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 testis¹.
- 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 disrepecy: 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.](image)

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

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 inhibit 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: