

Stereotypies in Children With a History of Early Institutional Care

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Objectives: To investigate the prevalence of stereotypies in children with a history of early institutional care, evaluate the efficacy of a foster care intervention compared with institutional care on the course of stereotypies, and describe correlates in language, cognition, and anxiety for children who exhibit stereotypies.

Design: Randomized controlled trial.

Setting: Institutions in Bucharest, Romania.

Participants: One hundred thirty-six children with a history of early institutional care.

Intervention: Comparison of a foster care intervention with continued care as usual in an institution.

Main Outcome Measures: The presence of stereotypies as well as outcomes in language, cognition, and anxiety.

Results: At the baseline assessment prior to placement in foster care (average age of 22 months), more than 60%

of children in institutional care exhibited stereotypies. Follow-up assessments at 30 months, 42 months, and 54 months indicated that being placed in families significantly reduced stereotypies, and with earlier and longer placements, reductions became larger. For children in the foster care group, but not in the care as usual group, stereotypies were significantly associated with lower outcomes on measures of language and cognition.

Conclusions: Stereotypies are prevalent in children with a history of institutional care. A foster care intervention appears to have a beneficial/moderating role on reducing stereotypies, underscoring the need for early placement in home-based care for abandoned children. Children who continue to exhibit stereotypies after foster care placement are significantly more impaired on outcomes of language and cognition than children without stereotypies and thus may be a target for further assessments or interventions.

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STEREOTYPIES ARE DEFINED AS repetitive, invariant movements with no obvious goal or function.¹ Stereotypies may occasionally be seen in typically developing children but are more commonly associated with a number of different medical conditions, including autism and mental retardation.² In addition, stereotypies are known to develop in association with atypical and especially restricted sensory environments or deprivation.¹ The association of stereotypies with disorders of the central nervous system suggests a neurological basis, but the underlying cause, pathophysiology, and possible approaches for treatment all require further investigation.³

In this article, we focus on stereotypies associated with early psychosocial deprivation. Most of our understanding of stereotypies associated with sensory-restricted environments comes from animal models. Stereotypies are the most common form of abnormal behavior found in caged animals, and animal models of deprivation-induced

stereotypies have been found across many species.⁴⁻⁶ In these animal models, stereotypies have been linked to alterations in the cortical-basal ganglia circuitry.⁷

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Human models of severe psychosocial deprivation in early childhood are understandably more limited. However, there are case reports of previously healthy children who developed stereotypies after exposure to prolonged extreme deprivation. In 1 case, a typically developing, healthy, 3-year-old girl in Thailand exhibited a number of motor stereotypies after being isolated in a cage for 6 years. Four years after her reintegration into the community, she showed considerable improvement including the elimination of stereotypies, but she continued to have many developmental difficulties, particularly with speech.⁸

Children raised in institutions provide another unfortunate but important group

in which to study the effects of extreme early psychosocial deprivation, including the presence of stereotypes. Institutionalized children are known to exhibit deficits across numerous domains of functioning, including physical, psychiatric, and cognitive outcomes.⁹⁻¹¹ There are a few reports in the literature suggesting an increased incidence and severity of motor stereotypes in this population, believed to be caused by the restricted sensory environment limiting typical input.¹²⁻¹⁴ Hypotheses for the functional significance of stereotypes in this environment include attempts at self-stimulating, coping mechanisms for self-soothing, or expressions of frustration or anxiety, particularly in children who lack adequate verbal communication skills.^{13,14}

An important issue that has not been addressed to date, to our knowledge, is a systematic study of the reduction or elimination of deprivation-induced stereotypes by restitution of enhanced caregiving environments. One report in the literature suggests a reduction in stereotypes after adoption and calls for additional research in this area. In this study of 46 children adopted from Romanian orphanages by families in British Columbia, Canada, 84% of caregivers reported the presence of stereotypes at the time of placement. The average age of the children at adoption was 18.5 months, and the mean time spent in institutions prior to adoption was 17.5 months. By the time the interview for the study was conducted (a median of 11 months after placement with the adopted family), 98% of stereotyped behavior problems were reported by caregivers to have shown some improvement or to be completely resolved.¹³ However, since children in this study were not randomly assigned to foster care placement, the application of these results to all children with a history of early institutional care is limited by selection bias. In addition, the study reports findings from only 1 assessment after placement and therefore does not provide data on stereotypes at later ages.

