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Case Report

Testicular fibroma of gonadal stromal origin in an adult male

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ABSTRACT

Para-testicular masses are a rare entity, and therefore the diagnosis and management nearly always lead to clinical doubts. Aside from the doubts that arise from these masses being uncommon, it is always necessary to rule out the malignancy process of them. Sexual cord tumors are extremely rare. Testicular fibroma of gonadal stromal origin is a proliferative process that can develop in para-testicular structures. The objective of our study is to present a rare case report of testicular fibroma of gonadal stromal origin as well as the well-documented diagnostic process and the successful therapeutic management that was subsequently carried out.

We report a case of a 68-year old male who came in for a consult due to the casual finding of a nodule in his left testicle with normal tumor markers. Ultrasonography showed a nodular image that was well-defined with a diffusely homogeneous echotexture; it was also hypoechoic, vascularized and demonstrated hydrocele. MRI revealed a solid tumor with extrinsic growth to the left testicle and epididymis, and the lesion was relatively hyperintense in T1-weighted image and hypointense in T2. A surgical exeresis of the para-testicular tumor and hydrocelectomy was performed. The pathological anatomy and immunohistochemistry revealed a fibroma of gonadal stromal origin.

Histopathological analysis made a diagnosis, although its clinical and radiological characteristics make it one of the differential diagnoses to consider in testicular tumors. Its characteristics, radiological and histopathological, allow for conservative management in clinical practice.

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Fibroma testicular de origen gonadal estromal en un varón adulto

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RESUMEN

Las masas paratesticulares son una entidad rara, por lo que su diagnóstico y tratamiento casi siempre dan lugar a dudas clínicas. Más allá de las dudas que surgen por el hecho de que estas masas sean poco comunes, siempre hay que descartar el proceso de malignidad de las mismas. Los tumores del cordón sexual son extremadamente raros. El fibroma testicular de origen estromal gonadal es un proceso proliferativo que puede desarrollarse en estructuras paratesticulares. El objetivo de nuestro estudio es presentar un reporte de un caso raro de fibroma testicular de origen del estroma gonadal así como el proceso diagnóstico bien documentado y el manejo terapéutico exitoso que se llevó a cabo posteriormente.

Presentamos el caso de un varón de 68 años que acude a consulta por el hallazgo casual de un nódulo en el testículo izquierdo con marcadores tumorales normales. La ecografía mostró una imagen nodular bien definida con una ecotextura difusamente homogénea; además era hipoecoico, vascularizado y demostraba hidrocele. La resonancia magnética reveló un tumor sólido con crecimiento extrínseco en el testículo izquierdo y el epidídimo, y la lesión era relativamente hiperintensa en la imagen potenciada en T1 e hipointensa en T2. Se realizó exéresis quirúrgica del tumor paratesticular e hidrocelectomía. La anatomía patológica y la inmunohistoquímica revelaron un fibroma de origen estromal gonadal.

El análisis histopatológico permitió establecer el diagnóstico, aunque sus características clínicas y radiológicas lo convierten en uno de los diagnósticos diferenciales a considerar en los tumores testiculares. Sus características, radiológicas e histopatológicas, permiten un manejo conservador en la práctica clínica.

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1. INTRODUCTION

Para-testicular tumors are uncommon; the most common benign para-testicular tumors are the adenomatoid and lipoma. Testicular fibroma is a benign, proliferative process of intratesticular and para-testicular structures; its most frequent location is in the scrotal tunics, especially in the tunica vaginalis, although it can also be found in the epididymis, tunica albuginea and the spermatic cord [1].

Fibromas of gonadal stromal origin are rare; only a few cases have been reported. They are analogous to similar tumors in the ovary. The tumor presents as a slow, unilaterally growing and sometimes painful mass, and is sometimes the casual finding of a testicular nodule - the most frequent symptom. Associated hydrocele is also frequent, but recurrences or metastases have never been reported in literature [2].

