

Fig. S1 Ophn1 localized to highly motile structures during neuronal development.

a. to d. Cortical neurons from $ophn1^{+/y}$ were stained with antibodies against ophn1 at 1 (a and b.), 7 (c.) and 21 (d.) days of in vitro culture (div) together with antibodies against tubulin (Tuj-1) (a. and b.) or MAP2 (c. and d.). At 1 div, ophn1 is localized in neuritic (a.) and axonal growth cones (b.) at div1, which are the sites of most active morphological changes at this stage of development. Later, at 7 div, ophn1 is concentrated at the tip of growing dendrites (c.; note that the luminosity was artificially increased in the frame). At 21 div, ophn1 labeling showed a predominant localization in the dendritic spines (d. and inset).

Fig. S2. Immuno-localization of ophn1 in mouse sagittal brain sections

a. The hippocampus shows relatively strong ophn1 expression in pyramidal cells in CA1 and the dentate gyrus. Note that ophn1 is also present in the ependymal cells lining the ventricle. A higher magnification in the stratum radiatum of the CA1 area (inset) shows ophn1 staining in the cell bodies and dendritic shafts of pyramidal neurons.

b. In the cerebellum, the cell bodies of Purkinje cells and their dendrites in the molecular layer are brightly stained for ophn1. Higher magnification of the molecular layer shows the staining for ophn1 in the dendritic shaft of a Purkinje cell.

c. and d. Specificity of the ophn1 antibody. Ophn1 immuno-staining of $ophn1^{-/y}$ brain slices showing the absence of signal in the hippocampus c. and in the cerebellum d.

Supplementary data Table 1:

Summary of different analyses related to activity and anxiety in $ophn1^{+/y}$ (WT) and $ophn1^{-/y}$ mice (KO) as tested in the open-field, O-maze, light-dark box and Y-maze. In each case are detailed the mean of the variable, the standard error of the mean in each genotype as well as the results from the statistical analyses. Please note that $ophn1^{-/y}$ mice performed more distance in all tests. To overcome the bias linked to this hyperactivity, we calculated the percentage of distance or time spent in the light area to total distance. Only the open-field test showed significant results with $ophn1^{-/y}$ mice being less anxious than $ophn1^{+/y}$ mice.