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Fibro osseous lesions: a review on classification system and proposal of new classification

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Abstract
Benign fibro-osseous lesions of the maxillas constitute a varied group of lesions with a common histological characteristic: the substitution of normal bone by tissue composed of collagen and fibroblasts, with variable amounts of a mineralized substance that may be bone, cementum or both. On the basis of histo-pathology these lesions comprise fibrous dysplasia, periapical cemento-osseous dysplasia, focal cementoosseous dysplasia, florid cemento-osseous dysplasia and cemento-ossifying fibroma. The term fibro-osseous is descriptive, nosologically limited and diagnostically non-specific. Nomenclature, classification and diagnosis of these lesions is problematical, partly because of a lack of agreement about terminology, but also because of a significant overlap in histological features. The group includes developmental and reactive or dysplastic lesions as well as neoplasms. A number of classifications have been proposed by various workers like Waldron , Speight, Carlose , Eversole , Reichart (WHO) etc but non have been universally accepted.

Key Words
Benign, Fibro, Maxillas, Dysplasia, WHO, focal cementoosseous

Introduction

The term Fibroosseous lesions is a generic designation of a group of disorders characterized by the replacement of normal bone by connective tissue, with varying degrees of mineralization in the form of woven bone or cementum-like round acellular basophilic structures.[1]

Waldron in 1970 described fibro osseous lesions as a group of pathological changes with in the jaw bones in which normal bone is replaced by fibrous tissue ,with or with out calcification.[2] Fibro-osseous lesions (FOL) of the jaws constitute a group of conditions which are remarkable for their clinico-pathological similarities.[1] FOLs of the face and jaws include ossifying fibroma, fibrous dysplasia, cementoossifying fibroma, benign cemento-blastoma and periapical cemental dysplasia (also known as periapical fibrous dysplasia). They differ, with the exception of fibrous dysplasia, from those found in the rest of the skeleton. Fibro – osseous lesion may be non neoplastic or neoplastic & of odontogenic or non odontogenic origin.[3]

A number of classifications have been proposed by various workers like Waldron, Speight, Carlose , Eversole , Reichart (WHO) etc but non have been universally accepted. .In 1972, Eversole concluded that fibrous dysplasia and cemento-ossifying fibroma are clinically and radiologically distinct disease entities that nevertheless are not always histologically distinguishable.[4] In 1972, the World Health Organization (WHO) considered ossifying fibroma to be a tumor of bone origin, and cementifying fibroma a tumor of odontogenic origin.[5] However, in 1992, the WHO grouped such lesions under the common denomination of cemento-ossifying fibromas, on the grounds that they represented histological variants of one same type of lesion.[5]

Till date fibro-osseous lesions are a poorly defined group of lesions affecting the jaws and craniofacial bones. All are characterized by the replacement of bone by cellular fibrous tissue containing foci of mineralization that vary in amount and appearance. Various workers have not agreed on an exact terminology, classification , clinical features , histopathology and radiographic features, but a concept has emerged which has culminated in the latest WHO classification[5]. The core of this classification is the concept of a spectrum of clinicopathological entities in which the diagnosis can only be made on the basis of a full consideration of clinical, histological and radiological features.[2] It is important to appreciate that a histologic diagnosis of fibro-osseous lesion is non-specific and has a limited value in predicting biologic behavior or guiding treatment. A specific diagnosis i important because of the different treatment modalities available for different lesions.[3]

Discussion:

