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Beyond the Pelvis: Giant Iliopsoas Endometriosis Revealing an Undiagnosed Müllerian Malformation

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Abstract

Unicornuate uterus with a non-communicating functional rudimentary horn associated with ipsilateral renal agenesis is a rare congenital Müllerian and Wolffian duct anomaly. We present a rare case of a 35-year-old woman with a known history of endometriosis presenting with a one-year history of cyclical left-sided lumbar pain. Pelvic MRI demonstrated a unicornuate right uterus with a non-communicating functional left rudimentary horn, left hematosalpinx, and ipsilateral renal agenesis on uro-CT. A large retroperitoneal mass measuring 11 cm arising from the left iliopsoas muscle, highly suggestive of a deep infiltrating endometriotic implant, was identified on imaging. Given the high risk of femoral nerve and external iliac vascular injury with surgical excision, a multidisciplinary decision was made to perform percutaneous ultrasound-guided sclerotherapy as a minimally invasive alternative. This case highlights an exceptionally rare association between a unicornuate uterus with a non-communicating functional rudimentary horn, ipsilateral renal agenesis, and giant retroperitoneal endometriosis of the iliopsoas muscle, underscoring the critical role of multimodal imaging in establishing the diagnosis and guiding management in such complex presentations.

Categories: Urology, Obstetrics/Gynecology, Radiology
Keywords: deep infiltrating endometriosis, iliopsoas endometriosis, müllerian duct anomaly, multimodal imaging, percutaneous sclerotherapy, renal agenesis, rudimentary uterine horn, unicornuate uterus

Introduction

Müllerian duct anomalies (MDAs) are congenital malformations of the female reproductive tract resulting from abnormal development, fusion, or resorption of the paramesonephric ducts. Their prevalence is estimated at 6.7% in the general female population and rises to 16.7% in women with recurrent miscarriages [1]. Among these anomalies, the unicornuate uterus with a non-communicating functional rudimentary horn, classified as Class U4a (hemi-uterus with a functional rudimentary cavity) according to the European Society of Human Reproduction and Embryology (ESHRE) and the European Society for Gynecological Endoscopy (ESGE) consensus [2], represents a particularly significant subtype. In a large literature review, 92% of rudimentary horns were non-communicating, and renal anomalies were present in 36% of cases, typically ipsilateral to the rudimentary horn. Non-communicating horns containing functional endometrium are the most clinically significant, as they are more likely to be associated with pelvic pain from hematometra or endometriosis due to retrograde menstruation [3].

When the non-communicating rudimentary horn contains functional endometrium, the menstrual blood shed at each cycle has no outlet toward the main uterine cavity or the vagina. It therefore accumulates within the horn, leading to hematometra, progressive distension, and increasing intracavitary pressure, which causes cyclical, often unilateral, pain. This pressure drives menstrual blood in a retrograde direction through the ipsilateral fallopian tube, resulting in hematosalpinx and spillage of endometrial cells into the peritoneal cavity, where they can implant and give rise to pelvic endometriosis. In rare instances, the disease extends into the retroperitoneum and reaches extra-pelvic structures [3]. Retroperitoneal endometriosis involving the musculoskeletal system remains exceptionally uncommon, and endometriosis of the psoas or iliopsoas muscle has been described in only a few case reports [4,5]. To our knowledge, there is a limited availability of cases associated with a unicornuate uterus or ipsilateral renal agenesis.

Because the contralateral unicornuate uterus menstruates normally, menstrual cycles usually appear regular, and the anomaly may remain unrecognized for years, with symptoms attributed to endometriosis alone. Several clinical features should therefore prompt dedicated pelvic imaging, ideally MRI, to look for an obstructive Müllerian anomaly. These include severe or progressive dysmenorrhea beginning soon after menarche, cyclical pain that is predominantly unilateral or felt in the lumbar region, pain that persists despite hormonal therapy, endometriosis with a strictly unilateral distribution, a unilateral pelvic mass with hemorrhagic content on ultrasound, an adverse obstetric history such as recurrent miscarriage, preterm delivery, or fetal malpresentation, and known unilateral renal agenesis. Conversely, the discovery of

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unilateral renal agenesis in a woman should prompt evaluation of the genital tract [3].

