

# World Journal of *Clinical Cases*

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## Cutaneous ciliated cyst on the anterior neck in young women: A case report

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### Abstract

#### BACKGROUND

A cutaneous ciliated cyst (CCC) is a rare, benign tumor in young female adults, which is usually found on the lower extremities.

#### CASE SUMMARY

We found an uncommon location of CCC in the anterolateral cervical area and reviewed the literature. A 20-year-old female complained of a well-defined, painless, palpable mass that started several years ago. The mass was tense and movable and located at the anterolateral aspect of the neck. Imaging showed a non-enhancing round mass. Surgical excision biopsy was performed, and the cystic mass was revealed to be a CCC.

#### CONCLUSION

The rare location of CCC can be found in anterior neck area, which should be another diagnostic option for mass on anterior neck.

**Key Words:** Head and neck neoplasms; Female; Young adult; Subcutaneous mass; Mixed tumor; Mullerian; Case report

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**Core Tip:** Cutaneous ciliated cysts are benign tumor and commonly found on lower extremities of young female adults. This case shows a rare location of cutaneous ciliated cyst found on anterior neck area, which can be another diagnostic option for evaluating anterior neck mass.

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## INTRODUCTION

Morphologic evaluation or physical examination is helpful in the differential diagnosis of benign skin lesions in the cervical region. However, if the lesion is located deep in the subcutaneous area and does not have pathognomonic findings, such as central punctum of the epidermoid cyst, the differential diagnosis can be challenging and requires further imaging modalities. Otherwise, the location of the mass can be associated with its origin. Congenital neck masses can be classified by their location. Thyroglossal duct cyst, cervical clefts, and teratomas are mostly located at the midline, and branchial cleft anomalies, lymph nodes, and thyroid lesions are mostly located in the lateral neck<sup>[1]</sup>.

Cutaneous ciliated cyst (CCC) was first reported in 1978 by Farmer and Helwig. It is a rare benign tumor of Mullerian heterotopias, and it has been reported as frequent in young females after puberty<sup>[2]</sup>. The most common location is the lower extremities. There have been serial case reports that reported unusual locations of CCC, such as the back<sup>[2]</sup>, abdominal wall<sup>[3]</sup>, or scalp<sup>[4]</sup>. It rarely occurs in male patients, and the locations reported are in the perineum, inguinal, and shoulder areas.

To our knowledge, there are limited reports of CCC that develop in the anterolateral neck area. We report our experience and summarize previous literature.

## CASE PRESENTATION

### Chief complaints

A 20-year-old female patient visited the clinic with a palpable nodule in her right anterior neck area.

### History of present illness

The mass was first noticed a few years previous, and she could barely remember, but it had not grown since then.

### History of past illness, personal and family history

She did not have any specific medical history, including congenital anomaly. However, she had a family history of papillary thyroid cancer of her father.

### Physical examination

The mass was located in the subcutaneous tissue of the mediolateral area of the anterior neck, close to the right head portion of the clavicle. It was movable and tense and not accompanied by pain, fever, or redness. No specific abnormality was seen on her thyroid gland and midline cervical area.

### Laboratory examinations

Ultrasonography showed a well-demarcated low echoic round mass, measuring 2.2 cm × 1.5 cm. The complete blood count showed that the white blood cell count was 4500/mm<sup>3</sup>, hemoglobin 13.9 g/dL, hematocrit 42.8%, and platelet count 264000 /μL. Neutrophil showed 55.82% in differential count, and lymphocyte showed 32.45%, monocyte 9.32%, eosinophil 1.70%, and basophil 0.71%. Blood chemistry results, including aspartate aminotransferase, alanine aminotransferase, total bilirubin, alkaline phosphatase, uric acid, gamma guanosine triphosphate, and lactate dehydrogenase, were within normal limits. Serum β-human chorionic gonadotropin detected to be less than 1.20 mIU/mL.

### Imaging examinations

Computed tomography (CT) was performed for the differential diagnosis and spatial evaluation. Enhanced CT imaging showed a non-enhancing well-defined ovoid mass in the right paramidline supraclavicular fossa that measured 2.2 cm × 1 cm × 1.5 cm



(Figure 1A and B), and the differential diagnosis from the image was dermoid or epidermoid cyst.

