

Supplementary Information

Table S1. Conservation of dynein complex genes * between *Chlamydomonas reinhardtii* and *Mus musculus*.

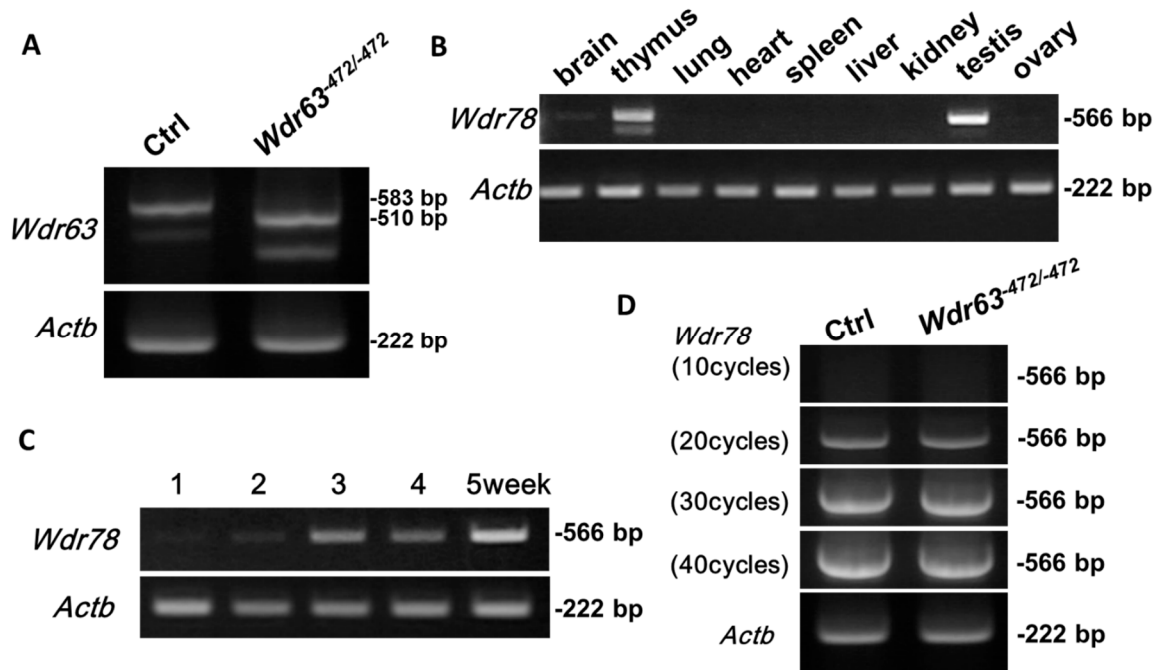
<i>Chlamydomonas</i> Gene	Mouse (HUMAN) Gene	MGI/HGNC Number **	KO Mouse
<i>Outer arm dynein (OAD)</i>			
<i>IC69</i>	<i>Dnaic2 (DNAI2)</i>	2685574/18744	No
<i>IC78</i>	<i>Dnaic1 (DNAI1)</i>	1916172/2954	Yes (ciliary dyskinesia)
<i>LC1</i>	<i>Dnal1 (DNAL1)</i>	1921462/23247	No
<i>LC2</i>	<i>Tctex1d1 (TCTEX1D1)</i>	1914594/26882	No
	<i>Tctex1d2 (TCTEX1D2)</i>	1913311/28482	No
	<i>Tctex1d4 (TCTEX1D4)</i>	3045358/32315	No
	<i>Tcte3 (TCTE3)</i>	98642/11695	Yes (infertile; impaired spermatogenesis)
<i>LC3/LC5</i>	<i>Nme8 (NME8)</i>	1920662/16473	Yes (fertile)
	<i>Nme9 (NME9)</i>	4359686/21343	No
<i>LC4</i>	<i>No determined (ND)</i>	-	-
<i>LC6/LC8/LC10</i>	<i>Dnal4 (DNAL4)</i>	1859217/2955	No
	<i>Dynll1 (DYNLL1)</i>	1861457/15476	Yes (pre-weaning lethality)
	<i>Dynll2 (DYNLL2)</i>	1915347/24596	No
<i>LC7</i>	<i>Dynlrb1 (DYNLRB1)</i>	1914318/15468	No
	<i>Dynlrb2 (DYNLRB2)</i>	1922715/15467	No
<i>DC1</i>	<i>ND</i>	-	-
<i>DC2</i>	<i>Ccdc114 (CCDC114)</i>	2446120/26560	No
	<i>Ccdc63 (CCDC63)</i>	3607777/26669	No
<i>DC3</i>	<i>ND</i>	-	-
<i>Inner arm dynein (IAD) (f/II)</i>			
<i>IC138</i>	<i>Wdr78 (WDR78)</i>	2385328/ 26252	No
<i>IC140</i>	<i>Wdr63 (WDR63)</i>	3045269/30711	No
<i>IC97</i>	<i>Casc1 (CASC1)</i>	2444480/29599	Yes (higher incidence of lung tumour)
<i>FAP120</i>	<i>Ank2 (ANK2)</i>	88025/493	Yes (embryonically lethal)
<i>TCTEX1</i>	<i>Dynlt1a (DYNLT1)</i>	3807506/11697	No
	<i>Dynlt1b (DYNLT1)</i>	98643/11697	No
	<i>Dynlt1c (DYNLT1)</i>	3807476/11697	No
	<i>Dynlt1f (DYNLT1)</i>	3780996/11697	No
<i>TCTEX2B</i>	<i>Tctex1d1 (TCTEX1D1)</i>	1914594/26882	No
	<i>Tctex1d2 (TCTEX1D2)</i>	1913311/28482	No
	<i>Tctex1d4 (TCTEX1D4)</i>	3045358/32315	No
	<i>Tcte3 (TCTE3)</i>	98642/11695	Yes (infertile; impaired spermatogenesis)

