REVIEW

Survival of dental implants in patients with bone dysplasia: A systematic review

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ABSTRACT

Objective: This study used published studies to assess the survival rate of dental implants placed in patients with bone dysplasia of the maxillofacial region.

Material and methods: An electronic search without a specified date range was performed using the MEDLINE, PubMed, EMBASE, Web of Science, and Cochrane databases. No gender or age restrictions were applied.

Results: Eighteen publications were found that met the study's criteria, reporting data on 18 patients with bone dysplasia including cleidocranial dysplasia (CDD), fibrous dysplasia (FD), florid cemento-osseous dysplasia (FCOD), and odonto-maxillary segmental dysplasia (SOMD), who received a total of 130 implants, an average of 7.2 implants/patient (range 1 to 16). The mean age of the patients was 36.7 years (range 15 to 70 years). For implants placed in bone dysplasia, the survival rates were 100% for patients with CDD (n = 8), FD (n = 5), SOMD (n = 2), FCOD with implants inserted far from the lesions (n = 2) and 0% for dental implants inserted within FCOD (n = 1). The mean follow-up was 38.2 months (min 6, max 60).

Conclusions: Dental implants placed in patients with dysplastic bone lesions show high survival rates, similar to those in the general population for CDD, FD, and SOMD. For FCOD, the failure rate was 100%.

Key Words: Dental implants, Bone, Osseous dysplasia, Fibrous dysplasia, Florid cemento-osseous dysplasia, Bone diseases, Treatment outcome, Osteomyelitis

1. INTRODUCTION

Since the introduction of dental implants, we have learned that the initial quality and quantity of alveolar bone directly influence the success of the implants.^[11] Their osseointegration also depends on bone remodeling,^[2] which can be altered in certain bone conditions. Today, many different bone dysplasias are known, some of which also affect the alveolar bone. One of the possible lesions is characterized by anarchy of bone remodeling, leading to bony structure and stability changes. Hence, it is essential to identify to

what extent bone dysplasias influence the osseointegration and survival of implants.

2. MATERIAL AND METHODS

We applied the PRISMA checklist and established a focused question according to the PICO scheme (patient, intervention, comparison, outcome) as follows: Do patients with bone dysplasia in the jaw region (P) undergoing dental implantation (I) have a similar implant survival (O) compared to the average population (C).

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We included all available pro- and retrospective longitudinal studies up to July 2020.

Our local ethics committee exempted the study from its review process because we did not use any primary patient data from our clinic.

2.1 Research strategy

We searched MEDLINE, PubMed, EMBASE, Web of Science, and Cochrane library using an open date ending with July 2020. The keywords were: "dental implant" AND "dysplasia;" "dental implant" AND "bone dysplasia."

2.2 Inclusion and exclusion criteria

The following articles were included: prospective randomized controlled trials, cohort studies, case series, case reports. The exclusion criteria included: secondary data from metaanalysis and systematic reviews; reports with one specific bone lesion with less than two articles published in the entire medical literature.

2.3 Variables

The Primary outcome was implant survival. The secondary outcomes included the number of patients, age, sex, number of implants, bone augmentation, the material of bone augmentation, and follow-up time.

2.4 Choice of articles

Two authors (AP, SD) independently identified the articles according to the criteria mentioned above. An initial assessment was done based on the titles. The abstracts of the selected papers were then read. Finally, all articles that matched the inclusion criteria were read, and data was extracted from those that remained appropriate.

3. RESULTS

3.1 Literature review

The initial search retrieved 397 articles, 274 of which were excluded (235 not matching the topic, 39 animal studies). There were 105 duplications, with a remainder of 18 pieces that were used for the final analysis (see Figure 1).



Figure 1. PRISMA flow chart describing the data selection

3.2 Types of bone dysplasias

The publications ranged from 1997 to 2019 and were primarily case reports. The four types of conditions included cleidocranial dysplasia, fibrous dysplasia, florid cemento-

osseous dysplasia, and segmental odonto-maxillary dysplasia. These are discussed separately in the following sections. The articles are summarized in Table 1.

