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Tailgut Cyst in a Newborn: Report of a Case and Literature Review

Jaffar Al-Khuzaie, MD* Saeed Al-Hindi,MD,FRCST, CAB**
Aziz Hassan,*** Raja Al-Youssif ****

We report a case sacrococcygeal tailgut cyst in a newborn. Although this condition is extremely rare it should be one of the differential diagnosis of sacrococcygeal swellings.

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Tailgust cyst (retro-rectal cystic hamartoma) is a rare congenital lesion, which is present in the pre-sacrococcygeal space. It is made up of multi-loculated cyst lined by gastrointestinal epithelium, often mucus producing cells and squamous, transitional and columnar epithelium. Most of the lesions are reported in adult women; half are discovered incidentally[1,2]. The definitive treatment is surgical excision.

THE CASE

A newborn girl was found to have a cystic right gluteal mass on routine examination. She is a product of full term with a birth weight of 4.3 Kg. No other abnormality detected on systemic examination. Rectal examination was negative except for some vague fullness. Plain spine x-ray did not show any abnormality while spinal and pelvic ultrasound showed large (10 x 8 x 5 cm) multilocular cyst anterior to sacrum and posterior to rectum, with no solid or calcified components. Laboratory tests for complete blood count, urea, creatinine, electrolytes, liver function tests, alpha fetoprotein and Beta HCG were normal. At operation a huge cystic lesion was found occupying the retro-rectal space and adherent to coccyx. Complete excision along with the coccyx was performed (Fig.1). Postoperative course was uneventful. Gross histopathological examination showed a cyst measuring 8 x 5 x 1 cms multi-loculated in nature, it was filled with serous fluid and had smooth lining. Microscopically, the cysts were lines by ciliated pseudo-stratified low columnar to cuboidal epithelium. Bundles of smooth muscle were found adjacent to the cyst, which was not organized in a circular and longitudinal fashion and it also lacked a myenteric plexus. There was no evidence of malignancy or immature elements (Fig. 2,3)

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* Resident
** Chief Resident
*** Consultant
**** Consultant in Pathology
Department of Surgery
Salmaniya Medical Complex
Kingdom of Bahrain

Figure 1. Intraoperative finding

Figure 2. Low power view showing the cyst wall with a simple lining (H & E x 40)

Figure 3. High power view showing a lining of ciliated epithelium (H & E x 400)

DISCUSSION

The retro-rectal space is bounded by rectum anteriorly, the sacrum posteriorly, the peritoneal reflection superiorly and the leavators ani and coccygeus muscles inferiorly. The ureteres and iliac vessels are the lateral margins.

The embryology of the retro-rectal space is complex and all three grm layers play part in the development of the structures of this end of the embryo. The cloacal membrane becomes ventral and bounds a distal recess of the hind gut known as the tailgut (post anal gut) during the fourth week of the primitive gut development. This tail is maximally developed around the 8-mm stage (35 days gestational age) and usually completely regresses by the 35-mm stage (56 days gestational age). The anus is formed cephalad to the tail. Because the primitive gut extends into the tail beyond the

point at which anus develops, it is called the tail gut or post anal gut. Remnants of the tail gut gives rise to congenital cysts[3,4].

The largest series was presented in a paper by Hjermastad and Helwig[3] which shows that the condition is extremely rare and most description are in case reports. The age ranged from 4 days to 73 years, with an average age of 35 years. Most of the cases are females, of caucasian origin. Half of these cases are symptomatic in terms of anal or low back pain, painless rectal bleeding, recurrent pilonidal sinus, urinary frequency and retention of urine. Most of the asymptomatic patients were detected on routine physical examination. The lesion is an extrinsic cystic fluctuant mass that may appear either mobile or fixed. A postanal funnel-shaped dimple is stressed as an indicator of an underlying tailgut cyst[5,6].

Results of radiographics studies (Barium enema, CT and MRI) showed a posterior extrinsic mass compressing the rectum and spina bifida occulta was detected in few cases[3,7,8]. The multicystic nature is not specific. The presence of calcification favours the diagnosis of teratoma. It is not generally a feature of tailgut cyst although it was reported in association with a tailgut cyst harboring malignancy[9].

The presence of ciliated epithelium should not be considered as evidence of respiratory epithelium and therefore teratomatous. In fact, it is ordinarily found in fetal and neonatal gastrointestinal tract. The lack of ectodermal differentiation and the presence of gastrointestinal type epithelial lined cyst favours the diagnosis of tailgut cyst over teratoma, while the absence of a well organised muscle coat and the lack of myenteric plexus favours the diagnosis of a tailgut cyst over a duplication cyst[3].

Differential diagnosis from which cysts should be distinguished include: teratomas (which always contain elements of three germ cell layers), dermoid cysts (which contain hair and other dermal appendages), duplication cyst (lined with intestinal epithelium with characteristic villi and cryps, contains well developed layers of smooth muscle, a myoenteric plexus and a serosa), anal glands cysts (which is thought to be acquired lesions located closer to the anal sphincter) and chordomas of the sacrum (which are malignant and arise from embryonic remains of the notochord)[10].

Late occurance of carcinoma in tailgut cyst is rare and can occur at any age in adults; only a few cases have been described[11,12,13].

We reviewed 36 literature reports of tailgut cyst since 1966 to February 2000 and we found it a very rare condition. Only one case was reported in a newborn[14].

CONCLUSION

Complete surgical excision is the treatment of choice for all tailgut cysts. In most cases total excision is best done by a posterior approach. An abdominosacral approach is advised for large cysts.

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