In the current study, we draw on data from the Bucharest Early Intervention Project (BEIP), a study of current and formerly institutionalized children in Romania. The BEIP is unique in its ability to evaluate directly the effects of early family placement rather than institutional care for young children because the participants were institutionalized children randomly assigned either to continued care as usual in the institution or to a foster care intervention. The BEIP is the first such randomized clinical trial to evaluate foster care as an alternative to institutional care for abandoned children.¹⁵ Because the children enrolled in this study were closely followed up longitudinally, this study also provides the unusual opportunity within the population of institutionalized children to investigate the course of stereotypes in children exposed to extreme deprivation and the presence of sensitive periods for the development or remediation of stereotypes.

This study addressed the following questions: (1) How many young children being raised in institutions exhibit stereotypes? (2) For children with a history of early institutionalization, does placement in a foster care intervention reduce or eliminate stereotypes? If so, is there an effect of timing of placement? (3) To characterize those children who continue to exhibit stereotypes compared with those whose stereotypes resolve, are there any correlates of stereotypes that we can identify in this population, particularly focusing on outcomes in language, cognition, and anxiety?

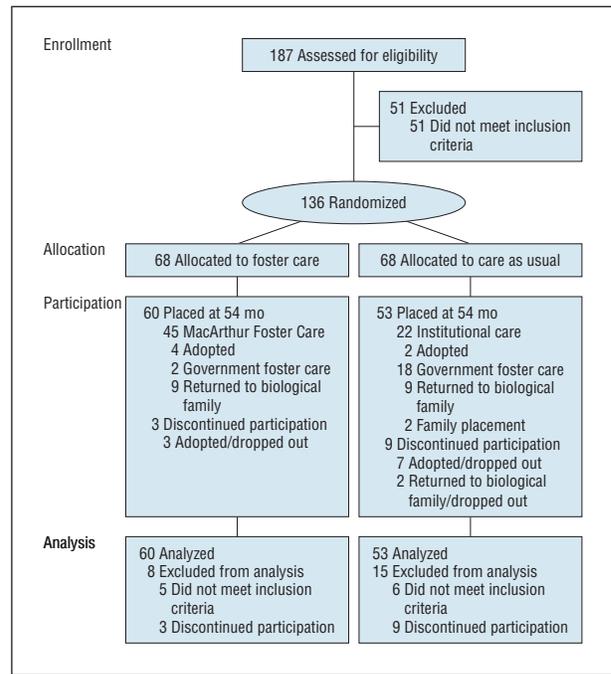


Figure 1. Flowchart of participants randomized to a foster care intervention or care as usual in the institution. Information is presented for the final assessment at 54 months.

METHODS

PARTICIPANTS

Participants for this study were children with a history of early institutional care enrolled in the BEIP. Children younger than 31 months living in 6 institutions in Bucharest, Romania, underwent screening for enrollment in this study. These children were originally placed in an institution at or close to the time of birth. Of 187 children who were screened, 51 were excluded for medical reasons, including genetic syndromes, fetal alcohol syndrome, and microcephaly.⁹ Of the remaining 136 children, 68 were randomly assigned to foster care placement (referred to as the foster care group [FCG]) and 68 were assigned to continued care as usual in the institution (the care as usual group [CAUG]). The foster care intervention was created through collaboration between study investigators and Romanian authorities, and the characteristics of the foster care system are discussed at length elsewhere.¹⁵ A thorough discussion of the ethical issues inherent to this study has been covered in previous publications.^{9,16,17}

The average age at foster care placement was 22.9 months with a range of 6 to 33 months. After a baseline evaluation prior to foster care placement, all children were reassessed at ages 30, 42, and 54 months. The flow of participants and causes of attrition by the last assessment at 54 months are described in **Figure 1**. All analyses were done following an intent-to-treat model, with children considered within their original group assignment even though many of the CAUG children were later placed in foster care.

