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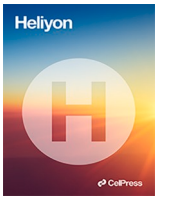

Case report

# Comprehensive treatment of primary pelvic synovial sarcoma: A 28-month follow-up case report and review of the literature

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ABSTRACT

*Background:* Primary synovial sarcoma originating in the pelvis is an extremely rare malignancy, and only a few cases have been reported. Usually, the tumor is detected at an advanced stage, making treatment more difficult. Lacking high-quality research, there are no consensus guidelines for the systemic treatment of the disease.
*Case presentation:* We admitted a 32-year-old male patient with an MRI suggestive of an 8.2 × 7.7 x 8.9 cm mass in the pelvis with bleeding. After a comprehensive evaluation, the patient underwent surgery, and postoperative pathology suggested biphasic synovial sarcoma. Once the diagnosis was clear, the patient was treated with chemotherapy, radiotherapy, and targeted therapy. Unfortunately, the patient died due to a recurrence of pelvic synovial sarcoma with multiple metastases throughout the body, with a survival period of 28 months.
*Conclusion:* Synovial sarcoma, a highly malignant soft tissue tumor, and primary intrapelvic synovial sarcoma are even rarer, with a poor prognosis. Ultrasound, Computed Tomography, and MRI can help doctors detect the tumor at an early stage and treat it aggressively, especially with surgical treatment, which can effectively improve the survival rate. Combined with the course of diagnosis and treatment of this case, it is possible to deepen the understanding of primary pelvic synovial sarcoma and discuss appropriate treatment strategies for this rare disease.

## 1. Background

Synovial sarcoma (SS) is a relatively rare soft tissue sarcoma commonly found in the joints of the extremities and rarely in other areas. In general, the larger the tumor size is, the higher the risk of metastasis and recurrence will be, and tumors less than 5 cm in diameter have a good prognosis [1,2]. The aim of this study was to investigate the clinical, imaging, and histological features and treatment of this rare tumor. Here, we report a case of primary pelvic synovial sarcoma.

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## 2. Case presentation

A 32-year-old male patient was admitted to our hospital with difficulty urinating and lower abdominal and perianal pain for three days. The patient had no symptoms of urinary frequency, urgency, hemorrhagia, or fever, and no other management was performed during the onset. The patient had mild pressure pain in the lower abdomen, and no abnormalities in the reproductive system or in the external examination of the anus. Based on clinical and empirical judgment, we believe that the patient may have acute prostatitis. After admission, the patient completed blood tests, except for WBC (+) in urine; inflammatory indicators such as WBC and CRP in his blood were normal; and tumor markers such as PSA, CEA, and AFP were not abnormal. Ultrasound of the abdomen indicates a mass in the lower posterior bladder. Magnetic Resonance Imaging (MRI) suggested an intrapelvic cavity mass, approximately 8.2× 7.7 x 8.9 cm in size, with high, equal, and low mixed signals, locally visible cystic foci, and fluid planes, suggesting a tumorous lesion with possible hemorrhage and cystic changes. The border between the mass and the prostate is not smooth; there are obvious compressed changes of the bladder and bilateral seminal vesicle glands, the pelvic bowel structural disorder, and there are no visibly enlarged pelvic lymph nodes (Fig. 1A–D). The pathological findings of the transrectal ultrasound-guided puncture biopsy of pelvic tumor suggested a soft tissue spindle cell tumor. The patient was diagnosed with a pelvic tumor and was treated with surgery after multiple metastases were ruled out by computed tomography of the head, thorax, and abdomen. Intraoperatively, a tiny amount of dark red bloody fluid was seen in the pelvis, and a solid cystic mass with a pseudo-envelope behind the bladder was observed. The pseudo-capsule adheres to the bladder, small intestine, rectum and pelvic tissue. A sinus duct of approximately 1x1 cm in size was seen on the upper surface inside the mass, and dark red blood and blood clots were spilling out. Removing the blood clots from the sinus duct opening, a large amount of bloody fluid, approximately 110 ml, was seen leaking out, and the tumor was removed entirely. Post-operative pathology suggested pelvic synovial sarcoma, biphasic type (Fig. 2A–C); immunohistochemistry showed partial positivity for CK, weak positivity for CD99, and strong positivity for Bcl-2 (Fig. 2D–F). Fluorescence in situ hybridization (FISH) diagnosis: SYT signaling was distributed in dots, and SYT gene rearrangement was positive (Fig. 2G–H). The patient started 6 consecutive cycles of chemotherapy (ifosfamide 3 g + epirubicin 100 mg) 1 month after surgery, during which no tumor recurrence occurred. Follow-up continued after chemotherapy, and patients found pelvic tumor recurrence with peripheral lymph node metastases at follow-up at the 8th month follow-up. Tumor recurrence severely undermines the patient's confidence in continuing to receive chemotherapy. Even though other more appropriate treatment strategies were presented, the patient decided to choose radiotherapy.