Table 1. Features	of the included	studies $(N = 18)$
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Articles	Dysplasia	Number of patients	Age (years)	Sex M/F	Number of implants	Bone augmentation	Material of bone augmentation	Implant Survival rate (%)	Follow-up time (months)
Lombardas & Toothaker 1997 ^[3]	cleido-cranial dysplasia	1	15	Н	10	Yes	Iliac crest	100	6
Bektor et al. 2002 [4]	segmental odonto-maxillary dysplasia	1	16	F	1	No		100	6
Petropoulos et al. 2004 ^[5]	cleido-cranial dysplasia	1	42	F	16	Yes	Autogenous bone (Unspecified)	100	6
Petropoulos et al. 2011 [6]	cleido-cranial dysplasia	1	45	F	14	Yes	Autogenous bone (unspecified) + DFDBA	100	60
Whitt et al. 2011 [7]	segmental odonto-maxillary dysplasia	1	17	F	2	No		100	96
Bencharit et al. 2013 [8]	florid cemento-osseous dysplasia	1	58	F	5 (Implants placed far from the lesions)	Yes		100	36
Mendoca & Platas 2014 [9]	fibrous dysplasia	1	35	F	6	Yes	Iliac crest + DFDBA	100	84
Noh et al. 2014 [10]	cleido-cranial dysplasia	1	27	F	2	No		100	60
Petrocelli & Kretschmer 2014 ^[11]	fibrous dysplasia	1	13	F	6	Yes	Iliac crest	100	36
Schnutenhaus et al. 2015 ^[12]	cleido-cranial dysplasia	1	47	Н	12	No		100	
Sosin et al. 2015 [13]	fibrous dysplasia	1	29	F	3	Yes	Iliac crest + DFDBA		
Atil et al. 2018 [14]	cleido-cranial dysplasia	1	48	F	10	Yes	Iliac crest		
Esfahanizadeh & Yousefi 2018 ^[15]	florid cemento-osseous dysplasia	1	62	F	2 (Implants placed far from the lesions)	No		100	18
Bajwa et al. 2018 [16]	fibrous dysplasia	1	32	F	16	No		100	60
Ahmad et al. 2019 [17]	cleido-cranial dysplasia	1	24	F	14	No		100	36
Adnot et al. 2019 [18]	fibrous dysplasia	1	64	F	5	No		100	24
Ambard et al. 2019 [19]	cleido-cranial dysplasia	1	17	F	4	No		100	36
Shin et al. 2019 ^[20]	florid cemento-osseous dysplasia	1	70	F	2 (Implants placed within the lesions)2 (Implants placed far from the lesions)	No		0 100	9

3.2.1 Cleido-cranial dysplasia (CCD)

A total of 82 implants were found in 8 patients. 6 were male, 2 were female. On average, 10 implants were placed for each patient with a range of 2 to 16 implants. The mean age was 33 years, the field 15 to 48 years. In addition, bone augmentation was performed in 4 patients (2 patients with bone from the iliac crest; 2 patients with autologous bone mixed with demineralized freeze-dried bone allograft (DFDBA).

Implant survival was 100%. The average follow-up time was 34 months (2.8 years), ranging from 6 months to 60 months (5 years). In 4 studies, the follow-up time was more than 12 months,^[6, 10, 17, 19] in 2 studies more than 6 months,^[3,4] and in 2 studies, the follow-up time is unknown.^[12, 14]

3.2.2 Fibrous dysplasia (FD)

All 5 patients were female and treated with a total of 36 implants. On average, 7.2 implants were placed with a range from 3 to 16 implants. The average age was 36.4 years (range = 13 to 64 years). In addition, bone augmentation was performed in 3 patients (1 patient with bone from the iliac crest; 2 patients with bone from the iliac crest mixed with DFDBA).

Implant survival was 100%. The mean survival was 51 months (4.25 years) with a minimum of 24 months to 84 months (7 years). Four studies had a follow-up time of more than 12 months.^[9, 11, 16, 18] In one article, the follow-up time is unknown.^[13]

3.2.3 Florid cemento-osseous dysplasia (FCOD)

11 implants were inserted in 3 female patients with an average of 3.7 implants (range of 2 to 5 implants). The mean age was 63 years ranging from 58 to 70 years. In one patient, 2 implants were placed remotely from the dysplastic area;^[15] in another patient, 2 implants were inserted in a diseased and healthy region, respectively.^[20] The third patient was treated with partial excision of dysplastic bone before 5 implants were placed remotely from the residual lesions.^[8]

Implants inserted in the diseased areas all as failed early to lack of osseointegration in the 8 first weeks after implant placement resulting in a success rate of 0%.^[20] However, the 9 implants inserted remotely had a 100% success rate. The mean follow-up was 27 months (2.3 years) for these implants, ranging from 18 to 36 months.

3.2.4 Segmental odonto-maxillary dysplasia

Two female patients received 3 implants. One patient was 17 years old, and the age is unknown for the second patient. The survival rate was 100% after a mean follow-up of 51 months (4.25 years), ranging from 6 to 96 months (8 years).

4. DISCUSSION

4.1 Cleido-cranial dysplasia

Cleidocranial dysplasia is a rare bone disease that occurs in 1 in a million patients.^[21] It is caused by an autosomal dominant mutation of the CBFA1 gene, also known as RUNX2 on the 6p21 chromosome, and 30%-40% are de novo mutations.^[21] Clinical signs are supernumerary teeth, late tooth eruption, a hypoplastic maxilla, hypoplastic or completely missing clavicular bones, brachycephaly, and abnormal closure of the fontanelles.^[22–24] Most of the craniofacial effects only develop during adolescence,^[25] leading to delayed diagnosis of otherwise healthy patients.^[26,27]

The treatment from a dental and maxillofacial perspective aims to reestablish masticatory function and dental aesthetics.^[28] Early-onset of orthodontic treatment in children is more likely to be successful alone,^[29,30] while adult patients require implant-borne prosthetic restorations.

