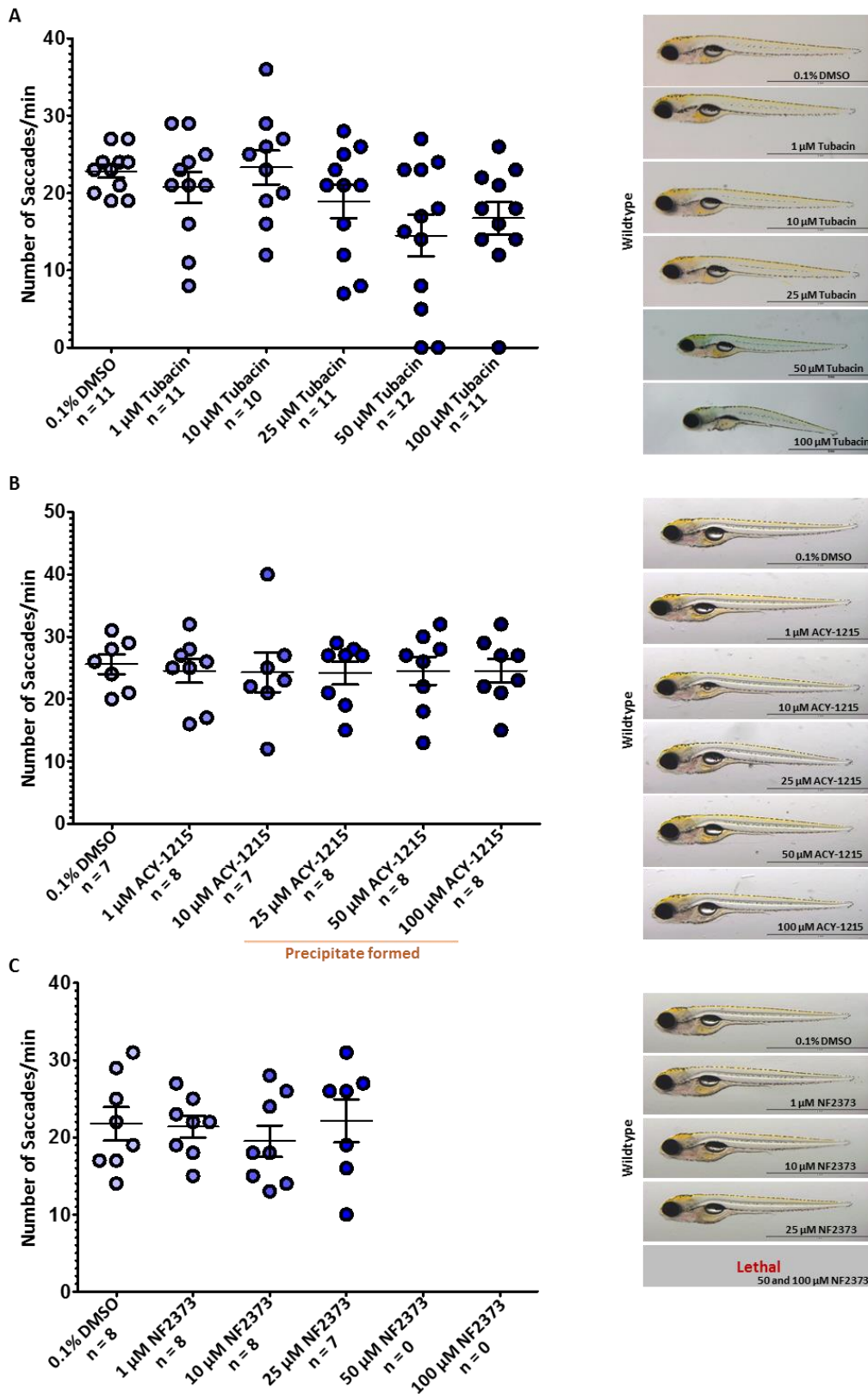
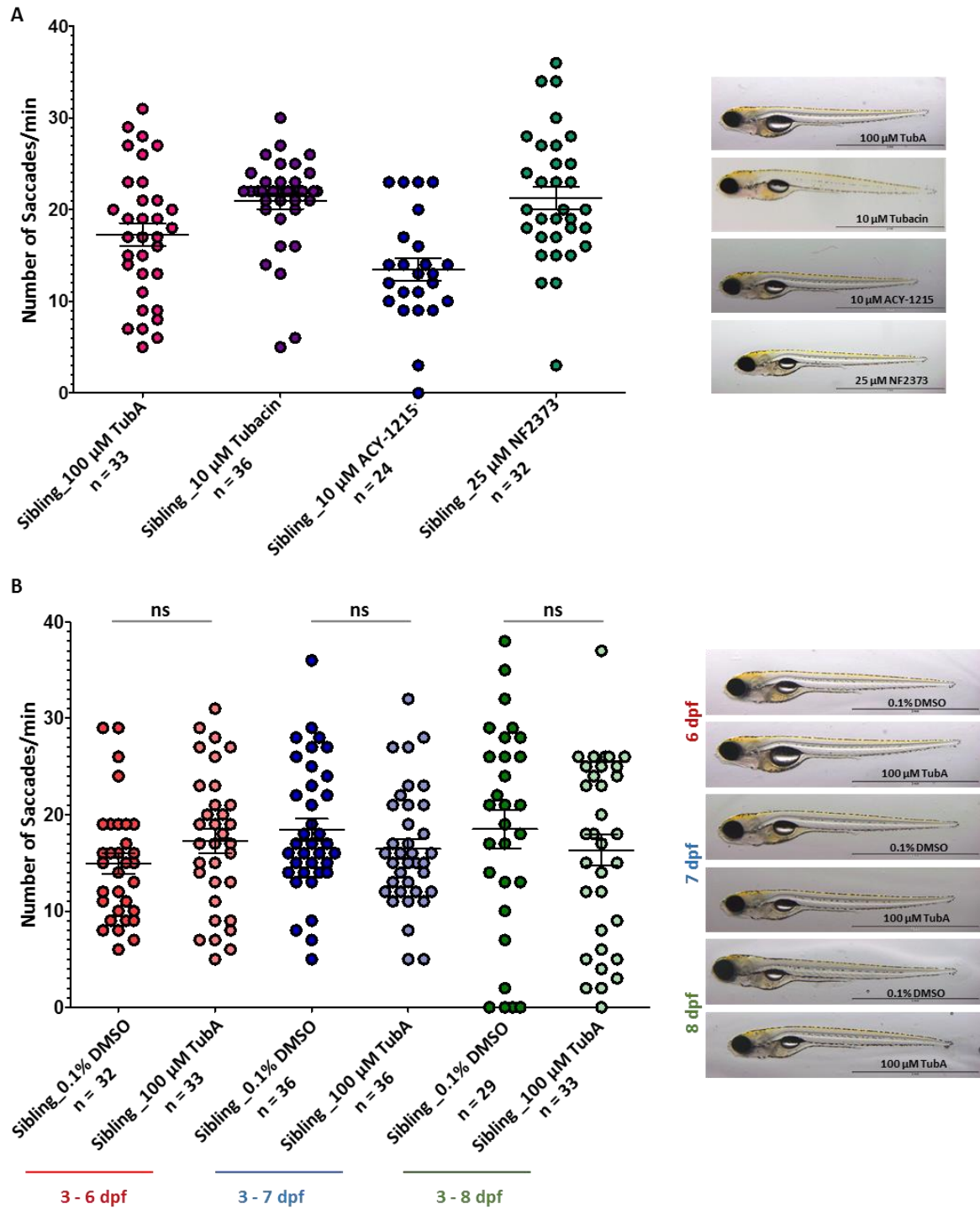


