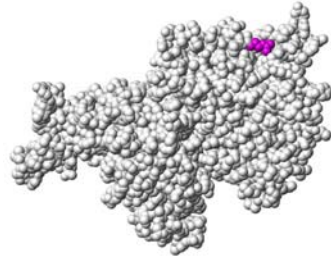


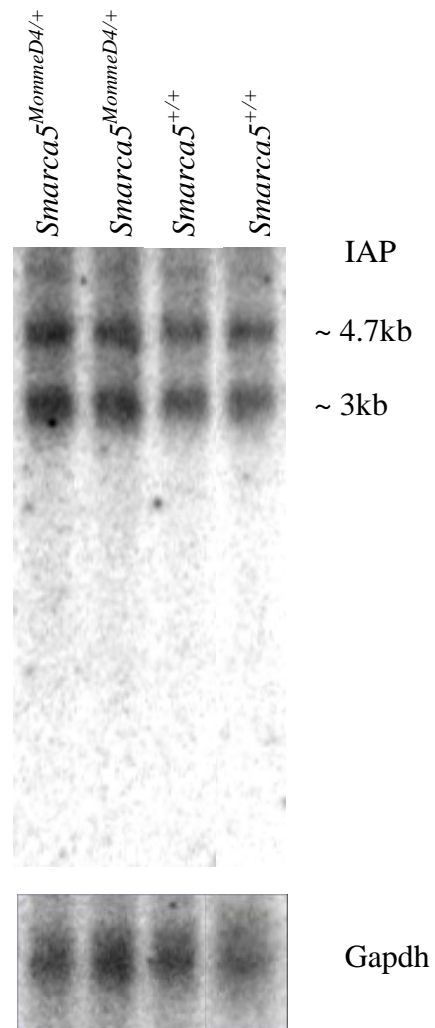
*a*



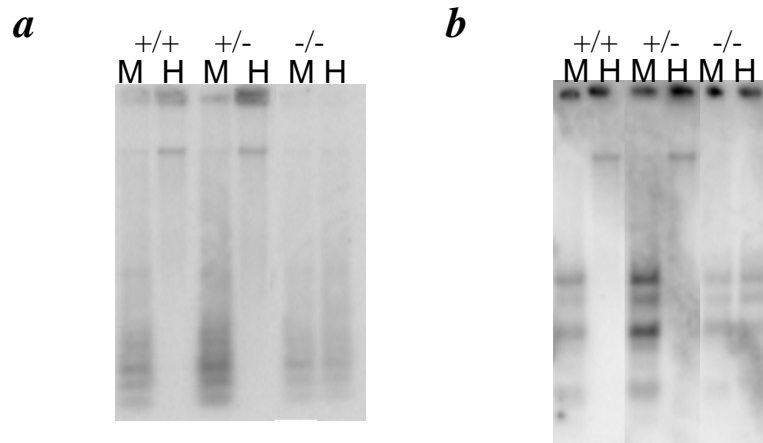
*b*



**Supplementary Figure 1. Homology model of mut-snf2h.** **a.** Ribbon representation showing the sidechain of Arg520 in yellow. **b.** CPK representation showing the sidechain of Arg520 in magenta on the surface of the molecule.

