International Journal of Innovative Research in Medical Science (IJIRMS)
Volume 07, Issue 12, December 2022,
https://doi.org/10.23958/ijirms/vol07-i12/1585

Review article

Haglund Syndrome: A Systematic Review of the Current Possible Management

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Received 24 November 2022; Accepted 14 December 2022; Published 19 December 2022

Abstract

Background: Haglund's syndrome is considered a triad of posterosuperior prominence of the calcaneus or Haglund deformity, which is also known as retrocalcaneal bursitis and insertional Achilles tendinopathy. However, many cases are resolved depending on the conservative treatment only, in patients with persistent signs and symptoms after six months of conservative management, surgery is required. In this review, we aimed to systematize the possible management of Haglund's syndrome among studies published after the 2015. ***Methodology:*** This is a systematic review of the literature which was conducted following Cochrane Handbook of Systematic Review of Intervention and Preferred Reporting Items for a Systematic Reviews and Meta-Analyses (PRISMA) for selection of the studies. Data were extracted and recorded for a stepwise analysis from each study. Statistical analysis was conducted using IBM SPSS version 260 (IBM Corp., Armonk, N.Y., USA). ***Results:*** Through the search of literature, a total of 1474 studies were identified for initial screening. After removal of 1191 duplicated studies, preliminary evaluation was conducted among 283 papers which were available for the title and abstract screening. One- hundred- twenty-one studies (121) were excluded because being research focused on pathologies not related to Haglund' deformity while 91 studies were excluded because reporting data on surgical management of other hindfoot condition. Therefore, 71 studies were available for full-text analysis and after application of exclusion criteria, 12 studies were available for final evaluation. ***Conclusion:*** Surgical correction of Haglund's syndrome in case of failure of the conservative treatment provides overall good clinical outcomes with higher superiority for minimally invasive approaches which have similar good clinical outcomes in term of AOFAS, VISA, and VAS scores with less rate of complications and failure rate when compared to open surgeries.

Introduction

Haglund's syndrome is considered a triad of posterosuperior prominence of the calcaneus or Haglund deformity, which is also known as retrocalcaneal bursitis and insertional Achilles tendinopathy [1]. Haglund's syndrome is in usual occurred bilateral and affects the middle- aged individuals with a higher prevalence among women. Haglund syndrome is associated with heel pain, swelling, post-static dyskinesia and redness [2-6].

In most cases, this condition is diagnosed by combining the results of clinical evaluation with the imaging tests [7]. Radiography is considered the most commonly used test for confirmation of the diagnosis of Haglund's syndrome however, MRI is indicated in doubtful case [8,9]. Moreover, several recent studies showed that ultrasonography has 100 % specificity in diagnosis of retrocalcaneal bursitis however has 50 % sensitivity and it is even less sensitive in determining whether the superficial Achilles tendon bursa is involved [3,4,9,10]. The diagnosis of the Haglund syndrome aims to assess the cause of the symptoms including traumatic causes as stress fractures of the calcaneus or malunited fracture of the calcaneus, infection causes as tuberculosis of the calcaneus, neoplastic causes as osteochondroma of the calcaneus and the inflammatory causes including negative spondyloarthropathies [8-11].

The first line treatment of Haglund's syndrome is conservative including the following indications: rest, a change of habit considering impact sports, modification of the used shoe, and the use of non-steroidal anti-inflammatory drugs. However, many cases are resolved depending on the conservative treatment only, in patients with persistent signs and symptoms after six months of conservative management, surgery is required [12-15].

In the recent decades, surgical management of the Haglund's syndrome, mainly because of the better understanding of the pathophysiology of this condition. There has been recent discussion considering the best surgical approaches, including minimally invasive surgery, endoscopy, and techniques for repairing the tendon to the bone with suture anchors. Percutaneous screws with reliable and stable fixation are available as a modern technique that allow recovery of the functional consideration with less injury in the tissue because of the smaller portals [12,15-20]. However, up to the research's knowledge, there is no study that aimed to compare between different approaches in management of Haglund's syndrome. Thus, in this review, we aimed to systematize the possible management of Haglund's syndrome among studies published after the 2015.

