

Bilateral Testicular Microlithiasis with Leydig Cell Hyperplasia: A Case Report

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ABSTRACT

Testicular microlithiasis is a rare finding in intratesticular pathology, and its association with Leydig cell hyperplasia is not widely reported. We present a case of such an association. 19-year-old male presented with bilateral testicular microlithiasis on ultrasound, and elevated tumor markers. Inguinal orchiectomy revealed Leydig cell hyperplasia. Discussion: Testicular microlithiasis is increasingly detected due to advancements in ultrasound technology, but its clinical significance remains unclear. Leydig cell hyperplasia, characterized by testicular nodules, is rarely associated with microlithiasis.

This case underscores the need for vigilance in evaluating testicular abnormalities, as severe microlithiasis and Leydig cell hyperplasia may mimic malignant lesions.

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INTRODUCTION

Testicular microlithiasis is a rare finding when studying intratesticular pathology; The literature has shown that testicular microlithiasis represents between 2.7% to 5% of testicular ultrasound findings.⁽¹⁾

Testicular microlithiasis is a rare and little-known disease that can be associated with cancer and infertility.⁽¹⁾

Its association with Leydig cell hyperplasia does not seem to be reported in the bibliography. A rare case of such an association is presented.

CASE PRESENTATION

A 19-year-old male with no history of cryptorchidism, with a healthy brother, presents with right testicular pain of a week's duration and induration of the right testicle. On physical examination, apparent age and secondary sexual development were normal, little beard, no gynecomastia, no abdominal or supraclavicular masses, testicles with slight induration and little pain but on the right side. Ultrasound (Pic 1) presence of significant bilateral testicular microlithiasis. Laboratory tests with elevated values of alpha-fetoprotein 12.0 ng/mL and beta fraction of human chorionic gonadotropin 2.0 mIU/mL. Normal spermatobioscopy. The patient underwent inguinal orchiectomy and the pathology report was testicular

parenchyma with lamellar dystrophic microcalcifications and Leydig cell hyperplasia. (Pic 2)

DISCUSSION

The superficial position of testes within the scrotum makes them ideal for ultrasound evaluation. Recent advancements in ultrasound technology have improved image quality, leading to increased detection of testicular microlithiasis. Microliths in testes, comprised of calcifications surrounded by collagen fibers, result from a failure of phagocytosis by Sertoli or spermatogenic cells.⁽³⁾ This accumulation may trigger an immune response, causing deposits in the seminiferous tubules. The clinical significance of microlithiasis and its association with testicular germ cell tumors remains unclear.⁽³⁾ However, studies suggest a correlation between microlithiasis and intra-tubular germ cell neoplasia (ITGCN), a precursor to invasive testicular germ cell tumors.⁽³⁾ Various risk factors such as cryptorchidism, testicular atrophy, subfertility, family or personal history of testicular cancer, and testicular dysgenesis syndrome are associated with an increased prevalence of microlithiasis and raise the risk of developing testicular tumors.⁽⁴⁾ Therefore, its presence in a first-degree relative might indicate a predisposition to testicular cancer.⁽³⁾ The management of patients with

