

RELAPSE OF PRIMARY TESTICULAR NON-HODGKIN'S LYMPHOMA IN THE MANDIBLE – A CASE REPORT

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Abstract

Metastasis of primary tumors to the jaws from distant sites of the body is rare. In the majority of cases, the primary malignancy is already known at the time of admission while the occult primary may be detected only later. Cellular malignancies especially those of haematopoietic origin, diagnosed in the oral and maxillofacial region are usually found to be secondaries from various locations in the body. However non-Hodgkin's lymphoma (NHL) appearing in the mandible subsequent to a primary tumour elsewhere in the body is scantily reported in the literature.

We report a case of relapse of previously diagnosed testicular non-Hodgkin's lymphoma in the mandible. The patient had undergone treatment for primary testicular non-Hodgkin's lymphoma in the left testis one year ago. He remained symptom-free until the appearance of a tumour in the mandible. It is to be noted that there was no sign of metastasis elsewhere in the body other than the present lesion.

Key Words: Non-Hodgkin's lymphoma, relapse, metastasis, mandible, primary testicular lymphoma.

Introduction

Primary testicular non-Hodgkin lymphoma (PTL) comprises around 9% of testicular cancers and 1-2% of all non-Hodgkin lymphomas.(1) It primarily affects older men, with a median age at presentation of around 67 years.(1) By far the most common histological subtype is diffuse large B-cell lymphoma, accounting for 80-90% of PTLs.(1)

Specific homing, however, determines the site of many extra-nodal lymphomas, as exemplified by cutaneous T-cell lymphomas, which seem to be derived from skin-homing T-cells and mucosa-associated lymphoid tissue lymphomas that show features of the mucosal immune system.(2)

Testicular lymphoma often disseminates to other extranodal organs, such as contralateral testis, central nervous system (CNS), lung, pleura, Waldeyer's ring and soft tissues.(3)

The duration of survival after relapse

seems to be poor. Failures usually occurred within 1 to 3 years after the initial therapy. However, several late relapses have been observed up to 14 years after diagnosis, especially in the CNS and the contralateral testis.(4)

Involvement of Waldeyer's ring either as the extranodal primary focus or as part of more generalized disease, is far more frequent with NHL than with Hodgkin's disease (Banfi et al. 1972) and occurs in about 5% of cases at presentation (Rosenberg et al. 1961).(5)

The entities mimicking metastases of PTL are sarcoidosis, mushroom worker's lung, lymphoma and pheochromocytoma.(6) which need to be ruled out.

We present a case of non-Hodgkin's lymphoma of the mandible in an elderly male with a known history of testicular NHL for which he received treatment approximately one year ago. No sign of metastases was noted anywhere in the body until the appearance of the present lesion in the mandible.

Case Report

A 72-year-old male patient reported to the dental outpatient clinic with complaint of a painful mobile back tooth and swelling of the right side of face since 2 weeks (Fig 1). The swelling had been rapidly increasing in size.

On intraoral examination, a non tender diffuse growth was noted in relation to grade II mobile 48. The present size of the lesion was approximately 7 X 6 cm and extended from the retromolar trigone to the distal aspect of 43 enveloping the alveolus. The lesion was purplish in color and was boggy in consistency. The surface of the lesion was ulcerated (Fig 2). Homolateral lymph nodes were palpable.

The patient previously had non-Hodgkin's lymphoma of the left testis. It had been diagnosed as High grade diffuse large cell lymphoma and treated by left orchidectomy followed by chemotherapy with 4 cycles of CHOP comprising of Cyclophosphamide, doxorubicin (Hydrodoxorubicin), vincristine (Oncovin) and Prednisolone.

A CT scan of the mandible revealed a large osteolytic lesion involving the body which extended from the angle to the midline. The radiolucency was 5 X 3.5 X 3.5 cm in size and had an ill-defined, ragged border (Fig 3,4 & 5).

FNAC of the lesion was done initially (Fig 6). Biopsy of the lesion was taken from a nonulcerated area. The sections showed large cells (nuclei \geq twice the size of a small lymphocyte) in a diffuse growth pattern. The cells had vesicular nuclei, prominent nucleoli and a basophilic cytoplasm. There was moderate to high growth fraction. The features were suggestive of large cell lymphoma (Fig 7&8)

Then immunohistochemistry was undertaken which was positive for CD20 and negative for CD3, which conclusively proved the lesion to be as a B-cell lymphoma. Based on histopathology and immunohistochemistry, a diagnosis of relapse of NHL in the mandible was made.

