ISSN: 2158-284X Volume 11, Number 7, July 2020

Scientific Research Publishing

International Journal of Clinical Medicine

ISSN: 2158-284X
9 772158 284007 07

https://www.scirp.org/journal/ijcm

Journal Editorial Board

ISSN: 2158-284X (Print) ISSN: 2158-2882 (Online)
https://www.scirp.org/journal/ijcm

International Journal of Clinical Medicine, 2020, 11, 439-464
https://www.scirp.org/journal/ijcm
ISSN Online: 2158-2882
ISSN Print: 2158-284X

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Volume 11 Number 7 July 2020

International Journal of Clinical Medicine (IJCM)
Journal Information

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International Journal of Clinical Medicine, 2020, 11, 439-453
https://www.scirp.org/journal/ijcm
ISSN Online: 2158-2882
ISSN Print: 2158-284X

Syncope Beginning in People Over 50 Years Old—Experience in 52 Cases

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How to cite this paper: Pedro, J.-C., Maximiliano, A., Teresa, V.M., Yaninna, C., Tomás, P.J., Sebastián, B. and Francisco, T. (2020) Syncope Beginning in People Over 50 Years Old—Experience in 52 Cases. *International Journal of Clinical Medicine*, **11**, 439-453.
https://doi.org/10.4236/ijcm.2020.117037

Received: May 23, 2020
Accepted: July 12, 2020
Published: July 15, 2020

Abstract

It is not common to start suffering from syncopes after age 50. They are mainly male patients who present causes other than vasovagal syncope, which predominates at an early age. Orthostatic hypotension is the predominant causal factor, which is attributed in many cases to advanced age, metabolic, cardiovascular or neurological diseases, to failure of baroreflexes, all of the above may be associated with the use of hypotensive drugs alone or in combination with psychotropic drugs. Furthermore, causes such as carotid sinus syncope, postprandial syncope and situational syncope become more frequent. Therefore, as people age, they present a favorable pathological terrain for the production of syncope. The older you are, the more likely you are to start with syncope. Finding the definitive diagnosis for their syncopes can be difficult, given the multiplicity of interacting factors. Their study is more exhaustive and requires a good anamnesis, knowing the drugs used by the patient, concomitant diseases and careful surveillance to get closer to the diagnosis.

Keywords

Vasovagal Syncope, Fainting, Dysautonomia, Older Adults

1. Introduction

Syncope and/or lipothymia are common at all ages. However, going over 50 and just starting to present them is unusual. The first peak is during adolescence at age 15 [1]. Its frequency is usually twice as high in women as in men [2] [3]. Neurally mediated reflex syncope (vasovagal syncope (VVS)) predominates as the first cause of these [2] [3] [4].

There is another peak, somewhat smaller after 65 years [4] [5] [6]. This group is special, since other causes for syncope appear and it is more complex to find a single cause, because there may be more than one, in the same person. 40% - 60% of cases, could be multicausal [7] [8] [9].

We show our results, in terms of sex, age, associated factors, probable causes, and follow-up, in 52 patients who started their syncopes at age 50 or later. They were sent to us after a cardiac cause was ruled out and there were doubts about the cause of the condition. They were studied with head up tilt (HUT) from 2012 to 2018 and followed during their evolution, between 15 months and 7 years (Mean age: 4.5 years).

2. Material and Methods

Pre-Tilt test study and inclusion criteria

This study included retrospective data analysis of 212 patients studied with HUT (60% female), conducted between 2012 and 2018. The mean age of these patients was 31.8 years (range: 6 - 89 years).

Only 52 subjects (25%) who consulted for syncope or presyncope, beginning at 50 years or older were included. None had a history of prior syncope.

To rule out a cardiac or other causes, subjects were screened with a medical history, physical examination, an evaluation by a cardiologist and examinations: electrocardiogram (12 lead), echocardiogram, heart rate holter, and sometimes an electrophysiological study. If this review is negative or doubtful, the patient is sent for our neurological evaluation and HUT.

If a cardiac cause was proven the patient was not included. The same if the patient was epileptic or suffered from pseudosyncopes. 80 volunteers of the same age and sex served as controls.

Subjects were recruited from patients of the Militar Hospital from Santiago.

3. Tilt Test Exam Conditions

Fasting patient, between 8 and 12 hours. Quiet room, with dim light at a temperature between 20˚C - 22˚C. Supervised by a neurologist, a cardiologist and a medical technologist. Cardiology personnel installs continuous EKG monitoring.

A hemoglucotest is performed prior to the exam.

Electrocardiographic monitoring and continuous measurement of blood pressure, heart rate and surveillance of doctors and nurses are performed.

Drugs, venous lines, a defibrillator, and equipment for cardiopulmonary resuscitation are available.