Methodology

This is a systematic review of the literature which was conducted following Cochrane Handbook of Systematic Review of Intervention [21] and Preferred Reporting Items for a Systematic Reviews and Meta-Analyses (PRISMA) [22] for selection of the studies (**Figure 1**). The systematic search was conducted among all studies published between 2015 and now and was conducted in the following databases: Web of Science, ScienceDirect, PubMed, Embase, Cochrane Central Register of controlled Trials (CENTRAL) and MEDLINE. The following Keywords were used for the research: "Haglund syndrome", "Haglund deformity",

"endoscopic calcaneoplasty", "calcaneal osteotomy", "calcaneal resection", "endoscopic decompression", "Achilles", "insertional Achilles tendinopathy", "retrocalcaneal decompression", "retrocalcaneal bursitis". The inclusion criteria of choosing studies used in this study included being original studies that reporting clinical and functional outcomes of patients who underwent different management approaches to correct Haglund's deformity and including at least 10 patients to be eligible for this study. Technical notes, ex-vivo, pre-clinical studies (either on animal or cadaveric models), abstracts, clinical studies without adequate quantitative or qualitative data and editorial commentaries were excluded from the analysis in this review.

The analysis of this review was selectively focused on patients who had any intervention used to management of symptomatic Haglund's syndrome. Data were extracted and recorded for a stepwise analysis from each study. Basic information of each study including setting of the study and date of publishment as well as population characteristics including number of the samples, male/ female percentages, age, technique used in management which is classified into open surgeries and minimally invasive management were extracted as well as follow-up duration. To assess the outcomes of the intervention, we assessed the functional and complications outcomes. Complication rate was assessed as a secondary outcome including type of the complication. Among the studies, three tools were used to assess the improvement after the surgery, including Ankle-hindfoot scale (AOFAS) which consists of three subscales: pain, function and alignment and includes a total of nine items. The minimum score is 0 points (indicating severe pain and impairment), the maximum score is 100 points (no impairment) [23], visual analog scale (VAS) (where 0 describes no pain,10 describes the most pain imaginable) [24], and the Victorian Institute of Sports Assessment-Achilles questionnaire (VISA-A) which is ranging between very poor, (score around 24) to excellent (a score of 90) [25]. Statistical analysis was conducted using IBM SPSS version 260 (IBM Corp., Armonk, N.Y., USA). Frequency of events and percentage were used to describe the categorical variables while continuous variables were reported as mean and standard deviation. T test and Pearson's test were used to assess the difference between open surgery and minimally invasive techniques considering the primary and secondary outcomes. A p value of 0.05 or less was considered statistically significant.

Results

Studies collection

Through the search of literature, a total of 1474 studies were identified for initial screening. After removal of 1191 duplicated studies, preliminary evaluation was conducted among 283 papers which were available for the title and abstract screening. One-hundred- twenty-one studies (121) were excluded because being research focused on pathologies not related to Haglund' deformity while 91 studies were excluded because reporting data on surgical management of other hindfoot condition. Therefore, 71 studies were available for full-text analysis and after application of exclusion criteria, 12 studies were available for final evaluation (**Figure 1**).

General characteristics of the studies

In this review, we included 12 studies [8,26-36] where only study was systematic review [29]. The pooled studies included 1542 patients with Haglund's symptom with mean age 41.98 years old (Standard deviation of 7.1 years, minimum and maximum age of 32.5 and 59.3 years old). Male/ female percent were not mentioned in two studies, in the other 10 studies, male represented 59 % (N=230) and females (41 %, N=161). Moreover, 43 % of affected foot was the left one (N=168) while 171 (57%) were at the right foot. Among the study, the mean follow up was 33.1 months (SD= 23.4, min =3.5, and max=86.5). Considering the used intervention, 11 studies included strategies of using open surgical procedures (73.3 %) while 4 studies included strategies of using minimally invasive methods (**Table 1**).

