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Systematic Reviews/Meta-analyses

Clinical and radiologic outcomes of posterior column extension, pedicle subtraction, and vertebral column resection osteotomies in adult chin on chest deformity: A systematic review

Ergin Coskun, MD[b], Ian J. Wellington, MD[a], Chirag Chaudhary, MD[c], Kathleen Crea, MLS[d], Mark P. Cote, DPT, MSCTR[e], John M. Rhee, MD[f], Scott Mallozzi, MD[a], Isaac L. Moss, MDCM, MASc, FRCSC[a], Hardeep Singh, MD[a,*]

[a] Department of Orthopedics, The University of Connecticut, 120 Dowling Way, Farmington, CT 06032, United States
[b] Riley Hospital for Children, Indiana University Health, 705 Riley Hospital Dr, Indianapolis, IN 46202, United States
[c] Insight Surgical Hospital, 21230 Dequindre Rd Warren, MI 4809, United States
[d] Lyman Maynard Stowe Library, UConn Health, The University of Connecticut, 120 Dowling Way, Farmington, CT 06032, United States
[e] Massachusetts General Brigham Sports Medicine, Harvard Medical School, The University of Connecticut, 120 Dowling Way, Farmington, CT 06032, United States
[f] Department of Orthopedic Surgery, Emory Spine Center, Emory University, 59 Executive Park South, Atlanta, GA 30327, United States

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ABSTRACT

Background: Chin-on-chest deformity is a rare and severely disabling condition characterized by kyphotic deformity in the cervicothoracic spine. To treat this deformity, various osteotomy techniques were described.
Methods: A comprehensive literature search of biomedical databases including MEDLINE (via PubMed), Scopus (via Elsevier), Embase (via Elsevier), and Cochrane Library in English from 1/1/1990 to 3/31/2022 was conducted using a combination of text and Medical Subject Headings (MeSH).
Results: The final analysis included 16 studies. All the studies were assigned a level of evidence of four. Except for two articles, all of the articles were non-comparative studies. A total of 288 patients were included in this review. Of the 288 patients, 107 underwent posterior column extension osteotomy (PCEO), 108 underwent pedicle subtraction osteotomy (PSO), and 33 underwent vertebral column resection osteotomy (VCRO). The most common osteotomy level in fifteen of the studies was C7/T1. The studies included in this review described several techniques for cervical sagittal balance correction. The range of preoperative and postoperative visual analogue scale (VAS) scores was 5.5–8.6 to 1.7–4.91, respectively. The range of preoperative and postoperative neck disability index (NDI) was 34.2–65.4 to 22.1–51.3, respectively. The most common complications were upper extremity paresthesia and hand numbness through the C8 dermatome distribution.
Conclusions: Corrective osteotomies provide satisfactory results in patients with chin-on-chest deformity; however, the quality of the included studies limits the evidence.

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* Corresponding author. Department of Orthopedics, The University of Connecticut, 120 Dowling Way, Farmington, CT 06032, USA
E-mail address: hasingh@uchc.edu (H. Singh).

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Background

Chin-on-chest deformity is a rare and severely disabling condition characterized by kyphotic deformity in the cervicothoracic spine [1]. It is classically characterized by severe pain, often accompanied by myelopathy and radiculopathy [2,3]. Functionally, patients suffer from limitations in horizontal gaze, restriction of upright posture, swallowing dysfunction, aspiration risk, and social impairment [2,4–6]. Risk factors for chin-on-chest deformity include ankylosing spondylitis (Fig. 1A), previous trauma, or iatrogenic following cervical laminectomy [7–9].

Another cause of chin-on-chest deformity is dropped head syndrome which is defined by severe weakness of the cervical paraspinal muscles resulting in progressive cervical kyphosis [10,11]. It has been associated with a number of different etiologies, including aging, neuromuscular disorders such as amyotrophic lateral sclerosis, myasthenia gravis, and polymyositis, or secondary to radiation therapy [10]. Dropped head syndrome can be distinguished from a rigid chin-on-chest deformity in its early stages by a patient's ability to extend the neck either actively or passively, which may progress to a rigid flexion deformity in the later stages [11].

Regardless of the etiology, a chin-on-chest deformity frequently requires surgical correction to alleviate symptoms and restore function [8,12]. The surgical aims are to restore cervical sagittal balance, maintain a comfortable horizontal gaze, alleviate compression of neurologic elements, ease chewing and swallowing, and improve the overall quality of life [6,13]. Surgical treatment of this deformity is extremely challenging and associated with a high rate of complications due to the extensive dissection required for correction, exposure to critical vascular and neurologic structures, and proximity to the trachea and esophagus [6,14]. Additionally, fused/ankylosed deformities may be impossible to correct with instrumentation and fusion alone. In these cases, osteotomies are often needed to correct sagittal and coronal spine imbalances [6,15]. Various osteotomies, including pedicle subtraction osteotomy (PSO), vertebral column resection osteotomy (VCRO) and posterior column extension osteotomy (PCEO) (Fig. 1B) such as Smith-Peterson osteotomy (SPO), have been described for use in these situations [16–19].