After imaging to assess the absence of systemic metastases, the patient received a radiotherapy regimen with Dt: 6000cGy/30Fx/6W. Pelvic magnetic resonance examination at the 1st, 2nd, and 3rd months after radiotherapy showed that the tumor and peripheral lymph node metastases were significantly reduced, and the efficacy evaluation was partial response(PR), PR, and stable disease(SD), respectively. Unfortunately, the patient's chest and abdomen computed tomography at the 5th month revealed that the tumor had developed multiple metastases in the lungs and abdominal cavity. Due to disease progression, the patient received 3 cycles of

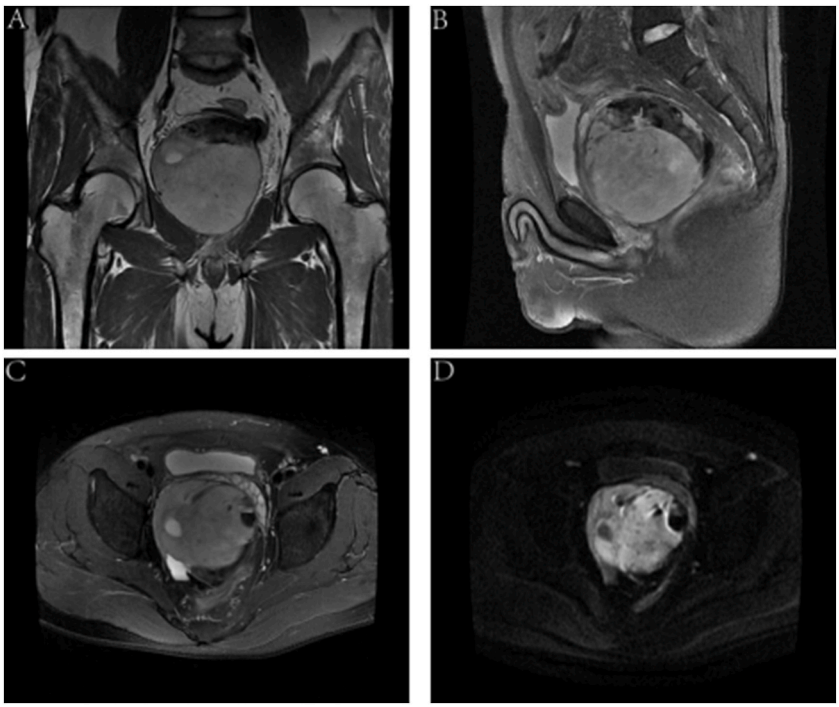


**Fig. 1.** (A–C) Intrapelvic masses were observed in coronal, sagittal, and transverse views on T1 MRI sequences. (D) The coexistence of high, medium, and low signals on the magnetic resonance-diffusion weighted imaging (MR-DWI) sequence, namely, the "triple signal sign."