Management techniques and outcomes

1. Open surgeries

In this review, 10 studies tested the efficacy of different open surgeries [8,26-31,33-35] including central Achilles tendon splitting in three studies, double row suture ancho in two studies, dorsal closing wedge calcaneal osteotomy in two studies, resection of calcaneal tuberosity in two study and posterosuperior prominence resection (PPR) in one study.

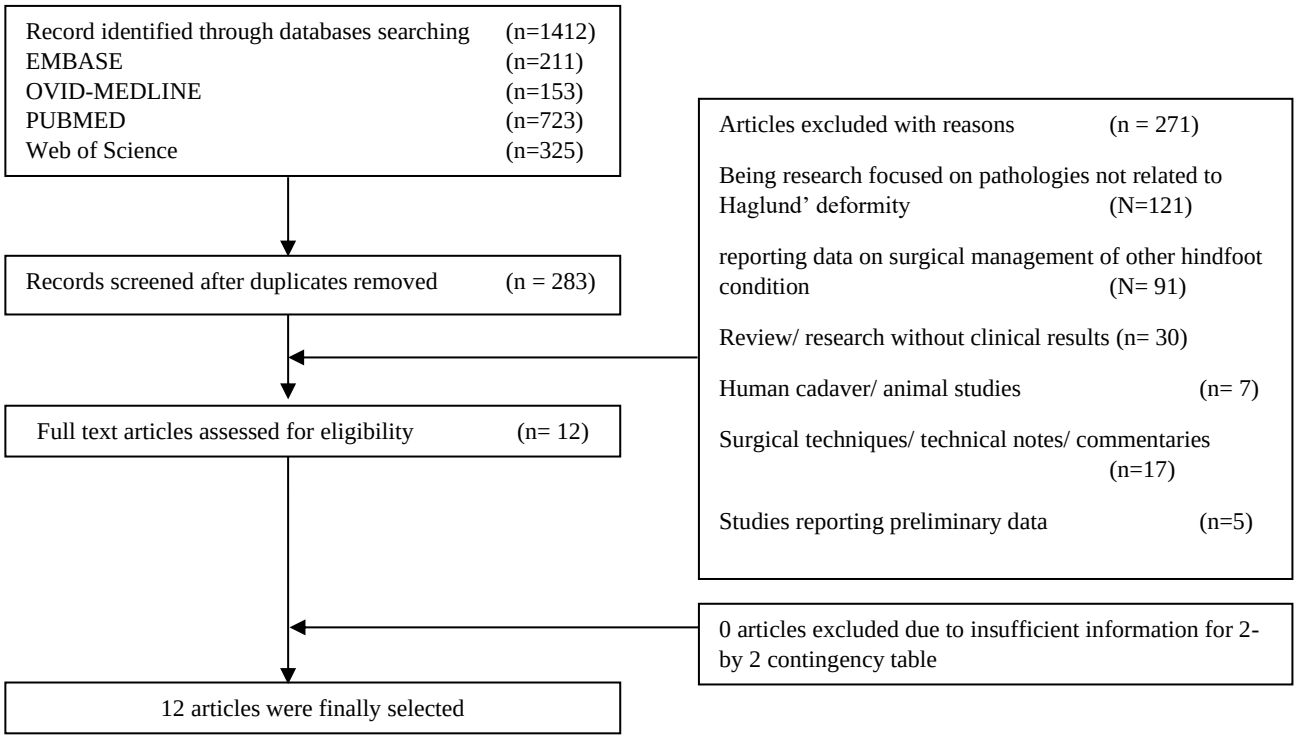


Figure 1: PRISMA flowchart for studies selection

Table 1: General characteristics of the pooled studies (N=12).

