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 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 für 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 '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 endocrinial 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

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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).