



Surgical excision of a giant solitary keratoacanthoma in the cheek: a case report and literature review

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Background: Giant keratoacanthoma, a rare variant solitary of keratoacanthoma is characterized by a diameter exceeding 20 mm, rapid growth, and destruction of underlying tissue. Traditionally, it has been considered to be a self-resolving or low-grade squamous proliferation. Due to their atypical appearance, Giant keratoacanthomas may present a diagnostic challenge. Histopathological examination remains the reference standard for distinguishing squamous cell carcinoma (SCC) from keratoacanthoma. Surgical management is still the standard treatment for solitary keratoacanthoma.

Case Description: We present the case of a 75-year-old man with a giant keratoacanthoma of the cheek, with a transverse length of 25 mm and a longitudinal length of 30 mm located 30 mm below the earlobe that was surgically excised with no obvious recurrence during follow-up. We used the Intraoperative frozen-section examination, which played a crucial role in ensuring clear margins and complete tumor removal.

Conclusions: This case demonstrates that keratoacanthoma can be considered surgically curable and should be distinguished from SCC and other crateriform tumors. After surgical resection of giant isolated keratoacanthoma on the face, the high risk of cosmetic problems should be considered. We suggest free-flap transplantation be performed for functional repair, when the invasion range of the tumor is large and a perforation defect is formed.

Keywords: Giant; keratoacanthoma; histopathology; squamous cell carcinoma (SCC); case report

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Introduction

Keratoacanthoma is a low-grade skin tumor characterized by a distinct subset of self-regressing, locally invasive and destructively expanding keratinocytic skin neoplasms with the micromorphology of squamous cell carcinoma (SCC) (1). Clinically, keratoacanthoma can be classified as solitary

keratoacanthoma, multiple keratoacanthoma, eruptive keratoacanthoma (2) and keratoacanthoma centrifugum marginatum (3). Giant keratoacanthoma is a rare variant of solitary keratoacanthoma characterized by a diameter exceeding 20 mm, a self-limiting course, rapid enlargement within several weeks and spontaneous regression within several months (4). Complete surgical lesion

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removal is recommended for the treatment of solitary keratoacanthoma (5). Among reported cases, giant keratoacanthomas are very rare. We report a case of a giant keratoacanthoma of the cheek that was surgically excised and not observed to reoccur during follow-up. We present this case in accordance with the CARE reporting checklist (available at <https://acr.amegroups.com/article/view/10.21037/acr-23-100/rc>).

Case presentation

A 75-year-old male patient presented with a left facial mass that had persisted for more than 6 months. Throughout the aforementioned period, the patient experienced accelerated mass growth, itching, pain, swelling, purulent ulceration, and other discomforts. The patient had no family history of similar diseases or history of trauma preceding onset and had no obvious history of drug addiction or chemical exposure. At initial visit, the patient underwent history taking and physical examination, which revealed an exophytic mass on the left cheek with a transverse length of 25 mm and a longitudinal length of 30 mm located 30 mm below the earlobe (*Figure 1*). Cervical lymph nodes were not swollen. Based on the clinical history and physical examination, the preliminary consideration is “cheek mass, suspected to be giant keratoacanthoma”. To further confirm

the diagnosis and proceed with treatment, the patient was admitted to Hospital of Stomatology, Tianjin Medical University on November 12, 2021 for further evaluation and treatment.

Surgery was performed under local anesthetic infiltration with 2% articaine. Meilan was used to mark the surgical incision within 5 mm of the tumor’s periphery. The lesion was completely removed from the left cheek, and approximately 5 mm from the edge of the lesion was visible to the naked eye. The skin flap was extended for esthetic purposes, and the flap was brought along the direction of skin striations for tension-reducing sutures to close the gap (*Figure 2A-2E*). The wound was subsequently closed with a 3-0 silk thread. After completely removing the neoplasm, the tissue was processed for intraoperative frozen section histopathological examination. The histopathological results are as follows: The surface of the keratoacanthoma is verrucous and cup-shaped architecture with a central keratin-filled crater; inflammatory infiltrate, including lymphocytes and neutrophils surrounding the tumor; focal mild epithelial dysplasia; absence of significant cellular atypia or invasion into the surrounding tissues (*Figure 3A,3B*).

Postoperatively, the surgical site was scrubbed daily with iodophor for one week. No lesions recurred within the first postoperative week, and the wound healed properly. The lesion resolved completely, and the cheek function was normal (*Figure 1D*). The patient was followed up since the day of surgery, and his last follow-up was on July 17, 2023. There was no cheek discomfort during the treatment or follow-up periods. The patient had no obvious adverse reactions and had good prognosis, no recurrence, uncomplicated wound healing, and satisfactory cosmetic results. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for the publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Discussion

Keratoacanthoma, also known historically as molluscum sebaceum, is a self-healing SCC. In the recent World Health Organization (WHO) classification of cutaneous tumors, keratoacanthoma was classified as a low-grade SCC, suggesting that it is biologically stable and that some

Highlight box

Key findings

- Our report highlights the successful surgical excision of a giant keratoacanthoma, with no evidence of recurrence during the follow-up period. Intraoperative frozen-section examination can quickly evaluate the tumor margins and develop a surgical plan.

