

Menstruating umbilicus – An interesting case report on primary umbilical endometriosis

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Abstract: Primary umbilical endometriosis (PUE) is defined as the presence of ectopic endometrial tissue in the umbilical region of the abdomen. Although extra-pelvic endometriosis is not uncommon, a spontaneous development of endometriosis in the umbilical region without any previous surgical intervention is rare and accounts for only 0.5% to 1% of all cases. In the current case report, we present an interesting case of a young woman with a history of pain in the peri-umbilical region followed by bleeding through the umbilicus during menstrual cycles for the past 6 months. On examination, a vague nodularity was felt just below the umbilicus in the subcutaneous plane. Imaging studies indicated the benign nature of the lesion which was subsequently excised and sent for histopathological examination. Microscopically it was found to be a focus of extra-pelvic, primary endometriosis in the umbilical region.

Keywords: Extra-pelvic, Primary umbilical endometriosis, Subcutaneous plane

Key message: Case of primary umbilical endometriosis in a young, parous woman with no previous surgical history – an interesting case report.

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I. Introduction:

Endometriosis is the presence of functional endometrial tissue outside the uterine cavity.^[1] It affects 10–15% of all women of childbearing age and 6% of perimenopausal women.^[2-3] It can involve any of the anatomic sites like pelvic peritoneum, fallopian tubes, ovaries, urinary tract, bladder, colon, appendix, rectum, heart, kidneys, lungs, pancreas, muscles, central nervous system and rarely the subcutaneous tissue.^[4] In women with previous Caesarean-section, umbilical endometriosis can occur in 30% of cases.^[5] However, primary umbilical endometriosis still accounts for only 0.5% to 1% of all cases^[1], with less 100 cases reported in literature so far, making it a rare entity in clinical practise.

II. Case Report:

A 34-year-old woman, who has completed her family, delivered twice naturally, with no previous surgical history, presented with complaints of vague pain in the peri-umbilical region and bleeding through the umbilicus during menstrual cycles. She claims to be having regular menstrual cycles with normal flow. On clinical examination, a nodular area was felt just beneath the umbilicus, measuring 4x4cm. It was firm in consistency and was located in the subcutaneous plane. Further imaging studies indicated the lesion to be benign and that it did not have any intra-abdominal extension. Surgical excision was done followed by histopathological examination. Grossly the excised tissue was grey brown, soft to firm in consistency with focal areas of hemorrhage and ill-defined grey white areas on cut section (Figure 1). Microscopically, the material received showed fibromuscular tissue with islands of endometriosis comprising of tubular glands lined by a single layer of proliferating columnar epithelium, bordered by a compact endometriotic stroma (Figure 2). There were also foci of hemorrhage. Immunohistochemistry was done for confirmation, which showed strong positivity for estrogen receptor (ER) and progesterone receptor (PR) in the endometrial glands while CD10 (cluster differentiation 10) was strongly positive in the stromal cells (Figure 3), thus authenticating the diagnosis of primary umbilical endometriosis.

III. Discussion:

Primary (spontaneous) umbilical endometriosis was first defined by Villar in 1886.^[6] According to hypothesis proposed by Sampson in 1920, endometriosis was caused by retrograde menstruation passing thorough the fallopian tube into the pelvis.^[7] Several theories / hypotheses have been proposed to understand the origin of this disease. Latcher has classified these theories into three main categories: the embryonal rest theory,

the coelomic metaplasia theory and the migratory pathogenesis theory.^[8] Although there are several theories explaining the potential origin of endometriosis, the pathogenesis still remains a much-debated issue.

In the present case report, we share a riveting history of a young woman, in the reproductive age group, who has completed her family, delivered twice naturally, presented with complaints of abdominal pain and bleeding through the umbilicus during menstrual cycles for the past 6 months. She claims to have regular menstrual cycles with normal flow and without any pelvic pain or discomfort. She had no previous surgical history, did not undergo sterilization post child birth. On imaging studies, a fairly circumscribed heterogeneously hypoechoic vascularised and non-vascularised nodule in the subcutaneous plane just below the umbilicus was observed. The lesion had no intra-abdominal extension. At this point, differential diagnoses of haemangioma, vascular malformation, urachal anomaly and cutaneous endometriosis were considered. Surgical excision was planned subsequently and the tissue was sent for histopathological examination. Although on gross examination it wasn't pointing at a specific diagnosis, microscopic examination revealed the presence of islands of endometriosis in the fibromuscular tissue. It was further authenticated by immunohistochemical markers for endometrial glandular (ER and PR) and stromal components (CD10).