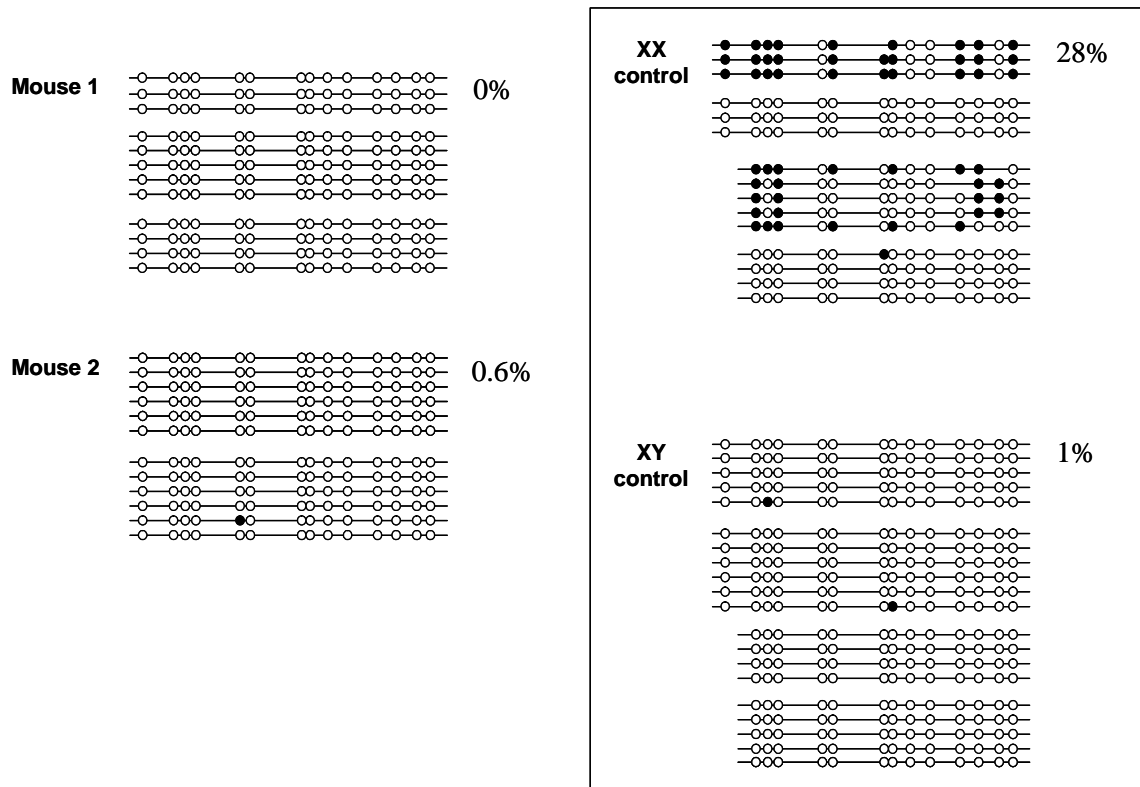


**Supplementary Figure 2. Northern analysis of IAP RNA in *Smarca5*<sup>Momme D4/+</sup> testes. No abnormal IAP mRNA transcripts were detected in *Smarca5*<sup>Momme D4/+</sup> testes.**



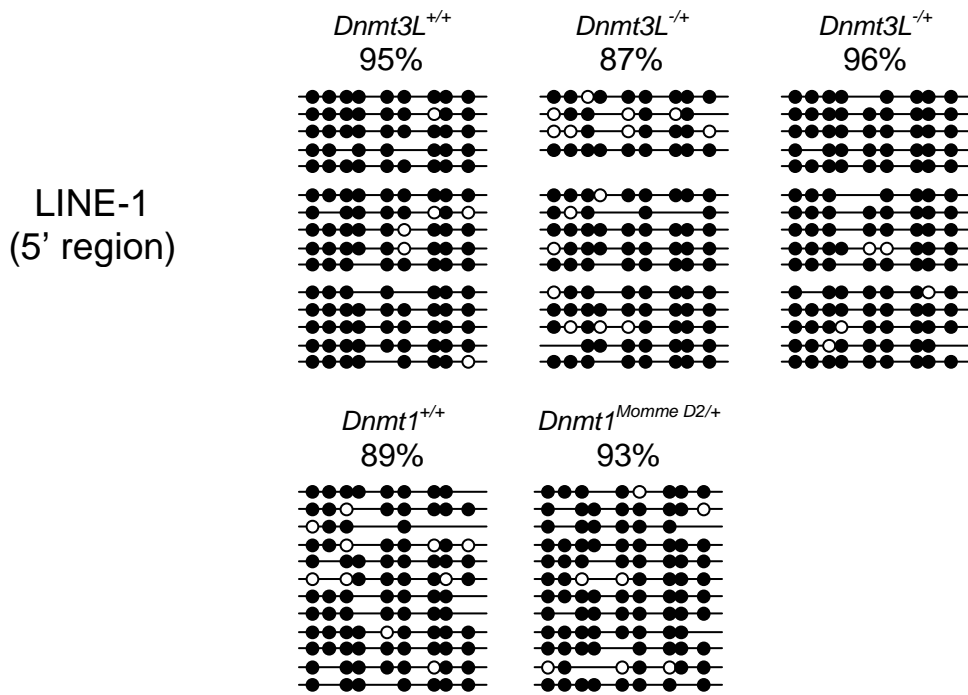
**Supplementary Figure 3. DNA methylation analysis of repeats in *Dnmt1*<sup>MommeD2</sup> embryos .** DNA from *Dnmt1*<sup>MommeD2</sup> wildtype, heterozygous and homozygous embryos (8.5 dpc) were digested with HpaII (H) and MspI (M) and hybridised with **a.** a probe for minor satellite DNA and **b.** a probe for an IAP retrotransposon.

