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SUBJECTS WITH DOWN SYNDROME DISPLAY BODY TONICITY AND STABILITY IMPAIRMENTS AFTER LATE ADOLESCENCE

Mirela GAVRIŞ-ŞTEF, Dana CRISTEA, Anca POP[*]
University of Oradea, Geography, Tourism and Sport Faculty,
Physical Education, Sport and Kinetotherapy Department, mirelagav@yahoo.com, popancacristina@yahoo.com,

Beatrice FERRY, Daniel COURTEIX[**]
Clermont Université, Université Blaise Pascal, Clermont-Ferrand,France, e-mail: beatrice.ferry@brive.unilim.fr, daniel.courteix@univ-bpclermont.fr

Abstract: Down syndrome (DS) is the most common genetic cause of developmental disability, characterized by mental retardation and musculoskeletal disorders. As reported in the literature, individuals with Down syndrome display low muscle tone, or low stiffness. The purpose of this study was to study in a large cohort of DS subjects the static stabilometric characteristics as surrogate of body tonicity and stability assess the bone ultrasound properties in a cohort of subjects with DS with reference to age and body tonicity. One hundred ninety-three subjects with DS and 246 healthy subjects participated in this study. Stabilometric performances were obtained from a force platform. Subjects were divided into four age groups (mean age 10.8 yrs, 15.2 yrs, 20.5 yrs, 33.4 yrs respectively) using a decision tree procedure. Stabilometric performances of the controls were higher at each age group, showing an improvement of stability and tonicity with age. The stabilometric performances displayed by the controls were higher for age group 3 and 4, showing an improvement of stability and tonicity with age. These improvements correlated negatively with age. The present results demonstrated that body stability and balance continue to evolve until adulthood in ordinary people whereas they reach a plateau at adolescence in DS subjects.

Key words: Down syndrome, Muscle hypotonia, Tonicity, Stability.

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BACKGROUND

Down syndrome (DS) is a genetic disabilities caused by the presence of all or part of an extra 21st chromosome (Lejeune, Turpin et al. 1959) which may be responsible for skeletal abnormalities, short stature and other mechanisms of premature aging (Roth, Sun et al. 1996) and generates physiological and physical developmental disorders. Down syndrome (DS) is the most common genetic cause of developmental disability, characterized by mental retardation and

[*] Mirela Gavriş Ştef

musculoskeletal disorders (Sago, Carlson et al. 2000) Expression of the DS phenotype includes cardiac malformations and hypotonia. Clinically, children with DS are described as having low tone, or low stiffness ("American Academy of Pediatrics: Health supervision for children with Down syndrome," 2001), which may affect muscle strength and motor skills (Apache 2005). Individuals with Down Syndrome have reduced resting metabolic rates, which contribute to a higher frequency of obesity than in other individuals (Rubin, Rimmer et al. 1998).

The significant increase in the life expectancy of this population may explain the onset of osteoporosis. Moreover, thyroid dysfunction, abnormalities of sexual development and musculoskeletal troubles (peripheral muscle strength) as well as poor calcium and vitamin D intakes, may contribute to the development of osteoporosis. Moreover, both pediatric and adult cohorts, display a lower level of physical fitness (Fernhall, Pitetti et al. 1996) than ordinary persons.

Usually the measures of the variance of speed by means of stabilometric platforms are able to characterize the tone of the muscular posterior chain. This measure could allow the assessment of body tonicity, because through this device, the lower the variance, the higher the tonicity.

Therefore, the purpose of this study was to study in a large cohort of DS subjects the static stabilometric characteristics as surrogate of body tonicity and stability.

MATERIAL AND METHODS

All active individuals with Down syndrome were eligible for recruitment into this study which was conducted with the same ambulatory devices in France and Romania. A group of 193 subjects with Down syndrome (104 males and 89 females) aged between 8 and 37 years participated in this study. Concomitantly, we measured a total of 246 healthy subjects (107 males and 139 females) aged between 10 and 61 years. Measurements were performed at special events with support of Special Olympics organization and French Federation of Adapted Sport. Written informed consent was obtained from all parents or subjects.

