nurses had been trained by the first author to use the scale, in the course of four training sessions using a set of 30 video clips of infants, until they reached good inter-rater reliability assessed by a Kappa coefficient of 0.8 for withdrawn vs. non-withdrawn, and no more than a one point difference on each item of the scale on the training video clips. Their reliability was checked during the study, by random selection of one eligible child for each nurse, to be seen in a live examination by the first author and by monitoring of assessment difficulties.

2.3.2. The Revised Brunet-Lézine Test

This early childhood psychomotor developmental test, inspired by Gesell's scale, was developed in France from 1943 onwards for infants aged 2–30 months [19]. The test was revised between 1994 and 1996 on a sample of 1032 French children from 6 French regions. In the present study 3 experienced psychologists performed the Brunet-Lézine examination for the eligible infants, working blind to the ADBB scores. Only the global Developmental Quotient (QD, i.e. Developmental Age/Real Age) was used in this study. The cut-off of 100 was used to define developmental delay ($DQ < 100$) or not ($DQ \geq 100$).

2.3.3. Parental report of behavioural symptoms in the child (PRBSC)

This variable is defined as the parent's report on 4 of the following items: infant sleep problems, feeding problems, relational difficulties between parents and infant and behaviour problems with the child. These symptoms are routinely assessed by the nurse during her examination, and entered into the DEPSE database. In the Appendix are displayed the data items which are routinely recorded on a standardised form in the DEPSE, and which were used in the analysis for all infants included in the study.

2.4. Statistical analysis

First the overall ADBB score distribution was studied and the proportion of withdrawal and its 95% confidence interval were estimated using the proportion of children with an ADBB score of 5 and over. For the second objective (identification of risk factors) univariate analyses followed by a multivariate analysis were performed. In the univariate analysis, the associations between presence (ADBB $\geq$5) or absence (ADBB $<$5) of withdrawal and all the variables defined in Appendix, were tested using a χ^2 test with 1 degree of freedom or a Fisher exact test as required. In order to find the variables that were independently associated with withdrawal a multivariate analysis with forward stepwise logistic regression was performed. All variables that had a p-value <0.10 in the univariate analysis were kept for this step. Two analyses were performed, one without the medical variables and one including them. All statistical analyses were performed using SAS software.

3. Results

Six hundred and forty infants were included, between September 2004 and July 2005 with a mean age of 16 months (range 13–19). Among infants eligible and present for a screening on the day the ADBB trained nurses were on duty, refusal rate was very low (1%, $n = 7$, mostly because the parents did not understand French sufficiently). Thirteen percent of the 640 infants ($n = 83$, 95% CI [10.4%; 15.6%]) had an ADBB score at 5 and over 5 on the ADBB. The

Table 1
Variables significantly associated with withdrawal behaviour ($N = 640$)

Variables	Not withdrawn ADBB <5% (n/N^*)	Withdrawn ADBB ≥5% (n/N^*)	χ^2 or Fisher (F)	p value
Gender (boy)	51% (285/557)	66% (55/83)	6.0	0.01
Family situation (difficulty)	1.5% (9/555)	6% (5/80)	F	0.03
Adoption (yes)	0.5% (2/557)	5% (4/83)	F	0.007
Twins (yes)	1.8% (10/556)	7% (6/82)	F	0.01
Type of care of the child (home)	60% (221/552)	73% (22/81)	4.4	0.03
Failure to thrive, obesity or Endocrine troubles (yes)	7% (37/556)	14% (12/83)	5.2	0.02
Parent's Report of Behavioural Symptoms in the Child PRBSC (yes)	14% (76/556)	39% (32/83)	30.1	410^{-8}
Developmental delay (yes)	16% (9/556)	13% (10/83)	23.7	110^{-6}

* n = number of case, N = number of children in this category; N is not always the same due to missing values.

mean ADBB score was 1.9, with a standard deviation of 2.5, a median of 1 and a range of 0–19.

Table 1 shows the variables recorded in the DEPSE screening that were found to be significantly associated with being withdrawn.

