

Table S1. Oligonucleotide sequences

Figure S1. *Thm1* cko cystic kidney disease initiates by P15. Histological analyses of P10, P15 and P20 wt and *Thm1* cko kidneys.

Figure S2. Proliferation persists in P20 *Thm1* cko medulla. (A) Immunostaining of P20 wt and *Thm1* cko kidney sections for PCNA. In wt kidney, band of PCNA+ cells on left side of section is cortex. Note marked reduction in PCNA+ cells in medulla, right of cortex. Similar corticomedullary region is photographed for *Thm1* cko kidney. Note absence of a reduction in PCNA+ cells in *Thm1* cko medulla. All scale bars represent 500 μ m. (B) Proliferation rates in cortices and medullas of 3 wt and 3 *Thm1* cko mice.

Figure S3. Deletion of *Thm1* at 5 weeks of age does not cause renal cysts 3 months following deletion. Histological analyses of wt and *Thm1* cko kidneys 3 months following injection of mice with tamoxifen at 5 weeks of age.

Figure S4. GLI protein levels are similar between wt and *Thm1*^{aln/aln} whole kidney extract. Representative Western blot analyses of protein extracts from (A) E13.5 wt and *Thm1*^{aln/aln} kidneys cultured in cAMP for 4 days and (B) E18.5 wt and *Thm1*^{aln/aln} kidneys. Three cultured kidneys were pooled each for wt and *Thm1*^{aln/aln} from 2 organ culture experiments. Kidneys of 2 E18.5 wt and 2 *Thm1*^{aln/aln} mice were analyzed separately.

Figure S5. Small molecule Wnt inhibitors, IWR-1 and IWP-2, do not reduce cystogenic potential of CD1 kidney explants. E13.5 CD1 kidneys were cultured in the presence of 8-bromo-cAMP, in combination with 10 μ M, 20 μ M or 40 μ M IWR-1 or 10 μ M, 20 μ M or 40 μ M IWP-2 for 4 days. Contralateral kidneys were cultured in control DMSO at concentrations present in IWR-1 and IWP-2 treatments. Graphs represent quantitative assessment of kidney images following 4-day culture. Bars represent mean \pm SEM. $n=4$ kidneys. $*P<0.05$

Figure S6. SMO inhibitor, Sant1, prevents tubular dilation in CD1 kidneys cultured with cAMP. E13.5 CD1 kidneys were cultured in the presence of 8-bromo-cAMP, in combination with either control DMSO or 20 μ M Sant1 for 4 days. Graphs represent quantitative assessment of kidney images following 4-day culture. Bars represent mean \pm SEM. $n=10$ kidneys from 2 experiments. $*P<5.0 \times 10^{-6}$.

Table S1. Oligonucleotide Sequences

alndiag F	5'-CGCTGATTA ACTACTATGGTC-3'
alndiag R	5'-GCGTGGTAAAATCGGAAGAC-3'
jck-F	5'-CTTCCCACCTGTTGCTGTTT-3'
jck-R	5'-CAGTGGGCTTACCACCATCT-3'
Pkd1 ^{m1Bei} -F	5'-CTTCCCACCTGTTGCTGTTT-3'
Pkd1 ^{m1Bei} -R	5'-CCTCAGAGCCCTTCAAGTTCTCTCA-3'
Pkd1 ^{m1Bei} probe	5'-CTTCCCACCTGTTGCTGTTT-3'
Pkd1f-F	5'-CCTGCCTTGCTCTACTTTCC-3'
Pkd1f-R	5'-AGGGCTTTTCTTGCTGGTCT-3'
mPtch1-F	5'-CCTCCTGAACCCCTGGAC-3'
mPtch1-R	5'-CATGCCAAAGAGCTCAACG-3'
mPtch2-F	5'-CCTAGAACAGCTCTGGGTAGAAGT-3'
mPch2-R	5'-CCCAGCTTCTCCTTGGTGTA-3'
mHhip-F	5'-GGCCAGATGGAAGACTACGA-3'
mHhip-R	5'-CCACTCATGACCTCCTGGAC-3'
mGli2-F	5'-GCAGACTGCACCAAGGAGTA-3'
mGli2-R	5'-CGTGGATGTGTTCATTGTTGA-3'
mGli3-F	5'-CACCAAAACAGAACACATTCCA-3'
mGli3-R	5'-GGGGTCTGTGTAACGCTTG-3'
mOaz1-F	5'-GCCTGAGGGCAGTAAGGAC-3'
mOaz1-R	5'-GGAGTAGGGCGGCTCTGT-3'

