



# Case Report Jacob's Disease: Case Series, Extensive Literature Review and Classification Proposal

Luca Raccampo <sup>1</sup>, Giorgio Panozzo <sup>2</sup>, Alessandro Tel <sup>1</sup>, Michele Di Cosola <sup>3</sup>, Gianluca Colapinto <sup>4</sup>, Lorenzo Trevisiol <sup>2</sup>, Antonio D'agostino <sup>2</sup>, Salvatore Sembronio <sup>1,\*</sup> and Massimo Robiony <sup>1</sup>

- <sup>1</sup> Maxillofacial Surgery Department, Academic Hospital of Udine, Department of Medicine, University of Udine, 33100 Udine, Italy
- <sup>2</sup> Section of Oral and Maxillofacial Surgery, Department of Surgical Sciences, Dentistry, Gynaecology and Paediatrics, University of Verona, 37129 Verona, Italy
- <sup>3</sup> Section of Dentistry and Dental Prosthetics, Department of Medicine, University of Foggia, 71122 Foggia, Italy
- <sup>4</sup> Independent Researcher, 70121 Bari, Italy
- \* Correspondence: salvatore.sembronio@uniud.it

**Abstract:** Jacob's disease is a rare entity consisting of the formation of a pseudojoint between an abnormal coronoid process of the mandible and the inner surface of the zygomatic bone. First described by Jacob in 1899, its diagnosis and definition have never been entirely univocal. In this paper, we present three emblematic cases and an extensive review of the literature on Jacob's disease. Given the variability observed in the presentation of the disease, we have developed a proposal for the classification, here reported.

Keywords: Jacob's disease; coronoid hyperplasia; osteochondroma; temporomandibular surgery



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# 1. Introduction

In 1899, Oscar Jacob first described the eponymous disease. He noticed, as a postmortem relief, a restricted jaw range of motion in a patient caused by an impingement between the inner side of the zygoma and the coronoid process of the mandible (CPM) [1]. He observed that the two bony surfaces were joined by bands of fibrous tissue, mimicking a new joint. Nearly 50 years earlier, in 1853, Langenbeck described the first case of an enlargement of the CPM [2]. Subsequently, several cases of Jacob's disease (JD) have been reported in the literature but without a clear and unambiguous definition of the condition ever being given. This confusion over the classification of the different forms of coronoid enlargement and JD is partly due to the misuse of the terms osteoma (OS), cartilage-capped exostosis (CCE) and osteochondroma (OC) and the latter's apparent link with JD [3]. Differences in the proportion of cartilage and bone elements in the sample justified different histological diagnoses. All these pathological entities, as well as true coronoid hyperplasia (CH), can determine an enlargement of the coronoid and potentially lead to JD. While, for some authors, the presence of an osteochondroma of the CPM must be a prerequisite for diagnosing JD [4–7]; this is not a defined and agreed point. In fact, it is defined by most as the formation of a pseudojoint between the inner surface of the zygoma and the CPM, which can be deformed or elongated by several pathological processes [8–14]. The authors report here three cases of JD that express well the broad spectrum of the pathology, as well as an extensive and meticulous literature review, aiming to propose a possible clinical and radiological classification of this disease.

## 2. Review of the Literature

After having established the definition of JD as a condition where a pseudojoint forms between an abnormal coronoid process and the inner aspect of the zygoma, with a broad spectrum of presentation and pathology, we conducted a comprehensive literature review to identify the cases that matched this definition or were already identified as JD by the authors. Articles were retrieved from PubMed, Cochrane and Semantic scholar databases using the following search terms: "Jacob's disease", "Coronoid Osteochondroma", "Coronoid hyperplasia", "Coronoid process pseudojoint" and "Coronoid process enlargement". Articles were also retrieved indirectly by screening the references of papers identified through the aforementioned keywords. The results were filtered, limiting the selection to papers written in the English language. Cases in which a certain JD diagnosis could not be established through author declaration, imaging or inferences were excluded. Full texts of all articles meeting the inclusion criteria were thoroughly reviewed for data extraction. We extracted reports of age, gender, the side and type of the CPM anomaly, MMO before and after surgery, surgical treatment approach, follow-up period and reported recurrence. A total of 116 cases, including the 3 cases hereafter reported, were selected from a total of 107 articles, and the data collected are summarized in Table 1 [1,4,6–8,11–111].

**Table 1.** Results of the literature review performed, reporting various parameters of interest that will be discussed in the discussion section.