Regardless of subtype, all fibro-osseous lesions demonstrate replacement of normal bone by fibrous connective tissue with an admixture of mineralized product including osteoid, mature bone, and or cementum like calcifications.[6] Thus histologic diagnosis of a fibro-osseous lesion is in many cases, relatively uncomplicated. The main challenge lies in the classification of fibro-osseous lesions. The late Charles Waldron wrote “In absence of good clinical and radiologic information a pathologist can only state that a given biopsy is consistent with a fibro-osseous lesion. With adequate clinical and radiologic

information most lesions can be assigned with reasonable certainty into one of several categories."[7] Conversely in the absence of such information Eisenberg and Eisenbud stated that "pathologists today will often rightly decline to render a definitive diagnosis. Instead, the pathologist will resort to the noncommittal designation of benign fibro-osseous lesions. This is the only acceptable approach considering the potential for inappropriate treatment otherwise." Therefore the identification of the majority of fibro-osseous lesions is made upon clinical and radiological features and their classification is not merely an academic exercise because the therapeutic management of fibro-osseous lesion varies depending on the actual disease process. To further complicate matters, a number of other disease processes demonstrate clinical, radiographic, and microscopic features that bear resemblance to those encountered in recognized fibro-osseous conditions.[8]

Since 1930's, numerous classifications have been proposed and variety of lesions have come under the umbrella of fibro-osseous lesion, which includes developmental lesions, reactive lesions, and benign fibro-osseous neoplasms.[7] In the first edition of the WHO classification of "odontogenic tumours" (1971) four lesions containing cementum-like structures were identified,[9] and they were benign cementobolastoma (true cementoma) ,cementifying fibroma, periapical cemental dysplasia (periapical fibrous dysplasia) and the gigantiform cementoma (familial multiple cementoma).They were grouped under benign category as cementomas which in turn was placed within the "neoplasms and other tumours related to the odontogenic apparatus" category. Other lesions, those are frequently histologically indistinguishable from those four lesions, fibrous dysplasia and ossifying fibroma, were placed in the category of "neoplasms and other tumours related to bone".

The observation of identical cementum-like tissue in lesions in extra-gnathic sites suggested that this tissue may be a merely normal variant of bone,[10] and that dental cementum itself is a specialized form of "bundle-bone". Therefore, in the second edition of the WHO's classification in 1992, three of the "cemental" lesions were transferred to the "neoplasms and other tumours related to bone" (periapical cemental dysplasia, florid cemento-osseous dysplasia,other cemental dysplasias) group, leaving the benign cementoblastoma (true cementoma) as the sole true neoplasm of dental cementum.[9]

Although the term fibro-osseous lesion is not mentioned by the authors of WHO's second edition, their broad reclassification of these lesions, based both on behaviour and histopathology, is entirely consistent with Waldron's recommendations made in 1985.[11]

The fibro-osseous lesions are now a subset of "neoplasms and other tumours related to bone". Historically, the nosology of fibro-osseous lesions has been fraught with inconsistency, confusion, and a seemingly endless array of terminology. However, a classification of fibro-osseous lesions proposed by Waldron has gained wide recognition over the years and remains, to date, the most accepted.[6] Recently, Brannon and Fowler proposed a slightly modified categorization of fibro-osseous lesions, which, with further study, eventually may become the standard. Nevertheless, despite advances in our understanding of these conditions, occasional lesions still defy classification.[4]

Depending upon above mentioned features, various investigators have attempted to classify FOL. Some have included lesions originating from PDL or medullary bone, others have included lesions containing giant cells and nongiant cells (pure fibro-osseous) Many other attempts at classification have been offered in the past, but in light of newly described entities and changing concepts, Waldron classified FOL as –

Charles A. Waldron (1985)[10]

I. Fibrous dysplasia
   A. Polyostotic
   B Monostotic
II. Fibro-osseous (cemental) lesions presumably arising in the periodontal ligament
   A. Periapical cemental dysplasia
   B. Localized fibro-osseous-cemental lesions (probably reactive in nature)
   C. Florid cement-osseous dysplasia (gigantiform cementoma)
   D. Ossifying and cementifying fibroma
III. Fibro-osseous neoplasms of uncertain or debatable relationship to those arising in the periodontal ligament (category 11)
   A Cementoblastoma,osteoblastoma , and osteoid osteoma
   B. Juvenile active ossifying fibroma and other so called aggressive, active ossifying / cementifying fibromas

In essential agreement with Waldron's classification, many investigators believe that other entities are also within the spectrum of FOL, such as Chronic diffuse sclerosing osteomyelitis, Cherubism, Aneurysmal bone cyst and Central giant cell granuloma.