Blood samples were drawn and biochemical examination was done for serum

lactate dehydrogenase (LDH) and liver function test (LFT). Serum LDH gave a value of 517 IU/L (Normal-164-412 IU/L) which favored the presence of secondaries. Serum bilirubin was 0.2-0.8mg% (Normal-0.34mg%). The routine blood picture showed Hb-9.5gm% which decreased to 7.5gm% after a week. Peripheral blood smear was done which was normal. To rule out metastasis in the internal nonpalpable lymph nodes, CT scans of the abdomen and chest were done. No abnormal findings were detected. Roentgenographic skeletal survey of limbs, chest, vertebral column and skull revealed no other secondaries. Bone marrow aspirate and biopsy of the posterior superior iliac crest showed no detectable abnormalities. Staging of non-Hodgkin's lymphoma was done by assessing - Tumor load - Extent of spread. This is not always an indicator of aggressiveness in NHL.

In accordance to the tables-1 & 2, the tumor was staged as I E (single location/extranodal) relapsed diffuse large B-cell lymphoma. The patient was then referred to an oncologist for treatment.



Fig.-1: Diffuse swelling on the right side of the face near the angle of the mandible.



Fig.-2: Diffuse ulcerative growth in the region of 46, 47, 48 was seen.



Fig.-3

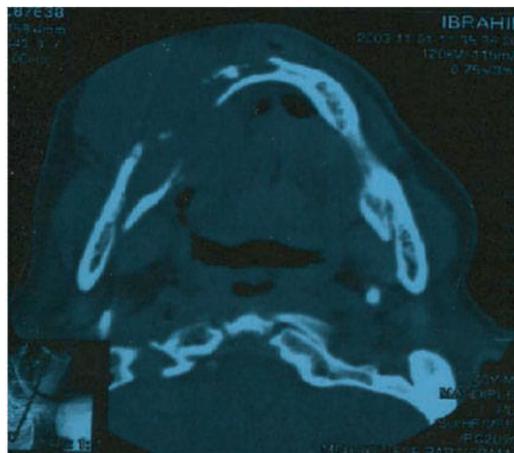


Fig.- 4

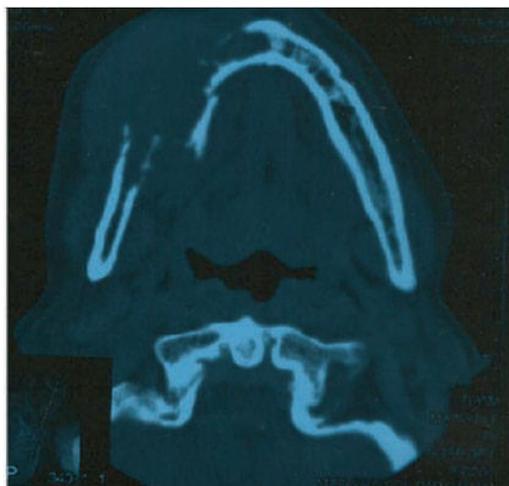


Figure- 5

Figure-3, 4, & 5: CT scans of the mandible in different plains showing large osteolytic lesion involving body of mandible.

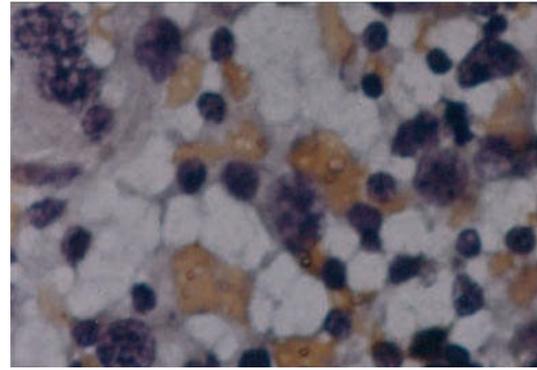


Fig-6: FNAC of the lesion

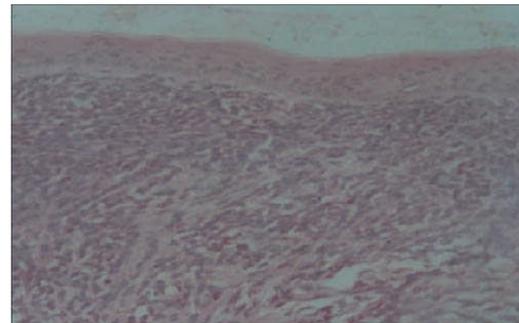


Fig-7: Hematoxylin & Eosin section (10x) showing sheets of atypical lymphoid cells.

