

## Supplementary Figure S1. Non-photoreceptor cilia show abnormalities in *ift80* morphants.

(A, B) Cilia within the pronephros of 30 hpf wild type and dynein morphant embryos were immunolabeled with an anti-acetylated tubulin antibody. Images show the anterior pronephros. Anterior is to the left dorsal is up. Gaps were seen in regions of the pronephric duct of *ift80* morphants (brackets). (C, D) 24-hpf ears develop tether cilia (arrows) at the anterior (a; left) and posterior (p; right) poles of the otic vesicle and numerous short cilia that disappear between 27-30 hpf (see text for details). Scale bar, (A, B) 30 μm; (C, D) 25 μm.

## Supplementary Text

As *ift80*-MO injected animals showed photoreceptor defects and IFT proteins are required for cilia integrity, we asked if cilia in other tissues were also affected. We performed whole-mount immunostaining on embryos injected with *ift80*-MO with an antibody for acetylated tubulin to examine cilia in the ear and pronephric duct at 30 hpf. Zebrafish embryos possess bilateral pronephric ducts that serve as simple blood filtration organs. The lumens of these simple kidneys are populated with numerous motile cilia that are necessary to eliminate urine<sup>54</sup>. Wild type embryos exhibited a strong immunofluorescence signal from cilia protruding from the

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pronephric duct (Supplementary Fig. S1A). Cilia in *ift80* morphant pronephric ducts were shorter and present at lower density (Supplementary Fig. S1B), likely resulting in reduced motility and culminating in kidney cysts. The kinocilia of the vertebrate inner ear are also microtubule-based structures with an axoneme similar to that of photoreceptors<sup>55</sup>. At 30 hpf, wild type zebrafish possess 1-2 hair cells in the anterior and posterior regions of the otic vesicle that extent cilia to "tether" the mineralized otoliths<sup>55</sup>. In addition, numerous short cilia protrude into the otic vesicle between 19-24 hpf but then gradually shorten and disappear by 36 hpf (Supplementary Fig. S1C). In the *ift80* morphant embryos, tether cilia appeared shorter than wild type whereas the numerous non-tether cilia were much longer than wild type. Early reports suggested these short cilia are motile, but the exact function for these cilia and the reason they disappear is not entirely understood<sup>55</sup>. Therefore, loss of *ift80* does not prevent cilia formation but does disrupt the normal patterning and density of cilia in both the pronephros and ear of zebrafish.