

# Management of Scrotal Aggressive Angiomyxoma: A Case Report

R LAKSHMANA<sup>1</sup>, JAYAKUMAR<sup>2</sup>, N SNIGDHA<sup>3</sup>, NEETHISH K PAUL<sup>4</sup>, PA SETHU RAMAN<sup>5</sup>

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## ABSTRACT

Aggressive Angiomyxoma is most commonly identified as a slow growing mesenchymal neoplasm most commonly seen in women than in men. It has a marked tendency for local recurrence but with low instances of metastasis. Radiological investigations may assist in arriving at a provisional diagnosis and provide an arbitrary idea about the margins of the tumour but the gold standard diagnosis is always be histopathological examination. Surgery is the first line and the most reliable form of treatment. However, around 40-70% of patients came with complaints of recurrence. This is a case of a 61-year-old male patient who came with complaints of scrotal swelling which increased in size over a period of six months. The swelling was firm in consistency without warmth or tenderness. Contrast-enhanced Magnetic Resonance Imaging (CE-MRI) was taken to discern the borders of the mass and it was suspected to be a soft tissue sarcoma. Mass was excised in toto and a confirmatory diagnosis of aggressive angiomyxoma was made only after postoperative histopathological examination. The patient was on regular follow-up for 8 months to date and no evidence of local recurrence was detected. This case report presents to be a curious case as aggressive angiomyxomas are usually tumours seen in females. It also describes the management of scrotal swelling without a preoperative diagnosis.

**Keywords:** Histopathological examination, Mesenchymal neoplasm, Metastasis, Recurrence

## CASE REPORT

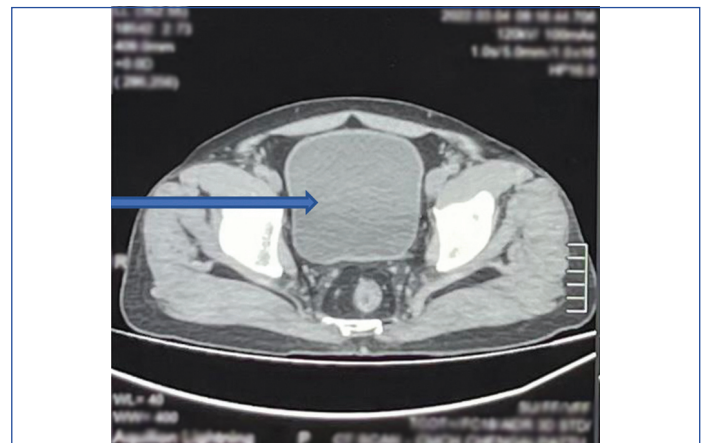
A 61-year-old male came with complaints of scrotal swelling for six months. There was a gradual increase in size, without complaints of pain. The patient neither had complaints of fever, abdominal pain, altered bowel or bladder habits, loss of weight, loss of appetite, chest pain, or breathlessness. The patient had no known comorbidities and had no history of any previous surgery.

On general examination, the patient had no pallor, no icterus, no pedal oedema. Vitals were stable. Systemic examination was normal. On local examination, a 6×5 cm in size, cystic, non tender, non transilluminant, irreducible swelling was palpated in the Bilateral (B/L) scrotum extending into the perineum. The skin over the swelling had minimal rugosities. No cough impulse [Table/Fig-1]. B/L testes were palpated separately. Penis was in midline.



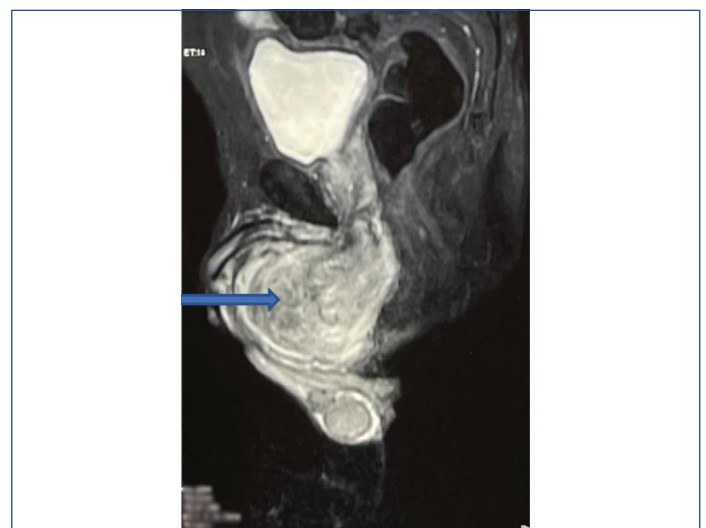
[Table/Fig-1]: Swelling on examination.