Given the challenges posed in the management of this pathology, a comprehensive review of the current literature surrounding the use of osteotomies for chin-on-chest deformities would be beneficial for treating surgeons. The purpose of this review is to identify the clinical and radiologic outcomes of PCEO, PSO, and VCRO in the treatment of adult chin-on-chest deformity.

Methods

A systematic review of the available literature was performed to identify the clinical and radiologic outcomes of PCEO, PSO, and VCRO in the treatment of adult chin-on-chest deformity based on the Preferred Reporting Items for Systematic Reviews and Meta- analyses (PRISMA) guidelines. The review protocol was registered to the PROSPERO (CRD42021285999).

We conducted a comprehensive literature search in biomedical databases including MEDLINE (via PubMed), Scopus (via Elsevier), Embase (via Elsevier), and Cochrane Library. A primary literature search in PubMed from 1/1/1990 to 3/31/2022 using a combination of text and Medical Subject Headings (MeSH) terms was performed. The searches were then performed in three additional databases, Scopus, Embase, and Cochrane Library to ensure a thorough review of the available literature. A detailed search strategy is available in the Appendix.

The combined searches produced 11205 references for screening. A total of 1477 duplicate citations were excluded, resulting in 9728 unique citations for review. An additional 893 articles were identified outside of the initial search by reviewing the references of included studies (Fig. 2).

Inclusion and exclusion criteria

Randomized controlled trials, non-randomized controlled trials, cohort studies, case-control studies, and case series were included. Additionally, only studies used patients aged 18 and older who underwent PCEO, PSO, and VCRO due to adult chin-on-chest deformity were included.

Animal studies, cadaveric studies, case reports, literature reviews, meta-analyses, technical notes, expert opinions, and editorial letters were all excluded.

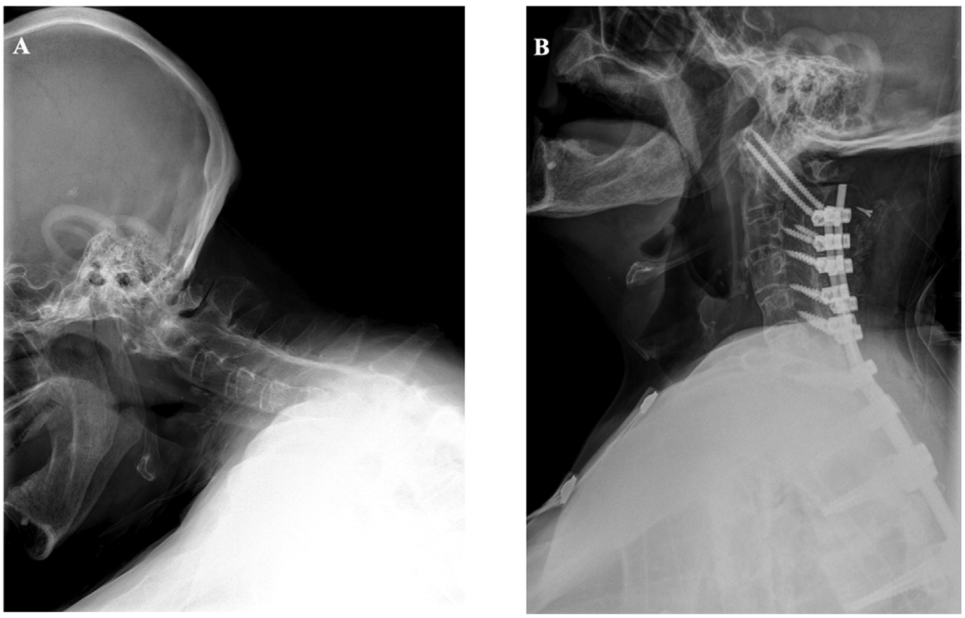

Fig. 1. (A–B): Pre-operative lateral cervical spine X-ray of 75-year-old male with chin-on-chest deformity due to ankylosing spondylitis (A). Post-operative X-ray of the same patient after a T1-T4 osteotomy with internal fixation (B).

