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Prenatal testosterone and stuttering

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ABSTRACT

Background: The prevalence of stuttering is much higher in males compared to females. The biological underpinnings of this skewed sex-ratio is poorly understood, but it has often been speculated that sex hormones could play an important role.

Aims: The present study investigated a potential link between prenatal testosterone and stuttering. Here, an indirect indicator of prenatal testosterone levels, the Digit Ratio (2D:4D) of the hand, was used. As numerous studies have shown, hands with more "male" characteristics (putatively representing greater prenatal testosterone levels) are characterized by a longer ring finger compared to the index finger (represented as a lower 2D:4D ratio) in the general population.

Study design, subjects, outcome measures: We searched for differences in the 2D:4D ratios between 38 persons who stutter and 36 persons who do not stutter. In a second step, we investigated potential links between the 2D:4D ratio and the multifaceted symptomatology of stuttering, as measured by the Overall Assessment of the Speaker's Experience of Stuttering (OASES), in a larger sample of 44 adults who stutter.

Results: In the first step, no significant differences in the 2D:4D were observed between individuals who stutter and individuals who do not stutter. In the second step, 2D:4D correlated negatively with higher scores of the OASES (representing higher negative experiences due to stuttering), and this effect was more pronounced for female persons who stutter.

Conclusions: The findings indicate for the first time that prenatal testosterone may influence individual differences in psychosocial impact of this speech disorder.

1. Introduction

Stuttering is a neurodevelopmental disorder affecting the fluency of speech, with a prevalence of about 1% among adults and a skewed sex ratio [1]. People who stutter are more likely to be male than female. The sex ratios range from about 2:1 in younger children to roughly 4:1 in adolescents and adults [2]. This change in sex ratio over time indicates a much higher rate of spontaneous recovery in girls who stutter [3]. As with many other predominantly male phenotypes and disorders, it has been often speculated that developmental effects of sex hormones like testosterone or estrogen could account for the sex biases in stuttering [1,4,5]. A very prominent and pioneering but nevertheless contentious theory that addresses the possible biological underpinnings of stuttering in this context is the so-called Geschwind-Behan-Galaburda (GBG) model of cerebral lateralization [4]. The authors hypothesized that many predominantly male disorders might result from a high level of testosterone during embryonic development, such that it delays left-hemisphere growth, causes right-hemisphere dominance for speech and language, and results in left-handedness. On this basis, the GBG model hypothesizes relationships among maleness, prenatal testosterone exposure, cerebral lateralization, left-handedness, and disorders such as dyslexia, autism, delayed speech and language development, and also stuttering. Of note, handedness is also discussed to be associated with stuttering [6,7]. The GBG model has been extensively debated and criticized during the last decades, but there are also findings that seem to confirm a relationship among prenatal testosterone exposure and cerebral lateralization in general [8–10].

The relationship between prenatal testosterone and (language) lateralization is of interest in the context of the aetiology and pathogenesis of stuttering, because atypical hemispheric language processing is characteristic for adults who stutter. In the 1920s, Orton and Travis discussed a lack of hemispheric dominance and a resulting conflict in the

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neuromotor control of speech as one of the multiple causes of stuttering [7]. Moreover, newer brain imaging studies commonly reported results that suggest increased right hemisphere participation in speech production of people who stutter [e. g. 11–13]. This right hemispheric overactivity was considered to be compensatory because it is associated with decreased functional and anatomical anomalies in the left hemisphere of people who stutter [13,14, see also review 15].

Taking all these findings together, it is possible that prenatal testosterone could influence the aetiology and the development of stuttering. To the best knowledge of the authors of the present article, there are no studies that have investigated these relationships in the context of stuttering. There are only anecdotal case reports that indicate a potential association between testosterone and stuttering. For example, Kartalci et al. [16] showed an association between testosterone administration for treating hypogonadism and stuttering in an adolescent boy. Because of the lack of empirical evidence in the field, the present study sought to explore whether prenatal testosterone could be linked to the occurrence and/or the symptomatology of stuttering.

As it is difficult to assess early prenatal testosterone levels in humans (e.g., at the end of 1st Trimester or beginning of 2nd Trimester), and because it is unknown if a person will stutter in the future, indirect measures are required. One of the most often-used indirect indicators of prenatal testosterone is the 2D:4D ratio of the hand (D = digit or finger). Longer ring fingers (4D), as compared to the index finger (2D;), characterize the typical male hand (lower 2D:4D ratio). This pattern is associated with higher prenatal testosterone levels. In contrast, longer index fingers, as compared to ring fingers, characterize a more typical female hand. This pattern is associated with lower prenatal testosterone levels (note that the ratio is not related to *actual* testosterone levels in adolescence/adulthood; it only reflects the prenatal environment). The robust statistical difference in 2D:4D between male and female hands has been abundantly researched and confirmed in a large number of samples [e. g. 17]. Hönekopp et al. [18], Breedlove [19] and Manning et al. [20] have summarized the sizable body of evidence linking digit ratio to prenatal testosterone.