We report a case of a unicornuate uterus with a non-communicating functional left rudimentary horn associated with ipsilateral renal agenesis, complicated by a giant endometriotic implant of the iliopsoas muscle measuring 11 cm in its longest axis. This case highlights the diagnostic contribution of multimodal imaging, particularly pelvic MRI, in delineating this complex and rare association, distinct from obstructed hemivagina and ipsilateral renal agenesis (OHVIRA) syndrome [6], and reports the use of percutaneous ultrasound-guided sclerotherapy as a potential minimally invasive approach, the safety and efficacy of which remain under evaluation.

Case Presentation

A 35-year-old woman (G2P2, with one live birth and one premature delivery complicated by neonatal death at one year of age) presented with a one-year history of cyclical left-sided lumbar pain temporally correlated with menstrual periods. The patient had a known history of endometriosis and had been receiving oral contraceptive therapy for nine months prior to presentation. Clinical examination revealed isolated left lumbar tenderness without neurological deficit. No urinary or digestive symptoms were noted.

Pelvic MRI was performed on a 1.5 Tesla unit as the initial imaging modality. A uterine malformation was identified, consisting of a normally developed right uterus (Figure *1*) and a non-communicating functional left rudimentary horn measuring 49x44 mm containing hematometra, classified as Class U4a according to the ESHRE/ESGE consensus (Figure *2*). No vaginal anomaly, obstruction, or septum was identified on imaging (Figure *3*). Proximal and distal pelvic endometriosis was demonstrated, involving the left parametrium and left fallopian tube, showing T2 shadowing and T1 fat-saturated hyperintense foci, consistent with endometriotic hematosalpinx. A large retroperitoneal formation arising from the left iliopsoas muscle was identified, measuring 72x44x113 mm, appearing multilocular with high T2 and T1 signal intensity, without suppression on fat-saturation sequences and without significant contrast enhancement, consistent with a deep infiltrating endometriotic implant (Figure *4*). The main differential diagnoses considered were psoas abscess, retroperitoneal sarcoma, schwannoma, and necrotic lymphadenopathy; these were considered unlikely given the absence of any solid component or significant enhancement, the absence of rim enhancement, and the persistence of high T1 signal on fat-saturated sequences, indicating hemorrhagic content consistent with endometriosis.



FIGURE 1: Sagittal T2-weighted MRI sequence demonstrating the normally developed right uterus

