International Surgery Journal
Save S et al. Int Surg J. 2024 Nov;11(11):1896-1899
http://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902

DOI: https://dx.doi.org/10.18203/2349-2902.isj20243252

Case Report

A large long standing keratinous cyst on the cheek: a case report

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Received: 27 August 2024
Accepted: 03 October 2024

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ABSTRACT

Epidermal inclusion cysts or keratinous cysts can be congenital or acquired. These slow-growing, chronic cysts can sometimes be neglected, unless they affect cosmesis or present the fear of turning malignant. We present a case of a keratinous cyst on a patient's cheek, overlooked for many years, which slowly and steadily kept on growing and finally presenting as a disfiguring ovoid swelling over the patient's cheek. Epithelial inclusion cysts have a tendency of slow but steady growth, with a rare of real risk of malignant change. Secondary infection, abscess formation, pressure effect and cosmetic effect are a few complications seen in such cases. Improper excision of these cysts leads to recurrence due to left over residual cyst tissue. This unearths the importance of timely investigation and treatment of swellings that we might at first assume to be insignificant.

Keywords: Otolaryngology, Cystic swelling, Keratinous cyst, Sebaceous cyst

INTRODUCTION

A variety of lesions can develop within the layers of the skin and the dermal appendages. Among such cutaneous cysts, epidermal cysts, which are retention cysts from skin appendages, are reported to be the most common type.[1,2] They originate from implanted epithelium, following trauma, surgery or localised inflammation of hair follicles or can be congenital.[1,3]

Epithelium inclusion cysts, pilar/trichilemmal cysts, dermoid cysts and steatocystoma are different types of keratinous cysts.[1] These cysts were previously documented as sebaceous cysts; however, this term is seldom used nowadays.[1] This is because chromatography studies of the lipids from these cysts show lipid patterns that resemble that of the epidermis more than sebaceous glands.[1] Other accepted terms for these cysts are epidermal cysts, epidermal inclusion cysts, keratin cysts, epithelial cysts and epidermoid cysts.[1]

These cystic masses are reported to be most commonly seen in the face, scalp, neck and trunk.[4] These cysts are usually benign however malignant degeneration can occur in some cases. This can lead to invasion of adjacent tissues or distant metastases.[1,5] Complete excisions is the usual choice of treatment and the condition can recur of there is any incomplete excision.[1,5]

CASE REPORT

A 40-year-old male patient came to the outpatient department (OPD) with complains of a swelling over his right cheek. The swelling was noticed incidentally by the patient 6 years ago however was not troublesome to him in any way. The swelling was gradually increasing in size over the years and the patient finally reported to the OPD due to the cosmetic effect of the swelling. The patient did not recollect any history of trauma over the cheek or any pain or pus discharge from the swelling. There was no sudden increase in the size of the swelling.

On examination, there was a single, ovoid swelling around 4×3×2 cm in size over the right cheek (Figure 1). There was no local rise of temperature on palpation. It was firm in consistency, mobile, non-indurated and not adherent to the surrounding structures. There was no tenderness on

palpation and the skin over swelling was normal with no pus point or scarring over the skin.

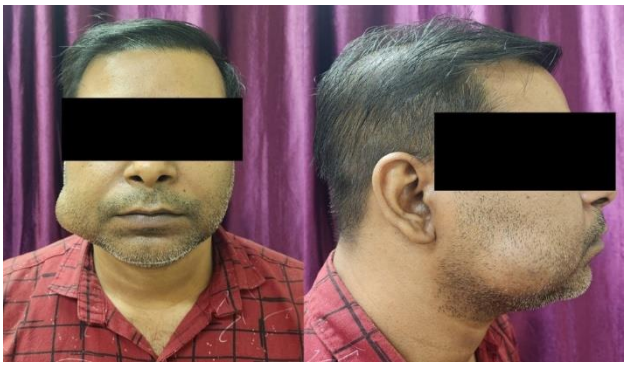

Figure 1: Patient presented with a right cheek swelling.

The patient was asked to do an ultrasonography (USG) of the swelling which showed a 4×2.3×4.1 cm well defined heterogenous hyperechoic lesion with no internal vascularity and was reported as an epidermoid cyst. Excision of the cyst was planned and for further delineation of the extension of the swelling a computed tomography (CT) scan was done. The report mentioned a thin wall, encapsulated cyst in the subcutaneous plane most likely being an epidermoid cyst. The CT further mentioned that the cyst was abutting the superficial lobe of the parotid gland (Figure 2).

