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Sylvie Schneider-Maunoury, Alexia Mahuzier, Héliori-Mael Gaudé, Isabelle Anselme, Flora Silbermann, et al.. Dishevelled stabilisation at the cilium by RPGRIP1L is essential for planar cell polarity. First International Cilia in Development and Disease Scientific Conference, pp.O21. inserm-00752956

HAL Id: inserm-00752956

<https://www.hal.inserm.fr/inserm-00752956>

Submitted on 16 Nov 2012

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ORAL PRESENTATION

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

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From First International Cilia in Development and Disease Scientific Conference (2012)
London, UK. 16-18 May 2012

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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Published: 16 November 2012

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doi:10.1186/2046-2530-1-S1-O21

Cite this article as: Schneider-Maunoury *et al.*: Dishevelled stabilisation at the cilium by RPGRIP1L is essential for planar cell polarity. *Cilia* 2012 **1**(Suppl 1):O21.

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