

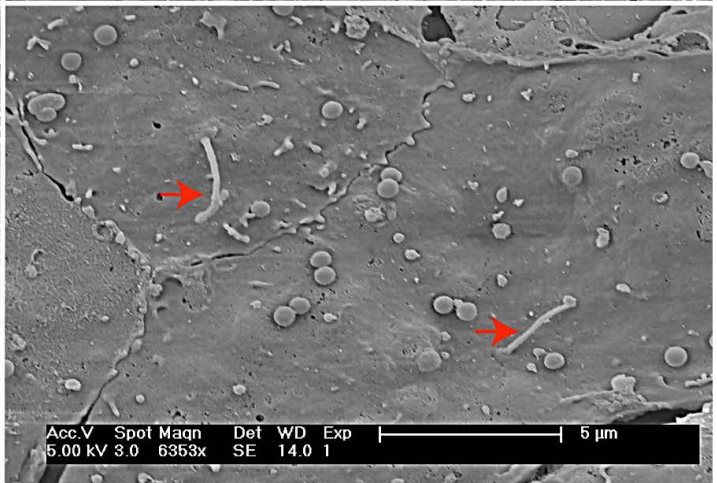
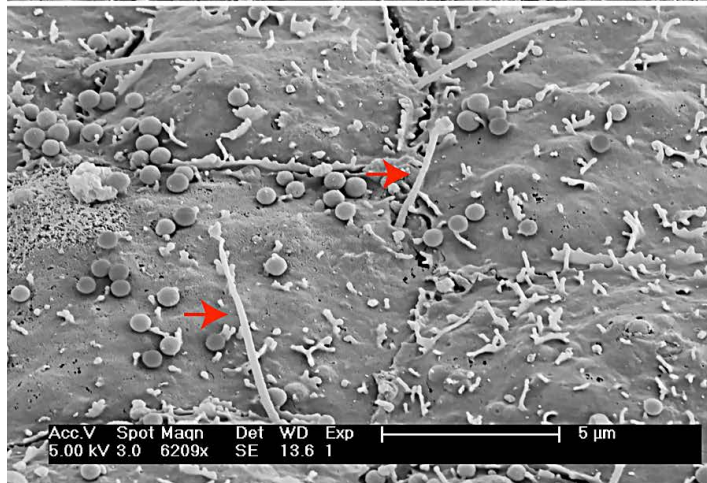
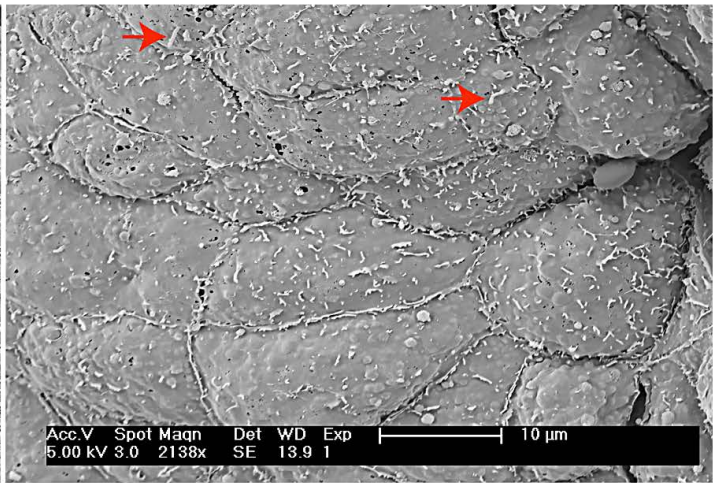
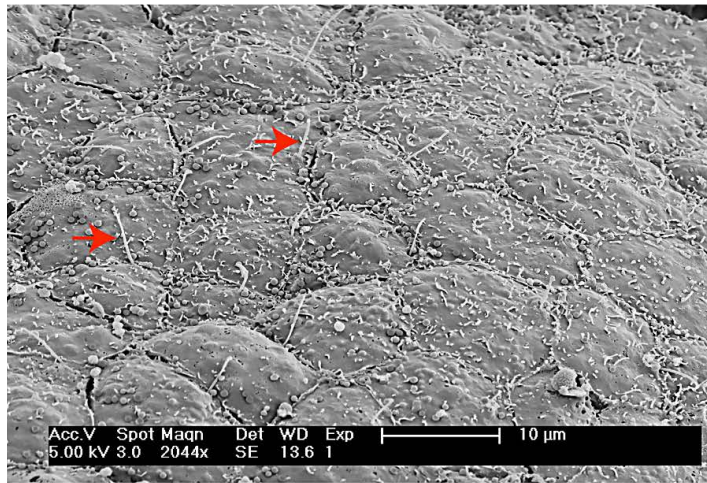
**Figure S1: Early markers of LR patterning in *Xenopus*.**

(a) *In situ* hybridization for *coco* in the *Xenopus* LRO showing normal and abnormal expression patterns. Images are ventral views with anterior to the top.

