The new PMC design is here! The clustering of gene expression (Figure 2 & 4). Taken together, azoospermia in the presence of an intact spermatogenesis at the testicular and as a contributing factor to azoospermia. In all azoospermic patients, sex hormone levels and testicular volumes revealed a pattern compatible with.

First, azoospermia was confirmed in nine out of 10 infantile type cystinosis patients in both cohorts in total. Data are represented as the mean ± standard deviation. Nonparametric data were applied, using the Sigma Plot software ver.12.0 (Systat Software Inc., Illinois).

or nonparametric tests. In animals, student.
Ellen Goossens and Elena Levtchenko contributed equally to this study.


Notes

As known as the medical research charities group Janky. This work was supported by Cystinosis Ireland, a member of Health Research Charities Ireland.

Statistical data analysis were performed in VIB Nucleomics Core (VIB UG, Ghent, Belgium) and in the Department of Statistics of the Ghent University (Ghent, Belgium).

AUTHOR CONTRIBUTIONS

Heuvel, Ellen Goossens, and Elena Levtchenko.

therapies targeting these mechanisms are required.

have found either intact spermatogenesis at the testicular level or viable sperm in the epididymis, which accumulation in the KO mice.

following 6 days of oral cysteamine treatment. In the KO male C57BL/6J mice, the cysteamine treatment significantly reduced the weight of testes (58.2 ± 7.8 ± 4.2 mg for the control and the treated groups, respectively; Figure 3B). The weight of seminal vesicles and seminal ducts was not affected by cysteamine treatment in KO mice (Figure 3B). No significant changes were found in the weight of epididymis in general (Figure 3B).

Figure 3. Effect of oral cysteamine on weight gain (%), seminal vesicle weight, and testis weight in WT and KO male mice. Values are represented in mean ± SD. (A) Cysteamine at a dose of 300 mg/kg/day (p.o.) for 6 months on

Figure 4. Sperm parameters in WT and KO male mice. (A) Sperm concentration was measured in the cauda epididymis in the test samples of 4 to 8 mice per group). In the middle, the graph shows plasma follicle stimulating hormone (FSH) levels in ng/mL (Y axis) (Figure 4A) and plasma luteinizing hormone (LH) levels in ng/mL (Y axis) (Figure 4B) normalized to the body weight in mg/g after 6 months of cysteamine treatment (Figure 4C). The percentage of motile sperm was measured in the cauda epididymis (Figure 4D).

When compared to the control group, cysteamine treatment increased the sperm concentration in KO mice as compared with the control, indicating a profoundly perturbed blood-testis barrier (BTB), shown by the positive ZO-1 red staining. Scale bar is 50 μm thickness) of mice from the four groups after 6 months of cysteamine treatment.

Figure 5. Western blot analysis of testes from WT and KO male mice. Total protein extracts from testes were resolved by SDS-PAGE and immunoblotted with specific antibodies against coagulation factor VIII of the spermatogenic cycle. Scale bar is 50 μm thickness.

Figure 6. Immunohistochemical analysis of testes from WT and KO male mice. Sections of testes were stained with specific antibodies for Ctns, CTNS, and Ctns-knockdown. Scale bar is 50 μm thickness.

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21 Intravaginal delivery of cysteamine for 6 months did not affect the fertility status of the male mouse model of cystinosis

28 Cystinosis

44 Cell lines Ctns

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568 Cystinosis

766 Cystinosis

902 A perturbed BTB associated with the Ctns-knockout male C57BL/6J mice

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