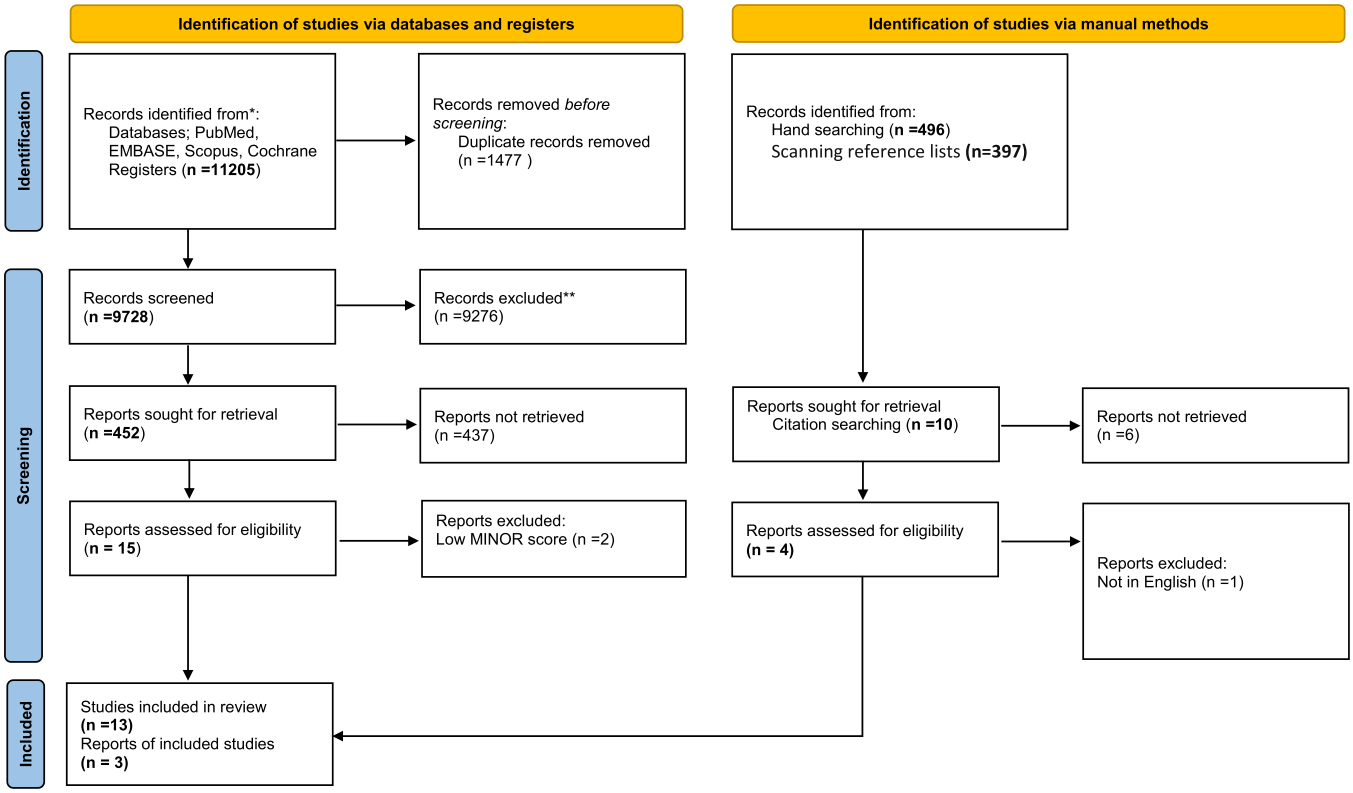

Fig. 2. PRISMA 2020 flow diagram for new systematic reviews which included searches of databases, registers and other sources.

Study selection and data extraction

The article selection process was done in two steps after the databases had been searched, titles and abstracts of studies had been uploaded into Mendeley via Critical Appraisal Skills Program Checklist including questions to help eliminate bias and make for a better systematic review.

First, two authors performed independent reviews of titles and abstracts in Mendeley using the predefined inclusion and exclusion criteria detailed above. The authors then reviewed the full text of the included manuscripts to determine relevancy. Any conflicts regarding inclusion were resolved by the third author. After assessing the eligibility of the 19 full-text studies, we agreed to include 16 studies in this systematic review.

A standardized electronic form was created for the extraction of data. This form contained the following data: name of the journal, title of study, authors, year of publication, study design, level of evidence, Methodological Index for Non-Randomized Studies (MINORS) score, statistical methods, potential conflicts of interest, location, demographics of the participants in the study, sample size, randomization, baseline data, interpretation, generalizability, description of the intervention, description of the control group or alternative intervention, results of the intervention and statistical methods.

Additionally, surgical data was collected such as technique, level of operation, method of anesthesia, patient positioning, blood loss, and operative time. Clinical outcomes recorded were visual analogue scale (VAS), neck disability index (NDI), Oswestry disability index (ODI), SF-36 PCS, and SF-12 PCS. Radiological parameters evaluated were mean preoperative and postoperative cervical kyphosis, mean radiologic correction, mean preoperative and postoperative chin-brow to vertical angle (CBVA), mean preoperative and postoperative cervical sagittal vertical axis (C-SVA). Intraoperative complications such as dural tear, pneumothorax, respiratory arrest, excessive blood loss, intraoperative pedicle or vertebral body fracture, and neuromonitoring changes were each included. Acute postoperative complications such as DVT, pulmonary embolism, dysphagia, temporary and persistent neurologic changes, neurologic deficit, superficial wound infection, deep wound infection and vertebral subluxation, revision surgery for subluxation were included. Finally, long-term postoperative complications such as pseudarthrosis, revision surgery due to pseudarthrosis, implant failure, and revision surgery due to implant failure were recorded.

Quality assessment

We assessed the methodological quality using the Methodological Index for Non-Randomized Studies (MINORS) guidelines which includes 12 items. Each item has two scores for a total of 24; less than 16 points indicates low quality, while more than 16 points indicates high quality [20]. The studies were assigned a level of evidence score according to the Oxford Centre for Evidence-Based Medicine [21].