Routine blood investigations were done. Tumour markers like Alpha-fetoprotein (AFP), Carcinoembryonic Antigen (CEA), Cancer Antigen (CA) 19-9, CA242, and CA125 were negative. Initial radiological evaluation was done at another center. Ultrasonogram (USG) Scrotum showed it to be an extra testicular mass. No other findings were described through the ultrasound. Computed Tomography (CT) abdomen was done to further confirm the swelling to be extratesticular. It showed a heterogeneously enhancing extratesticular mass of 9.4×5.6 cm in the right scrotum [Table/Fig-2].



[Table/Fig-2]: CT Image showing the extratesticular mass (Blue arrow).

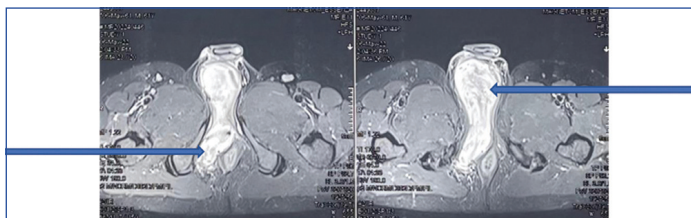
MRI showed a 14.5×7.8×5.1 cm hyperintense, T1 iso intense partially fat suppressible lesion noted in the right ischio-rectal fossa extending inferior to the root of penis and displacing the penis left laterally entering the root of the scrotum [Table/Fig-3].



[Table/Fig-3]: MRI Image showing the hyperintense mass (Blue arrow).

Due to financial constraints on the patient's part, a repeat of the previous investigations was avoided. To get a better orientation regarding the origin and the structures around the tumour a CE-MRI was done at the tertiary care centre.

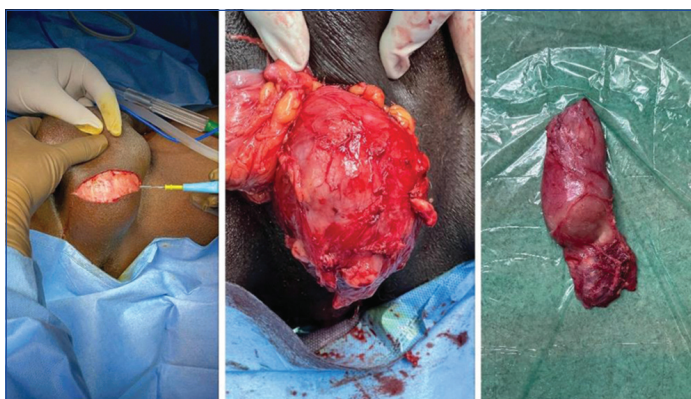
The CE-MRI showed a right perianal/perineal mass lesion of size 13.7×8.3×5.05 cm (AP×CC× TR), hyperintense, noted in the right ischioanal fossa and ischioanal fossa extending anteriorly along the perineum into the posterior border of the scrotum. It was seen abutting the anterior wall of the anal canal, the root of the penis, and scrotum without any evidence of local infiltration. Suspected to be a myxoid/spindle cell neoplasm [Table/Fig-4].



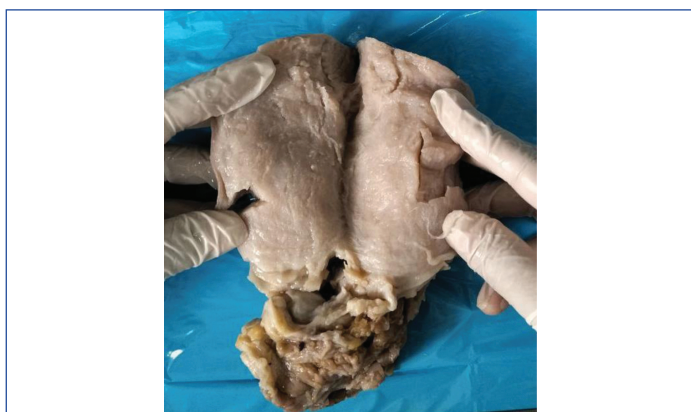
[Table/Fig-4]: CE-MRI Image showing the origin of the mass.

