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Disrupted sensorimotor synchronization, but intact rhythm discrimination, in children treated for a cerebellar medulloblastoma

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ABSTRACT

The aim of this study was to investigate the temporal abilities of children treated by surgery for a malignant tumor in the cerebellum, both in the perception and the production of rhythm. Children with a diagnosed medulloblastoma and age-matched control children were tested in a rhythm discrimination task and a sensorimotor synchronization task. Their motor and cognitive capabilities were also assessed through a battery of age-adapted neuropsychological tests. The results did not show any significant difference in performance between groups for the discrimination task. On the contrary, children with cerebellar lesions produced longer and more variable inter-tap intervals (ITI) in their spontaneous motor tempo (SMT) than did the control children. However, the length and, to a lesser extent, the variability of their SMT decreased after a synchronization phase, when they had been instructed to tap in synchrony with a beep. During the synchronization task, the children with medulloblastoma succeeded to modify the length of their ITI in response to an auditory rhythm, although with better success when the inter-stimuli intervals (ISI) were shorter than when they were longer than the ITIs of their own SMT. Correlational analyses revealed that children's poorer synchronization performance was related to lower scores in neuropsychological tests assessing motor dexterity and processing speed.

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1. Introduction

Medulloblastoma is a highly malignant embryonic tumor of the cerebellum (von Hoff et al., 2010). It is the major cause of malignant brain tumors in childhood that is diagnosed in 75% of cases before the age of 9 years (Becker & Jay, 1990; Roberts, Lynch, Jones, & Hart, 1991). The treatment of a medulloblastoma is surgery of the tumor in the cerebellum, with craniospinal radiotherapy and chemotherapy (Taillandier et al., 2011). The consequences of the surgery are cognitive and motor deficits (Callu et al., 2008, 2009; Grill, Kieffer, & Kalifa, 2004; Grill, Viguier, et al., 2004; Kieffer-Renaux et al., 2005). Some authors have argued that it could also affect the processing of temporal rhythms (e.g., Clarke, Ivry, Grinband, Roberts, & Shimizu, 1996; Ivry & Keele, 1989; Rao et al., 1997; for a recent review see Spencer & Ivry, 2013). The impairment of children with cerebellar lesions in social interaction, speech control or music could originate from basic deficits in the processing of rhythm (e.g., Feldman, 2007; Patel, 2006; Wing, 2001). The aim of our study was thus to examine if children with cerebellar lesions due to surgery of a medulloblastoma show deficits in discrimination and motor production of temporal rhythms.

The abilities in human beings to discriminate rhythms and to produce motor rhythms have been extensively studied since Fraisse's (1974) seminal book on the psychology of rhythm (for a review see Repp & Su, 2013). However, although education of children in rhythm has often been recommended, only a few studies have experimentally investigated the development of rhythmical activities. Some studies have examined spontaneous motor tempo (SMT) in children of different ages, by asking them to tap with their finger on a button at their preferred rate (Drake, Jones, & Baruch, 2000; McAuley, Jones, Holub, Johnston, & Miller, 2006; Provasi & Bobin-Bègue, 2003). Results have revealed that the SMT slows down with age during childhood (Bobin-Bègue & Provasi, 2008; Drake et al., 2000; McAuley et al., 2006; Provasi & Bobin-Bègue, 2003) and adulthood (Vanneste, Pouthas, & Wearden, 2001). The mean tapping interval shifts from 400–500 ms at ages of 2–7 years old, to 600 ms for young adult ages, and 700 ms for older ages (75 years). More importantly, the variability (*SD*) of SMT, i.e., the variability in inter-tap interval (ITI), decreases with age. Concerning sensorimotor synchronization, when participants have to tap in synchrony with isochronous metronome rhythm, adults often anticipate the beat, showing negative asynchrony between their finger tap and the beat of the metronome of about 10 ms (Miyake & Yohei, 2004). This negative asynchrony is rarely found in young children. In addition, as for the SMT, the variability of asynchronies appears to be greater in children than in adults (Repp, 2005). Recently, Bobin-Bègue and Provasi (2008) used a synchronization–continuation task to examine the abilities of very young children, aged from 1 to 3 years old, to synchronize their manual taps with external rhythms that were similar, faster or slower (10, 20, 30 and 40%) than their own SMT. Their data revealed that these young children were able to synchronize their taps to an external rhythm (rhythmic sounds), but only when it was close to their own SMT. In the post-synchronization–continuation phase, when the sound was removed and the child was supposed to continue to tap at the same rhythm, the children succeeded better in decreasing than in increasing the tapping rhythm, but returned to their initial SMT when the external rhythm was farther away (i.e., faster or slower than 20%) from their own SMT.

There has been no research on the capabilities in discrimination and production of rhythms of children with cerebellar lesions. However, studies in adults with cerebellar lesions have reported higher variability in SMT (finger tapping) compared to healthy adults (Ivry, Keele, & Diener, 1988; Molinari et al., 2005; Molinari, Leggio, & Thaut, 2007). Adult cerebellar patients also produced more variable ITIs during synchronization–continuation phases (e.g., Claassen et al., 2013; Schlerf, Spencer, Zelaznik, & Ivry, 2007; Witt, Laird, & Meyerand, 2008). Cerebellar dysfunction thus impairs performance in a sensorimotor synchronization task (Nicolson & Fawcett, 2011). We can thus assume that children with cerebellar deficits due to treatment of medulloblastoma may also show poorer performance than their age-matched control children both in SMT and during a synchronization task.

However, there is a debate, which is not yet settled, on the components responsible for deficits in sensorimotor synchronization in cerebellar patients. According to Wing and Kristofferson's model (1973), in the sensorimotor synchronization task, the perception of temporal intervals in a rhythmic sequence must be differentiated from the production of a motor rhythm synchronized to this rhythmic sequence. In effect, two separate components can explain the timing variance in sensorimotor synchronization: a "central timekeeper" determining when to initiate the response, and motor effectors involved in the motor implementation of the response. The timekeeper-related variance would be the major source of the variability in ITI of temporal synchronization, as it depends on the value of the inter-stimulus intervals (ISI), with a greater variance for longer ISIs. On the contrary, the contribution of the motor component to the variance of synchronization would be lower, at least in healthy individuals, as it would be constant for different ITI values. In relative terms, the proportion of variance introduced by this motor component would be more substantial for short ITIs than for long ones, i.e., opposite to the timekeeper effect.

It is now well established that lesions of the cerebellum affect motor control and the integration of sensory and motor information underlying coordinated movements (Manto & Oulad Ben Taib, 2013). Consequently, it is reasonable to assume that low performance in a sensorimotor synchronization task for patients with lesions of the cerebellum would be mainly due to a deficit in the basic motor component of synchronization, i.e., causing motor variability during repetitive tapping (Ivry & Keele, 1989; Spencer & Zelaznik, 2003). Ivry (1996) nevertheless suggested that the perception and production of rhythms share a common temporal system. It has been reported, however, that the impact of cerebellar lesions on synchronization may be a function of their localization, with an increase in variability associated with the motor response after medial cerebellar lesions, but an increase in clock-related variability after lateral lesions (Fierro et al., 2007; Ivry et al., 1988). Finally, it remains unclear whether there is a region in the cerebellum that would be related to timing functions, independently of the motor component (Rao, Mayer, & Harrington, 2001; Tracy, Faro, Mohamed, Pinsk, & Pinus, 2000).