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16et al. [27]195820////////noidectomy reportedreportedreported17Shira et al. [28]195814MR/L/8/Extraoral coro- noidectomyNotNot18Lebo [29]196118MLOC1429Extraoral coro- noidectomyNotNot19Mohnac [30]196218MR/LOC18/Extraoral coro- noidectomyNotNot20Van de Vijver [31]196218/R/L///Intraoral coro- noidectomyNotNot21Dechaume et al. [32]196413/////Intraoral coro- noidectomyNotNot22Rottke et al. [33]196729/////Intraoral coro- noidectomyNotNot23Allan et al. [34]196732FRCCE1640Intraoral coro- noidectomyNotNot24Allan et al. [34]196722MLCCE1340Intraoral coro- noidectomyNotNot25Meyer [35]197210FROC20/Intraoral coro- noidectomyNotNot		Chemin								Intraoral coro-	Not	Not
17Shira et al. [28]195814MR/L/8/Extraoral coro- noidectomyNotNot18Lebo [29]196118MLOC1429Extraoral coro- noidectomyNotNot19Mohnac [30]196218MR/LOC18/Extraoral coro- noidectomyNotNot20Van de Vijver [31]196218/R/L///Extraoral coro- noidectomyNotNot21Dechaume et al. [32]196413//////Intraoral coro- noidectomyNotNot22Rottke et al. [32]196729/////Intraoral coro- noidectomyNotNot23Allan et al. [34]196732FRCCE1640Intraoral coro- noidectomyNotNot24Allan et al. [34]196722MLCCE1340Intraoral coro- noidectomyNotNot25Meyer [35]197210FROC20/Intraoral coro- noidectomyNotNot	16	et al. [27]	1958	20	/	/	/	/	/	noidectomy	reported	reported
17Shira et al. [25]195814MK/L/8/noidectomyreportedreported18Lebo [29]196118MLOC1429Extraoral coro- noidectomyNotNot19Mohnac [30]196218MR/LOC18/Extraoral coro- noidectomyNotNot20Van de Vijver [31]196218/R/L////Intraoral coro- noidectomyNotNot21Dechaume et al. [32]196413//////Intraoral coro- noidectomyNotNot22Rottke et al. [33]196729//////Intraoral coro- noidectomyNotNot23Allan et al. [34]196732FRCCE1640Intraoral coro- noidectomyNotNot24Allan et al. [34]196722MLCCE1340Intraoral coro- noidectomyNotNot25Meyer [35]197210FROC20/Intraoral coro- noidectomyNotNot	17	China at al [20]	1059	14	м	D/I	/	0	/	Extraoral coro-	Not	Not
18Lebo [29]196118MLOC1429Extraoral coro- noidectomy or reportedNotNot19Mohnac [30]196218MR/LOC18/Extraoral coro- noidectomy reportedNotNot20Van de Vijver [31]196218/R/L////Intraoral coro- noidectomy reportedNotNot21Dechaume et al. [32]196413//////Intraoral coro- noidectomy reportedNotNot22Rottke et al. [33]196729//////noidectomy reported reportedreported23Allan et al. [34]196732FRCCE1640Intraoral coro- noidectomy reported reportedNot noidectomy reported reported24Allan et al. [34]196722MLCCE1340Intraoral coro- noidectomy reported reportedNot noidectomy reported reported25Meyer [35]197210FROC20/Intraoral coro- noidectomy reported reportedNot noidectomy reported reported	17	Shira et al. [28]	1958	14	11/1	K/L	/	8	/	noidectomy	reported	reported
10       Leto [27]       101       10       M       L       OC       14       25       noidectomy       reported       reported         19       Mohnac [30]       1962       18       M       R/L       OC       18       /       Extraoral coro- noidectomy       Not       Not         20       Van de Vijver [31]       1962       18       /       R/L       /       /       /       Intraoral coro- noidectomy       Not       Not         21       Dechaume et al. [32]       1964       13       /       /       /       /       /       Intraoral coro- noidectomy       Not       Not         22       Rottke et al. [32]       1967       29       /       /       /       /       /       /       noidectomy       reported reported         23       Allan et al. [34]       1967       32       F       R       CCE       16       40       Intraoral coro- noidectomy       Not       Not         24       Allan et al. [34]       1967       22       M       L       CCE       13       40       Intraoral coro- noidectomy       Not       Not         25       Meyer [35]       1972       10       F       R	18	Lebo [29]	1961	18	м	т	00	14	29	Extraoral coro-	Not	Not
19Mohnac [30]196218MR/LOC18/Extraoral coro- noidectomy reportedNotNot20Van de Vijver [31]196218/R/L///Intraoral coro- noidectomy reportedNotNot21Dechaume et al. [32]196413//////Intraoral coro- noidectomy reportedNot22Rottke et al. [33]196729/////Intraoral coro- noidectomy reportedNot23Allan et al. [34]196732FRCCE1640Intraoral coro- noidectomy reportedNot noidectomy reported24Allan et al. [34]196722MLCCE1340Intraoral coro- noidectomy reportedNot noidectomy reported25Meyer [35]197210FROC20/Intraoral coro- noidectomyNot	10		1701	10	141	L	00	14	27	noidectomy	reported	reported
20       Van de Vijver [31]       1962       18       /       R/L       /       /       Intraoral coro- noidectomy       Not       Not         21       Dechaume et al. [32]       1964       13       /       /       /       /       /       Intraoral coro- noidectomy       Not       Not         22       Rottke et al. [33]       1967       29       /       /       /       /       /       Intraoral coro- noidectomy       Not       Not         23       Allan et al. [34]       1967       32       F       R       CCE       16       40       Intraoral coro- noidectomy       Not       Not         24       Allan et al. [34]       1967       22       M       L       CCE       13       40       Intraoral coro- noidectomy       Not       Not         25       Meyer [35]       1972       10       F       R       OC       20       /       Intraoral coro- noidectomy       Not       Not	19	Mohnac [30]	1962	18	М	R/L	OC	18	/	Extraoral coro-	Not	Not
20       Variate Vijver       1962       18       /       R/L       /       /       /       Intraorial coro-       Not       noidectomy       reported         21       Dechaume et al. [32]       1964       13       /       /       /       /       /       /       Intraorial coro-       Not       Not         22       Rottke et al. [33]       1967       29       /       /       /       /       /       Intraorial coro-       Not       Not         23       Allan et al. [34]       1967       32       F       R       CCE       16       40       Intraorial coro-       Not       Not         24       Allan et al. [34]       1967       22       M       L       CCE       13       40       Intraorial coro-       Not       Not         25       Meyer [35]       1972       10       F       R       OC       20       /       Intraoral coro-       Not       Not		Van de Viiven								noidectomy	reported	reported
21       Dechaume et al. [32]       1964       13       /       /       /       /       //       Intracriticity       Not noidectomy       reported         22       Rottke et al. [33]       1967       29       /       /       /       /       /       Intraoral coro- noidectomy       Not       Not         23       Allan et al. [34]       1967       32       F       R       CCE       16       40       Intraoral coro- noidectomy       Not       Not         24       Allan et al. [34]       1967       22       M       L       CCE       13       40       Intraoral coro- noidectomy       Not       Not         25       Meyer [35]       1972       10       F       R       OC       20       /       Intraoral coro- noidectomy       Not       Not	20	[31]	1962	18	/	R/L	/	/	/	noidectomy	reported	reported
21       betal. [32]       1964       13       /       /       /       /       /       Intribute of the indication of the indindication of the indication of the indicatio		Dechaume								Intraoral coro-	Not	Not
22       Rottke et al. [33]       1967       29       /       /       /       /       Intraoral coro- noidectomy       Not       Not         23       Allan et al. [34]       1967       32       F       R       CCE       16       40       Intraoral coro- noidectomy       Not       Not         24       Allan et al. [34]       1967       22       M       L       CCE       13       40       Intraoral coro- noidectomy       Not       Not         25       Meyer [35]       1972       10       F       R       OC       20       /       Intraoral coro- noidectomy       Not       Not	21	et al. [32]	1964	13	/	/	/	/	/	noidectomy	reported	reported
22       et al. [33]       1967       29       /       /       /       /       noidectomy       reported       reported         23       Allan       1967       32       F       R       CCE       16       40       Intraoral coro-       Not       Not         24       Allan       1967       22       M       L       CCE       13       40       Intraoral coro-       Not       Not         24       Allan       1967       22       M       L       CCE       13       40       Intraoral coro-       Not       Not         25       Meyer [35]       1972       10       F       R       OC       20       /       Intraoral coro-       Not       Not	22	Rottke	1067	20	/	/	/	/	/	Intraoral coro-	Not	Not
23Allan et al. [34]196732FRCCE1640Intraoral coro- noidectomyNotNot24Allan et al. [34]196722MLCCE1340Intraoral coro- noidectomyNotNot25Meyer [35]197210FROC20/Intraoral coro- noidectomyNotNot	22	et al. [33]	1967	29	/	/	/	/	/	noidectomy	reported	reported
et al. [34]     100     10     10     noidectomy     reported     reported       24     Allan et al. [34]     1967     22     M     L     CCE     13     40     Intraoral coro- noidectomy     Not     Not       25     Meyer [35]     1972     10     F     R     OC     20     /     Intraoral coro- noidectomy     Not     Not	23	Allan	1967	32	F	R	CCE	16	40	Intraoral coro-	Not	Not
24     Allan et al. [34]     1967     22     M     L     CCE     13     40     Intraoral coro- noidectomy     Not     Not       25     Meyer [35]     1972     10     F     R     OC     20     /     Intraoral coro- noidectomy     Not     Not	_0	et al. [34]	1707		•		002		10	noidectomy	reported	reported
25 Meyer [35] 1972 10 F R OC 20 / Intraoral coro- Not Not	24	Allan	1967	22	М	L	CCE	13	40	Intraoral coro-	Not	Not
25 Meyer [35] 1972 10 F R OC 20 / Initiadia toro- Not Not		et al. [34]								Induction Intraoral core	Not	Not
noidectomy reported reported	25	Meyer [35]	1972	10	F	R	OC	20	/	noidectomy	reported	reported

Case No.	Author	Year	Age	Gender	Side	Туре	MMO before Treatment (mm)	MMO after Treatment (mm)	Treatment	Treatment Follow-Up (Months)	Treatment Recurrence
26	James	1974	52	F	R	OC	5	40	Extraoral coro-	12	No
27	Cooper	1074	42	г	т	00	1	1	Intraoral coro-	Not	Not
27	et al. [37]	1974	43	F	L	UC	0	/	noidectomy	reported	reported
28	et al. [38]	1975	14	F	L	OC	10	/	Extraoral coro- noidectomy	Not reported	Not reported
29	Singer et al. [39]	1976	64	М	R	OC	5	/	Intra/extraoral coronoidec- tomy	Not reported	Not reported
30	Ramon et al. [40]	1977	45	М	R	OC	/	/	Intra/extraoral coronoidec- tomy	Not reported	Not reported
31	Michel et al. [41]	1977	30	F	R	/	10	/	Intraoral coro- noidectomy	Not reported	Not reported
32	Norman et al. [42]	1980	21	М	L	OC	3	/	Intraoral coro- noidectomy	Not reported	Not reported
33	Ito et al. [43]	1981	20	F	L	OC	3	/	Extraoral coro-	Not	Not
34	Boland	1983	25	/	/	/	/	/	Intraoral coro-	Not	Not
25	et al. [44]	1002	22	E	T	00	7		Intraoral coro-	reported	reported
35	Isnii et al. [45]	1983	32	F	L	UC	1	41	noidectomy Intra/extraoral	reported	reported
36	Ishii et al. [45]	1983	53	М	R	OC	20	37	coronoidec- tomy	Not reported	Not reported
37	Revington [46]	1984	24	М	R	CCE	9	/	Extraoral coro- noidectomy	6	reported
38	Tucker et al. [47]	1984	16	М	R	СН	22	/	Intraoral coro- noidectomy	Not reported	Not reported
39	Macleod [48]	1987	46	F	/	CCE	5	/	Intraoral coro- noidectomy	Not reported	Not reported
40	Schwartz et al [49]	1987	15	М	L	OC	18	/	Extraoral coro-	Not	Not
41	Halling	1989	22	М	R	/	21	/	Intraoral coro-	Not	Not
40	et al. [50] Goudot	1000	45		T	,		,	noidectomy Intraoral coro-	reported Not	reported Not
42	et al. [51]	1989	45	/	L	/	/	/	noidectomy	reported	reported
43	et al. [52]	1990	37	F	L	OC	2	28	noidectomy	reported	reported
44	Rames et al. [53]	1990	36	/	R/L	СН	/	/	Intraoral coro- noidectomy	Not reported	Not reported
45	Asanami et al. [54]	1990	17	М	L	CCE	8	64	Intra/extraoral coronoidec- tomy	36	Not reported
46	Honig et al. [55]	1993	22	М	R	CH	21	/	Intraoral coro- noidectomy	Not reported	Not reported
47	Kerscher et al. [56]	1993	45	М	L	OC	14	32	Intraoral coro- noidectomy	Not reported	Not reported
48	Smyth et al. [57]	1994	15	М	R/L	СН	4	40	Extraoral coro- noidectomy	96	Yes (96 months)
49	Gibbons	1995	34	М	R/L	OC	19	34	Intraoral coro-	Not	Not
50	Cenetoglu	1004	10	м	т	00	10	19	Intraoral coro-	reported	Not
50	et al. [59] Kormor	1990	19	IVI	L	00	12	40	noidectomy	12 Not	reported
51	et al. [60]	1996	40	М	L	OC	/	/	noidectomy	reported	reported
52	Mizumoto et al. [61]	1996	43	М	L	OC	11	49	Extraoral coro- noidectomy	12	Not reported
53	Constantinides et al. [62]	1997	31	М	R	OC	10	/	Extraoral coro- noidectomy	12	Not reported
54	Gross et al. [63]	1997	22	М	L	OC	20	40	Intraoral coro-	Not reported	Not
55	Chen et al. [64]	1998	57	F	L	OC	14	42	Extraoral coro-	72	No
56	Takenobu	1998	56	F	L	OC	2	30	Intraoral coro-	12	Not
57	Manganaro	1998	26	F	L	OC	/	/	Intraoral coro-	Not	Not
58	[66] Ishii et al. [67]	1998	25	М	R	СН	22	40	noidectomy Intraoral coro-	reported Not	reported
50	Chicareon	1000		141	n	00		1	noidectomy	reported Not	reported Not
59	et al. [68] Hernandoz-	1999	5	M	K	00	/	/	Not reported	reported	reported
60	Alfaro et al. [8]	2000	22	М	L	OC	21	52	Extraoral coro- noidectomy	6	Not reported
61	Escuder y de la Torre et al. [14]	2001	19	М	L	/	7	/	Intraoral coro- noidectomy	Not reported	Not reported