In the last two decades, more articles dealing with implantborne restorations have been published, some of which report implants combined with various types of bone augmentation due to disease-typical atrophy of the alveolar bone.^[5, 6, 14]

Our review shows a 100% success rate for dental implants in 8 patients with up to 5 years of follow-up.

4.2 Fibrous dysplasia

Fibrous dysplasia was first described by von Recklinghausen in 1891 and named osteitis fibrosa generalization, and it represents 2.5% of all bone diseases with an equal male/female ratio.^[31] The etiology is genetic with a mutation of the GNAS1 gene (guanine nucleotide-binding protein, alpha stimulating) on the 20q13 gene.^[32] From a pathophysiological perspective, this mutation induces an abnormal differentiation of osteoblasts over several biochemical steps resulting in dysplastic bone.^[31] In other words, the healthy bone may be replaced progressively by the more fragile dysplastic variant.

There are different forms of the disease. The first affects only one monostotic site, while the polyostotic form involves several areas.^[33] Fibrous dysplasia is also part of the McCune-Albright syndrome and endocrine effects and cutaneous "café au lait" macules.^[16] Rarely is a biopsy necessary to confirm the diagnosis since conventional or CT imaging is highly typical.^[34] Imaging shows well-circumscribed lesions with a ground-glass matrix and either completely sclerotic or lucent bone depending on the cellular composition.^[34]

Our review found a 100% success rate for dental implants in 5 patients with up to 7 years of follow-up.^[9,11,13,16,18] However, it was observed that the contact between diseased bone and the implants is less stable than in healthy bone.^[35–37] Hence, several authors suggested inserting longer implants (up to 16 mm) for compensation.^[11,16] Other precautions included low-speed drilling, ample irrigation, and late loading of the implants at 6 months.^[18] The latter time point is questionable since all loading strategies (2-12 months) were successful.^[9,11,13,16,18]

4.3 Florid cemento-osseous dysplasia

Florid cemento-osseous dysplasia is a rare fibro-osseous lesion^[38] described first in 1976 by Melrose.^[39] Many names were given to this pathology, but it was renamed FCOD in the second edition of the WHO classification of odontogenic tumors in 2013.^[40] The exact etiology remains unclear, but genetic factors are probable since familial predisposition.^[41,42]

Regarding pathophysiology, normal bone is progressively replaced by avascular cementoma-like tissue with impaired capacity for regeneration.^[38] The typical patient is female, of African origin, and aged 30 to 50 years.^[42] The lesions are frequently bilateral in the posterior mandible.^[38] Most are discovered by chance on routine dental radiographs and are generally asymptomatic.^[41–43] However, they can also present as secondary infections with pus, fistula, and sequestra.^[43,44] In the absence of such complications, no surgical treatment is required.

In conventional or CT imaging, radio-lucent, radio-opaque, or mixed lesions are present and typical enough that biopsy is rarely needed to confirm the diagnosis.^[40]

In our review, there were two distinct groups of patients. One included patients with implants inserted within the lesions, and the other had implants that were far from the lesions. In the first group (n = 1), only one patient had an early loss of both inserted implants.^[20] In contrast, the second group (n = 2) had a 100% success rate with an observation time from 18 to 36 months.^[8, 15] Hence, the bone remote to the affected area seems sufficient to allow implantation.^[8, 15]

4.4 Segmental odonto-maxillary dysplasia

Segmental odonto-maxillary dysplasia is a sporadic mesoectodermal dysplasia apparent in early life, possibly even in utero, with a male predominance and unknown etiology.^[45]

The condition is characterized by enlarged connective tissues and/or bone on one or both sides of the maxilla, with possible asymmetry of the face.^[7] The bone lesions are sclerotic, dense, and not well circumscribed due to enlarged vertical trabecular bone.^[7] In addition, developmental problems such as delayed tooth eruption and congenital agenesis of the premolars have been associated with the condition.^[45]

The two reported cases in our review had a 100% success rate up to 8 years of observation. Thus, dental implants seem to be an excellent therapeutic option in this group.^[4,7]

5. CONCLUSION

With a success rate of 100%, dental implants are a valid therapeutic option in CCF, FD, SOMD, and FCOD if not placed within the affected site. Dental implants inserted within FCOD were shown to fail in 100% of patients reported in this review. The limitation of the present review is represented by the small number of reported cases resulting from the rarety of such bone lesions. In addition, the fact that the published studies are based only on one patient finding.

CONFLICTS OF INTEREST DISCLOSURE

The authors declare they have no conflicts of interest.

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