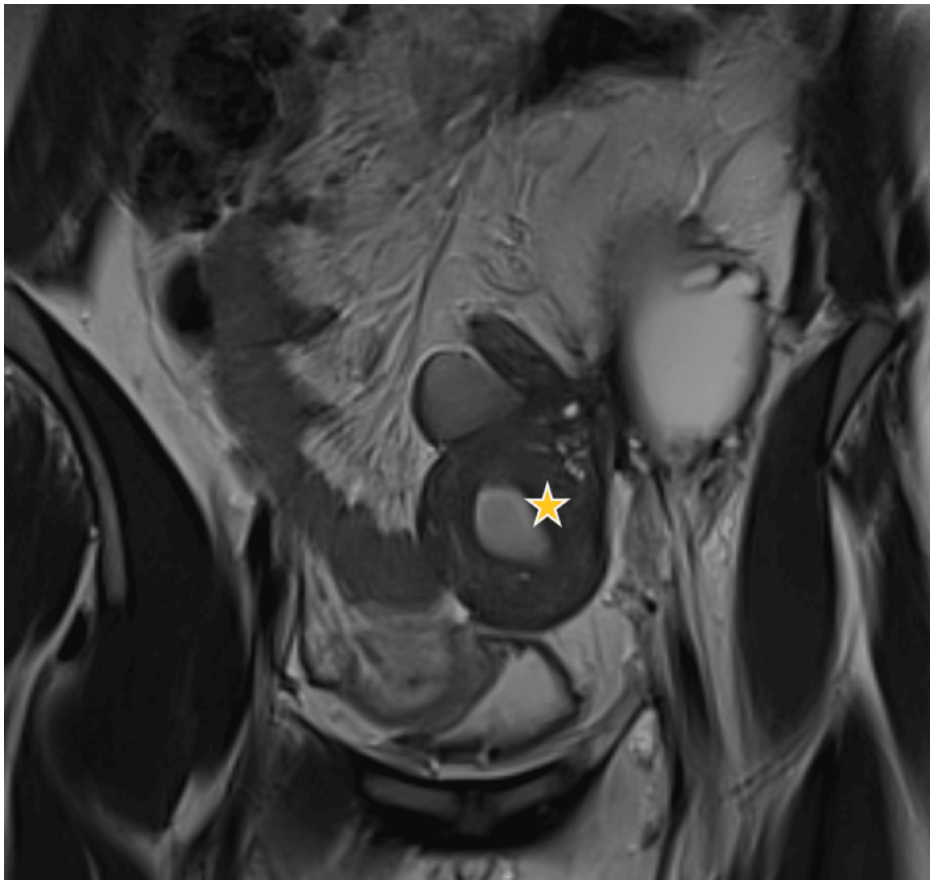


FIGURE 2: Coronal T2-weighted MRI sequence demonstrating the non-communicating left rudimentary uterine horn containing hematometra (marked with a star)

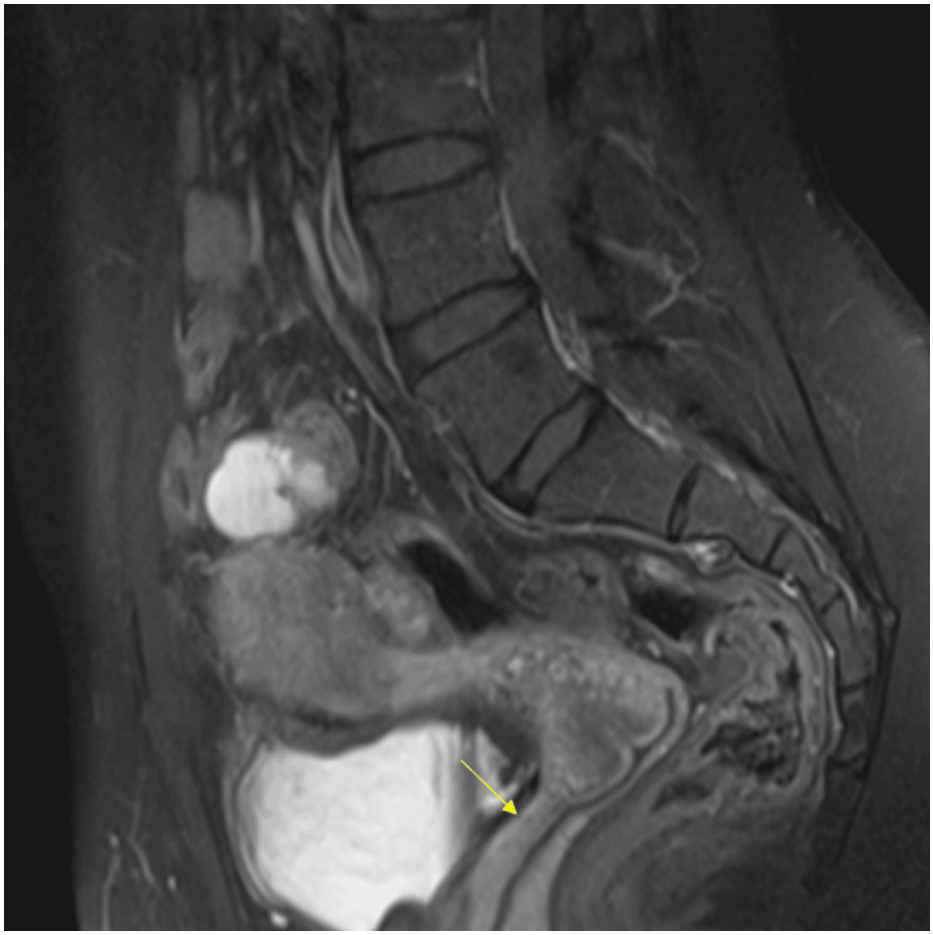


FIGURE 3: Sagittal post-contrast T1 fat-saturated MRI sequence demonstrating a patent vaginal canal (see arrow)