Supplementary Figures

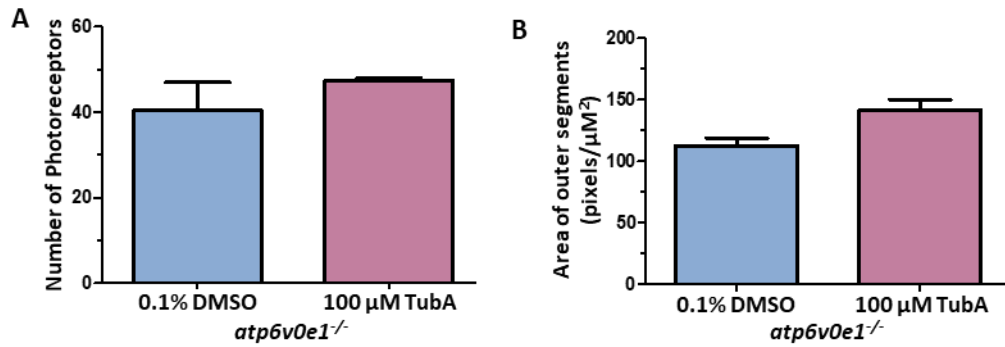


Supplementary Figure 1: Determining maximum tolerated concentration of various HDAC6i compounds in wildtype larvae. (A - C) Wildtype larvae were treated for 3 days

