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Case Report

Orthodontic movement of impacted cuspid in fibrodysplastic bone: A case report

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ABSTRACT

A case of orthodontic replacement of impacted cuspid in fibrodysplastic maxillary bone in a 12-year-old girl is reported. Fibrodysplastic bone is classically described as a fibrous bone without osteoblastic rimming. It is well known that orthodontic forces lead to tooth movement through proliferation and increased activity of bone cells (osteoblasts and osteoclasts). The reported case clinically support the latest histological studies suggesting that in fibrodisplastic bone osteoblasts are present but altered in shape and therefore difficult to recognize in sections.

In February 2003, a 12-year-old girl was referred to the Department of Oral and Maxillofacial Surgery, 2nd University of Naples, for evaluation and treatment of a painless, hard, right maxillary swelling. The swelling started on the right incisor-premolar region of the maxilla in September 2002, as a small, painless growth. The patient referred to a general dentist that noticed the inclusion of the right cuspid and proceeded to surgical exposition and direct bonding for orthodontic traction.

After the exposition of the impacted canine, the swelling increased in size resulting in asymmetry of the face prompting the patient to report to our department.

Extraoral examination showed bony, hard swelling of the right maxilla, extending from the right infra-orbital region to the upper lip. The nasolabial fold had been obliterated. The overlying skin was normal. Oral examination revealed a flattening of the upper buccal fold in canine area and the wire emerging between right first premolar and lateral incisor. The orthopantomogram revealed the impacted cuspid with the wire leaved from the previous surgery.

Computed tomography showed a mass with mixed aspect in right maxilla extending to right orbit and right maxillary sinus (Fig. 1B). A diagnosis of fibrous dysplasia was made on the basis of an incisional biopsy (Fig. 2A, B). The surgical resection was performed with recontouring of the maxilla, and sparing all the teeth (Fig. 3). Histological examination revealed compact, collagenous fibrous tissue associated with bone trabeculae with Chinese letter pattern; osteoblastic rimming was not evident. The orthodontic treatment of the impacted tooth was performed until the complete eruption (2 years later) (Fig. 4).

Although recent articles reported an adequate healing of fibrodysplastic bone in orthognatic surgery [1,2] at best of our knowledge there are no studies in English language on the orthodontic tooth movement in dysplastic bone.

Fibrous dysplasia (FD) of bone is a congenital, nonheritable skeletal disorder. The disease affects both sexes equally and usually is diagnosed in childhood or adolescence. Clinically, the lesions of fibrous dysplasia tend to become static as skeleton maturity is reached. The disease may affect one or several bones and may involve extraskeletal organs. Skeletal involvement may be limited to one bone (monostotic forms) or extended to multiple bones (polyostotic forms) or the entire skeleton (panostotic forms) [3].

The proximal part of the femur and craniofacial bones are the two most commonly affected sites. Fibrous dysplasia, in the stomatognatic apparatus, occurs more frequently in the maxilla than mandible [4,5].

At the bone tissue level, FD is characterized by dysplastic lesions that consist of abnormal fibrous tissue in the marrow space intertwined with poorly oriented, irregular trabeculae of woven bone [6].

FD as a disease of bone as an organ is a disease of skeletal stem cells. Clinically oriented descriptions would invariably bring in "altered bone remodeling" as the basis of the disease.

Classical histopathological features of FD consists of cellular fibrous tissue with spindle shaped cells and immature, isolated trabeculae of woven bone generally without rimming of osteoblasts [7].

Either way, two facts remained unnoticed: osteoblasts, of course, do form FD bone, but are altered in shape and therefore difficult to recognize in sections, and the "fibrous" tissue filling the marrow

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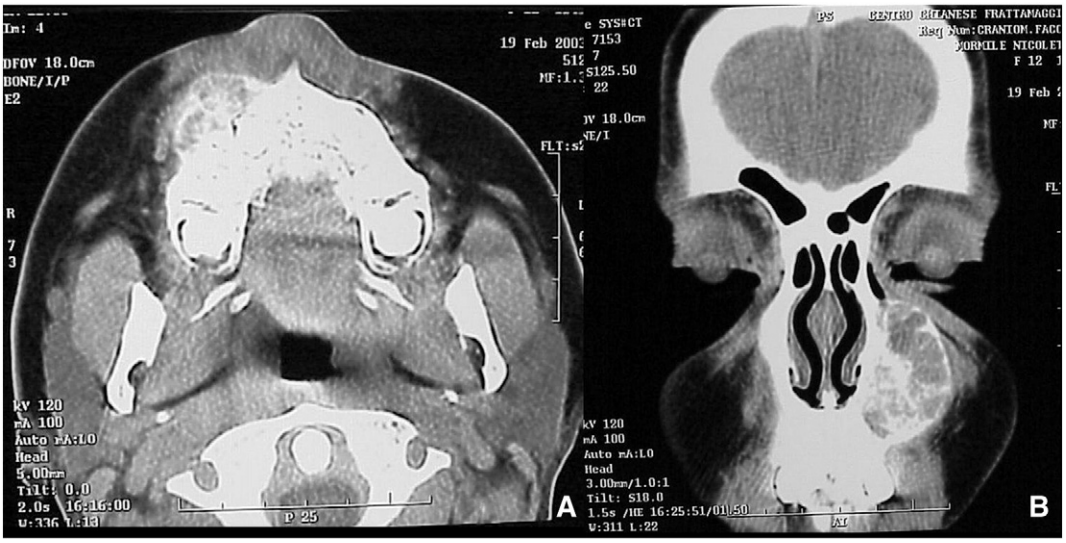