INSTRUMENTS

Trained and reliable Romanian staff administered all assessments. For the instruments that rely on caregiver report, for children living with foster parents, the foster mother served as the reporter. For children living in institutions, an institutional caregiver reported on the child's behavior. If the child

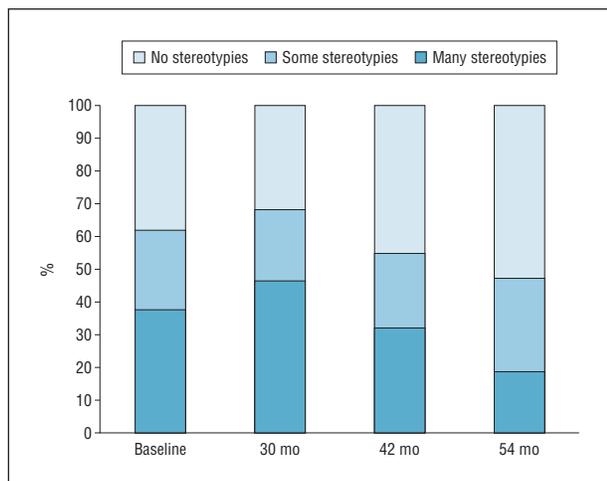


Figure 2. Percentage of children exhibiting none, some, or many stereotypies in the care as usual group at baseline and at follow-up assessments at 30 months, 42 months, and 54 months.

was identified by staff as having a favorite caregiver, this person was selected. If no favorite was identified, a staff member who knew the child well was chosen.

STEREOTYPIES

The Disturbances of Attachment Interview,¹⁴ a semistructured interview administered by a trained clinician to the child's primary caregiver, was used to assess stereotypies. Based on probes detailing the child's behavior, caregivers' responses were coded by interviewers as 0=none, 1=somewhat or sometimes, and 2=many or often. The Disturbances of Attachment Interview was administered to all children in the study at baseline, 30 months, 42 months, and 54 months.

LANGUAGE

The Reynell Developmental Language Scales¹⁸ were used to assess verbal comprehension (receptive language skills) and expressive language skills. The Reynell Developmental Language Scales are designed to measure language skills in young or developmentally delayed children and are intended for use with children between 18 months and 7 years of age. This measure was administered to all children in the study at the assessments at 30 months and 42 months.

COGNITION

Cognitive development was assessed at baseline, 30 months, and 42 months using the Bayley Scales of Infant Development¹⁹ and at 54 months with the Wechsler Preschool and Primary Scale of Intelligence.²⁰ The Bayley Scales of Infant Development, intended for infants from 1 to 42 months of age, assess a child's development in 3 domains: cognitive, motor, and behavioral. The child's score is reported as the developmental quotient (DQ). The Wechsler Preschool and Primary Scale of Intelligence measures intellectual functioning in verbal and performance domains and also provides a measure of the child's general intellectual ability (full-scale IQ).

ANXIETY

The Preschool Age Psychiatric Assessment,²¹ a structured psychiatric interview, was administered to the child's primary

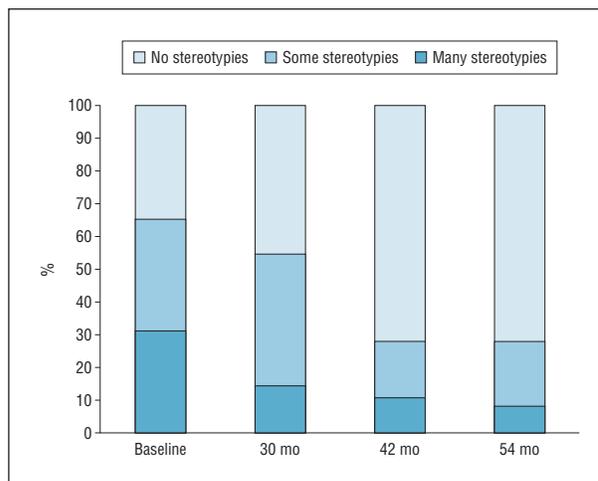


Figure 3. Percentage of children exhibiting none, some, or many stereotypies in the foster care group at baseline and at follow-up assessments at 30 months, 42 months, and 54 months.