Given the above-mentioned evidence on males being more affected in the context of stuttering, as well as the laterality issues indicating that prenatal testosterone might be involved in stuttering, we hypothesized in a first step of this study that people who stutter would be more likely to exhibit male hand characteristics (lower 2D:4D ratio – hence higher prenatal testosterone) than people who do not stutter. In a second step, we were interested in linking 2D:4D to the individual degree of the multifaceted symptomatology within the stuttering group. Here, we hypothesized that a lower 2D:4D (higher prenatal testosterone levels) would be linked with a higher symptomatology, as measured by the Overall Assessment of the Speaker's Experience of Stuttering (OASES) [21]. Besides the two mentioned research hypotheses, we aimed to replicate the classical sex effect of the 2D:4D ratio, with females being associated with higher ratios compared to males.

2. Methods

2.1. Participants

In the first part of the study, 38 persons who stutter (28 males and 10 females) and 36 persons who do not stutter (24 males and 12 females) were invited. This sample size, which is comparable to other studies also investigating the 2D:4D marker in patient groups such as schizophrenia [e. g. 22,23], represents a starting point to investigate the link between prenatal testosterone and stuttering. We aimed to achieve the same number of males and females in the non-stuttering group after the persons who stutter were recruited. The mean age of persons who stutter was 30.63 years (SD = 10.65). The mean age of the non-stuttering group was 31.53 years (SD = 10.54). The persons who stutter were recruited in speech therapy settings in 2012-2013 and at a self-help congress for stuttering in 2012. The persons who do not stutter reported no lifetime history of stuttering and no occurrence of stuttering in their families. They were recruited at the university and in the personal environment of the researchers. No significant between-group differences were found with respect to age ($F_{(1,72)} = .13$, $p = .72$) or sex ($Chi^2 = .44$, $df = 1$, $p = .51$). The present study was approved by the local ethics' committee of the university and all participants gave written consent.

In the second part of the study, the association between 2D:4D and experiences due to stuttering in everyday life were investigated in the group of people who stutter. In addition to the original described sample described above, 6 additional participants who stutter were recruited. Thus, the total sample of persons who stuttered examined in the second part of this study consisted of 44 individuals (34 males and 10 females). The mean-age of the participants was 31.95 years (SD = 11.90).

2.2. Questionnaire

The German translation of the Overall Assessment of the Speaker's Experience of Stuttering (OASES, [21]; translation by [24]) was administered to all persons who stutter. This questionnaire includes 100 items that examine the overall impact of stuttering on people's lives. Each item is assessed as a self-report with a five-point Likert scale. The questionnaire is divided into four subscales: (I) general perspectives about stuttering, (II) affective, behavioral and cognitive reactions to stuttering, (III) functional communication difficulties, and (IV) the impact of stuttering on the speaker's quality of life. The German version has been externally validated with the NEO-FFI, a measure for human personality [25]. High neuroticism and low extraversion was associated with high OASES scores (hence high negative experience of stuttering in everyday life).

2.3. 2D:4D ratio

All participants were first asked if one of their index or ring fingers had been broken. If so, the afflicted hand was excluded from the analysis. There were four instances of a broken left ring finger, one instance of a broken right ring finger, and instance of a broken right hand. Therefore, different sample sizes (degrees of freedom) can be observed in the statistical analysis reported below.

Scans of both the left and right hand were conducted with a Canon Scanner. Every scan was implemented in the software GIMP 2.0 to calculate 2D:4D. A line was drawn from the middle of the lowest crease from the index and ring finger to the tip of each finger. Each hand was measured by two independent raters in pixels. The inter-rater-reliability between the two measures for the left and right 2D/4D fingers was high and ranged from .978 to .998. The correlations between measures was $r = .85$ for the left 2D:4D ratio and $r = .86$ for the right 2D:4D ratio.

2.4. Statistical analyses

An analysis of variance (ANOVA) was used to investigate potential differences in the 2D:4D ratio by subject group (persons who stutter/persons who do not stutter) and sex (male/female). In a second step, partial correlations were computed between the 2D:4D measures of the left/right hand and the OASES scores, controlling for age, because this variable correlated significantly (though not strongly) with the OASES total scale, ($r = -.30$, $p < .05$) and the subscale I ($r = -.33$, $p < .05$). The other correlations between OASES subscales and age barely missed significance. Sex was not associated with significant differences in the OASES. All analyses were conducted for the total sample and for the male/female subsamples independently. Considering a potential link between the four subscales of the OASES and 2D:4D, the alpha to be reached for significance was adjusted to .0125 for these analyses (.05 divided by four tests).