

ORAL PRESENTATION

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# Dishevelled stabilisation at the cilium by RPGRIP1L is essential for planar cell polarity

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Cilia are involved in planar polarity in different systems but the mechanisms by which they influence the polarization process are unclear [1]. In order to clarify this issue, we investigated the function of the ciliary gene *Rpgrip1l* (*Ftm/NPHP8/MKSS*) in the mammalian cochlear sensory epithelium and in the zebrafish floor plate. We and others have previously shown that mutations in the human *RPGRIP1L* gene cause Meckel and Joubert type B syndromes [2]. The *Rpgrip1l* protein is localised at the ciliary transition zone and is required for transduction of the Hh/Gli pathway [3]. Our recent work has shown that *Rpgrip1l* patterns the telencephalon via the regulation of Gli3 proteolytic cleavage [4]. Here we show that in both the mammalian cochlear sensory epithelium and the zebrafish floor plate, *Rpgrip1l* is required for correct positioning of the basal body along the planar polarity axis. Our results strongly suggest that *Rpgrip1l* is essential for stabilizing the adaptor protein dishevelled at the basal body and/or cilium. Finally, we demonstrate that, in the zebrafish floor plate, the function of *Rpgrip1l* in basal body positioning is mediated by dishevelled. We propose that *Rpgrip1l* participates in a protein complex required for stabilizing dishevelled at the cilium, and that this stabilization is essential for asymmetric localization of the basal body along the planar polarity axis.

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