Anthropometric measurements. Body height was measured using a portable stadiometer calibrated to the nearest 0.1 cm. Body mass was measured with subjects wearing light clothing and without shoes, to the nearest 0.1 kg, using a digital scale calibrated daily. The body mass index (BMI) was calculated from the previous parameters.

Stabilometric measures. The measures were obtained in standard conditions, with subjects required to keep their eyes open and teeth clenched; conditions in which all the external afferences are active, the parameters measured reflecting the postural control of the subject under examination (Cultrera, Pratelli et al. 2010). Because we had no interest in the sensory input, we did not perform tests with “eyes closed”. Body sway area was recorded by means of a 3-strain gauge platform with automatic weight correction (Stabilotest, PostureWin – Platform V328). The forces acting on the platform were sampled at 40 Hz. Participants stood barefoot facing forwards at a target placed at 90 cm distance, respecting standard and validated conditions (Kapteyn, Bles et al. 1983; Hsu, Kuan et al. 2009). Feet were placed at an angle of 30°and the distance between the heels was 2 cm. Although the reference values for stabilometry testing are established at 51.2 s we ascertained a second measurement at 12.8 s because we postulated that a shorter period was more suitable for DS population (mental retardation). The analysis technique is based on the measurement of the centre of pressure (CoP) sway in a standing position. The following parameters were used to assess the balance: the postural sway area (in mm2, corresponding to the area of the 90% confidence intervals for the ellipse surface area which contains 90% of the CoP positions sampled - *Area*), the CoP‟ s total path length over the time (in mm - *Length*), the variance of speed (*VFY*), the average sway velocity (a*V*) and the CoP‟ s total path length per unit surface (*LFS*).

Statistics. All values were expressed as mean and standard deviation (SD). The Gaussian distribution of the variables was assessed using the Shapiro-Wilk test. In case of non Gaussian distribution, non parametric tests were used and for a complex statistical analysis, the data were logtransformed. Analyses were conducted for the whole group and separately for gender. Because the age range was not continuous, subjects were divided into age groups using a decision tree procedure. The Decision Tree procedure creates a tree-based classification model.

The areas (AUC) under the receiver operating characteristic curves (ROC) were employed for Area, Length, VFY, aV and LFS (all measured at 51s) to evaluate their power to discriminate DS from healthy patients.

All analyses were conducted using SPSS software (PASW version 18). Significance was set at $p<0.05$. In order to preclude the outliers (out of range data), we excluded the data with values beyond plus or minus 2 standard deviations.

RESULTS

Subjects characteristics. As displayed in table 1, CTL total group and DS total group were similar in age and weight. DS subjects were smaller and had higher BMI than CTL group ($P<.001$). The classification obtained from the decision tree test is displayed figure 1. Four age groups were characterized by the relation age-BUA. The mean ages were for group 1: 10.8 ± 1.1 yrs old, group 2: 15.2 ± 1 yrs old, group 3: 20.5 ± 2.3 yrs old and group 4: 33.4 ± 7 yrs old. Characteristics of subjects per class of age are shown table 1. In this experimental design, no significant difference was observed in the distribution of gender within age groups (Pearson Chi-Square = 3.55, p >0.05).

Table 1. Characteristics of the subjects

	CTL (mean±SD)	DS (mean±SD)	Between groupes		CTL (mean±SD)	DS (mean±SD)	Between groups
Age (yr.)	21.2±8	20.8±6	NS	Group 1	10.8±0.7	10.8±1.4	NS
				Group 2	15.4±0.7	14.9±1.2	NS
				Group 3	20.0±1.7	21.1±2.7	p<.01
				Group 4	36.6±8.5	30.1±3.2	p<.001
Height (cm)	167.5±11	151.0±13	p<.001	Group 1	145.6±7.0	144.4±14.5	NS
				Group 2	169.1±8.3	150.0±12.9	P<.001
				Group 3	169.9±9.2	152.3±14.6	P<.001
				Group 4	168.4±9.8	153.2±11.1	P<.001
Weight (kg)	61.7±14	58.6±15	NS	Group 1	40.7±8.0	51.5±17.5	P<.05
				Group 2	61.5±12.7	55.0±18.5	NS
				Group 3	63.2±12.9	61.7±12.9	NS
				Group 4	67.4±15.9	60.4±12.7	NS
BMI kg/m^2	21.9±3	26.1±12	p<.001	Group 1	19.1±3.3	23.8±4.7	P<.005
				Group 2	21.3±3.3	23.8±6.0	NS
				Group 3	21.8±3.6	28.1±17.7	P<.001
				Group 4	23.9±4.2	26.0±5.5	P<.05