We detail an exhaustive case report on the clinical, immunohistochemical and radiological description of a para-testicular fibroma case associated to hydrocele. This lesion can lead to confusion among urologists because it often simulates a malignant testicular tumor and it is treated with aggressive or radical surgery. Therefore, it is important to know its characteristics. Our careful characterization from different, multidisciplinary foci can help clinicians in the recognition of and decision-making regarding this rare tumor.

2. CASE REPORT

A 68-year old male, with no medical history of interest, came in for a follow-up for lower urinary tract syndrome with elevated prostate specific antigen (PSA) (11,60 ng/ml) and a prostate biopsy four years ago (non-tumor findings in pathological review). The patient chose to come in for a consult because of a spontaneous finding of a mass around his left testicle.

On physical examination, both testicles were of normal size, morphology and structure, but there was a palpable, hard and firm nodular lesion in the upper pole of the left testicle that seemed extra-testicular.

The ultrasonographic examination (Figure 1) showed a nodular image measuring 16x12 mm, well-defined, near the left epididymis with a diffusely homogeneous echotexture, and which was hypoechoic and vascularized. In addition, a tiny hydrocele was observed in the left scrotal sac. Standard

blood and urine biochemical analyses tests and testicular tumor markers were within the standard limits: alpha-fetoprotein (4.1 UI/mL), b-human chorionic gonadotropin (1.98 mUI/mL) and lactic dehydrogenase-LDH (180 UI/L).

six months later.

A new ultrasonography was then performed, and it showed both slow and minimal tumor growth (1-2 mm greater than the previous control), with a moderately increased left

