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Review

Efficacy of proportional versus high condylectomy in active condylar hyperplasia — A systematic review



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ABSTRACT

Objective: To perform a systematic literature review to test the efficacy of proportional condylectomy versus high condylectomy in patients with active condylar hyperplasia, in terms of avoiding secondary surgeries.

Method: Following a search of Medline (Pubmed), Embase, Scopus, Web of Science and Cochrane databases, ten studies were included for qualitative analysis, and two studies were included for meta-analysis. Quality assessment was performed using the Newcastle–Ottawa scale for cohort studies and the 18-item modified Delphi technique for case series.

Results: 259 patients were included in the qualitative analysis, with a weighted arithmetic mean age of 20.4 years, and a female:male ratio of 2:1. Meta-analysis was carried out for 52 patients, and it was found that proportional condylectomy reduced the need for secondary surgery ($p = 0.0003$). Although this evidence had limitations, excised bone on proportional condylectomy was superior when compared with excised bone on high condylectomy, re-establishing the occlusal plane, resulting in fewer asymmetries, and therefore reducing the need for further surgery.

Conclusions: This systematic review showed a tendency for proportional condylectomies to avoid additional surgeries; however, more comparative studies are necessary.

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1. Introduction

Mandibular condylar hyperplasia (CH) is defined as a growth abnormality of the temporomandibular joint caused by hyperactivity of the growth center of the mandibular condyle or a tumor in the condyle, resulting in progressive asymmetry (Karssemakers et al., 2018; Mouallem et al., 2017; Saridin et al., 2010a, 2010b; 2009; Vernucci et al., 2018; Walters et al., 2013). It is an idiopathic condition, but it is associated with hormonal, genetic, and/or traumatological factors, among others reported in the literature (Ghaws et al., 2016; Karssemakers et al., 2018; López et al., 2018; Martin-Granizo et al., 2017; Mouallem et al., 2017; Vernucci et al., 2018). This condition is more prevalent in women than in men,

although compared with other mandibular bone conditions, CH is uncommon (Vásquez et al., 2017).

Several classifications of CH have been reported (Rodrigues and Castro, 2015). Delaire, in 1983, considered two biotypes, one predominantly vertical and one transverse (Rajkumar et al., 2012; Mouallem et al., 2017). Obwegeser and Makek, in 1986 considered three classifications: hemimandibular hyperplasia (HH), hemimandibular elongation (HE), and a hybrid type that combines characteristics of HH and HE (Abuzinada and Alyamani, 2012; Deleurant et al., 2008; Fariña et al., 2015; Lippold et al., 2007; Martin-Granizo et al., 2017). Wolford, in 2014, established the following types: CH type 1 (CH1), causing condylar and horizontal mandibular growth, which could be bilateral (CH1A) or unilateral (CH1B); CH type 2, which is caused by an osteochondroma with a condylar enlargement (CH 2A) or exophytic osteochondroma (CH 2B); CH type 3, which includes rare benign tumors originating from the mandibular condyle; and CH type 4, including malignant tumors originating from the mandibular condyle (Ji et al., 2017; Martin-Granizo et al., 2017; Wolford et al., 2014, 2009, 2002).

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These three classifications have two points in common: first, patients with a predominantly horizontal growth (transverse as described by Delaire; hemimandibular elongation according to Obwegeser and Makek; and CH1B as stated in Wolford), present an important laterognathia, causing a posterior crossbite tendency or edge to edge bite, with lateral displacement on the non-affected side (Mouallem et al., 2017; Posnick et al., 2017; Rodrigues and Castro, 2015). This can develop an ipsilateral class III malocclusion, prognathism, chin deviation, and an opened gonial angle with no major discrepancy in the ramus shape between both sides (Deleurant et al., 2008; Gray et al., 1990; Iannetti et al., 1989; Posnick et al., 2017; Wolford et al., 2009). A slight occlusal cant might also be present, and a depressed commissure of the lips on the affected side (Ji et al., 2017; Posnick et al., 2017; Walters et al., 2013).

The second point, is that vertical growth predominance (vertical as described by Delaire; hemimandibular hyperplasia according to Obwegeser and Makek's classification; and CH2A as stated by Wolford — excluding the histopathological finding of osteochondroma) presents an enlargement of the condylar head and neck, increasing the height of the ramus on the affected side, and displacing the mandible body inferiorly (Abuzinada and Alyamani, 2012; Fariña et al., 2015; Han et al., 2018; Hernández-Alfaro et al., 2016; Ji et al., 2017; Jones and Tier, 2012; Lippold et al., 2007; Motamedi, 1996; Nolte et al., 2016; Rodrigues and Castro, 2015; Xu et al., 2014). It is possible to observe vertical facial discrepancy, a canted occlusal plane, and a downward tilt of the lip commissure on the affected side (Walters et al., 2013).

To decide on the appropriate treatment for CH, it is first necessary to determine the pattern of growth activity, and thus determine the progression of asymmetry (Di Blasio et al., 2015; Rodrigues and Castro, 2015; Rushinek et al., 2016; Wolford et al., 2014, 2009). The combination of methods such as progressive radiographs and dental casts may give an idea of this activity. In some studies, those methods were complemented with single photon emission computed tomography (SPECT), assuming a difference of 10% between the affected condyle and the normal condyle (AlSharif et al., 2014; Hamed et al., 2017; Martin-Granizo et al., 2017; Mouallem et al., 2017; Rushinek et al., 2016). In other studies only SPECT was applied to perform the diagnosis (Vernucci et al., 2018; Fariña et al., 2016, 2015; Hernández-Alfaro et al., 2016; Elmozen et al., 2015). However, the accuracy of SPECT was highly variable in several reports, showing it to be a controversial method in need of more evidence to justify its use (Chan and Leung, 2018; Hamed et al., 2017; Rushinek et al., 2016; Saridin et al., 2009; Wen et al., 2014). It is also important to consider the distributions of growth and biotypes that can be found in condylar hyperplasia (Ghaws et al., 2016; Walters et al., 2013).