# Table 1. Cont.

Table 1. Cont.

Becklery of brokki Becklery of Prokki Becklery Decket Prokki Becklery Decket Becklery D	Case No.	Author	Year	Age	Gender	Side	Туре	MMO before Treatment (mm)	MMO after Treatment (mm)	Treatment	Treatment Follow-Up (Months)	Treatment Recurrence
66.         Prinkli Pr	62	Escuder y de la Torre et al. [14]	2001	16	/	R/L	OC	14	49	Intraoral coro- noidectomy	12	No
interking         Lineking         Dots         21         M         R         OC         is         40         Line(s) extremal cores         Note reported         Note reported         Note reported         Note reported         Note reported         Note reported         Note reported         Note reported           66         Maxamaba et al. [1]         205         23         F         L         OC         12         41         Constrained reported         12         Not reported           66         Maxamaba et al. [1]         205         23         K         L         OC         12         41         Constrained         Not reported         Not r	63	Emekli et al. [4]	2002	26	F	R	OC	10	35	Intraoral coro- noidectomy	6	No
66         Moy chooldhary (a)         202         32         M         L         OC         0         39         Instraction instraction         >12         Net reported           66 $C_{11}$ 2005         25         F         L         CCE         30         40         Instraction         12         Not reported           67         Maximuch         2005         38         M         L         OC         12         41         Correction of reported         3         Not reported           68         Attan         2005         44         F         1.         OC         30         43         Instract core reported         Not reported         Not reported         Not           71         Discler call [2]         2006         44         F         1.         OC         6         1         Instract core reported         Not         Not           71         Discler call [2]         2009         35         M         R/L         OC         14         30         Instract core reported         Not           72         ortal [2]         2009         35         M         R/L         OC         12         S2         Enstracal core reported         Not     <	64	Emekli et al. [4]	2002	21	М	R	OC	8	40	Intra/extraoral coronoidec- tomy	Not reported	Not reported
66         Capade (1)         205         23         F         L         CCE         30         40         Initianal converting interaction on the properting interaction on thepropering interaction on the properting interaction on t	65	Roychoudhury et al. [6]	2002	32	М	L	OC	0	39	Extraoral coro- noidectomy	>12	Not reported
66*         Matsumoto         2005         38         M         L         OC         12         41         Intro of compy         3         Not           66         Alan         2006         24         M         8/L         CCE         15         20         Introord compy         Popered           67         Wat L [71]         2006         44         F         I.         OC         30         43         Introord compy         Popered           70         Dede et al. [72]         2007         20         M         8/L         OC         6         /         Introord comp         Popered           72         Dede et al. [72]         2009         30         F         R         OC         8         31         Introord comp         6         Not           73         Deman         2009         30         M         K/L         CCU         15         44         Introord comp         6         Not           74         Deman         2009         39         M         L         OC         12         28         Popered         Not         Not           75         deta. [77]         2011         21         M         R	66	Capote et al. [11]	2005	23	F	L	CCE	30	40	Intraoral coro- noidectomy	12	Not reported
66         eta L[6]         200         24         M $k/l$ CCE         15         30         Intracef core- mode core- bindectory         Not mode core- bindectory         Not mode core- bindectory         Not mode core- bindectory         Not mode core- bindectory         Not bindectory         Not mode core- bindectory         Not mode core- bindectory<	67	Matsumoto et al. [69]	2005	38	М	L	OC	12	41	Intra/extraoral coronoidec- tomy	3	Not reported
699         Villaturera         2006         44         F         L         OC         30         43         Intrace one- ment of an addetinary ment of a mode time provided and extrany         Not ment of a mode responded         Not ment of responded         Not ment of responded         Not ment of responded         Not ment of responded         Not ment of responded           70         Deck et al. [7:]         2009         39         F         R         OC         8         31         Intrace on a mode node comp         F         No           72         Thorn         2009         15         M         R/L         CCE         15         44         Intrace on a comp         14         No           73         derating         2009         43         F         R         OC         12         52         Belander comp         6         Not Not         Not           74         derating         210         16         M         R         OC         20         48         Intrace one- mode comp         reported	68	Akan et al. [70]	2006	24	М	R/L	CCE	15	30	Intraoral coro- noidectomy	Not reported	Not reported
70         Dade et al. [2]         207         20         M         R/L         OC         6         /         Indicators         Not         Not           71         detal. [7]         2009         99         F         R         OC         8         31         modelexany         9         reported           72         Thea         2009         15         M         R/L         CCE         15         44         modelexany         9         reported           73         Osman         2009         43         F         R         OC         14         30         modelexany         Not           74         Dridu         1(3)         2009         9         M         L         OC         12         52         Extraord or period         14         reported           75         vstildag         2010         16         M         R         OC         20         48         Intraoral core- neported         Not         noidectomy         14         reported         Not           76         atal [7]         2011         52         F         R         OC         20         40         Intraoral core- neported         reported         reported	69	Villanueva et al. [71]	2006	44	F	L	OC	30	43	Intraoral coro- noidectomy	Not reported	Not reported
71         2009         2009         39         F         R         OC         8         31         Infrareal cor- noidectomy         9         Reported period           72         et al. [7]         2009         43         F         R         OC         14         40         moldectomy         14         No           73         Osman et al. [73]         2009         43         F         R         OC         14         30         moldectomy         6         Reported           74         D'Ambroan et al. [73]         2010         16         M         R         OC         12         52         Bitaterial cor- noidectomy         Not reported         molectomy         Not reported         Not reported         Not reported         Not reported         Not reported         Not reported         Not noidectomy         12         No           75         detal. [71]         2011         25         F         R         OC         20         48         Infrareal cor- noidectomy         12         No           76         ct al. [71]         2011         25         F         R         OC         20         40         infrareal cor- noidectomy         24         No           78	70	Dede et al. [72]	2007	20	М	R/L	OC	6	/	Intraoral coro- noidectomy	Not reported	Not reported
72       atta [71]       2009       15       M       R/L       CCE       15       44       intraced core-indications       14       No         73       Costal [73]       2009       43       F       R       OC       14       30       indications       6       propried         74       D'Ambrato       2009       43       K       OC       14       30       indications       6       propried         75       Textand core-insported       reported       reported       nodectomy       14       Not         76       Taking       2011       21       M       R       OC       7       /       intraced core-insported       nodectomy       Not         76       Taking       2011       25       F       R       OC       7       /       intraced core-insported       nodectomy       6       Not         79       Acasta-feria       2011       55       F       R       OC       51       61       intraced core-insported       nodectomy       24       No         80       Sreeramaneri       2011       45       F       L       OC       51       61       intraced core-insported       nodectomy	71	Zhong et al. [73]	2009	39	F	R	OC	8	31	Intraoral coro- noidectomy	9	Not reported
73         Obstram         2009         43         F         R         OC         14         30         Introductory- nodectory         6         Not reported           74         td Ambrasio         2009         39         M         L         OC         /         /         Indicational corp- nodectory         Not         Reported Not           75         td [15]         2010         16         M         R         OC         12         52         Participation         Not         Not           76         tdafuti         2011         21         M         R         OC         20         48         Intronal corp- nodectory         Not         Not           77         Aria text [77]         2011         25         F         R         OC         8         41         Intranal corp- nodectory         6         Reported           78         Coll-Anglafa         2011         55         F         R         OC         5         40         Particetory         3         Not           80         Srccramment etal.[7]         2011         45         F         L         OC         5         40         Intranal corp- nodectory         3         Not	72	Thota et al. [74]	2009	15	М	R/L	CCE	15	44	Intraoral coro- noidectomy	14	No
74         D'Ambrosi         2009         39         M         L         OC         /         /         Intraorations         Note         Note noidections         Note noidections         Note noidections         Note noidections         Note noidections         Note noidections         Note noidections         Note noidections         Note noidections           76         dt al. [7]         2011         21         M         R         OC         20         48         Intraorations         No           77         Ajila et al. [7]         2011         25         F         R         OC         7         /         Intraorations         No           78         Costal-Arglada et al. [2]         2011         55         F         R         OC         20         40         Extraorations         No           80         Streamanenti et al. [2]         2011         45         F         L         OC         51         61         Intraorations         No           81         Aokie et al. [80]         2012         18         M         R         OC         2         40         Extraorations         Not reported           82         Pacheo Ruiz et al. [81]         2012         18         M	73	Osman et al. [75]	2009	43	F	R	OC	14	30	Intraoral coro- noidectomy	6	Not reported
