



**Cover:** Aggregates of dVAPB(P58S), in red, are visualized in the *Drosophila* third instar larval brain using an antibody generated against the coiled-coil domain of the protein. The mutant protein is expressed in neurons using the UAS-Gal4 system. The aggregates/puncta mimic inclusions seen in human ALS patients with the hVAP(P56S) mutation. The authors find that ROS levels modulate aggregates by triggering their clearance through the ubiquitin proteasomal system. ROS, in the context of their experiments, is in turn regulated by SOD1 activity and also by the strength of mTOR signaling. This study uncovers gene networks in neurons that regulate aggregate dynamics. See article by Chaplot et al. (dmm033803). Cover image is licensed under a Creative Commons Attribution 4.0 International license.

## EDITORIALS

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From gene to treatment: supporting rare disease translational research through model systems  
**Hmeljak, J. and Justice, M. J. (Editor-in-Chief)**  
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## SPECIAL ARTICLE

The Human Cell Atlas: making 'cell space' for disease  
**Ponting, C. P.**  
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## REVIEW

Wnt signaling in orofacial clefts: crosstalk, pathogenesis and models  
**Reynolds, K., Kumari, P., Sepulveda Rincon, L., Gu, R., Ji, Y., Kumar, S. and Zhou, C. J.**  
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## RESEARCH ARTICLES

Differential regulation of the unfolded protein response in outbred deer mice and susceptibility to metabolic disease  
**Havighorst, A., Zhang, Y., Farmaki, E., Kaza, V., Chatzistamou, I. and Kiaris, H.**  
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Leptin induces muscle wasting in a zebrafish *kras*-driven hepatocellular carcinoma (HCC) model  
**Yang, Q., Yan, C., Wang, X. and Gong, Z.**  
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VPS13A is closely associated with mitochondria and is required for efficient lysosomal degradation  
**Muñoz-Braceras, S., Tornero-Écija, A. R., Vincent, O. and Escalante, R.**  
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Loss of *Frrs1l* disrupts synaptic AMPA receptor function, and results in neurodevelopmental, motor, cognitive and electrographical abnormalities  
**Stewart, M., Lau, P., Banks, G., Bains, R. S., Castroflorio, E., Oliver, P. L., Dixon, C. L., Kruer, M. C., Kullmann, D. M., Acevedo-Arozena, A., Wells, S. E., Corrochano, S. and Nolan, P. M.**  
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Serotonin inhibits axonal regeneration of identifiable descending neurons after a complete spinal cord injury in lampreys  
**Sobrido-Cameán, D., Robledo, D., Sánchez, L., Rodicio, M. C. and Barreiro-Iglesias, A.**  
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ApoE-associated modulation of neuroprotection from A $\beta$ -mediated neurodegeneration in transgenic *Caenorhabditis elegans*  
**Griffin, E. F., Scopel, S. E., Stephen, C. A., Holzhauer, A. C., Vaji, M. A., Tuckey, R. A., Berkowitz, L. A., Caldwell, K. A. and Caldwell, G. A.**  
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Early detection of pre-malignant lesions in a KRAS<sup>G12D</sup>-driven mouse lung cancer model by monitoring circulating free DNA  
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A double-hit pre-eclampsia model results in sex-specific growth restriction patterns  
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SOD1 activity threshold and TOR signalling modulate VAP (P58S) aggregation via reactive oxygen species-induced proteasomal degradation in a *Drosophila* model of amyotrophic lateral sclerosis  
**Chaplot, K., Pimpale, L., Ramalingam, B., Deivasigamani, S., Kamat, S. S. and Ratnaparkhi, G. S.**  
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Liver-specific insulin receptor isoform A expression enhances hepatic glucose uptake and ameliorates liver steatosis in a mouse model of diet-induced obesity

**Lopez-Pastor, A. R., Gomez-Hernandez, A., Diaz-Castroverde, S., Gonzalez-Asequinolaza, G., Gonzalez-Rodriguez, A., Garcia, G., Fernandez, S., Escribano, O. and Benito, M.**

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A yeast-based screening assay identifies repurposed drugs that suppress mitochondrial fusion and mtDNA maintenance defects

**Delerue, T., Tribouillard-Tanvier, D., Daloyau, M., Khosrobakhsh, F., Emorine, L. J., Friocourt, G., Belenguer, P., Blondel, M. and Arnauné-Pelloquin, L.**

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