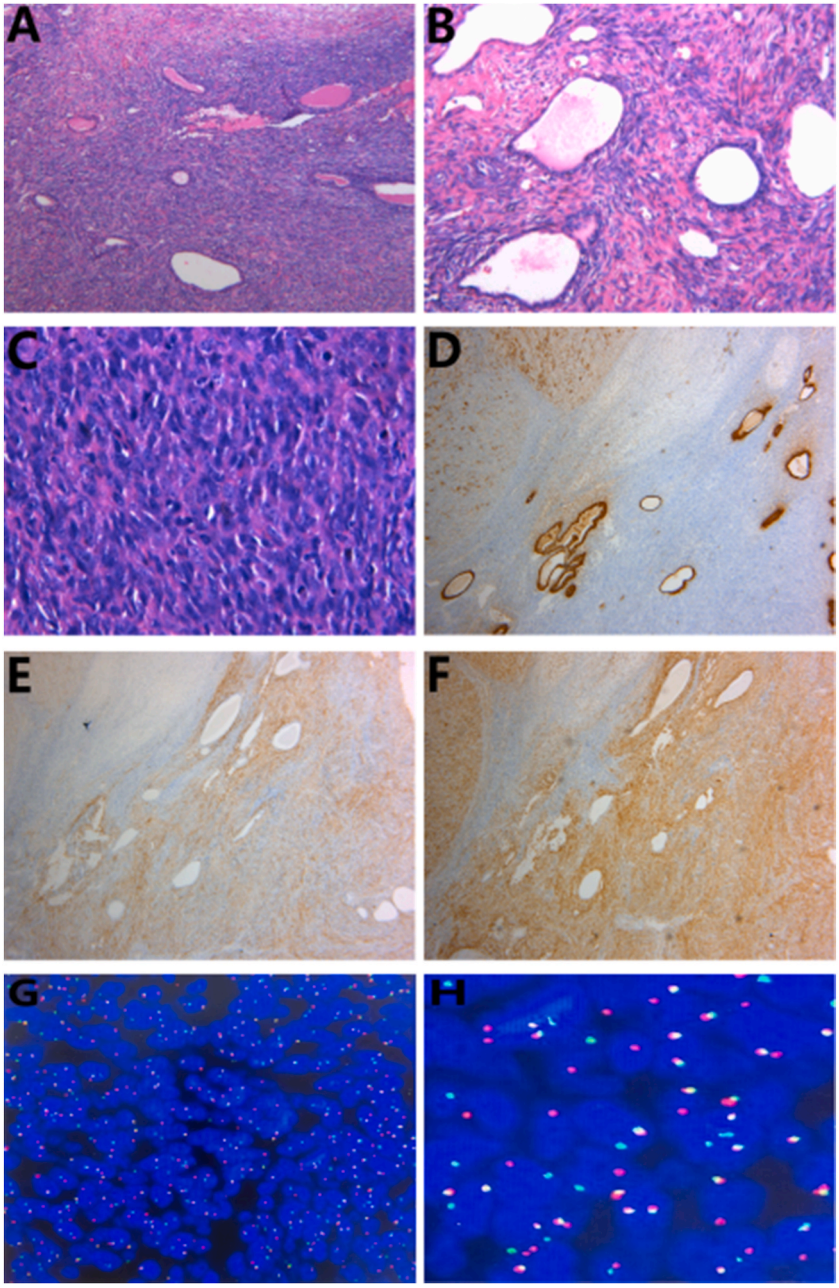


**Fig. 2.** (A) Tumors consist of a mixture of spindle cells and epithelial components (H & E, magnification 40 × ). (B) The epithelial components are arranged in a glandular duct-like pattern, and the glandular lumen contains eosinophilic secretions (H & E, magnification 100 × ). (C) The spindle cells were dense and long- or fat-spindle-shaped, and nuclear schisis was visible (H & E, magnification 200 × ). (D–F) Immunohistochemistry was partially positive for CK, weakly positive for CD99, and strongly positive for Bcl-2(magnification ×40). (G–H) FISH isolation probes confirmed a positive SYT gene rearrangement (split red and green signals)(magnification ×100 and x400).

chemotherapy combined with anlotinib-targeted therapy (ifosfamide 3g + epirubicin 100mg + anlotinib 10mg). Because the patient developed severe myelosuppression with fever after the third cycle of treatment, we adjusted the fourth and fifth cycle regimens (ifosfamide 2.25 g + epirubicin 75 mg + anlotinib 10 mg). One month after the end of the 5-cycle chemotherapy combined with targeted therapy, the patient was admitted to the hospital with heart failure. Patients reported that chest tightness, fatigue, nocturnal cough, increased frequency of bowel movements, and symptoms of loss of appetite gradually worsened in the past month. We consider that the tumor has progressed to the terminal stage, but the patient's systemic assessment does not support the continuation of chemotherapy, so we recommend genetic testing to find a new targeted treatment for the tumor. Unfortunately, the patient eventually decided to abandon all treatment after learning about the condition and died due to the rapid progression of the disease. From onset to

mortality, the overall survival time was 28 months.