Fig. 1. Coronal and axial computed tomography scan of the lesion involving the maxilla from the right orbital rim above to the alveolar ridge below.

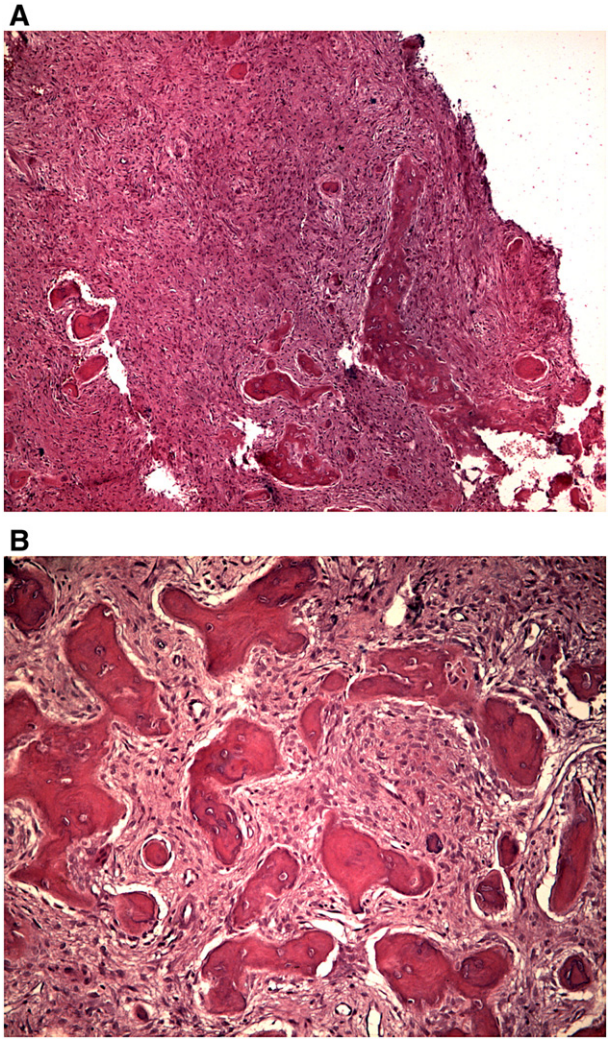

Fig. 2. Fibrous dysplasia consisting of small, principally nonmineralized, irregular and poorly oriented trabeculae of woven (immature) bone in bland cellular and collagenous matrix. A. 10×, B. 40× magnification.

spaces between the abnormal bone trabeculae is made of cells phenotypically resembling bone marrow stromal cells.

Whereas highlighting the basic fact that there is a change in the bone marrow structure in FD and that morphologically abnormal osteoblasts are likely abnormal in function as well, these observations opened the way to the adoption of a novel conceptual angle on the disease. This can now be seen as a disease of the osteoblastic lineage, and therefore of the skeletal (stromal) stem cells from which the lineage emanates [3].

FD occurs when bone marrow cells are affected by missense mutations. This leads to a block in the differentiation of the primitive bone marrow stromal cells (sometimes referred to as mesenchymal stem cells) to mature bone cells (osteoblasts and osteocytes) [8].

It has been recently reported the presence of activating somatic mutations of the Gsα gene in osteoblastic cells derived from fibrotic lesions in patients with monostotic fibrous dysplasia, suggesting that the mutation may induce abnormal osteoblastic cell proliferation or function in this disorder. Mutational activation of the α-subunit has been associated with increased proliferation in a number of endocrine tissues and in fibroblasts. It is, therefore, conceivable that mutations of the Gsα gene may induce abnormalities in the control of osteoblast growth and/or differentiation, resulting in fibrous dysplasia [9].