Figure S1. *Thm1* cko renal tubular dilations begin by P15

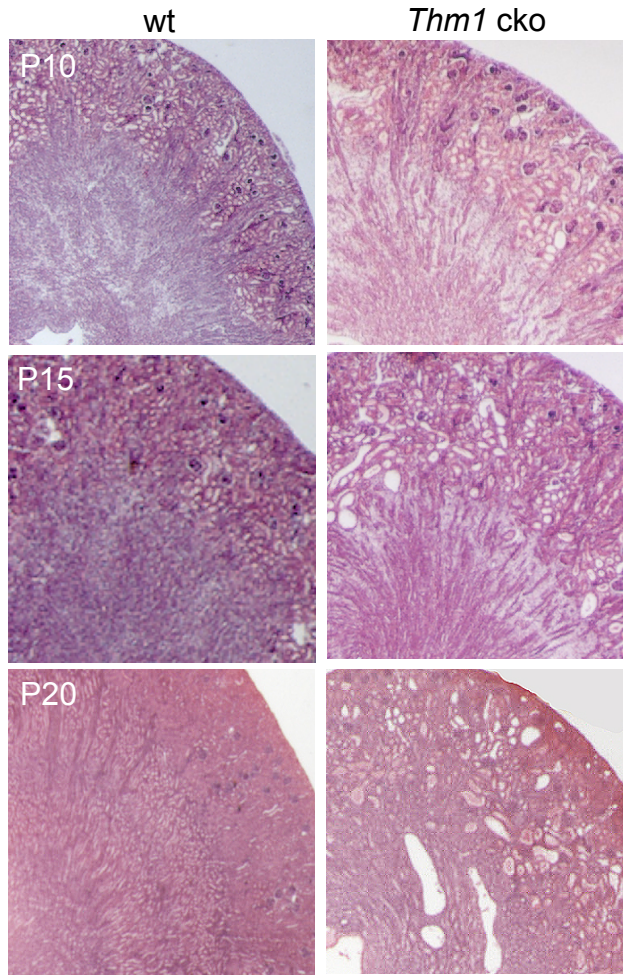


Figure S2. Proliferation persists in P20 *Thm1* cko kidneys

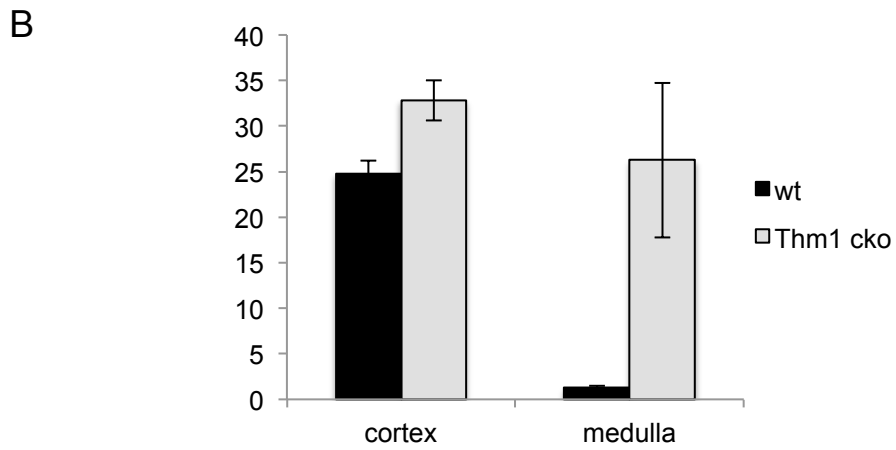
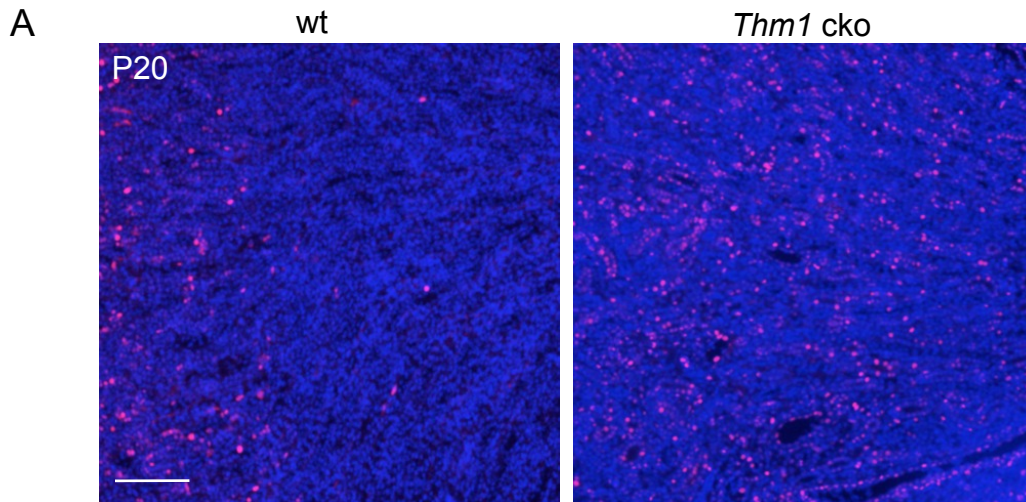