## 3. Discussion

Synovial sarcoma is a rare soft tissue sarcoma with unclear differentiation, accounting for approximately 5–10 % of all soft tissue sarcomas. It can occur in patients of any age but is commonly seen mainly in adolescents and young adults [3,4]. According to the statistics, the incidence of SS is 0.81/1,000,000 among children and 1.42/1,000,000 among adults [5,6]. It can occur anywhere in the body, though most often in the joints of the extremities, and presents as a growing soft tissue mass. Other areas, such as the oropharynx, larynx, mediastinum, lungs, kidneys, prostate, and abdominal wall, can also show SS growth [7].

Primary pelvic SS is relatively rare. In reviewing the literature, we identified 15 cases of pelvic SS (Table 1 shows selected elements of the reported cases), with patients ranging in age from 17 to 77 years. Tumor locations also vary, including Pelvis, Ovary, Oviduct, Uterus, Prostate, and others, with the greatest number of cases occurring in the pelvis, which is consistent with epidemiological studies showing that SS tends to occur in the bones and joints of the human body. These reports were 33 % male and 67 % female, suggesting that women may be more likely to develop pelvic SS. The most common symptom of pelvic SS is abdominal pain and distension. In the pelvic region, physical examination can be useful in determining the exact size and location of the lesion, but limitations exist for tumors in the deep pelvis. Radiological examination is usually an important method of detecting occult pelvic SS, but it is not possible to rely on imaging evidence for a direct diagnosis. The pelvic anatomy is complex, and its differential diagnosis is more varied. Notably, pathologic testing combined with cytogenetic or molecular analysis can accurately diagnose SS, which is the currently accepted diagnostic standard.Surgery is the primary treatment option for pelvic SS, and in some cases, preoperative or postoperative radiation and chemotherapy can reduce the rate of disease recurrence and improve survival [8–21].

The clinical manifestations of SS are not obvious. It is usually a slow-growing, painless mass, and when detected, the tumor has grown very large and shows symptoms of pressure on the surrounding organs [22,23]. Given the young age of onset, insidious course, and atypical clinical manifestations, it is easily misdiagnosed as a benign tumor, leading to treatment delay. In this case, the first symptom was the pressure of SS on the bladder, rectum, and other organs, which manifested as difficulty in urination and painful swelling in the lower abdomen and perianal area. We initially suspected acute prostatitis, so we did not perform a digital rectal examination, which led to a misjudgment of the disease.

The diagnosis of SS relies on imaging tests such as computed tomography and magnetic resonance. On computed tomography, the mass appears as a round or lobulated mass with a density similar to or slightly lower than that of muscle. It is heterogeneous and often accompanied by punctate peripheral calcifications [24]. Magnetic resonance is an imperative test to determine the diagnosis and staging of SS, and the imaging characteristics are diverse. Masses smaller than 5 cm appear as homogeneous masses on all sequences, favoring benign imaging features. Among sarcomas larger than 5 cm, there is significant heterogeneity. The heterogeneous mixed signal of the high, medium, and low intensities on the magnetic resonance-diffusion weighted imaging (MR-DWI), the so-called "triple signal sign," is due to calcification, cystic changes, hemorrhage, and fibrosis in the process of tumor growth [25,26]. The MRI of our patient showed a clear triple-signal sign, with bleeding and cystic changes visible inside the tumor, and the intraoperative views confirmed the presence of hemorrhage inside the tumor.

In general, imaging is inadequate to confirm the diagnosis, and the final diagnosis is based on pathological findings. Synovial sarcomas are gray or yellowish-brown in appearance, and most are 3–10 cm in size [27]. Histologically, SS originates from mesenchymal cells and manifests as spindle cell sarcoma, which can be classified into monophasic, biphasic, and hypo-differentiated types according to the ratio of spindle cells to epithelial cells. The monophasic type is the most common type, accounting for approximately 50–60 % of all SS and consisting of only spindle cells. This is followed by biphasic SS, which makes up approximately 20–30 % of all synovial sarcomas and comprises both epithelial and spindle cells. Poorly differentiated SS is the least common type, accounting for 10–15 % of cases [28]. In our patient, preoperative mass puncture biopsy suggested soft tissue spindle cell sarcoma, and postoperative gross specimen pathology indicated a biphasic synovial sarcoma with the coexistence of epithelial cells and spindle cells. Immunohistochemistry was positive for EMA, CK, CK19, CD99, and Bcl-2 proteins and negative for S100 proteins. Molecularly genetically expressed as t(X:18) translocations, including the common SS18:SSX1 and SS18:SSX2 and the less common SS18:SSX4 translocations, this translocation is present only in synovial sarcoma and is seen in approximately 95 % of cases [29–31]. FISH and reverse transcription polymerase chain reaction (RT-PCR) assays can confirm this translocation. Of course, the diagnosis of synovial sarcoma cannot be excluded in patients with no detectable t(X:18) translocation and imaging showing synovial sarcoma features (approximately 5 % of all patients), as this group of patients may be associated with t(X; 20) and SS18L1-SSX1 fusion transcripts [32].

