CASE REPORT

Benign Spindle Cell Tumour of Mandible and Points of Modification in Reconstruction with Nonvascularised Iliac Crest Graft

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Abstract Benign fibrous histiocytoma (BFH) is a rare spindle cell tumor occurring in the jaw. It is a benign lesion composed of spindle shaped fibroblasts arranged in a storiform pattern. Literature shows that there are less than hundred reported cases of BFH worldwide and only six reported cases in the mandible. In this article, we are reporting a rare case of BFH in mandible and novel points of modification in reconstruction with nonvascularised iliac crest graft.

Keywords Benign fibrous histiocytoma · Spindle cell tumor · Reconstruction · Iliac crest · Graft

Introduction

Benign fibrous histiocytoma (BFH) is a well-established mesenchymal tumor that was first described by Stout and Lattes [1]. Most commonly this tumor occurs on the skin of the extremities. Cutaneous BFH is a solitary, slowly growing nodule usually occurring in early to mid-adult life. It is a mesenchymal tumour that occurs predominantly on the skin as well as in bone including femur, tibia, and ilium [2].

According to the WHO histological classification of tumors, primary benign fibrous histiocytoma of bone is defined as a benign lesion composed of spindle shaped

fibroblasts, arranged in a storiform pattern, with a variable admixture of small, multinucleated osteoclast like giant cells. According to this classification, there are less than hundred reported cases of BFH worldwide and only six reported cases in the mandible [3, 4].

BFH has been reported in the oral and maxillofacial regions. The majority were found in the soft tissues [2]. All of the central lesions were located in the mandible within the angle and ramus. The lesions appeared as well-defined radiolucencies associated with clinical swelling.

In the extensive lesions, resection and reconstruction is the treatment of choice. The resection of a mandibular segment without adequate reconstruction produces serious esthetic and functional sequelae leading to a loss of quality of life. The use of non-vascularized iliac bone graft in reconstruction of mandibular defect is well recognized. The drawback with the use of this method includes donor site complications, failure of the bone graft and inability of the nonvascular graft to tolerate post operative radiation.

The graft success rate in non-vascularized iliac bone graft varies from 76 to 100 % [5]. Non-vascularized iliac bone grafts have been found to create a better contour and bone volume for facial esthetics and subsequent implant insertion, and may be the treatment of choice for reconstruction of defects <5 cm in diameter.

Case Report

A 46 year old female reported to the Department of Oral and Maxillofacial Surgery with the chief complaint of gradually increasing swelling of the posterior left mandible for a period of 3 months (Fig. 1). Intermittent and dull aching pain with frequent pus discharge was present for the last 3 days. She was under medications for diabetes and

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Fig. 1 Pre operative photograph

hypertension and the general physical examination revealed no other abnormalities.

Extraoral examination revealed presence of a diffuse swelling on the left side of the face. Regional lymph nodes were not palpable. On intraoral examination a well-defined, firm, non-tender swelling was present on the left side, extending from the mandibular first molar to the ascending ramus and causing expansion of the cortical plates. Also, loss of buccal vestibular depth was noted. The overlying mucosa was erythematous with pus discharge and the involved teeth did not show signs of mobility or tenderness.

Conventional radiographs revealed a well-defined multilocular radiolucency involving the left mandibular body and ascending ramus (Fig. 2). There was horizontal and vertical expansion with thinning of the cortex. The

Fig. 2 Pre operative OPG

computed tomographic (CT) scans showed bony destruction and the lesion demonstrated a heterogeneous soft tissue density mass expanding the mandible (Fig. 3). There was buccal and lingual cortical plate expansion with destruction of lingual cortical plate. According to the clinical, radiographic and initial histopathological features, our impression was that of a benign tumor.