Post-surgery the patient was keeping well with no similar bleeding episodes from the umbilicus. Although excision of the umbilicus along with the nodule is the preferred management, in the present case, only local excision was done as pre-operatively other differentials of haemangioma and urachal anomaly were also considered. Further, the lesion was solitary, fairly circumscribed and was confined to the subcutaneous plane. Post-operatively the patient was symptomatically relieved and hence she was put on follow up with no insistence on surgery unless she presents with a local recurrence.

IV. Conclusion:

Endometriosis being a common condition seen in women of reproductive age group, its unusual site of presentation reinforces the fact that it could potentially involve any of the anatomical structures and organs including brain, lungs, breast etc. However, since it is a benign condition, pharmacotherapy along with surgical excision provides excellent cure rates.

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Images with Legends:

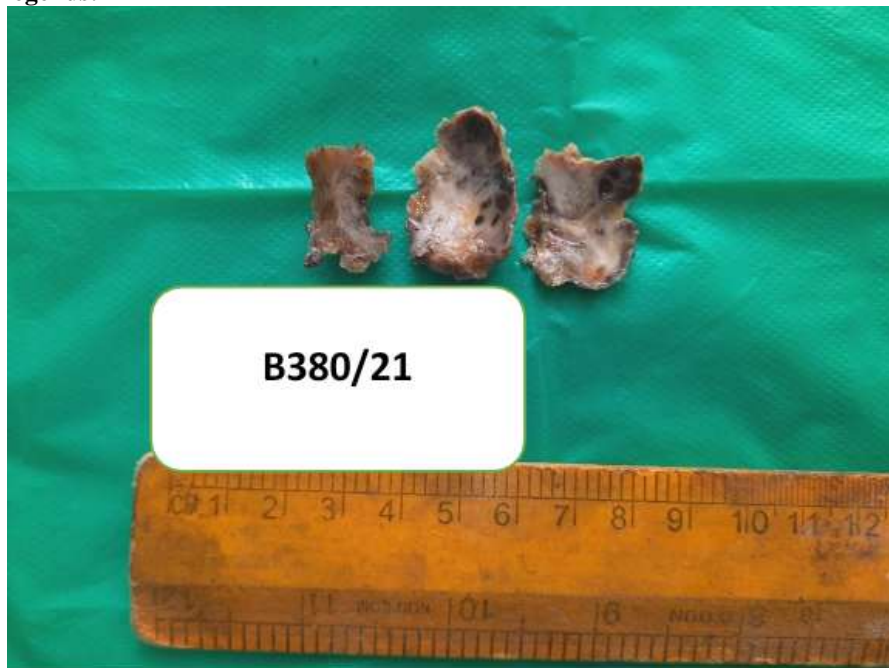


Figure 1: Resected specimen of the nodule in the subcutaneous plane from the umbilical region showing focal areas of hemorrhage and ill-defined grey white areas.