Synovial sarcoma is a rare tumor type, and there are no consensus guidelines for systemic treatment of the disease due to a lack of high-quality studies. Improving the survival rate of patients with this type of tumor and controlling the recurrence rate of this tumor are the main goals of the ongoing research on this disease. Currently, surgery is the preferred treatment for early SS. A complete surgical resection of the tumor is key to reducing postoperative recurrence and improving survival, and postoperative supplemental chemoradiotherapy can significantly improve treatment outcomes [33,34]. For relapsed, metastasized, and unresectable SS, chemoradiotherapy or targeted therapy plus chemoradiotherapy are also effective palliative treatments for delaying tumor progression, but there are differences in the effectiveness of treatment to prolong survival.

It is undeniable that radiotherapy and chemotherapy, as conventional and effective methods for treating tumors, also have corresponding drawbacks. In terms of radiotherapy, surgical resection of tumors combined with radiotherapy has been widely used in patients with synovial sarcoma and has significantly prolonged the survival period. NCCN guidelines recommend radiotherapy as standard adjuvant therapy after surgery for SS [35]. It has also been suggested that, when complications such as infection, local hemomatoma, and tissue necrosis can be controlled, early preoperative radiotherapy can inhibit further growth of the SS tumors and

**Table 1**
Review of characteristics of Synovial sarcoma.

| case No | First author Year | age (years) Sex | Clinical Symptoms | site | size(cm) | Diagoosis | follow (moths) | Immunohistochemistry | Molecular | therapy | Condition |
|---|---|---|---|---|---|---|---|---|---|---|---|
| 1 | Zheng L et al., /2023 | 42/F | Bellyache | Pelvis | 3.5 × 2.5 × 1.6 | Biphasic SS | 60 | Pancytokeratin+ , PAX8+ , ER+ , EMA+ BCL2 patchy+ , CK7 focally+ | t(X; 18)SS18-SSX RT-PCR | Surgery Radiotherapy | Alive |
| 2 | Zheng L et al., /2023 | 54/F | Bellyache | Pelvis | 10 × 7.5 × 5.8 | Biphasic SS | NA | Epithelioid cells-AE1/AE3+, Cam5.2+ EMA+, PAX-8+, CK7+, CD10 weakly+ Spindle cells-BCL-2+, PAX-8+, $CD10^{+}$, TLE-1+ | SS18 (SYT) (18q11) RT-PCR | Surgery Chemotherapy | NA |
| 3 | Gupta D et al., /2023 | 32/F | Abdominal distension | Ovary | 13.9 × 10 × 9.1 | Biphasic SS | 21 | BCL2+ , cytokeratin+ , TLE1+ | NA | Surgery Chemotherapy | Alive |
| 4 | BandeguddaS et al., /2022 | 35/M | Acute urinary retention | Prostate | NA | Monophasic SS | 1 | TLE-1+ , CD99+ , FLI+ , BCL-2+ | SS18-SSX | Surgery Chemotherapy | Died |
| 5 | Yordanov et al., /2022 | 77/F | Bellyache | Ovary | 7 × 5 | Monophasic SS | 16 | EMA+ , CD99+ | NA | Surgery | Died |
| 6 | Minig L et al., /2008 | 40/F | Vaginal bleeding | Vagina | 5 | PD biphasic SS | 25 | NA | SYT-SSX1 RT-PCR | Surgery Chemotherapy Radiotherapy | Alive |
| 7 | Ioannidis A et al., /2019 | 54/M | Abdominal pain Vomiting | Sigmoid colon | 20 × 17 × 12 | Monophasic SS | 3 | CD99+ , BCL2+ , S-100+ | NA | Surgery Chemotherapy | NA |
| 8 | Hasan R et al., /2013 | 17/M | Abdominal distension | Pelvis | NA | PD biphasic SS | NA | Vimentin+ , CD 99+ , Bcl2+ , Mic2+ , EMA focally+ | SSX-SYT RT-PCR | Chemotherapy Surgery | NA |
| 9 | Olivetti L et al., /2015 | 46/M | Acute urinary retention | Prostate | 8 × 7.5 × 8.5 | Monophasic SS | 3 | CK+,AE1/AE3+,BCL-2 diffusely+, CD99 diffusely+ | t ( X; 18 ) ( p11q11 ) SSX-SYT | Surgery Chemotherapy | Alive |
| 10 | Rekhi B et al., /2013 | 19/M | Swelling pain | Pelvis | 13.3 × 12.4 x9.9 | PD biphasic SS | NA | Vimentin+、bcl-2+、calponin+, MIB 1+,P53+, MIC 2 focally+ | t(X; 18) SYT-SSX2 RT-PCR | NA | NA |