Among these infants, only 447 underwent a complete DQ assessment using the Brunet-Lézine test. Sixty two percent of the children for whom a Brunet-Lézine score was obtained had a score ≥100, 37% had a score between 80 and 100 and 1% had a score <80.

Withdrawal behaviour as assessed with the ADBB was significantly linked with being a boy rather than a girl ($p < 0.01$), with not living with both parents or not living with the mother, i.e. in foster-care or living with a relative ($p < 0.08$), with being adopted ($p < 0.0005$), with being one of twins ($p < 0.01$), with being taken care of at home rather than in day care ($p < 0.03$), with having psychological problems ($p < 0.0001$) and with having developmental delay ($p < 0.0001$). No significant correlation was found between withdrawal and the SES of the family, ethnic origin, gender, rank of birth, or with other medical pathology, except failure to thrive, endocrine problems or DQ. Among psychological difficulties, as reported by parents to the nurse, sleep disorders come first ($n = 59$, 43%), then relational and behavioral difficulties, ($n = 34$, 25%), feeding disorders ($n = 5$, 4%). Stressful family events ($n = 4$, 3%) and isolated motor retardation ($n = 4$, 3%) were less frequent.

Multivariate analysis was performed with all the variables in Table 1, since no variable had a p-value between 0.05 and 0.1. Five variables were found to be independently associated with withdrawal behavior in the final model when the medical variable of psychological disorders was included in the analysis (Table 2). When this variable was not included in the analysis, the same four other variables were found in the final model with odds ratios very similar to those in Table 2.

4. Discussion

This research is among the first large studies to assess the level of withdrawal behaviour in non-clinical infants [21,29]. Among 640 infants with a mean age of 16 months, prevalence of withdrawal behaviour is quite high in a low or medium risk sample. A Finnish study in Tampere [30] shows a comparable one-time incidence, among 144 infants aged 2–18 months with 8% at one assessment and only 3% in two assessments at a two week interval.

Correlation between ADBB scores and being a twin are strong and stable (OD = 7.2), (this is known to be a difficult situation for caregivers), as they are with the global level of psychological difficulties in the child (OR = 4.4), and with difficult family situations (OR = 4.5). As expected, no correlation was found between ADBB global score and family SES, their occupation or kind of work, the ethnic origin of the parents, or the age of parents. In an earlier study in Paris [14] with 60 infants aged 2–24 months in a baby welfare clinic, no significant correlation was found between the total score on the ADBB provided by the nurse and the age of the mother, parity, the age of the father, the age of the infant, or birth rank. No correlation was found between the number of days in perinatal hospitalization and the global ADBB score, but only 6.7% of the children were admitted in hospital postnatally. In this study withdrawal behavior was found more frequently among boys, which is in line with what is generally found at a later age, with a rather high odds ratio (OR = 2.1). More unexpectedly, children cared for at home are more likely to present withdrawal behaviour than children in day care centres. This may be linked with the high prevalence of post-natal depression in Paris [16,37,38], and with the generally widespread use of day care in France, which also involves middle class parents seeing it as a better choice for the child's development [39]. The higher level of withdrawal behaviour in adopted children is more unexpected (OR = 12.1), but not surprising,

Table 2
Variables independently associated with Withdrawal Behaviour (ADBB ≥5) using multiple logistic regression

	OR	95% CI	p value
Adoption (yes)	12.1	1.9–75.9	0.008
Twins (yes)	7.2	2.4–21.4	0.0004
Gender (boy)	2.1	1.2–3.5	0.007
Family situation (difficulty)	4.5	1.4–14.7	0.01
Parent's Report of Behavioural Symptoms in the Child PRBSC (yes)	4.4	2.6–7.6	<0.0001

considering it is a high-risk situation for care giving. However, with only 7 adopted children in the study, the confidence interval for this odds-ratio is very large (1.9; 75.5) so that its level should be interpreted with caution. No correlation was found between certain particular medical disorders or sensory difficulties and infant withdrawal behaviour, with the exception of failure to thrive or excessive weight, or endocrine disorders. A previous study had found a correlation between a score of 5 and over on the ADBB and some disorders in the follow-up of the child during the following year, particularly ENT infections, (otitis media and rhinitis), but also Gastro Oesophageal Reflux (GOR) [28], and another study showed that withdrawal behavior and infections were more frequent in securely attached children in day care when returning from holidays to day care [39].