Results

Our final analysis included 16 studies, only one of which was a prospective study that despite having a short follow-up period, was included due to its prospective multicenter design and standardized collection of detailed clinical and radiographic data [22]. All studies were assigned a level of evidence of four. With the exception of two studies that compared lower cervical osteotomy (LCO) and upper thoracic three-column osteotomy (UTO) [15], and one that compared SPO, PSO, and Anterior-Posterior Osteotomy (APO) [19], all other studies were non-comparative studies (Table 1).

Four studies reported results of PCEO including SPO [2,23–25], five studies reported outcomes of PSO [3,6,9,14,26], and five studies assessed outcomes of PSO and PCEO [8,10,13,19,27]. Two studies evaluated the outcomes of VCRO [15,22] (Table 3).

Table 1
Descriptive data of included studies.

Authors	Year	Study design	Level of evidence	MINOR score	Statistical methods
McMaster et al. (1997)	1997	Non-comparative retrospective case series	4	8	-
Willems et al. (2005)	2005	Non-comparative retrospective case series	4	10	-
Belanger et al. (2005)	2005	Non-comparative retrospective case series	4	9	-
Langeloo et al. (2006)	2006	Non-comparative retrospective case series	4	10	-
Tokala et al. (2007)	2007	Non-comparative retrospective case series	4	9	-
Samudrala et al. (2010)	2010	Non-comparative retrospective case series	4	10	-
Deviren et al. (2011)	2011	Non-comparative retrospective case series	4	8	-
Lee et al. (2012)	2012	Non-comparative retrospective case series	4	10	-
Theologis et al. (2014)	2014	**Comparative** retrospective cohort	4	15	Student t test Chi-square test
Kim et al. (2015)	2015	**Comparative** retrospective case-control	4	15	Student t test Kruskal-Wallis test
Smith et al. (2017)	2017	Non-comparative **prospective case series**	4	15	Student t test
Tobin et al. (2017)	2017	Non-comparative retrospective case series	4	8	-
Sabou et al. (2018)	2018	Non-comparative retrospective case series	4	10	Student t test
Shin et al. (2019)	2019	Non-comparative retrospective case series	4	8	-
Meng et al. (2020)	2020	Non-comparative retrospective case series	4	10	-
Verla et al. (2021)	2021	Non-comparative retrospective case series	4	7	Student t test

One study reported complications of both cervical and lumbar osteotomies in patients with ankylosing spondylitis [24]. Finally, one study presented a case series of patients undergoing PSO with computer guidance [14].

One study received a MINORS score of seven [10]. Four studies [3,9,14,23] were given a MINORS score of eight, two studies [2,6] were given a score of nine, and six studies [8,13,24–27] were given a score of ten. Two comparative studies [15,19] and one prospective study [22] received a MINORS score of fifteen (Table 1).

In all studies, metric data was expressed as the mean standard deviation. In five studies, it was compared using the Student‘s t-test [10,13,15,19,22] (Table 1).

Demographic data

Nine studies were performed in United States [2,3,9,10,14,15,19,22,27], five were performed in Europe [6,13,23–25], and two were performed in Asia [8,26]. A total of 288 patients were included, with sample sizes ranging from four to sixty-one per study. The average age ranged between 48 and 70 years. The mean length of follow-up was between three and fifty-four months. Etiologies of chin-on-chest deformity included post-laminectomy, post-traumatic, post-radiation therapy, and most commonly ankylosing spondylitis (Table 2). In all sixteen studies, the most common surgical indication was impaired horizontal gaze [2,3,6,10,23,25–27].

Surgical data

The type of anesthesia used during the procedure was not specified in six studies [3,10,13,19,22,26]. In one study, 26 patients were operated on while under local anesthesia [2]. General anesthesia was used in the remaining studies [6,8,9,14,15,23–25,27]. Of the 288 patients, 107 underwent PCEO, 108 underwent PSO, and 33 underwent VCRO (Table 3).

The most common osteotomy level in fifteen of the studies was C7/T1, while in one study, the level of osteotomy was not specified [19]. In most studies, stabilization was achieved through internal fixation with postoperative external halo fixation. Surgical time was reported in nine studies and ranged from 194 to 450 minutes [3,6,8,13,15,19,22,25,26]. Twelve studies reported blood loss ranging from 232 ml to 2400 ml [3,6,8,10,13,15,19,22,24–27] (Table 3). The average hospital stay after surgery was reported in 5 studies and ranged from 7.5 to 16.1 days [3,10,15,22,27].

Radiological parameters

Each study used a different set of radiographic parameters to determine outcomes. Cervical kyphosis and C2–C7 SVA were the two most commonly reported sagittal plane parameters. During the follow-up period, loss of cervical alignment correction was observed in two of the studies, with a mean of between 2.6 and 6 degrees [2,23]. Six studies recorded mean CBVA pre- and postoperatively [3,6,13,25–27]. The range of the mean CBVA correction was 32.25 to 38 degrees [3,6,8,25,27]. Additionally, two studies compared preoperative and postoperative T1 slopes [15,22] (Table 4).

The studies which reviewed only PCEO demonstrated a mean radiographic correction of cervical kyphosis of between 24.3 and 54 degrees [2,23–25]. For studies using only PSO, the mean radiographic correction of cervical kyphosis ranged from 20 to 57 degrees [3,6,9,14]. Kim et al. found that when SPO, PSO, anterior osteotomy (ATO), and ATO+SPO were compared, the PSO group had the highest correction degree (mean 44.8 degrees) [19].