To rule out pelvic herniation before proceeding with surgical excision of the tumour, sigmoidoscopy and cystoscopy were done and it was confirmed that bowel or bladder were not the contents.

The patient was taken up for surgery and through a trans-scrotal incision, the mass was excised in toto and sent for histopathological examination [Table/Fig-5]. Gross pathology showed a soft tissue mass of size 13×7×3 cm with a grey-white external surface. The cut surface was grey white, homogeneous, soft, gelatinous, solid area. No necrosis or haemorrhage was noted [Table/Fig-6].



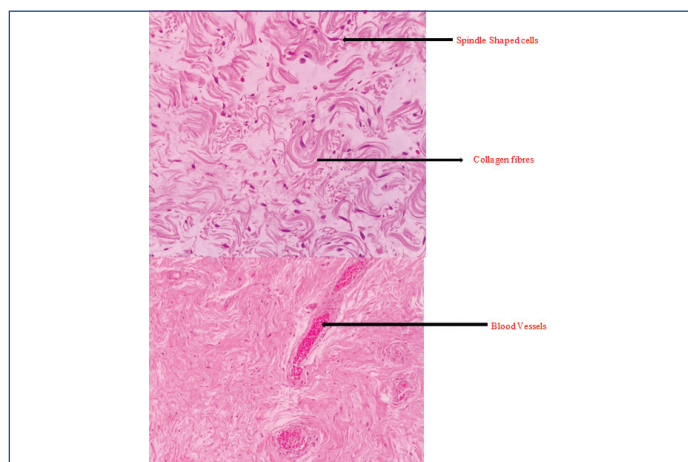
[Table/Fig-5]: Intraoperative pictures showing incision, intraoperative mass and postoperative mass.



[Table/Fig-6]: Gross specimen under examination.

Microscopy showed an encapsulated infiltrative hypocellular lesion composed of spindle-shaped tumour cells with delicate cytoplasmic processes and bland nuclear chromatin. No evidence of atypia or increase in mitosis was detected. The tumour cells were embedded in a myxoid stroma with scattered delicate collagen fibers. Focal areas showed entrapped mature adipose

tissue within the stroma. Numerous small and medium-sized thick walled hyalinised blood vessels were also seen. Attached skeletal muscle bundle also was noted [Table/Fig-7].



[Table/Fig-7]: Histopathological picture showing encapsulated infiltrative hypocellular lesion composed of spindle-shaped tumour cells (40X, Haematoxylin & Eosin).

Based on the above microscopic findings, a diagnosis of aggressive angiomyxoma was made amongst other differential diagnoses such as superficial angiomyxoma, angiomyofibroblastoma, myxoid neurofibroma, myxoid smooth muscle tumour and myxoid liposarcoma. Aggressive angiomyxoma is an unencapsulated, hypocellular and a locally infiltrative tumour. Stellate to spindle cells with small capillary sized blood vessels with a myxoid stroma and abundant thin and thick walled blood vessels are seen. Superficial angiomyxomas were ruled out as they mostly composed of epidermoid or squamoid elements are evident. Angiomyofibroblastomas on the other hand show epithelioid to plasmacytoid tumour cells. Myxoid neurofibromas portray a characteristic feature of buckled nuclei. Typical cigar-shaped nuclei are seen with eosinophilic cytoplasm in myxoid smooth muscle tumours. Lipoblasts, plexiform vasculature with less differentiated round cell component are key features of Myxoid liposarcomas. Each of these tumours has specific microscopic features that differentiate them from each other despite their similar clinical features and gross features. Spindle-shaped tumour cells with delicate cytoplasmic processes with evidence of infiltrative nature are the key features of aggressive angiomyxoma that differentiate it from the rest of the differential diagnoses.