#	Authors	Year	Aim of the study	No. of total sample	Age	Gender		Side		Follow up period (in months)	Intervention	Type
						Male	Female	Left	Right			
1	Yasin Güler [26]	2019	Evaluate the clinical and radiographic results of central Achilles tendon splitting and double-row suture anchor technique	27	47	13	14	13	14	31	Central Achilles tendon splitting and double-row suture ancho	Open surgical procedures
2	Mattia Alessio-Mazzola [29]	2020	Report the clinical outcome following surgical correction of Haglund's deformity summarizing different surgical strategies and comparing clinical outcomes, failures, complications and return to activities of patients underwent open and endoscopic techniques.	650	47.1	NA	NA	NA	NA	37.4	NA	Open surgical procedures
				461	40.1					38.5	NA	Minimally invasive
3	Zhan Xia [8]	2017	Evaluate the surgical outcomes of Haglund's triad using a central tendon-splitting approach	22	59.3	10	12	11	11	15.1	Central tendon-splitting approach, with Achilles tendon partial detachment and debridement	Open surgical procedures
4	Zilu G [30]	2020	This study compared the clinical outcomes of (DCWCO) and (PPR) for the treatment of Haglund syndrome.	12	32.8	9	3	5	7	86.5	Dorsal closing wedge calcaneal osteotomy (DCWCO)	Open surgical procedures
				32	36.5	6	26	18	14	71.8	Posterosuperior prominence resection (PPR)	Open surgical procedures
5	Jun-Yi Liu [31]	2021	This trial is aimed to evaluate the functional and surgical outcomes of MMIR comparing with Achillon	44	38.2	36	8	26	18	24	Achillon	Open surgical procedures
				70	40.19	61	9	38	32	24	Modified minimally invasive repair (MMIR)	Minimally invasive
6	Kord et al [32]	2019	Introduce a modified technique for minimally invasive Achilles tendon (AT) rupture repair using gift box sutures.	24	38	17	7	11	13		Modified minimally invasive AT repair using two mini-incisions and gift box sutures.	Minimally invasive
7	Zhang [33]	2021	To analyses the imaging changes in bone marrow oedema of the calcaneal prominence, retrocalcaneal bursa and degenerative Achilles tendon after the surgical treatment of insertional Achilles tendinopathy (IAT).	30	44	21	9	10	20	36	Achilles tendon calcification with an open incision	Open surgical procedures

8	Natarajan [34]	2015	Determine the outcome of calcaneal tuberosity resection through lateral approach for refractory Haglund deformity.	40	44					13	Resection of the postero-superior calcaneal tuberosity through lateral approach	Open surgical procedures
9	Naseer A. Mir [35]	2018	Evaluate the clinical and functional outcome of osteotomy of the calcaneal tuberosity with debridement of the retrocalcaneal bursa and the Achilles tendon	25	38.7	9	16	11	14	13.5	Calcaneal tuberosity with debridement of the retrocalcaneal bursa	Open surgical procedures
10	Sergio1 [36]	2018	Evaluate the reliability and effectiveness of minimally invasive percutaneous surgical procedure for Achilles tendon decompression	21	40	15	6	10	11		Percutaneous Achilles tendon decompression	Minimally invasive
11	Jiang [27]	2016	Investigate whether double-row suture technique is a better option for the treatment of Haglund syndrome than single-row suture technique regarding the surgical outcomes.	32	51.4	11	21	15	17	3.5	Double-row suture technique	Open surgical procedures
12	Georgiannos et al [28]	2017	Dorsal wedge calcaneal osteotomy would be an effective and safe method for the treatment of athletes with insertional Achilles pathology unrelieved by nonoperative measures.	52	32.5	22	30			36	Dorsally based wedge calcaneal osteotomy	Open surgical procedures

