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LEFT FOOT MONOPHASIC SYNOVIAL SARCOMA IN A 28-YEAR OLD MALE; A CASE REPORT AND REVIEW OF THE LITERATURE

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ABSTRACT

Synovial sarcoma though a malignant soft tissue sarcoma of unknown histogenesis by definition, generally tends to arise about or near joints. By morphologic or histopathologic characterization, the subtypes identified are the biphasic, monophasic and undifferentiated or poorly differentiated variants. While the monophasic variant is the most common of the three, 10% of soft tissue sarcomas in all are represented by synovial sarcoma, and commonly affect the extremities. In view of the challenges of accurate diagnosis or misdiagnosis, we report a case of a left foot monophasic synovial sarcoma in a 28 year old male. The swelling which was initially painless, later showed pressure symptoms and was eventually subjected to surgical excision. The histopathological examination of the excised mass confirmed it to be a Monophasic synovial sarcoma. A recurrence developed while the patient was awaiting uptake of neo adjuvant chemo and radiation therapy.

KEYWORDS: Synovial sarcoma, Left Foot, Monophasic, Soft tissue sarcoma.

INTRODUCTION

Although a well defined malignant soft tissue neoplasm which arises about or near joints generally, synovial sarcoma is characterized in definition by unknown histogenesis, and expresses the biphasic, monophasic and poorly or undifferentiated morphologic variants.[1,2] Even though the term 'synovial sarcoma' implies an origin from joint linings, less than 10% of them are intra-articular, signifying that the tumour does not necessarily develop from synoviocytes.[1,2] They constitute about 10% of all soft tissue sarcomas, most occurring in the age brackets of the 20s and 40s, and 60 to 70% are seen to involve the lower extremity, particularly around the knee and thigh areas.[3,4] It suffices therefore to admit that synovial sarcoma is rarely encountered in routine clinical practice. Nevertheless it is a highly malignant type of soft tissue sarcoma, for which Clinicians and Pathologists are called upon to entertain a high index of suspicion in order not to miss detecting likely cases.[7]

The peak incidence is reported in the fourth decade of life, and further to that most authors agree that synovial sarcoma is one of the most commonly misdiagnosed malignancies of soft tissues, owing to its ability to elicit pain similar to that caused by common trauma, ability to change size, and the slow growing pattern.[3-5] For example it can be misinterpreted or misdiagnosed initially as ganglion cyst or glomus tumour when it involves the hand-wrist complex.[7] Apart from the foot sarcomas, synovial sarcoma developing in the hand is very uncommon, and going by the literature, it is an extreme rarity that bears a very grim prognosis.[7]

Considering the background that early and accurate diagnosis is usually a challenge, encountering a synovial sarcoma on the foot dorsum (an unusual location) of a 28 year-old is a surgical or even pathologic curiosity which the authors wish to share with the community of scholars.

CASE REPORT

A 28 year-old male had presented with a soft tissue swelling around the left ankle and dorsum of the foot which was of two years duration at the time of presentation. This swelling was initially painless, but began to develop pressure symptoms in the last two months prior to presentation. Incidentally, the index patient has been on HAART (highly active anti-retroviral therapy) following a positive retroviral screen test.

On examination, an irregularly shaped mass was seen on the anterior surface of the left ankle, and extending to the dorsum of the foot. It measures approximately 8 x 10 cm in size, felt firm in consistency, not attached to the overlying skin, but was firmly attached to the underlying structures. It was non tender, but exhibited differential warmth to touch, whilst regional lymphadenopathy was absent.

The investigations requested include; Left ankle X-ray and ultra-sound scans which demonstrated the mass lesion, packed cell volume (PCV of 42%), serum E/U/Cr (electrolyte, urea and creatinine assays) which show normal kidney function test, serology (retroviral screen test was positive), ECG (electrocardiography), and echocardiography which revealed normal cardiac function.

The patient was worked up for surgery, whilst a unit of blood was grouped and cross matched. The mass on the dorsum of the left foot was surgically excised, but was reported to have recurred about 8 months later with pain associated in the current presentation.

The histopathology report had earlier diagnosed the mass lesion as a synovial sarcoma. The tissue sections show a variably cellular proliferation of neoplastic spindle shaped cells disposed as haphazard fascicles within a collagenous matrix. The cells are characterized by a pale, poorly defined cytoplasm, and in some of them are seen moderate to abundant abnormal mitotic figures, that is typical of such a malignant mesenchymal disease. On the basis of a monomorphic or monotonous picture of the neoplastic spindle cells, it was finally signed out as a Monophasic synovial sarcoma.

The managing Orthopedic team has written to refer the patient to the Radio-oncology unit for a review, expert evaluation, and possible co-management.

