Accessory Penis and Scrotum in a Male Infant

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An 11-day-old infant was admitted to the hospital with an accessory penis and scrotum on the posterior thoracic region. He was a full-term vaginal delivery and there were no perinatal problems. His physical examination was unremarkable except for a midline, thoracic, soft tissue mass that measured 5 × 4 cm and rose 1.5 cm above the plane of the back and an accessory small penis with well-formed glans, urethral meatus, and scrotum over the swelling. There were no formed testes (►**Fig. 1A, 1B**). The ectopic phallus was firm and well defined but it became larger after tactile stimulation. Magnetic resonance imaging (MRI) of the spine revealed no evidence of the extension of the mass into the intraspinal space or defect of the vertebra but demonstrated a fusion of the spinous processes adjacent to the base of the mass (►**Fig. 2**). Based on these features, a preoperative diagnosis of fetus in fetu (FIF) was made. The mass was excised. The resected specimen consisted of the "scrotal" structure with attached phallus and no testes were present. Pathological examination revealed abundant cavernous sinuses as well as smooth muscle cells present in the *corpus cavernosum* (►**Fig. 3A**) as in the primary penis; the small cystic spaces lined by intestinal-type epithelium could be seen at the bottom of the penis near the scrotum (►**Fig. 3B**). The infant had an uneventful postoperative course and physical examination as well as neurologic examination. Both were within the normal limits. Clinical follow-up at 18 months did not show recurrence. The parents were fully informed and consent was obtained. All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

The appearance of an ectopic genital organ is an extraordinarily rare event. Davis in 1949 reported a case of a peniform structure associated with a dermoid of the perineum.[1] More recently, Baht et al described a gluteal pseudophallus in a male child.[2] An accessory scrotum is also an unusual developmental anomaly with only 53 cases reported so far.[3] Histological confirmation of penile and scrotum tissue is available in both published cases and our case. Hitherto, the correct classification of this rare deformity is not clear. Historically,

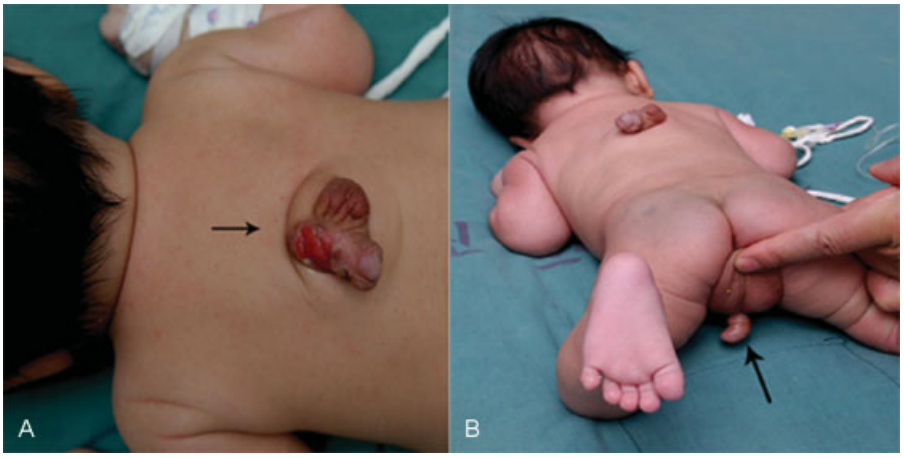

Fig. 1 (**A**) Accessory penis and scrotum on thoracic area over dorsal midline. (**B**) In the normal position is a penis of normal size and contour.

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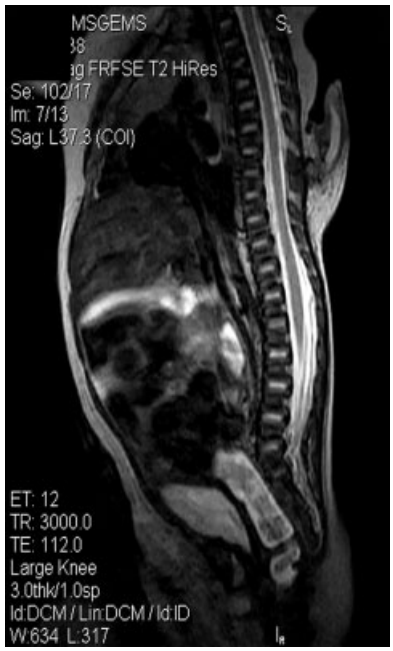

Fig. 2 Magnetic resonance imaging of the spine revealed no evidence of the extension of the mass into the intraspinal space or defect of the vertebra.

differentiated teratoma.[8] However, it is necessary to differentiate between FIF and teratoma because of a slight risk of malignancy associated with retroperitoneal teratomas.[6–8] In contrast, FIF is almost always benign.[6–8] In our case, the presence of intestinal-type epithelium found together with abundant cavernous sinuses and smooth muscles in the accessory penis implied that the accessory penis was non-single differentiation. This result validates the preoperative diagnosis of FIF. The recommended treatment for FIF is the complete surgical removal. As it is essentially a benign condition, FIF always has a good long-term prognosis. A regular follow-up should also be considered because one case of malignant recurrence after removal of FIF has been reported.[9]

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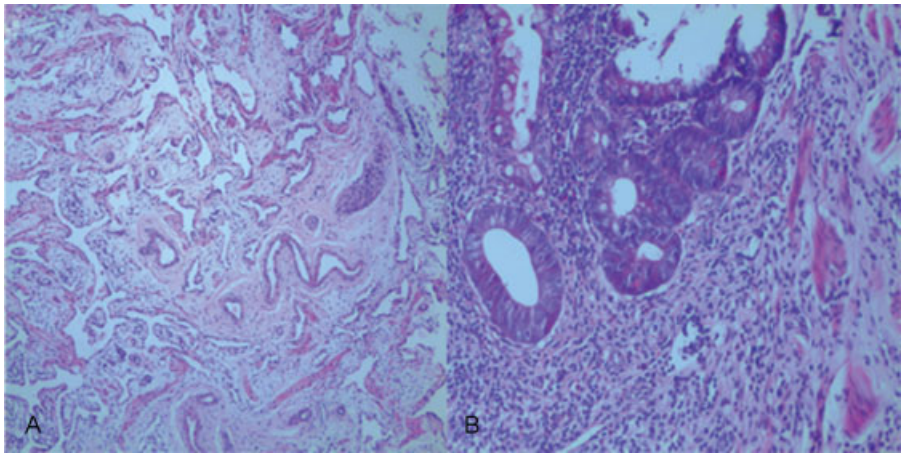

Fig. 3 (**A**) Abundant cavernous sinuses and smooth muscle cells showed in the *corpus cavernosum* of the accessory penis (hematoxylin-eosin, original magnification ×100). (**B**) The small cystic spaces lined by intestinal-type epithelium surrounded by inflammatory cells at the root of the accessory near the scrotum (hematoxylin-eosin, original magnification ×100).

authors have classified similar events as either FIF or teratoma.[4,5] FIF (also known as parasitic twins) is a rare congenital anomaly in which a vertebrate fetus is found inside a normal cotwin's body.[6] The most widely accepted theory is that FIF results from the unequal division of the totipotent cells of a blastocyst.[6] The smaller twin then gets incorporated within the normally developing twin by unknown mechanisms.[6] The most common site of FIF is the retroperitoneum.[6,7] Unlike teratoma, which often occurs in the lower abdomen and pelvis, FIF generally occurs in the upper abdomen, and vertebral column and limbs often can be seen in these fetuses.[7] Because some FIFs lack a spinal column as noticed in this case, investigators have suggested that FIF is a type of highly

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