Nine studies included data on AOFAS at pre-surgery and post-surgery and all studies used = open surgical procedures showed that there is a significant improvement of AOFAS with mean score at pre-surgery of 52.27 (SD=5.97, min = 39.3 and max= 59.5) and post-surgery of 88.62 (SD=8.7, min = 67.3 and max = 98.2) with mean increase of 36.3 (SD=7.42, min = 28 and max = 46.2). VISA-A was used in five studies and all studies showed a significant improvement in the VISA-A from mean VISA-A score at pre-operation of 50.1 (SD=13.5, Min= 35.7, Max=65.9) to mean VISA-A score at post-operative of 91.86 (SD=4.4, Min= 87.4, Max= 98.2). VAS score was reported in four studies and all these studies showed a significant improvement of pain score which decrease from mean VAS score of pre-operation of 7.8 (SD=0.82, Min= 7.23, and Max= 9) to VAS at post-operative of 2.02 (SD=1.533, Min= 0.433, and Max= 4.1). (**Table 2**).

Table 2: The outcomes of open surgical procedures in term of AOFAS. VISA-A and VAS scores

#	Authors	Intervention type	AOFAS			VISA-A			VAS score		
			Pre	Post	P	Pre	Post	P	Pre	Post	P
1	Yasin Güler [26]	Open surgical procedures	47	92	0.001	NA	NA	NA	9	2	0.001*
2	Mattia Alessio-Mazzola [29]	Open surgical procedures	53.9	87.1	0.001	60.1	89.2	0.001	7.3	1.56	0.001*
3	Zhan Xia [8]	Open surgical procedures	39.3	67.3	0.001	NA	NA	NA	7.8	4.1	0.001*
4	Zilu G [30]	Open surgical procedures	52	98.2	0.001	37.1	98.2	0.001	NA	NA	NA
		Open surgical procedures	50.7	93.4	0.001	35.7	94.3	0.001	NA	NA	NA
5	Jun-Yi Liu [31]	Open surgical procedures	NA	NA	NA	NA	NA	NA	NA	NA	NA
6	Zhang [33]	Open surgical procedures	50.6	94.27	0.001	NA	NA	NA	7.23	0.433	0.001*
7	Natarajan [34]	Open surgical procedures	58	86	0.001	NA	NA	NA	NA	NA	NA
8	Naseer A. Mir [35]	Open surgical procedures	54	86	0.001	NA	NA	NA	NA	NA	NA
9	Jiang [27]	Open surgical procedures	57.7	86.2	0.001	51.6	87.4	0.001	NA	NA	NA
10	Georgiannos et al [28]	Open surgical procedures	59.5	95.7	0.001	65.9	90.2	0.001	NA	NA	NA
* Significant in p value ≤ 0.05.											