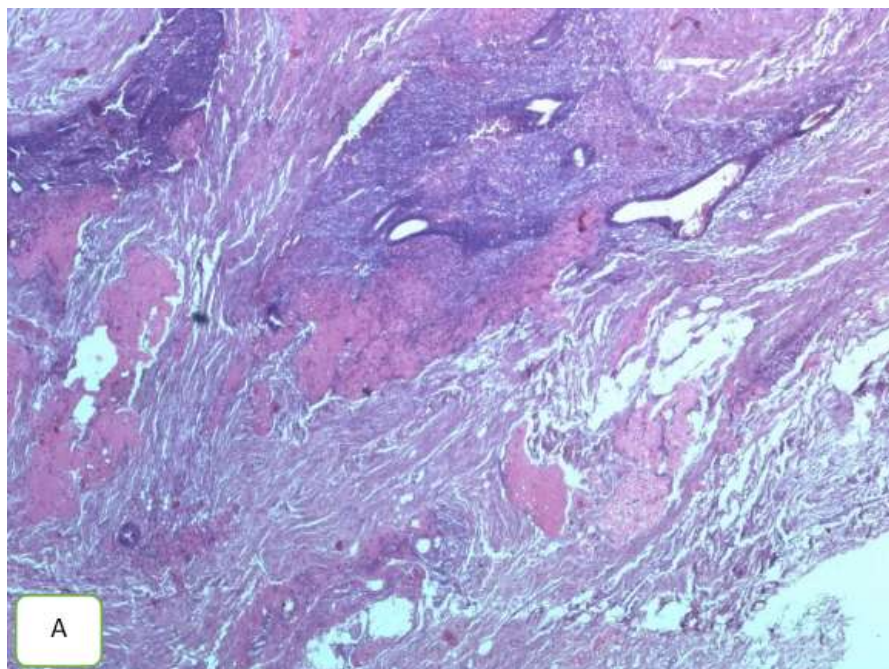
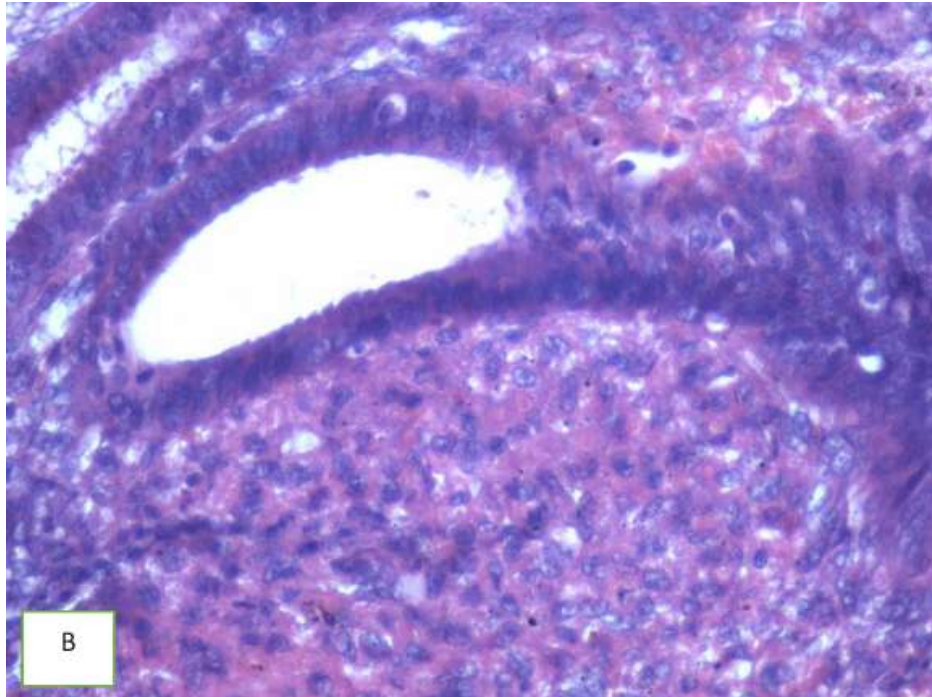


Figure 2: (A) Photomicrograph showing fibromuscular connective tissue with interspersed islands of endometrial tissue comprising of tubular glands and stroma. (H and E stain, x40 magnification)



(B) Photomicrograph showing a tubular endometrial gland lined by single layer of columnar epithelium, surrounded by a compact endometrial stroma. (H and E stain, x400 magnification)