DISCUSSION

The authors report a case of a left foot dorsum mass which was earlier assigned a histological diagnosis of monophasic synovial sarcoma, following a surgical excision. About 10 months to a year later the patient is suffering a recurrence of the same tumor in same location. It is one among the several soft tissue sarcomas.

Alegria-Landa et al had reported a case of primary subcutaneous synovial sarcoma, but prior to their report, only four of such cases have been captured in published literature.

On the basis of histologic or morphologic appearance, synovial sarcoma is divided into three subtypes – biphasic, monophasic, and poorly differentiated morphologies. The monophasic form being the most common is characterized by ovoid or spindle shaped cells disposed in a monomorphic or monotonous distribution within a collagenous stroma.[4,8] Whereas we report here a case signed out as monophasic synovial sarcoma of the left foot, it is note worthy to observe that another quite uncommon or extreme rarity is the synovial sarcoma of the hand, which going by the literature conveys a very poor prognosis.[6]

It is to be noted that imaging appearance is non-specific, and therefore a biopsy is necessary to confirm the diagnosis in all cases. Synovial sarcoma is not only locally aggressive, but also demonstrates a higher metastatic potential than most other soft tissue sarcomas and so the patients have an overall poor prognosis.[3]

It is important to differentiate it from other entities that may mimic it, for example: extra-skeletal chondrosarcoma, extra-skeletal osteosarcoma, MPNST (malignant peripheral nerve sheet tumour), and solitary fibrous tumour. MPNST may display similar histologic features, but a lack of haemorrhage, surrounding oedema, and lack of association with major nerves with the sarcoma makes MPNST less likely.[4]

Other locations or body sites which can be involved by synovial sarcoma include; head and neck, chest wall, mediastinum, kidneys, retroperitoneum, and subcutaneous tissue.[8-10]

There may not be many signs to detect on general or systemic examination, owing to the several body regions and sites that can be involved, except for the localized changes produced at the sites of involvement e.g a focal mass or secondary pain.[9-11] Further buttressing how this sarcoma has no origin from synovial structures or synoviocytes, some particularly rare involvements and presentations are referenced herewith. Synovial sarcoma in the mediastinum, tonsils, kidney, vertebral spine, intraneural (tibial nerve), and the tongue.[16,18-22] The case of mediastinal sarcoma reported by Abu-Zaid et al only presented with right-sided chest pain and dyspnea.[24]

In the current literature, Lodhia J et al.[25] published the first case reported in Tanzania, while another report from Sub-saharan Africa too by Nademo et al.[26] obviates the need to encourage and call for more reportings of these cases from SSA (Sub-saharan Africa). Like the index case, the Tanzanian patient was positive for HIV infection. Such cases may reveal novel associations such as a possible link to viral infection, e.g HIV, and also improve our understanding of the disease presentation and its pathology.

The peculiar translocation between the chromosomes X and 18 which produces the fusion gene SS18-SSX – the known genetic lesion that undergirds synovial sarcoma is described as pathognomonic of this soft tissue sarcoma as referenced by many authors.[11-18]

Reports from West Africa were not left out of this review. Kessely et al (in Chad)[27] reported a rare case of thoracic spinal synovial sarcoma, whereas Babalola et al.[28] reported a case of posterior thigh sarcoma from the South western Nigeria region.

Although the mainstay of treatment is surgery, depending on the stage of the disease neoadjuvant chemotherapy and radiotherapy can be introduced in some cases.[11,27] Local recurrence and metastatic disease are quite common despite aggressive wide surgical excision that may be done initially. Consistent with this poor outcome was the case reported by Babalola et al, although the patient in this case was said to have had the largest tumour size so far and declined further adjuvant chemotherapy post-operation. He subsequently developed an aggressive recurrence, and passed away four months post excision.[28] The patient in the index report is still under follow up with the hope of taking adjuvant radiotherapy and chemotherapy, with a guarded prognosis in view.

CONCLUSION

While we stay expectant of the outcome of the patient reported on, the cases reviewed have highlighted the diagnostic challenges and issues encountered with the management and prognosis of synovial sarcoma from different parts of the world. Due to some associated deceptively benign features, many researchers admit it is one of the most readily misdiagnosed soft tissue sarcomas. To this end, Clinicians and Pathologists are called upon to appropriately entertain a high index of suspicion in order not to miss likely cases.

Recommendation

Researchers globally and particularly in the African subcontinents are by this report encouraged to communicate more case reports, and cohort study findings so as to enhance more understanding of the presentation and pathology of this disease. We also recommend that governments and partner organizations cooperate to provide or develop facilities and capacity for cytogenetic testing. This will enable multidisciplinary care teams and researchers to more accurately diagnose synovial sarcoma whilst overcoming the dilemma of misdiagnosis.

Consent

Informed consent was duly sought and obtained from the patient, whilst assuring of their confidentiality in publishing this report.

Declarations

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