4. Tilt Test Protocol

A record of heart rate (HR) and blood pressure (BP) and of symptoms reported by the patient is kept every 5 minutes. The reason for stopping the examination or any important incident is noted and recorded at any time. The sublingual ni-

troglycerin protocol is based on Del Rosso [10].

Carotid massage is performed on all patients over 60 years of age. Previous discard of murmur or carotid stenosis or stroke in the last 6 months. Five minutes on each side [11].

Tilt Test ends if a "positive HUT" is obtained: This is syncope (loss of consciousness) or presyncope (dizziness, nausea, paleness, etc., announcing that syncope is imminent). Associated with low blood pressure (systolic BP < 70 mmHg) or low blood pressure plus bradycardia, or if intolerable patient discomfort occurs.

If there are no symptoms, it is terminated due to the end of the protocol.

In addition, sympathetic and parasympathetic function tests (Valsalva maneuver and deep breathing test) are performed in order to support or rule out failure in the baroreflexes.

The equipment consists of: Digital monitor (Ohmeda 2300 Finapres BP Monitor USA). Digital cuff placed on the index or middle finger to measure BP and HR continuously.

Electric tilting table (Magnetic Manumed USA) and electrocardiogram monitor (Quinton Q4500 USA). The patient is fastened to the table with two velcro straps (knees and chest).

For the statistical comparison, Anova and logistic regression are used, and depending on the sample size, a non-parametric test is used.

5. Ethical Clearance

Our study was analyzed and approved by the institutional ethics committee of the Hospital Militar, and was carried out in accordance with the ethical standards of the Helsinki Declaration 1964. Patients and controls signed an informed consent before inclusion.

6. Results

In 52 syncope started from the age of 50 or later, 34 men (64%). Mean age 67.5 years (range: 50 - 89 years). Of these, 54% were 65 or older (range: 65 - 89 years/43% female). **Table 1** shows the patients according to age and sex.

Table 2 shows the number of patients and the age of onset of their syncopes by decade.

The predominance of males is clear in these adults who have just started their syncope, even worse after 65 years (57% at that age). The opposite occurs in the youth population [1] [2] [3] [4] [12].

The reasons for consultation in our patients were: syncope, lipothymia, orthostatic dizziness and different combinations between them. All these account for 88% of cases (n: 46), which is comparable to the youth population [1] [2] [3] [4] [12] [13].

Table 3 shows the reason for consultation of the patients and the positivity of the HUT.

Table 1. Sex and age of onset of syncopes.

Sex and age at the onset of symptoms			
	Female	Male	Total
N° of cases	18 cases	34 cases	52 cases
Mean Age	69.9 years	66.1 years	67.5 years

Table 2. Number of patients and age of onset of their syncopes by decade.

Number of cases and age of onset of syncope per decade		
Age (years)	N° of cases	%
50 - 59	18 patients	34%
60 - 69	10 patients	19%
70 - 79	15 patients	29%
80 - 89	9 patients	17%
TOTAL	52 cases	100%

Table 3. Reason for consultation and Tilt Test result in 52 patients.

Reason for consultation	N° of cases	HUT positive %	HUT Negative %
Syncopes	26	20 (77%)	6 (23%)
Syncopes and lipothymia	7	6 (86%)	1 (14%)
Lipothymia	6	6 (100%)	Zero
Postprandial syncope	5	4 (80%)	1 (20%)
Postural dizziness	4	3 (75%)	1 (25%)
Dizziness and syncopes	1	1 (100%)	Zero
Convulsive syncope	1	1 (100%)	Zero
Laziness and drowsiness	1	Zero	1 (100%)
Parkinson and dizziness	1	Zero	1 (100%)
Total	N: 52 (100%)	N: 41 (79%)	N: 11 (21%)

7. Findings in the Tilt Test

The overall positivity of the HUT was 78%, (n: 41), which is common in patients selected by trained doctors and sent to HUT [14] [15] [16].

The most important final events found in the positive HUT were: symptomatic orthostatic hypotension (OH) (n: 24 cases/46%), with final collapse of the patient. Followed by mixed vagal syncope mainly vasodilator n: 11 (21%) and cardioinhibitory vagal syncope with 8% (n: 4). This is shown in **Table 4**.

Negative Tilt Test: Eleven patients (21%), who consulted for syncope, lipothymia, or orthostatic dizziness had a negative HUT. They were followed up, trying to find a possible cause of their symptoms.

The possible explanation of symptoms in patients with negative HUT can be seen in Table 5.

8. Orthostatic Hypotension (OH)

It is frequently found in our patients n: 24 (46%).

Hypotension can occur early, within the first 3 minutes (4 cases) or more frequently after 3 minutes (late OH), in 20 patients [17] [18] [19].