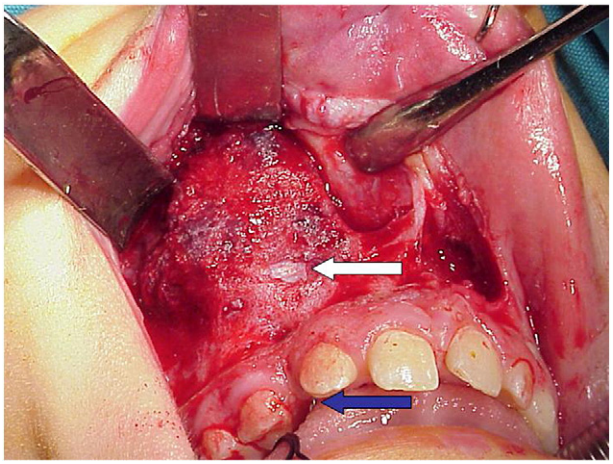

Fig. 3. Surgical recontouring of maxilla. Note the presence of the wire (blue arrow) for the orthodontic traction of the impacted cuspid (white arrow).

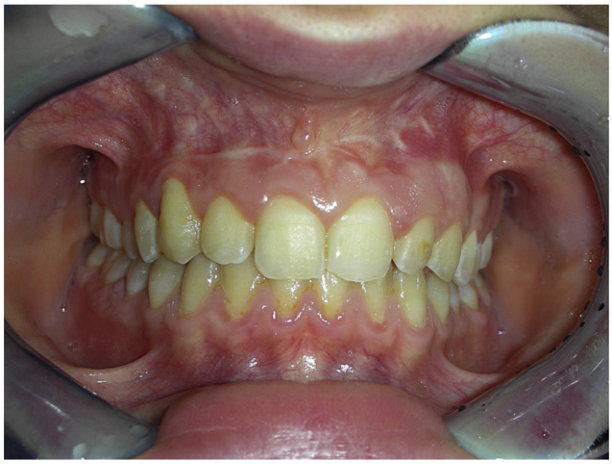

Fig. 4. The patient at the end of the orthodontic treatment.

It was recently recognized that FD of bone expresses the impact of the GNAS1 mutations in cells of osteoblastic lineage, such that each FD lesion can be seen as the result of abnormally functioning osteogenic cells in the bone/bone marrow environment [10].

It is known that bone cells (osteoblasts and osteoclasts) respond to orthodontic forces by proliferation and increased activity; however, the mechanisms for conversion of orthodontic forces into biologic activity are not completely understood [11].

Cells that produce specialized products usually appear round, a shape that might facilitate exposure of specific parts of their genome. In orthodontic tooth movement, such transformations in cellular shape are readily visible in mechanically stressed paradental cells. In unstressed periodontal ligament (PDL) sites, alveolar bone osteoblasts appear flat, while those in areas of PDL tension seem large and round.

In areas of PDL compression, PDL fibroblasts assume a round shape. Histologic studies by Reitan [12–14] and Rygh [15] have demonstrated that activated osteoblasts in PDL tension sites are engaged in producing a new bone matrix, while PDL cells in compression sites are primarily involved in enzymatic degradation of the compressed extracellular matrix [12,16].

Histologic studies made it clear that tissues can be remodeled only by the action of cells. In the case of the PDL, the cells that form and degrade the periodontal extracellular matrix are primarily the fibroblasts. In the case of the alveolar bone, the cells that remodel it are the osteoblasts, osteoclasts, and osteocytes. Osteoclastic activity is quite intense in such a case, and it is the activity of these multinucleated cells that removes the alveolar bone that stands in the way of the moving teeth.

The fibroblasts, in areas of tension, proliferate and synthesize new matrix components, and in areas of compression they degrade the necrotic PDL.

However, in both sites of tension and compression, the fibroblasts seem to produce factors that activate neighboring bone cells. Recent evidence suggests that osteoclasts are regulated by factors derived from adjacent osteoblasts, and PGE2 was proposed as being a major part of this bridge.

Thus, in tooth movement, PDL fibroblasts may not only be responsible for the remodeling of the periodontal matrix, but may also be actively involved in the regulation of the activity of the cells that remodel the alveolar bone. Osteocytes also seem to be sensitive to applied loads, and it was suggested that these cells, which are capable of recognizing and responding to molecular reorientation in their surrounding matrix, communicate these alterations to bone surface cells (primarily osteoblasts), providing them with an osteogenic stimulus [12].

Despite the fibrous dysplasia affect osteoblastic activity and proliferation, the present case suggests a normal response of dysplastic bone to the orthodontic forces and thus the possibility of an orthodontic treatment.

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