Despite of the presence of common factors, therefore, diagnosis as well as treatment procedures have not been standardized, and it is possible to find several surgical management protocols and algorithms in the literature (Chiarini et al., 2014; Fariña et al., 2015; Ghaws et al., 2016; Nelke et al., 2018; Rodrigues and Castro, 2015; Saridin et al., 2010a; Vernucci et al., 2018; Villanueva-Alcojol et al., 2011; Wolford et al., 2014, 2002; Xu et al., 2014). The most common methods reported in the literature are: the application of orthognathic surgery alone (Motamedi, 1996; Nelke et al., 2018; Posnick et al., 2017; Vernucci et al., 2018); the exclusive surgical treatment of the temporomandibular joint (TMJ) (Nelke et al., 2018; Vernucci et al., 2018), including high condylectomy (Karssemakers et al., 2018; López et al., 2018; Martin-Granizo et al., 2017) or low, 'proportional' condylectomy (Mouallem et al., 2017; Vásquez et al., 2017); and a combination of condylectomy and orthognathic surgery performed in either one or two surgical sessions (Hampf et al., 1985; Vernucci et al., 2018; Wolford et al., 2014).

For active condylar hyperplasia (ACH), condylectomies are an adequate treatment option. Low condylectomy is used for removing TMJ tumors. This surgical approach involves not only removing the hyperactive portion, but also restoring the occlusal plane by resecting the necessary quantity of bone to match the non-affected side. For this reason it is called proportional condylectomy (Fariña et al., 2016, 2015; Mouallem et al., 2017; Wolford et al., 2014). In some reports the term 'proportional condylectomy' is synonymous with low condylectomy, regardless of the histological diagnosis. In cases where low condylectomy is performed for osteochondroma removal, the healthy side is used as a comparison in order to level the occlusal plane, reducing the asymmetry in the same way as a proportional condylectomy (Fariña et al., 2016, 2015; Hernández-Alfaro et al., 2016; Mouallem et al., 2017; Vásquez et al., 2017). With high condylectomy, progressive growth of the asymmetry can be avoided simply by removing the necessary bone. Subsequently, depending on the condition of the patient, it can be treated using an orthodontic approach (Di Blasio et al., 2015; Chen et al., 1996; Elmozen et al., 2015). Either method can avoid the need for further intervention, which is preferable for the patient.

In consideration of the above, the objective of this systematic review was to evaluate the efficacy of proportional condylectomy versus high condylectomy in patients with active condylar hyperplasia, in terms of avoiding additional surgical procedures.

2. Materials and methods

2.1. Design and eligibility criteria

This systematic review was carried out according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) (Shamseer et al., 2015) and the recommendations of the Cochrane Handbook for Systematic Reviews of Interventions (Higgins and Green, 2011). The PROSPERO database registration was made under number CRD42018108368. The search terms were established using the PICO system: P — patients with active condylar hyperplasia; I — proportional condylectomy; C — high condylectomy; O — additional surgical procedures.

The inclusion criteria included randomized clinical trials, cohort studies, case-controlled studies, and case series studies published in the English language that described the main treatment and information about additional surgical procedures. Also, only studies with at least five patients with confirmed active condylar hyperplasia (ACH) who underwent a condylectomy (high and/or proportional), with reports of absence or presence of additional surgical treatments, were considered. There was no restriction on year of publication. Articles that included data for inactive condylar hyperplasia, in-vitro studies, case reports, reviews and studies involving animals, were excluded.

2.2. Search strategy

Searches of articles published up to 4 June 2018, from five databases (MEDLINE (PubMed), Embase, Scopus, Web of Science, and Cochrane), were performed using the following terms: "(Condylar AND hyperplasia) OR (unilateral AND condylar AND hyperplasia) OR (hemimandibular AND elongation) OR (hemimandibular AND hyperplasia) OR (early AND high AND condylar AND hyperplasia)".

Article selection by reading titles and abstracts was carried out by two blinded and independent researchers (TN-S and FM). Differences of opinion relating to inclusion or exclusion of an article in the revision were resolved by consensus, with a third researcher (BV) being consulted to make a final decision. If necessary, additional information was requested from authors. A kappa test was

used to determine the level of agreement between the researchers after the selection of titles and abstracts.

2.3. Data collection process

After reading the articles, three evaluation tables were created using the most relevant data for our systematic review.

For the assessment of the risk of bias, the Cochrane collaboration tool for randomized clinical trials (Higgins and Green, 2011), the Newcastle–Ottawa quality assessment form for cohort studies and case control studies (Wells et al., 2011; Zeng et al., 2015), and the 18-item modified Delphi technique for appraising case series studies (Moga et al., 2012; Zeng et al., 2015), were planned.

2.4. Statistical analysis

LibreOffice Calc was used to perform descriptive statistics. In order to evaluate the efficacy of the condylectomies in avoiding unnecessary secondary surgeries, Review Manager 5.3 was employed to perform a meta-analysis. Considering the dichotomous nature of the data, in terms of presence/absence of subsequent surgical procedures, an inverse variance method and a determination of the relative risk (RR) with a fixed effect were applied.

3. Results

1364 studies were identified: 412 from PubMed/MEDLINE, 367 from Embase, 318 from Scopus, 264 from Web of Science, and three from the Cochrane database. After reviewing titles and abstracts, 202 articles were preselected. Duplicates were removed and 55 studies were submitted to full-text analysis.

Kappa coefficients had a high level of agreement according to Landis and Koch (1977). Values of 0.85, 0.91, and 1.0 were obtained for Pubmed/MEDLINE, Scopus, and Cochrane respectively. Coefficients of 0.89 were achieved for the Embase and Web of Science databases.