| 11 | Mitsuhashi A et al., /2006 | 23/F | Bellyache | Oviduct | 14 × 12 × 4 | Monophasic SS | 23 | Bcl-2+,AE1/AE3+, CAM 5.2+, in the epithelial cells Focal+ in spindle cell, vimentin+ and desmin+ | SYT-SSX1 RT-PCR | Surgery Chemotherapy | Alive |
| 12 | Zhang J et al., /2021 | 48/F | Rectal bleeding | Rectum | 6.3 × 4.1 × 4.0 | Monophasic SS | 48 | Vimentin+, $CD99^{+}$, BCL2+ MiB-1 diffusely+ AE1/AE3 focally+, EMA focally+ | SYT/SSX1 RT-PCR | Surgery Chemotherapy | Died |
| 13 | Williams PJ et al., /2021 | 67/F | Rectal bleeding | Rectum | 6.8 × 4.5 | Biphasic SS | 3 | TLE1+, CAM5.2+, BCL-2+, $CD56^{+}$, SMM+ | SS18-SYT | Surgery | Alive |
| 14 | Cui W et al., /2023 | 60/F | Urethral bleeding | Urethra | 2.5 × 2.0 | Biphasic SS | NA | $CD99^{+}$, INI-1+,Ki-67+ , bcl-2+ , Vimentin+ CK focally+,CK-L focally+ | SS18-SYT | Surgery | NA |
| 15 | Dundr P et al., /2011 | 52/F | Metrorrhagia | Uterus | 3.5 × 3.5 × 2.5 | Biphasic SS | 56 | EMA+,S-100+,CD99+,NSE+ Vimentin+, in poorly differentiated cells CAM5.2+,AE1/3+, in larger epitheloid cells | t ( X; 18 ) SYT-SSX 1 RT-PCR | Surgery Radiotherapy | Alive |

F, Female; M,man; PD, poorly differentiated; SS, synovial sarcoma; RT-PCR, reverse transcription polymerase chain reaction, NA, Not available.

make the tumor edges clearer, facilitating complete surgical resection [36,37]. Following adjuvant chemotherapy, concurrent radiation therapy can also enhance the effect of eliminating tumor cells and, as a result, lessen distant micrometastases, which lowers the tumor-specific mortality rates [38]. There is no question that radiation therapy has a therapeutic effect on tumors; however, it is unclear when radiation therapy should be administered. When radiotherapy is administered prior to surgery or during a local recurrence, there is an uncontrollable risk of tumor progression or multiple metastases; conversely, if radiotherapy is given after surgery or chemotherapy, the improvement in clinical efficacy is also accompanied by an increase in complications, such as non-healing of postoperative wounds, bone marrow suppression, drug toxicity, etc [39].

From the perspective of chemotherapy, the clinical efficacy brought about by chemotherapy is very significant. The majority of chemotherapy regimens start with isocyclophosphamide and doxorubicin or epirubicin together as the first line of treatment. Furthermore, anthracyclines such as adriamycin are commonly used in chemotherapy. The primary advantages of chemotherapy include its capacity to lower tumor size, obstruct the blood supply to the tumor, hasten the death of tumor cells, lessen or postpone the likelihood of tumor rupture and metastasis, and facilitate the removal of tumors completely through surgery [40]. Inevitably, while chemotherapy brings curative effects, it is also accompanied by varying degrees of risks of drug side effects. The treatment outcome and survival duration of patients may be significantly impacted by acute or subacute toxic reactions to chemotherapy drugs, such as gastrointestinal disturbances and bone marrow suppression, as well as drug-specific side effects like liver damage, heart damage, and pulmonary fibrosis [41]. The patient in this case study report also experienced good outcomes such as tumor shrinkage and delayed metastasis of the tumor during treatment, but he also unavoidably suffered from bone marrow suppression, cardiac damage, and other toxic reactions to the chemotherapy drugs. Unfortunately, epirubicin induced heart failure, which indirectly led to the patient's death.

