Case Report

DOI: https://dx.doi.org/10.18203/issn.2455-4510.IntJResOrthop20242408

A case report of a rare incidence adnexal nodular hidradenoma mimicking as simple ganglion cyst in hand

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Received: 13 June 2024 Revised: 18 July 2024 Accepted: 24 July 2024

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ABSTRACT

Soft tissue swelling of hand is a common condition presenting in orthopedic outpatient department (OPD). Simple ganglion cyst is the most prevalent presenting disorder among the various differential diagnoses for this ailment, which include ganglion cyst, inclusion cyst, giant cell tumor of tendon sheath, enchondroma, carpal bossing, leiomyoma, and adnexal tumors. A 75-year-old male patient presented with soft tissue swelling over dorsum of his left hand, which was insidious in onset and painless in nature at initial stages, but over the past 6 months it grew over to become of approximately 7×3 cm in size, and now causing slight pain and discomfort to patient. Primary investigation like X-ray, ultrasonography and magnetic resonance imaging (MRI) was done. Clinical and radiological investigation was suggestive of simple ganglion cyst. So patient was operated for it as surgical excision and tissue biopsy was sent for histological examination to arrive at definitive diagnosis of the condition and further line of management. Histological finding of tissue biopsy was suggestive of adnexal nodular hidradenoma with nodular and cystic areas of differentiation. Thus, before drawing a final diagnosis, a rare tumor like adnexal hidradenoma should also be taken into account as a differential diagnosis. This aids in the planning of the patient's future course of treatment. This instance emphasizes the significance of tracing reports prior to repeat surgery and submitting biopsies of all hand lesions for histological testing.

Keywords: Cystic swelling of hand, Hidradenoma, Ganglion cyst, Adnexal tumour, Hand

INTRODUCTION

Ganglion cysts are soft tissue swellings which are most frequently encountered in orthopaedic outpatient department (OPD). This swelling is commonly found in the hand or wrist. Second most common site is ankles and feet. Rarely it may present in sites like distal interphalangeal joint, hip, knee, ankle, foot and other body parts.

Though exact etiology of ganglion cyst is unclear, various theories have been proposed to explain its occurrence.⁴ Among them most widely accepted theory is myxoid degeneration of connective tissue after repetitive injury to the capsular and ligamentous structures. Repetitive microtrauma results in mucinous degeneration of

connective tissue leading to production of hyaluronic acid from fibroblasts and accumulation of mucin jelly-like material to form the cyst.^{3,4}

Pathophysiological study of cystic fluid shows gelatinous material containing mainly hyaluronic acid and lesser amounts of glucosamine. Since there is no epithelial lining exists in these structures, they should not be classified as true cysts.⁵

Most cases of ganglion cyst are asymptomatic or painless but it may present with pain which may be due to nerve compression or less frequently because of inflammatory alterations brought on by consequences from rupture, bleeding, or infection.⁴

Clinically other differential diagnosis of cystic swelling over dorsum of hand includes Inclusion cyst, giant cell tumor of tendon sheath, enchondroma, carpal bossing, leiomyoma, adnexal tumors, and dermatofibroma.³ So, tissue examination and immunohistochemistry of the lesion should be carried out to arrive at final diagnosis of the condition.

CASE REPORT

A 74-year-old male patient presented to the OPD of P.D.U. Medical College and Hospital Rajkot, with complain of swelling on dorsum of left hand. Initially painless and insignificant, the swelling grew over the course of the previous six months, leading the patient to experience slight discomfort. The patient used some analgesics on his own over the course and has not sought medical attention until now.

On clinical examination swelling was approximately 7×3 cm in size, soft, mobile, skin-colored, globular with a bosselated surface proximal to the base of left fifth finger on dorsum of hand. The skin over the swelling was shiny with no signs of inflammation on the surface.



Figure 1: Clinical appearance of presenting swelling.

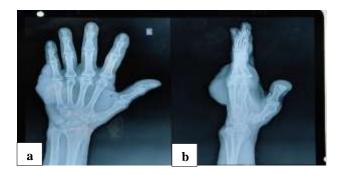


Figure 2 (a and b): X ray image suggestive of soft tissue swelling with no bony involvement.

X-ray of left hand showed soft tissue swelling with no underlying bony involvement. Ultrasonography was advised which showed well defined multiloculated complex cystic lesion with multiple septations suggestive of benign mucin containing cystic lesion. The findings described above pointed to a possible ganglion cyst, therefore we performed an MRI to check for extent of soft tissue involvement and to rule out vascular anastomosis.

An MRI result revealed a distinct loculated cystic lesion infiltrating the nearby hypothenar muscles, and encasing the extensor tendon of the fifth finger suggestive of ganglion cyst. So, the patient was posted for an elective surgical excision of ganglion cyst.

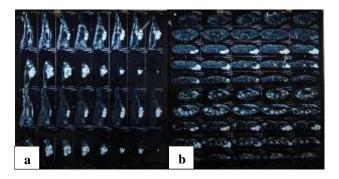


Figure 3 (a and b): MRI finding displaying extent of soft tissue involvement.

Surgical procedure

Patient was posted for elective ganglion cyst excision. The patient was operated under regional anesthesia in supine position. Wide-local excision was done through a skin incision extending from Proximal interphalangeal joint of the left fifth finger to base of fifth metacarpal. Careful dissection was done. The swelling appeared to be 7×3 cm of size with bosselated nodular surface and few cystic areas. The grayish bosselated soft tissue mass was excised totally from either side of the extensor tendon with care to preserve normal tendon anatomy and function. Followed by Thorough wash given with normal saline and skin closure was done and sterile dressing was placed. Excised specimen was sent for a histopathological examination.

