International Journal of Science and Research (IJSR)
ISSN: 2319-7064
Impact Factor 2024: 7.101

Primary Squamous Cell Carcinoma at Proximal Tibia: A Rare Case Report

Dr. Jyoti Singh[1], Dr. Reena Jain[2], Dr. Jyoti Priyadarshini[3], Dr. Sudha Iyengar[4]

[1]PG Student, Department of Pathology, GRMC, Gwalior (MP)

[2]Professor, Department of Pathology, GRMC, Gwalior (MP)

[3]Professor, Department of Pathology, GRMC, Gwalior (MP)

[4]Professor and HOD, Department of Pathology, GRMC, Gwalior (MP)

Abstract: ***Introduction: Primary squamous cell carcinoma of bone is rare in the skeletal system. The majority of these carcinomas when found in the extremities are metastatic from another primary. Case Summary: We report a case of 40 year female presented with complain of right sided knee pain, tenderness since 3 month, insidious in onset also having difficulty in weight bearing there was no history of trauma and fever previously. Pain was aggravated on movement and reduced on taking rest or on medication. The excised tissue was sent for histopathological examination for histopathological examination in our department. Conclusion: This report is the first to describe primary keratinizing SCC of the tibial bone. However, there is still no consensus on the standard treatment method or prognosis of primary SCC of non-skull bones because of its rarity, with only a few cases having been reported and followed up. Our findings demonstrate that clinicians must exhaust all available means for the diagnosis of primary SCC of bones, so greater attention can be paid to timely treatment and effective management.***

Keywords: Keratinizing SCC, Proximal Tibia, p63, Histopathological examination

1. Introduction

Primary squamous cell carcinoma of bone is rare in the skeletal system other than in the skull, with only two such cases reported in the English literature [1, 3]. This is attributed to the absence of native squamous epithelium in bones. The majority of these carcinomas when found in the extremities are metastatic from another primary. In 1997, Gangopadhyay and Saha [3] reported a case of a primary squamous cell carcinoma of the left iliac bone, and in 2003, Abbas et al. [1] reported the case of a patient with a primary squamous cell carcinoma of the distal tibia.

2. Case Report

A 40- year female presented with complain of right sided knee pain, tenderness since 3 month insidious in onset also having difficulty in weight bearing there was no history of trauma and fever previously. Pain was aggravated on movement and reduced on taking rest or on medication.

CT scan revealed focal osteolytic lesion involving upper tibial shaft with small cortical erosion seen mainly along the posterior and adjacent medial/lateral aspect of tibia, with extraosseous soft tissue seen. Features are more in favour of neoplastic etiology (? Sarcoma). A biopsy of bony tissue from tumor was taken and histopathologically squamous cell carcinoma was diagnosed.

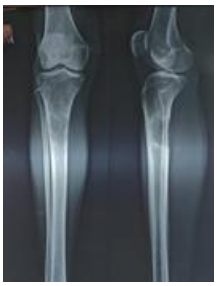

Figure 1: X-ray knee joint

3. Methodology

Gross examination:
Specimen received consist of multiple, irregular, greyish white to greyish brown soft to hard tissue pieces altogether measuring 1.5x1.5 cm

Microscopic Examination
Histological section shows tissue predominantly composed of tumor cells showing features of keratinizing squamous cell carcinoma interspersed with bony spicules. Findings are suggestive of Keratinizing Squamous Cell Carcinoma of right proximal tibia.

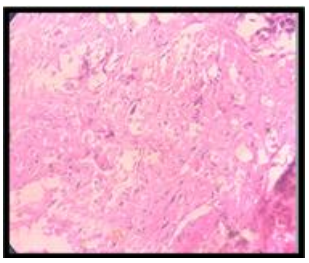

Figure 2: Histological section shows tissue predominantly composed of tumor cells showing features of keratinizing squamous cell carcinoma.

