REGIONAL CENTRE FOR BIOTECHNOLOGY

Journal Club

"Behavioral and Other Phenotypes in a Cytoplasmic Dynein Light Intermediate Chain 1 Mutant Mouse"

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Abstract

The cytoplasmic dynein complex is fundamentally important to all eukaryotic cells for transporting a variety of essential cargoes along microtubules within the cell. This complex also plays more specialized roles in neurons. The complex consists of 11 types of protein that interact with each other and with external adaptors, regulators and cargoes. Despite the importance of the cytoplasmic dynein complex, we know comparatively little of the roles of each component protein, and in mammals few mutants exist that allow us to explore the effects of defects in dynein-controlled processes in the context of the whole organism. Here we have taken a genotype-driven approach in mouse (*Mus musculus*) to analyze the role of one subunit, the dynein light intermediate chain 1 (*Dync1li1*). We find that, surprisingly, an N235Y point mutation in this protein results in altered neuronal development, as shown from *in vivo* studies in the developing cortex, and analyses of electrophysiological function. Moreover, mutant mice display increased anxiety, thus linking dynein functions to a behavioral phenotype in mammals for the first time. These results demonstrate the important role that dynein-controlled processes play in the correct development and function of the mammalian nervous system.