Postoperatively weekly follow up was taken. In 13 days wound healing was there with primary intention. With proper physiotherapy patient regained his normal wrist and finger range of motion with adequate grip strength within a month.

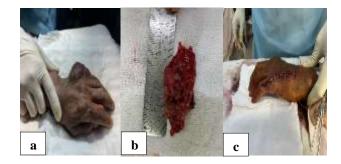


Figure 4 (a-c): Intraoperative clinical image and excised mass.

Histopathological findings of tissue sample showed features of fibrous tissue containing a multi nodular basaloid tumor showing peripheral palisades and high nuclear to cytoplasmic ratio.

There were two distinct pattern appeared Solid and cystic; wherein: the solid section is made up of two different cell types: polyhedral cells with basophilic cytoplasm and clear cells with round, eccentric nuclei; and the cystic portions are the result of tumor cell degeneration. There was evidence of ductal differentiation which was further confirmed by positive staining with epithelial membrane antigen and carcinoembryonic antigen. Above findings and further immunohistochemistry assay suggested that these are the features of "atypical solid and cystic hidradenoma."

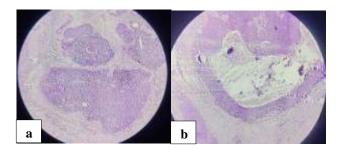


Figure 5 (a and b): Histological slides showing nodular and cystic areas.

DISCUSSION

Nodular hidradenoma is a rare benign adnexal tumour that arises from the distal excretory duct of eccrine sweat gland, but recent study shows that it could be either of eccrine or apocrine in origin and so reclassified as apocrine. Nodular hidradenoma is most commonly seen in age group of 20-50 years. Though current case was 74 years old male. This swelling occurs in female twice as commonly as male. Predominant site being scalp and axillae.

A study conducted by Ramish et al reported a female with similar swelling in scalp, which is common site of this swelling. In current study swelling was approximately 7×3 cm in size, soft, mobile, skin-colored, globular with a bosselated surface proximal to the base of left fifth finger on dorsum of hand. However, some studies like, study conducted by Farah et al, another study by Jyotsna et al. have found atypical locations like abdomen and breast. However, involvement of distal extremities particularly dorsum of hand is very rare which was seen in current case. They either occur sporadically or may present with multiple adnexal tumours and should be evaluated for rare genetic syndromes like Birt-Hogg-Dubé syndrome, Cowden syndrome, and Muir-Torre syndrome. 10,11

This swelling usually presents as a slow growing, solitary, freely movable nodule, solid, or cystic mass measuring on an average 1-2 cm in diameter, but may reach up to 6.0 cm or more. This lesion is typically located on the head, neck and limbs. ^{12,13}

Hidradenoma is a slow-growing benign adnexal tumour, that presents as a solitary nodule and can be either apocrine

or eccrine, with variants including solid-cystic, clear cell, nodular, and poroid types. ^{13,14} Rapid growth may represent trauma, haemorrhage, or a malignant change. Malignant transformation can be characterised by a larger size, infiltrative growth pattern, poor circumscription, nuclear enlargement and pleomorphism, increased mitotic activity, necrosis and ulceration. ^{10,13}

Even though histology confirms the clinical diagnosis of hidradenoma, misunderstandings may arise if histologic data is overlooked as evidenced by our case. There is perhaps a need to apply a more cautious approach with cystic lesion on hand or wrist as hidradenoma may be present masking ganglion cyst. This case highlights the importance of considering adnexal tumours in the differential diagnosis of cystic lesions.

Certain lesions such as lipomata, ganglia, fibro lipomatous hamartoma, and GCTTS have characteristic appearances. Unfortunately, where the lesion does not exhibit typical features, differentiation from malignancy cannot be categorically made through imaging. ¹⁵ In such instances fine needle aspiration cytology and histopathology is a useful tool to aid in diagnosis. ¹⁶

A study conducted by Jin et al, has shown malignant potential of this tumor so every case should be sent for histopathological examination.¹⁷ Current case was also confirmed by histopathological examination which showed atypical solid and cystic hidradenoma.

Other differential diagnosis includes ganglion cyst, Epidermal inclusion cyst, vascular malformation, and basal cell carcinoma. Furthermore, due to the wide overlap in the clinical symptoms of cutaneous adnexal tumours, tumours with ambiguous histopathologic features can be diagnosed by biopsy and immunohistochemistry. 18

CONCLUSION

This case is deviating from common presentations in terms of age, gender, and location of lesion. So, this case highlights the importance of including even rare tumors like adnexal nodular hidradenoma in differential diagnosis of soft tissue swellings of hand. Understanding unusual tumours and their exclusion is essential for efficient therapeutic treatment. If adequate investigation is not done, tumours with similar clinical presentations may be misdiagnosed for one another. Our case study highlights the need to send biopsies of all hand lesions for histological analysis along with appropriate follow-up to verify the diagnosis. By doing this, rare disorders can be detected and patients could be counselled of the potential for a recurrence.

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

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Cite this article as: Merja HR, Ramavat SD, Mehta PS. A case report of a rare incidence adnexal nodular hidradenoma mimicking as simple ganglion cyst in hand. Int J Res Orthop 2024;10:1080-3.