Surgical removal of the lesion along with segmental resection was done followed by reconstruction with iliac crest. Submandibular surgical approach was modified by a step incision 2 cm below the inferior border of mandible. Layer by layer dissection was done to reach the body and ramus of the mandible (Fig. 4) and segmental resection was done for the complete removal of the lesion (Fig. 5). A reconstruction plate was molded in the mandibular arch before resection in order to preserve facial contour. Simultaneously with the resection another surgical team harvested an iliac crest bone graft. A step was made on the proximal and distal part of mandible and the reverse made on the graft for proper interlocking and contact of the graft. Multiple bur holes were made on the graft, an application of cranes principle to increase the osteoclastic and osteoblastic activity. Finally fixation was done with 2 cm continuous plate with bicortical screws and with 2 mm two hole plate (Figs. 6, 7). Layer by layer closure was done with vicryl and ethilon. Specimen was sent for histopathological studies (Fig. 8). After intermaxillary fixation patient was discharged on the third day post-op, without any complications.

Fig. 3 Pre operative CT

Fig. 4 Exposure

Fig. 5 Resected specimen

Fig. 6 Reconstruction

Discussion

Benign fibrous histiocytoma is a mesenchymal tumour that occurs predominantly on the skin of extremities, as well as

Fig. 7 Post operative OPG

Fig. 8 Histopathological specimen with whorled arrangement of plump spindle shaped cells and cells with a foamy cytoplasm resembling histiocytes

in bone including femur, tibia and ileum [4]. The tumour is rare in the jaw bone and only one has been reported in the maxilla and four in the mandible [6]. The etiology of oral BFH is obscure. Chronic irritation, continuous trauma, and spontaneous development have been reported for those located within the oral cavity. BFH has been recently included in the fibrohistiocytic tumours of the soft tissues that are further divided into cutaneous, noncutaneous and fibrohistiocytic tumours of bone. BFH is reported at any age with male preponderance of 2.5:1, mostly above 25 years [4]. The clinical features mimic those of painless solitary tumour which is slowly enlarging and can be progressive. Later it can present with cortical plate expansion and pain. Mostly it is accidentally diagnosed in radiographs (Fig. 9).

Radiographic appearance in the mandible can vary from well defined or ill defined unilocular radiolucency to multilocular radiolucency with or without hyperostotic border [2]. The differential diagnoses include ameloblastoma, myxoma and very importantly non-ossifying fibroma for its similarity in histopathological picture [6]. Due to the rarity

Fig. 9 Post operative image of the patient

of lesion, the characteristic radiographic findings make it difficult to differentiate BFH from other multilocular lesions of the mandible [7]. The final diagnosis should ultimately be based on histopathology and immunohistochemistry.

The treatment plan generally consists of wide surgical resection, but can vary considering the age and site. The prognosis for BFH is good and recurrence is seen only if there is incomplete removal of lesion. The recurrence rate of BFH is low. Bielasiewicz et al. [8] stated that only 2 cases (11 %) of 18 patients had a recurrence after a local excision in cases of BFHs occurring in the head and neck area and metastasis had never been reported. Reconstruction of mandibular defects represents a challenge due to the functional and cosmetic importance [9]. These deformities depend on the size and location of the defect. Free bone grafting was the first method of reconstructing mandibular defects and was initially reported by Bardenheuer in 1881 [10]. Evaluation of full extent of the proposed defect is the most important aspect.

Though the use of vascularised grafts still hold the gold standard, the lack of appropriate facilities and the high cost of these newer modalities has made the use of non vascularised iliac bone graft in the reconstruction of the mandible defect of smaller span (<5 cm) and lateral defects, a debatable option in developing countries. The success of the graft depends on the well adjusted stumps, well vascularised receiving bone borders, tight sealing of oral mucosa, stability and proper occlusion [11, 12].

The points of modification to increase the take of graft that is, step osteotomy for better incorporation, multiple bur holes to enhance vascularity and eventual resorption, initial reconstruction plate for contour and later with smaller dimension mini plates have been followed which we believe has helped in the survival of the graft.

Conclusion

The use of autogenous non-vascularised iliac bone grafts with the mentioned modification can give reasonable outcomes for mandibular lateral defects of <5 cm dimension. The gold standard vascularised grafts and novel developments such as distraction osteogenesis, tissue engineering and modulary endoprosthesis can be reserved for more larger, central, and residual defects of mandible.

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