2. Minimally invasive surgery

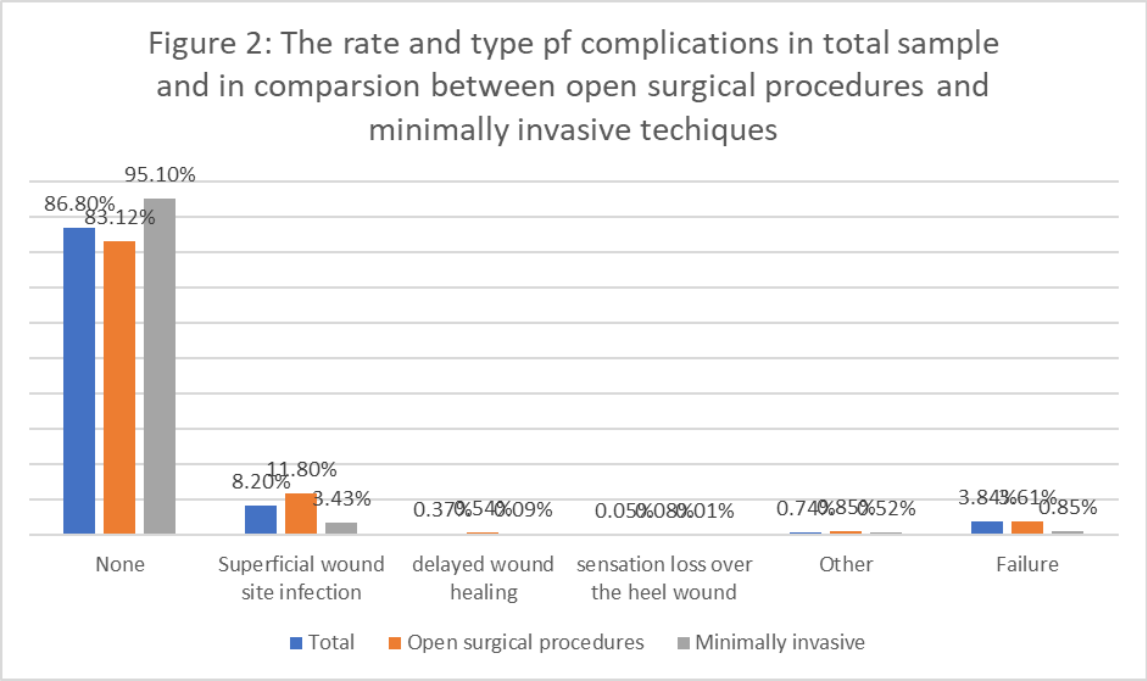
Four studies reported using of some of techniques known as minimally invasive surgery including modified minimally invasive repair (MMIR), modified minimally invasive AT repair using two mini-incisions and gift box sutures and percutaneous Achilles tendon decompression. AOFAS was also used among these studies

and showed a significant improvement from 53.57 (SD=4.86, Min= 50.47 and Max = 60.8) at pre-operative to 90.78 (SD=5.95, Min=83 and Max = 97.43). The data showed that there is no difference in the improvement of AOFAS between open surgical procedures and minimally invasive techniques (P=0.708). Patients with minimally invasive operation showed improvement in the VISA-A score from 52 at pre-operative to 89 at post-operative with no significant difference between these scores and scores of VISA-A in patients treated with open surgical procedures. VAS was reduced from 6.9 at pre-operation to 0.78 at post-operation in patients treated with minimally invasive surgery which is not significantly different in the VAS improvement in patients treated with open surgical procedures (P=0.243). The only variable that shows a significant difference between patients treated with minimally invasive and open surgical procedures is improvement of Fowler-Philip angle (FPA) which decreased with 25.8 points in minimally invasive- patients (from 79.57 to 53.76) compared with 20.9 points in patients with open surgical procedures from 54 to 33.1 (P=0.018). (**Table 3**).

Table 3: Comparison between minimally invasive techniques and open surgical procedures in the final outcomes.

	Type								
	Minimally invasive				**Open surgical procedures**				
	Mean	**Standard Deviation**	**Minimum**	**Maximum**	**Mean**	**Standard Deviation**	**Minimum**	**Maximum**	
AOFAS pre	53.57	4.86	50.47	60.80	52.27	5.98	39.30	59.50	0.708
AOFAS post	90.78	5.95	83.00	97.43	88.62	8.72	67.30	98.20	0.660
VISA pre	52	2	50	54	50	13	36	66	0.820
VISA post	89	3	86	92	92	4	87	98	0.417
VAS pre	6.90	.28	6.65	7.20	7.83	.82	7.23	9.00	0.123
VAS post	.78	.52	.19	1.15	2.02	1.53	.43	4.10	0.243
CAL pre	.	.	.	.	23.87	2.49	21.00	25.50	
CAL post	.	.	.	.	23.43	2.98	20.00	25.40	
FPA pre	79.57	.	79.57	79.57	54.00	1.00	53.00	55.00	0.002
FPA post	53.76	.	53.76	53.76	33.10	2.44	31.40	35.90	0.018

In figure 2, we showed the incidence of complications among total patients, those with open surgical procedures and those with minimally. In general, 86.8 % of the patients showed no complications while superficial wound/ site infection was the main complications that occurred in 8.2 % of the patients (173) while failure rate as reported in three studies was 3.84 %. In comparison of the two groups, we found a significant lower incidence of complications among those treated with minimally invasive technique (4.9 %) compared with 16.88 % among those treated with open operation procedures where superficial infection reported in 11.8 % of the open operation group compared with 3.43 % in the other group. Failure rate of both procedures was 3.61 % in open operation group compared with 0.85 % in minimally invasive procedures (**Figure 2**).



