

Original Article

Presentation and Management of Intermammary Epidermoid Cyst: A Case Series

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Abstract**Introduction**

This study aims to demonstrate the clinical presentation and treatment of epidermoid sinuses occurring in the intermammary region. They are formed by dome-shaped nodules resulting from burst pilosebaceous follicles.

Methods

The study is a retrospective multicenter case-series study. The cases were presented and treated for 12 years. It includes cases with a ruptured epidermoid cyst. The cases were managed through excision and primary closure of the lesion.

Results

The case series included 19 cases; 18 (95%) patients were female, and the age ranged between 15 to 30 years. All of the patients were obese. All the female cases had hyperpigmented patches in the intermammary region. All of them complained of discharge from the intermammary region for a prolonged period, ranging from 6 weeks to 2.5 years. Two cases (10.5%) developed recurrence and were managed by re-excision and primary closure.

Conclusion

The intermammary epidermoid cyst is a very rare form of the disease. The most effective management strategy is wide local excision with primary closure.

1. Introduction

Epidermoid, keratin, or sebaceous cysts are dome-shaped nodules that develop from burst pilosebaceous follicles. It appears on shoulders or back, face, and neck, or wherever

sebaceous glands are found [1]. It is regarded as a kind of adnexal tumor of pilosebaceous origin. The epidermoid cyst can be one of the three major subtypes: (1) Epidermal inclusion cyst (EIC) or infundibular cyst is lined by keratinizing stratified squamous epithelium with a granular layer; (2) trichilemmal cyst is characterized by abrupt keratinization and is lined by stratified squamous epithelium that does not have a granular layer; (3) sebaceous cyst has the similar lining as EIC, but it also has adnexal structures [2]. Because they surround crucial structures, diseases of the intermammary area are of particular health significance [3,4]. The appearance of epidermoid cysts in the intermammary region is a very unique condition [5]. Deep epidermal cysts of the breasts are extremely rare and are usually caused by traumatic implantation [6]. No published report addresses the presentation, management, and outcome of intermammary epidermoid cysts in genuine literature [7].

The purpose of this study is to illustrate the manifestations and treatment of intermammary epidermoid sinuses and cysts.

2. Methods

2.1. Study design

This is a multicenter retrospective case-series study. The patients were consecutively ordered.

2.2. Setting

The patients have been managed in different private, academic, and community practice settings. They were in Iraq and Egypt. The patients were presented and managed for 12 years (from March 2010 to March 2022). The average time of follow-up was years. The data collection lasted one month (from the first of February 2023 to the first of March).

2.3. Participants

The study included all patients with ruptured epidermoid cysts who have been admitted to one of the five main tertiary hospitals and whose diagnoses were confirmed by pathological examination of the resected specimens. The clinical and sociodemographic information was obtained from the patients, patient's medical records, and health care providers.

2.4. Pre-intervention considerations

All patients underwent planning and investigations for general anesthesia. Preoperatively, the patients were assessed for a chest x-ray (posteroanterior and lateral views), vital signs, and level of hemoglobin (complete blood counts).

2.5. Types of intervention(s) deployed

All surgeries were performed in the supine position under local anesthesia. Before the operation, the patients were kept fasting for at least eight hours and injected with broad-spectrum antibiotics (ceftriaxone vial 1gm i.v).

2.6. Peri-intervention considerations

At the time of the intervention, continuous electrocardiography (ECG) monitoring was used to monitor the patients. According

to the vital signs, intravenous fluid in the form of crystalloid was infused. Wide local excision and primary closure were done. An open drain was left.

2.7. Post-intervention considerations

All patients were advised to receive oral antibiotics and analgesics at least for 5 days. The drains were detached after 5 days. The diagnosis of the disease has been approved by the results of the pathological examination of the specimens.

3. Results

3.1. Participants

The case series included 19 patients, 18 (95%) of them were female, ranging in age from 15 to 30 years, with a mean age of 24 years. All of the patients were obese, with a body mass index (BMI) ranging from 31 to 35 kg/m² and a mean BMI of 32 kg/m². All the female cases had hyperpigmented regions between the two breasts, and all of the female cases had large breasts and used tight bra (Figure 1). They lacked intermammary hairs. All of them complained of discharge from the intermammary region for a long time, ranging from 6 weeks to 2.5 years, with a mean duration of presentation of 15 months. Two cases (10.5%) developed recurrence and were managed by re-excision and primary closure.



Figure 1. Intermammary epidermoid cyst in a patient with large breast size and history of tight bra

3.2. Outcomes and follow-up

Within two months of follow-up, 15 cases (78.9%) were cured of the condition. Four cases (21.1%) recurred. All four cases underwent re-operation, which included wide local excision with primary closure.

