



CYLD tumour supressor regulates ciligenesis

Thibaut Eguether, Marion Bonnet, Maria Ermolaeva, Manolis Pasparakis,
Gilles Courtois, Anne-Marie Tassin

► To cite this version:

Thibaut Eguether, Marion Bonnet, Maria Ermolaeva, Manolis Pasparakis, Gilles Courtois, et al..
CYLD tumour supressor regulates ciligenesis. First International Cilia in Development and Disease
Scientific Conference, May 2012, Londres, United Kingdom. pp.P66. inserm-00752966

HAL Id: inserm-00752966

<https://inserm.hal.science/inserm-00752966>

Submitted on 16 Nov 2012

HAL is a multi-disciplinary open access archive for the deposit and dissemination of scientific research documents, whether they are published or not. The documents may come from teaching and research institutions in France or abroad, or from public or private research centers.

L'archive ouverte pluridisciplinaire **HAL**, est destinée au dépôt et à la diffusion de documents scientifiques de niveau recherche, publiés ou non, émanant des établissements d'enseignement et de recherche français ou étrangers, des laboratoires publics ou privés.

POSTER PRESENTATION

Open Access

CYLD tumour suppressor regulates cilogenesis

T Eguether^{1*}, BM Bonnet², EM Ermolaeva², PM Pasparakis², CG Courtois³, TAM Tassin¹

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

The tumor suppressor CYLD gene encodes a deubiquitinating enzyme that removes lys-63-linked ubiquitin chains. CYLD regulates, by its catalytic domain, NF- κ B, c-Jun kinase and Wnt/ β -catenin signalling pathways. When mutated in its catalytic domain CYLD cause skin appendages tumors as in familial cylindromatosis. We have found CYLD as a partner of the centrosomal protein CAP350 in immunoprecipitation experiments followed by mass spectrometry. CAP350 was shown to play a role in microtubule stabilization (Hoppeler-Leber et al, 2007). Here, we show that CYLD localizes to the centrosome in cells and is enriched in purified centrosomes. To understand the functional interaction between CYLD and CAP350, we studied mice carrying deletion of part of the deubiquitinase domain and mimicking the human pathology. These homozygous mice *cyld* (*del17/del17*) die perinatally due to respiratory dysfunction and exhibit immature lung phenotype. To test if CYLD is involved in ciliogenesis, we studied the presence of motile cilia in the trachea. *Cyld* (*del17/del17*) embryo tracheas exhibit a decreased number of ciliated cells compared to wild type ones of the same litter. In addition, these ciliated cells have fewer and shorter cilia. Similar results were obtained on ependymal cell culture. Transmission electron micrographs demonstrate that most of the basal bodies fail to anchor to the plasma membrane. We demonstrate that in MEFs derived from *cyld* (*del17/del17*) embryo, CYLD and CAP350 do not longer interact in contrast to MEFs derived from wild type embryo. In addition, these mutant MEFs exhibit primary cilia growth defect compared to MEFs derived from wild type mice.

Author details

¹Institut Curie INSERM U759, France. ²Cologne University, Germany.
³INSERM, France.

Published: 16 November 2012

* Correspondence: thibaut.eguether@curie.fr

¹Institut Curie INSERM U759, France

Full list of author information is available at the end of the article

doi:10.1186/2046-2530-1-S1-P66

Cite this article as: Eguether et al.: CYLD tumour suppressor regulates cilogenesis. *Cilia* 2012 **1**(Suppl 1):P66.

Submit your next manuscript to BioMed Central and take full advantage of:

- Convenient online submission
- Thorough peer review
- No space constraints or color figure charges
- Immediate publication on acceptance
- Inclusion in PubMed, CAS, Scopus and Google Scholar
- Research which is freely available for redistribution

Submit your manuscript at
www.biomedcentral.com/submit