To consider it as the cause of the condition, hypotension must produce orthostatic symptoms during the HUT [19].

Table 6 shows the number of patients and the probable cause of OH, postulated after an average follow-up of 4.5 years. The n is greater than 24, because

Table 4. Tilt Test results 52 patients.

Results	N°	%
Orthostatic hypotension	24	46%
Vagal syncope > vasodilator	11	21%
Negative Tilt Test	11	21%
Vagal syncope cardio inhibitory	4	8 %
Carotid sinus syncope	2	4%
TOTAL	52	100%

Table 5. Possible etiological explanation of consultation symptoms in patients with a negative Tilt Test (HUT).

Sex	Age	Reason of Consultation	Explanation	N
Male	63	Parkinson's disease screening	Parkinson's disease with no dysautonomia	1
Female	76	Syncope	Cardiac arrythmia	1
Male	57	Cough and syncope	Cough Syncope	1
Male	56	Daytime sleepiness	Sleep Apnea	1
Female and Male	76 and 52	Postural dizziness	Stroke + High blood pressure (uncontrolled)	2
Female	66	Syncope	Drug effect	1
Male	65	Syncope after feeding	Postprandial syncope	1
Male (both)	62 and 71	Syncope and dizziness	Carotid sinus syncope	2
Male	59	Orthostatic dizziness	Not explained	1

Table 6. Factors associated with the existence of orthostatic hypotension.

Factors associated with OH	N	%	Mean age
Cardiovascular diseases	13	25	78
Hypotensive drugs	13	25	72
Baroreflexes failure*	8	15	75
Autonomic NS neuropathy	3	6.0	72
Joint hypermobility ≥ 7	2	4.0	65
Postprandial syncope	1	2.0	73
Diabetic neuropathy	3	2.0	63
No apparent explanation	5	10	68
Total combinations	48	89%	70.5 years

*Includes 3 patients with Parkinson's disease.

several factors were combined in most of the patients, without being able to assure which is the influence that each one of them weighs. 48 possible combinations were found, which would explain 89% of the causes of OH.

In 5 patients with clear OH, we were unable to find any associated concomitant factors.

9. Drug Use and Orthostatic Hypotension

19 patients (36%) took medications potentially related to the production of OH and syncope or lipothymia: Antihypertensive, antidepressant or antipsychotic drugs [7] [20].

Antihypertensive drugs found were losartan n = 6 (11%), enalapril 5 (10%) and atenolol 4 (8%). Combined with diuretics, or with psychotropic drugs n = 4 (8%).

Antipsychotics or antidepressants used were: amitriptyline, trazodone, haloperidol and quetiapine. All were combined with antihypertensive drugs (n: 4). Five patients were using sertraline, but their syncopes had already started before starting the drug.

Eight of these patients (42%) benefited from the withdrawal or decrease of antihypertensive drugs.

10. Cardiovascular Comorbidities

In 31 patients we found comorbidities in the cardiovascular area (60%), and the use of drugs related to that area.

High blood pressure (HBP) 20 cases, diabetes mellitus (DM) 10 cases, stroke 5 cases, heart failure 2 cases, pacemaker 1 case, arrhythmia one case, dyslipidemia 3 cases, and different combinations between them (n: 23).

11. The Deterioration or Loss of the Baroreflexes

In 10 cases (24% of positive HUTs), we found damage to the baroreflexes, corroborated as orthostatic hypotension, without accompanying tachycardia [21] plus abnormalities in blood pressure response in the Valsalva Maneuver and in deep breathing test [22] [23].

12. Situational Syncope

We found two patients with situational syncope. Both males. One of them when coughing presented syncope with severe drop in BP and HR, consistent with his symptoms ("cough syncope"). However, his orthostatic HUT was negative.

The other case presented episodes of very prolonged laughter ("gelastic syncope"), with choking and syncope. His orthostatic HUT was positive (vasodilator).

13. Postprandial Syncope

We found postprandial syncope (PPS) in 5 patients (9.6%). Mean age: 71.2 years. Four males. Syncope only appeared after feeding. Four with positive HUT (3 with vasodilator VVS and one with OH). They all suffered from HBP. Three were type 2 diabetics and two had hypercholesterolemia. In patients under 60 years of age, we have only found one case of postprandial syncope.

14. Carotid Sinus Syncope

Carotid sinus compression was practiced in all patients 60 years of age or older, according to what international protocols dictate [11].

Compression was positive in 2 males (4%). Mean age 66 years. Both had a negative orthostatic HUT for vasovagal dysautonomia. They consulted for presenting dizziness or syncope when changes in posture of the head. This endorses the usefulness of using such compression in the study of syncope in older adults. [11].

15. Parkinson's Disease

Four patients (3 males) Mean age: 78 years. Three with Parkinson's over 7 years of evolution, in treatment associated with hypotensive drugs. Sent by syncopes and lipothymia.