After full-text reading of the selected articles, 45 were excluded: 26 for not having clear data on the surgical procedures performed after the first surgery (Brusati et al., 2010; Deng et al., 2009; Elbaz et al., 2014; Fariña et al., 2011; Götz et al., 2007; Gray et al., 1990; Guo et al., 2016; Hamed et al., 2017; Ji et al., 2017; Jones and Tier, 2012; Karssemakers et al., 2018, 2014; Lippold et al., 2007; López et al., 2018; Martin-Granizo et al., 2017; Nelke et al., 2018; Olate et al., 2015, 2014; 2013; Rodrigues and Castro, 2015; Saridin et al., 2010a, 2010b; 2009; Vásquez et al., 2017; Wen et al., 2014); six for having data with inactive cases (AlSharif et al., 2014; Han et al., 2018; Mouallem et al., 2017; Norman and Painter, 1980; Rushinek et al., 2016; Vernucci et al., 2018); five for not specifying clearly the type of condylectomy carried out (Chen et al., 1996; Rajkumar et al., 2012; Hampf et al., 1985; Henderson et al., 1990; Vásquez et al., 2016); four for not including condylectomy in the initial surgical treatment (Iannetti et al., 1989; Motamedi, 1996; Posnick et al., 2017; Xu et al., 2014); three for not having a minimum of five patients undergoing a condylectomy (Abuzinada and Alyamani, 2012; Choung and Nam, 1998; Mehrotra et al., 2011); and one (Wolford et al., 2002) because it used the same sample as a later study (Wolford et al., 2009) — the latter was chosen because it used a larger sample of patients and provided a better description of the data.

Ten articles were chosen for the qualitative analysis — six were retrospective case series (Di Blasio et al., 2015; Chiarini et al., 2014; Deleurant et al., 2008; Fariña et al., 2015; Hernández-Alfaro et al., 2016; Villanueva-Alcojol et al., 2011) and four were retrospective cohort studies (El.mozen et al., 2015; Fariña et al., 2016; Wolford

et al., 2014, 2009). No randomized clinical trials or case-controlled studies were found. From those ten selected articles, two performed a quantitative comparison between proportional and high condylectomy (Fariña et al., 2016; Hernández-Alfaro et al., 2016). Fig. 1 shows the article selection process.

3.1. Demographic data and diagnosis

Table 1 shows the demographic characteristics of patients and diagnosis of condylar hyperplasia. 259 patients were considered, with a weighted arithmetic mean age of 20.4 years; 175 were female and 84 were male.

114 patients were reported to have a vertical development of the pathology. These included 77 females (Di Blasio et al., 2015; Chiarini et al., 2014; Fariña et al., 2016; Hernández-Alfaro et al., 2016; Villanueva-Alcojol et al., 2011; Wolford et al., 2014), 34 males (Di Blasio et al., 2015; Chiarini et al., 2014; Fariña et al., 2016; Hernández-Alfaro et al., 2016; Villanueva-Alcojol et al., 2011; Wolford et al., 2014), and three of unknown gender (Fariña et al., 2015). Of those patients, 42 had histological osteochondroma (Hernández-Alfaro et al., 2016; Wolford et al., 2014) — 32 females and 10 males. Horizontal development was reported in 101 patients. These included 61 females (Deleurant et al., 2008; Hernández-Alfaro et al., 2016; Villanueva-Alcojol et al., 2011; Wolford et al., 2009), 27 males (Deleurant et al., 2008; Villanueva-Alcojol et al., 2011; Wolford et al., 2014, 2009), and 13 of unknown gender (Fariña et al., 2015). Four patients (all female) were reported as hybrid type (Villanueva-Alcojol et al., 2011). Growth tendency was not reported for the other 40 patients (El.mozen et al., 2015).

The left side was affected in 68 patients (Di Blasio et al., 2015; Chiarini et al., 2014; Deleurant et al., 2008; El.mozen et al., 2015; Fariña et al., 2015; Hernández-Alfaro et al., 2016; Villanueva-Alcojol et al., 2011; Wolford et al., 2014), and the right side in 88 patients (Di Blasio et al., 2015; Deleurant et al., 2008; El.mozen

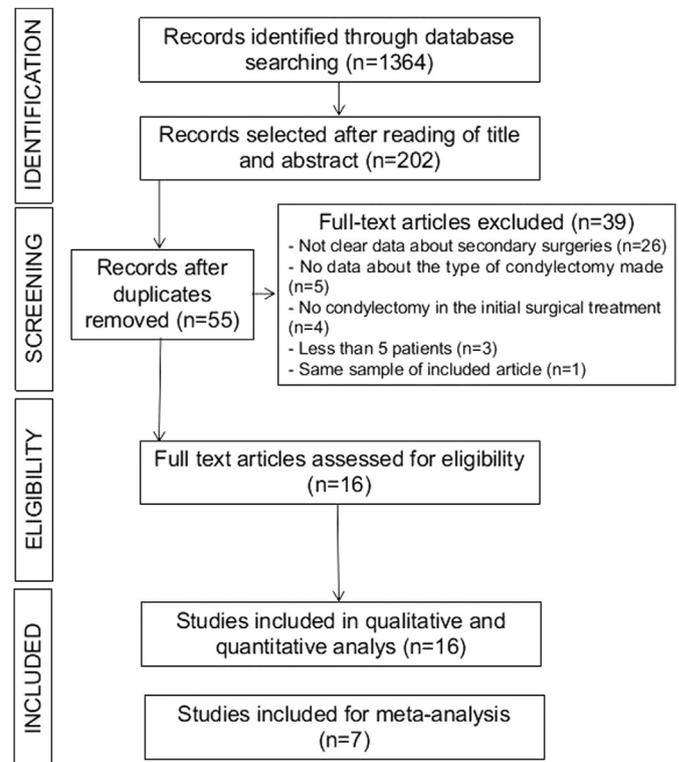


Fig. 1. Article selection process.