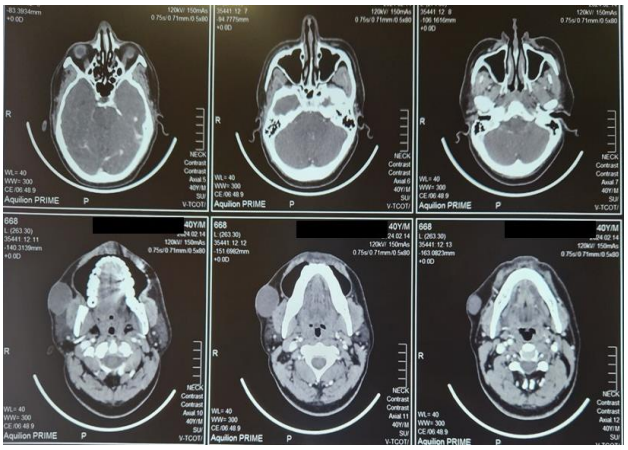

Figure 2: CT scan film showing mass in the subcutaneous plane.

Excision was the cyst was carried out under local anaesthesia (Figures 3 and 4). The cyst was removed in toto (Figure 5) and sent for histopathological examination.

The histopathology report confirmed the pre-operative diagnosis. Microscopic findings showed a cyst with ulcerated linings and the cyst wall showing granulation tissue with lymphoplasmacytic infiltration and giant cells in the cyst wall. The patient had a normal post-operative period with complete wound healing. No recurrence of the swelling was noted.

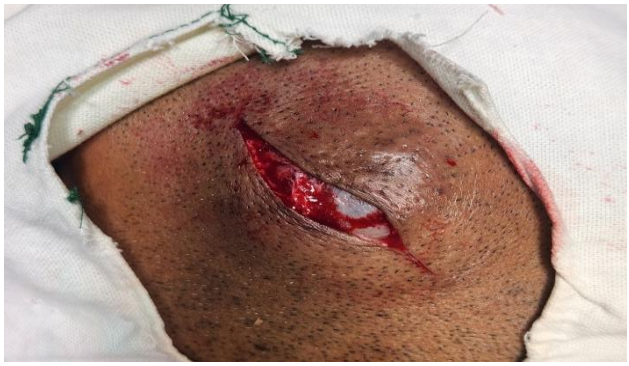

Figure 3: Capsule of swelling during excision.

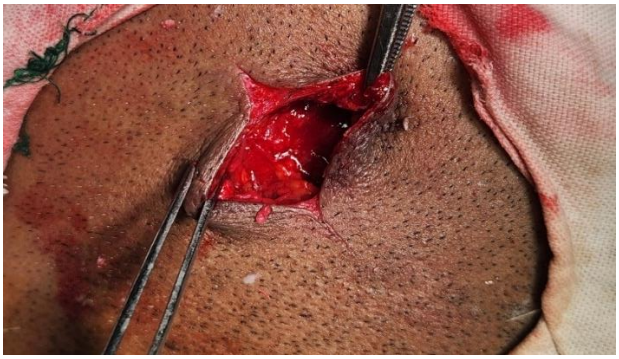

Figure 4: Cavity left behind after excision of the swelling.

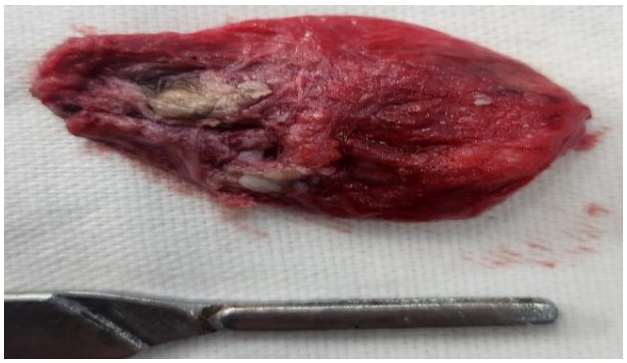

Figure 5: The excised swelling with capsule.

DISCUSSION

Invaginations and cystic expansion of the epidermis or the epithelium around the hair follicle cause keratinous cysts.[1] This cyst gets filled with lipids and keratin debris which looks like a thick cheesy material. However, in a study from an Indian population, it was found that 63% of these cysts also had melanin pigmentation.[6] Most cases present within the age group of 15-50 years and there is no sex predilection.[3] The exception is in Gardner syndrome where they occur at an average age of 13 years.[6]

These cysts can either be congenital or acquired.[1,3] Congenital cases are due to entrapment of ectoderm in lines of fusion during embryogenesis.[3,5] Another theory

that these cysts may be due to ectodermal differentiation of multipotent cells that had been pinched off during anterior neuropore closure.[4] Acquired cases are because of implanted epithelial, following trauma, surgery or localised inflammation of hair follicles.[1] Trauma is the most common cause for these cysts.[3] Human papilloma virus infection is also reported as a causative agent in occasional cases.[3] Gorlin syndrome, Gardner syndrome and Lowe syndrome are certain hereditary syndromes which commonly have keratinous cysts.[3]

They most commonly occur on skin containing hair, however, rarely can occur on the palms and soles.[3] Major sites of predilection are, face, neck, scalp and back.[3,4] On examination they are reported as firm, fluctuant and unattached to underlying tissue.[3] They have very slow growth and are usually asymptomatic until they get secondarily infected, ruptured or become large and affect nearby structures.[3,6]