caregiver at the 54-month assessment. If the caregiver reported symptoms, the interviewer collected information about frequency, duration, and dates of onset. This information was used to determine whether the symptoms met *Diagnostic and Statistical Manual of Mental Disorders* diagnostic criteria for various disorders. This assessment, intended for children aged 3 to 6 years, has been found to have test-retest reliability comparable with that of well-established measures for older children and adults.²² In this analysis, the outcome measure used from the Preschool Age Psychiatric Assessment was the presence of any anxiety diagnosis.

STATISTICAL ANALYSIS

To examine the prevalence of stereotypies, the Wilcoxon Mann-Whitney test was used. This test was chosen because the dependent variable, stereotypy score, is ordinal. Timing-of-placement analyses were done using Fisher exact test. Other analyses were done using independent-sample *t* tests for continuous variables and χ^2 tests or Fisher exact test for categorical variables. A *P* value lower than .05 was considered significant.

RESULTS

No significant relationship was found between stereotypies and sex, ethnicity, or birth weight at the baseline assessment or any of the follow-up assessments.

PREVALENCE OF STEREOTYPIES

The prevalence of caregiver-reported stereotypies is summarized for the CAUG in **Figure 2** and for the FCG in **Figure 3**. As expected, there was no statistically significant difference in the underlying distributions of stereotypies in the CAUG and the FCG at the baseline evaluation, prior to placement in foster care ($z = -0.20$; $P = .84$). At each follow-up assessment, the results indicated that there was a statistically significant difference between the distribution of stereotypies in the CAUG and in the FCG, with children in the FCG exhibiting fewer stereotypies (30 months, $z = -2.99$; $P = .003$; 42 months, $z = -3.36$; $P = .001$; 54 months, $z = -2.06$; $P = .04$).

As a comparison, the same assessments were administered to a group of children raised with their biological families in the greater Bucharest area. At the initial baseline evaluation, 1 of 61 children (1.64%) in this never-institutionalized group had many stereotypes, 12 of 61 (19.7%) had some stereotypes, and 48 of 61 (78.7%) had no reported stereotypes. At 30 months, no children in this group had many stereotypes, 1 of 52 (1.9%) had some stereotypes, and 51 of 52 (98.1%) had no stereotypes. At 42 months and 54 months, no children in this group were reported to exhibit stereotypes.

TIMING OF PLACEMENT IN FOSTER CARE INTERVENTION

Next, we looked within the FCG to see if there was a relation between timing of placement into foster care and subsequent stereotypes. Children in the FCG were divided into 3 groups based on age at placement into foster care: younger than 12 months, between 12 and 24 months, and greater than 24 months. At each follow-up assessment, the percentage of children who exhibited any stereotypes was lowest for the children placed youngest and highest for the children placed oldest (**Table**). The difference between the 3 age-at-placement groups was significant at the 30-month ($P=.006$) and 54-month ($P=.03$) assessments. The 42-month data show the expected ordering, but the difference was not significant ($P=.34$).

CORRELATES OF STEREOTYPES

For each of the following assessments conducted after the baseline evaluation, analyses were performed with subjects stratified by group assignment (CAUG or FCG). Within each of these groups, we compared outcomes for children with and without stereotypes.

LANGUAGE

At all assessments, children with stereotypes had lower mean scores for verbal comprehension and for expressive language than children without stereotypes, but these differences were significant only in the FCG and not in the CAUG. In the CAUG at 30 months, the mean comprehension score was 17.1 for children with stereotypes and 19.7 for children without stereotypes ($P=.19$). The mean expressive language score was 5.30 for children with stereotypes and 6.31 for children without stereotypes ($P=.53$). At 42 months in the CAUG, the mean comprehension score was 32.0 for children with stereotypes and 33.1 for children without stereotypes ($P=.52$). The mean expressive language score was 17.4 for children with stereotypes and 19.1 for children without stereotypes ($P=.39$).