Stabilometric parameters

Because all statistical results was similar for the two duration tests (12.8 s and 51.2 s), we chose to present only the data corresponding to 51.2 s recording test. Independent of the parameter studied, DS values were consistently less efficient than CTL. Females presented better stabilometric values than males for CTL whereas there was not a gender difference for DS. The female CTL had higher stabilometric performance than female DS for all parameters. The same observation was apparent in males, with the exception of LFS and VFY, which not differ. Results for age categories are displayed figure 1. No differences existed at age group 1 and 2, whereas at age group 3 and 4, CTL groups showed better a performance than DS. DS group displayed no difference in stabilometric values irrespective of age group. No difference was observed between age group for DS with the exception of LFS, which demonstrated a agerelated decrease. Inter age performances increased for each parameter in CTL (figure 1). Negative correlation existed between age and all stabilometric parameters for CTL group: area (Spearman‟ s rho = -.396, p<0.001), length (Spearman‟ s rho = -.350, p<0.001), LFS (Spearman‟ s rho = -.276, p<0.001), aV (Spearman‟ s rho = -.351, p<0.001), and VFY (Spearman‟ s rho = -.445, p<0.001), whereas age did not correlate with values from the DS groups.

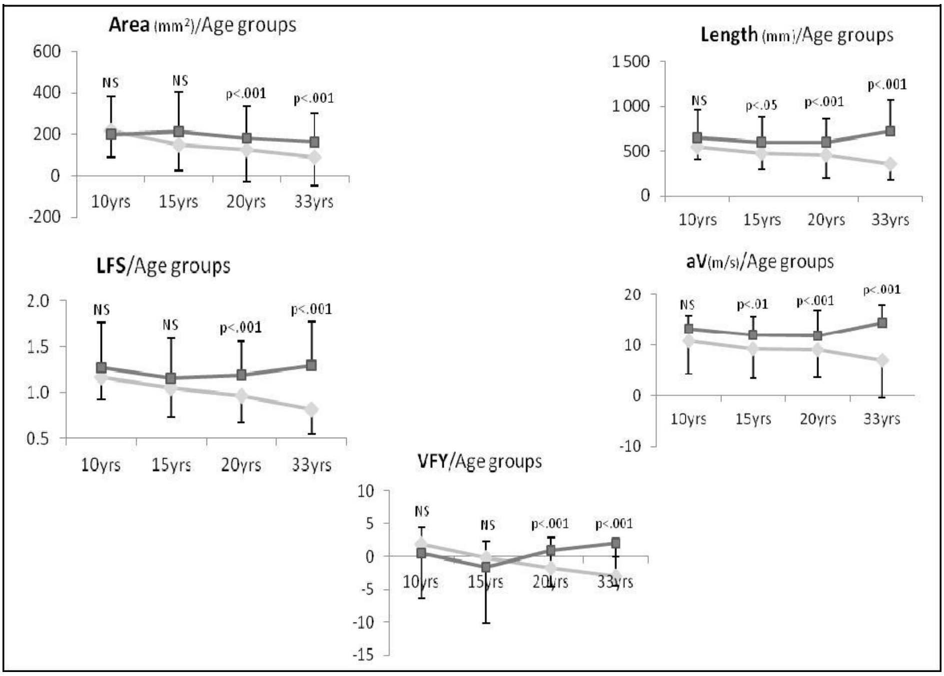

Figure 1. Comparison between DS (dark) and CTL (light) of stabilometric parameters in function of age group.