The patient was discharged six days after surgery. The patient is on follow-up for the past eight months and is asymptomatic. The wound site is healthy and no local swelling was palpated on examination. An ultrasound of the scrotal region is planned after one year of surgery to rule out recurrence. No specific medications have been given.

## DISCUSSION

Aggressive angiomyxoma is a rare benign mesenchymal tumour most commonly occurring in the pelvic soft tissues and perineum of women more often than in men [1-5]. Aggressive angiomyxoma was first described by Steeper TA and Rosai J in 1983 [6]. The term "aggressive angiomyxoma" was chosen for this neoplasm to emphasise the neoplastic nature of the blood vessels and its locally infiltrative and recurrent nature [6]. Aggressive angiomyxoma shows female predominance, as less than 100 male cases have been reported [1,3,5-7]. In men, aggressive angiomyxoma usually arises from the pelviperineal tissue mostly involving scrotum and sometimes involving the bladder. The tumour originates most commonly from the scrotum (42%), pelvis (11%), perineum (10%) and rarely from the groin (9%), spermatic cord (5%) and prostate (4%) [1,2,7]. Tabulation was made comparing the key features of a case such as this with others across the world [Table/Fig-8] [1-4].

Variables	This case report	Sabbagh AJ et al., [1]	Sun, Juan et al., [2]	Nyandwi L et al., [4]	Law YX et al., [3]
Place of study	SRM University, Tamil Nadu, India	Aleppo University, Syria	Peking Union Medical College, China	National Hospital of Niamey, Niamey, Niger	National University Hospital, National University Health System, Singapore
Age/Sex	61-year-old male.	72-year-old male	2 patients. 45-year-old male. 64-year-old male.	10-month-old infant boy.	38-year-old male
Presenting complaints	Painless swelling in B/L scrotum for 6 months with a gradual increase in size.	Painless swelling in the right testicle. No other symptoms.	1-year history of a growing left scrotal mass. No pain. Enlarged right scrotum.	Swelling detected during neonatal period with gradual increase in size.	Swelling in right side of scrotum enlarged over a year.
Findings on examination	A 6x5 cm size, cystic, non tender, non transilluminant, irreducible swelling was palpated in B/L scrotum extending into the perineum. Skin over the swelling had minimal rugosities. No cough impulse. B/L testes were palpated separately.	Hard, moving, painless mass above the right testicle.	The mass had a clear boundary, moderate mobility, and normal overlying skin. The light transmittance test was negative. Enlarged right scrotum, nearly 10 cm in diameter with no pain or tenderness, and the scrotum did not disappear while lying flat. The light transmittance test was negative.	Painless, spherical, soft, regular in shape and 10cm in diameter, with some collateral venous circulation. A mass effect on the penis and perineum could be observed. There was no transillumination. The right testicle was not found, and the left testicle was in the testicular bursa and appeared to be healthy.	Right testis felt firm and enlarged measuring 10x6.5 cm. Non tender.
Investigations : Blood Work - Radiology -	Routine blood investigations normal. Alpha fetoprotein, carcinoembryogenic antigen, Cancer Antigen (CA) 19-9, CA242, CA125 were negative.	Biochemical and hormonal tests normal. Beta-human chorionic gonadotrophins (B-HCG), alpha fetoprotein and Prostate Specific Antigen (PSA) under normal limits.	Routine blood parameters within normal limits. Alpha fetoprotein, Carcinoembryonic antigen, Cancer Antigen (CA) 19-9, CA242, CA125, and CA72-4 were negative. Routine blood parameters within normal limits.	Alpha fetoprotein and Beta human chorionic gonadotropin levels were within normal limits.	Lactate dehydrogenase, alpha-fetoprotein, and beta-human chorionic gonadotropin levels were not elevated.
	Ultrasonogram (USG) Scrotum showed it to be an extratesticular mass. Computed Tomography (CT) It showed a Heterogeneously enhancing extratesticular mass 9.4x5.6 cm in the right scrotum. Magnetic Resonance Imaging (MRI) showed a 14.5x7.8x5.1 cm hyperintense, T1 iso intense partially fat suppressible lesion noted in the right ischiorectal fossa extending inferior to root of penis and displacing the penis left laterally entering the root of scrotum. Contrast Enhanced MRI showed Right perianal/perineal mass lesion of size 13.7x8.3x5.05 cm (APxCCxTR), hyperintense noted in the right ischiorectal fossa and ischioanal fossa extending anteriorly along the perineum into the posterior border of the scrotum. It was seen abutting anterior wall of anal canal, root of penis and scrotum without any evidence of local infiltration. Suspected to be a myxoid/spindle cell neoplasm	A scrotal ultrasound showed a well-defined, oval, hypoechoic, homogeneous tissue above the right testis measuring 6.1x4.5 cm with internal vascularity. It squeezes the medial part of the right epididymis head	The US of the scrotum showed that a 1.6x1.2x1.0 cm area of clear boundary and low echo could be seen on the left spermatic cord with a meager blood flow signal on colour Doppler flow imaging (CDFI). The US of the scrotum revealed that a gelatinous uneven echo could be seen at the right spermatic cord and the upper part of the scrotum, and Colour Doppler Flow Imaging CDFI showed negligible blood flow signal. MRI showed an elliptical long T1 and long T2 signal mass in the right scrotum, with a clear boundary and size of about 9.6x6.9x6.2 cm, which presented as an iso-height signal on the diffusion weighted imaging sequence and small spots of enhancement on the enhanced scan	Ultrasound examination revealed a large structural mass, with the heterogeneous echo measuring 10x7x6 cm. The two testes were upwardly displaced by the mass. Abdominopelvic Computed Tomography (CT) demonstrated a large intrascrotal tissue mass communicating with the abdomen with no visible air component, leading to the decision that the mass was probably an inguinal mesenteric hernia.	Ultrasound (US) scrotum revealed a large heterogeneous solid mass in the right scrotum. There was internal vascularity. Although the mass appeared distinct from the right testis at some areas, the invasion could not be entirely excluded.
Preoperative suspicion of diagnosis	Suspected to be a spindle cell/myxoid neoplasm based on the MRI findings.	No specific pre operative diagnosis.	Suspected to be lipoma of spermatic cord. No specific preoperative diagnosis.	Suspected to be a testicular tumour on clinical examination. Suspected to be an inguinal hernia.	Suspected to be testicular carcinoma.
Treatment	Complete excision of the tumour through a trans-scrotal incision.	Radical orchidectomy.	Tumour Resection. Intraoperative frozen section showed that the tumour originated from mesenchymal tissue. Scrotal mass resection	Two combined surgical incision approaches were adopted, the right inguinal approach helped locate the healthy right testicle and the scrotal route was for wide excision.	Right radical orchidectomy through an inguinal approach.
Postoperative Period	Diagnosis made based on Histopathological Examination (HPE).	Diagnosis made based on HPE.	Diagnosis made based on HPE both cases.	Diagnosis made based on HPE.	

