

## Cutaneous Ciliated Cyst Over Knee

Sir,

Cutaneous ciliated cysts are very unusual benign lesions exclusively occurring on the lower extremity of young females shortly after puberty.<sup>[1]</sup> They have been widely regarded as Müllerian heterotopias because of the morphological similarity of the cyst lining cells to the epithelium of fallopian tubes.<sup>[2]</sup> We report a case of an 18-year-old female presented in orthopaedic outpatient department with gradually increasing, painless swelling, over right knee joint since 4 years. On examination, a soft to cystic, solitary, movable, nontender, fluctuant swelling, measuring 4 × 4 cm in size was noted. There was no abnormality of the overlying skin. Ultrasonography confirmed the cystic nature of the lesion. There was no continuation between the cyst and knee joint. Surgical removal of the cyst was carried out under local anaesthesia. Grossly, we received a skin covered, cystic specimen measuring 3.9 × 3.5 × 3.0 cm in size. On cutting open, an uniloculated cyst, measuring 3 cm in diameter was identified, which contained serous fluid. The cyst

wall was thin, smooth, and greyish white in colour. Light microscopy revealed a cyst in the deep dermis, which was predominantly lined by single layer of ciliated cuboidal to columnar cells [Figure 1]. At places the lining epithelium showed stratification and squamous metaplasia [Figure 2]. These lining epithelial cells did not contain mucin. The cyst wall was thin, fibrocollagenous without any inflammatory infiltrate. On immunohistochemical staining, the lining epithelial cells showed strong membrane positivity for epithelial membrane antigen and cytoplasmic positivity for cytokeratin (PanCK AE1/AE3) [Figure 3]. Strong nuclear positivity for oestrogen receptor (ER) and progesterone receptor (PR) was noted within the epithelial cells [Figure 4]. Immunohistochemical staining for carcinoembryonic antigen (CEA) and S-100 was negative.

Cutaneous cysts are rare benign lesions, lined by a simple cuboidal to columnar ciliated epithelium, seen typically on lower extremity in females in the second or third decade of life.<sup>[3]</sup> One case has been reported in a 51-year-old postmenopausal female patient.<sup>[4]</sup> Ciliated cutaneous cysts have also been reported at unusual sites, such as

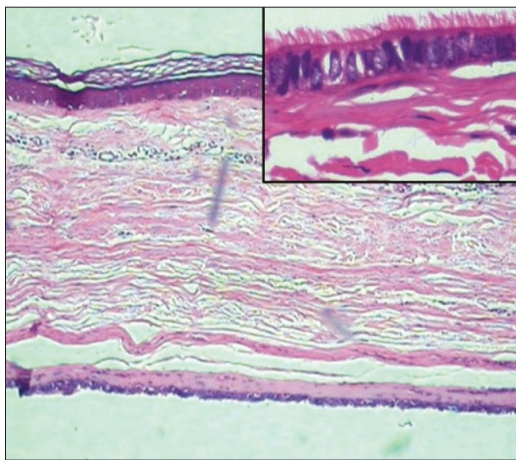


Figure 1: Photomicrograph showing a thin-walled cyst in the deep dermis lined by ciliated cuboidal to columnar cells (H and E, ×40). Inset: closer view of the ciliated epithelium (H and E, ×400)

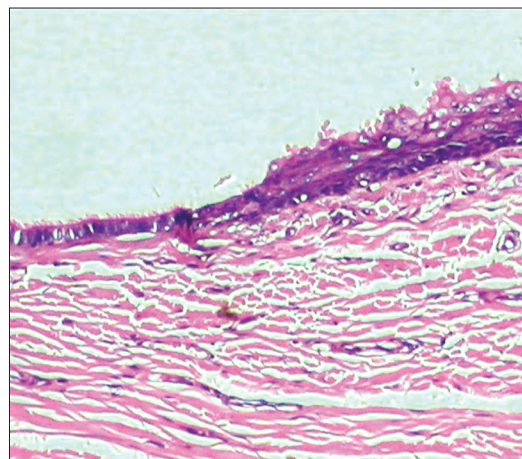


Figure 2: Focal squamous metaplasia of the lining epithelium of cyst (H and E, ×100)

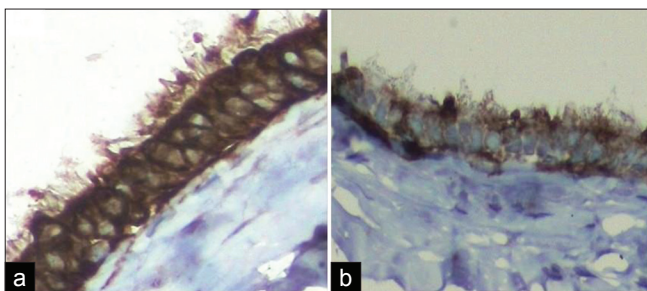


Figure 3: Lining epithelial cells showing (a) cytoplasmic positivity for cytokeratin (×100) and (b) strong membrane positivity for epithelial membrane antigen