Table 1
Demographic data and diagnosis

Author/year	Patients	Age (mean/range)	Sex	Type of hyperplasia	Affected side
Hernández-Alfaro et al. (2016)	7	30.85	F: 6	CH2: 5	Left: 6 F: 5 M: 1
		22–42	M: 1	CH1B: 2 F: 2	Right: 1 F: 1 NR
Fariña et al. (2016)	49	19.7 ± 3.72	F: 32	HH (based on clinical characteristics)	
	G1: 11	G1: 19.27 ± 3.65	M: 17		
	G2: 38	G2: 19.83 ± 3.72	F: 7 M: 4 G2: F: 25 M: 13		
Fariña et al. (2015)	16	19.125	F: 10	HE: 13	Left: 6 F: 4 M: 2
		14–33	M: 6	HH: 3	Right: 10 F: 6 M: 4
El.mozen et al., 2015	40	14–33	F: 23 M: 17	NR	Left: 12 Right: 28
	G1: 24	G1: 19.83 ± 3.26	G1: F: 13 M: 11		G1: Left: 8 Right: 16
	G2: 16	G2: 19.93 ± 4.13	G2: F: 10 M: 6		G2: Left: 4 Right: 12
Di Blasio et al. (2015)	8	T0: 13.3 11.2–13.8 T1: 15.2 13–18 T2: 17.1 14.10–20 26.3	F: 7	CH2 (no osteochondroma, vertical vector): 8	Left: 6
		13–48	M: 1		Right: 2
Wolford et al. (2014)	37	26.3 13–48	F: 28 M: 9	Horizontal growth initially diagnosed as CH1B, but osteochondroma present: 1 CH2: 36	Left: 17 Right: 20
Chiarini et al. (2014)	5	16.8 14–17	F: 2 M: 3	CH2 with enlargement of condyle head, no osteochondroma: 5	Left: 5
Villanueva-Alcojol et al. (2011)	36	22.7 ± 6.7	F: 25	CH1 (HE): 24 F: 17 M: 7	Left: 14 F: 10 CH1: 8 CH2: 1 Hybrid: 1
		11–42	M: 11	CH2 (HH): 8 F: 4 M: 4	M: 4 CH1: 3 CH2: 1 Right: 22 F: 15 CH1: 9 CH2: 3 Hybrid: 3 M: 7 CH1: 4 CH2: 3
				Hybrid: 4 F: 4	Hybrid: 3 M: 7 CH1: 4 CH2: 3
Wolford et al. (2009)	54	G1: 17.5	F: 36 M: 18	CH1: 54	Group 1, both sides: 12
	G1: 12	13–25	G1: F: 8 M: 4	CH1A: 36	Group 2, both sides: 24
	G2: 42	G2: 16.6 13–24	G2: F: 28 M: 14	CH1B: 18	Unilateral NR: 18
Deleurant et al. (2008)	7	16.3 13–25	F: 6 M: 1	HE: 7	Left: 2 Right: 5

F, female; M, male; HH, hemimandibular hyperplasia; HE, hemimandibular elongation; CH, condylar hyperplasia; G1, group 1; G2, group 2; NR, not reported.

et al., 2015; Fariña et al., 2015; Hernández-Alfaro et al., 2016; Villanueva-Alcojol et al., 2011; Wolford et al., 2014). 36 cases were reported as bilateral (Wolford et al., 2009). In 67 patients the side was not specified (Fariña et al., 2016; Wolford et al., 2009).

To determine the growth activity of the condyles, clinical and radiological evolution were observed in two studies (Di Blasio et al., 2015; Wolford et al., 2009), SPECT was applied in six reports (Chiarini et al., 2014; Deleurant et al., 2008; El.mozen et al., 2015; Fariña et al., 2016, 2015; Hernández-Alfaro et al., 2016; Villanueva-Alcojol et al., 2011), and both methods were applied in one (Wolford et al., 2014).

3.2. Surgical procedure characteristics

As shown in Table 2, surgery comprising condylectomy only was carried out in 168 condyles — 109 were high condylectomy and 59 were proportional. Condylectomy in conjunction with orthognathic surgery was performed in 79 cases (43 high condylectomy (Wolford et al., 2014, 2009) and 36 proportional condylectomy (Wolford et al., 2014)), and orthognathic treatment alone was applied in 12 patients (Wolford et al., 2009).

To perform the condylectomy, a piezoelectric tool was used in four studies (Chiarini et al., 2014; Deleurant et al., 2008; Fariña et al., 2016; Hernández-Alfaro et al., 2016) and a burr in one study (Fariña et al., 2015), with apparatus not reported in the remaining five studies (Di Blasio et al., 2015; El.mozen et al., 2015; Villanueva-Alcojol et al., 2011; Wolford et al., 2014, 2009). The surgical approach intraoral in one study (Hernández-Alfaro et al., 2016), while periauricular access was used in five studies (Chiarini et al., 2014; El.mozen et al., 2015; Fariña et al., 2016, 2015; Villanueva-Alcojol et al., 2011).

The weighted arithmetic mean for excised bone was 9.23 mm for proportional condylectomy (Fariña et al., 2016, 2015) and ranged from 3 to 6 mm for high condylectomy (Di Blasio et al., 2015; Chiarini et al., 2014; Villanueva-Alcojol et al., 2011; Wolford et al., 2009). Minimal complications relating to surgery were reported in two studies (Fariña et al., 2015; Hernández-Alfaro et al., 2016). Orthodontic procedures were reported in 139 patients (Chiarini et al., 2014; El.mozen et al., 2015; Fariña et al., 2016, 2015; Hernández-Alfaro et al., 2016; Villanueva-Alcojol et al., 2011; Wolford et al., 2014). To control the occlusion postsurgically, and maintain a stable condylar position, these orthodontic treatments were applied in conjunction with intermaxillary elastic therapy for 1–2 weeks (Wolford et al., 2009, 2014), or 1–3 months (Fariña et al., 2015, 2016). El.mozen et al. also mentioned using a multi-loop edgewise arch wire with intermaxillary elastic traction in 24 patients, but the period of application was not specified (El.mozen et al., 2015).