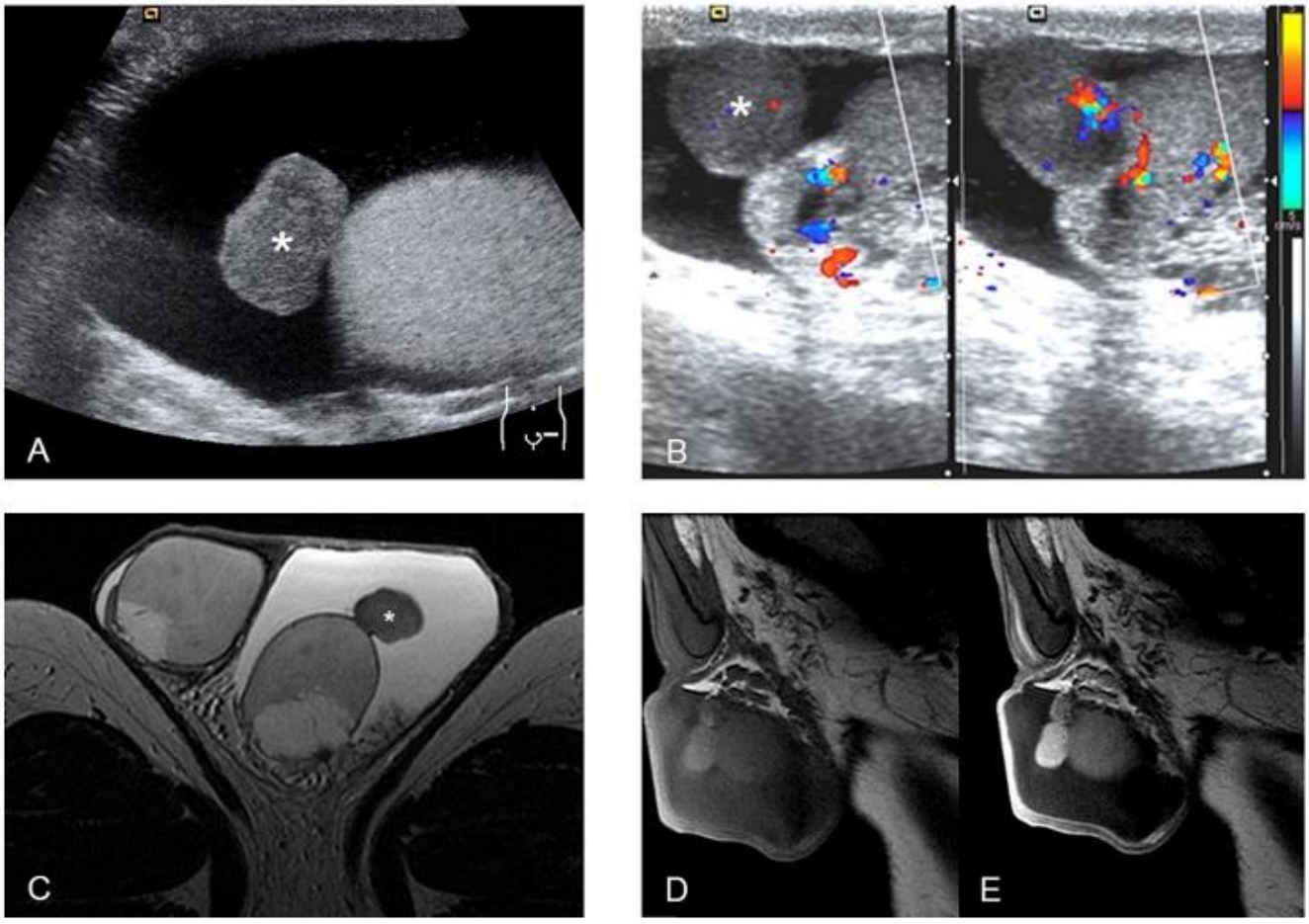

Figure 1*: 1A-1B: Ultrasound images show a nodular lesion, well defined, near the left epididymis, with homogenous and hypoechoic echotexture. Color Doppler images show marked internal vascularity of the tumor. 1C: Transverse, T2-weighted image shows a solid para-testicular tumor in contact with the tunica albuginea. The lesion appears with low signal on T2 weighted sequences. The image also demonstrates a bilateral tubular ectasia of rete testis. 1D-1E: Sagittal before and after contrast T1-weighted images present an intense, homogenous enhancement of the lesion.*

Because of these findings, it was decided that the best course of action would be to control the lesion and reevaluate it six months later. In the ultrasonogram, there were no changes regarding the size and morphology compared to the previous control. The radiologic study was extended with the request of a Magnetic Resonance Imaging (MRI). MRI (Figure 1) revealed a 19x16x13 mm solid tumor with extrinsic growth to the left testicle and epididymis, which also seemed to contact the tunica albuginea. The lesion was relatively hyperintense compared to the testicular parenchyma in the T1-weighted image and hypointense in T2, with intense homogeneous post-contrast enhancement. The differential diagnosis was oriented as a benign lesion. As a result, it was decided to control and repeat the medical imaging process

hydrocele that had started to become physically and esthetically uncomfortable.

Therefore, because of the increased hydrocele and the high suspicion of benignity, trans-scrotal surgical exploration was decided instead of an inguinal approach as in the malignant masses on which we practice. We performed a para-testicular tumor exeresis and hydrocele drainage without eversion of the tunica vaginalis. A malignant process was not suspected, but just in case, we tried to minimize any oncological risks by preserving anatomical spaces. The hydrocele's liquid was sent for cytological evaluation. Finally, the tunica vaginalis was sutured, closing the cavity around the testis. During the surgery, it was observed that the tumor was connected to the albuginea tunic

of the superior left testicle pole, and the resection was subsequently completed (Figure 2).