## FINAL DIAGNOSIS

She had undergone an excisional biopsy for therapeutic and diagnostic purposes. In the gross examination, the lesion was an ovoid-shaped, solid, soft mass, and thin fibrous capsules surrounded the entire lesion. The microscopic findings revealed a well-defined and thin-walled, ovoid, unilocular cystic lesion lined by ciliated pseudostratified cuboidal to columnar epithelium showing focal intraluminal papillary projections in the subcutaneous tissue. The luminal surface of the cystic lesion was filled with amorphous proteinous materials without keratinous and solid mass-like lesion. The outer surface of the cystic lesion was completely covered with thin collagenous tissue without smooth muscle bundles. It also showed focal chronic inflammatory change with no lymphoid follicles or other components. The peritumoral subcutaneous tissue revealed no thyroid, cartilaginous, or skin appendageal tissue (Figure 2).

## TREATMENT

After complete excision of the lesion, there was no development of seroma or wound infection.

## OUTCOME AND FOLLOW-UP

After six months of follow-up, there was no evidence of local recurrence.

## DISCUSSION

The location of CCC is mostly around the perineum or lower extremities, and is also reported in the abdominal wall, back, fingertips, and scalp. We reviewed the literature available of full-text files using PubMed search between 1978 and 2019 (Table 1). Most of the clinical features were palpable cystic masses remaining indolent for months to decades, except one report of a rapid growth two months after two years from its first notice<sup>[5]</sup>. The case reports included patients of various races, including African, Caucasian, and Asian. When the mass was ruptured during operation, the cystic contents showed clear serous or yellowish fluid materials. Limited evidence of long-term follow-up was found. Ross *et al.*<sup>[6]</sup> reported a 36-year-old female patient with CCC on the dorsal aspect of the foot, found indolent since the patient's late teens. Santos *et al.*<sup>[7]</sup> reported a 35-year-old male patient who had CCC in his right perineum; they reported no evidence of recurrence or malignant change during follow-up.

CCC developed in male patients was found in a few reports<sup>[7-11]</sup>; the locations were the scalp, perineum, and back. There has been a debate whether it is a persistent Mullerian cyst or ciliated metaplasia in the eccrine cyst lining<sup>[12]</sup>. Because CCC has been regarded as Mullerian in origin, in addition to the morphologic evaluation under hematoxylin and eosin stain, there were reports of the use of estrogen receptor (ER), progesterone receptor, WT1, or PAX-8 immunostaining<sup>[13]</sup>. Like other studies, the results of the immunohistochemical staining for ER, S-100, and desmin in this patient were negative<sup>[9]</sup>. The choice of the diagnostic modality of a neck mass is sometimes challenging, especially if the mass is cystic nature. Cystic lesions on the lateral neck in young adult can have malignant potential, such as metastatic thyroid cancer<sup>[14,15]</sup>. Fine needle aspiration cytology (FNAC) can be useful in solid lesions or metastatic thyroid cancer, but the diagnostic yield might not be sufficient for a definite diagnosis in cystic masses. Moreover, FNAC can be performed for the diagnosis of thyroglossal duct cysts<sup>[16]</sup> or epidermal cysts. However, it might cause an infection that interferes with the subsequent surgical procedure.