*Mtm1*

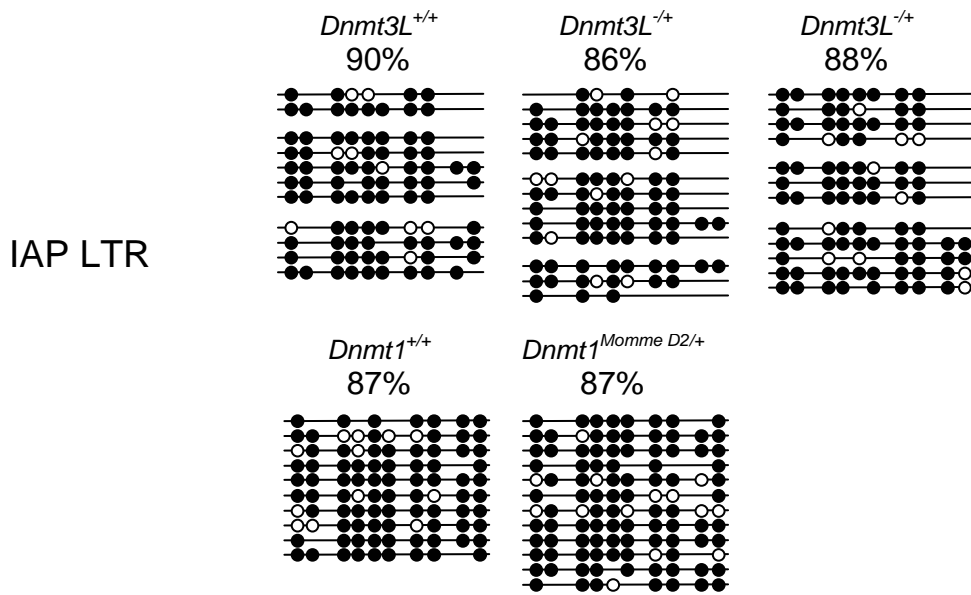


**Supplementary Figure 4. Methylation at the *Mtm1* CpG island in Mouse 1 and Mouse 2.** Mouse 1 and Mouse 2 are two of the three female mice which displayed no methylation at the X-linked *Hprt* gene (see Figure 5). No DNA was available from Mouse 3.

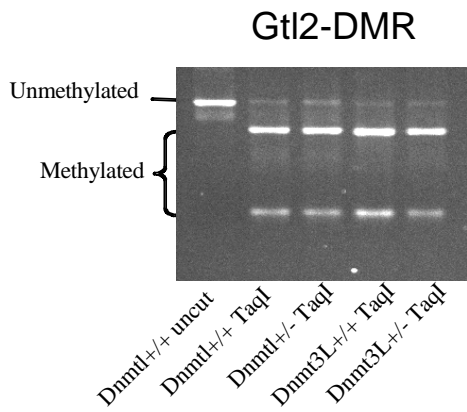
**a**



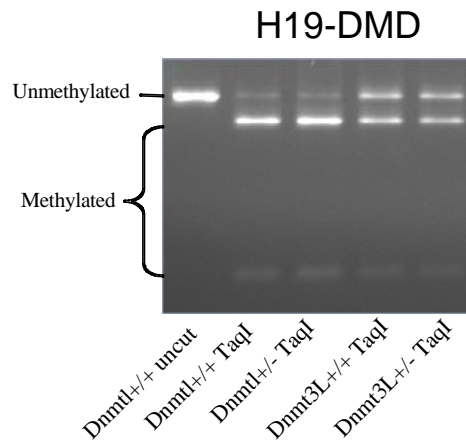
**b**



**c**



**d**



**Supplementary Figure 5. Methylation analysis of DNA from the sperm of *Dnmt1*<sup>Momme D2/+</sup> and *Dnmt3L*<sup>-/-</sup> mice.** Bisulphite sequencing of **a.** LINE-1 and **b.** IAP LTR. (for primers see <sup>46</sup>) **c.** Methylation at the Gtl2-DMR. **d.** Methylation at the H19-DMD (for methods see <sup>49</sup>).

