Alarm Distress Baby Scale (ADBB) in Assessing Social Withdrawal in Infants with an Underlying Organic Illness – Congenital Heart Disease, Prader Willi Syndrome, Cleft Lip and/or Palate – An Overview

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Abstract

Background
Infant social withdrawal, recognizable from two months onwards as assessed by the Alarm Distress Baby Scale (ADBB), provides a further measure of infant well-being. It has been widely used studying normal infants and those subjected to major adverse psychosocial environments. An overview is presented together with its use to evaluate infants with underlying organic illness and to understand the importance of the psychosocial well-being of the parents.

Methods
The theoretical basis for the ADBB is followed by a description of the observations and interactions that constitute the scale and how they are measured. Normative values are provided together with the prevalence and risk factors of infant withdrawal. Three papers are reviewed where the ADBB was scored in infants subjected to cardiac surgery, who were born with Prader-Willi Syndrome, or who had a cleft lip.

Results
Half of the infants with severe congenital heart disease (CHD) requiring cardiac surgery had elevated ADBB scores. That correlated not with the severity of the CHD or the surgery required but more to the maternal distress, anxiety, and depression. Almost two-thirds of infants with Prader-Willi syndrome exhibited social withdrawal. Intranasal oxytocin which improved the infants' sucking and interactions dropped the incidence down to one-fifth with improvement in parental sensitivity and dyadic reciprocity. Infants with a cleft lip ± a cleft palate had by 12 months decreased their ADBB scores to normative values. A high incidence of depression was noted in both parents which dropped following early repair of the cleft lip. The maternal well-being was found to be of greater importance than the nature of the abnormality and/or the surgery required.

Conclusion
Infant social withdrawal found in infants subjected to adverse psychosocial environments was also observed in infants born with significant organic abnormalities. The interrelationship to parental, especially maternal, well-being provides multiple avenues for potential interventions.

Introduction

Developmental psychology has taught us that infants are social beings, actively seeking interactions and contact, both physically and psychologically, with their environment. Infants are born with social and cognitive capacities that enable them to participate in human interaction[1,2]. However, it is important to consider that infant social behavior develops in the context of early parent-infant interaction as suggested by Feldman[3].

Infant's interaction skills include the ability to initiate and maintain eye contact with another person, to vocalize, to use and to imitate facial expressions, and to mimic body and head movements to initiate and sustain an interaction or provoke a reaction in others[2,4,5].

1.1. Social withdrawal

Social withdrawal behavior, from birth onwards but clearly as early as 2 months of age, is indicated by diminished or absence of either positive (e.g., smiling, eye contact, cooing) or negative behaviors (e.g., dampening of protest, and diminished crying) as described by Guedeney[6]. He suggested that ongoing social withdrawal is similar to a state of learned helplessness.

Dollberg et al.[7], argued that withdrawal behavior that persists has its roots in the organism's evolutionary biological-temperamental repertoire, as evidenced from animal studies. They suggested that under certain contextual circumstances, the natural mechanism of episodes of social withdrawal may lead to the development of a defensive strategy of sustained withdrawal, which acts against the infant's natural tendency to reach out to the social environment.

Microanalytic studies have shown that brief episodes of infant withdrawal appear frequently during parent–infant interactions, playing an important role in the regulation of early interactions[8-11]. Closing the eyes, turning the head and looking away are the infant's means of regulating the flow of interaction[12].That suggests infants might subsequently develop sustained social withdrawal behaviors as a way of handling repetitive or durable violations of his or her expectations within social interactions[13,14]. If such interaction is continually over intrusive or unresponsive, infants may learn that they have to consistently withdraw to reduce their arousal level[15]. Over a longer period of time, stronger protesting behavior can lead to increasingly pathological behavior in terms of avoidance, as observed in the Still Face experiment[16].

1.2. History of the concept

René Spitz[17] was one of the first authors to use this term in his clinical description of anaclitic depression in young infants aged around nine months. Robertson and Bowlby[18] described a three stage emotional reaction in young children —protest, despair, and withdrawal behavior—and eventually detachment in face of prolonged separation.

Subsequently, Engel and Reichsman[19,20] described sustained social withdrawal behavior as a defense mechanism in a 14 months infant, during a long hospitalization. Based on this experience, Engel and Schmale[21] developed the concept of conservation-withdrawal threshold as a biological mechanism which allows the system, under certain severe conditions, to disengage from the external environment in the service of conserving energy and assuring the organism's survival.