Meta-analysis was evaluated in 52 patients (Fig. 2). Proportional condylectomy significantly reduced the need for secondary surgery compared with high condylectomy ($p = 0.0003$; MD: 0.54; CI: 0.34–0.84) (Fariña et al., 2016; Hernández-Alfaro et al., 2016).

3.3. Comparison of pre- and postsurgical measurements

Table 3 shows the occlusal, symmetric, and temporomandibular status before and after surgery. Facial asymmetry is shown to be a main characteristic of condylar hyperplasia, with progressive growth in active cases (Di Blasio et al., 2015; Chiarini et al., 2014; Deleurant et al., 2008; El.mozen et al., 2015; Fariña et al., 2016, 2015; Hernández-Alfaro et al., 2016; Villanueva-Alcojol et al., 2011; Wolford et al., 2014). This asymmetry is generally accompanied with chin deviation to the contralateral side under the influence of either horizontal or vertical growth factors (Fariña et al., 2016; Hernández-Alfaro et al., 2016). Vertical growth is

determined by a ramus discrepancy (Di Blasio et al., 2015; Fariña et al., 2015), sometimes with bowing of the lower border. After proportional condylectomy, improvement in facial symmetry was observed, as a result of the reduction in vertical discrepancy between both sides (Fariña et al., 2016, 2015; Hernández-Alfaro et al., 2016).

Asymmetry was also reduced after high condylectomy, with satisfactory results (Di Blasio et al., 2015; Chiarini et al., 2014; Deleurant et al., 2008; El.mozen et al., 2015; Fariña et al., 2016; Hernández-Alfaro et al., 2016; Villanueva-Alcojol et al., 2011), although with less improvement compared with proportional condylectomy, according to Fariña et al. (2016) (Fariña et al., 2016). Moreover, with this type of condylectomy, immediate results were not achieved and subsequent evaluations were necessary (Di Blasio et al., 2015; Deleurant et al., 2008). Satisfactory results with high condylectomy were associated with orthodontic treatment performed after surgery (El.mozen et al., 2015; Fariña et al., 2016; Hernández-Alfaro et al., 2016; Villanueva-Alcojol et al., 2011), or with additional surgery (Fariña et al., 2016; Hernández-Alfaro et al., 2016; Wolford et al., 2014). On the other hand, condylectomy in conjunction with orthognathic surgery was able to address changes relating to chin and dental deviation (Wolford et al., 2014, 2009).

In relation to occlusal changes, it can be seen that anterior and posterior crossbite, as well as tilted occlusal plane, were corrected after proportional condylectomy (Fariña et al., 2015). High condylectomy reduced occlusal changes, including tilted occlusal plane (Deleurant et al., 2008; El.mozen et al., 2015; Wolford et al., 2014), but when not accompanied with postsurgical orthodontics, it could take longer to adjust to normal occlusion or simply to maintain any occlusal alterations (El.mozen et al., 2015).

When combining high condylectomy with orthognathic surgery, mandibular measures were statistically different from those patients with orthognathic surgery only, after the longest follow-up (Wolford et al., 2009). When high condylectomy and orthognathic surgery were carried out at different times, there was an increase in the angle formed by the S–N and palatal planes, and a reduction in the angle formed by the palatal and mandibular planes. According to Deleurant et al., worsening of presurgical conditions towards class II was presented in four patients, contributing to a change in ANB (Angle that determines the anteroposterior relationship of the maxilla with the mandible, formed by the A point (A), located in the deepest region of the anterior maxillary contour, by Nasion (N), located in the anterior part of the frontonasal suture, and by the B point located in the deepest zone of the anterior mandibular contour) mean from 3.1 to 5.1 (Deleurant et al., 2008).

Regarding the TMJ after surgical intervention, there was an improvement in pathological condition (Villanueva-Alcojol et al., 2011; Wolford et al., 2009), most of the healthy presurgical characteristics were maintained, and there were no significant TMD complications (Di Blasio et al., 2015; Chiarini et al., 2014; Fariña et al., 2015; Hernández-Alfaro et al., 2016; Villanueva-Alcojol et al., 2011; Wolford et al., 2014, 2009).

3.4. Risk of bias

Table 4 reports the results of the Newcastle–Ottawa quality assessment for cohort studies included in this systematic review. Table 5 summarizes the application of the 18-item modified Delphi technique to the case series studies included in this review.

4. Discussion

This systematic review evaluated the efficacy of proportional and high condylectomies in terms of avoiding secondary surgery after the first surgical procedure. The literature included in this

Table 2
Surgical characteristics of studies selected.