**Supplementary Table 1. Genes in the *Momme D4* linked interval.**

Gene	Ensembl Gene ID	Gene start (bp)	Gene end (bp)
<i>HIP</i>	ENSMUSG00000064325	82866417	82954078
	ENSMUSG00000044691	83006878	83007712
<i>Gypa</i>	ENSMUSG00000051839	83389800	83405220
<i>4930505O20Rik</i>	ENSMUSG00000044588	83408135	83408878
<i>Frem3</i>	ENSMUSG00000042353	83506826	83591102
<b><i>Smarca5 (Snf2h)</i></b>	<b>ENSMUSG00000031715</b>	<b>83595689</b>	<b>83635205</b>
<i>Gab1</i>	ENSMUSG00000031714	83660180	83776225
<i>Usp38</i>	ENSMUSG00000038250	83876479	83910652
	ENSMUSG00000037982	83909322	83934330
	ENSMUSG00000056170	84093373	84094094

Based on Ensembl build 36

**Supplementary Table 2. *Smarca5*<sup>MommeD4</sup> phenotypes.**

**a. Number of homozygous mutant embryos produced from *Smarca5*<sup>Momme D4/+</sup> intercross in the inbred FVB background**

Genotype	14.5 dpc		17.5 dpc	
	Observed	Expected	Observed	Expected
Homozygous	15 <sup>†</sup>	19	12 <sup>‡</sup>	13.5
Heterozygous	40	38	30	27
Wildtype	22	19	12	13.5
Total	77		54	

<sup>†</sup> Three of these embryos displayed an abnormal phenotype

<sup>‡</sup> All but one of these embryos displayed an abnormal phenotype: four were white and regressing, consistent with macrophage invasion, and seven were anatomically normal but small.

**b. Body weight at weaning of *Smarca5*<sup>MommeD4/+</sup> mice**

Weight at 3 weeks of age (g)

Male offspring		Female offspring	
<i>Smarca5</i> <sup>+/+</sup>	<i>Smarca5</i> <sup>MD4/+</sup>	<i>Smarca5</i> <sup>+/+</sup>	<i>Smarca5</i> <sup>MD4/+</sup>
11.9 ± 0.5	10.7 ± 0.9	11.5 ± 0.5	10.3 ± 0.8
n=10	n=20	n=13	n=12
p<0.005		p<0.005	

Offspring were generated from *Smarca5*<sup>Momme D4</sup> heterozygous males crossed with wildtype females

**Supplementary Table 3. Primers**

Name	Sequence
DXMit1F	caagcaaccgaggaagacat
DXMit1R	caggatgctaataccacctgc
DXMit117F	gtaaaaccgcactgactaagcc
DXMit117R	ttcgcctctatcttactctggc
Hprt4-L	gatttttgagtttggttagatgtt
Hprt4-R	cacaaatataaattataacatctac
Hprt4-Lint	gtggggatgtttttagtgagtt
Hprt4-Rint	caaacctaaacataccctctcatac
Mtm1SEQ-L	ggggatttaggattggaaaggttgaagga
Mtm1SEQ-R	taaccttcctaaacccaattccccattc
Mtm1SEQLshort	aggaaaaagtagggatttttgaga
Mtm1SEQRshort	taacacaaaaacactaaccaaaaa
Mtm1L	ttggttttattgtttattgttttag
Mtm1R	acattcttaataactattcacacttc
MD4L	ggaatggctggcctaggta
MD4R	ctcgtatgcaatacaatacaaaaca
MD2L	cggcattccagatgattcc
MD2R	tgtcagcctcggctctaac
<i>Smarca5-ex11L</i>	cagcaggggaagatggacaaa
<i>Smarca5-ex11R</i>	aacaccaccatcttgcact