Four studies reported mean correction C2–C7 SVA ranged from 24.5 mm to 45 mm[3,9,19,26]. Theologis et al. revealed a greater decrease in the mean T1 slope in the UTO group compared to the LCO group [15]. Smith et al. also observed a decrease in mean T1 slope from 48.1 to 40.5 with PSO and VCRO [22] (Table 4).

Clinical outcomes

Five studies reported preoperative and postoperative VAS scores for neck pain [3,13,15,26,27]. The range of preoperative and postoperative VAS scores was 5.5–8.6 to 1.7–4.91, respectively. Four studies used NDI [3,13,15,19]. The range of preoperative and postoperative NDI was 34.2–65.5 to 22.1–51.3, respectively. Theologis et al. also used ODI to compare LCO and UTO groups, finding that UTO reduced ODI score more than LCO [15]. Two studies demonstrated an increase in SF-36 scores following PSO surgery [3,26]. The preoperative and postoperative SF-36 PCS ranges from 20.7 to 30.2 and 35.8 to 53.3, respectively. One study used SF-12 scores to assess the preoperative and postoperative quality of life [15] (Table 5).

Complications

Intraoperative

No intraoperative pedicle fracture was reported in any of the included studies following cervical osteotomies. One study reported a case

Table 2
Demographic data.

Author	Number of cases	F/M	Mean ages (years)	Etiology	Mean FU (months)
McMaster et al. (1997)	15	2/13	48	Ankylosing spondylitis:15	18
Willems et al. (2005)	22	-	-	Ankylosing spondylitis:22	>12
Belanger et al. (2005)	26	1/25	51	Ankylosing spondylitis:26	54
Langeloo et al. (2006)	16	2/14	50.8	Ankylosing spondylitis:16	>12
Tokala et al. (2007)	8	2/6	54	Ankylosing spondylitis:5 Psoriatic spondyloarthropath:3	24
Samudrala et al. (2010)	8	3/5	63	Postlaminectomy:4 Posttraumatic:2 Neurofibromatosis:2	15.3
Deviren et al. (2011)	11	4/7	70	Ankylosing spondylitis:1 Other:10	23
Lee et al. (2012)	7	2/5	52.6	Ankylosing spondylitis:5 Postinfectious:1 Idiopathic:1	16.5
Theologis et al. (2014)	48	LCO:4/11 UTO:20/13	61	Ankylosing spondylitis:9 Posttraumatic:6 Poslaminectomy:6 Proximal Junction Kyphosis:23 Other:4	LCO:21.6 UTO:22.2
Kim et al. (2015)	61	-	58.2	Ankylosing spondylitis:13 Posttraumatic:22 Postlaminectomy:8 Degenerative:15 Klippel-Feil:2 Muscular Dytropy:1	30
Smith et al. (2017)	23	16/7	62.3	-	>3
Tobin et al. (2017)	4	4/0	59	Spondyloarthropathy:1 Posttraumatic:2 Postlaminectomy:1	25.7
Sabou et al. (2018)	13	0/13	57.5	Ankylosing spondylitis:13	37.6
Shin et al. (2019)	12	7/5	61.1	-	-
Meng et al. (2020)	7	0/7	-	Ankylosing spondylitis:7	32.9
Verla et al. (2021)	7	1/6	69	Postradiation:7	-

F, female; M, male; Mean FU, mean follow-up.

Table 3
Surgical data.

Author	Technique	Surgery level	Anesthesia	Position	Surgical time (minutes)	Blood loss (ml)
McMaster et al. (1997)	PCEO:15	C7/T1:15	General	Prone:15	-	-
Willems et al. (2005)	PCEO:22	C6/C7	General	Sitting:22	-	702
Belanger et al. (2005)	PCEO:26	C5/C6: 1 C6/C7: 2 C7/T1:23	Local	Sitting:26	-	-
Langeloo et al. (2006)	PCEO:16	C7:16	General	Prone:5 Sitting:11	194	940
Tokala et al. (2007)	PSO:8	C7:8	General	Prone:8	300	2400
Samudrala et al. (2010)	PSO:7 SPO:1	C7:1 T1:5 C6/T1:1 T2/T3:1	General	Prone:8	-	800
Deviren et al. (2011)	PSO:11	C7:9 C6/C7:1 T1:1	-	-	258	1100
Lee et al. (2012)	PSO:5 APO:2	C3/C6:1 C6:1 C7:5	General	Prone:6 Sitting:1	312	548
Theologis et al. (2014)	LCO: 12-PSO, 3-VCR UTO:12-PSO, 21-VCR	C7:15 T1-4, T2-7, T3-9, T4-9, T5-4	General	-	LCO: 272 UTO:333	LCO:1530 UTO:1160
Kim et al. (2015)	SPO:13 PSO:10 ATO: 16 ATO+SPO:22	-	-	-	SPO:230.7 PSO:344.7 ATO:206.3 ATO+SPO:321.5	SPO:232 PSO:712.5 ATO:183 ATO+SPO: 325
Smith et al. (2017)	PSO:14 VCR:9	PSO: C7-1, T1-9, T2-3, T3-1 VCR: T2-4, T3-4, T4-1	-	-	348	1300
Tobin et al. (2017)	PSO:4	T1:3, T1/T2:1	General	Prone:4	-	-
Sabou et al. (2018)	SPO:10 PSO:3	C7/T1:13	-	-	450	1200
Shin et al. (2019)	PSO:12	C7:8 T1:4	General	Prone:12	-	-
Meng et al. (2020)	PSO:7	C7:7	-	-	255	1475
Verla et al. (2021)	PSO:3 PCEO:4	C7:3-PSO C+T:4 EO	-	-	-	917