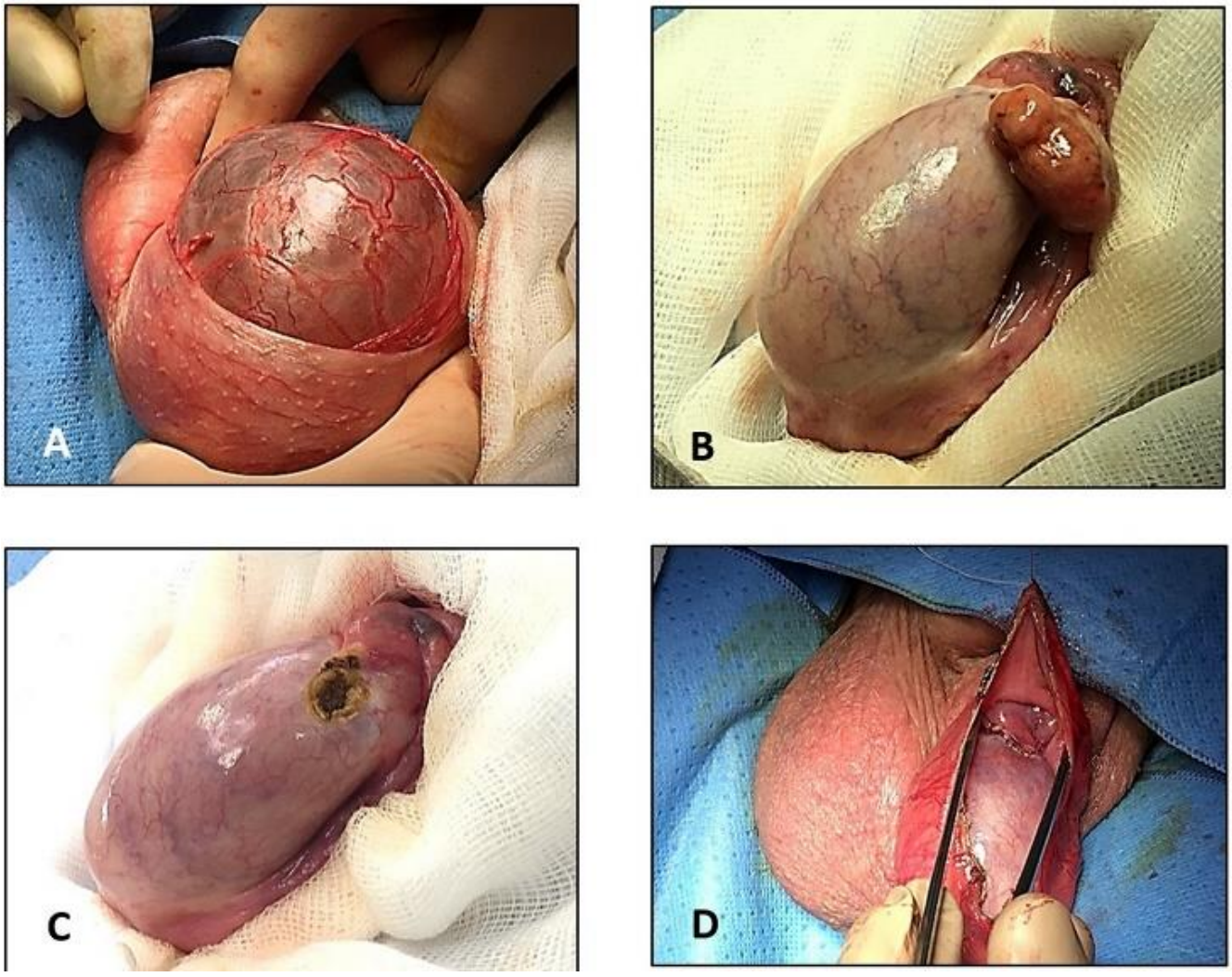

***Figure 2**: 2A: Transescrotal incision and exposure of voluminous hydrocele. 2B. Left testicle, in the upper pole, we can see a well-circumscribed mass of approximately 3 cm. 2C. Surgical site after conservative management. 2D. Left testicle after surgical excision of paratesticular mass and hydrocele drainage. Closure of tunica vaginalis.*

The pathological anatomy was reported as a testicular fibroma of gonadal stromal origin. It was a multi-nodular, 3x2 cm tumor, and its sections showed bundles of bland spindle-shaped cells without necrosis, alternating more densely cellular areas with other more lax and perivascular areas. The cells had minimal cytoplasm and oval nuclei with scant mitosis (Figure 3). Necrosis was not observed. The surgical limits presented a normal testicular parenchyma without histological lesions, and cytological analysis of the hydrocele was also negative for malign cells.

On immunohistochemical analysis, the tumor cells showed a diffuse expression of vimentin, actin, inhibin, calretinin, BCL2, WT1, CD56 and progesterone receptors. There was focal positivity for cytokeratins (AE1-AE3), RE, WT1 and desmin, and negativity for S100, CD31, EMA, CD34 and Caldesmon (Figure 3).