Author/year	Surgical treatment	Surgical approach	Tool used in the condylectomy	Bone excised (mm)	Orthodontic treatment	Complications	Secondary surgical treatment	Follow-up (months)
Hernández-Alfaro et al. (2016)	High condylectomy: 2 Low condylectomy: 5	Intraoral	Piezoelectric	NR	Conventional full preparation: 7	Minimal postoperative swelling: 7	Orthognathic surgery: 7	8.7
Fariña et al. (2016)	G1, high condylectomy: 11 G2, proportional condylectomy: 38	Preauricular	Piezoelectric	G1: 5.81 ± 0.93 G2: 9.28 ± 2.55	Postsurgical: 49	NR	Orthognathic surgery: 16 G1: 10 G2: 6 None: 33 G1: 1 G2: 32	≥18
Fariña et al. (2015)	Low (proportional) condylectomy: 16	Preauricular	Burr	9.125 5–15	Presurgical orthodontic braces used, but teeth not necessarily aligned or in good occlusion, postsurgical: 16 G1, postsurgical: 24 G2, did not take orthodontic therapy: 16 NR	Transitory facial paresis of the frontalis branch, recovered after 2 months: 1 No long-term complication NR	Orthognathic surgery: 2 Not presented: 14	62.25
El.mozen et al., 2015	High condylectomy: 40	Preauricular	NR	NR	NR	NR	None: 40	Postsurgical: 18.4 Postorthodontic: 13.8
Di Blasio et al. (2015)	High condylectomy: 8	NR	NR	4–5	NR	NR	None: 8	T0–T1: 23 T1–T2: 23 48
Wolford et al. (2014)	High condylectomy and orthognathic surgery: 1 Low condylectomy and orthognathic surgery: 36	NR	NR	NR	Postsurgical: 2	NR	Low condylectomy: 1 (patient with high condylectomy) None: 36	48
Chiarini et al. (2014)	High condylectomy: 5	Preauricular	Piezoelectric	6	Presurgical: 5	Not presented in long term	None: 5	24
Villanueva-Alcojol et al. (2011)	High condylectomy: 36	Preauricular	NR	4–5	Postsurgical: 36	NR	Orthognathic surgery: 6	51.6
Wolford et al. (2009)	Orthognathic surgery, G1: 12 High condylectomy + orthognathic surgery, G2: 42	NR	NR	3–5	Not clearly specified	NR	G1, additional surgical interventions: 12 G2, secondary surgery, repeating maxillary surgery: 1	G1: 67.2 G2: 61.2
Deleurant et al. (2008)	High condylectomy (condylar shaving): 7	NR	Piezoelectric	NR	Yes, but not specified	NR	Orthognathic surgery: 7	52

G1, group 1; G2, group 2; NR, not reported.

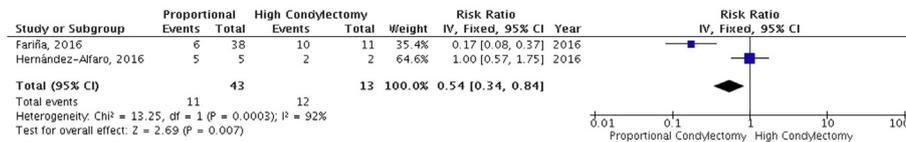


Fig. 2. Comparison between proportional condylectomy and high condylectomy.

study was very heterogeneous. Risk of bias assessment showed that most of the case series presented a moderate risk (Table 5), while cohort studies were of good quality, with the exception of one study described as a retrospective cohort study, although its methodology suggested more of a case series study (Wolford et al., 2014) (Table 4).

Despite the heterogeneity of the included studies and their methodological limitations, in agreement to data found in this study, balance leaning towards proportional condylectomy. Besides meta-analysis results, this tendency can be also found slightly in proportions. Of the 95 patients submitted to proportional condylectomy (Fariña et al., 2016, 2015; Hernández-Alfaro et al., 2016; Wolford et al., 2014), 13.68% underwent secondary surgery. Of the 152 high condylectomy patients, 17.76% underwent secondary surgery (Deleurant et al., 2008; Fariña et al., 2016; Hernández-Alfaro et al., 2016; Villanueva-Alcojol et al., 2011; Wolford et al., 2014, 2009). The remaining 12 patients were submitted to orthognathic surgery only, without condylar intervention. As expected, all of these required secondary surgery after redeveloping class III deformities (Wolford et al., 2009).

An explanation for this might be the presurgical conditions. In the two studies included in the meta-analysis, there were no statistically significant differences between those patients submitted to high condylectomies and those submitted to proportional condylectomies in terms of chin deviation or discrepancies between the condylar process and mandibular ramus. However, in the study by Hernández-Alfaro, although a proportional condylectomy was performed in five patients, a double-surgery approach was planned (Hernández-Alfaro et al., 2016). Meanwhile, for Fariña et al. (2016), the objective was to compare both condylectomies in patients with similar characteristics, in terms of avoiding secondary surgery. Presurgical conditions were not ideal for avoiding additional surgery following high condylectomy because the discrepancies were greater than 9 mm; hence, 10 out of 11 patients were submitted to orthognathic correction (Fariña et al., 2016). On the other hand, the cases in studies by El. Mozen et al., Di Blasio et al., and Chiarini et al. did not require postsurgical orthognathic surgery after a high condylectomy of no more than 6 mm, because the surgical technique was enough to reach the necessary occlusal plane and, in the absence of other skeletal discrepancies, no further surgeries were required (El.mozen et al., 2015; Di Blasio et al., 2015; Chiarini et al., 2014).

Likewise, in the case of Fariña et al. (2015), proportional condylectomy significantly reduced the chin deviation and any discrepancies between both sides. The marked preexistence of dentoskeletal changes was a relevant reason for subsequent surgery, which was not directly related to the presence of condylar hyperplasia or the type of condylectomy (Fariña et al., 2015). This was also the case with Deleurant et al. (2008).

This indicates that severe dentoskeletal deformations are variables that should be controlled in this type of study, in order to observe in a more objective way the efficacy of condylectomies in terms of avoiding further surgery. This was demonstrated in the study by Villanueva-Alcojol et al., where secondary surgery was applied to correct any residual occlusal and facial asymmetry after the initial high condylectomy, when it could have been avoided

using proportional condylectomy (Villanueva-Alcojol et al., 2011). Despite these findings, evidence based on presurgical measures regarding discrepancy, chin deviation, and midline deviation, did not provide enough quantitative data for our systematic review to propose a treatment algorithm based on clinical measures. This is an issue that needs to be addressed in future studies.

Based on the above findings, evaluating the hyperactivity of the condyle is a crucial factor in determining the success of treatment. Although many of the included articles based their diagnosis on SPECT, this is considered by many authors as a diagnostic aid that should be applied when there is a suspicion of progressive condylar growth after imaging and clinical diagnosis (AlSharif et al., 2014; Rushinek et al., 2016; Wolford et al., 2014).

According to Chan and Leung, serial radiographs and clinical assessment are irreplaceable compared with scintigraphy, and SPECT is not justifiable if clinical and radiographic data are available (Chan and Leung, 2018). Scintigraphy does offer important specificity and sensitivity for some evaluation parameters, but does not represent an exact diagnosis, compared with clinical and radiographic evolution, because an accuracy of 100% has not yet been reported (Hamed et al., 2017; Rushinek et al., 2016; Saridin et al., 2009; Wen et al., 2014).

