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The role of stereotypies in overselectivity process in Rett syndrome

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Abstract

Ten Rett syndrome (RS) girls and 10 control girls executed an attentional task in which a complex stimulus was shown followed by individual stimuli presented with distractors. Participants had to discriminate previously presented stimuli from distractors. RS girls carried out the task both in a condition with the containment of stereotypies and in a no-containment condition. Overselectivity occurred in RS since patients failed to discriminate about 1/3 of the individual stimuli. There were no statistical differences with respect to the number of correct responses in the two conditions; RS girls learned quickly when their stereotypies were contained as opposed to when the containment of stereotypies was lacking.

Keywords: Rett syndrome; Stereotypies; Learning; Overselectivity; Attention

Rett syndrome (RS) is a developmental disorder, predominantly affecting females (incidence 1/20,000–1/15,000), resulting in severe mental retardation and neuro-behavioral disability. In late infancy, after a period of apparent normal development (Nomura & Segawa, 1990), RS patients undergo striking developmental regression (Hagberg, 1993; Moeschler, Charman, Berg, & Graham, 1991) characterised by the loss of pre-existing hand use (such as object reach, grasp, and manipulation) and by the appearance of distinctive hand stereotypies (hand wringing, tapping, and mouthing). In spite of severe disabilities in the post-regression phase, often

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individuals regain social interest and are relatively stable for an extended period before they manifest progressive motor deterioration as a result of weakness, wasting, and dystonia (Hagberg, 1993, 2002). During this period, crude self-feeding capabilities may be retained, but voluntary hand use is generally exceedingly limited and hand stereotypies are pervasive (Fontanesi & Haas, 1988; Keer, Montagne, Mus, & Stephenson, 1987). Some studies (Perry, Sarlo-McGravy, & Haddad, 1991; Witt-Engerström, 1990) have reported that patients often remain visually attentive to objects and people, tracking their movements and even showing preferences by means of ''eye pointing'' (Baptista, Mercadante, Macedo, & Schwartzman, 2006). This may reflect their cognitive abilities however stereotypic behaviors could interfere with their learning process. Recent research has suggested that this conceptualization of stereotypy is accurate but incomplete (Guess & Carr, 1991; Kennedy, Meyer, Knowels, & Shukla, 2000), for example, Kennedy et al. (2000) analyzed the stereotypy of autistic students and found several patterns. For some students, stereotypy was a function of sensory and social reinforcement, while for others an undifferentiated patter of behavior occurred across conditions. These findings suggest that the causes of stereotypy can be a function of (a) positive sensory reinforcement, (b) negative sensory reinforcement, (c) positive social reinforcement, and/or (d) negative social reinforcement. As Berkson and Tupa (2002) showed, there is an increased probability of stereotypy inversely related to a person's intellectual functioning level. This impairment may occur because the resources of attention can be allocated to the involuntary movement (stereotypic) so the residual resources are very few for the codification process. The purpose of this study is to analyse how stereotypies influence attention processes in RS girls and to test whether stereotypy containment improves the attention.

One of the paradigms to study selective attention is overselectivity. This refers to atypically limited learning with respect to the range or to the number of stimulus features (Lovaas, Koegel, & Shreibman, 1979). Overselectivity is a widely acknowledged problem in the education of individuals with developmental disabilities like mental retardation and autism (Allen & Fuqua, 1985). Although the problem is widespread, literature about its remediation is remarkably limited (Schreibman, 1997).

Lovaas et al. (1979) reported restricted stimulus control in mentally retarded and autistic people. Their study involved initial discrimination training with complex stimuli followed by testing with individual stimuli, which were included in the complex ones, to determine how many individual stimuli had been previously coded. For example, if initial training used the complex ABC stimulus (that is, a stimulus constituted by the individual stimuli A, B, and C) presented as correct and the XYZ stimulus as incorrect (see Fig. 1), then, in the test phase, pairs of individual stimuli were presented in combination (A versus Y, B versus X, and so forth: Fig. 2) with the request to recognise which stimulus was the correct one.

When given such tests, individuals with development disabilities may respond appropriately only to a lower number of individual stimuli as compared to non-disabled individuals. Wilhelm and Lovaas (1976) reported reliable stimulus encoding by all three correct stimuli in typically developing children, two correct stimuli in children with moderate mental retardation, and only one or two correct stimuli in children with severe retardation. Restricted stimulus encoding has been documented with multiple stimuli differentiated along the same dimension (Koegel & Wilhelm, 1973; Wilhelm & Lovaas, 1976) as well as along different dimensions (differences in colour, form, and so on: Kovattana & Kraemer, 1974). The finding has been replicated by controlling the discrimination of stimuli presented individually (Dube, Kledaras, Iennaco, Stoddard, & McIlvane, 1993) and the discrimination of stimuli presented in multiple arrays (Stromer, McIlvane, Dube, & Mackay, 1993).