What is known and what is new?

- Keratoacanthomas are typically small, dome-shaped lesions that resemble squamous cell carcinomas (SCCs). Histopathological examination remains the reference standard for distinguishing SCC from keratoacanthoma.
- Giant keratoacanthomas may present a diagnostic challenge due to their atypical appearance. They can grow rapidly and become locally invasive. Intraoperative frozen-section examination played a crucial role in ensuring clear margins and complete tumor removal.

What is the implication, and what should change now?

- Clinicians should consider the possibility of giant keratoacanthomas in patients presenting with large, rapidly growing skin masses. Surgical excision with intraoperative frozen-section examination is an effective approach for the confirmation of diagnosis.



Figure 1 Clinical and postoperative healing photographs. Clinically, the patient’s face was asymmetrical from left to right, with the left part being slightly larger than the right (A). On the left, there is an ovoid extraneous mass (arrow) with a transverse length of 25 mm and a longitudinal length of 30 mm located 30 mm below the earlobe (A-C). One week postoperatively (D), the patient’s sutures were removed, and his clinical condition was stable. These images are published with the patient’s consent.

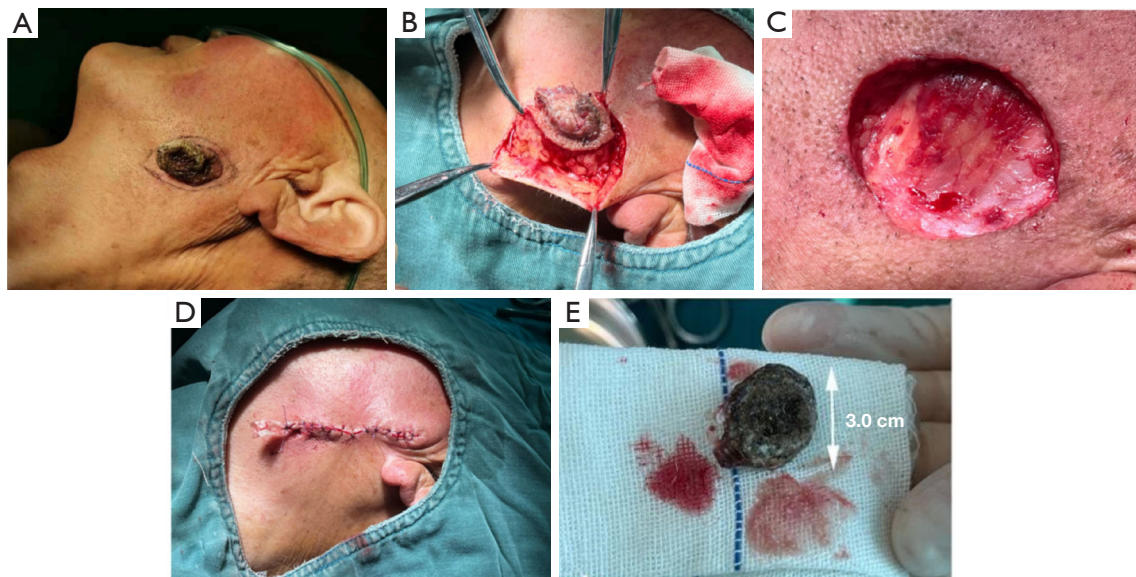


Figure 2 Intraoperative photographs. On the left, the border outside the lesion has a diameter of 5 mm and an oval tag line of approximately 30 mm; lines were painted along the dermatoglyph (A) and the mass layered around the local infiltration anesthesia, layered cut skin, subcutaneous tissue separation neoplasm and subcutaneous tissue and adipose tissue adhesion around the lesion (B). After the neoplasm was completely removed (C), a relaxation incision was made along the long axis, and a tension-reducing suture was inserted to close the gap (D). Finally, a drainage strip was placed (D). Vegetable-patterned substances from the surface of the tumor were completely removed (E). These images are published with the patient’s consent.

keratoacanthomas evolve into conventional SCCs with a gradual loss of capacity for spontaneous regression (6). Individuals with keratoacanthoma are most likely to be men aged ≥ 60 years (7).

The histopathological stages of keratoacanthoma

are as follows: early, proliferative, well-developed, regressing, and regressed (6). Based on distinctive clinical and histological features, keratoacanthomas can also be separated into follicular and non-follicular categories (8). Follicular lesions are located on the hair-bearing skin.