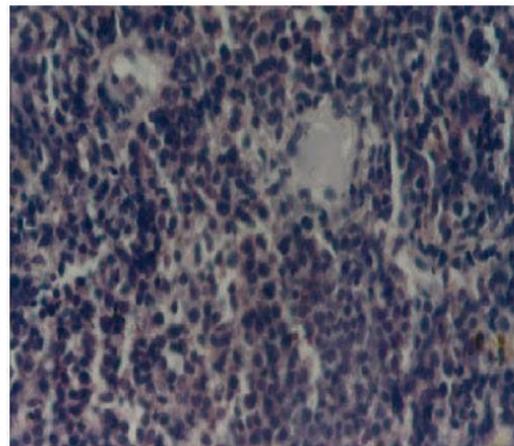


Fig-8: Hematoxylin & Eosin section (40x) showing sheets of atypical lymphoid cells.

Discussion

Primary testicular lymphoma represents 1% to 2% of all non-Hodgkin's lymphomas, with an estimated incidence of 0.26/100,000 per year. Approximately 85% of patients are older than 60 years of age. Even though lymphomas account for only 1% to 7% of all testicular malignancies, they represent the most common testicular tumor in men older than 50 years of age.

Histologically, 80% to 90% of primary testicular lymphomas are of diffuse large-cell type with B-cell immunophenotype (4).

Metastasis of primary tumours to the oral cavity is very rare and represents only 1% of all malignant oral tumours. In 25% of the cases, oral metastases are found to be the first sign of the metastatic spread; and in 23% of the cases, it is the first indication of an undiscovered malignancy at a distant site (8). Primary NHL of the bone is rare, accounting for <5%, and that of the mandible is 0.6% (9). Also, there is scanty information on the incidence of metastatic non-Hodgkin's lymphoma to the jaws. The major primary sites presenting oral metastases to the jaw bones in men were 22% in the lung and 11 % in prostate (10), while metastasis of malignancies from the testes comprise 3 % only (11).

Nearly all NHLs derive from lymphocytes, usually B-lymphocytes, few are of genuinely histiocytic (monocyte-macrophage) origin. Lesions of large and small lymphocytes are the lymphoblastic and lymphocytic lymphomas respectively. Lymphoblastic lymphomas have a poor prognosis with median survival from 06-11 years. Tumors of large cleaved cells have a more diffuse pattern with relatively slow progression. Non nodular (diffuse) lymphomas have a worse prognosis (5).

Lymphoblastic lymphomas (reviewed by Nathwani et al.1981) is a distinct lesion, more common in males than females. There is a biphasic age incidence – the second and third decade and the seventh decade. The lesion is a diffuse lymphoma composed of immature large lymphocytes or lymphoblasts whose nuclei have a delicate chromatin and a few nucleoli and sometimes a linear 'chicken's foot' accumulation of chromatin. Crush artifact is common and the cells often arrange in single files and infiltrate the capsule of the lymph node (5).

In NHL, the disease is not contiguous and may recur in any lymph nodes or extranodal site. Extranodal disease occurs

with greater frequency in NHLs (particularly diffuse type) than Hodgkin's disease and may account for nearly a quarter of cases. Widespread disease is more commonly seen with a diffuse than with a nodular histologic pattern (Jones et al. 1973) (5).

Anemia of a non-specific type with a normocytic, normochromic blood film can occur as a presenting feature in NHL; low Hb and elevated ESR at presentation are unfavorable prognostic indices. The patient often presents with bone pain or with local pressure effects and the lesions on radiological examination are usually osteolytic. Blood samples should be taken for liver function tests (serum bilirubin, alkaline phosphatase, liver enzymes, proteins); for urea, calcium and electrolyte assessment (looking particularly for renal abnormalities). Uric acid levels must be assessed before and after treatment (5)

To conclude, carcinomas and other sarcomas are most commonly seen as metastasis to the jaws. However, NHL in the jaws may in rare cases be a metastasis from a primary site and hence proper investigations of the underlying pathology and necessary screening should be done.

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