	Soft tissue mass of size 13x7x3 cm with a grey white external surface. Cut surface was grey white, homogeneous, soft, gelatinous, solid area at one end. No necrosis/haemorrhage.	Cut section showed para-testicular well-defined mass measuring 6.5 cm with a greater diameter.	Not described in the article	The lump had two different appearances: one portion had the appearance of a typical solid tumour and the other showed mixed tumour growth.	The tumour was paratesticular. No necrotic or haemorrhagic areas. It was well circumscribed enveloped by an intact rim of fibrous capsule.
HPE: Gross features -Microscopic features-	Unencapsulated, hypo cellular lesion, lesion composed of spindle to stellate shaped tumour cells with delicate cytoplasmic process. Delicate collagen fibers. Focal areas show entrapped mature adipose tissue within stroma. Many small & thick walled blood vessels. Attached skeletal muscle bundle.	Poorly circumscribed infiltrative tumour made up of bland, spindle, and stellate cells with delicate cytoplasmic processes that surrounded blood vessels, on the myxoid stroma. There were no atypia or mitotic figures.		Histological appearance of a benign fibroblastic tumour, which was initially suggestive of angiomyxoma	Hypocellular, containing abundant vessels and stromal cells in a background of collagenous stroma. The vessels were of varying caliber. There was no evidence of cytological atypia, pleomorphism, or increased mitotic activity in cellular areas.
Immuno-histochemistry		ER, PR positive Calretinin, CD34, desmin and S100 negative.	CD 31 (+), CD 34 (+), S-100 (+), BCL-2 (-), SMA (-), and Ki-67 (index 6%)	CD 34 (+)	Weak positive for S100 and Oestrogen Receptor (ER) protein. Moderate expression for desmin.
Follow-up and Recurrence	On follow-up for 7 months. No signs of recurrence detected.	On follow-up for 2 years. No signs of recurrence determined by MRI	Both patients on follow-up for 6 years. No signs of recurrence.	On follow-up for 6 months. No signs of recurrence.	On follow-up for 12 months. no signs of recurrence.