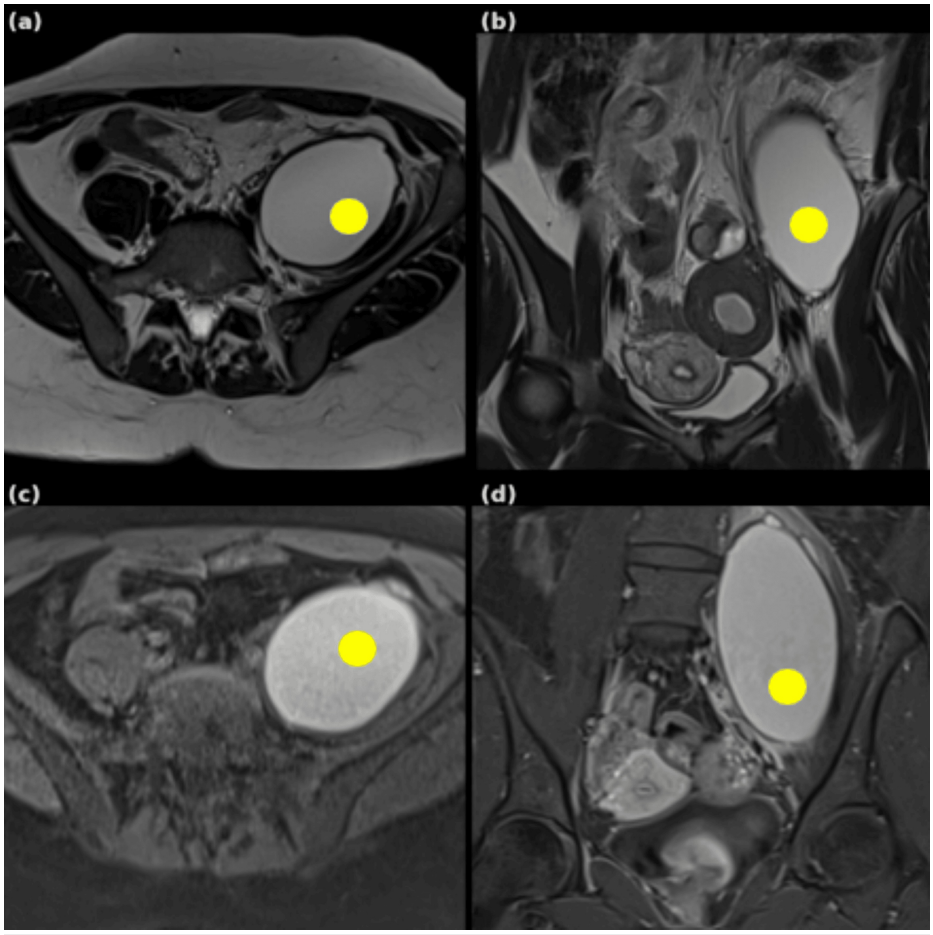


FIGURE 4: MRI showing a left-sided pelvic formation arising from the iliopsoas muscle, appearing hyperintense on axial and coronal T2-weighted sequences (a, b), hyperintense on axial T1 fat-saturation sequence (c), and without significant contrast enhancement on the coronal post-contrast sequence (d)

A complementary uro-CT scan was performed to assess the urinary tract. It confirmed the absence of the left kidney with compensatory right renal hypertrophy measuring 60x61x140 mm (Figure *5*) and demonstrated a large retroperitoneal mass arising from the left iliopsoas muscle, measuring 67x36x112 mm, with thick liquid content without contrast enhancement, extending to the left external iliac pedicle with infiltration of the adjacent fat and adhesive changes.



FIGURE 5: Contrast-enhanced abdominopelvic CT scan, coronal reconstruction, showing an empty left renal fossa consistent with renal agenesis and a fluid collection within the left psoas muscle (marked with a star)

Imaging findings were reviewed in a multidisciplinary team meeting involving interventional radiologists, gynecologists, and surgeons. Given the persistence of symptoms despite hormonal therapy, the size of the retroperitoneal lesion, and its close relationship with the left femoral nerve and external iliac vessels, which entailed a significant risk of neurovascular injury with surgical resection, percutaneous sclerotherapy was selected as a minimally invasive alternative. The procedure was performed under ultrasound guidance, using a puncture route that avoided the femoral neurovascular bundle. The cyst content was completely aspirated before injection of the sclerosing agent, which was re-aspirated at the end of the procedure. Aspiration yielded thick, chocolate-colored fluid, consistent with old hemorrhagic content. Surgical excision of the non-communicating functional left rudimentary horn was also proposed after the patient had been informed of the associated risks, but she declined surgery. The patient is currently under close clinical and radiological follow-up.