3. Conservative management

Conservative management includes in general reassessment of shoe of the patients and heel pads or heel lifts in the cases of arched feet. Casting may be necessary for reduction of the pain while ice bag may be necessary to reducing the impact of swelling. The using of anti-inflammatory medication, stretching exercises and physiotherapy may have an effect on relieving of tension from the calcaneal tendon. In this review, no study investigated the impact of conservative management however, all studies included patients who conservative management failed with them.

Discussion

The main finding of this systematic review is that surgical correlation of Haglund's symptom is associated with good clinical outcomes and is related with low incidence of complications. The

analysis of the results pooled for all types of interventions showed that there is not significant difference between open surgeries and minimally invasive approaches considering the postoperative AOFAS score, VAS score and VISA score however, minimally invasive approaches showed lower complications and failure rates. Comparative studies by Leitze et al [37] and Mansour et al., [38] showed similar results considering no significant difference of clinical outcomes scores, time to return to normal or physical activities however, these studies did not report significant difference between both approaches considering failure rate or rate of complications. However, these both studies observed numerically better clinical scores and lower of complications in endoscopic techniques without significant difference. On the other hand, another systematic review conducted by Mattia Alessio Mazzola showed that endoscopic approach has significantly better postoperative AOFAS scores than open surgeries however, it reported similar results considering the superiority of endoscopic approaches considering complications rate and failure rate [29].

The result of this review supports that minimally invasive operative treatment as an adequate option for this syndrome as it has the same clinical outcomes with less complication and failure rate. Most of the surgical treatment options for Haglund syndrome could be performed with endoscopic or with minimally invasive approaches. These results is in agreement with the results of previous studies [4,5,39-41,6,8-10,12,14-16]. Most of the minimally invasive approaches concentrate on peritendinous tissues to eliminate neovascularization and its accompanying nerves which is the main cause of pain and disease progression. Therefore, minimally invasive approaches address pain as main outcome while open techniques aim to treat of the degenerated tendon tissue. However, inadequate bone resection in open surgeries could be associated with recurrence of the symptoms with other post-surgical complications including scar formation, nerve entrapment, nonunion of the calcaneal osteotomy and weakening or rupture of the Achilles tendon [42].

Therefore, this study confirms that conservative management including heel elevations, program with eccentric exercises, orthosis prepare ad hoc, avoiding exercise that involve dorsal flexion and physical therapy should be the first line management of Haglund's symptom [43]. However, in case of failure of conservative treatments, surgical management should be considered where endoscopic or minimally invasive approaches have a higher advantageous over open surgery considering having the same clinical outcomes with lower complications and failure rates. Endoscopic and minimally invasive approaches have an extra advantage reported by other studies of shorten of the time of post-operative recovery and to return to normal life [44]. However, endoscopic approach is considered a challenging technique where all the possible pitfalls should be known to avoid them during the surgery. One of the important risks including not completely removing of the exuberant posterosuperior bone portion especially in its more lateral and lateral portions.

Conclusion

Surgical correction of Haglund's syndrome in case of failure of the conservative treatment provides overall good clinical outcomes with higher superiority for minimally invasive approaches which have similar good clinical outcomes in term of AOFAS, VISA, and VAS scores with less rate of complications and failure rate when compared to open surgeries.

Conflicts of Interest

The authors declare that there is no conflict of interest regarding the publication of this paper.

Funding Statement

No funding and no financial support

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