Looking back on the whole treatment process, this also inspired us to reflect more thoroughly. Firstly, during the period from the discovery of the tumor to the surgery and postoperative adjuvant chemotherapy, the patient's disease condition was in an improving state, and the tumor showed no signs of recurrence or metastasis, which indirectly proved that the treatment plan at that stage was appropriate and effective. Secondly, whether to continue radiotherapy following adjuvant chemotherapy is also an important point for us to consider. Although the best timing of radiotherapy has not been clearly determined, based on the study of this case, we think that continuing to supplement radiotherapy after adjuvant chemotherapy may better delay the period of tumor recurrence or metastasis. Even if there is no evidence of tumor recurrence after adjuvant chemotherapy, further radiation therapy is still necessary. Finally, in the event of a local recurrence or peripheral metastasis of the tumor, we think that the optimal strategy should be concurrent chemoradiotherapy combined with targeted therapy rather than radiotherapy or chemotherapy alone. When treating recurrent or metastatic SS with radiation therapy or chemotherapy alone, the therapeutic benefit is usually limited, and the probability of tumor progression or multiple metastases is not greatly decreased [42,43]. While chemoradiotherapy plus targeted therapy or immunotherapy is also a palliative treatment for tumor recurrence, it will limit tumor progression and extend patient survival more effectively than radiotherapy or chemotherapy alone [44,45]. When drug side effects are controlled stably, concurrent comprehensive treatment may be the most comprehensive and effective strategy for the early stages of tumor recurrence and progression. With the progress of research, new treatment options such as targeted drugs, immunotherapy, and metabolic therapy are in the research and exploration stages, and it is hoped that future studies can clearly reveal the biological characteristics of the tumor and provide more effective treatment options for patients with synovial sarcoma [46–51].

Synovial sarcoma, a highly malignant soft tissue sarcoma, and primary intrapelvic synovial sarcoma are even rarer and have a poor prognosis. Ultrasound, Computed Tomography, and MRI can help doctors detect the tumor at an early stage and treat it aggressively, especially with active surgical treatment, which can effectively improve the survival rate. Combined with the course of diagnosis and treatment of this case, it is possible to deepen the understanding of primary pelvic synovial sarcoma and discuss appropriate treatment strategies for this rare disease.

## 4. Conclusion

This case report highlights that SS is a rare tumor type, that it is not easy to clearly diagnose SS in the pelvic region, and that it is prone to erroneous empirical judgment. Imaging tests such as ultrasound, Computed Tomography, and MRI are important for the early diagnosis of tumors. For rare tumors with abnormal locations, pathology and immunohistochemistry are most critical in diagnosis and treatment. FISH or RT-PCR analyzes the molecular genetic characteristics of tumors, which provides more accurate guidance in diagnosis and treatment.

## Ethics declarations

Review and/or approval by the ethics committee of Nanchang First Hospital was not needed for this study because it is only a case report.

## Consent to participate and publication

Patient provided written informed consent for the publication of their anonymized case details and images.

## Availability of data and materials

All data and figures generated or analyzed during this study are included in this published article.

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No financial support was received for this submission.

## CRediT authorship contribution statement

**Zhiwei Li:** Writing – review & editing, Writing – original draft, Supervision, Formal analysis, Data curation, Conceptualization. **Kaibing Xiao:** Writing – original draft, Investigation, Formal analysis, Conceptualization. **Shaorui Niu:** Writing – review & editing, Methodology, Investigation, Data curation, Conceptualization. **Qiqi Zhu:** Writing – review & editing, Methodology, Data curation. **Zhiyang Xiao:** Resources, Investigation, Data curation. **Pang Yang:** Writing – review & editing, Writing – original draft, Resources, Methodology, Investigation, Formal analysis, Data curation, Conceptualization.

## Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

## Data availability

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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We would like to thank the patient for his participation and his consent to the publication of the case details and associated images.

## Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.heliyon.2024.e38807.

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