Discussion

Unicornuate uterus with a non-communicating functional rudimentary horn is a rare Müllerian anomaly, with a prevalence of approximately 0.1% in the general female population, 0.5% in infertile women, and 2% in those with a history of miscarriage [7]. It results from the unilateral failure of one Müllerian duct to develop normally during the seventh to eighth week of gestation, while the contralateral duct undergoes complete differentiation [1]. The intimate embryological relationship between the Müllerian and Wolffian ducts explains the frequent association with ipsilateral renal agenesis, reported in up to 40% of cases [2,3]. According to the ESHRE/ESGE classification, this anomaly is categorized as Class U4a when the rudimentary horn contains functional endometrium without communication with the main uterine cavity [2].

OHVIRA syndrome shares the same embryological substrate but is defined by a distinct triad: uterus didelphys, obstructed hemivagina, and ipsilateral renal agenesis [6]. The absence of uterine duplication, vaginal obstruction, and hematocolpos in the present case excludes this diagnosis. To date, the closest published case is that of Moawad et al., describing a Class U4a unicornuate uterus with ipsilateral renal agenesis and a confirmed normal vagina, with OHVIRA explicitly excluded by vaginoscopy; however, no retroperitoneal endometriosis was reported [7]. To our knowledge, the limited availability of previously published cases combining all four features of the present case precludes its classification within any existing syndrome and supports its recognition as a rare and novel phenotypic association within the spectrum of Müllerian and Wolffian duct anomalies, rather than a distinct clinical entity [6,7,8].

A review of 366 rudimentary horn presentations reported a mean age of diagnosis of 23 years for gynecologic presentations, with 78% of cases identified in the third decade of life or later [9]. In the present case, diagnosis was established at age 35, reflecting a significant diagnostic delay. The presence of a normally functioning right uterus maintaining regular menstrual cycles rendered the underlying Müllerian malformation clinically silent, leading to the attribution of symptoms to endometriosis alone without identification of the congenital anomaly [3,7]. Beyond endometriotic complications, unicornuate uterus with a non-communicating rudimentary horn is associated with significant obstetric morbidity, including preterm delivery and neonatal death, as illustrated by the obstetric history of the present patient [3].

The non-communicating functional left rudimentary horn, unable to evacuate its menstrual output, generated progressive hematometra and ipsilateral hematosalpinx, with retrograde spillage of endometrial cells into the peritoneal cavity, accounting for the strictly ipsilateral distribution of endometriotic lesions observed on imaging [3,10]. Retroperitoneal extension to the iliopsoas muscle is hypothesized to have occurred via lymphatic or hematogenous dissemination of endometrial cells, consistent with mechanisms proposed for other cases of extra-pelvic skeletal muscle endometriosis [11].

Endometriosis of the iliopsoas muscle is an exceptionally rare entity, with only five cases identified in a systematic review of skeletal muscle endometriosis [11]. Tsukasaki et al. reported a retroperitoneal endometriotic cyst infiltrating the iliopsoas, incidentally discovered in a patient presenting with acute back pain, highlighting that this diagnosis may be overlooked in the absence of a high clinical index of suspicion [5]. Cyclical lumbar pain correlated with menstruation represents the hallmark clinical feature across all reported cases and was the key symptom that prompted radiological investigation in our patient [4,11]. Andriamanjay et al. reported the only case successfully managed by hormonal therapy alone, establishing medical treatment as a viable alternative to surgery in selected patients [12]. In the present case, the exceptional volume of the lesion, measuring 72x44x113 mm, representing one of the largest iliopsoas endometriotic implants reported to date, combined with its proximity to the left femoral nerve and external iliac vessels, made surgical resection particularly hazardous [4,12].

Pelvic MRI is the modality of reference for characterizing Müllerian anomalies and deep infiltrating endometriosis, providing superior soft-tissue contrast and reliable differentiation from other retropsoas lesions, including abscess, schwannoma, and retroperitoneal sarcoma [13,14]. Uro-CT offers complementary urological assessment, particularly for identifying associated renal anomalies and delineating retroperitoneal disease extension [3]. In the present case, the combination of high T2 signal intensity and T1 fat-saturated hyperintense foci without contrast enhancement on MRI, together with the cyclical nature of the pain, the coexisting ipsilateral pelvic endometriosis, and the aspiration of chocolate-colored fluid, was highly suggestive of the endometriotic nature of the iliopsoas lesion, while uro-CT confirmed ipsilateral renal agenesis and mass extension to the external iliac pedicle [9,13,14]. However, the diagnosis was not confirmed histopathologically, which is a limitation of this report.

