Original Research Article

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Cytohistopathological appraisal of unusual locations of epidermoid inclusion cysts

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ABSTRACT

Background: Epidermoid inclusion cysts are common benign lesions of the pilosebaceous origin that encountered anywhere in the body. Most common sites are hairy and non hairy areas mainly scalp, back, neck and trunk. Objective of present study was to highlight and document the cytohistomorphological features of epidermoid inclusion cysts (EIC) at unusual locations of the body in our tertiary care center.

Methods: The present study is retrospective, cross sectional study done on cytohistopathological specimen over a period of 30 months from June 2016 to December 2017. A total of 82 cases of EIC were included in the study. All the sites were included in the study that is usual as well as unusual. Sebaceous and dermoid cysts were excluded from the study. Unusual sites of EIC were collected, tabulated and compared with existing literature.

Results: The most common affected site of EIC in our study was head, neck and face (42.7%) followed by extremities and axilla (29%), back (11%) and chest/abdomen (8.6%). Peak incidence of EIC was found in 31 to 40 years (34.1%) with male preponderance (1.5:1). The unusual sites encountered were oral cavity and tonsil in head neck region, sole in the extremities, breasts and testis.

Conclusions: The study highlights the importance of FNAC in early diagnosis and histopathology for confirmation and treatment purposes in epidermoid inclusion cysts. It provides prompt response to avoid cosmetic and functional disabilities in unusual locations of common lesions like EIC because the differentials may from inflammatory to malignant ones.

Keywords: Cytology, Epidemoid inclusion cysts, FNAC, Histopathology, Unusual

INTRODUCTION

Epidermal inclusion cyst (EIC) also called as epidermal cyst/epidermoid cyst are benign entity of the pilosebaceous origin can occurs anywhere in the body lined by squamous epithelium.¹ The origin of EIC is controversial but it is developmental lesion from implanted epithelium arising from trauma or surgery so the name epidermal inclusion cyst. Also think that abnormal ectodermal tissue components formed during fetal period can form EIC2. The most common

presentations of EIC are on head, neck, face and scalp, trunk and extremities.² Grossly they are smooth, round, regular unilocular swelling of varying sizes from few millimeters to centimeters without a punctum.³ The light microscopical hallmark is cyst lined by stratified squaomus epithelium with a granular layer and lumina containing abundant whitish laminated keratinous material.^{4,5}

Authors tried to highlight the unusual locations of EIC with cytohistopathological overview in this article. We

found the few unusual sites of epidermal inclusion cyst in the breasts, testis, tonsil, sole of foot and oral cavity etc. The prime and early diagnosis of EIC is as important as treatment for the benefit of the patients as the prognosis matters.

METHODS

The present study was retrospective, cross sectional, observational on cytohistopathological specimens received from the various department of our institute to Pathology department after informed consent. The study includes 83 cases of EIC diagnosed on cytology and subsequently confirmed on histopathology over a period of 2.5 years from June 2016 to Dec 2017. All the cases of EIC were included in the present study and sebaceous and dermoid cysts were excluded. The detailed history and clinicopathological records were assessed and unusual locations of EIC were highlighted, data was analyzed and tabulated.

RESULTS

Present study comprised of 82 cases of epidermoid cysts sent to our Pathology department from various department in our institute over a period of two and half years. The age group of the EIC patients ranged from 8 years to 58 years with mean of 38.1 years. The largest age group with the lesion of EIC was 31-40 years (28 cases), followed by 21-30 years (14 cases) as summarized in Table 1.

Table 1: Age group distribution of EIC cases.

| Age group | No. of cases |
|-----------|--------------|
| 0-10 | 02 |
| 11-20 | 10 |
| 21-30 | 14 |
| 31-40 | 28 |
| 41-50 | 10 |
| >51 | 18 |
| Total | 82 |

The males were outnumbered the females with overall M:F ratio of 1.5:1, as Males encountered were 50 and females were 32.

