Introduction. The etiology of idiopathic scoliosis and Adolescent Idiopathic scoliosis (AIS) remain unclear as well as the biological mechanism behind this phenotype. Since the genetic implication is accepted, many association analysis (GWAS) and some candidate gene analysis (linkage analysis, WES) seems to reveal the implication of ciliary genes. The aim of this study is to clarify the implication of ciliary pathway on the onset of the spinal curvature that occurs in AIS patients through functional studies of two genes: POC5 and TTLL11, a gene coding for a tubulin polyglutamylase. Methods. The characterization of these two proteins was assessed by qPCR, Western blot and immunofluorescence in vitro using control cells and cells (osteoblast and fibroblast) derived from AIS patients. We then study the impact of genetic modification of these genes on the functionality of the proteins in vitro and in vivo in zebrafish model created by CRISPR/Cas9 that we phenotyped by using microCT and histologic analysis. Results and Discussion. Our study revealed that mutant cells, for both gene, were less ciliated and the primary cilia was significantly shorter compare to control cells. We also observed a default in cilia glutamylation by immunofluorescence and Western Blot. Moreover, we observed in both zebrafish model, a 3D spine curvature similar to the spinal deformation in AIS. Interestingly, our preliminary results of immunohistology showed a retinal defect, especially at the cone cell layer level. Conclusion and Significance. This study strongly supports the implication of the ciliary pathway in the onset of AIS and this is the first time that a mechanism is described for AIS. Indeed, we show that shorter cilia could be less sensitive to environmental factors due to lower glutamylation and result in altered signaling pathway. Identifying the biological mechanism involved is crucial for elucidating AIS pathogenesis. Support : Yves Cotrel Foundation, FRQS