PCEO, posterior column extension osteotomy; PSO, pedicle subtraction osteotomy; SPO, Smith-Peterson osteotomy; VCR, vertebral column resection osteotomy; ATO, anterior osteotomy; APO, anterior-posterior osteotomy; LCO, lower cervical osteotomy; UTO, upper thoracic osteotomy.

of dural tear during the osteotomy [15]. The incidence of dural tear was 0.34%.

One study reported a case of pneumothorax and two cases of vertebral body fracture following UTO [15]. The incidence of pneumothorax and vertebral body fracture were 0.34% and 0.69%, respectively.

In one study, only one case of screw misalignment was reported [19]. The incidence of screw misalignment was 0.34%.

Acute postoperative

Theologis et al. reported two patients with DVT during the early postoperative period in their series [15]. The incidence of DVT was 0.69%. In one study, a patient had pulmonary embolism without DVT [22]. Following corrective osteotomy for chin-on-chest deformity, the incidence of pulmonary embolism was 0.34%.

In ten studies, neurologic complications were reported in 46 patients [2,13–15,22–27]. The incidence of neurologic complication was 15.9%. Thirty-eight of them (82.6%) experienced temporary neurologic symptoms. However, the symptoms of 8 patients (17.3%) persisted. The most common complications were new upper extremity paranesthesia and hand numbness through the C8 dermatomal distribution which were seen following C7 and T1 osteotomies [6,13,15,23–25]. The most severe complication was quadriparesis (0.34%), quadriplegia (0.34%), and C6 spinal cord injury (0.69%) [2,23–25].

In five studies, 17 patients with dysphagia were reported [2,3,10,22,23]. The rate of dysphagia was 5.9%. Five studies reported

Table 4
Radiological parameters.

Author	Mean preop. cervical kyphosis or lordosis	Mean postop. cervical lordosis	Mean radio-graphic correction	Mean preop. Chin-brow to vertical angle	Mean postop. Chin-brow to vertical angle	Mean correction Chin-brow to vertical angle	Mean preop. SVA (mm)	Mean postop. SVA (mm)	Mean correction SVA (mm)	Mean preop. T1 slope	Mean postop. T1 slope
McMaster et al. (1997)	C7/T1 Kyphosis:23	C7/T1: 31	54	-	-	-	-	-	-	-	-
Willems et al. (2005)	-	-	24.3	-	-	-	-	-	-	-	-
Belanger et al. (2005)	C2/C6 Lordosis:12	-	38	-	-	-	-	-	-	-	-
Langeloo et al. (2006)	-	33.5	-	42	5	38	-	-	-	-	-
Tokala et al. (2007)	C7/T1 Kyphosis:15	41	57	41	6	34.7	-	-	-	-	-
Samudrala et al. (2010)	C7/T1 Kyphosis:38.67	5.87	35.63	36.25	4.13	32.25	-	-	-	-	-
Deviren et al. (2011)	C2/T1:25.8	C2/T1:24.1	49.9	39.9	3.2	36.7	C2/C7:79	C2/C7:34	C2/C7:45	-	-
Lee et al. (2012)	-	-	39.7	-	-	37.1	-	-	-	-	-
Theologis et al. (2014)	C2/T1 Kyphosis LCO:20.6 UTO:6	C2/T1 LCO:23.8 UTO:17.2	-	-	-	-	C2/C7 SVA LCO:8.1 UTO:6	C2/C7 SVA LCO:4.3 UTO:4.2	-	LCO:42.1 UTO:50.7	LCO:385 UTO:35.3
Kim et al. (2015)	SPO:13.8 PSO:20.3 ATO:13.9 ATO+SPO:30.9	SPO: -5.6 PSO: -16.2 ATO: -4.9 ATO+SPO: -4.8	SPO:19.4 PSO:44.8 ATO:22.4 ATO+SPO:32.5	-	-	-	-	-	SPO:35 PSO:28 ATO:13 ATO+SPO:36	-	-
Smith et al. (2017)	C2/C7 Lordosis:2.8	C2/C7:12.7	-	-	-	-	C2/C7:65.7 C7/S1:0.4	C2/C7:43.6 C7/S1:30.5	-	48.1	40.5
Tobin et al. (2017)	C2/T1:-3.25	C2/T1:6.25	20	-	-	-	C2/C7:58	C2/C7:33.5	24.5	-	-
Sabou et al. (2018)	C2/C7:4.92	C2/C7:25.53	-	54	7	-	C2/C7:93.66	C2/C7: 39.18	-	-	-
Shin et al. (2019)	C2/T1 Kyphosis: 30.2	C2/T1:19.2	49.3	-	-	-	C2/C7:84	C2/C7: 32	-	-	-
Meng et al. (2020)	C2/T1 Kyphosis:26.3	C2/T1:5.4	-	43.7	0.9	-	C2/T1: 66	C2/T1: 30	37	-	-
Verla et al. (2021)	C2/C7 Lordosis:-21.65	C2/C7:-0.03	-	-	-	-	69.6	30.4	-	-	-

