2. Historical Background

Our present understanding of the concept of fibro-osteo-cemental lesions is the result of many years of treatment and observation. The process can be said to have its origins both in the individual observations of such surgeons as Syme [1828], Maisonneuve [1856], Ferguson [1865], Menzel [1872] (fig. 1), Bryant [1874] etc., and in later synoptic works such as those by Hyfelder [1857], Weber [1866], Bayer [1884], and Kentenich [1896]. These were later followed by the first attempts to consider the clinically observed phenomena from a morphological point of view. In 1864, for example, Virchow introduced the term 'leontiasis ossea' to describe diffuse enlargements of the maxillo-facial bones. He chose this term because he had observed that patients suffering from 'hyperostosis' often had a monstrous, almost leonine facial appearance, similar to that observed in cases of leprous leontiasis. Though undoubtedly useful, Virchow's work could not claim the distinction of having characterized an etiologically defined pathological entity. A breakthrough in the field of osteopathology was made by Paget in 1877, when he defined the clinical and pathological characteristics of an entity which he called 'osteitis deformans'. Though largely ignored on the Continent, this entity gained rapid recognition in Britain. One of the main reasons for the Europeans' initial unwillingness to accept

Fig. 1. 35-year-old woman with osteofibroma operated on by A. Menzel (Vienna, 1870). The tumor had been growing for more than 25 years [from Menzel, 1872].

Paget's findings is undoubtedly the diverging
approach taken by Friedrich Daniel von
Recklinghausen (fig. 2). His monumental
work was dedicated to Rudolf Virchow and
was published on the 13th of October 1891,
to mark the occasion of the latter’s 70th
birthday. In his treatise, von Recklinghausen

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Fig. 2. Friedrich Daniel von Recklinghausen
(1833-1910).

[1891] introduces the term ‘osteitis fibrosa’,
and applies it to all the cases of skeletal
lesions he had observed where the bone marrow
was either partially or totally transformed
into fibrous tissue. This would of
course include those cases whose fundamental
morphology is identical to that of Paget's
disease. Christeller [1926] speaking to a
meeting of the Deutsche Gesellschaft fr Pathologie,
put forward this unitarist point of
view as late as 1926, though he did differentiate
between two forms of ‘osteitis fibrosa’.
One was hyperostotic and could in fact be
taken to correspond to Paget's disease, while
the other was hypostotic and corresponded to
‘osteodystrophia fibrosa’. On the very same
day, Schmorl [1926] who was also present,
managed to produce empirical proof that
these two bone lesions were not only clinically
different, but that they were also fundamentally
dissimilar both from a pathological
and an anatomical point of view. His findings
were based on 25 autopsy cases. In the
same year, the unitarist approach was refuted
for once and for all by findings made by
Mandl [1926]. He was the first person to succeed
in removing an adenoma of the parathyroid
gland from a patient with osteitis fibrosa
generalisata, thus contributing substantially
to a better understanding of this systemic
bone lesion. This newly developed parathyroid
 gland surgery could thus be used in conjunction
 with chemical and physiological examinations
 to isolate the syndrome of hyperparathyroidism.
 These investigations made it
 possible to isolate this syndrome conclusively
 and also to establish that it was (a)
 characteristic of osteitis fibrosa generalisata,
 and (b) in no way related to Paget's osteitis
 deformans. This, however, was but a first
 step. It soon emerged that a number of conditions,
 which should in fact have been classified
 in a separate group of their own, were
 often being diagnosed as 'osteitis fibrosa generalisata'.
 Indeed, if one looks through old
 case histories or old medical journals, one
 occasionally comes across cases which are
 described as 'atypical Recklinghausen',
 'doubtful Recklinghausen', 'Recklinghausen's
 localized fibrocytic osteitis' or 'Recklinghausen
 without lesion in the parathyroid
 gland'. Faced with this muddled terminology,
 Konjetzny[1922] introduced the term 'osteitis
 fibrosa localisata', with a view to distinguishing
 localized processes featuring fibrous
 bone transformation from more generalized
 alterations. However, since this resulted in
 the most diverse bone lesions being diagnosed
 either as 'osteitis fibrosa localisata' or

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 'osteitis fibrosa generalisata' merely according
 to whether they occurred generally or
 locally, Konjetzny himself soon decided to
 reject this definition.
 Montgomery[1927] was the first to use
 the term 'ossifying fibroma'. He used this
term to denote those tumorous conditions
 occurring in the maxillo-facial bones which
 were sharply delineated and featured fibrosis
 and a degree of new bone formation. He
describes 3 cases of his own and cites a further 14 cases drawn from the literature which he regards as similar.

Jaffe [1933] described a number of cases of osteitis fibrosa cystica which were remarkable in that they did not feature tumors of the parathyroid gland. He suggested that these cases were not classifiable either as Recklinghausen's bone disease or as Paget's disease, and that they should be regarded as a third and separate condition which had yet to be described in greater detail. At the time, he assumed that these conditions were of congenital origin.