(b) *In situ* hybridization for *pitx2* in *Xenopus* lateral plate mesoderm showing normal and abnormal expression pattern. Lateral views with dorsal to the top. Embryo sides are indicated above figure.

## Control

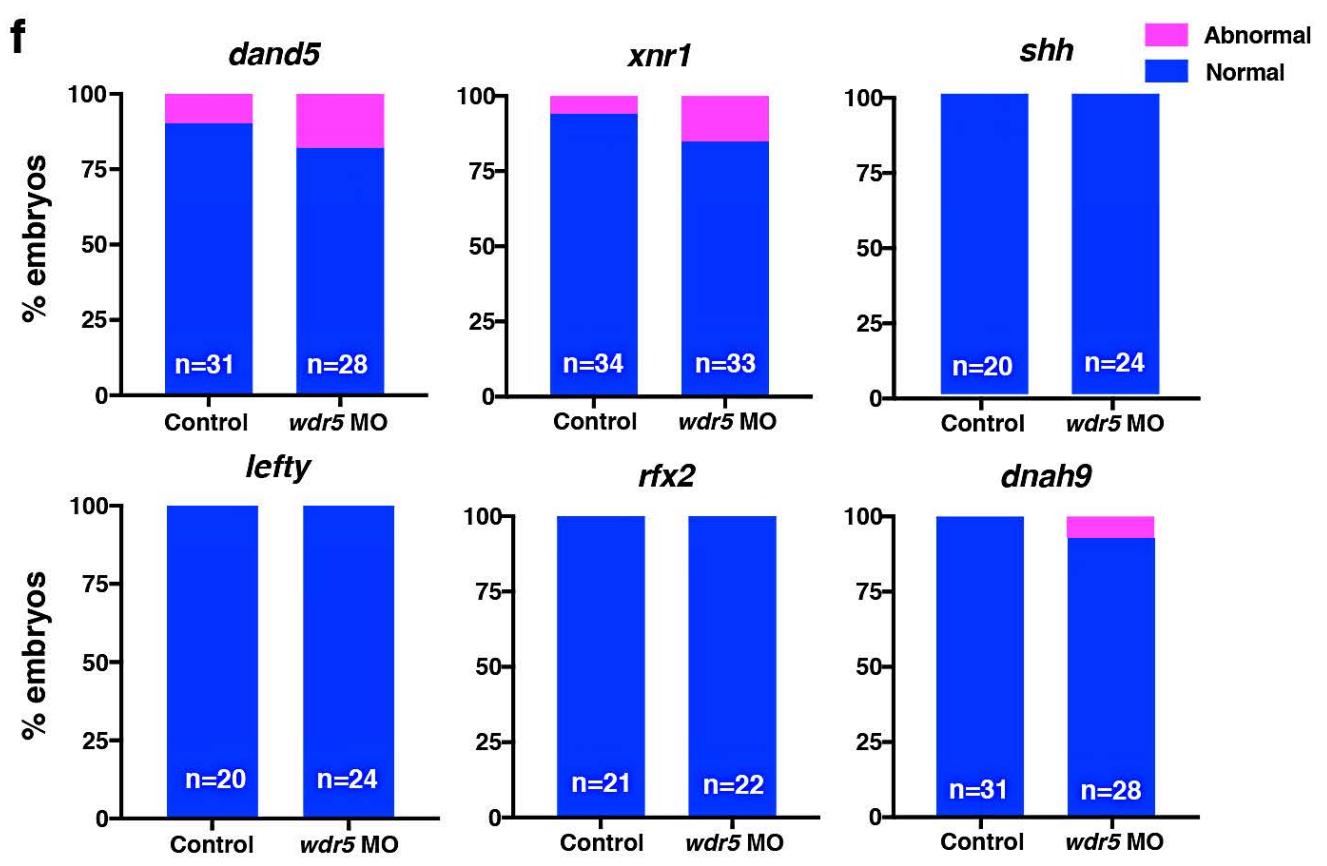
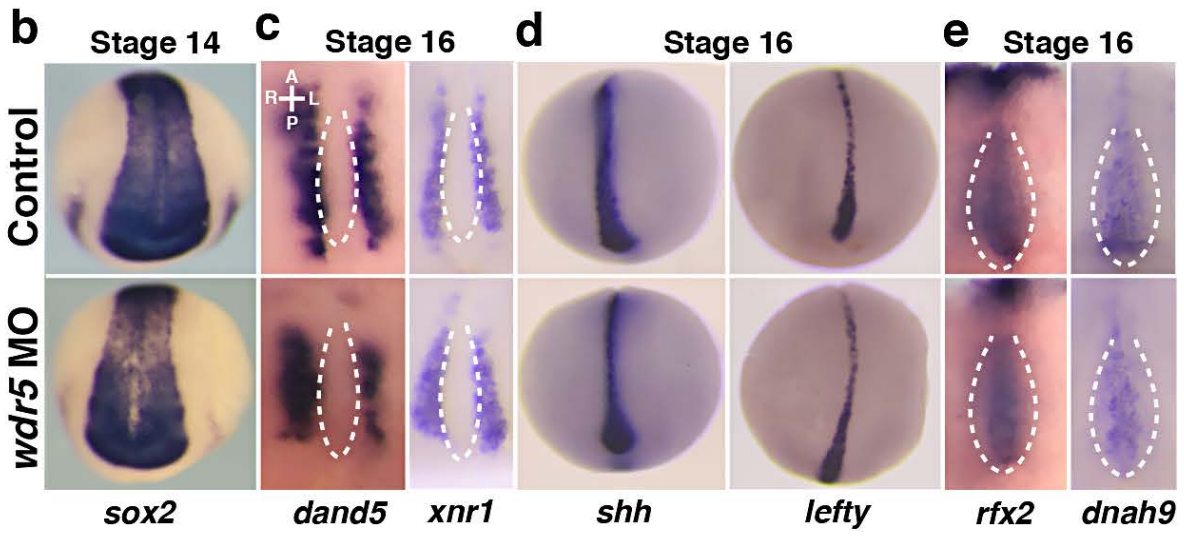
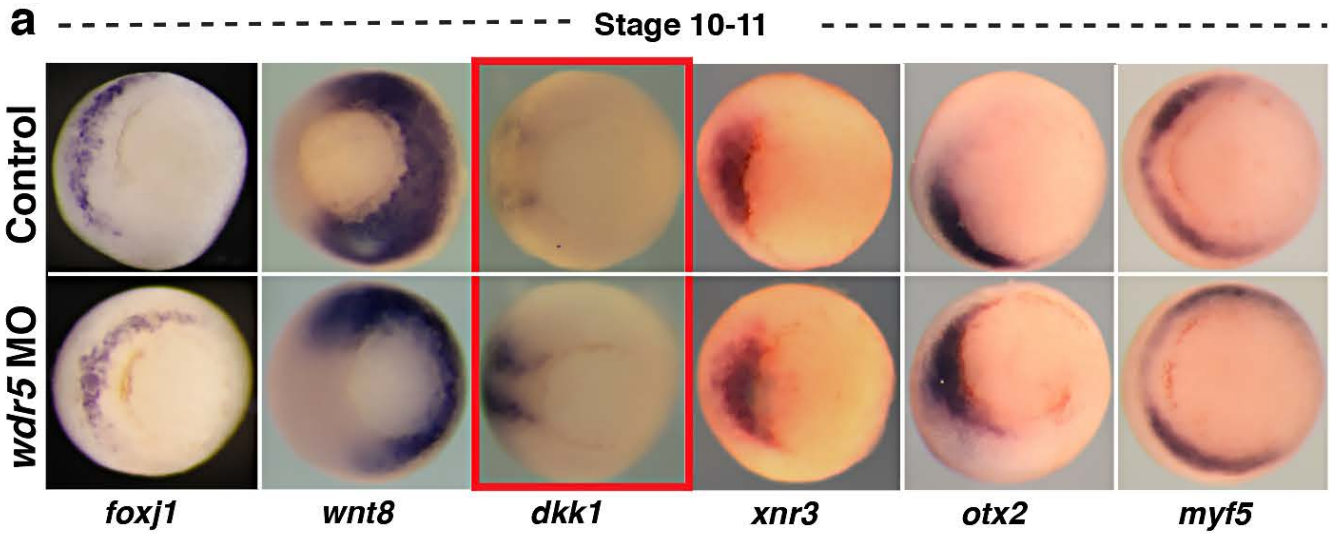
## *wdr5* MO