* Note; only intermediate, light chains and docking complexes are listed. ** MGI URL: <http://www.informatics.jax.org/>; HGNC URL: <http://www.genenames.org/>.

Table S2. Mean computer assisted sperm analysis (CASA) values for *Dnaic1^{em1Osb/em1Osb}* mice vs. control mice.

Mouse Genotype	Incubation Time (min)	Motility (%)	Progressive Motility (%)	Average Path Velocity (VAP) ($\mu\text{m/s}$)	Linear Velocity (VSL) ($\mu\text{m/s}$)	Curvilinear Velocity (VCL) ($\mu\text{m/s}$)
Ctrl	10	87.0 \pm 7.9	67.7 \pm 5.0	119.7 \pm 6.7	90.1 \pm 7.6	234.4 \pm 5.4
	120	89.3 \pm 4.0	57.0 \pm 3.5	114.4 \pm 9.4	75.5 \pm 6.1	244.8 \pm 34.7
<i>Dnaic1^{em1Osb/em1Osb}</i>	10	91.0 \pm 6.1	73.3 \pm 5.1	125.6 \pm 7.3	96.2 \pm 2.2	217.6 \pm 29.5
	120	78.7 \pm 17.0	54.0 \pm 6.9	120.9 \pm 4.7	73.3 \pm 4.0	255.0 \pm 8.8

Data are expressed as means \pm SD ($n = 3$).

**Figure S1.** (A) RT-PCR for *Wdr63* in control and sKO testes; (B) RT-PCR for *Wdr78* in various tissues and (C) testes samples at different ages; (D) Semi-quantitative RT-PCR of *Wdr78* in control and sKO testes; *Actb* (β -actin) as control.**Table S3.** Mean computer assisted sperm analysis (CASA) values for *Wdr63^{-472/-472}* mice vs. control mice.