The list of "additional entities" seems almost endless and therefore will not be further pursued. Suffice it to say that lesions with no appreciable fibrous or osseous component do not fulfill the criteria for FOL as defined by Waldron.Later, to simplify the controversy of FOL, World Health Organization (1991)[12] put forward a simplified classification:

1. **Osteogenic neoplasm** – Cemento-ossifying fibroma
2. **Non - neoplastic –**
   a) Fibrous dysplasia
   b) Cemento-osseous dysplasia
      i) Periapical cemento-osseous dysplasia
      ii) Florid cemento-osseous dysplasia
      iii) Focal cemento-osseous dysplasia
3. **Cherubism**

Brannon and Fowler (2001)[3] gave another classification which was quite different from that of Waldron and WHO classification. This was done to include more number of lesions which were also showing features like FOL

I. Osseous Dysplasia (OD) (Reactive)
Nonhereditary
- Periapical
- Focal
- Florid

Hereditary (developmental)
- Familial Gigantiform
- Cementoma

II. Fibro-osseous neoplasms
Ossifying fibroma (OF)
"Juvenile", "active" or aggressive variant of OF

III. Fibrous displasia (FD)
Polyostotic FD

Monostotic FD
Craniofacial FD

IV. Giant cell lesions
Central giant cell granuloma
Aneurysmal bone cyst
Cherubism

IV. Miscellaneous benign fibro-osseous lesions
Cementoblastoma
Tori/exostoses
Osteoma

Later on, to overcome the demerits of his classification, Waldron reviewed the subject of BFOL in 1993 and suggested a modification of his earlier classification. More recently, Slater, Slootweg, Eversole and Melrose have made recommendations or modifications in classifying FOL. Based on the aforementioned classification by Waldron, the following categorization is suggested:

Fibro-Osseous Lesions by Waldron (1993)[11]

I. Osseous dysplasia (OD)
Nonhereditary
- Periapical
- Focal
- Florid Hereditary
- Familial gigantiform cementoma

II. Fibro-osseous neoplasms
- Conventional ossifying fibroma (OF)
- So-called "juvenile" or "aggressive" forms OF

III. Fibrous dysplasia (FD)
- Polyostotic FD with endocrinopathy
- Polyostotic FD
- Craniofacial FD

This modified classification has merit, but further study and evaluation of FOL are needed.[5]

Pieter J. Slootweg & Hellmuth Muller (1996)[13]

Group I : Fibrous dysplasia
Group II : Juvenile ossifying fibroma
Group III : Ossifying fibroma
Group IV : Cemento-osseous dysplasia

This classification lays emphasis primarily on the histopathological features, and they underscore that this classification requires inclusion of adjacent normal bone to make a diagnosis. However in the absence of this, the clinical and radiological features have to be taken in to consideration.

Paul M Speight and Roman Carlos (2006) gave a classification based on new WHO classification also from Waldron (1993), Slootweg (1996) and Brannon and Flower (2001). The core of the classification given by WHO in 2005 was concept of a spectrum of clinic-pathological entities in which diagnosis can only be made on the basis of a full consideration of clinical , histological and radiological features. Although the terminology was still problematic so Speight and Carlos used a new classification and concentrated on the histopathological features that may guide the working surgical pathologist towards a diagnosis.