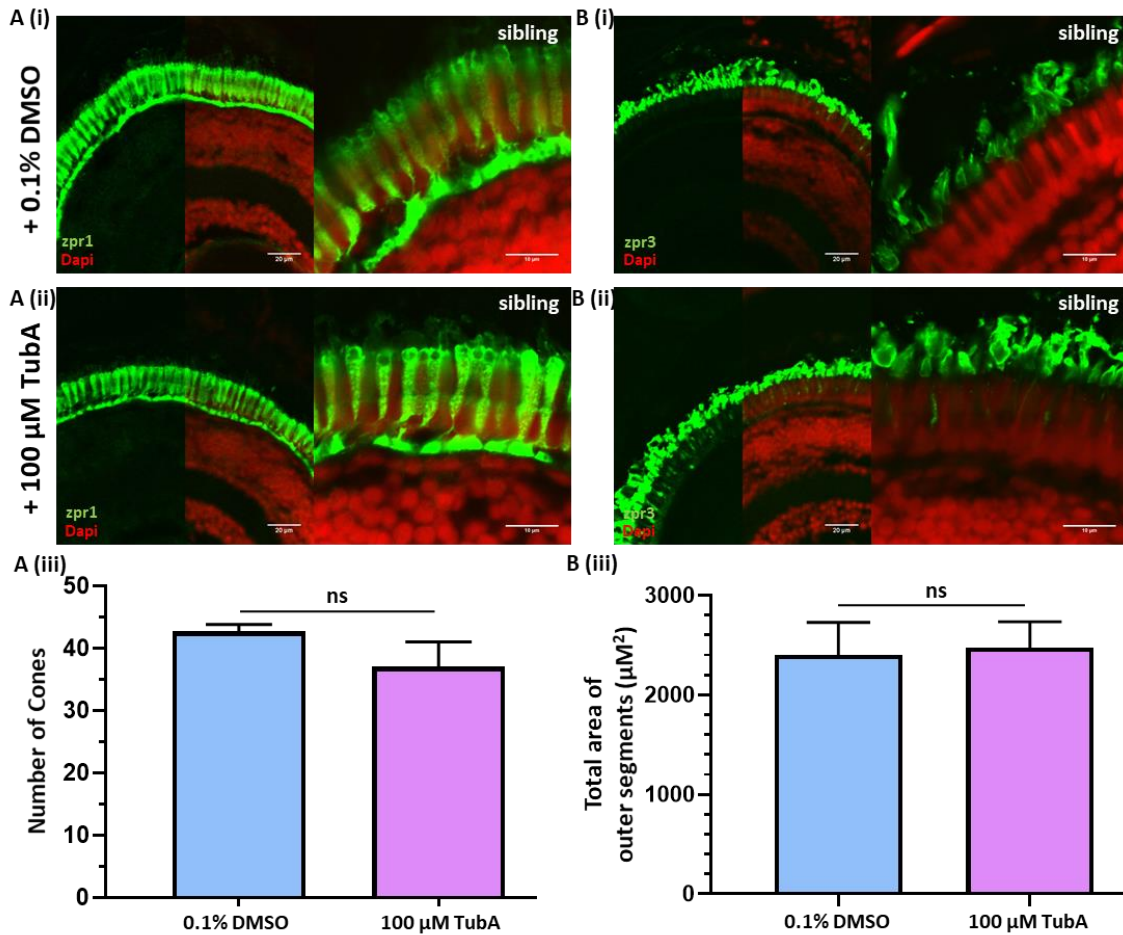
from 3 dpf with selected HDAC6i, at increasing (1, 10, 25, 50 and 100 μ M) doses and 0.1% DMSO was used as vehicle control (N = 1, n = 8 - 12 larvae/treatment group). MTC was determined based on the given criteria 1) an adverse effect was not observed in overall morphology of all surviving (> 80% survival rate) larvae and 2) visual function should not reduce by >15% of average number of saccades. (A) In all concentrations of tubacin, the larvae presented with a visual response, 10 μ M of Tubacin treatment was selected as the MTC, as concentrations at 25 μ M and higher resulted in a reduction in average number of saccades, which could be indicative of potential toxicity in the eye. (B) HDAC6i ACY-1215 precipitated at higher concentrations (25, 50 and 100 μ M), though a visual response was obtained at said concentrations and the larvae did not present with toxicity. 10 μ M ACY-1215 was chosen as the MTC. (C) 50 and 100 μ M of NF2373 was lethal to the larvae. 25 μ M of NF2373 was safe without any adverse effects and determined to be the MTC. Representative larval images are shown on the right.



Supplementary Figure 2: OKR data of siblings for treatment conditions. (A - B) Siblings from *atp6v0e1* line were used as internal controls for the HDAC6i treatment conditions (N = 3 and n = 12/treatment group). Representative larval images are presented on the right.



Supplementary Figure 3: EM analysis of photoreceptor number and outer segment area in *atp6v0e1* mutant retina treated with Tubastatin A. (A) TubA treatment did not result in a marked difference in the number of photoreceptors in *atp6v0e1*^{-/-} compared to vehicle-treated *atp6v0e1*^{-/-}. (B) EM analysis suggests an increase in outer segment area of 25.7% in *atp6v0e1* mutants following TubA treatment, but this has not been demonstrated to be statistically significant (3 sections from the central retina per sample were counted from EM images, N = 2).



Supplementary Figure 4: Immunohistochemical analysis of retina from siblings treated with Tubastatin A. (A - B) Siblings from *atp6v0e1* line were used for analysis of cone and rod photoreceptors histology. Representative retinal images are presented. Graphical presentation of number of cone cells (zpr-1 staining) and total area of photoreceptor outer segments (zpr-3 staining), respectively (3 sections per sample was analyzed, N = 3). Student's unpaired T-test was used for statistical analysis.