75       Vesidage (al., [7])       2010       16       M       R       OC       12       52       Extraord core- noidectomy       14       reported reported         76       et al. [7]       2011       21       M       R       OC       20       48       Intraoral core- noidectomy       Not       Not         77       Ajila et al. [7]       2011       25       F       R       OC       7       /       Intraoral core- noidectomy       12       No         78       Cell Angleda et al. [7]       2011       55       F       R       OC       20       40       Extraoral core- noidectomy       6       meported         79       Acosta-Ferra et al. [7]       2011       55       F       R       OC       51       61       Intraoral core- noidectomy       15       No         80       Sevenameneni et al. [7]       2012       18       M       R       OC       21       40       Extraoral core- noidectomy       15       No         81       Aokiet et al. [81]       2012       13       M       R/L       CH       10       48       Intraoral core- noidectomy       Not       noidectomy       noridec- nomidec-       Not         8	74	D'Ambrosio et al. [13]	2009	39	М	L	OC	/	/	Intraoral coro- noidectomy	Not reported	Not reported
76         Takafuj et al. $[77]$ 2011         21         M         R         OC         20         48         Intronal corb- moted ecropy- reported         Not proported         Not proported         Not proported         Not proported         Not proported         Not proported         Not proported           77         Ajila et al. $[73]$ 2011         52         F         R         OC         8         41         notidectomy         6         Root           78         Coll-Anglada et al. $[73]         2011         55         F         R         OC         20         40         Intraoral corb-noidectomy         24         No           80         Srearmaneriet al. [71]         2011         45         F         L         OC         5         40         Intraoral corb-noidectomy         24         No           81         Aoki et al. [80]         2012         18         M         R         OC         2         40         Coronoidec-noidectomy         3         reported           83         Taxassol         2012         13         M         R/L         CCE         10         48         Intraoral corb-noidectomy         Not         reported         reported         reported         repo$	75	Yesildag et al. [76]	2010	16	М	R	OC	12	52	Extraoral coro- noidectomy	14	Not reported
77       Ajila et al. [78]       2011       28       M       L       OC       7       /       Introval core- introval core- table core table core- table core- core	76	Takafuji et al. [77]	2011	21	М	R	OC	20	48	Intraoral coro- noidectomy	Not reported	Not reported
78       Coll-Anglada et al. [12]       2011       52       F       R       OC       8       41       Intraoral corp- noidectomy       6       Not reported         79       Acosta-Feria et al. [7]       2011       55       F       R       OC       20       40       Extraoral corp- noidectomy       24       No         80       Sreeramaneri et al. [7]       2011       45       F       L       OC       5       40       corronoide- tomy       3       Not reported         81       Aokie tal. [80]       2012       18       M       R       OC       2       40       model corronoide- tomy       3       Not reported         83       Tavassol       2012       13       M       R/L       CH       10       48       Initiaoral corp- noidectomy       Not reported         84       Choi et al. [83]       2013       13       M       R/L       CCE       10       40       Initiaoral corp- noidectomy       Not reported       reported         85       Choi et al. [83]       2013       13       M       R/L       CCE       20       45       Initiaoral corp- noidectomy       Not reported       Not reported         86       Lei Minor <t< td=""><td>77</td><td>Ajila et al. [78]</td><td>2011</td><td>28</td><td>М</td><td>L</td><td>OC</td><td>7</td><td>/</td><td>Intraoral coro- noidectomy</td><td>12</td><td>No</td></t<>	77	Ajila et al. [78]	2011	28	М	L	OC	7	/	Intraoral coro- noidectomy	12	No
79         Acosta-Ferria et al. [79]         2011         55         F         R         OC         20         40         Extraoral croo- noidectomy         24         No           80         Sreeramaneni et al. [7]         2011         45         F         L         OC         5         40         Intra-/extraoral coronoidec- noidectomy         3         Reported reported           81         Aokiet al. [80]         2012         18         M         R         OC         51         61         Intra-/extraoral form         Not coronoidec- tomy         3         Reported reported           82         Pacheo Ruiz et al. [82]         2012         13         M         R/L         CH         10         48         Intra-/extraoral form         Not coronoidec- tomy         7         Not         Not           83         Tavassol         2013         13         M         R/L         CCE         15         40         Intraoral coro- noidectomy         Not         reported           84         Choi et al. [83]         2013         13         M         R/L         CCE         20         45         Intraoral coro- noidectomy         Not         Not           86         Stringer et al. [81]         2013         15	78	Coll-Anglada et al. [12]	2011	52	F	R	OC	8	41	Intraoral coro- noidectomy	6	Not reported
80         Steeramanni et al. [7]         2011         45         F         L         OC         5         40         Intra-acco- connoidec- tomy         Not economide- tomy         Not connoidec- tomy         Not intra-acco- noidectomy         Not intra-acco- noidectomy         Not intra-acco- reported         Not intra-acco- noidectomy         Not intra-acco- reported         Not reported           81         Aoki et al. [80]         2012         18         M         R         OC         2         40         Intra-acco- noidectomy         Not reported           82         Pacheo Ruiz et al. [82]         2012         13         M         R/L         CH         10         48         Intra-acco- noidectomy         Not reported         Not reported           83         Tavassol         2013         13         M         R/L         CCE         15         40         Intra-acco- noidectomy         Not reported         Not reported           84         Choi et al. [83]         2013         13         M         R/L         CCE         20         45         Intra-acco- noidectomy         Not reported         Not           86         Stringer et al. [81]         2013         15         M         L         OC         20         41         Indiaccomy	79	Acosta-Feria et al. [79]	2011	55	F	R	OC	20	40	Extraoral coro- noidectomy	24	No
81         Aoki et al. [80]         2012         18         M         R         OC         51         61         Intraoral coro-noidectomy intra/extraoral coronoidec.         15         No           82         Pacheco Ruiz et al. [81]         2012         28         M         R/L         OC         2         40         coronoidec- tomy         3         Not reported           83         Tavassol et al. [82]         2012         13         M         R/L         CH         10         48         noidectomy noidectomy         Not reported         reported         report	80	Sreeramaneni et al. [7]	2011	45	F	L	OC	5	40	Intra/extraoral coronoidec- tomy	3	Not reported
82Pacheco Ruiz et al. [81]201228M $R/L$ OC240Intra / extraoral conoidec- tomyNot reported $83$ Tavassol et al. [82]201213M $R/L$ CH1048Intraoral coro- noidectomyNot reportedNot reported $84$ Choi et al. [83]201313M $R/L$ CCE1540Intraoral coro- noidectomyNot reported $85$ Choi et al. [83]201313M $R/L$ CCE2045Intraoral coro- noidectomyNot reported $86$ Stringer et al. [84]201327MLOC1040Intraoral coro- noidectomyNot reported $87$ Hosin Hosin15MLOC2241Intraoral coro- noidectomyNot reported $88$ Losa-Munoz201442MROC2041Intraoral coro- noidectomyNot reported $89$ et al. [87] et al. [86]201442MR/35/None reportedNot reported $90$ Zarembinski et al. [89]201420FLOC1540Intraoral coro- noidectomyNot reportedNot reported $91$ Fan et al. [89]201420MLOC22/Not modectomy reportedNot reported $94$ Schine et al. [94]201420 <td>81</td> <td>Aoki et al. [80]</td> <td>2012</td> <td>18</td> <td>М</td> <td>R</td> <td>OC</td> <td>51</td> <td>61</td> <td>Intraoral coro- noidectomy</td> <td>15</td> <td>No</td>	81	Aoki et al. [80]	2012	18	М	R	OC	51	61	Intraoral coro- noidectomy	15	No
83         Tavassol et al. [82]         2012         13         M         R/L         CH         10         48         Intraoral coro- noidectomy         Not         Not         Not           84         Choi et al. [83]         2013         13         M         R/L         CCE         15         40         Intraoral coro- noidectomy         4         reported           85         Choi et al. [83]         2013         13         M         R/L         CCE         20         45         Intraoral coro- noidectomy         Not         Not           86         Stringer et al. [85]         2013         27         M         L         OC         10         40         Intraoral coro- noidectomy         Not         Not         Not           87         Hosein et al. [85]         2013         15         M         L         OC         22         41         Intraoral coro- noidectomy         Not         not         Not         reported           88         Lesa-Munoz et al. [86]         2014         42         M         R         OC         15         40         Intraoral coro- noidectomy         Not         Not           90         Zarembinski et al. [87]         2014         50         M	82	Pacheco Ruiz et al. [81]	2012	28	М	R/L	OC	2	40	coronoidec- tomy	3	Not reported
84       Choi et al. [83]       2013       13       M       R/L       CCE       15       40       Intraoral coro-noidectomy       4       Not proported proported         85       Choi et al. [83]       2013       13       M       R/L       CCE       20       45       Intraoral coro-noidectomy       Not       Yes (36)         86       Stringer et al. [84]       2013       15       M       L       OC       10       40       Intraoral coro-noidectomy       Not       Not         87       Hosein       2013       15       M       L       OC       22       41       Intraoral coro-noidectomy       Not       Not         88       Losa-Munoz et al. [86]       2014       42       M       R       OC       20       41       Intraoral coro-noidectomy       Not       reported       repor	83	Tavassol et al. [82]	2012	13	М	R/L	СН	10	48	Intraoral coro- noidectomy	Not reported	Not reported