Meanwhile, Tizard and Rees[22] observed that institutionalized children who had been admitted to a residential nursery before the age of 4 months were largely unresponsive and emotionally withdrawn at 4 years. Fraiberg[23] described a group of pathological defenses -avoidance, freezing and fighting- observed in infants between 3 and 18 months of age who experienced severe danger and deprivation. These early defenses are embedded in the biological repertoire, and are related to the concept of withdrawal.

Another study that has contributed to the development of social withdrawal as a construct is the description of two infants with non-organic failure to thrive (NOFTT)[24]. This study highlighted the possibility to a better understanding of NOFTT within the context

of conservation-withdrawal. Infants with failure to thrive (FTT) were described as sad, depressed, withdrawn and irritable. Indeed, findings confirm that some FTT children prefer interacting with inanimate objects, make little eye contact, rarely vocalize, seem to dislike cuddling, and may engage in self-stimulatory activities, or in rumination. These behaviors are considered as signs of social withdrawal.

Nowadays, social withdrawal is recognized as a sign of infant distress in both organic disease and/or relational problems[7, 25-27]. It has been suggested that under certain contextual circumstances, the natural mechanism of episodes of social withdrawal may lead to the development of a defensive strategy of sustained withdrawal, which acts against the infant's natural tendency to reach out to the social environment[7].

1.3. The Alarm Distress Baby Scale (ADBB)

Infant psychological well-being have been investigated mostly based on mothers' perceptions[28-30]. Such perceptions may be prone to influences such as parental anxiety, depression or psychopathology and hypersensitivity to their infant's behavior or illness.

In 2001, Guedeney and Fermanian[31] created the Alarm Distress Baby Scale (ADBB) to facilitate a more structured observation of social behavior in infants from 2 to 24 months old, using an independent trained observer in a standardized situation[27, 31, 32]. The methodology is based on Winnicott's famous 'set situation'[33], as well as on the Neonatal Behavioral Assessment Scale[34] both of which provide a given stimulation and observe the way the infant makes use of it. The ADBB is a scale that therefore neither involves special apparatus nor a special sequence of prescribed interactions, and which does not require the parent's active interaction with the infant.

The ADBB scale consists of eight items concerning the behavior and features of the infant: 1) facial expression, 2) eye contact, 3) general level of activity, 4) self-stimulating gestures, 5) vocalizations, 6) response to stimulation, 7) relation between the infant and the clinician, and 8) attraction, defined as the ability of the infant to attract and maintain the clinician attention.

The assessment of social withdrawal is appreciated by the infant's reaction to stimulation. Pediatric consultation presents various stimuli in a short period of time and with a sufficiently identical sequence. Additionally, it is important that the clinician attempts to socially engage the infant –by talking, smiling, and touching-. The rating is done immediately after observation in a live situation or on videotape by a clinician trained in the scale. Previous studies have indicated that a total score of 5 or more is thought to be deviant and a sign of distress in the infant[31,35-39]. Higher total scores reflect higher levels of social withdrawal. One advantage of assessing an infant's social behavior with a relative stranger, rather than with his parent, is that it does not put the parent under any perceived pressure. However, the scale has been also validated and used in parent-infant settings.

The ADBB is able to detect a broad range of concurrent or emergent socioemotional problems, as excessive withdrawal from social interactions is a sign of infant distress, regardless of the cause. In this line, Feldman[3] argued the importance of infant withdrawal behavior as a sign of a dysregulation of parent–infant synchrony.

1.4. Prevalence

Infant social withdrawal has been found to have a prevalence in the community of between 3% and 13%[14,40], with elevated scores found in clinical samples[41] and those with mother–infant interaction difficulties[39]. In a recent study conducted by Tauber et al.[42], in infants with a genetic diagnosis of Prader-Willi Syndrome, the prevalence of social withdrawal was 62%. These findings suggested the necessity of identifying the risk factors for social withdrawal.

1.5. Risk factors

The relationship between the baby and her environment is a set of bidirectional processes, where the baby is not only affected

by the influences of his environment, but also is very active. According to Feldman[3] the interaction is the mutual influence between two subjects with reciprocity and interdependence. Given the bidirectional relationship between infants and their caregivers, both influence each other by their expressive displays, behavior and affective states[43]. When one of the partners is not healthy, whether it is the mother (e.g. depression; neglectfulness) or the infant (e.g. being ill, emotional distress), the interactive process is affected[44].