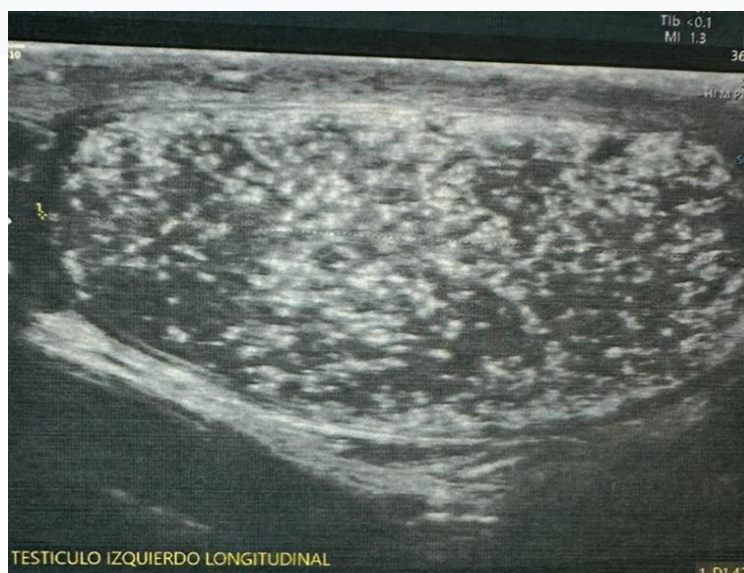
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testicular microlithiasis varies significantly among practitioners. According to EAU guidelines, patients without testicular germ cell tumor risk factors should be encouraged to perform self-examination. ⁽³⁾ However, there is no consensus on managing patients with microlithiasis and risk factors for testicular germ cell tumors. ⁽⁵⁾ Testicular biopsy remains the gold standard to detect intra-tubular germ cell neoplasia (ITGCN), while immuno-cytological markers in semen are not recommended due to high false negative rates. ⁽²⁾ An individualized approach based on patient age, concurrent features of testicular dysgenesis syndrome, fertility status, desire for paternity, and ultrasound pattern is recommended. ⁽⁷⁾ Observation versus testicular biopsy is debatable in various scenarios, including patients previously treated with orchiectomy for testicular cancer and harboring microlithiasis in the contralateral testis. ⁽⁴⁾ Immediate biopsy can detect ITGCN, a highly curable condition with radiotherapy alone, thereby avoiding orchiectomy and the

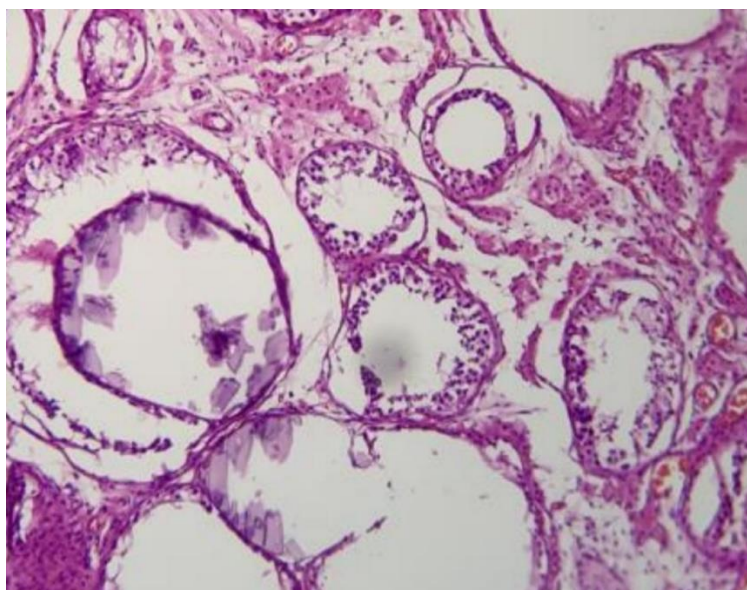
need for testosterone replacement therapy. ⁽³⁾ Leydig cell hyperplasia is a rare benign condition characterized by small, multifocal and frequently bilateral testicular nodules, and can present primarily, producing precocious puberty in male children, or secondary, as a testicular mass with gynecomastia and development of marked sexual characteristics. ⁽⁵⁾ To our knowledge, the association of testicular microlithiasis and leydig cell hyperplasia has not been reported, which is the reason for the present case. ⁽⁵⁾

CONCLUSION

The present case made us suspect a malignant lesion with pain and slight elevation of tumor markers, the pathology report confirmed severe testicular microlithiasis and the finding of Leydig cell hyperplasia, with the patient not presenting any clinical condition of suspicion for this pathology. It should alert us to the possible scenarios of testicular alterations and the close monitoring of these patients.



(Pic 1) USG ultrasound showing testicular microlithiasis.



(Pic 2) Testicular pathology sample shows multifocal lamellar dystrophic microcalcifications.

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