85       Choi et al. [83]       2013       13       M       R/L       CCE       20       45       Intraoral coro-noidectomy reported months)         86       Stringer et al. [84]       2013       27       M       L       OC       10       40       Intraoral coro-noidectomy reported months)         87       Hosein et al. [84]       2013       15       M       L       OC       22       41       Intraoral coro-noidectomy reported months)         88       Losa-Munoz et al. [86]       2014       42       M       R       OC       20       41       Intraoral coro-noidectomy reported months)         89       Dandrival cet al. [86]       2014       20       F       L       OC       15       40       Intraoral coro-noidectomy reported metod motot Not exported       Not exported metod metod motot Not exported motot Not exported metod metod motot Not exported metod motot Not exported metod metod motot Not exported metod metod motot Not exported metod metod Not exported metod metod Not exported metod motot Not exported metod metod Not exported metod motot Not exported metod metod motot Not exported metod metod Not exported metod Not exported metod metod metod Not exported exported exported tomy         90       Zarembinski et al. [89]       2014       20       M       L       OC	84	Choi et al. [83]	2013	13	М	R/L	CCE	15	40	Intraoral coro- noidectomy	4	Not reported
86Stringer et al. [84]201327MLOC1040Intraoral coro- noidectomyNotNot87Hosein et al. [85]201315MLOC2241Intraoral coro- noidectomyNotNot88Losa-Munoz et al. [86]201442MROC2041Intraoral coro- noidectomyNotNot89Dandriyal et al. [87]201420FLOC1540Intraoral coro- noidectomyNotNot90Zarembinski et al. [89]201450MR/35/NoneNot reportedNot reported91Fan et al. [89]201420MLOC22/Not reportedNot reported92Rahim et al. [80]201419MLOC22/Not reportedNot reported93Reddy et al. [94]201421FROC1242Extraoral coro- noidectomyNot reportedNot reported94Sinha et al. [93]201421FROC1242Extraoral coro- noidectomyNot reportedNot reported95Sawada et al. [93]201458MLOC1020Intraoral coro- noidectomyNot reportedNot reported96Shin et al. [94]201639FL <td>85</td> <td>Choi et al. [83]</td> <td>2013</td> <td>13</td> <td>М</td> <td>R/L</td> <td>CCE</td> <td>20</td> <td>45</td> <td>Intraoral coro- noidectomy</td> <td>Not reported</td> <td>Yes (36 months)</td>	85	Choi et al. [83]	2013	13	М	R/L	CCE	20	45	Intraoral coro- noidectomy	Not reported	Yes (36 months)
87       Hosein et al. [85] et al. [86]       2013       15       M       L       OC       22       41       Intraoral coro- noidectomy       18       Reported reported         88       Losa-Munoz et al. [86]       2014       42       M       R       OC       20       41       Intraoral coro- noidectomy       Not       Reported reported         89       Dandriyal et al. [87]       2014       20       F       L       OC       15       40       Intraoral coro- noidectomy       Not       Not         90       Zarembinski et al. [89]       2014       50       M       R       /       35       /       None       Not       Not       reported         91       Fan et al. [89]       2014       20       M       L       OC       22       45       Intraoral coro- noidectomy       No       No         92       Rahim et al. [90]       2014       19       M       L       OC       22       /       Not reported reported       Poot reported       Not         93       Reddy et al. [91]       2014       21       F       R       OC       12       42       Extraoral coro- noidectomy       Not       Not noidectomy       Not       Not       <	86	Stringer et al. [84]	2013	27	М	L	OC	10	40	Intraoral coro- noidectomy	Not reported	Not reported
88Losa-Munoz et al. [86]201442MROC2041Intraoral coro- noidectomyNotNot89Dandriyal et al. [87]201420FLOC1540Intraoral coro- noidectomyNotNot90Zarembinski et al. [89]201450MR/35/NoneNot reportedNot reported91Fan et al. [89] et al. [89]201420MLOC2545Intra/extraoral coronidec- tomy20No92Rahim et al. [90] et al. [91]201419MLOC22/Not reported reportedNot reported93Reddy et al. [92] et al. [92]201421FROC1242Extraoral coro- noidectomy reportedNot reported94Sinha et al. [92]201458MLOC1020Intra/extraoral reportedNot reported95Sawada et al. [93]201639FL/2141coronoidec- reported36No96Shin et al. [94] et al. [95]201618MROC1140Extraoral coro- rodidectomy36No97Mohanty et al. [95]201659FROC949Extraoral coro- rodidectomy6Not reported98Robiory et al. [95]20	87	Hosein et al. [85]	2013	15	М	L	OC	22	41	Intraoral coro- noidectomy	18	Not reported
89Dandriyal et al. [87]201420FLOC1540Intraoral coro- noidectomy54No90Zarembinski et al. [89]201450MR/35/NoneNot reportedNot reported91Fan et al. [89]201420MLOC2545coronoidec- tomy20No92Rahim et al. [90]201419MLOC22/Not reportedNot reportedNot reported93Reddy et al. [91]201421FROC1242Extraoral coro- noidectomyNot reportedNot reported94Sinha et al. [92]201458MLOC1020Intraoral coro- noidectomyNot reported95Sawada et al. [93]201514MLOC1020Intraoral coro- noidectomyNot reported96Shin et al. [94]201639FL/2141coronoidec- roported36No97Mohanty et al. [96]201618MROC1140Extraoral coro- noidectomy36Not reported98Robiony et al. [96]201659FROC949Extraoral coro- noidectomy6Not reported99Imen et al. [97]201629MLOC5<	88	Losa-Munoz et al. [86]	2014	42	М	R	OC	20	41	Intraoral coro- noidectomy	Not reported	Not reported
90Zarembinski et al. [88]201450MR/35/NoneNot reportedNot reportedNot reported91Fan et al. [89]201420MLOC2545Intra/extraoral coronoidec- tomy20No92Rahim et al. [90]201419MLOC22/Not reported reportedNot reportedNot reported93Reddy et al. [91]201421FROC1242Extraoral coro- noidectomyNot reportedNot reported94Sinha et al. [92]201458MLOC///Intraoral coro- noidectomyNot reportedNot reported95Sawada et al. [93]201514MLOC1020Intraoral coro- noidectomyNot reportedNot reported96Shin et al. [94]201639FL/2141coronoidec- tomy36No97Mohanty et al. [96]201618MROC1140Extraoral coro- noidectomy36Not reported98Robiony et al. [96]201659FROC949Extraoral coro- noidectomy6Not reported99Imme et al. [97]201629MLOC555Extraoral coro- noidectomyNot </td <td>89</td> <td>Dandriyal et al. [87]</td> <td>2014</td> <td>20</td> <td>F</td> <td>L</td> <td>OC</td> <td>15</td> <td>40</td> <td>Intraoral coro- noidectomy</td> <td>54</td> <td>No</td>	89	Dandriyal et al. [87]	2014	20	F	L	OC	15	40	Intraoral coro- noidectomy	54	No
91Fan et al. [89]201420MLOC2545coronoidec- tomy20No92Rahim et al. [90]201419MLOC22/Not reported reportedNot reportedNot reported93Reddy et al. [91]201421FROC1242Extraoral coro- noidectomyNot reportedNot reported94Sinha et al. [92]201458MLOC//Intraoral coro- noidectomyNot reportedNot reported95Sawada et al. [93]201514MLOC1020Intraoral coro- noidectomyNot reportedNo96Shin et al. [94]201639FL/2141coronoidec- tomy36No97Mohanty et al. [95]201618MROC1140Extraoral coro- noidectomy36No98Robiony et al. [96]201659FROC949Extraoral coro- noidectomy6Not reported99Imen et al. [97]201629MLOC555Extraoral coro- noidectomyNot	90	Zarembinski et al. [88]	2014	50	М	R	/	35	/	None	Not reported	Not reported
92Rahim et al. [90]201419MLOC22/Not reported reportedNot reportedNot reported93Reddy et al. [91]201421FROC1242Extraoral coro- noidectomyNotNot reported94Sinha et al. [92]201458MLOC//Intraoral coro- noidectomyNotNot95Sawada et al. [93]201514MLOC1020Intraoral coro- noidectomyNot reportedNot96Shin et al. [94]201639FL/21141Extraoral coro- noidectomyNot reportedNo97Mohanty et al. [95]201618MROC1140Extraoral coro- noidectomy36No98Robiony et al. [96]201659FROC949Extraoral coro- noidectomy6Not reported99Imen et al. [97]201629MLOC555Extraoral coro- noidectomyNot	91	Fan et al. [89]	2014	20	М	L	OC	25	45	Intra/extraoral coronoidec- tomy	20	No
93Reddy et al. [91]201421FROC1242Extraoral coro- noidectomyNotNot94Sinha et al. [92]201458MLOC//Intraoral coro- noidectomyNotNot95Sawada et al. [93]201514MLOC1020Intraoral coro- noidectomyNot96Shin et al. [94]201639FL/2141coronoidec- noidectomy36No97Mohanty et al. [95]201618MROC1140Extraoral coro- noidectomy36No98Robiony et al. [96]201659FROC949Extraoral coro- noidectomy6Not reported99Imen et al. [97]201629MLOC555Extraoral coro- noidectomyNot	92	Rahim et al. [90]	2014	19	М	L	OC	22	/	Not reported	Not reported	Not reported
94       Sinha et al. [92]       2014       58       M       L       OC       /       /       Intraoral coro- noidectomy       Not       Not         95       Sawada et al. [93]       2015       14       M       L       OC       10       20       Intraoral coro- noidectomy       Not       No         96       Shin et al. [94]       2016       39       F       L       /       21       41       coronoidec- tomy       36       No         97       Mohanty et al. [95]       2016       18       M       R       OC       11       40       Extraoral coro- noidectomy       36       No         98       Robiony et al. [96]       2016       59       F       R       OC       9       49       Extraoral coro- noidectomy       6       Not reported         99       Imen et al. [97]       2016       29       M       L       OC       5       55       Extraoral coro- noidectomy       Not	93	Reddy et al. [91]	2014	21	F	R	OC	12	42	Extraoral coro- noidectomy	Not	Not
95       Sawada et al. [93]       2015       14       M       L       OC       10       20       Intractal cory- noidectomy reported       No         96       Shin et al. [94]       2016       39       F       L       /       21       41       coronoidec- tomy       36       No         97       Mohanty et al. [95]       2016       18       M       R       OC       11       40       Extraoral coro- noidectomy       36       No         98       Robiony et al. [96]       2016       59       F       R       OC       9       49       Extraoral coro- noidectomy       6       Not reported         99       Imen et al. [97]       2016       29       M       L       OC       5       55       Extraoral coro- noidectomy       Not	94	Sinha et al [92]	2014	58	М	L	OC	/	/	Intraoral coro-	Not	Not
96       Shin et al. [94]       2016       39       F       L       /       21       41       Intra/extraoral         97       Mohanty et al. [95]       2016       18       M       R       OC       11       40       Extraoral coro- noidectomy       36       No         98       Robiony et al. [96]       2016       59       F       R       OC       9       49       Extraoral coro- noidectomy       36       No         99       Imen et al. [97]       2016       29       M       L       OC       5       55       Extraoral coro- noidectomy       Not	95	Sawada et al [93]	2015	14	М	L	OC	10	20	Intraoral coro-	Not	No
97Mohanty et al. [95]201618MROC1140Extraoral coro- noidectomy36No98Robiony et al. [96]201659FROC949Extraoral coro- noidectomy6Not reported99Imen et al. [97]201629MLOC555Extraoral coro- NotNot	96	Shin et al. [94]	2016	39	F	L	/	21	41	Intra/extraoral coronoidec-	36	No
98       Robiony et al. [96]       2016       59       F       R       OC       9       49       Extraoral coro- noidectomy       6       Not reported         99       Imen et al. [97]       2016       29       M       L       OC       5       55       Extraoral coro- noidectomy       Not	97	Mohanty	2016	18	М	R	OC	11	40	Extraoral coro-	36	No
er al. [96] noidectomy reported 99 Imen et al. [97] 2016 29 M L OC 5 55 Extraoral coro- Not Not	98	Robiony	2016	59	F	R	OC	9	49	Extraoral coro-	6	Not
the second se	99	et al. [96] Imen et al. [97]	2016	29	М	L	OC	5	55	Extraoral coro-	Not	reported Not