In the FCG at 30 months, the mean comprehension score was 17.9 for children with stereotypes and 25.4 for children without stereotypes ($P=.003$). The mean expressive language score was 5.4 for children with stereotypes and 10.8 for children without stereotypes ($P=.009$). At 42 months in the FCG, the mean compre-

Table. Percentage of Children in the Foster Care Group Exhibiting Stereotypes by Age Group at Entry Into Foster Care^a

Age at Placement, mo	Children With Stereotypes, No. (%)		
	30 mo	42 mo	54 mo
<12	8 (25)	7 (14)	7 (0)
12-24	23 (39)	25 (20)	23 (17)
>24	31 (74)	32 (38)	30 (43)

^aNumbers of children differ because results were not available for some children at all evaluations. For a summary of the flow of participants and causes of attrition by the last assessment at 54 months, see Figure 1.

hension score was 34.2 for children with stereotypes and 38.5 for children without stereotypes ($P=.08$). The mean expressive language score was 17.8 for children with stereotypes and 24.6 for children without stereotypes ($P=.001$).

COGNITION

At all assessments, children with stereotypes had lower mean scores for cognitive outcomes than children without stereotypes, but this difference only achieved significance within the FCG. In the group of all ever-institutionalized children (both the CAUG and FCG) at baseline, the difference on DQ between children with and without stereotypes was not statistically significant (71.08 vs 75.75; $P=.07$). Within the CAUG, differences on DQ or full-scale IQ between children with and without stereotypes were not statistically significant at 30 months (74.74 vs 78.75; $P=.21$), 42 months (72.46 vs 78.72; $P=.09$), or 54 months (70.91 vs 75.21; $P=.25$). At 54 months, in addition to full-scale IQ, scores were also obtained for performance IQ and verbal IQ. Neither performance IQ (71.00 vs 76.25; $P=.14$) nor verbal IQ (76.22 vs 78.86; $P=.49$) was significantly different for children with and without stereotypes in this group.

In the FCG, differences on DQ or full-scale IQ between children with and without stereotypes were statistically significant at 30 months (77.46 vs 84.92; $P=.02$), 42 months (75.12 vs 87.53; $P=.004$), and 54 months (72.18 vs 84.55; $P=.02$). In the FCG, the difference between the mean score for children with and without stereotypes was statistically significant for verbal IQ (74.24 vs 88.26; $P=.007$), but not performance IQ (75.71 vs 84.48; $P=.08$).

ANXIETY

In the CAUG, 12 of 24 children (50%) with stereotypes had an anxiety diagnosis, and 9 of 28 children (32%) without stereotypes had an anxiety diagnosis, a difference that was not statistically significant ($P=.19$; Pearson χ^2 test). In the FCG, 5 of 17 children (29%) with stereotypes had an anxiety diagnosis, and 5 of 42 children (12%) without stereotypes had an anxiety diagnosis, a difference that was not statistically significant ($P=.13$, Fisher exact test).

To our knowledge, this study is the first randomized trial of foster care as an alternative to institutional care and therefore the first clinical trial to determine if family care can remediate stereotypies in children being raised in conditions of social deprivation. We believe that this study has several important findings. First, a striking number of children who have a history of institutional care exhibit stereotypies, with the majority of children in institutional care at our baseline evaluation showing such behaviors. At follow-up evaluations, we described a general course of declining prevalence of stereotypies over time in both the CAUG and FCG. The stereotypy scores in the FCG, however, were significantly lower than in the CAUG, suggesting a beneficial/moderating role of the foster care intervention. This pattern of resolution of stereotypies with time contrasts with the literature on the longitudinal course of stereotypies seen in typically developing, healthy children, suggesting a different course for deprivation-induced stereotypies.²³ This is consistent with a previous report in the literature on the resolution of stereotypies in children adopted out of institutions.¹³

To investigate further the foster care intervention, we next considered timing of placement into foster care. Results indicate that being placed in families significantly reduces stereotypies, and with earlier and longer placements, reductions become larger. These results underscore the need for early placement in home-based care for abandoned children.

Finally, we investigated possible correlates in language, cognition, and anxiety among children to characterize children who exhibit stereotypies compared with those who do not. The results indicated a pattern of significant differences on language and cognition measures within the FCG, but not within the CAUG, for children with and without stereotypies.

These data suggest that for children with a history of early severe deprivation who are subsequently exposed to a family environment, 2 subgroups emerge. Most children experience remediation of stereotypical behavior, suggesting that recovery is possible. However, a significant subset of this population continues to exhibit stereotypies. These children are also significantly more impaired on outcomes of language and cognition than the children who do not exhibit stereotypies. Since stereotypies are an easily observable measure (more so than extensive cognitive or language assessments, for example), this may point to a potential strategy for identifying young children with a history of early deprivation who may be most at risk for poor outcomes in language and cognition and who may benefit from further assessments. Further research is needed to clarify what might be different about these 2 groups.

Though stereotypies may serve as a marker of poor outcomes in these domains, we do not believe it is appropriate to suggest from these data that stereotypies are a cause of lower cognitive outcomes. It remains unclear what precisely the presence of stereotypies may indicate in respect to brain damage or development in this population. Previous research has identified a variety of ways

in which early institutionalization of young children affects brain development, including alterations in metabolic, physiological, and neurochemical activity; changes in the size of certain areas of the brain; and diminished white matter connectivity.²⁴⁻²⁷ However, the relation between these identified neurobiological changes and the development of deprivation-induced stereotypies requires additional research.

There are several limitations of this study. First, the data on stereotypies used in these analyses were gathered through caregiver report, not direct observation by study staff. This approach was chosen because the caregiver had observed the child in multiple settings on numerous occasions and thus we felt was able to provide a more accurate assessment than could be obtained through a brief observed session. Previous reports on stereotypies in typically developing children have suggested that children are most likely to exhibit this behavior when alone in a crib or when irritable or upset,²⁸ and we believe that these situations are more likely to be observed by a caregiver than during a time in which the child is actively engaged in a study assessment. However, relying on data from caregivers introduces the possibility of reporter bias. For example, foster parents may be more likely to report stereotypies than institutional caregivers if they spend more time observing the child or if they are more likely to believe that reporting the behavior may be beneficial to the child. Conversely, it is also possible that foster parents may minimize this type of behavior if they believe it will reflect poorly on the care they are providing or that it will mark their child as abnormal.

Additional limitations include that motor stereotypies were grouped together in these analyses and the impact of a particular type of stereotypy (for example, rocking back and forth as compared with a more elaborate stereotypy) was not considered separately. To address this, further analyses could consider the quality and nature of stereotypies in more detail. In addition, the data reported in this study extend only through age 54 months. Future work should be done to investigate if the results found herein persist into adolescence and adulthood.

In summary, these findings contribute to our understanding of the prevalence of deprivation-induced stereotypies and to associated deficits in language and cognition in children who exhibit this behavior. The results also provide evidence that young children with a history of early institutional care who are placed into foster care may experience significant recovery from stereotypies. These findings have implications beyond the unique population of children in orphanages, as the extreme example of institutional care can help us to better understand the impact of deprivation on children in many settings.

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Fox. *Drafting of the manuscript*: Bos, Fox, and Nelson. *Critical revision of the manuscript for important intellectual content*: Zeanah, Smyke, Fox, and Nelson. *Statistical analysis*: Bos and Fox. *Obtained funding*: Zeanah and Nelson. *Administrative, technical, and material support*: Zeanah and Nelson. *Study supervision*: Zeanah, Smyke, Fox, and Nelson.

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Care more particularly for the individual patient than for the special features of the disease.
—Sir William Osler, address to the students of the Albany Medical College, 1899