Figure S3. Deletion of *Thm1* at 5 weeks of age does not cause renal cysts 3 months following deletion

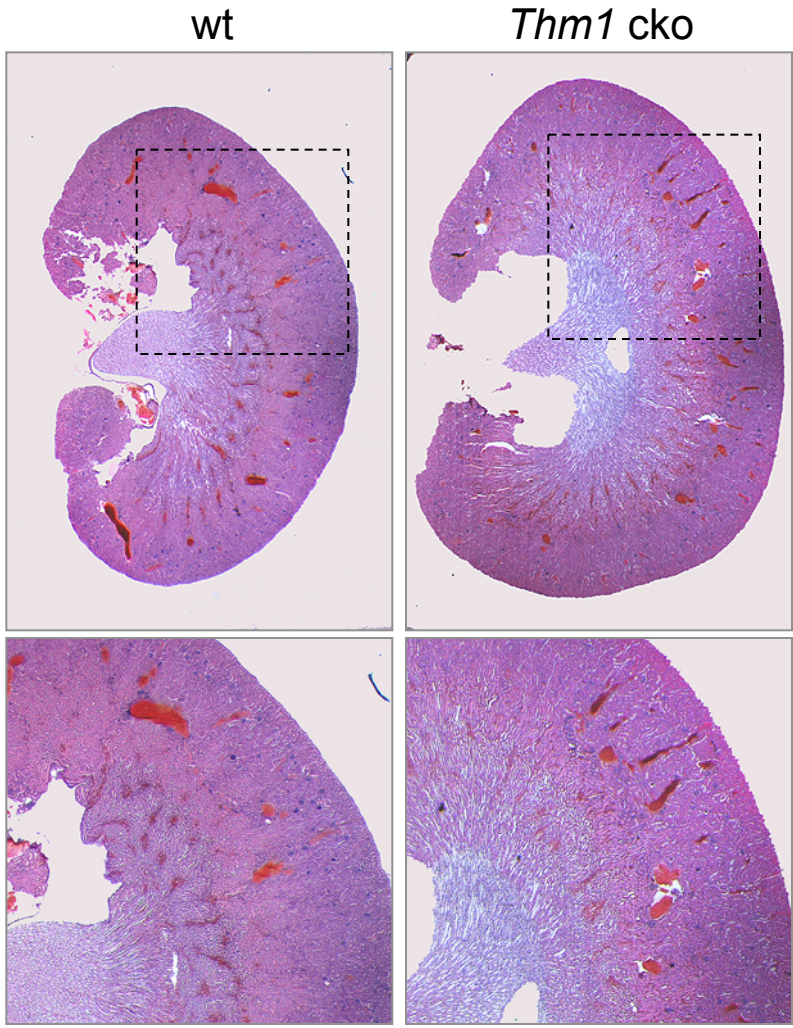


Figure S4. GLI protein levels are similar between wt and *Thm1^{aln/aln}* whole kidney extracts

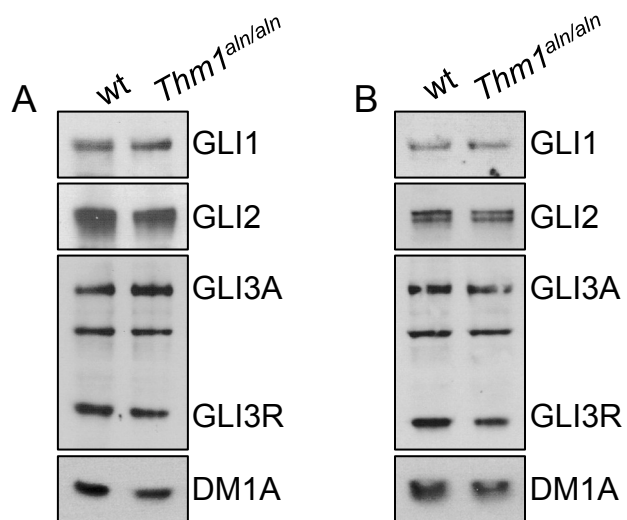


Figure S5. Small molecule Wnt inhibitors do not reduce cystogenic potential in CD-1 kidneys cultured with cAMP