**[Table/Fig-8]:** Comparison of this case report with prominent studies [1-4].

The clinical appearance of aggressive angiomyxoma differs according to the site and size of the lesion but usually presents as painless swelling over a stated time in the scrotum.

Misdiagnoses have been recognised as testicular neoplasm, inguinal hernia, hydrocele, or spermatocele [1,8]. Aggressive angiomyxoma in the prostate causes lower urinary symptoms, while in other regions presents with the symptoms of mass that compress the neighbouring organs. Physical examination in most cases reveals a firm, non tender, and non transilluminated mass [1,2,9]. USG is usually the first choice for screening pelvic lesions before performing further imaging techniques. Aggressive angiomyxoma lesions were found to be irregular, heterogeneous and hypoechoic masses with a large region of involvement and relatively well-defined margins. The lesions had intermediate to high echogenicity with a layered or swirled arrangement. The testicles and epididymis commonly demonstrated normal echogenicity and vascularity [1-3,9-11]. A finger-like growth pattern on the ultrasonic imaging indicated aggressive growth into surrounding tissues [9].

MRI provides a better insight in identifying the characteristics of such mesenchymal tumours, including isointense or hypointense characteristics on T1-weighted images and hyperintense characteristics on T2-weighted images. The swirled sign observed on both USG and MRI has also been mentioned in a recent study [1,3,10,11]. Typical CT features are a mass hypodense or isodense to muscle with well-defined margins that show variable enhancement on administration of intravenous contrast. Considering that the appearance of the tumour is unspecific on a CT, the suspected diagnosis must be based again on the characteristic morphology and localisation of the mass, and should be complemented with MRI. Angiography shows a hypo attenuating mass. the tumour tends to grow around the structures of the pelvic floor without penetrating the muscles. They can increase in size, infiltrate the surrounding tissue but do not show the tendency to metastasise [12].

In most cases, the diagnosis of this tumour was made when here was a local recurrence or when an intraoperative frozen section was sent for histopathological examination. In addition, none of these radiologic investigations can discriminate aggressive angiomyxoma from different malignancies, including sarcomas [1,9,10].

Hence, although detailed radiological investigations such as USG, CT and MRI assist in delineating the margins of the tumour, preoperative diagnosis of aggressive angiomyxoma can be extremely challenging [1-3,7,10,12,13]. Wide surgical excision is the treatment of choice, including broad free margins around the tumour in the area of

resection. This can be tricky because the consistency of the tumour tissue the same as the adjacent tissue [1,9,14]. To avoid incomplete resection of the tumour, a thorough radiological investigation is important, which was performed in this case.

Gross sections of the postoperative specimens of these tumours are soft, partly circumscribed with a gelatinous appearance on the cut section. Microscopically, aggressive angiomyxomas consist of mesenchymal uniform spindle-shaped or stellate cells embedded in a loose myxoid and collagenous stroma with abundant fibroblasts, myofibroblasts and variably sized vessels. Angiomyxomas show low mitotic activity with an absence of nuclear atypia, which corresponds with our histology [1-3,6,10]. The most decisive immunohistochemistry profile, in reference to various studies, is the positivity of Vimentin and Cluster of Differentiation 34 (CD34) with the negativity of S-100. Mammary Serum Antigen (MSA), Smooth Muscle Actin (SMA), and Desmin are of medium sensitivity and specificity because they reported positivity in about 50% of the cases. Smooth muscle actin highlights myoid bundles and was found to be positive in individual tumour cells [2-4]. Stunningly, Estrogen Receptor (ER) and Progesterone Receptor (PR) receptors reacted positively in 50% of cases (n=32), even when the tumour was in men [2,15]. One or both of these hormone receptors displays strong nuclear positivity in most of the tumour cells. This proposes a hormonal effect on the tumour growth and may arise from specialised cells of the stroma of the perineum. Thus, hormonal therapy might be useful as an adjuvant treatment [1,15]. Labeling for Ki-67 consistently demonstrates a low proliferative index (<1% of tumour cells) [8]. Chromosomal translocation of the 12q13-15 band involving the HMGA2 gene has been described [15].