Table 2: Region wise distribution of EIC cases.

| Region | No. of cases | Percentage |
|----------------------------|--------------|------------|
| Head, neck and face | 35 | 42.7 |
| Chest and abdomen | 07 | 8.6 |
| Back | 09 | 11.0 |
| Upper extremity and axilla | 08 | 9.8 |
| Lower extremity and axilla | 16 | 19.5 |
| MGT (testis) | 01 | 1.2 |
| Breast | 02 | 2.4 |
| FGT (ovary) | 04 | 4.8 |
| Total | 82 | 100 |

In the region wise distribution of present study, the head and neck (42.7%) was the most frequently affected region followed by extremities and axilla (29%). Back (11%) and chest/abdomen (8.6%) was the after the head/neck and extremities as summarized in Table 2.

Table 3: Distribution of head, neck and face cases.

| Location | No. of cases |
|-------------|--------------|
| Eyebrow | 09 |
| Cheek | 06 |
| Scalp | 04 |
| Forehead | 05 |
| Ear/pinna | 05 |
| Oral cavity | 02 |
| Tonsil | 02 |
| Neck | 02 |
| Total | 35 |

Distribution of head neck and face patients of EIC was summarized in Table 3, as the most common site is eyebrow (09) and unusual site is tonsil (02 cases).



Figure 1: a) and b) Gross and cut section of intratonsillar EIC; c) Gross and cut section of intratesticular EIC; d) Cut section of tonsillar EIC with keratinous material in the lumen; e) Gross appearance of mobile lump in inner quadrant of right breast.

The most unusual sites observed in present study were breasts, testis, tonsil, oral cavity, left sole of foot, cheeks and eyebrows (Figure 1 a-e). Regarding morphology the size of the EIC ranged from 0.8 to 6 cms in diameter with the mean of 3.2cms. The clinical manifestation ranges from pain, foreign body sensation, hampered daily routine function and cosmetic reasons. The patients of swelling/mass with clinical features were encountered in OPDs of various departments commonly surgery, gynecology, medicine and pediatrics etc. The prime investigation was offered as fine needle aspiration

cytology (FNAC) as first diagnostic measure with clinical suspicious of inflammatory lesion as abscess or tumorous mass varied from benign to malignant. The FNAC was performed under all aseptic precautions with 23G needle in majority of accessible sites, while in some cases, authors received the excised specimen to rule of malignancy. To our and clinicians surprise, it turned out to be epidermoid inclusion cyst on cytology/histopathology. All the cytological aspirates were grey whitish and granular in EIC. The microscopical examinations were done under rapid Pap and Toluidine blue stain for prompt diagnosis.

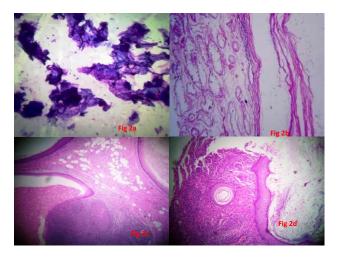


Figure 2: a) FNAC of EIC showing numerous scattered anuclated squames with keratinous material (Toluidune blue x100); b) Photomicrograph showed testicular tissue and cyst with squamous lining and abundant keratinous material in its lumina (H&E, x100); c) Chronic tonsillitis with many cysts lined by stratified squamous epithelium and keratin debris in lumen (H &E, x100); d) Photomicrograph of infected EIC with multinucleated foreign giant cells reaction (H & E, X100).

The FNA smears showed numerous sheets, clusters and scattered anucleated squames with keratinous flakes confirming the diagnosis (Figure 2a). Occasional giant cells were noted in few cases. We received the well circumscribed specimen and on c/s showed cysts filled with whitish material (Figure 1c, d). On subsequent histopathology, revealed the cysts lined by stratified squamous epithelium with a granular layer. The lumina were filled with laminated keratinous material with keratin flakes (Figure 2 b, c) At place, ruptured cyst with foreign body giant cells reaction to calcification was noted to keratinous material with dense and diffuse mononulcear cells infiltration (Figure 2d).