Case No.	Author	Year	Age	Gender	Side	Туре	MMO before Treatment (mm)	MMO after Treatment (mm)	Treatment	Treatment Follow-Up (Months)	Treatment Recurrence
100	Gangoli et al. [98]	2017	15	F	L	OC	9	34	Intraoral coro- noidectomy	Not reported	Not reported
101	Choontharu et al. [99]	2018	16	F	L	OC	43	/	Intraoral coro- noidectomy	6	No
102	Roscher et al. [100]	2018	18	F	L	OC	20	41	Intraoral coro- noidectomy	12	Not reported
103	Villegas Cisneros et al. [101]	2018	10	М	R/L	СН	19	35	Intra/extraoral coronoidec- tomy	Not reported	Not reported
104	Lan et al. [102]	2019	34	F	R	OC	5	36	Intraoral coro- noidectomy	21	No
105	Kono et al. [103]	2019	51	М	L	OC	20	42	Intraoral coro- noidectomy	30	No
106	Samandari et al. [104]	2019	28	М	L	CH	25	/	Intraoral coro- noidectomy	Not reported	Not reported
107	Gomez et al. [105]	2020	54	F	L	OC	12	22	Intraoral coro- noidectomy	12	Not reported
108	Jimenez Alvarez et al. [106]	2020	15	М	R	OC	10	40	Intraoral coro- noidectomy	72	Not reported
109	Avelar et al. [107]	2020	6	М	R/L	СН	10	28	Extraoral coro- noidectomy	Not reported	No
110	Okazawa et al. [108]	2020	27	М	L	OC	4	60	Intra/extraoral coronoidec- tomy	12	No
111	Khadembaschi et al. [109]	2020	14	М	R/L	CCE	8	45	Extraoral coro- noidectomy	18	Not reported
112	Alam et al. [110]	2021	32	М	R	OC	8	38	Extraoral coro- noidectomy	Not reported	Not reported
113	Leal et al. [111]	2021	11	F	L	CCE	20	25	Intraoral coro- noidectomy	Not reported	Not reported
114	Raccampo et al.	2022	19	F	R	CCE	25	40	Intraoral coro- noidectomy	12	No
115	Raccampo et al.	2022	18	М	R	OC	20	44	Intraoral coro- noidectomy	12	No
116	Raccampo et al.	2022	23	М	R/L	СН	20	37	Intraoral coro- noidectomy	14	No