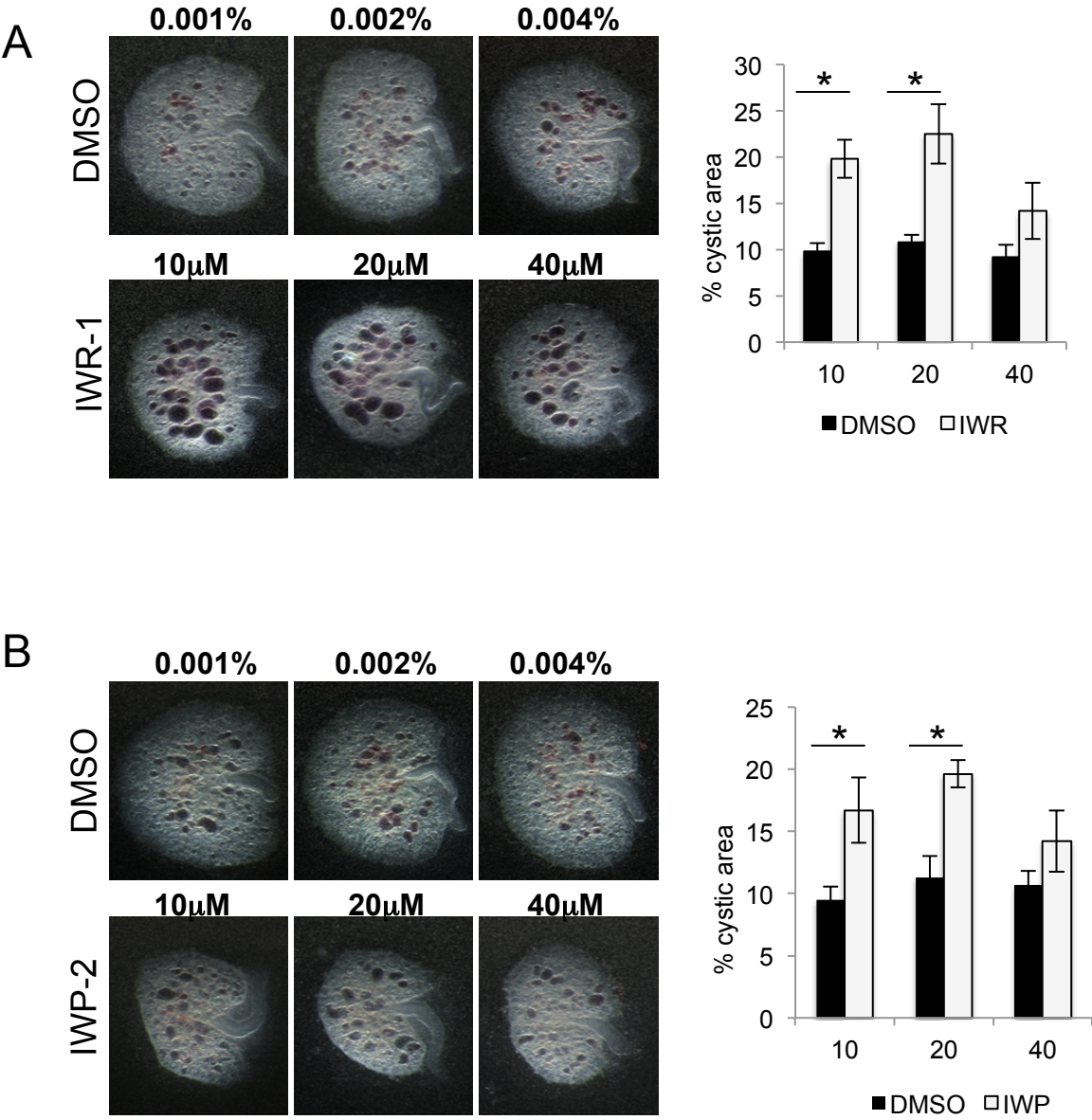


Figure S6. SMO inhibitor, SANT1, prevents tubular dilation in CD1 kidneys cultured with cAMP

