**Purpose** - Children with syndromic craniosynostosis are at risk for neuropsychological impairment. The cause of this developemental delay is unclear. With this prospective diffusion tensor imaging (DTI) study we aim to assess white matter disorders in patients with syndromic craniosynostosis.

**Materials and Methods** - The Medical Ethics Committee approved this study. A prospective cohort study was performed in 45 children with syndromic craniosynostosis aged six to fourteen years. Data of 7 agematched controls were available for comparison. For DTI, an echo planar sequence with diffusion gradient (b=1,000s/mm²) applied in 25 non-collinear directions was used. Regions of interest (ROIs) were placed in different white matter structures. Eigenvalues were measured in all ROIs, from which Fractional Anisotropy (FA) were calculated.

**Results -** FA values of the corpus callosum (splenium), the medial cerebellar peduncle, the fornix, the uncinate fasciculus and the mean white matter (MWM), which is given by the mean of the right and left frontal and occipital white matter measurements, were significantly lower in our patients as compared to controls (respectively p < 0.05).

**Conclusion -** DTI measurements of white matter tracts show significant white matter differences between children with craniosynostosis and healthy controls. This could imply the presence of a primary disorder of the white matter micro-architecture causing the developmental delays seen in these patients.