Table 1. Cont.

#### 3. Case Presentation

3.1. Case 1

A 19-year-old woman was referred to us, presenting with a history of progressive limitation in mouth opening over the previous 18 months. She complained of a dull pain just in front of her right ear, exacerbated by palpation and mandibular movements. She had no medical history and did not report any previous local trauma. She was initially diagnosed with temporomandibular disease (TMD) dysfunction and treated conservatively with an occlusal bite with no symptomatologic relief. At physical examination, the maximum mouth opening (MMO) was reduced to 25 mm, but mandibular movements were preserved. No articular noises during temporomandibular joint (TMJ) bilateral palpation were perceived, but she complained of pain in the masticatory muscles bilaterally. No malocclusion, facial asymmetry or swelling were identified. The orthopantomogram (OPG) (Figure 1) and the magnetic resonance imaging (MRI) were negative. Given the absence of clinical improvement but a rather slow and progressive reduction in MMO, a computed tomography (CT) was performed, revealing an enlargement and an abnormally shaped right CPM, establishing a close relationship with the zygomatic arch (Figure 2). Furthermore, single-photon emission computed tomography (SPECT) was prescribed, and the late images of bone uptake showed a focal increase in the correspondence of the so-called pseudojoint between the jaw and the right zygomatic bone compared to the left side (Figure 3). According to this, a provisional diagnosis of JD was made. The patient underwent a right coronoidectomy through an intraoral approach under general anesthesia with awake, blind, nasal intubation. The right, mushroom-shaped coronoid process was identified and resected, and a sort of fibrotic capsule between the inner aspect of the zygoma and the CPM was highlighted, establishing the pseudojoint, with this confirming the diagnosis of JD (Figure 4). Histopathologically, the specimen showed a growth pattern of hyperplastic bone tissue covered by cartilaginous layers and an external coating of dense fibrous tissue. There were no intra-operative or immediate postoperative complications. Postoperative CT was obtained for baseline follow-up. The MMO increased to about 30 mm in the immediate postoperative period, and this improved to 40 mm after a month of aggressive physiotherapy. She underwent clinical and radiological follow-up, and after 12 months, the MMO was stable, and no recurrence was observed.



Figure 1. Patient's OPG showing no evident anomalies.



**Figure 2.** Cinematic 3D reconstruction (Mimics, Materialise NV, Leuven, Belgium) showing the pseudojoint established between the altered right CPM and the inner aspect of zygomatic bone.

## 3.2. Case 2

An 18-year-old male patient with a 24-month history of worsening reductions in mouth opening was referred to our attention for an evaluation of suspected TMD. No previous history of trauma was reported by the patient, and the medical history did not report any relevant elements. A physical examination showed an MMO of 20 mm with a deviation to the right during mouth opening (Figure 5). The patient did not complain of pain at the palpation of TMJ bilaterally. The masticatory muscles were painful bilaterally and especially on the right side. No TMJ clicking or crepitus were detectable. The endfeel was rigid and painful on the right side. At the OPG, the right glenoid fossa and condyle

were not totally distinguishable, so an MRI and CT scan were requested to examine the case in depth. The MRI was negative for TMD, but in the CT scan, an enlarged right CPM with a mushroom-shaped end was highlighted, establishing a close relationship with the inner aspect of the right zygoma, which seemed impressed by the CPM forming a pseudojoint (Figure 6). It is interesting to notice how also the contralateral CPM was slightly elongated (Figure 7). The suspected diagnosis was of a right JD. The patient was subsequently submitted to an intraoral right coronoidectomy under general anesthesia. An incision along the vestibular aspect of the ascending mandibular ramus was performed, then a sub-periosteal dissection exposing the right maxillary zygomatic arch and the anterior rim of the mandibular ascending ramus was executed. At this point, the CPM was detected as showing a protuberance covered with fibrous tissue mimicking a capsule. Immediately after the completion of the coronoidectomy, the MMO increased to 40 mm (Figure 8). The histopathology of the CPM showed sections of compact, trabecular bone tissue covered by a layer of hyaline cartilage and fibrous cartilage overlaid by dense fibrous connective tissue compatible with the diagnosis of osteochondroma (Figure 9). At 1 week, postoperative jaw-opening exercises and articular physiotherapy with TheraBite jaw motion rehabilitation system <sup>TM</sup> (Atos medical, Padua, Italy) were prescribed. At 1 month follow-up, the patient had an MMO of 20 mm, 18 mm at 2 months and 10 mm at 3 months. The cause of this negative trend was initially identified as the fibrotic retraction of the intraoral scar of the surgical access. At 5 months follow-up, the MMO was 32 mm and increased to 38 mm 1 month later. At 12 months follow-up, the MMO was 44 mm. A control CT scan was conducted and showed no recurrence and no significant changes on the contralateral side.



**Figure 3.** SPET-CT showing bone reworking in correspondence to the joint between the jaw and the right zygomatic bone compared to the contralateral homologous site.

#### 3.3. Case 3

A 23-year-old male patient presented complaining of a progressive limitation in mouth opening. He did not report any pain. The reduction in the MMO had been worsening over the prior 5 years. There was no history of trauma. Clinical examination revealed an MMO of 20 mm with no deviation (Figure 10). There was no pain during jaw opening. He did not experience pain on palpation of the preauricular area bilaterally. The endfeel was rigid but not painful, and crepitus was perceived at the right TMJ. OPG and TMJ projections showed a lack of translation of the mandibular condyles in the opening and an alteration in the morphology of the CPM bilaterally. MRI was negative for intra-articular causes of ankylosis. CT scans showed a bilateral enlargement of the CPMs with a mushroom-shaped bony outgrowth forming a pseudojoint with the inner aspect of the zygomatic bones, which appeared modeled to the CPMs. This is particularly evident in the 3-dimensional dynamic reconstruction for the virtual surgical planning (VSP) we performed before the surgery (Figure 11). The diagnosis was bilateral CPM hyperplasia, determining a bilateral JD. The

patient was scheduled for a bilateral endoscopically assisted intraoral coronoidectomy under general anesthesia with awake fiberoptic intubation. Fibrotic shoots were detected at the pseudojoint between the CPMs and the zygoma. The entire CPM was then removed. At the end of the surgery, the interincisal distance was 50 mm with a mechanical forced opening (Figure 12). The resected specimens showed cortical bone tissue with a regular structure and orientation of the bone lamellae, determining a bilateral JD due to a true CPM hyperplasia. Articular physiotherapy was started one week after surgery using TheraBite jaw motion rehabilitation system<sup>TM</sup> (Atos medical, Padua, Italy). At 1 month after the surgery, the MMO was 25 mm and increasable after forcing, 2 months later it was 30 mm and 35 mm at 3 months. At 14 months follow-up, the patient MMO was stable at 37 mm, and the radiographic evaluation was negative for recurrence.



Figure 4. Right mushroom-shaped coronoid process excised.



**Figure 5.** Patient showing an MMO of 20 mm and a slight deviation to the right at first physical examination.



**Figure 6.** Cinematic 3D reconstruction (Mimics, Materialise NV., Leuven, Belgium) showing the established right JD.



Figure 7. 3D CT reconstructions showing an enlarged left CPM.



**Figure 8.** Surgical sequence of the intraoral coronoidectomy. The patient shows an immediate improvement in the MMO up to 40 mm.







(**b**)

**Figure 9.** Hematoxylin–eosin staining of the lesion. Sections of compact, trabecular bone tissue covered by a layer of hyaline cartilage and fibrous cartilage overlaid by dense fibrous connective tissue (**a**,**b**).



Figure 10. MMO of 20 mm at first clinical examination.



**Figure 11.** 3-dimensional dynamic reconstruction showing the bilateral pseudojoint determining a bilateral JD.



Figure 12. MMO of 50 mm right after bilateral coronoidectomy.