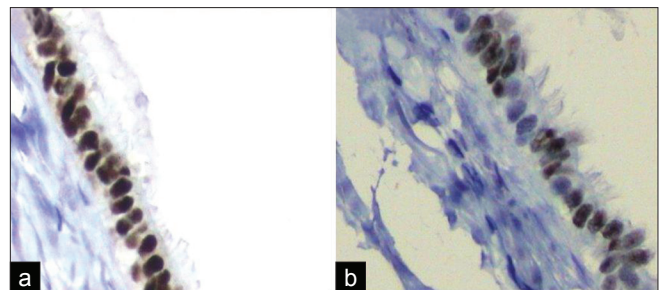


Figure 4: Lining epithelial cells showing nuclear positivity for (a) progesterone receptor and (b) oestrogen receptor (×100)

**Table 1: Summary of the previous reports on cutaneous ciliated cysts**

Previous reports	Age and sex of the patient	Site of the cutaneous ciliated cyst
Fontaine <i>et al</i> <sup>[1]</sup>	14-Year-old female	Abdominal wall
Farmer <i>et al</i> <sup>[3]</sup>	11 Females of 2 <sup>nd</sup> -3 <sup>rd</sup> decade	Lower extremity
Torisu-Itakura <i>et al</i> <sup>[4]</sup>	51-Year-old female	Leg
Businger <i>et al</i> <sup>[5]</sup>	54-Year-old female	In the posterior mediastinum
Ashton <i>et al</i> <sup>[6]</sup>	19-Year-old male	leg
Yokozaki <i>et al</i> <sup>[7]</sup>	23-Year-old female	Right leg
Tachibana <i>et al</i> <sup>[8]</sup>	19-Year-old female	buttock
Ohba <i>et al</i> <sup>[9]</sup>	Male	on the cheek
Santos <i>et al</i> <sup>[10]</sup>	Male	Perianal region
Valcárcel <i>et al</i> <sup>[11]</sup>	15-Year-old male	Scrotal skin ciliated cyst
Chong <i>et al</i> <sup>[12]</sup>	16-Year-old female	Leg
Dini <i>et al</i> <sup>[13]</sup>	12-Year-old female	Perineal region

abdominal wall and posterior mediastinum.<sup>[1,5]</sup> Cases of cutaneous ciliated cysts in males in perianal and inguinal area have also been documented in the literature.<sup>[6]</sup> In our case, the patient was an 18-year-old female with a cyst over right knee joint. Table 1 summarises the previous reports of cutaneous ciliated cysts highlighting the age and sex of the patient with their sites.

On light microscopy, the cysts are uniloculated and are lined by ciliated cuboidal to columnar epithelium without mucous cells, morphologically similar to the epithelium of fallopian tubes.<sup>[2]</sup> Immunohistochemical staining for PR, ER, cytokeratin, and epithelial membrane antigen were positive, whereas it was negative for CEA, which supports the theory of heterotopia of the ciliated epithelium from the Müllerian epithelium in its histopathogenesis.<sup>[12]</sup> The positivity of lining epithelium for ER and PR and occurrence of this lesion in second decade after puberty also suggests Müllerian origin. This supports the hypothesis, which suggests that the cells from the fimbrial ends of the fallopian tubes developing from the Müllerian ducts possibly detach and become incorporated into the lateral mesoderm where the lower limb buds arise. These arrested cells then remain dormant until puberty after which, under the influence of ovarian hormone stimulation, the heterotopic Müllerian epithelium produces serous fluid, resulting in cystic formation.<sup>[2]</sup> Ciliated metaplasia of eccrine or apocrine duct epithelial is another hypothesis documented for histogenesis of ciliated cyst, which explains rare occurrence of cutaneous ciliated cysts in male.<sup>[14,11]</sup> There is marked similarity between the cutaneous ciliated cyst lining and normal salpingeal epithelium in the mode of staining for dynein.<sup>[13]</sup>

This lesion shares its cutaneous origin with other cutaneous cysts, such as bronchogenic cyst and thyroglossal cyst.<sup>[14]</sup> However, the location of lower extremity, absence of mucous glands, cartilage and inflammation, positivity for ER and negativity for CEA establishes the diagnosis of cutaneous ciliated cyst.

Surgical removal under local anaesthesia is the recommended treatment for cutaneous ciliated cyst.<sup>[4]</sup> The recurrence has not been reported in the literature. In the present case also we followed-up the patient for 6 months without any signs of recurrence.

In conclusion, although rare, the possibility of ciliated cutaneous cyst should be considered in a young female presenting with cystic lesion on lower extremity because of its distinct Müllerian histogenesis.

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