

# Pathological Fracture of Humerus in a Case of Adenoid Cystic Carcinoma of the Submandibular Gland: A Rare Case Report

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## Abstract

Adenoid cystic carcinoma of salivary glands usually presents as a locally aggressive malignancy. Bone

**Keywords:** Cancer; Bone metastases; Pathological fracture; Palliative care; Multidisciplinary care

Adenoid cystic carcinoma (ACC), a rare head and neck cancer often involving the salivary glands, follows an indolent course and is locally aggressive with a propensity for perineural invasion. Distant metastasis is usually a late event, lungs being the preponderant site. The median survival after onset of distant metastases has been reported to be about three years [1]. Pathologic fracture is a devastating skeletal related event (SRE) in patients with metastatic bone disease. These are more often reported with solid tumors of breast, lung, thyroid, and kidney and hematolymphoid malignancies like multiple myeloma. With regards to ACC, patients with bone metastases are rare and they fare poorer than patients with pulmonary metastases [2]. To our knowledge, pathological fracture of long bones in patients with ACC of submandibular gland has not been reported in literature till date. We present the first case report on a patient with ACC of submandibular gland presenting with a pathologic fracture of humerus in a specialist palliative care clinic (SPC) at a tertiary care cancer center.

Mrs. RPG was a 65 year old homemaker and mother of two children. She was diagnosed with ACC of right submandibular gland 3.5 years ago for which she underwent radical treatment. A year and five months after her diagnosis, her disease progressed and she developed subcutaneous and pulmonary metastases. She was referred to SPC for early palliative care and was started on oral Morphine (titrated to 40mg/day), adjuvant analgesics and local measures for her pain, which was predominantly over the ulcerated subcutaneous metastatic nodules. Additionally, palliative chemotherapy continued. Her response-evaluation scans revealed new onset bone metastases in the left acetabulum and sixth cervical vertebra for which she was completely asymptomatic. She was started on bis-phosphonate therapy to delay the occurrence of consequent SREs.

Three years after the onset of her disease, she presented to SPC with complaints of pain over the left arm for a duration of two weeks.

The pain was insidious in onset, localized, sharp in nature, severe in intensity, gradually progressive, triggered by movement of the arm, not relieved with her analgesics and impacting her sleep as well as

activities of daily living (ADLs). On enquiry, there was no history of fall or trauma. On examination, tenderness was present over the left arm with restricted range of movements. X-Ray of the upper limb showed a lytic and mildly expansile intramedullary lesion with a wide zone of transition and overlying permeative pattern of cortical erosion involving the mid diaphysis of the humerus associated with subtle periosteal reaction (Figure 1). A diagnosis of a metastatic lytic



**Figure 1:** X-ray of left upper limb showing a lytic lesion with pathological fracture in the mid-diaphysis of the humerus

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humeral lesion with an associated pathological fracture was made taking into consideration the clinical and radiological characteristics. Being an un-displaced fracture, it was managed conservatively with a U-slab for immobilization. Additionally, palliative radiotherapy (12Gy in 2 fractions) was given to the lesion. Mrs. RPG was started on Sorafenib, a Vascular Endothelial Growth Factor (VEGF) inhibitor, with palliative intent. She eventually required 15 mg Morphine every 4 hours (90mg/day), Celecoxib 100 mg twice daily, Pregabalin 150 mg daily for adequate pain relief. She continued to receive zoledronic acid 4 mg once a month with intent to delay further SREs. She was enrolled under home care services, had good symptom control due to which she could make a visit to her native village.

Metastases to bones in patients with ACC of the submandibular gland is a late event. Sites of bone metastases from ACC of head and neck reported till date include vertebrae, ribs, pelvic bones, sternum, and femur [2-4]. Our patient developed bone metastases two years and eight months after her diagnosis and to our knowledge, metastasis to a long bone and in particular, to the humerus, from a primary ACC of the submandibular gland, has not been reported till date. Furthermore, this is the first case report in the English literature to describe pathological fracture of a long bone, in this histologic type of cancer of the submandibular gland.

Bone metastases confer poor prognosis in adenoid cystic carcinoma. We estimated the prognosis of our patient using Katagiri scoring system [5] which is one of the prognostication tools for patients with skeletal metastases and we estimated a median survival of around 6 months for our patient. Pathological fracture is a complication that increases symptom burden in patients with metastatic bone disease. Our patient had severe pain that limited her ADLs and required more than double the oral morphine equivalent dose that she needed previously for pain relief. Had our patient presented to us right at the onset of pain, the possibility for a fracture could have been avoided.

This emphasizes the need to educate patients and caregivers to look out for any new symptoms and to bring them to the attention of the treating team promptly. It also highlights how crucial it is for physicians to identify the cause of pain at a new skeletal site at the onset especially in patients with malignancies and to implement prophylactic measures to prevent complications such as pathological fractures, especially in long bones.

The management of pathological fractures depends upon the type of fracture and prognosis of the patient. Pathological fractures of long bones often require multi-disciplinary management including need for radiotherapy and surgery. In this case, a dedicated team of radiation oncologists, bone and soft tissue surgeons, medical oncologists, radiologists, and palliative care professionals were involved. In view of her poor prognosis and because it was an un-displaced fracture, our patient was managed conservatively. In addition to analgesics, radiotherapy was offered for palliation.

Our patient priorly had poor response to disease modifying therapies and this new onset bone lesion with associated pathological fracture indicated further disease progression. This event triggered a change in goals of care for our patient. She was commenced on targeted therapy with palliative intent. During this period, the SPC team provided holistic care to the patient and her family in the form of good symptom control, psychosocial support and rehabilitation because of which the patient's wish of visiting her hometown was also fulfilled.

This is a rare case report of a long bone pathological fracture in a patient with ACC of the submandibular gland. A multidisciplinary team of surgeons, radiation oncologists, medical oncologists, palliative care professionals, physiotherapists and nurses were involved in the management of the patient. Pathological fractures impair the quality of life of patients tremendously, affect their functionality and confer poor prognosis. They are almost always associated with severe pain and require frequent escalations of analgesics as the union of pathologic fractures is a tardy process. This case report underscores the need to evaluate cancer induced bone pain in long bones at the onset and consider prophylactic fixation, radiotherapy, and bisphosphonates to prevent the occurrence of pathological fractures.

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for the clinical information and images to be reported in the journal.

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