ROC results. The discriminatory ability of the stabilometric variables and BUA measurements is shown in figure 2. These parameters were able to discriminate between individuals with DS and healthy controls only for age groups 3 and 4 Among other, aV was one of the better discriminators between groups.

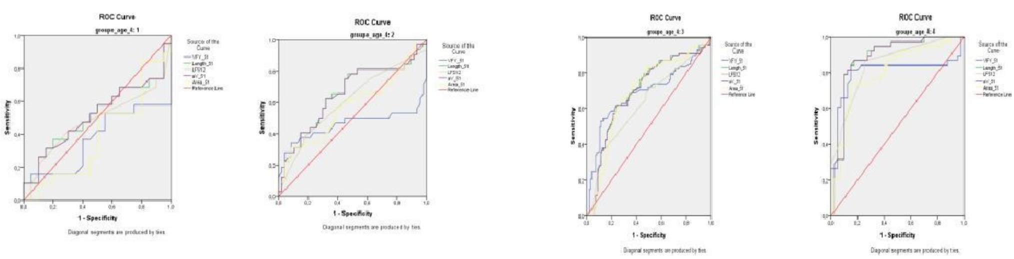

Figure 2. ROC curves for the six stabilometric variables for Down syndrome subjects and age-matched healthy subjects of age groups

DISCUSSION

The present results point out that these values are linked to significantly poorer stabilometric values. It appears that the weakness observed in individuals with Down syndrome may result from the lower muscle tonicity and stability as assessed by the stabilometric tests. The fact that the speed of sound failed to differ between groups and was higher in the younger age group is difficult to explain.

The differences observed between our groups could be linked to one or more specificities of this pathology. Another explanation can be argued. Even if little work has been done to quantify this characteristic in this group of patients, hypotonia is routinely reported in describing the characteristics of Down syndrome.

The techniques commonly used to assess the muscle tone (palpation, passively moving the limb, etc.) are highly subjective and give limited information about the state of the neuromuscular system (Webber, Virji-Babul et al. 2004). We chose to use stabilometric parameters as indexes of muscular tonicity because this device provides objective measures of body stability and tonicity. What is remarkable is that, for all parameters the values expressed by the Down syndrome group were largely unaffected by age. Conversely, the stabilometric performances displayed by the controls were higher at each age group, showing an improvement of stability and tonicity with age. These improvements correlated negatively with age, suggesting that lower the values, the higher the performances. It is noteworthy that a previous study (Hsu, Kuan et al. 2009) suggested that a child at the age of 12 years is supposed to reach balance level of an adult. The present results demonstrated that body stability and balance continue to evolve until adulthood in ordinary people. For instance, VFY, a parameter usually associated with a hyper- or hypo- muscle tone of the posterior chain (Vallier 1985) suggesting an association between the body tonicity and the bone status. Among the stabilometric parameters studied, some appeared to to be more sentence than others in discriminating between the differences between Down syndrome and non-Down syndrome subjects. In the study of Webber et al. (Webber, Virji-Babul et al. 2004) conducted in adults (30,8 ±6.2 yrs old), only the postural sway velocity was able to differentiate between the Down syndrome and non-Down syndrome groups. Our results are consistent with this statement. ROC analysis results showed that all stabilometric parameters were able to discriminate the two groups. Among other, aV was one of the better discriminators between groups.

Finally, if stabilometry is a method widely used by clinicians for measuring the balance and postural tonicity, it remains problematic as an instrument for measuring the muscle tone. Nevertheless, in the present study tends to support the use of stabilometric performance as a surrogate of total body tonicity.

CONCLUSION

The stabilometric performances displayed by the controls were higher for age group 3 and 4, showing an improvement of stability and tonicity with age. These improvements correlated negatively with age.

Concerning the DS subjects it is noteworthy that there was no improvement with age.

The present results demonstrated that body stability and balance continue to evolve until adulthood in ordinary people whereas they reach a plateau at adolescence in DS subjects.

Aknowlegments

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