Bilateral testicular atrophy and normal Inhibin B level : A paradoxal clinical finding for a rare biochemical cause !

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Background

- **Testicular atrophy is a rare complication following inguinal hernia repair particularly in children <2 years and those with an undescended testis[1] .**
- **Inhibin B is produced in the testis, principally by the Sertoli cells, and has been suggested as a good marker for spermatogenesis.**
- **Its value is expected to be very low in children with bilateral testicular atrophy.**

Case presentation

A 7-year-old child presented to pediatric endocrinology clinic for "bilateral testicular atrophy"

- **Past medical history:**
  - **Surgery at 6 years (2013) for undescended testis.**
  - **Intraoperative exploration showed two very small testis :**
    - ✓ The right one was fixed in the scrotum
    - ✓ The left one was fixed in the pubis (undescendable)
  - **Three years later (2016):**
    - ✓ **Testicular ultrasound**: the right testicle was hypotrophic and heterogeneous while the left testicle was not seen.
    - ✓ The 2 testis were non palpable
    - ✓ **Left orchidectomy** => Histopathology: testicular atrophy
- **Examination:**
  - Weight = 24 kg (-1DS; M); Height = 127 cm (-1DS; M)
  - Normal penis of 5 cm, apical meatus
  - 2 non palpable testes
- **Investigations:**
  - **Karyotype : 46,XY**
  - **Hormonal balance shows :**
    - ✓ **LH** = 0,4 UI/L (NR:0.02-3.6); **FSH** = 12,10 UI/L (NR≤2,3)
    - ✓ **Test HCG: Testosterone < 0,025 ng/mL before and after HCG stimulation test** (NR in prepubertal boys:<0.06)
    - ✓ **AMH < 0,01 ng/mL** (NR: 32-167)
    - ✓ **Inhibin B** = 113 ng/mL controlled at 139 pg/mL (NR:35-182)

➔ **We thought about extragonadal production of Inhibine B ???**

- **But:**
  - **AFP** = 1 ng/mL (NR<9)
  - **BHCG** < 0,5 mU/mL (NR<5)
- **We though seek for the ELISA assay technique (of Beckman Coulter) and blood samples were adressed for a dosage using the less common ELISA technique of Anshlabs:**
  - **Inhibin B < 4.6 pg/mL** (NR: 35-182)
    - **Cause of disrepency: Heterophilic antibodies**

➔ **Final diagnosis: bilateral testicular atrophy due to late surgery for undescended testis**

- **Follow-up:**
  - **Last examination: Age 10 years 8 months (Fig.1):**

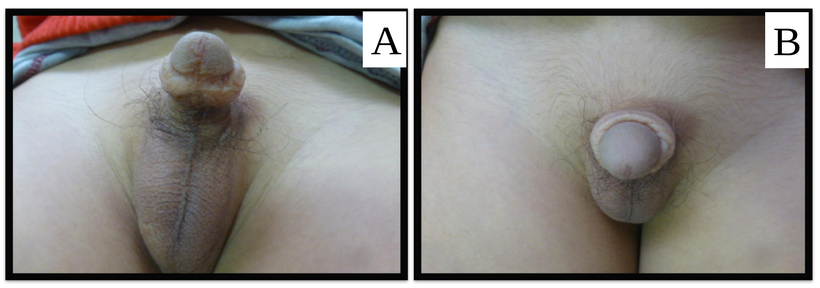

Figure 1: bilateral testicular atrophy with normal penis and pubic hair.

  - **Fertility is thus found to be compromised.**
  - **The child will receive a testosterone replacement therapy when aged 12 in addition to bilateral testicular prothesis.**

Discussion

- **Heterophilic antibodies are present in a significant proportion of the population, and are likely to give a false-increased result in sandwich assays (such as the inhibin assay)[2].**
- **They may arise in a patient in response to exposure to certain animals or animal products, due to infection by bacterial or viral agents, or nonspecifically.**
- **This interference can be found nowadays in 1 serum/10 000.**

Conclusion

- **Hormonal assays are often the diagnostic pivot in pediatric endocrinology.**
- **Being aware of biochemical causes of paradoxal hormonal dosages can be a key to avoid unnecessary additional explorations.**
- **Reanalyzing a specimen using a different assay platform is a straightforward approach to overcome assay interference.**

References:

1. Sonderman KA, Wolf LL, Armstrong LB, Taylor K, Jiang W, Weil BR, et al. Testicular atrophy following inguinal hernia repair in children. Pediatr Surg Int 2018;34:553–60.
2. Tate J, Ward G. Interferences in immunoassay. Clin Biochem Rev. 2004 May;25(2):105-20.

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Sex differentiation, gonads and gynaecology or sex endocrinology

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