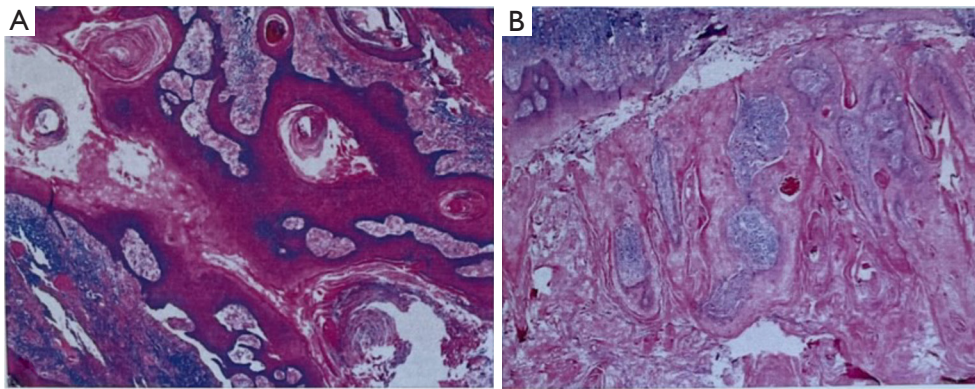


Figure 3 Intraoperative frozen tissue section of the patient and histopathological picture. Staining method: Hematoxylin-eosin staining. Magnification: $\times 40$. After completely removing the neoplasm, the lower edge of the upper edge of the 12° edge, 6° edge, 9° and 3° front edges and basal edge were used to excise a small amount of tissue for intraoperative frozen histopathological examination. The pathological results are as follows: the surface of the keratoacanthoma is verrucous; the nail process of the epithelium is growing deep (A), there is no obvious cell atypia, and marked inflammatory cell infiltration in the stroma, especially in the adjacent stroma and deep in the tumor, occurred. No lesions are observed on any of the cutting edges (B).

Non-follicular lesions included subungual and mucous membrane keratoacanthomas. Solitary keratoacanthoma is a benign epithelial neoplasm with follicular differentiation. An increasing number of reports suggest that solitary keratoacanthomas can be located in the conjunctival, vulvar, and subungual regions (5). Some lesions occur in the external auditory canal, and a risk factor for this is the frequent use of earbuds (9). Careful monitoring after biopsy is advocated even if the histopathological diagnosis is keratoacanthoma because a conventional SCC may remain in the residual tissue (10).

The origin of keratoacanthoma has not been completely elucidated; however, causative factors include immunosuppression, ultraviolet (UV) or X-ray exposure, drug treatments, foreign bodies, various traumas (5), and human papillomavirus DNA. Male sex, older age, smoking, and alcohol consumption have been reported to be hazardous variables (7).

The true incidence of keratoacanthoma may be underestimated because of diagnosis as SCC, physician underreporting, or spontaneous regression before diagnosis. An essential clinical characteristic of solitary keratoacanthoma is its self-limiting course; however, not all keratoacanthomas resolve spontaneously (7). Currently, histopathological examination remains the reference standard for distinguishing SCC from keratoacanthoma. Histopathologically, keratoacanthoma is characterized by mature lesions surrounding an arched epithelium that forms

a crater filled with keratin. An enlarged pale cytoplasm within cells is arranged in concentric-layered islands with central keratinization. These nests extend into the dermis to the level of sweat glands. Large cells with pink cytoplasm oriented towards the nest of central tumor cells are generally characteristic of keratoacanthoma (11).

In the literature concerning the use of dermoscopy and reflectance confocal microscopy in Chinese Han patients, the dermoscopic features of keratoacanthoma include concentric circles of the central crater, keratin mass, keratin scale, and polymorphic vascular pattern. Reflectance confocal microscopic features include refractile crust, atypical honeycomb pattern, dark-centered cells, large round nucleated cells, dendritic cells, and linear or round vessels traversing the dermal papillae (12). Currently, there is a web application in the latest research literature to process large numbers of free-text histopathology reports and classify diagnoses of keratinocytic cancers with a high degree of accuracy (13).

The treatment methods for keratoacanthoma can be divided into non-pharmacological, pharmacological, and surgical treatments (14,15). Surgical management is the standard treatment for solitary keratoacanthoma (5). Complete removal and follow-up are recommended for solitary keratoacanthoma within the proliferative phase. In the literature, treatment with intralesional methotrexate has been used as an important treatment for keratoacanthoma before surgical excision or as an adjuvant

treatment for postoperative management (16). Intralesional methotrexate has produced a greater response in patient with keratoacanthoma than in those with SCC, indicating its effectiveness and safety (17).

Conclusions

The high risk of cosmetic problems should be considered after the surgical resection of a giant isolated keratoacanthoma on the face. Our elderly male patient had loose facial skin, an obvious Langer's line, poor tension, and an incision of less than 40 mm. The skin flap was extended for esthetic purposes, and the flap was brought along the direction of skin striations for tension-reducing sutures to close the gap. When the invasion range of the tumor is large and a perforation defect is formed, free-flap transplantation should be performed for functional repair.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at <https://acr.amegroups.com/article/view/10.21037/acr-23-100/rc>

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Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for the publication of this case report and accompanying images. A copy of the written consent is

available for review by the editorial office of this journal.

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