In this systematic review, exclusive clinical and radiological evolution analysis was observed in two studies (Di Blasio et al., 2015; Wolford et al., 2009). Only one study confirmed condylar hyperactivity by using images, clinical evolution, and SPECT values (Wolford et al., 2014). In spite of this, one patient in this study was misdiagnosed as CH1B, but histological analysis found the presence of an osteochondroma, and the patient needed a secondary proportional condylectomy. Such issues need to be resolved to allow a real and complete diagnosis, and subsequently establish effective treatment algorithms.

In the latter case, there was no suspicion of osteochondroma because this finding is generally attributed to a vertical growth (CH2), and this patient had a prominent horizontal growth tendency (Wolford et al., 2014). For this reason, it is also necessary to reach a consensus on the diagnosis of condylar hyperplasia. For instance, some articles reported a Wolford CH2 classification as a vertical growth, independently of the histological finding (Di Blasio et al., 2015; Chiarini et al., 2014), but the articles by Wolford clearly state that this classification includes osteochondroma (Wolford et al., 2014, 2009, 2002).

In order to enable the unification of diagnosis based on biotype, histopathological, and histomorphometric findings in excised bone, as well as on cephalometric findings, it is important to include occlusal features in addition to pre- and postsurgical asymmetry. Because of this variability, for this systematic review analysis was mainly based on the type of surgery and not on the biotype or on histological findings.

There are weaknesses not only in CH diagnosis, but also in its etiology, which is neither defined nor even explored. For example, in some cases trauma is reported to be a possible cause, while in other studies CH patients with a history of trauma were excluded (Hamed et al., 2017; Norman and Painter, 1980; Posnick et al., 2017). Hormonal factors are a possible cause for manifestation in adolescents, with a female predominance. In our study, although a

Table 3
Pre- and postsurgical measurements

Author/year	Presurgical facial asymmetry	Postsurgical facial asymmetry	Presurgical occlusal characteristics	Postsurgical occlusal characteristics	Presurgical TMJ health	Postsurgical TMJ health
Hernández-Alfaro et al. (2016)	Present: 7/7	Improvement: 7	Posterior open bite ipsilateral side: 6/7 Contralateral crossbite: 6/7 Anterior open bite: 3/7	Satisfactory: 7	Satisfactory: 7	Pain VAS 1–2: 4
Fariña et al. (2016)	Present: 49/49	Moderate improvement: 11 (G1) High improvement: 38 (G2)	Tilted occlusal plane	NR	NR	NR
Fariña et al. (2015)	Difference between condylar process and mandibular ramus G1: 10.81 ± 1.40 G2: 9.26 ± 2.56 Present: 14/16 Discrepancy between sides: 5.87% Length mandibular ramus Affected: 73.87 mm Healthy: 65.81 mm	Difference between condylar process and mandibular ramus G1: 5.11 ± 1.27 G2: 0.52 ± 1.21 Improvement: 14 Discrepancy between sides: 1.75% Length mandibular ramus Affected: 64.62 mm Healthy: 66.12 mm	G1: 4.72° G2: 4.75° Posterior crossbite: 83.3% Anterior crossbite: 75%	NR Crossbite: 0% Anterior crossbite: 0%	Satisfactory: 16	Satisfactory: 16
El. Mozen et al., 2015	Present: 40/40	Improvement: 40/40	Zero overjet: 8.3% Normal overjet: 16.7% Occlusal plane: 4.71° Occlusal disturbance: 40/40	Zero overjet: 16.7% Normal overjet: 83.3% Occlusal plane: 1.14 G1, adjusted to normal occlusion during the follow-up period: 24 G2, open bite still remained: 7 G2, adjusted to normal occlusion during the follow-up period: 9	NR	NR
Di Blasio et al. (2015)	Present: 8/8 Total vertical area differences affected/unaffected: 6.3% Condylar area affected/unaffected: 10.6% Ramus area affected/unaffected: 4.6%	Moderate improvement: 8 Total vertical area differences affected/unaffected: -1.6% Condylar area affected/unaffected: -12.6% Ramus area affected/unaffected: 2.3%	NR	NR	Satisfactory: 8	Satisfactory: 8
Wolford et al. (2014)	Present: 37/37	Improvement: 36 Recurred facial asymmetry (high condylectomy): 1; improved after proportional condylectomy	Occlusal disturbance: 37/37 Incisal opening: 47.2 mm Right excursive movement: 7.8 Left excursive movement: 5.3	Class 1 occlusion stable: 34 Anterior open bite 2 mm: 1 Incisal opening: 44.9 mm Right excursive movement: 5.3 Left excursive movement: 5	Pain VAS: 3.6 Jaw function: 3.7 Diet: 3.2 Disability: 2.1	Pain VAS: 0.14 Jaw function: 1.5 Diet: 0.76 Disability: 0.24
Chiarini et al. (2014)	Present: 5/5	Improvement: 5/5	NR	Stable	Satisfactory: 5	VAS 1–3: 5
Villanueva-Alcojol et al. (2011)	Present: 36/36	Improvement: 36/36	Occlusal disturbance: 36	Satisfactory: 36/36	Unsatisfactory: 13 TMD clicking and mild pain: 8	Satisfactory: improvement in 13 with TMD
Wolford et al. (2009)	NR	NR	Occlusal disturbance: 54 Maximum incisal opening (mm) G1: 46.8 G2: 40.2 ANB: 3.1 SpaSPP-MeGO: 35.8 SN/SpaSpp: 5.6 Ar-Go-Me: 130.5	Satisfactory: 41 (G2) Unsatisfactory: 13 (12 G1; 1 G2) Maximum incisal opening (mm) G1: 46.7 G2: 49.5 ANB: 5.1 SpaSPP-MeGO: 31.2 SN/SpaSpp: 9.4 Ar-Go-Me: 129.8	Satisfactory, G1: 12 Pain VAS (G2): 0.6 Jaw Function (G2): 3.4	Satisfactory, G1: 12 Pain VAS (G2): 0.2 Jaw function (G2): 2.1
Deleurant et al. (2008)	Present: 7/7	Improvement: 7	ANB: 3.1 SpaSPP-MeGO: 35.8 SN/SpaSpp: 5.6 Ar-Go-Me: 130.5	ANB: 5.1 SpaSPP-MeGO: 31.2 SN/SpaSpp: 9.4 Ar-Go-Me: 129.8	Diet: 0.6 MMO: 53 mm	Diet: 0.4 MMO: 51 mm