Mouse Genotype	Incubation Time (min)	Motility (%)	Progressive Motility (%)	Average Path Velocity (VAP) ($\mu\text{m/s}$)	Linear Velocity (VSL) ($\mu\text{m/s}$)	Curvilinear Velocity (VCL) ($\mu\text{m/s}$)
Ctrl	10	93.3 \pm 4.7	75.0 \pm 3.6	115.5 \pm 4.6	87.4 \pm 4.9	221.0 \pm 21.6
	120	85.7 \pm 13.6	54.3 \pm 7.8	112.8 \pm 14.2	71.1 \pm 6.5	242.8 \pm 37.4
<i>Wdr63^{-472/-472}</i>	10	93.3 \pm 4.2	80.0 \pm 7.5	117.3 \pm 6.3	95.7 \pm 6.2	188.0 \pm 12.8
	120	91.0 \pm 6.9	57.0 \pm 5.0	103.3 \pm 2.4	64.1 \pm 4.4	188.7 \pm 15.0

Data are expressed as means \pm SD ($n = 3$).

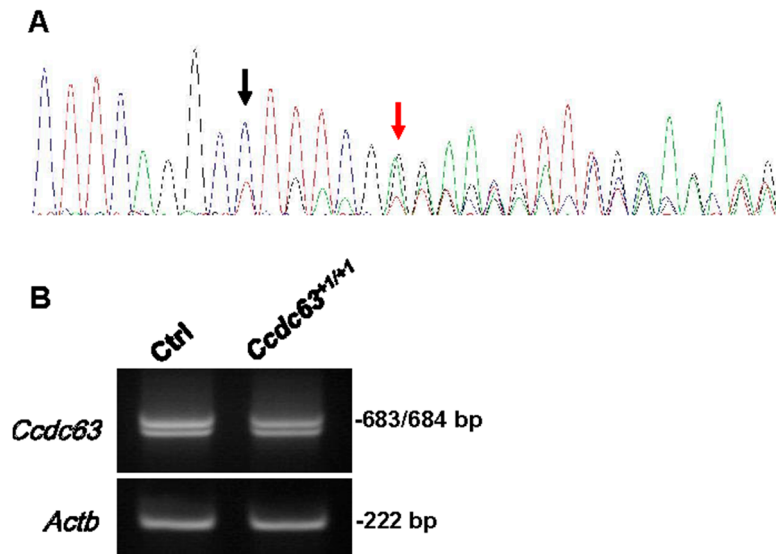


Figure S2. (A) Example of sequencing for a founder mosaic mouse. Black arrow indicates double wave (2 alleles) and red arrow indicates triple wave (3 alleles) (B) RT-PCR of *Ccdc63* in control and sKO testes. *Actb* (β -actin) as control.

Table S4. Off-target (OT) analysis of *Ccdc63* F0 mice.

Target	sgRNA Sequence + PAM*	Location	Mutation
<i>Ccdc63</i>	CCTTCTCG <u>GAAAGTTCCGAA</u> AGG	Chr5: 122129838-122129860	Yes (mosaic)
OT-1	AAAATGATG <u>GAAAGTTCCGAA</u> GGG	Chr1: 132472528-132472550	No
OT-2	CCTTTCGGA <u>ACTTTCTAAATTGA</u>	Chr4: 101217425-101217447	No
OT-3	CAGACTCTG <u>AAGTTCCGAA</u> GGG	Chr19: 18320482-18320504	No

* PAM—protospacer adjacent motif; shown in red; 12 nucleotides homologous to *Ccdc63* target sgRNA underlined in black.

