Granular Cell Tumor of Female Urethra – the Second Such Reported Case

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ABSTRACT
Granular cell tumors are very uncommon tumor specially in female urethra. In the literature, there has been only one case reported so far. Here we describe our experience with this very rare entity in a case of a 26-year-old female patient with granular cell tumor of urethra with its clinical and histopathological findings.

Keywords: urethra, granular cell tumor, female.

INTRODUCTION
Granular cell tumors (GrCTs) were first described by Abrikossoff, in 1926, as myoma. They are rare, usually benign soft tissue tumors of endo-mesenchymal origin, which can occur throughout the body and in any age or race (1). They occur more often in females than males, and in blacks than whites. The peak age incidence is in the fourth through fifth decade of life. While common sites are the tongue and breast, vulvar involvement has been reported in 7-16% of cases (2). However, the occurrence of such tumors in the urinary tract is rare. In most cases, these tumors usually arise from the urinary bladder or, less commonly, from the penile soft tissue (3).

Here we report the case of a granular cell tumor of a female urethra which, to the best of our knowledge, is the second one ever reported in the world so far, after the one described by Hitoshi Yokoyama et al in 2007 (3).

CASE REPORT
A twenty six-year old female presented with the complaint of a small swelling at urethral orifice of six month duration and dysuria. On clinical examination, a 1 cm x 1 cm firm non-tender swelling was found on the right side of the urethral orifice (Figure 1). There were no other clinical findings and regional lymph nodes were not enlarged. An excision biopsy was planned. Preoperative urine microscopy and cytology were normal. The tumor was excised under regional anesthesia. Intraoperatively, the tumor was found to arise from the terminal part of the urethral wall very near the external orifice. The urethra was repaired with absorbable suture with per urethral catheter in situ. Cut section of the tumor was grayish in...
color, which was firm and irregular in appearance. Histopathology revealed spaces lined by transitional epithelium to flattened epithelium in fibrous tissue (Figure 2). At places sheets of cells having pink granular cytoplasm with ovoid to round nuclei with mild lymphocytic infiltration were present consistent with benign Granular cell neoplasm of urethra. The postoperative period was uneventful and per urethral catheter was removed after two weeks. There has been no recurrence in two years of follow up.

DISCUSSION

Soft tissue granular cell tumors are very uncommon, usually benign neoplasms that most frequently originate from the skin and oral cavity. However, these tumors can also originate from the gastrointestinal tract (esophagus, anal canal), head neck region (tongue, parotid, thyroid, lacrimal gland), breast, biliary tract and even heart (5-9). In the genitourinary tract, most cases have been reported to originate from the bladder (11) and a few from the penile shaft. But it is extremely rare in female urethra. Although, less than 2% of all granular cell tumors are malignant (12), Fanburg-Smith et al. described a series of 46 cases of malignant granular cell tumors. The authors reported that cell necrosis, high mitotic index, high Ki-67 index, spindling tumor cells, vesicles with large nucleoli and muscle invasion were features of malignant granular cell tumors. Also, they found a median tumor size of 2 cm in benign cases and 4 cm in malignant cases (13). In our case, these malignant features were absent. In benign granular cell tumors, surgical excision with clear margins is sufficient, and more radical resections are not required (7-11).

CONCLUSION

There are several differential diagnoses of swellings in female urethral orifice from urethral polyps to urethral caruncle, etc. but granular cell tumor is an extremely rare entity. Considering the fact that these tumors can give rise to high grade malignancy, albeit infrequently (~2% of cases), care should be taken to always excise these swellings in totality and obtain proper postoperative histopathological analysis. Surgeons and pathologists need to be aware of this differential diagnosis when dealing with urethral swellings in female patients.

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Authors’ contribution: Dr Manoj Kumar Chaudhary was involved in primary management of the patient, while Dr Rubik Ray, Dr Tridip Dutta Baruah and Dr. Mrinal Shankar were involved in research regarding this rare entity as well as manuscript preparation.
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References