One limitation of the study is that it is not a sample that is representative of the population of the Region Ile de France, since the children assessed in the DEPSE have parents who had received a proposal for a free assessment from the *CPAM de Paris*, and are therefore listed in the social security system. This excludes the families who have not received a proposal, or which have no formal inscription in the social system whatsoever. This is not therefore an epidemiological study. Another limitation id the fact that no structured diagnosis procedure was used to yield a diagnostic of a disorder in the child, and that psychological difficulties in the child were assessed only through parent's assessment of sleep disorders, feeding problems, relational difficulties with the child or behavioral difficulties, without using a structured interview or a validated questionnaire.

5. Conclusion

This data does nevertheless tend to confirm the validity of ADBB as a screening instrument which could be used routinely at reasonable cost [6,40]. Positive and negative correlations are clinically relevant, as sustained withdrawal behavior is not linked with age, rank of birth or SES of the family, as was already found in a smaller sample [28], but is linked with sleep, behavioral or feeding difficulties with the child as assessed by the parents, and generally with situations known to be at risk for the interaction with the child (twins, adoption). The ADBB scale was tested in Finland with front line professionals against the GRS scale for mother-infant interaction, at 2 and 4 months with 127 infants aged from 8 to 11 weeks and the scale was found to be sensitive and specific enough to detect early parent-infant relationship disorders [29]. A study in Israel comprising 85 children compared clinical and non-referred children and found that the ADBB score was clearly higher in the clinical sample [7]. Therefore withdrawal behaviour seems to be an important alarm signal to screen for in infancy as early as 2 months of age and until language fully develops; the ADBB scale appears to assist in detecting withdrawal behavior in infancy, enabling causes to be identified and intervention to occur at an earlier stage.

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Appendix

The following data items were recorded on a standardized form and used in the analysis for all infants who were included in the study. Items that required a yes or no answer are indicated yes − no, for other items the range of options is given in brackets.

- Age (months)
- Gender (male − female)
- Rank of birth (number)
- Twin (yes − no)
- Adopted (yes − no)
- Born full term (yes − no)
- Term (premature, post mature)
- Weight at birth (under or over 2300 g)
- History of neonatal hospitalization (yes − no, if yes number of days)
- Occupation of father (home, full time work, unemployed, part-time work, student)
- Occupation of mother (home, full time work, unemployed, part-time work, student)
- Profession of father (home, agricultural, employee, technician, high level professional)
- Age of father (over 50 or under)
- Age of mother (over 40 or under)
- Profession of mother (home, agricultural, employee, technician, high level professional)
- Family on social welfare (yes or no)
- Family stress factors or difficulties (yes − no)
- One parent died (yes or no)
- Type of day care (at home with parent, day care, non-parental at home, alternate care)
- Existence of a known disease in the child (yes − no)
- Developmental delay (level of DQ with Brunet.-.Lézine, under 100 vs. over or equal to 100)
- Parent's Report of Behavioural Symptoms in the Child PRBSC: Sleep disorders (yes − no), Feeding disorders (yes − no), Relational difficulties (yes − no), Behaviour problems (yes − no)
- ENT problems (yes − no), if yes otitis, rhinopharingitis, laryngitis, hypoacousia, repetitive ENT (more than 3 episodes of infection a year)
- Ophtalmological problems (yes − no, if yes strabism, amblyopia, hypermetropism, myopia)
- Pneumological problems (yes − no, if yes bronchitis, asthma, pneumopathy)
- Dermatology problems (yes − no, if yes eczema, infections, recent burns)
- Digestive problems (yes − no, if yes gastro oesophageal reflux, rectal or anal disorder)
- Thriving and endocrine problems (failure to thrive or advanced stature for age, equal to or less than 3sd, Body Mass Index (BMI) over the 97th percentile)

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