## 4. Results and Discussion

JD is a rare condition in which an elongated CPM interferes with the inner surface of the zygomatic arch, establishing a pseudojoint. In our review, which represents, possibly, the widest review in the literature, we report a mean age of 28.7 years old (5–73), with a difference in the age of incidence of almost 10 years between the two genders (25.7 yearsold for males and 35.4 years old for females). In 12 cases, it was not possible to trace back the age of diagnosis of JD. We collected the data of 71 males (61.2%) and 34 females (29.3%). Unfortunately, 11 patients' genders were not reported. These data are in agreement with what was previously reported in the literature, as affecting mainly men between the second and fourth decade [6]. An overall prevalence of 0.5% has also been reported, but this data could be underestimated because the onset of symptoms, such as limited mouth opening, represents just the endpoint of a longer-term development [55]. The left side is reported to be the most affected [7], which is confirmed by our analysis, in which we identified 38 cases of right JD (32.8%), 50 of the left side (43.1%) and 21 bilateral cases (18.1%). For seven cases, we were unable to retrieve information about the side of the defect. As mentioned before, JD usually remains relatively asymptomatic in its first stages, with most patients reporting only a sensation of tension during chewing. Then, it usually evolves with the limitation of mandibular movements and worsening reduction in MMO. In the reviewed cases, the mean MMO before surgical treatment was 13.9 mm (2–51). Sometimes, a distortion in the zygomatic arch projection on the affected side or a palpable moving hard mass perceptible during mandibular movements are present, particularly if JD is caused by an osteochondroma [94]. In unilateral cases, deviation to the affected side during mouth opening can be seen [75]. Pain or paresthesia are not often reported. This insidious clinical onset may be confused with several disorders such as TMD mainly, trauma arthrosis and various other causes of intra- and extra-articular ankylosis. Therefore, JD is also often mistreated. The etiology of the disease has already been widely debated. Some authors theorize a genetic or endocrine cause, and others suggest a role for temporalis muscle hyperactivity, trauma, TMJ disc displacement or a family predisposition, but it is mostly considered idiopathic [3,52,112]. Although the cause remains unknown, some authors reported periosteal hyperactivity as a trigger for ectopic metaplastic cartilage formation [5]. As stated before, according to the different amounts and patterns of bone and cartilage tissues present, the abnormally enlarged CPM can be histologically diagnosed as OC, CCE, CH, benign tumors (such as osteoma or chondroma) or other developmental anomalies [4,7,10,14,70]. We found 17 cases of CCE (14.7%), 10 cases of CH (8.6%) and 76 cases of OC (65.5%), while in 13 cases (11.2%), it was not possible to establish a definite histological diagnosis. This marked prevalence of OC as a cause of JD may partly explain the confusion over its definition and its almost exclusive bi-directional association with pathology reported by various authors. It is also interesting to note how this pathology shows different clinical and radiological patterns, leading to a wide spectrum of presentations. Considering, for example, the cases we have reported, it is possible to see how they present a pattern of increasing severity from the first to the third patient. The first patient presented an early pathologic radiological pattern, with the pseudojoint not yet fully formed with an MMO of 25 mm. It should be remembered that ankylosis, in this case, extra-articular, is established when the MMO is less than 15 mm, and this is considered an incomplete ankylosis, when the MMO is less than 5 mm, the ankylosis is complete [113,114]. In contrast, the second patient showed a well-established ipsi-pseudo-articular framework and an MMO of 20 mm. In this patient, it is also interesting to notice how the contralateral CPM was also slightly longer than normal, although not yet pathological. The third patient, on the other hand, showed a well-established bilateral pattern with perfectly developed pseudojoints and a rapidly worsening 20 mm MMO. Our experience and analysis of the literature prompted us to wonder whether it was possible to classify, according to clinical and radiological features, the vast pathological spectrum that JD expresses. We, therefore, designed a classification proposal, as shown in Table 2, in order to speculate on the possibility of assigning an index of severity and stage to the

**Classification Proposal** An ipsilateral formation of a pseudojoint between an abnormal <u>Grade 1 A</u> CPM and the inner aspect of the zygomatic bone determining an extra-articular ankylosis with a MMO  $\geq$  20 mm. Grade 1 A bilateral formation of a pseudojoint between an abnormal Grade 1 B CPM and the inner aspect of the zygomatic bone determining an extra-articular ankylosis with a MMO  $\geq$  20 mm. An ipsilateral formation of a pseudojoint between an abnormal <u>Grade 2 A</u> CPM and the inner aspect of the zygomatic bone determining an extra-articular ankylosis with a  $20 \le MMO \ge 10$  mm. Grade 2 A bilateral formation of a pseudojoint between an abnormal Grade 2 B CPM and the inner aspect of the zygomatic bone determining an extra-articular ankylosis with a  $20 \le MMO \ge 10$  mm. An ipsilateral formation of a pseudojoint between an abnormal Grade 3 A CPM and the inner aspect of the zygomatic bone determining an extra-articular ankylosis with a MMO  $\leq$  10 mm. Grade 3 A bilateral formation of a pseudojoint between an abnormal Grade 3 B CPM and the inner aspect of the zygomatic bone determining an extra-articular ankylosis with a MMO  $\leq 10$  mm.

various connotations of JD. Obviously, this classification has to be intended as a proposal, considering that more cases have to be analyzed to evaluate its possible validity.

Table 2. Our proposal of JD's classification.

The diagnosis of JD is also often delayed because an OPG does not always permit suspecting a CPM enlargement, and the reported symptoms push the clinician to request an MRI with TMJ scans, which do not permit a proper CPM visualization. CT is frequently the last radiological examination performed. Multiplanar CT and 3D CT reconstructions, as well as VSP (Mimics, Materialise NV, Leuven, Belgium), represent the most effective tool for the correct diagnosis and surgical planning of JD cases. The definitive treatment of JD is surgical. Those submitted to surgery are patients that obviously cannot open their mouths properly, and this also entails anesthesiological issues, such as often having to resort to awake intubation or the use of a fibroscope. We saw that, despite this, in 66 cases (56.9%), the coronoidectomy was performed by an intraoral approach, while an extraoral approach was used 32 times (27.6%), and a combined approach was used 12 times (10.3%). This analysis shows slight differences in the rates reported in the literature but a significant 10% increase in the percentage of intraoral coronoidectomies previously reported [7]. Despite the difficulty of operating in such a confined space, made even more uncomfortable by the pathology, the intraoral approach is nevertheless preferred because of its relative lack of possible complications (almost no risk for facial nerve injury), such as eliminating surgical skin scars and a good possibility to reach the abnormal CPM. This approach can be implemented by various mini-invasive techniques, primarily endoscopy [115]. Other authors propose different approaches, such as coronal, hemi-coronal or trans-zygomatic [62,64]. In grade 1 cases, a conservative attitude can be taken into account, and surgery may be postponed depending on the rate of worsening of the MMO or radiological changes. In grade 2 and 3 patients, in our opinion, surgery should be considered in the first instance. We did not highlight any significant difference in MMO post-treatment, which resulted in 40.2 mm overall (20–64 mm), between patients treated via an intraoral approach (38.2 mm; 20–61 mm), extraoral (42.6 mm; 28–55 mm) and combined approaches (44.3 mm; 35–64 mm). A parameter that would be interesting to evaluate is the follow-up of these patients in order to verify any radiological or clinical recurrence. Unfortunately, in only 41 (35.3%) out of the 116 cases are the follow-up periods reported, with an average of 17.4 months. Regarding this, only two cases of recurrence (1.7%) were highlighted in our review—a percentage in

line with what is reported in the literature [4]. However, it should be noticed, as it was in the case of follow-up, that no explicit mention was made about the future fate of the patients in 92 (79.3%) of the cases examined.

#### 5. Conclusions

JD is a complex condition where a pseudojoint is established between an abnormal CPM and the inner aspect of the zygomatic bone, determining a progressive and worsening reduction of MMO. The diagnosis is often delayed because of its similarities with TMD and other more frequent causes of MMO reduction. CT scan that has to be performed for its provisional diagnosis, is often requested as the last radiological diagnostic examination. The spectrum of disease presentation is extremely wide, and a classification of the disease is certainly something useful that should be established internationally. Our proposal has to be intended as suggestive. A widely accepted classification may help physicians who detect this condition make a more specific diagnosis and more standardized treatment planning. The intraoral coronoidectomy approach is the most widely used procedure and allows a comparable clinical result to the extraoral approach with a lower risk of complications. More information about the follow-up of these patients is needed. A very low recurrence rate is confirmed.

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