DISCUSSION

Epidermoid inclusion cysts are benign subcutaneous developmental cystic mass of the skin most commonly seen on head, neck and extremities.² Authors found

handful of unusual cases of EIC and studied the literature. Authors compared present findings with available literature and found it rare incidence of EIC at unusual sites. The recent study by Nigam JS et al showed concordance with present study.⁶

Regarding unusual sites on head/neck/face, we have 4 cases in oral cavity and tonsil. The EIC are usually unilocular /solitary, we found single case of intratonsillar multiple EIC in right tonsil in a 40 year old female.⁷ She presented with difficulty in swallowing and pain in ear since 1 week. On L/E -hypertrophied right tonsil was noted and clinically diagnosed as malignant mass with tonsillitis. After tonsillectomy, we received multinodular tonsillar mass measuring 3x2 cms with multiple tiny cysts on C/S (Figure 1 a, b, d) with whitish material in the lumina. Histopathologically proved as rare entity- intratonsillar multiple epidermoid inclusion cysts with chronic tonsillitis in view of many cysts lined by stratified squaomus epithelium and lumina contained keratinous material, anuclated squames and cholesterol clefts with chronic tonsillitis (Figure 2c).7 EIC of head and neck region encountered 7% of cases and in oral cavity upto 1.6% of total outcome which constitute <0.01% of all cystic lesion of oral cavity.^{8,9} We haven't found any case of multiple, intratonsillar epidermoid cyst in immunocompetant patient in India. Hence, authors published this case.⁷

Authors encounter the interesting case of epidermoid cyst of breast. It was diagnostic dilemma on cytology. Authors diagnosed two cases of EIC of right breast in 52 year10 and 48 year old female (Figure 1e). To the best of our knowledge, though various databases, only 90 cases of EIC breast were reported upto 2016. 11 Authors diagnosed and highlighted this interesting case, diagnosed on cytology and confirmed on histopathology in right breast of 52 year old female. She presented with 2.5x2 cms solitary, firm, mobile lump in LOQ of right breast. Clinically thought to be fat necrosis/mastitis and FNAC was advised. FNAC was performed by 23 G needle under all aseptic precautions. Pap and Toluidine blue stain revealed numerous sheets, clusters and scattered anuclated squames diagnosed as EIC (Figure 2a). Subsequently confirmed on histopathology. In this case, we highlighted the role of FNAC as rapid, OPD, simple, non-invasive and cost effective method in such rare benign and diagnostically challenging lesions. As the differential vary from inflammatory to benign/malignant. Though the malignant transformation in EIC is very much rare that is 0.045 to 19% authors should take precautions by early diagnosing the lesion by FNAC. 11,12

Testicular EIC are rare benign lesion constituting 1-2% of all testicular neoplasm. We encountered intratesticular EIC in 30 year old male with conservative approach with testicular preservation and avoiding unnecessary orchidectomy. ¹³ Patient presented to surgical OPD with painless right testicular swelling since 3 months. No past/personal history was contributory. Local

examination revealed and irregular hard mass of 4cms felt at lower pole of the right testis and spermatic cord was normal. Enucleation of the mass through inguinal incision was done and histopathology confirmed as EIC with foreign body giant cells reaction and calcification. Grossly, well circumscribed, encapsuluated mass measuring 3.5x2.5 cms noted. C/S –filled with whitish chalky paste like material (Figure 1c). Microscopy confirmed EIC as lamellated keratin and anuclated squames in the lumina with stratified squamous epithelial lining (Figure 2b). The cytohistopathological examination is warranted in testis to rule out teratoma as of its malignant charactersitcs 14. Thus histopathology is gold standard for intratesticular EIC as treatment modalities differs from conservative to invasive procedures. Id

Regarding meta-analysis, by Bai Sakhiya N et al noted post aural and floor of mouth as most unusual sites.¹⁵ Nigam JS et al noted sole, prepuce, finger, palm and oral cavity as unusual site in their study.⁶ Lip and mandibular region was noted as unusual sites by Mahalaxmi S et al.¹⁶ Sahoo MR et al as caecum/GIT as unusual site, whereas Thyroid EIC reported by Kulkarni SS et al and Vulva BY et al.¹⁷⁻¹⁹ In the present study authors, encountered testis, breast and tonsil, oral cavity as unusual site of epidermoid inclusion cysts.

CONCLUSION

The knowledge of this common entity EIC at uncommon sites is important in view of its recurrence, diagnostic dilemma and malignant differentiation and behavior. Early and prompt diagnosis by FNAC and biopsy confirmation by histopathology is the gold standard for therapeutic purposes to avoid invasive surgeries. Due to rapid diagnosis by cytology, functional and cosmetic impairment can be avoided.

Hence the thorough knowledge common lesion (EIC) at unusual locations with vast differential is important on clinical grounds to medical personnel for better outcomes.

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Ethical approval: The study was approved by the

Institutional Ethics Committee

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