When dealing with such swellings, certain differential diagnoses should be kept in mind. Many conditions may mimic such swellings such as chronic infective lesions, benign neoplastic lesions like lipoma, haemangiomas or lymphangiomas or even extremely slow growing malignant neoplasms.[5] Diagnosis is usually done by USG. However, CT scans and MRI can be used to show their cystic nature and to differentiate from other swellings.[3] Datta et al also explain that fine needle aspiration cytology (FNAC) of these lesions can also help in diagnosis.[4]

Secondary infection of these cysts can lead to abscess formation.[3] This needs drainage and antibiotics before definitive management.[2] Further, long standing cases can develop malignant changes. Squamous cell carcinoma is the most common type followed by basal cell carcinoma, melanoma, Bowen disease and mycosis fungoides.[1,3,5,6]

Ruptured epidermal cysts have been reported in 21.3% to 38% cases as these cysts have a tendency to rupture easily.[1] Ruptured cysts can further cause a localised foreign body reaction.[1,3] This leads to the formation of the 'keratin granuloma' which are characterised by extruded keratin flakes surrounded by foreign body giant cells and inflammatory cells.[1]

We now discuss about the microscopic characteristics of these cysts. Keratinous cysts are lined by keratinized stratified squamous epithelium.[1,3] Depending on the type of keratinization and the formation of keratohyalin, keratinous cysts are divided into two types- Epidermoid cysts (contain keratohyalin) and Trichilemmal cysts (lack keratohyalin).[1] Also, focal calcification is frequently seen in Trichilemmal cysts. An epidermoid cyst forms when epidermal cells proliferate in a circumscribed space within the skin and retains keratinous debris, cholesterol or sebaceous material. Trichilemmal cysts, also called pilar cysts or wen, arise from the outer root sheath epithelium. They have a characteristic keratinization pattern called trichilemmal keratinization. Clinically, the two types of cysts are indistinguishable from each other however the epidermoid cysts are 4-5 times more common than trichilemmal cysts.[1]

Keratinous cysts are managed by a complete excision of the cyst and its wall.[3,5] Excision of the external port of the swelling is critical during the surgical procedure.[2] Incomplete excision can cause recurrence or a chronic inflammation.[3] If the cyst is tender or inflamed at the time of presentation, intralesional triamcinolone acetonide and oral antibiotics can be given followed by excision once the inflammation settles down.[3] The infected material can also be drained and cyst irrigated which is followed by antibiotics and thereafter surgical excision after the infection has settled down.[2] If there is no response by antibiotics, then a swab can be sent for microbiological culture. Golden et al report that in such resistant cases, methicillin resistant staphylococcus aureus (MRSA) was the usual causative organism.[2]

Ozan et al describe a rare case of a buccal mucosa epidermoid cyst which was seen in a 38-year-old female. Patient complained of a swelling in the inner aspect of her mouth, just distal to the oral commissure, for the past 6 months. Patient had no history of trauma or surgery over the site. On examination it was a soft, doughy, freely mobile mass which was non tender with no local rise in temperature. On histopathological examination, a diagnosis of epidermoid cyst was reported. They state that oral dermoid and epidermoid cysts are rare and account for less than 0.01% of all oral cysts.[5] Conversely, 1.6% of all epidermoid and dermoid cysts are found in the oral cavity.[4] Oral lesions are reported to occasionally cause difficulty in feeding, swallowing or phonation.[6]

In their case series, Datta et al, describe large keratinous cysts in various regions of the head and neck.[4] The various sites that they have mentioned include, the sublingual region extending till the neck, a cyst in the submandibular region presenting similar to a ranula in the sublingual region, a lateral neck swelling of a cyst that initially presented like an enlarged lymph node or an implantation dermoid over the eye causing mass below the eyebrow and cosmetic deformity, just to name a few.

CONCLUSION

Keratinous cysts, though common anywhere in the body, are relatively less common in the head and neck and there is a chance of misdiagnosis. A high index of suspicion is needed. Investigations like USG, CT scan, MRI scan and FNAC can help in diagnosis. In our case, a USG and CT scan of the swelling was done to help in the diagnosis.

Keratinous cysts may develop complications such as cystic rupture, secondary infections or abscess formation. This should be remembered when dealing with such cutaneous swellings. Further, these cysts have a higher recurrence rate in comparison to similar swellings like lipomas.

After successful excision, a histopathological examination and patient follow up is necessary as keratinous cysts have rare malignant potential.

In our case discussed here, a long-standing right sided cheek swelling was excised after USG and CT scan and the histopathological findings of the swelling were consistent with that of a keratinous cyst.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Save S, Mendonca E, Kumar R. A large long standing keratinous cyst on the cheek: a case report. Int Surg J 2024;11:1896-9.