Risk factors related to parent-infant interaction have been identified. It has been observed that poor mother-infant interaction patterns are related to infant social withdrawal[35,39,45]. Specifically, Dollberg et al.[7] observed more negative relational patterns in mother-withdrawn infant dyads in terms of higher maternal intrusiveness, lower reciprocity, and poorer infant involvement.

Infant social withdrawal may also stem from temperamental or genetic susceptibility[41], from sensory integration disorders, or from impairments and disabilities. Risk factors that have been associated to social withdrawal are prematurity[46,47], sleep and feeding disorders, crying and irritability[48], parent's mental health[49,50]; attachment disorders, acute and severe pain, auditory and /or visual deficits, and autistic spectrum disorders[3,27,32].

1.6. Implications on development

Infant social withdrawal, by definition, makes the infant less available for interaction, either because of factors relating to their own capacity to regulate stimulation or factors relating to the care, such as experiencing their caregiver as being emotional unavailable[51]. Besides, social withdrawal reduces infants' developmental opportunities afforded by the interpersonal interactions. Accordingly, infant social withdrawal has been associated with long-term developmental outcomes[52] with significant positive correlations between infant social withdrawal at six months and increased scores on atypical social behavior and with lower scores on attention scales at 30 months. Molteno et al.[53] observed that children diagnosed with Fetal Alcohol Syndrome -at five years- showed greater social withdrawal and lower levels of response and activity at six months of age. Infant social withdrawal behavior was the strongest predictor of children's IQ at nine years old. Hence, optimal development within the infant's potentialities is not achieved when the infant is withdrawn for any length of time. Social withdrawal behavior in infants is an important alarm signal to screen for, especially in the context of medical conditions.

1.7. Objectives

The aims of this study were to review previous reports of social withdrawal in infants with an underlying organic illness, and to determine the relative contribution of the mental well-being of parents compared to the severity of the infant's illness on the social withdrawal.

2. Review of previous studies of social withdrawal in infants with an underlying medical condition

Three medical conditions are reviewed in detail – namely social withdrawal in infants with congenital heart disease[54], Prader-Willi syndrome[55] and cleft lip and palate[56].

2.1. Maternal Distress and Infant Social Withdrawal Following Infant Cardiac Surgery for Congenital Heart Disease[54]

In the context of major organic illness in infants, previous studies have noted high levels of stress on parents[57-59] including those with congenital heart disease (CHD) where surgery in infancy is often required[60,61]. Symptoms of depression and anxiety[62] or trauma reactions[63] have been observed in mothers of children subjected to cardiac surgery[61,64,65].

The aims of Re's paper[54] were to review infant responsiveness using the ADBB scale as a standardized objective observational measure of social withdrawal, and to explore its association with measures of maternal distress (Edinburgh Postnatal Depression Scale[66], Spielberg State-Trait Anxiety Scale and Parenting Stress Index-Short form[67]).

This study involved 22 Australian mothers whose infants had cardiac surgery for congenital heart disease (CHD) which included the full spectrum of the more severe CHD abnormalities. High levels of infant social withdrawal and maternal distress were observed when the mothers and infants were assessed, the latter being 2 months or older.

Utilizing the ADBB scale, 10 of the 22 infants were socially withdrawn, 6 of whom had very high scores suggesting severe withdrawal. The elevated ADBB scores also suggested these infants' well-being was at risk beyond their physical well-being. The ADBB scores were among the highest levels reported in the literature[7,14,39,40]. Interestingly, infant social withdrawal was not significantly associated with the severity of the CHD and the complexity of the surgery required.

The EPDS revealed a markedly higher risk for depression in the mothers of those infants compared to those in the Australian community: 59% compared to 15.5% in the study by Buist et al.[68]. Mothers of infants needing cardiac surgery were almost five times more likely to experience depressive symptoms when the infant was two months of age than might be expected in the general community.

Two-thirds of mothers reported anxiety (according to the STAI-S, Speilberger[67]. Almost a third of mothers exhibited high levels of parenting stress on the PSI-SF[69]. Very clear associations between maternal distress (depression, anxiety and stress) and infant social withdrawal were found: when mothers reported more symptoms of depression, anxiety and stress, their infants were more withdrawn. Interestingly, all mothers whose distress scores were within normal levels (non-clinical) on each of the scales had "not withdrawn" babies.