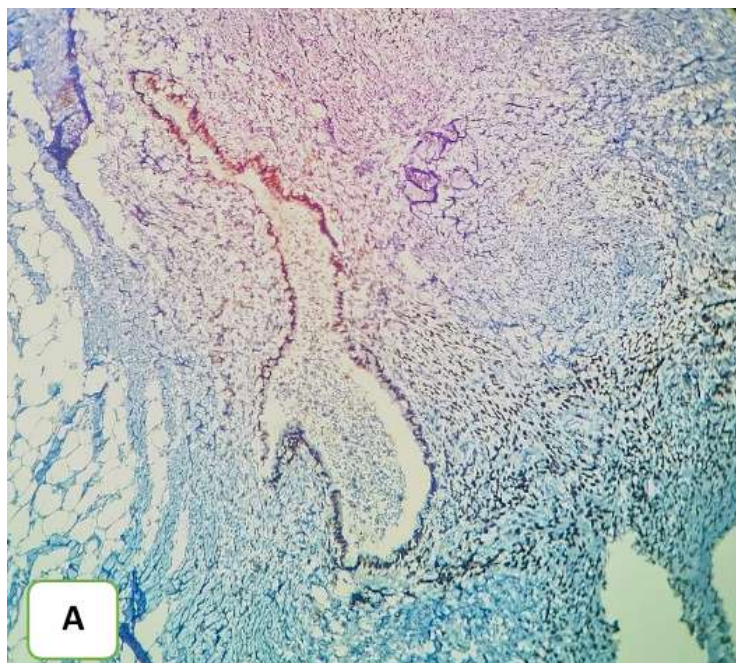
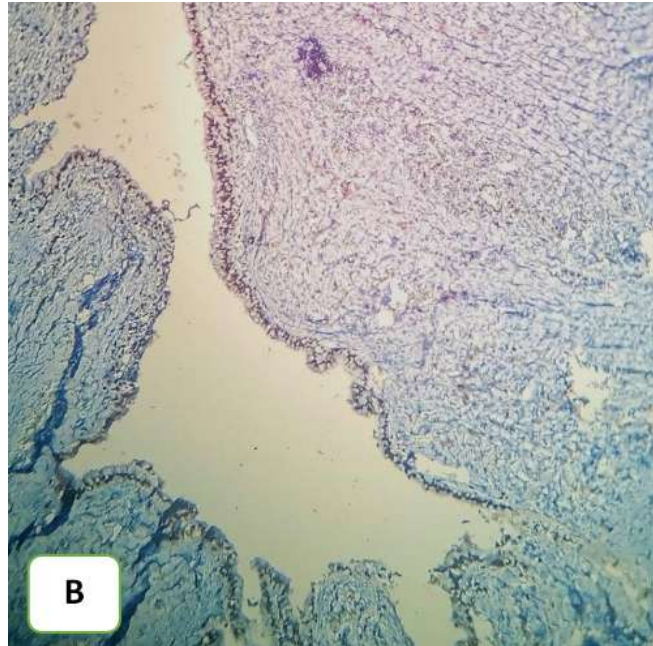
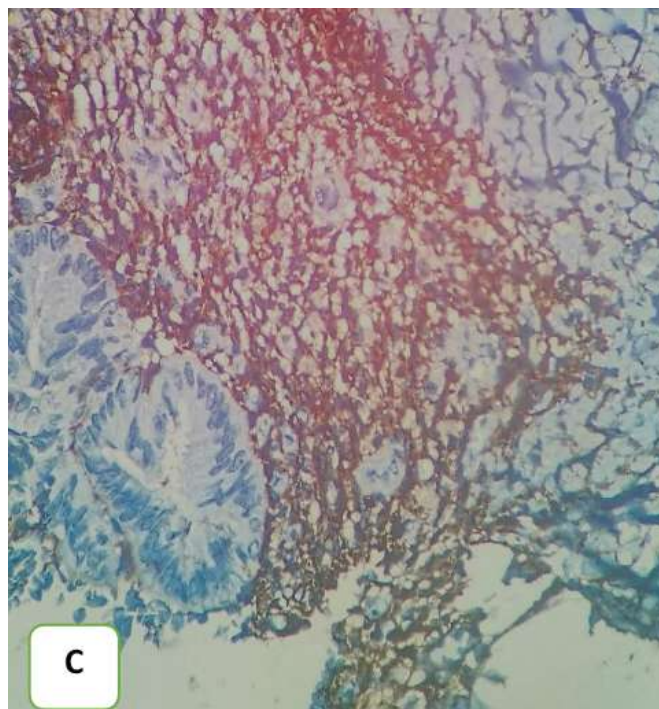


Figure 3: (A) Immunohistochemical marker -ER highlighting the endometrial glands (x100 magnification)



(B) Immunohistochemical marker -PR highlighting the endometrial glands (x100 magnification)



(C) Immunohistochemical marker – CD10 highlighting the endometrial stromal cells bordering the glands (x400 magnification)