**Figure S2: Wdr5 depletion leads to defective cilia in the LRO.**

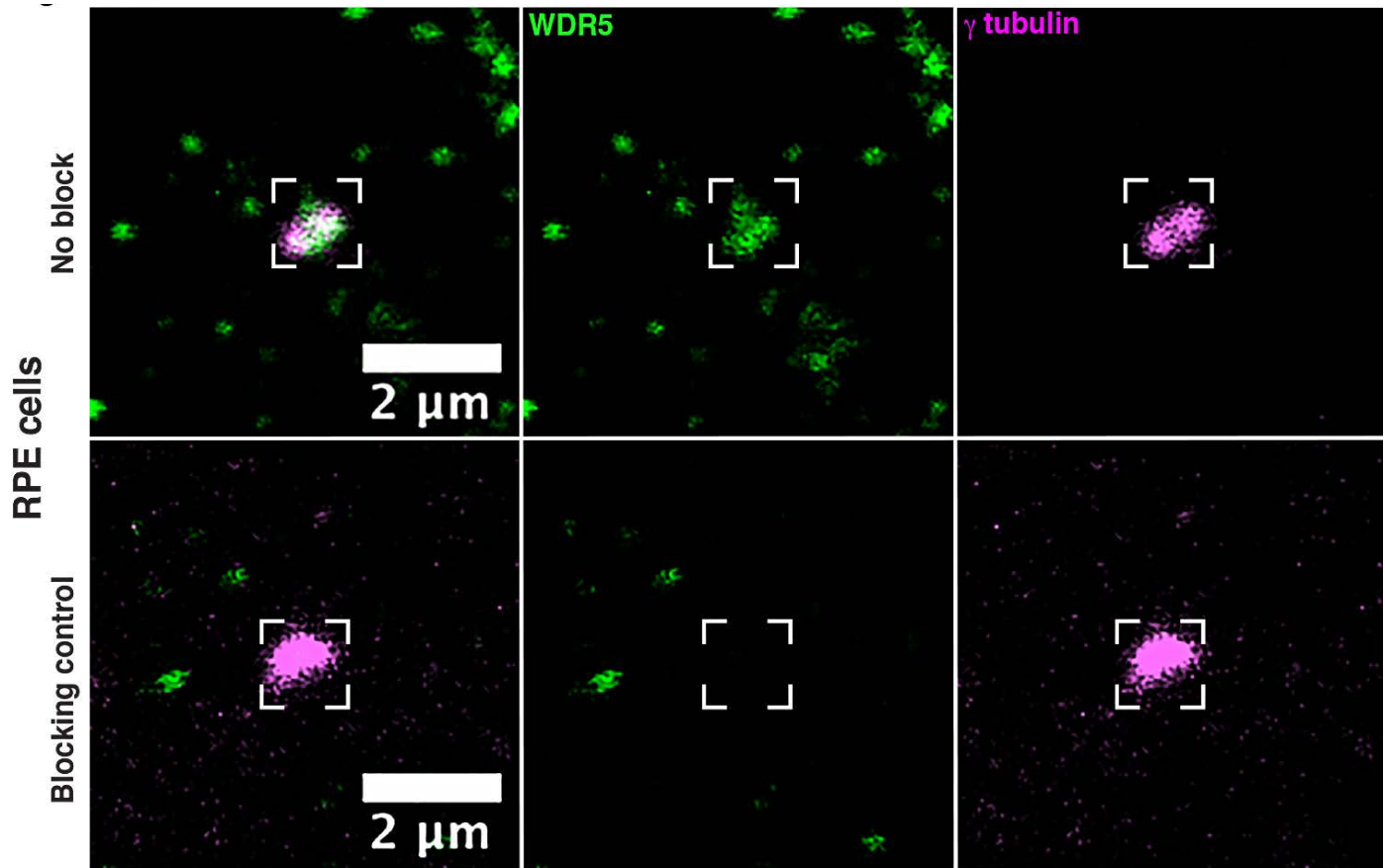
Scanning electron microscope (SEM) images showing cilia morphology (red arrows) in the LRO of uninjected controls and *wdr5* morphants.





**Figure S3: Early markers of dorso-ventral development and cilia cell fate.**

- (a) *In situ* hybridization for early developmental markers at stage 10-11 *Xenopus* embryos in uninjected controls (top row) and *wdr5* morphants (bottom row). All vegetal views with dorsal to the left (at stage 10-11).
- (b) *In situ* hybridization for *sox2* at stage 14, with an anterior view with dorsal to the top.
- (c) *In situ* hybridization for *dand5* and *xnr1* in the *Xenopus* LRO at stage 16. Margin of the LRO is indicated by dashed white lines.
- (d) *In situ* hybridization for *shh* and *lefty* at stage 16, with an anterior view with dorsal to the top.
- (e) *In situ* hybridization for *rfx2* and *dnah9* in the *Xenopus* LRO at stage 16. Margin of the LRO is indicated by dashed white lines.
- (f) Quantification of *in situ* hybridization for the midline marker *dand5*, *xnr1*, *shh*, *lefty*, *rfx2* and *dnah9*. Abnormal is either reduced or absent expression compared to wildtype expression (normal).
- Experiments were repeated 2-3 times. 'n' = number of embryos.



**Figure S4: WDR5 is specifically localized to the base of cilia**

Immunofluorescence showing localization of WDR5 (green) and  $\gamma$  tubulin (magenta) in monociliated human RPE cell. WDR5 signal is lost after incubating the antibody with the blocking peptide. Box region shows the base of cilia where WDR5 is localized in control but not in the blocking control. Experiment was repeated 2 times.