The paper concluded that maternal distress had a greater effect on infant social withdrawal as a sign of emotional distress than the severity of the cardiac abnormality. Besides, this study reported, probably for the first time, a standardized objective assessment of infant social withdrawal (ADBB) following cardiac surgery for CHD rather than relying on maternal reports[28,29].

2.2. Dysynchrony and Perinatal Psychopathology Impact of Child Disease on Parents-Child Interactions, the Paradigm of Prader Willi Syndrome[55].

Prader-Willi syndrome (PWS) is a rare genetic disorder which comprises several phases and is associated with behavioral disorders in the teenager. From birth to 9 months, infants with PWS display severe hypotonia, poor interactions, with a poor suck that may cause life-threatening complications. This research was based on the paradigm that infants with Prader-Willi syndrome display early endocrinal dysfunction associated with severe hypotonia and early feeding difficulties, which triggers impairments in parent-infant interaction. Tauber et al.[42] reported that early treatment with oxytocin (OXT) restored sucking after birth, which had been shown previously in animal studies. Tauber et al.[42] conducted a phase 2 escalating dose study of a short course (7 days) of intranasal OXT administration, in infants with PWS under 6 months old, who received 4 IU of OXT either every other day, daily, or twice daily. They found that OXT was well tolerated in infants with PWS and improved feeding and social skills.

In the study under review, Viaux-Savelon et al.[55] detailed early interaction improvement after OXT intranasal infusion based on reviewing infant social engagement and parent-infant synchrony. Eighteen French infants with a genetic diagnosis of Prader Willi syndrome (PWS) participated in this study. Social withdrawal behavior (ADBB scale[31]) and the mother-infant interaction (Coding Interactive Behavior CIB[70]) were evaluated while feeding, before and after the treatment of intranasal oxytocin. The authors reported that, at baseline, the median ADBB score was 6.5 with 62% of the infants clearly "withdrawn". After treatment, the median score significantly dropped to 3.5 and 81% of infants did not show social withdrawal. Significant improvement was observed on 4 of the 8 items: facial expression, eye contact, general level of activity and relationship. Of interest self-stimulating gestures were not

observed at baseline assessment nor after treatment.

Besides, Viaux-Savelon et al.[55] also observed significant improvements after treatment in parental sensitivity, dyadic reciprocity, child social engagement and child state evaluated by the Coding Interactive Behavior[70]. They reported that initially, the infants showed hypotonia, low expressiveness for positive or negative emotions, fatigability and poor involvement in the interrelationship, as they were socially withdrawn. The parents demonstrated a tendency to adapt to their infants' difficulties. However, their interactions were tense, restrictive and frequently intrusive with a "forcing" component that increased during the stressful feeding situation. After OXT treatment, the infants were more alert, less fatigable, more expressive, and had less social withdrawal. They initiated mutual activities and were more engaged in interactions through gaze, behavior, and vocalizations. The infants showed a improved muscle tone with better handling. It was suggested that these improvements helped the parents to be more sensitive, since they had become more aware of their infant's social signals. In general, the dyad displayed a better reciprocal exchange.

In summary, positive effects of OXT treatment on PWS infant behavior and social engagement and on parent-infant interaction were observed and reported for the first time in infants with PWS. That was reflected by the ADBB scores where the initial level of social withdrawal behavior in this sample was very high, compared to the level observed in community samples. The profile on the ADBB scale was specific with severe withdrawal and delay in response to stimulation but without self-stimulating gestures. In conclusion, the Prader-Willi paradigm highlighted the necessity to support early parent-infant relationship. OXT treatment showed in a relatively short time how the improvement of infant's ability (gaze, tonicity, suck, attentiveness) may impact at the same time on the parental state and the synchrony of the dyad in a positive way.

2.3. The impact of having a baby with cleft lip and palate on parents and on parent-baby relationship: the first French prospective multicenter study[56].

This article reported a prospective, longitudinal and multicenter French study of 158 infants with a cleft lip with or without a cleft palate (CLP). The main objective was to explore the effect of the malformation (CLP) on the infant's social withdrawal, the parent's mental health and the parent-infant relationship.

Social withdrawal behavior was evaluated by the Alarm Distress Baby scale (ADBB)[31] during follow-up consultations. The parent's mental health was assessed by the Parenting Stress Index (PSI)[69], the Edinburgh Postpartum Depression Scale (EPDS)[66] and the Impact on Family Scale (IOFS)[71]. All evaluations were done at 4 and 12 months after the infant's birth.