According to the 2022 ESHRE guidelines, surgical excision is the recommended treatment for deep infiltrating endometriosis refractory to medical management [15]. In the present case, symptoms persisted despite nine months of oral contraceptive therapy. However, several anatomical constraints argued against surgery. The lesion measured 11 cm in its longest axis and arose from the left iliopsoas muscle, so complete excision would likely have required partial resection of the muscle itself. The femoral nerve runs in the groove between the psoas and iliacus muscles, in direct contact with the lesion. On CT, the mass extended to the left external iliac pedicle, with infiltration of the adjacent fat and adhesive changes, which made a safe dissection plane unlikely. In this setting, complete excision carries a significant risk of femoral nerve injury, with potential permanent motor and sensory deficit of the lower limb, and of external iliac vascular injury [4,12].

Percutaneous image-guided sclerotherapy is well established for ovarian endometriomas, with reported cyst resolution rates exceeding 80% at one year [16-18]. Based on a systematic review by Ye et al., it has not previously been described for retroperitoneal iliopsoas endometriosis [11]. Its use in the retroperitoneal space nevertheless requires particular caution. An ovarian endometrioma is enclosed by a fibrous wall within the ovary. A retroperitoneal implant, by contrast, lies within loose connective tissue, in close contact with the femoral nerve and the external iliac vessels. Any leakage of the sclerosing agent into this space may cause chemical neuritis or nerve injury (ethanol, the most widely used agent, is itself used for therapeutic neurolysis) and perivascular inflammation or thrombosis. To minimize these risks, the procedure was performed under ultrasound guidance, using a puncture route that avoided the femoral neurovascular

bundle. The cyst content was completely aspirated before injection of the sclerosing agent, and the agent was re-aspirated at the end of the procedure, in line with techniques described for ovarian endometriomas [17,18]. In case of incomplete response or recurrence, the management strategy includes repeat sclerotherapy sessions, adjuvant hormonal therapy, and, as a last resort, surgical excision in a specialized center. This approach should therefore be regarded as investigational, and its safety and efficacy remain to be established. Outcomes in the present case are currently under evaluation.

The non-communicating functional rudimentary horn is the presumed source of the endometriotic lesions in this case. Surgical excision of a cavitated non-communicating rudimentary horn, usually by laparoscopy and often combined with ipsilateral salpingectomy, is the recommended treatment, as it relieves pain related to hematometra, removes the source of retrograde menstruation, and prevents the risk of rudimentary horn pregnancy and rupture [3,9]. In the present case, excision of the rudimentary horn was proposed by the multidisciplinary team, but the patient declined surgery. As long as the functional horn remains in place, persistent retrograde menstruation may lead to recurrence or progression of endometriosis, including at the iliopsoas site. This is a limitation of the current management and an important consideration for follow-up.

Conclusions

This case describes a rare association between a unicornuate uterus with a non-communicating functional rudimentary horn, ipsilateral renal agenesis, and presumed large retroperitoneal endometriosis of the iliopsoas muscle. Multimodal imaging proved indispensable in characterizing the full extent of the Müllerian malformation and its endometriotic sequelae, guiding the multidisciplinary therapeutic decision. This case underscores the importance of maintaining a high index of clinical suspicion for underlying Müllerian anomalies in patients presenting with cyclical lumbar pain, and suggests that percutaneous ultrasound-guided sclerotherapy, performed with strict precautions to protect adjacent neurovascular structures, may be a promising minimally invasive option for retroperitoneal endometriosis, although long-term follow-up data and larger series are needed to establish its efficacy and safety profile, particularly with regard to the risk of recurrence and potential neurovascular complications.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

Concept and design: Karima Sif Nasr, Samia Amar, Siham Elhaddad, Nazik Allali, Latifa Chat

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Supervision: Karima Sif Nasr, Siham Elhaddad, Nazik Allali, Latifa Chat

Disclosures

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