G1, group 1; G2, group 2; TMJ, temporomandibular joint; TMD, temporomandibular joint dysfunction; MMO, maximal mouth opening; VAS, visual analogue scale; NR, not reported.

Table 4
Risk of bias, Newcastle–Ottawa scale (NOS).

Cohort studies									
Author/year	SELECTION				COMPARABILITY	OUTCOME			Quality
	Representativeness of the exposed cohort	Selection of the non-exposed cohort	Ascertainment of exposure	Demonstration that outcome of interest was not present at start of study	Comparability of cohorts on the basis of the design or analysis controlled for confounders	Assessment of outcome	Was follow-up long enough for outcomes to occur?	Adequacy of follow-up of cohorts	
Fariña et al. (2016)	*	*	*	No (Not clearly specified)	**	No description	*	*	Good
El.mozen et al., 2015	*	*	*		*		*	*	Good
Wolford et al. (2014)	No description				No description		*	No statement	Poor
Wolford et al. (2009)	*	*	*		**		*	*	Good

Table 5
18-item modified Delphi technique for case series.

Author/year	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18
Hernández-Alfaro et al. (2016)	U	Yes	No	PR	Yes	No	Yes	Yes	PR	U	U	No	Yes	U	No	Yes	PR	Yes
Fariña et al. (2015)	Yes	Yes	U	Yes	U	No	Yes	No	Yes	Yes	Yes	Yes	Yes	U	No	Yes	Yes	Yes
Di Blasio et al. (2015)	Yes	Yes	No	Yes	Yes	No	PR	No	Yes	Yes	Yes	Yes	Yes	No	No	No	Yes	PR
Chiarini et al. (2014)	Yes	Yes	No	No	U	No	Yes	No	No	No	No	No	Yes	No	No	PR	No	PR
Villanueva-Alcojol et al. (2011)	Yes	Yes	U	Yes	U	No	PR	No	PR	U	U	Yes	Yes	No	No	No	Yes	No
Deleurant et al. (2008)	Yes	Yes	No	Yes	U	No	PR	Yes	Yes	Yes	Yes	Yes	Yes	No	No	No	Yes	No

PR, partially reported; U, unclear.

greater predominance was observed in women in most reports, with an approximate ratio of 2:1 (AlSharif et al., 2014; Di Blasio et al., 2015; Deleurant et al., 2008; El.mozen et al., 2015; Fariña et al., 2016, 2015; Hernández-Alfaro et al., 2016; Mouallem et al., 2017; Norman and Painter, 1980; Rushinek et al., 2016; Vernucci et al., 2018; Villanueva-Alcojol et al., 2011; Wolford et al., 2014, 2009), there was no significant suggestion of a hormonal role in the etiology of the CH in the included articles or in the literature.

It is important to highlight certain limitations and weaknesses of this study. One of these relates to the fact that condylar pathology was not clearly defined in some studies, hindering the homogenization of data (El.mozen et al., 2015; Fariña et al., 2016). In addition, the lack of information in some reports made it difficult to select articles with maxillary deformities associated with condylar hyperplasia. In severe cases, secondary orthognathic surgery was necessary to reestablish occlusal harmony, which could affect the results presented in this review. Also, postsurgical controls in many studies were restricted to panoramic radiographs, omitting relevant information about the neocondyle and its process of morphological adaptation that could be analyzed from CT scans.

Cephalometric information was absent in most articles, hindering a quantitative conclusion about the efficacy of each type of condylectomy at an anatomical level. This factor also limited results at the occlusal level, especially in determining the existence of a remaining transverse cant in the occlusal plane after condylectomy, especially in patients diagnosed with osteochondroma. A related issue was that five studies did not report the postsurgical occlusal stabilization treatment, and therefore the process used to reach an ideal occlusal outcome was not explained (Di Blasio et al., 2015; Deleurant et al., 2008; Villanueva-Alcojol et al., 2011; Chiarini et al., 2014).

Finally, a relevant aspect that is generally ignored, but is vital for the success of treatment, is patient satisfaction. Since condylar hyperplasia is a progressive condition, it is important to observe how the changes after condylectomy can improve the patient's quality of life. These aspects should be considered in order to improve the quality of further studies.

5. Conclusion

The studies included in this systematic review revealed different methodologies in every step, from diagnosis to treatment of condylar hyperplasia. Therefore, it is important to emphasize that this systematic review aims to set initial guidelines for planning future studies.

In spite of the heterogeneity presented in some primary articles, the results obtained in this review indicate that proportional condylectomy should be performed in cases of osteochondroma, because of its active and constant growth. This procedure can also be used as an initial treatment in place of a high condylectomy in patients with increased clinical growth activity and prominent asymmetry. In the absence of conspicuous dentoskeletal deformation, with mild or even moderate midline deviation and mandibular occlusal canting, and without a marked bowing of the jaw edge, proportional condylectomy could be considered as a sole treatment.

Proportional condylectomy is an effective method for avoiding unnecessary secondary surgeries in active condylar hyperplasia. However, the limitations of this study indicate that the methodology for treating ACH needs to improve in order to allow generalization of data and to create more effective treatment algorithms.

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Conflicts of interest

None.

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