Immunohistochemistry

IHC staining p63 done on the tissue biopsy submitted resulting in positivity in the tumor cells. P63 is generally considered a marker of squamous differentiation and is often positive in Squamous Cell Carcinoma where as it is less frequently found in most sarcomas.

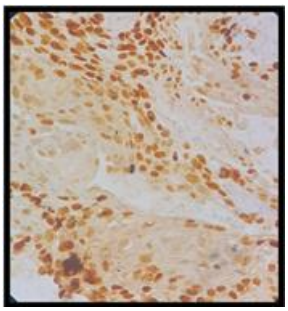

Figure 3: p63 positive at (400X)

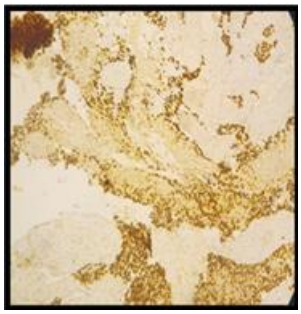

Figure 4: p63 positive at (100X)

4. Result

On the basis of histopathology, the case was diagnosed as Keratinizing Squamous Cell Carcinoma.

5. Discussion

Primary squamous cell carcinoma of bone usually is reported in the skull; it is rare elsewhere in the skeleton. A MEDLINE search from 1970 to 2008 with keywords “squamous cell carcinoma/cancer” and “bone/skeleton” listed only two cases outside the skull. The first report was of a case in the left iliac bone of a 20-year-old woman [3]. Extensive workup revealed no other foci of malignancy. Histologic analysis showed infiltration of bone tissue by malignant squamous cells with pearl formation.

SCC is a tumor of the epithelial tissue that typically originates from the epithelial linings of the skin, respiratory tract, digestive tract, and reproductive tract; thus, SCC can involve the head and neck, esophagus, lungs, cervix, and genital area [2]. Epithelial linings can be divided into layered squamous epithelium and non-squamous epithelium. The squamous differentiation phenotype of SCC depends on the type of oncogenic mutation involved and the cell of tumor origin, and this phenotype determines the degree of differentiation and therefore, the aggressiveness and invasiveness of these tumors[4]. In the case of most cancers, the initial target cells of the oncogenic mutations as well as the number of cancer stem cells in the tumor are unknown [5].

Therefore, it is difficult to know the source cells for primary SCC in the bones. SCC-derived cells share a common feature, in that they originate due to the mutation of proliferative basal cells, which are characterized by their ability to self-renew and produce terminally differentiated cells. Under the influence of oncogenic genes, both stem and progenitor cells can act as the origin cells of cancer [6]. A comparison of different SCCs shows that they are characterized by very similar mutant genes, including TP53, SOX2, TP63, CDNK2A (P16-INK4A), NOTCH1, KMT2D, PIK3CA, and PTEN [7].

Primary SCC of bone is commonly seen in the head and neck region [8, 9], and it is rarely found elsewhere in the skeletal system. It is not easy to make a diagnosis of primary SCC of a non-skull bone, as this depends not only on pathological and immunohistochemical examinations but also requires extensive workup to rule out metastasis. In addition to metastasis, the differential diagnosis of primary SCC should also include SCC caused by chronic osteomyelitis [10]. Keratin pearls are the pathological features of highly differentiated SCCs, and their presence in histopathological sections of well-differentiated SCCs is a common phenomenon [11].

Unlike the cases of primary SCC of a non-skull bone reported previously, our case was unique in that it had keratin pearls. This is because our patient had a well differentiated SCC. However, in our case, the tumor cells were also reactive to p63. In our patient, the final diagnosis of a primary SCC of the bone was supported by the immunohistochemical findings, the extensive workup for the identification of a primary source, and the fact that the patient remained disease-free during a 2-year follow-up period.