The post-operative period developed uneventful and at the most recent follow-up, 24 months following the surgery, the patient shows good status with neither evidence of tumor recurrence nor the hydrocele.

3. DISCUSSION

The para-testicular region is a complex anatomical zone that includes the spermatic cord, testicular layers, epididymis and vestigial remnants [1].

In this region, there is a great variety of tumors, the most frequent being lipoma, adenomatoid tumors, and leiomyoma [1, 3]. Para-testicular solid tumors are rare intra-scrotal masses, and, in general, are benign formations in more than 90% of cases [4].

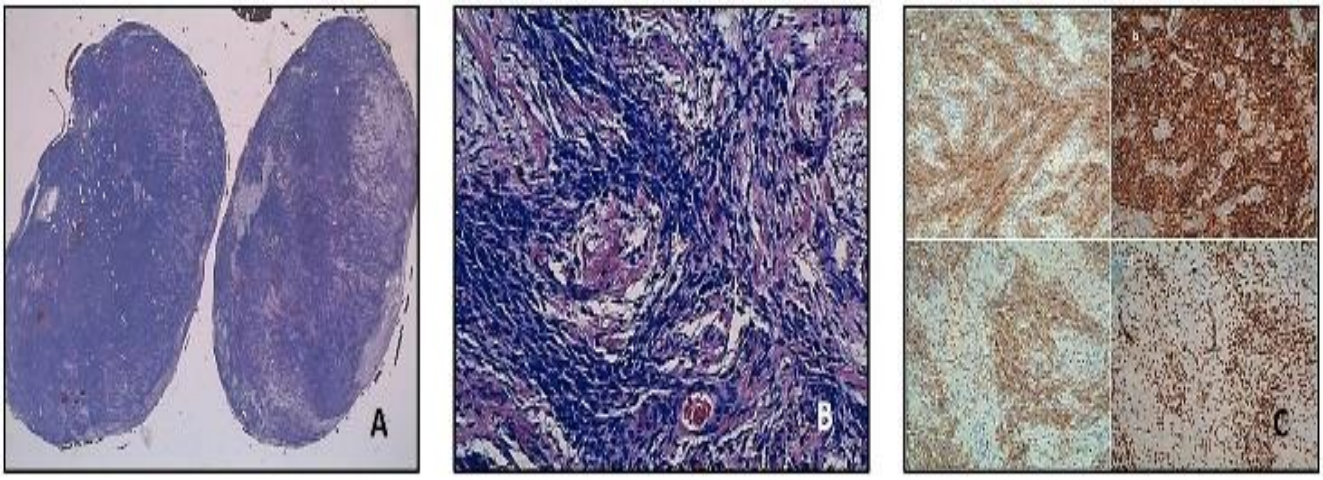

Figure 3*: 3A: Panoramic view of the tumor, partially covered by albuginea (HE 2x). 3B: At a higher magnification, a spindle cell tumor is observed, alternating loose and dense areas, with cells of sparse cytoplasm and oval and monomorphic nuclei (HE 20x). 3C: Immunohistochemical study. Expression of Calretinin (a), CD56 (b), Inhibin (c) and WT1 (d).*

Fibroma-thecoma is a type of sex cord and stroma tumor. Stromal cord tumors represent approximately 4% of testicular neoplasms [5]. They are tumors that contain Leydig cells, Sertoli, granulosa cells, or theca cells. Among them, tumors of the fibroma-thecoma group are very rare in the testicle; about 30 have been described, usually as isolated cases [6-10]. In this location, morphology and immunohistochemistry is similar to those of the ovary. The differentiation between fibroma and thecoma is not always evident, which is why some authors prefer a broader denomination of fibrothecoma [6] or tumors of the fibroma-thecoma group [11].

Most gonadal stromal fibromas have an intratesticular location, sometimes in contact with the albuginea. Our case has the peculiarity of having a para-testicular location without contact with the testicular parenchyma, but with contact with the albuginea and associated with a hydrocele. They are benign tumors with an age range between 5 and 69 years, with an average of 45 years and variable size, although they can reach 7.6 cm [6]. Very occasionally, a tumor with these characteristics has been dissected in a para-testicular location [12, 13].