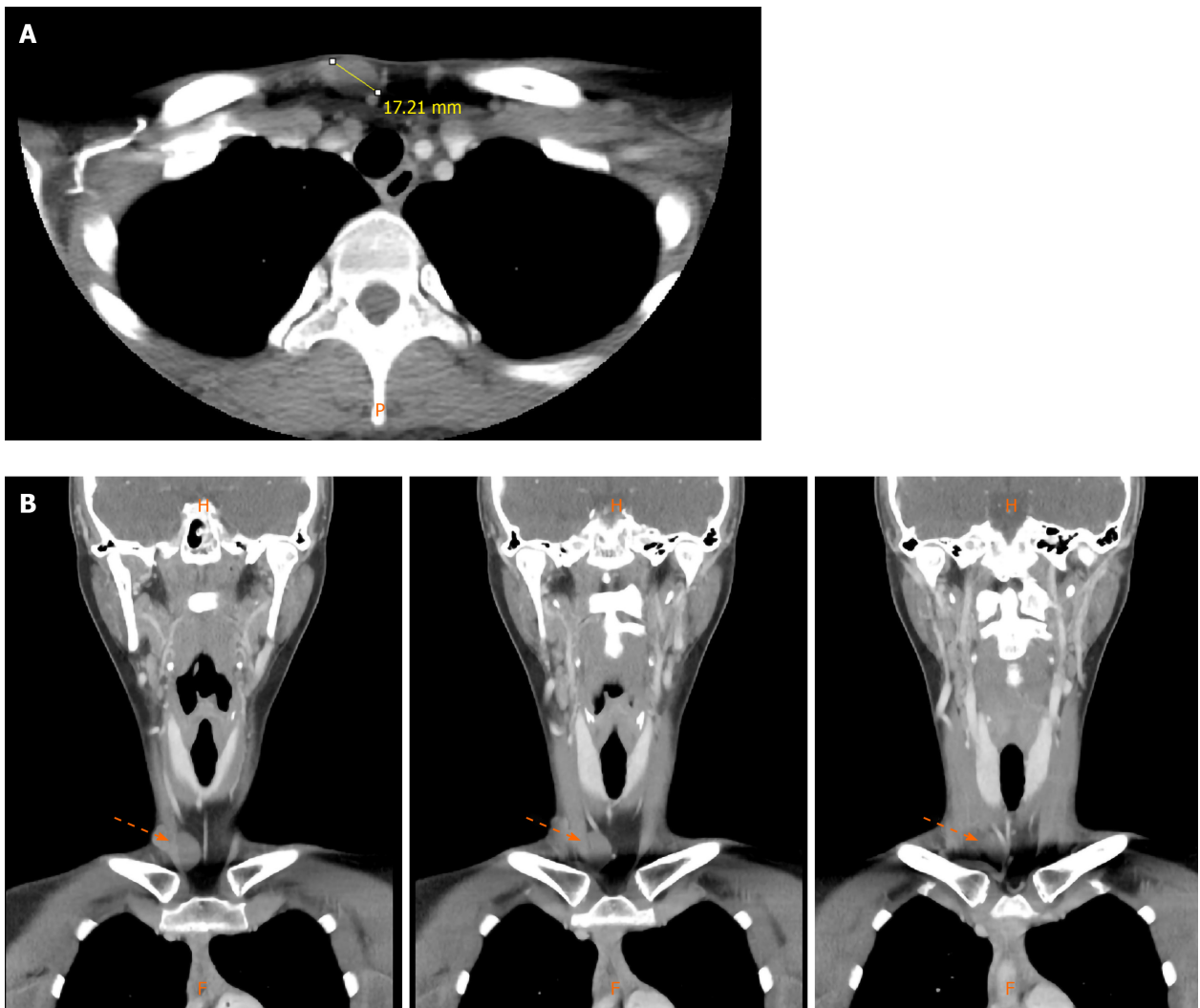
**Table 1 Literature review in clinical features of cutaneous ciliated cyst**

