Myotonic Dystrophy FOUNDATION

Fruit Fly – Myotonic Dystrophy Animal Models & Tools

Animal models play a key role in basic, translational and clinical research. The following tables highlight and summarize available animal models and tools for myotonic dystrophy research. Literature links connect to the original publication.

This table summarizes available **drosophila melanogaster ("drosophila", "fruit fly", "fly")** animal models used in myotonic dystrophy (DM) research. Drosophila melanogaster is used in research due to its rapid life cycle, relatively simple genetics with only four pairs of chromosomes, and large number of offspring per generation. The table also contains interesting review articles about this model system. This table was last updated and reviewed in June 2024.

To find additional animal models or learn more about each respective system, please examine and follow the associated literature links and references within each table.

To find additional information and resources focused on myotonic dystrophy, visit the Myotonic Dystrophy Foundation website at: www.myotonic.org.

Fruit Fly Models - Myotonic Dystrophy Animal Models & Tools

Fruit Fly Model	Article Type
Marzullo M, Coni S, De Simone A, Canettieri G, Ciapponi L. Modeling Myotonic Dystrophy Type 2 Using Drosophila melanogaster. Int J Mol Sci. 2023 Sep 16;24(18):14182. doi: 10.3390/ijms241814182. PMID: 37762484; PMCID: PMC10532015.	Review
Souidi A, Jagla K. Drosophila Heart as a Model for Cardiac Development and Diseases. Cells. 2021 Nov 8;10(11):3078. doi: 10.3390/cells10113078. PMID: 34831301; PMCID: PMC8623483.	Review, cardiac phenotypes
Chakraborty M, Llamusi B, Artero R. Modeling of Myotonic Dystrophy Cardiac Phenotypes in Drosophila. Front Neurol. 2018 Jul 16;9:473. doi: 10.3389/fneur.2018.00473. PMID: 30061855; PMCID: PMC6054993.	Review; focus on cardiac phenotypes
Koon AC, Chan HY. Drosophila melanogaster As a Model Organism to Study RNA Toxicity of Repeat Expansion-Associated Neurodegenerative and Neuromuscular Diseases. Front Cell Neurosci. 2017 Mar 21;11:70. doi: 10.3389/fncel.2017.00070. PMID: 28377694; PMCID: PMC5359753.	Review drosophila as model for RNA toxicity
Plantié E, Migocka-Patrzałek M, Daczewska M, Jagla K. Model organisms in the fight against muscular dystrophy: lessons from drosophila and Zebrafish. Molecules. 2015 Apr 9;20(4):6237-53. doi: 10.3390/molecules20046237. PMID: 25859781; PMCID: PMC6272363.	Review drosophila, zebrafish
Reiter L.T., Potocki L., Chien S., Gribskov M., Bier E. A systematic analysis of human disease-associated gene sequences in Drosophila melanogaster. Genome Res. 2001;11:1114–1125. doi: 10.1101/gr.169101.	General interest
Bier E. Drosophila, the golden bug, emerges as a tool for human genetics. Nat. Rev. Genet. 2005;6:9–23. doi: 10.1038/nrg1503.	General article about drosophila as tool

Fruit Fly Model	Article Type
Picchio L, Plantie E, Renaud Y, Poovthumkadavil P, Jagla K. Novel Drosophila model of myotonic dystrophy type 1: phenotypic characterization and genome-wide view of altered gene expression. Hum Mol Genet. 2013 Jul 15;22(14):2795-810. doi: 10.1093/hmg/ddt127. Epub 2013 Mar 21. Erratum in: Hum Mol Genet. 2023 Feb 19;32(5):883. PMID: 23525904.	
Rapisarda A, Bargiela A, Llamusi B, Pont I, Estrada-Tejedor R, Garcia-España E, Artero R, Perez-Alonso M. Defined D-hexapeptides bind CUG repeats and rescue phenotypes of myotonic dystrophy myotubes in a Drosophila model of the disease. Sci Rep. 2021 Sep 30;11(1):19417. doi: 10.1038/s41598-021-98866-0. PMID: 34593893; PMCID: PMC8484449.	
A. Bargiela et al., Increased autophagy and apoptosis contribute to muscle atrophy in a myotonic dystrophy type 1 Drosophila model. Dis. Model. Mech. 8, 679–690 (2015).	
Houseley J.M., Wang Z., Brock G.J.R., Soloway J., Artero R., Perez-Alonso M., O'Dell K.M.C., Monckton D.G. Myotonic dystrophy associated expanded CUG repeat muscleblind positive ribonuclear foci are not toxic to Drosophila. Hum. Mol. Genet. 2005;14:873–883. doi: 10.1093/hmg/ddi080.	
de Haro M., Al-Ramahi I., De Gouyon B., Ukani L., Rosa A., Faustino N.A., Ashizawa T., Cooper T.A., Botas J. MBNL1 and CUGBP1 modify expanded CUG-induced toxicity in a Drosophila model of myotonic dystrophy type 1. Hum. Mol. Genet. 2006;15:2138–2145. doi: 10.1093/hmg/ddl137.	
Garcia-Lopez A., Monferrer L., Garcia-Alcover I., Vicente-Crespo M., Alvarez-Abril M.C., Artero R.D. Genetic and Chemical Modifiers of a CUG Toxicity Model in Drosophila. PLoS ONE. 2008;3:e1595. doi: 10.1371/journal.pone.0001595.	
Yu Z., Teng X., Bonini N.M. Triplet Repeat–Derived siRNAs Enhance RNA–Mediated Toxicity in a Drosophila Model for Myotonic Dystrophy. PLoS Genet. 2011;7:e1001340. doi: 10.1371/journal.pgen.1001340.	
B. Llamusi et al., Muscleblind, BSF and TBPH are mislocalized in the muscle sarcomere of a Drosophila myotonic dystrophy model. Dis. Model. Mech. 6, 184–196 (2013).	