3.3. Complications and adverse or unanticipated events

Three hours after the surgery, two of the patients started to bleed, which was treated by packing and dressing. No other crucial complication developed.

4. Discussion

Sebaceous cysts sporadically develop inflammation and become fluctuant swellings encircled by erythematous skin and encompassing purulent material [8].

Sebaceous cysts could be categorized as acquired or congenital, although their presentations and pathological features are the same. There are several proposed etiopathogenetic theories. The most prevalent one is embryonal dysgenesis. According to this theory, congenital lesions are dysembryogenetic cysts that emanate from ectodermic entanglements at the time of fusion of the 1st and 2nd branchial arches during the first month of intrauterine life. Acquired cysts are derived from iatrogenic or traumatic inclusion of the epithelium or the blockage of the gland duct. Other investigators suggested that midline epidermoid cysts could be a variant of thyroglossal cysts [9]. Sebaceous cysts of the breast originating from fibrocystic change have been presented and discussed previously [5] The largest and oldest series belonged to Menville in 1936, in which, among three thousand specimens, 36 cases of breast epidermoid cysts were found, and seven (19%) of them were observed to be associated with malignancy. Up to date, no reports have been found in the literature reporting intermammary epidermoid cysts.

The main clinical feature of an epidermoid cyst is a constant discharge from the sinuses, along with occasional pain and tenderness. Clinical examination is the primary method of diagnosis, though fine needle aspiration cytology is occasionally required [5,8]. These features make it difficult to differentiate from pilonidal sinus [10]. The hallmark of the latter is the presence of foreign-body granuloma [11]. All of the current cases presented with chronically discharged sinuses. They have been diagnosed by history and examination and confirmed by histopathological inspection of the specimens.

Generally, a consensus has been reached to prescribe antibiotics combined with excision and drainage as a first line of management. When the inflammation subsided, elective excision and primary closure were recommended. Khafif and his colleagues, as well as Kitamura et al., have revealed gratifying results from one resection of infected epidermoid cysts followed by incision lavage and primary closure without preoperative surgical drainage [12,13]. Jun and his associates published a report containing 351 cases of inflamed epidermoid cysts. They compared the outcomes of the classical (drainage followed by excision and primary closure) and new (one-stage excision, evacuation, and primary closure) approaches to management. They concluded that non-classical management decreased the need for antibiotics by 50%, reduced the time of wound healing, and was cost-effective. In contrast, omitting drainage and direct excision and closure is not praised by Goldstein and Goldstein, as the inflamed edge is brittle and therefore impossible to eliminate, leading to a greater recurrence rate. In addition, there is a greater risk of perioperative hemorrhage which lengthens the duration of the operation, and primary surgical excision with direct closure results in greater ugly scars.

This is despite the higher risk of recurrence in comparison to a two-stage operation (drainage, followed by delayed excision, and primary closure) (6.4% versus 1.6%) [14]. Another conceivable management strategy, chronicled by Miyata and his colleague, is delayed primary closure, in which initial

debridement would be followed by primary closure within 7–10 days [15]. Minimal excision technique and laser marsupialization have been described for the management of epidermoid cysts but the risk and outcomes are not well defined [16,17]. In the current study, all the patients were managed by excision and primary closure. Previous reports regarding intermammary dermatological problems showed that wide local excision and primary closure of the intermammary area are quite acceptable, with pleasant outcomes both clinically and cosmetically [17,18]. There was a 10.5% recurrence rate, which is relatively higher than the recurrence of epidermoid cysts in other areas of the body. This may be explained by the larger breast size in the current cases and the nature of the intermammary region. Malignant transformation is a rare complication of an epidermoid cyst [2]. Menville and his colleagues reported 7 cases of sebaceous cysts with malignant conversion among 36 cases of epidermoid cysts [19].

5. Conclusion

Although the epidermoid cyst is the most common cyst in the skin, the intermediate epidermoid cyst is a very uncommon form of the disease with a benign clinical course. It presents with chronic discharging sinus. The most effective management strategy is wide local surgical excision with primary closure.

Declarations

Conflicts of interest: The author(s) have no conflicts of interest to disclose.

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Patient consent (participation and publication): Not applicable.

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Authors' contributions: RSA, and AMS were a major contributor to the conception of the study, as well as to the literature search for related studies, and participated in data collection. LRAP was involved in the literature review and designed the study. MLE and WZ participated in preparing the manuscript. MS, HOA, and YAS critically revised the manuscript and the processing of the figures. WMS and WZ confirmed the authenticity of the data. All authors approved the final version of the manuscript.

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