Short reports

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Endodermal sinus (yolk sac) tumor of vagina and cervix in an infant

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Abstract. A case of vaginal and cervical endodermal sinus (yolk sac) tumor in a 6-month-old female infant is reported. The patient presented with an intermittent bloody discharge on her diapers. Pelvic CT showed an irregular soft tissue density mass with heterogeneous enhancement within the vagina and extending to the cervix. The histopathologic features were identical to those of endodermal sinus tumors.

Endodermal sinus tumors (ESTs) were first described by Teilum in 1959 [1]. Most such tumors occur in the ovaries and testes of young patients and they are usually related to midline structures. Only 10%–15% are extragonadal. We present a case of an extragonadal EST arising from the vagina and cervix that was demonstrated by CT.

Case report

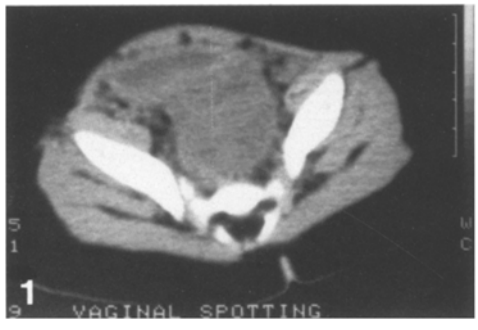
A 6-month-old Chinese female infant presented with a bloody discharge on her diapers. The results of urinalysis, urine and stool culture were negative. Sonography demonstrated a large pelvic mass. Subsequently her bloody-mucoid vaginal discharge became more frequent. Alpha-fetoprotein (AFP) levels were markedly elevated to 1270 ng/dl (normal value < 20 ng/dl at 6 months of age), but levels of human beta-chorionic gonadotropin (β-hCG), carcinoembryonic antigen (CEA) and the results of a luteinizing hormone-releasing hormone (LHRH) test were within normal limits. Repeat abdominal sonography demonstrated a 3.8 cm × 3.5 cm solid tumor mass behind the uterus with a high resistive blood flow.

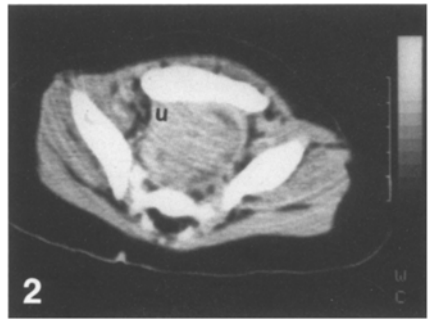
A pelvic CT study showed a non-specific soft tissue mass, with heterogeneous enhancement after contrast medium was injected (Figs. 1–3), located between the rectum and urinary bladder. The perivaginal fat planes were relatively clear.

At surgery a lobulated firm solid mass was found, originating from the vagina and extending into the cervix. The uterus, fallopian tubes, ovaries, bladder and rectum were grossly intact. Histologic examination revealed the characteristic Schiller-Dural bodies of an EST. Mitotic figures were numberous and the full thickness of the vagina had been invaded.

Discussion

The extragonadal location of ESTs in vagina and cervix could be the result of germ cells "lost" during their migration from the yolk sac along the dorsal wall of the embryo to the gonadal folds [2]. A review of the literature revealed only 3 cases of vaginal and cervical ESTs and 57 cases of vaginal ESTs [3, 4]. To our knowledge this is the first reported case of EST in an infant involving both vagina and cervix.





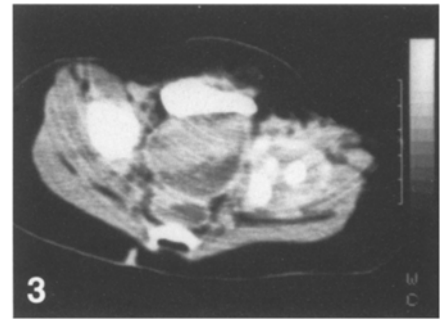


Fig. 1. Unenhanced CT scan reveals a heterogeneous low-density mass behind the urinary bladder

Fig. 2. Enhanced CT scan at the same level as Fig. 1 shows this mass has a broad base on the cervix and vaginal wall with heterogeneous enhancement and a lower – density component. The uterus *(u)* is displaced anteriorly

Fig. 3. Enhanced CT scan at a lower level than Fig. 2 discloses more caudal involvement of the vaginal wall

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The CT appearance of EST in the vagina is of a soft tissue mass with irregular margins that shows heterogeneous enhancement. The adjacent fat planes are frequently obliterated as a result of external extension of the tumor, sometimes through the uterus and fallopian tubes, due to its infiltrative character. Metastasis may occur to lung and liver. The differential diagnosis of vaginal tumors includes squamous cell carcinoma, clear cell carcinoma, melanoma, sarcoma and teratoma. In this case the elevated AFP level, normal β-HCG level and LHRH test and intermittent bloody discharge from the vagina led us to make the correct diagnosis.

Extragonadal ESTs have a worse prognosis than gonadal ESTs, because the diagnosis of extragonadal tumors is usually made at an advanced stage [5]. The management of gonadal and extragonadal ESTs may require a combination of surgery, radiotherapy and chemotherapy. An AFP radioimmunoassay and regular follow-up pelvic CT scanning can provide a guide to treatment response.

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