6. Conclusion

To the best of our knowledge, this report is the first to describe primary keratinizing SCC of the tibial bone. However, there is still no consensus on the standard treatment method or prognosis of primary SCC of non-skull bones because of its rarity, with only a few cases having been reported and followed up. Our findings demonstrate that clinicians must exhaust all available means for the diagnosis of primary SCC of bones, so greater attention can be paid to timely treatment and effective management. Regular and adequate follow-up is essential to help rule out metastasis and judge the prognosis.

References

[1] Abbas A, Bromage JD, Stocks PJ, Al-Sarireh B. Case of the Conference: Primary squamous cell carcinoma in a long bone. J Bone Joint Surg Br. 2005;87(suppl 1):6.

[2] Guan Y, Wang G, Fails D, Nagarajan P, Ge Y. Unraveling cancer lineage drivers in squamous cell carcinomas. Pharmacol Ther. 2020; 206: 107448. [RCA] [PubMed] [DOI] [Full Text] [Cited by in Crossref: 16] [Cited by in RCA: 17] [Article Influence: 3.4] [Reference Citation Analysis (1)]

[3] Gangopadhyay S, Saha S. Primary squamous cell carcinoma of bone. J Indian Med Assoc. 1997; 95: 521–523. [PubMed]
[4] Sánchez-Danés A, Blanpain C. Deciphering the cells of origin of squamous cell carcinomas. Nat Rev Cancer. 2018; 18:549-561. [RCA] [PubMed] [DOI] [Full Text] [Cited by in Crossref: 121] Cited by in RCA: 188] [Article Influence: 26.9] [Reference Citation Analysis (0)]
[5] Blanpain C, Fuchs E. Epidermal homeostasis: a balancing act of stem cells in the skin. Nat Rev Mol Cell Biol. 2009; 10:207-217. [RCA] [PubMed] [DOI] [Full Text] [Full Text (PDF)] [Cited by in Crossref: 1024] [Cited by in RCA: 919] [Article Influence: 57.4] [Reference Citation Analysis (0)]
[6] Guan Y, Wang G, Fails D, Nagarajan P, Ge Y. Unraveling cancer lineage drivers in squamous cell carcinomas. Pharmacol Ther. 2020; 206:107448. [RCA] [PubMed] [DOI] [Full Text] [Cited by in Crossref: 16] [Cited by in RCA: 17] [Article Influence: 3.4] [Reference Citation Analysis (1)]
[7] Sánchez-Danés A, Blanpain C. Deciphering the cells of origin of squamous cell carcinomas. Nat Rev Cancer. 2018; 18:549-561. [RCA] [PubMed] [DOI] [Full Text] [Cited by in Crossref: 121] [Cited by in RCA: 188] [Article Influence: 26.9] [Reference Citation Analysis (0)]
[8] Bodner L, Manor E, Shear M, van der Waal I. Primary intraosseous squamous cell carcinoma arising in an odontogenic cyst: a clinicopathologic analysis of 116 reported cases.
[9] Allanson BM, Low TH, Clark JR, Gupta R. Squamous Cell Carcinoma of the External Auditory Canal and Temporal Bone: An Update. Head Neck Pathol. 2018; 12: 407-418. [RCA] [PubMed] [DOI] [Full Text] [Cited by in Crossref: 36] [Cited by in RCA: 55] [Article Influence: 7.9] [Reference Citation Analysis (0)]
[10] Akoh CC, Chang J, Buckwalter J. Marjolin's Ulcer of the Tibia with Pelvic Lymph Node Metastasis. Iowa Orthop J. 2017; 37: 133-138. [PubMed]
[11] Sarode SC, Sarode GS, Sengupta N, Sharma NK, Patil S. Calcified keratin pearls in oral squamous cell carcinoma. Oral Oncol. 2020; 109: 104681. [RCA] [PubMed] [DOI] [Full Text] [Cited by in Crossref: 3] [Cited by in RCA: 1] [Article Influence: 0.2] [Reference Citation Analysis (0)]