Macroscopically, this type of tumor is a firm, well-circumscribed, rarely encapsulated nodule, with a gray-yellowish color, and a size of 0.8 to 7 cm in diameter, without hemorrhage or necrosis. Morphologically, it is characterized by spindle cells with scarce cytoplasm and oval nuclei arranged in irregular fascicles or in a storiform pattern, sometimes alternating more dense areas with others that are less dense. There are often mitoses, sometimes very frequently, but not atypia or necrosis [6]. In some cases, a minimum cordonal component has been demonstrated [7, 13].

The most classic immunohistochemical characteristics are the positivity for Inhibin and Calretinin, although it is possible that they express other, more constant markers such as Vimentin, CD56 and WT1. Other markers are expressed in a variable and focal way. These include estrogen and progesterone receptors, bcl2, CD34, Cytokeratin and MelanA [13].

Differential diagnosis should be made with other spindle cell tumors of testicular and para-testicular regions such as leiomyoma, schwannoma, neurofibroma, perineurioma or solitary fibrous tumor. A complete immunohistochemical profile may be necessary in these cases [7]. Some malignant para-testicular tumors, such as fibrosarcoma, rhabdomyosarcoma, leiomyosarcoma or liposarcoma, or malignant fibrous histiocytoma and stromal tumors or metastatic carcinomas, should also be considered [14].

In addition, other tumors of the sex cord-stromal group should be taken into account, such as Sertoli cell tumor or an undifferentiated cord tumor. A histochemical technique of examining reticulin and the pattern of cytokeratins may be helpful on this account. On the other hand, the myoid gonadal stromal tumor expresses markers of smooth muscle and S100 in an important way [13].

Testicular fibromas are reported in male patients who have unremarkable clinical features, no hormonal symptoms, and normal testicular tumor markers (a-fetoprotein and b-human chorionic gonadotropin). The most frequent symptom is unilateral scrotal swelling [8] and some cases are associated with a hydrocele, as in the reported case. It is of relevance that the hydrocele did not relapse, even without the tunica vaginalis' eversion, suggesting that it was generated by the para-testicular mass.

In our case, the patient had no pain in the testicle, no increase

in tumor markers nor any pathological findings in blood and urine analyses, which is the same as other cases found in the literature [7, 9].

Ultrasonographic examinations reported the presence of a hypoechoic or hyperechoic solid mass, depending on the quantity of fibrous tissue. On Doppler ultrasonography there was a characteristic increased vascularity, which could be confused with a malignant tumor. MR imaging allows for precise localization of scrotal masses and defines the anatomic relationship to the surrounding structures [15], like in our case with the tunica albuginea. In the MRI, testicular fibroma usually shows low intensity signals in T-1 and T-2 weighted images, due to the fibrous tissue content.

Fibromas of gonadal stromal origin occur in the testis, but some are adjacent to the tunica albuginea and may be confused in radiological images with fibromas originating from the testicular tunics. This being the case, clinical and radiological differential diagnosis includes the fibrous pseudotumor, which is associated more frequently with hydrocele. Some studies [16] suggest that fibrous pseudo-tumors might belong to IgG4-related diseases, which includes such diverse entities as retroperitoneal fibrosis, sclerosing pancreatitis and cholangitis, Riedel's thyroiditis, or sclerosing sialadenitis.

Tumors of scrotal structures are not common, and surgical exploration is usually required to rule out malignant processes. Thus, the role of an intra-operative frozen section may be important in cases that spur doubts. Awareness of both urologists and pathologists of para-testicular fibroma is useful because it may prevent the performance of an unnecessary radical orchiectomy, especially in young patients [14]. In addition, the treatment will depend on the lesion location; only in isolated cases can the lesion be removed without radical orchiectomy, as in our case, where radiological suspicious and a macroscopic view led us to a conservative surgical management.

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5. CONFLICT OF INTERESTS

The authors have no conflict of interest to declare. The authors declared that this study has received no financial support.

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