Aggressive angiomyxomas should be distinguished from other benign tumours affecting the pelvis and the genital tract, such as intramuscular myxoma, myxoid neurofibroma, myxoid or spindle cell lipoma, superficial angiomyxoma, angiomyofibroblastoma, and angiomyolipoma. One should also consider some malignant tumours with myxoid stroma, such as myxoid liposarcoma, myxoid malignant fibrous histiocytoma, and embryonal rhabdomyosarcoma [2-4,10].

Since, the consistency of an aggressive angiomyxoma is similar to that of local tissue, a gross margin clearance during surgery is difficult to achieve which in turn leads to a high propensity for local recurrence. A strict follow-up should be maintained to detect local recurrence in the early stages. Studies have referred to a high local recurrence rate between 35% and 72%, but no metastasis has been

reported to date in men [1,3,10,16]. Since metastasis is exceedingly rare, the prognosis is considered good [15].

Follow-up duration ranged from 1 to 144 months and MRI is the preferred method for detecting recurrences [11]. Ultrasound can also be applied for follow-up of patients after surgery, considering financial constraints [1,9].

However, most recent literature review revealed no differences in recurrence between the patients with negative margins and those with positive margins [15]. These results may be explained by the locally infiltrative nature of aggressive angiomyxoma. As this tumour may express ER and PR receptors, hormonal neoadjuvant or adjuvant therapy have been attempted in woman with aggressive angiomyxomas and a decrease in tumour size was observed in several reported cases [1,17]. These results establish a future type of therapy for such benign tumours. Four cases in men were reported using hormonal therapy with Gonadotropin-releasing Hormone (GnRH) agonist either as adjuvant therapy to prevent recurrence [10,18] or to manage recurrence before proceeding with a repeat surgery [19,20]. Additionally, many studies have showed promising results of using a single monthly 3.75 mg injection of leuprolide acetate, an agonist of (GnRH), for treatment of these lesions [1,13].

Vascular embolisation can diminish tumours by blocking their blood supply. It can also reduce intraoperative bleeding and operation risk. But, the tumour blood supply may come from multiple arteries, so the rapid development of alternative blood supply in tumour after embolisation may lead to the recurrence. Therefore, it is difficult to completely destroy tumours by embolisation alone, which is usually used for surgical adjuvant therapy [3].

## CONCLUSION(S)

This case report gives a broad perspective on the occurrence, presentation and the preferred treatment plan for a patient with testicular tumours without a clear pre operative diagnosis. Though resection of the tumour is the preferred mode of treatment, personalised treatment strategies assist in achieving a good clearance margin. Resection of the tumour with a good clearance margin proved to be an important step in the treatment in this case even though the diagnosis of the tumour was not known. Patient should be explained about high chances of local recurrence and the importance of a regular follow-up to detect any local recurrence at the earliest should be stressed upon.

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### PARTICULARS OF CONTRIBUTORS:

1. Professor, Department of General Surgery, SRM Medical College and Research Institute, Chennai, Tamil Nadu, India.
2. Professor, Department of Surgical Oncology, SRM Medical College and Research Institute, Chennai, Tamil Nadu, India.
3. Post Graduate, Department of General Surgery, SRM Medical College and Research Institute, Chennai, Tamil Nadu, India.
4. Assistant Professor, Department of General Surgery, SRM Medical College and Research Institute, Chennai, Tamil Nadu, India.
5. Senior Resident, Department of General Surgery, SRM Medical College and Research Institute, Chennai, Tamil Nadu, India.

### NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:

Neethish K Paul,  
Assistant Professor, Department of General Surgery, SRM Medical College and Research Institute, SRM Nagar, Potheri Village, Kattankulathur-603203, Chennai, Tamil Nadu, India.  
E-mail: neethisp@srmist.edu.in

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