Ref.	Sex (Age, yr)	Race	Clinical features of location, size and duration	Outcome
Park <i>et al</i> <sup>[17]</sup> , 1982	F (15)	NA	Upper half of right thigh, 2 cm, no increase in size over 7 mo	NA
Ross <i>et al</i> <sup>[6]</sup> , 1983	F (36)	African	Dorsal aspect of left foot, 3.5 cm, noticeable at late teens	No chance since late teens
al-Nafussi <i>et al</i> <sup>[18]</sup> , 1990	F (42)	NA	A 4.5 cm painless mass with no increase in size over 2 years	NA
Sickel <i>et al</i> <sup>[19]</sup> , 1994	F (20)	African	4 cm mass lasted over 2 years	NA
Trotter <i>et al</i> <sup>[8]</sup> , 1994	M (28)	Caucasian	dorsal aspect of left foot, 3 cm	NA
Ashton <i>et al</i> <sup>[9]</sup> , 1995	M (27)	NA	Sole of right foot 2 cm, 2 years	NA
Tachibana <i>et al</i> <sup>[5]</sup> , 1995	F (19)	Asian	Bottock, 5.5cm, 2 years	NA
Yokozaki <i>et al</i> <sup>[20]</sup> , 1999	F (23)	Asian	Right lower leg, 2.5 cm, growing during 12 months	NA
Dini <i>et al</i> <sup>[21]</sup> , 2000	F (12)	Caucasian	Sacrococcygeal area, 1.4 cm, enlarged slightly over 1 month	NA
Lee <i>et al</i> <sup>[22]</sup> , 2001	F (13)	NA	Midline coccygeal lesion, 2.5 cm	NA
Fontaine <i>et al</i> <sup>[3]</sup> , 2002	F (14)	NA	Right lower abdomen, 2 cm, 3 months, growing mass	NA
Ohba <i>et al</i> <sup>[10]</sup> , 2002	M (53)	NA	2 cm, 2 years of history	NA
Vadmal <i>et al</i> <sup>[23]</sup> , 2002	F (18)	NA	left flank, 0.7 cm, 6-7 months duration	NA
Santos <i>et al</i> <sup>[7]</sup> , 2004	M (35)	Asian	Right perineum, 3.5 cm	No evidence of recurrence
Kim <i>et al</i> <sup>[24]</sup> , 2006	F (41)	NA	1.5 cm, slowly growing during 3 mo	NA
Lee <i>et al</i> <sup>[11]</sup> , 2006	M (56)	NA	Right inguinal area, 1.5 cm, 3 years	NA
Chong <i>et al</i> <sup>[25]</sup> , 2006	F (16)	NA	1 year duration of painless nodule, increasing in size	NA
Torisu <i>et al</i> <sup>[26]</sup> , 2008	F (51)	Asian	Posterior aspect of left leg, gradually increase during 2 years	NA
Bivin <i>et al</i> <sup>[2]</sup> , 2010	F (13)	African	Right leg, 2 cm, slowly growing over 2 years	NA
Ashturkar <i>et al</i> <sup>[27]</sup> , 2011	F (18)	NA	Right knee, 3 cm, gradually increasing mass during 4 years	NA
Gelincik <i>et al</i> <sup>[28]</sup> , 2011	F (25)	NA	Subcutaneous area, 2 years	NA
Hung <i>et al</i> <sup>[29]</sup> , 2012	F (16)	NA	Left thumb, 1.1 cm	NA
Kavishwar <i>et al</i> <sup>[30]</sup> , 2014	F (38)	NA	Right popliteal fossa, 3 cm	NA
Oh <i>et al</i> <sup>[31]</sup> , 2014	F (13)	Caucasian	3 cm	NA
Reserva <i>et al</i> <sup>[4]</sup> , 2014	F (53)	African	Vertex mass, lifelong history	NA
Keisling <i>et al</i> <sup>[32]</sup> , 2015	F (14)	NA	Right lower leg, 2 cm	NA
Kim <i>et al</i> <sup>[12]</sup> , 2015	M (7)	NA	Left shoulder, 1 cm, 3 years	NA
Swarbrick <i>et al</i> <sup>[33]</sup> , 2015	M (14)	NA	Scrotal cyst	NA
Fabien-Dupuis <i>et al</i> <sup>[13]</sup> , 2016	F (16)	NA	Right thigh, 2.5 cm, 3 years	NA
Doğan <i>et al</i> <sup>[34]</sup> , 2018	F (13)	NA	Between scapula, 2 cm, few months	NA
Orleans <i>et al</i> <sup>[35]</sup> , 2019	F (37)	African	Mid back, 3.5 cm, years of history	NA