Paul M Speight and Roman Carlos (2006)[14]

A. Fibrous dyplasia
Monostotic fibrous dysplasia
Polyostotic fibrous dysplasia
Craniofacial fibrous dysplasia

B. Osseous dysplasias
Periapical osseous dysplasia
Focal osseous dysplasia
Florid osseous dysplasia
Familial gigantiform cementoma

C. Ossifying fibroma
Conventional ossifying fibroma
Juvenile trabecular ossifying fibroma
Juvenile psammomatoid ossifying fibroma

Another Classification of benign fibro-osseous lesions of the craniofacial complex was given by Roy Eversole in 2008. This classification includes neoplasms , developmental dysplastic lesions and inflammatory/reactive processes. The basis of this classification is that definitive diagnosis can rarely be rendered on basis of histopathological features alone rather, procurement of a final diagnosis is usually dependent upon assessment of microscopic, clinical and imaging features together.

Roy Eversole (2008)[15]

I. Bone dysplasias
   a. Fibrous dysplasia
      i. Monostotic
      ii. Polyostotic
      iii. Polyostotic with endocrinopathy (McCune-Albright)
      iv. Osteofibrous dysplasiaa
   b. Osteitis deformans
   c. Pagetoid heritable bone dysplasias of childhood
   d. Segmental odontomaxillary dysplasia
II. Cemento-osseous dysplasias
   a. Focal cemento-osseous dysplasia
   b. Florid cemento-osseous dysplasia
III. Inflammatory/reactive processes
   a. Focal sclerosing osteomyelitis
   b. Diffuse sclerosing osteomyelitis
   c. Proliferative periostitis
IV. Metabolic Disease: hyper parathyroidism
V. Neoplastic lesions (Ossifying fibromas)
   a. Ossifying fibroma NOS
   b. Hyperparathyroidism jaw lesion syndrome
   c. Juvenile ossifying fibroma
      i. Trabecular type
      ii. Psammomatoid type

Although, there is no universally accepted classification, Waldron's classification has been the most useful classification. Apart from these lesions as classified by Waldron, there are some other lesions which can also be characterized under the heading of FOL. So, for the description purpose, we have devised a working classification -

A. Developmental
   1. Solitary bone cyst
   2. Cherubism
   3. Osseous Choristoma
B. Reactive or Reparative
   I. Central (Intraosseous)
      1. Traumatic periostitis
      2. Garre's Osteomyelitis
      3. Sclerosing Osteomyelitis
      4. Periapical cemento-osseous dysplasia
      5. Central giant cell granuloma
      6. Aneurysmal bone cyst
   II. Peripheral (extraosseous)
      1. Peripheral giant cell granuloma
      2. Myositis Ossificans
C. Neoplasm
   1. Compact & Cancellous osteomas
   2. Osteoid Osteomas
   3. Osteoblastomas
   4. Benign cementoblastoma
   5. Ossifying fibroma
   6. Juvenile ossifying fibroma
      I. Trabecular type
      ii. Psammomatoid type
D. Endocrinal or Metabolic
   1. Brown tumor of hyper parathyroidism
E. Unknown Etiology
   1. Fibrous dysplasia
      i. Monostotic

ii. Polyostotic
iii. Polyostotic with endocrinopathy (McCune-Albright)
iv. Osteofibrous dysplasia
2. Paget's disease
3. Segmental odontomaxillary dysplasia

Precise diagnosis requires good clinical, radiological and histological correlation. This remains the most accepted and favored method of diagnosis. With the development of sophisticated immunological markers, we can expect a whole new wave of diagnostic splitting in approach to Fibro Osseous Lesions.[6]

Conclusion

Nomenclatures for benign Fibro osseous lesions have been historically been inconsistent and confusing so far. In recent years, significant progress has been achieved in understanding the histopathogenic similarities and differences of various fibro-osseous lesions, thereby enhancing one's ability to diagnose accurately and to manage many fibro-osseous conditions, including craniofacial Fibrous dysplasia, Ossifying fibroma, Focal and Florid osseous dysplasia. There is still a need for clarification of many aspects of these perplexing group of lesions. Elimination of confusing clinical terminology, such as "juvenile" and "aggressive," in histologic diagnosis and questionable terms, such as "cemento-" and "cementifying," for bone-forming neoplasms, would be a start in clearing the conflict.

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