SVA, Sagittal Vertical Axis; PCEO, posterior column extension osteotomy; PSO, pedicle subtraction osteotomy; SPO, Smith-Peterson osteotomy; VCR, vertebral column resection osteotomy; ATO, anterior osteotomy; APO, anterior-posterior osteotomy; LCO, lower cervical osteotomy; UTO, upper thoracic osteotomy.

Table 5
Clinical outcomes.

Author	Technique	Mean preop VAS	Mean postop VAS	Mean preop NDI	Mean postop NDI	Mean preop ODI	Mean postop ODI	Mean preop SF-36 PCS	Mean postop SF-36 PCS	Mean preop SF-12 PCS	Mean postop SF-12 PCS
McMaster et al. (1997)	PCEO:15	-	-	-	-	-	-	-	-	-	-
Willems et al. (2005)	PCEO:22	-	-	-	-	-	-	-	-	-	-
Belanger et al. (2005)	PCEO:26	-	-	-	-	-	-	-	-	-	-
Langeloo et al. (2006)	PCEO:16	-	-	-	-	-	-	-	-	-	-
Tokala et al. (2007)	PSO:8	-	-	-	-	-	-	-	-	-	-
Samudrala et al. (2010)	PSO:7 SPO:1	7.79	4.91	-	-	-	-	-	-	-	-
Deviren et al. (2011)	PSO:11	8.1	3.9	51.1	38.6	-	-	30.2	35.8	-	-
Lee et al. (2012)	PSO:5 APO:2	-	-	-	-	-	-	-	-	-	-
Theologis et al. (2014)	PSO:12, VCR:3 PSO:12, VCR:21	LCO:6.5 UTO:5.5	LCO:4.4 UTO:4.1	LCO:54.5 UTO:57.8	LCO:42.6 UTO:51.3	LCO:57 UTO:57	LCO:54 UTO:47	-	-	LCO:30.4 UTO:37.8	LCO:32.8 UTO:39.4
Kim et al. (2015)	SPO:13 PSO:10 ATO: 16	-	-	34.2	22.1	-	-	-	-	-	-
Smith et al. (2017)	PSO:14 VCR:9	-	-	-	-	-	-	-	-	-	-
Tobin et al. (2017)	PSO:4	-	-	-	-	-	-	-	-	-	-
Sabou et al. (2018)	SPO:10 PSO:3	8	1.7	65.54	22.9	-	-	-	-	-	-
Shin et al. (2019)	PSO:12	-	-	-	-	-	-	-	-	-	-
Meng et al. (2020)	PSO:7	8.6	1.7	-	-	-	-	20.7	53.3	-	-
Verla et al. (2021)	PCEO:4 PSO:3	-		-	-	-	-	-	-	-	-

PCEO, posterior column extension osteotomy; PSO, pedicle subtraction osteotomy; SPO, Smith-Peterson osteotomy; VCR, vertebral column resection osteotomy; ATO, anterior osteotomy; APO, anterior-posterior osteotomy; LCO, lower cervical osteotomy; UTO, upper thoracic osteotomy; VAS, Visual Analogue Scale; NDI, Neck Disability Index; ODI, Oswestry Disability Index; SF-36, Short Form Health Survey-36, SF-12, Short Form Health Survey-12; PCS, physical component score.

superficial wound infection in 10 patients [13,15,22,24,25]. The incidence of superficial wound infection was 3.47%. In addition, seven patients who had deep wound infection underwent wound debridement in four studies [6,15,19,26]. The incidence of deep wound infection was 2.43%.

In four studies [2,6,19,23], during the postoperative period, ten patients were determined to have a subluxation defined as a forward displacement of the upper vertebra on the lower vertebral body greater than 5 mm at the site of the osteotomy [2,23]. The incidence of subluxation was 3.47%. In patients who developed subluxation, four (40%) required revision surgery.

Late postoperative

Seven patients were reported to have pseudarthrosis in three studies [2,15,23]. The incidence of pseudarthrosis was 2.43% and all of them required return to the OR for revision surgery. Implant failure was observed in a patient who underwent revision surgery [3].

Discussion

Corrective osteotomies are a type of treatment option for chin-on-chest deformity [8,9,13,19,26]. Due to the requirement for multiple extensive approaches, exposure to critical vascular and neural structures, and working close to the trachea and esophagus, surgical treatment of this deformity is extremely difficult and associated with a high rate of complications [14,15,26]. Although the studies in this systematic review clarify that the corrective osteotomies provide improved health-related quality of life in patients with chin-on-chest deformity [13,15,26], we were unable to find sufficient studies with a high level of evidence demonstrating which osteotomy would be most effective in treating chin-on-chest deformity.