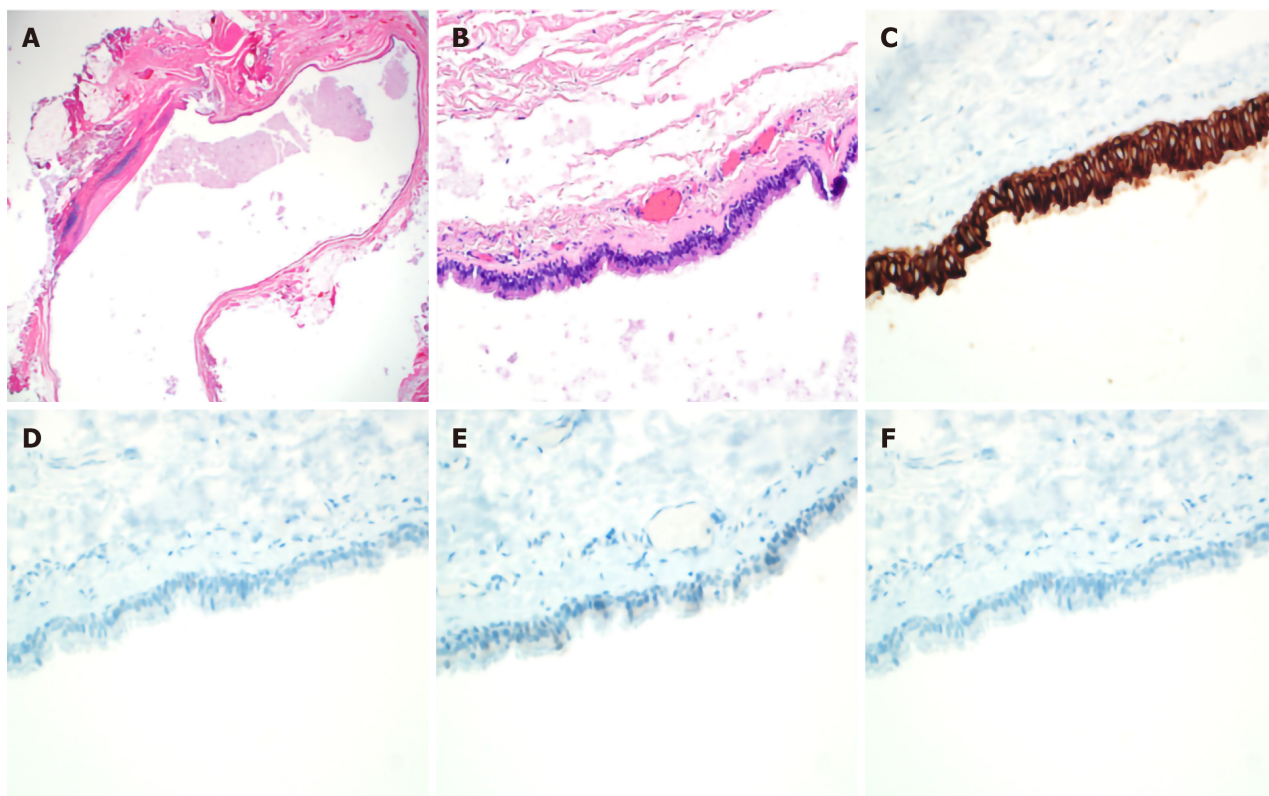
NA: Not available.

## CONCLUSION

The rare location of this case should be helpful to diagnostic decision of masses on the lateral neck in young female adults.



**Figure 1** Enhanced computed tomography of the neck. A: Non-enhancing round mass on the subcutaneous area in the anterior neck area in axial view; B: Serial images of coronal section.



**Figure 2** Pathologic findings reveal a well-defined ovoid unilocular cystic lesion lined by ciliated pseudostratified cuboidal to columnar epithelium. A and B: Focal intraluminal papillary projections in the subcutaneous tissue (Hematoxylin and eosin stain,  $\times 400$ ); C: The lining epithelium reveals immunoreactive cyokeratin (cytokeratin,  $\times 400$ ); D-F: It reveals negative findings for immunostainings (D: Estrogen receptor,  $\times 400$ ; E: Desmin,  $\times 400$ ; F: S-100,  $\times 400$ ).

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