Stafne [1933] described the radiological and clinical characteristics of 35 cases of cemental lesions in the jawbones. Stafne [1934] went on to describe what he termed 'periapical osteofibrosis', a condition which is characterized, histologically, by the simultaneous formation of new bone and fibrous tissue. In 1937, Thoma described a number of cases of cementoblastoma of the maxillo-facial bones, dividing their development into three distinct stages. In the same year, Phemister and Grimson [1937] published a study of 13 cases which they regarded as typically characteristic of 'fibrous osteoma'. In addition to these, their study also reevaluated two of Montgomery's [1927] cases and cited a further 30 drawn from the literature. The authors formulated the hypothesis that the relationship between 'fibrous osteoma' and the desmal bones was analogous to that already observed between osteochondroma and the hyaline bones. They also considered the possibility that compact osteoma might in fact be the final stage in the development of 'fibro-osteoma'.

Albright et al. [1937] published
a study dealing with five cases of a bone lesion which was characterized by osteitis fibrosa disseminata, large patches of abnormal skin pigmentation and endocrine dysfunction accompanied by pubertas praecox.

Lichtenstein [1938] described the characteristics of another condition which he termed 'polyostotic fibrous dysplasia'. A detailed and comprehensive comparison between the skeletal alterations associated with the condition described by Albright et al. [1937] and those which characterize 'fibrous dysplasia' as described by Lichtenstein [1938] shows that the two are absolutely identical. Both authors have observed exactly the same alterations to the bones of the jaw and skull. Eden [1939] published a study covering a number of cases of maxillo-facial bone lesions in which he concluded that 'ossifying fibroma' was in fact an immature form of 'benign fibro-osseous tumor of the membrane bone'. Uehlinger [1940] (fig. 3) published what is undoubtedly the most significant study of fibrous dysplasia to be written in German. He was the first to publish findings based on autopsy material secured from a patient with fibrous dysplasia. He described the condition as 'osteofibrosis deformans juvenilis'. One of the most important findings which this autopsy produced was that it was able to prove that the hypothalamus and the parathyroid gland were normal.

Lichtenstein and Jaffe [1942] again described the clinical, radiological and histological characteristics of fibrous dysplasia, in a new study based on an expanded body of case material which now comprised 24 cases. Thus, by the early 1940s, it was already apparent that von Recklinghausen's [1891]...
term 'osteitis fibrosa' would have to be divided into three quite distinct pathological entities. These were: osteitis deformans Paget, osteodystrophia fibrosa generalisata (primary and secondary hyperparathyroidism) and fibrous dysplasia in both its monostotic and polyostotic forms. Concerning the latter, it should be noted that the possibility of polyostotic forms of fibrous dysplasia also being accompanied by endocrinal disorders (Albright's syndrome) had also been recognized. All these alterations are known to occur in the bones of the jaw and skull. Besides the three already mentioned (Paget's disease, Recklinghausen's bone disease and fibrous dysplasia), other tumorous entities, such as cementoblastoma [Thoma, 1937], ossifying fibroma [Montgomery, 1927] and fibro-osteoma [Phemister and Grimson, 1937], had already been isolated by that time and were also known in the literature. Reparative alterations of the maxillo-facial bones had also been recognized as such, and had been diagnosed as chronic, hypertrophic, nonpurulent osteomyelitis [Axhausen, 1934]. Because of the morphological similarity between this condition and Paget's disease, Axhausen [1934] uses the term 'pseudo-Paget'. Regrettably, this process of pluralistic differentiation of these conditions then more or less came to a halt. In fact, subsequent work carried out in this field tended towards a more unitarist view. No sooner had the term 'fibrous dysplasia' been introduced as an entity, than a whole series of authors began to voice the assumption that ossifying fibroma and fibrous dysplasia were closely related. A study by Jaffe and Lichtenstein published as early as 1942 regarded the characteristics of several cases presented by Phemister and Grimson [1937] as indistinguishable from those associated with fibrous dysplasia. It
was in the same year, 1942, that Mallory conjectured that fibro-osteoma and fibrous dysplasia were in fact one and the same entity.

Schlumberger [1946a] published a study of 67 cases of monostotic fibrous dysplasia in which he put forward the view that 'ossifying fibroma' and 'nonossifying fibroma' of the bone were nothing other than two versions of one single entity, namely fibrous dysplasia.

In a second study, Schlumberger [1946b] made one of the first attempts to classify all the various fibro-osseous lesions of the bone as different versions of the same pathological process. Mammel [1948], in his histological study of ossifying fibroma and fibrous dysplasia, concurs with Schlumberger [1946b] on this point.

Over the past 30 years, this unitarist approach has resulted in the generic term 'fibro-osseous lesions of the maxillo-facial bones' being evolved [Berger and Jaffe, 1953; Cooke, 1957; Hamner et al., 1968; Panders, 1970; Waldron, 1970; Schmaman et al., 1970; Cangiano et al., 1971; Meister et al., 1973; Waldron and Giansanti, 1973a, b; Abrams and Melrose, 1975; Boysen et al., 1979]. It has been used to cover not only tumors (osteofibroma, ossifying fibroma, cementifying fibroma), but also inflammatory and reparative processes (pseudo-Paget, periostitis ossificans Garr), dysplastic alterations (fibrous dysplasia) and even some conditions whose etiological characteristics are unclear (Paget's disease, periapical cemental dysplasia).