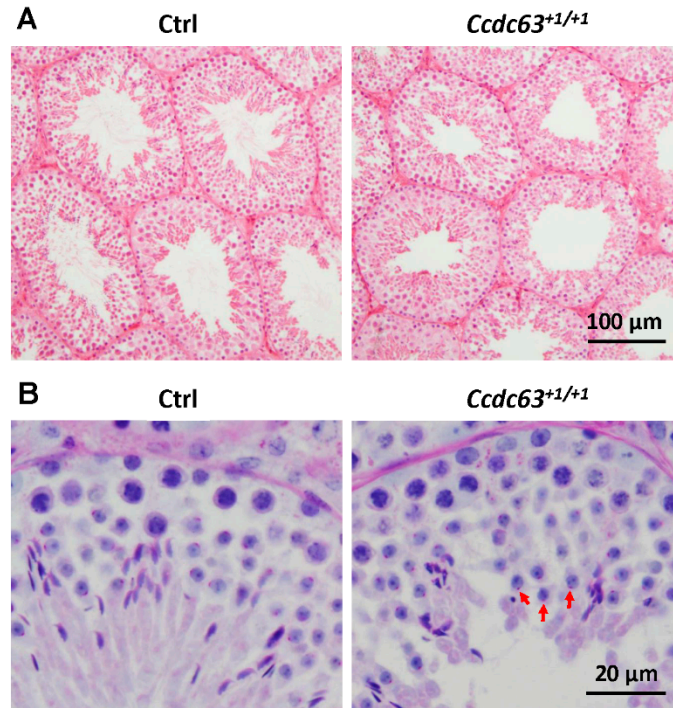


Figure S3. Morphological analysis of testes from control and *Ccdc63*^{+1/+1} mice. (A) Hematoxylin/Eosin stained seminiferous tubules; (B) PAS/Hematoxylin staining of seminiferous tubules (stage III–IV), red arrows indicate developing acrosomes.

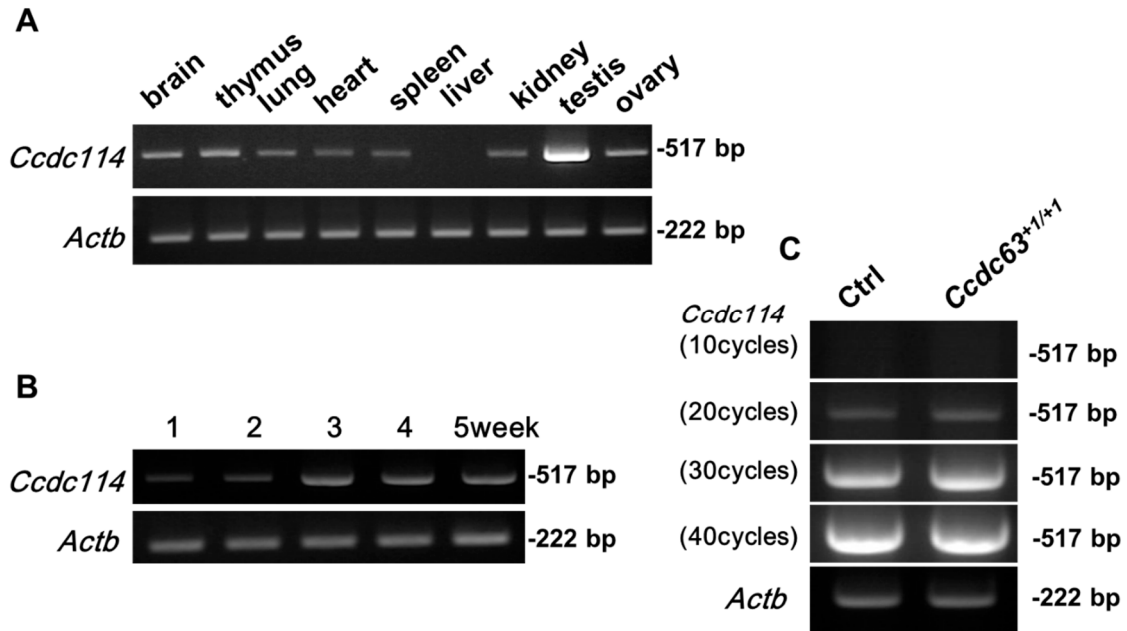


Figure S4. (A) RT-PCR for *Ccdc114* in various tissues and (B) testes samples at different ages. (C) Semi-quantitative RT-PCR of *Ccdc114* in control and sKO testes; *Actb* (β -actin) as control.