The incidence of social withdrawal among infants with CLP was 13% at 12 months of age, which is the same level as that observed in community studies in France[40]. The authors noted that the ADBB scores decreased from 4 months to 12 months. Interestingly, Grollemund et al.[56] found that the timing of surgery or the type of malformation did not have an effect on infant social withdrawal behaviors at 4 and 12 months postpartum. They also observed higher postpartum depression scores in both mothers and fathers of infants with CLP, compared to the general population at the 4 and 12 months assessments. Of interest, parents who had been informed of a prenatal diagnosis of CLP were better prepared to accept the waiting time between birth and the first surgical procedure, in comparison with the parents who learnt of the CLP diagnosis at birth. Furthermore, when the surgery was performed early (during the first three months of age) the maternal distress decreased significantly at the 12 month assessment compared to those where their infant had surgery later.

This was the first study using simultaneous and independent assessments of parents and infant's mental health in the context of CLP. The ADBB assessment of infants with CLP enabled a more objective study of the infant's

well-being and its impact on the parent-infant relationship. In conclusion, the authors suggested that the clinical evaluation by surgeons should include a parental screening for symptoms of depression and anxiety at the time of their infant's first evaluation. It is important that medical teams take into account the multiple challenges these parents have to overcome in this first year of their infant's life: accepting the fact that they have a baby with a malformation; seeing this cleft for the first time at birth; coping with the stares of others; persisting and not giving up as parents when confronted with the refusal, failure, and uncertainties of the first breast or bottle feeds; returning repeatedly to the hospital; coping with the anxiety of the first operation; having to witness the baby's discomfort after surgery; having to care for their baby; and once again accepting their baby after the changes brought about by surgery. The value of involvement of a mental health professional for those parents and baby becomes especially important for any infant with a CLP.

3. Discussion

The value of an objective assessment of infant well-being is well illustrated by the three studies detailed above which studied infants with a major underlying abnormality. The ADBB scale enables not only to provide a measure of infant well-being but also is able to highlight the importance of the parent-infant relationship. Smith-Nielsen et al.[27] suggested that adding the ADBB to existing routine developmental health follow-up practices may well increase the value of health care workers' practice by improving their awareness of the emotional well-being of infants under study.

As Re et al.[54] stated, it is not surprising to observe high levels of distress in parents whose infants have a life-threatening condition from their CHD, and being subjected to cardiac surgery at such an early age with potentially fatal outcomes. Similar concerns were found in parents with infants with Prader-Willi syndrome and cleft lip and palate, with their specific stressors on the families. The relationship between parents and their infant has been studied mainly by questioning the parents, most often focusing on the mother. Grollemund et al.[56] also included the father's perspective, emphasizing the need to consider and also address the mental health of fathers in these contexts.

The studies reviewed in this paper were the first to use simultaneous and independent assessment of parents and infant's mental health in the context of important organic conditions. Interestingly, the levels of social withdrawal in each of the three clinical situations (CLP, Prader-Willi, neonatal cardiac surgery) described were not related to the severity of the medical condition, but rather to the level of stress and distress shown by the mothers of these infants. That those infants and their parents need be followed and evaluated goes without saying though the importance of addressing the mental health issues of the parents especially the mother remains. The studies demonstrated links between the mother's (and father's) mental well-being and the infant's capacity to engage with the social environment (e.g. Moe et al.[47]). Finally, it is important to recall that social withdrawal is considered a defense mechanism when infants are faced with serious organic disease and/or relational problems. That suggests a joint assessment of the mental health of both infants and parents and the need to address both. The value of using the relative simple but informative objective assessment of infant's well-being may suggest to others the value of the ADBB scale when studying the impact of other illnesses or congenital abnormalities that manifest during infancy.

Conclusion

The ADBB scale was found very helpful in objectively reviewing the well-being of infants with major organic abnormalities. Its measurement and resultant elevated scores, while aligned for correlations with the severity of the abnormality and the interventions required, were found to be more directly related to the anxiety and depression exhibited especially by the mothers of those affected infants. These interactions provide multiple avenues for potential intervention to improve outcomes. Serial ADBB measurements were able to provide an objective assessment of

improvement in the infants. The relative ease of carrying out the ADBB scale and the accredited training required to score it, would favour its more widespread use in the study of infants born with various malformations and/or experiencing ill-health as a result of serious illness.

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