The majority of studies in this systematic review evaluated the clinical and radiological outcomes of PCEO or PSO for the correction of chin-on-chest deformity [2,3,6,8–10,13,14,23–27]. We were able to conclude from this systematic review that VCRO is used infrequently for the treatment of chin-on-chest deformity.

We found that various types of radiologic parameters were used in the included studies to assess the success of the correction. Although all studies reported cervical kyphosis correction angle, the level of the measurement varied among the studies. Meng et al. used CBVA measurements as the basis for kyphosis correction and treatment outcome evaluation because restoring the visual field has become the primary goal of cervical osteotomies [26]. Samudra et al. noticed a significant correlation between the angle of correction achieved through PSO and the correction in CBVA [27]. Tobin et al. assessed horizontal gaze impairment using the C2 slope, defined as the angle between the horizontal line and the inferior endplate of C2 [9]. Furthermore, the studies included in this review described several techniques to measure cervical sagittal balance, one of which is the C2–7 SVA measured as the deviation of a C2 plumb line from the center of C2 to the posterior superior endplate of C7 [3,9,13–15,22].

In this systematic review, we recognized that arm hypoesthesia was the most common neurologic complication caused by compression of the C8 nerve roots in the intervertebral foramen due to insufficient bone removal from the pedicles above and below the osteotomy and stretch or shifting of the spinal cord and nerve root at this level during correction [6,13,15,23–25]. Although dysesthesias usually resolve on their own, intrinsic hand-muscle weakness can persist [14,23,25,27].

The other cause of neurological complications is instability and subluxation, which can occur as a result of disruption of the stabilizers of the spine, including the posterior longitudinal ligament [2,6,23]. Belanger et al. revealed anterior subluxation in five of twenty-six patients. All of the subluxations occurred in patients who had not undergone internal fixation [2]. Langeloo et al. reported that internal fixation was consistently performed following extension correction and that clinical correction was satisfactory, with no translation or loss of correction [25]. In contrast to Belanger and Langeloo et al., Tokala et al. emphasized that subluxation can be caused not only by a lack of instrumentation but also by the PCEO surgical technique, which results in stretching of anterior structures such as the trachea and esophagus and rupture of the posterior longitudinal ligament [6]. They performed PSO on eight patients and concluded that PSO is superior to PCEO in terms of safety and efficacy because no anterior structures were stretched and rapid fusion can occur across the two cancellous surfaces with minimal correction loss [6]. Following the study of Tokala et al., Deviren et al. described a PSO with a wedge component that cleaves the vertebral body, resulting in a greater bone-on-bone load-bearing interface than an PCEO [3]. They reported that PSO not only provides a significant load-bearing surface area but also a more controlled closure than PCEO due to the absence of a sudden osteoclastic fracture [3]. Although they reported excellent radiologic results and no pseudarthrosis in eleven patients with severe cervicothoracic kyphotic deformities who underwent PSO, the majority of patients in their series (91%) did not have ankylosing spondylitis [3].

The other controversial issue in this systematic review is the level of osteotomy. Tobin et al. claimed that as a PSO is performed lower in the spine and through a larger T-1 pedicle, it allows for more sagittal correction [9]. Furthermore, they reported the T-1 PSO is superior to the C-7 PSO in terms of decreased risks of injury to the vertebral artery and clinically significant nerve root injury [9]. Theologis et al. assessed the level of osteotomy in 48 patients who underwent PSO or VCRO surgery [15]. In a series of patients with rigid cervicothoracic deformity, they compared the outcomes of the LCO and UTO, reporting that LCO provided greater cervical SVA correction and were shorter operations, but they were associated with more complications and longer hospital and ICU stays than UTO [15]. Smith et al. reported only one osteotomy in the cervical spine in their prospective case series [22]. They mentioned that because of the potentially increased safety of performing the PSO or VCR at more caudal levels, several of the study's co-authors have gradually shifted to performing osteotomy at the T2 and T3 vertebral levels when possible [22].

Our study has several limitations. First, this review included only studies with a level of evidence of 4, limited by a small sample size with just one prospective study. Second, demographic data, etiology of deformity, and surgical technique varied greatly between these studies. Additionally, the quality of the studies included in this review was low, scoring between 7 and 15 on the MINORS scale. Many of these scores were related to study design, lack of prospective data, and heterogeneity in surgical technique and patient population. This is likely due to the rare nature of these deformities, specific patient population, and lack of multicenter studies.

Conclusion

Various osteotomy options, including PCEO, PSO, and VCRO, are available to correct chin-on-chest deformities. Although corrective osteotomies provide satisfactory results in patients with chin-on-chest deformity, the quality of the included studies made this evidence limited. Nevertheless, this review provides a foundation for future research on corrective osteotomies in patients with chin-on-chest deformity.

Declaration of competing interests

One or more of the authors declare financial or professional relationships on ICMJE-NASSJ disclosure forms.

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Supplementary materials

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