All three with failure in the baroreflexes and positive HUT for OH.

In one with recent Parkinson's (3 years). HUT was performed to rule out dysautonomia. He did not suffer from syncope or lipothymia. His HUT was negative.

16. A Special Case: Patients over 65

They are the bulk of our cases, n = 28 (54%). 16 males (57%).

These patients suffer more frequently of: syncope (78%), orthostatic hypoten-

sion (85%), cardiovascular comorbidities (68%), postprandial syncope (9.6%), failure of baroreflexes (28%), use of hypotensive drugs (53%), concomitant diseases such as Parkinson's (14%), carotid sinus syncope (3.5%) and many associations between these factors.

17. Follow Up

This covers between 15 months and 7 years (X: 4.5 years). It allowed us to find causes that are unusual at younger ages: Use of hypotensive drugs (n: 8), one of them, added a diabetic autonomic neuropathy. Postprandial syncope (n: 4), post-stroke dizziness (n: 1). Arrhythmia (n: 1). Carotid sinus syndrome (n: 2). Sleep apnea with hypersomnia (n: 1) and degenerative neurological diseases such as Parkinson's disease with dysautonomia (n: 3). The total for this type of unusual cause is 20 cases (38%).

18. Unexplained Cases

Despite follow-up and complementary examinations, there were 6 patients (11%), in whom we did not find associated factors that made them prone to syncope.

They all had a positive HUT. Three with OH and three with vasovagal syncope.

In three of them, syncope occurred only once.

It is known that 10% of syncopes remain unexplained in any statistic [24] [25] [26].

19. Discussion

Approximately 50% of people present their first fainting, between 15 and 30 years of age [1], at that age, its frequency is usually double in women and neurally mediated syncope (VVS) predominates as the first cause [2] [3].

But up to 10% to 15% of the population would have it after the age of 65 [3].

In our patients over 50 years old, the relationship of sex is reversed, with the frequency being much higher in men (63% vs. 38%) than in women [27] [28].

The most important final event in HUT was orthostatic hypotension (46%). Vasovagal syncope is relegated to second place with 29%. This supports the observation that there are different causes for syncope in these patients than in young people [2] [4] [5] [6].

Certain factors alone or associated could explain the presence of a favorable terrain for syncopes and orthostatic hypotension [2] [4] [5] [6] [29]: cardiovascular comorbidities with structural damage to the vascular tree: HBP, atherosclerosis, heart failure, coronary disease, hypotensive drugs, diabetes mellitus, carotid sinus disease, failure in baroreflexes with autonomic insufficiency, Parkinson's disease, postprandial hypotension and autonomic peripheral neuropathy. All of them favor the occurrence of syncope and OH in older adults [2] [5] [6] [29]. Even if HBP [30] [31] [32] coexists.

The most frequent combinations in the same patient were: HBP plus hypotensive drugs (n: 13), HBP plus diabetes mellitus plus hypotensive drugs (n: 4), diabetes mellitus plus hypotensive drugs (n: 2), diabetes mellitus plus failure in baroreflexes (n: 6), HBP plus diabetes mellitus (7) and finally HBP plus failure in the baroreflexes (n: 4).

20. Hypotensive Drugs

Patients report that, when starting using a medication, fainting started or worsened, and stopped when they stopped using it. We can confirm it, by testing withdrawal or reducing doses. This occurred in 8 of 22 cases (36%) [8] [33] [34] [35] [36].

The effect of the sum of antihypertensive drugs in combination with psychotropic, anxiolytic, antidepressant drugs and the production of OH and syncope is clear [37].

Orthostatic hypotension is caused by failure of the mechanisms that compensate for the drop in venous return produced when standing. When OH is recent you should look for polypharmacy, or dehydration.

When it is chronic, it is more likely to find failure in the baroreflexes (worse after 65 years) [38] [39] [40], heart failure, arterial thickening, cardiovascular diseases, and central or peripheral neurological disorders that damage the autonomic nervous system such as diabetic neuropathy autonomic, and/or peripheral, idiopathic neuropathy [41] [42] [43] [44] or Parkinson's disease [44] [45]. Even worse if you add hypotensive drugs [46].

The factors found, concomitant with the OH [41] [47] [48] [49] are in **Table 6**.

OH, also asymptomatic, brings an increase in mortality and major cardiovascular events [50] [51] [52] [53].

We found 2 patients with joint ligamentous hypermobility (≥ 7), despite exceeding 50 years old [54] [55] [56]. In one of them, this was the only associated finding. The other patients also suffered from failure of the baroreflexes.

Situational syncope two cases. One of them presented a "cough syncope". The other case was a "gelastic syncope". Both are rare causes of VVS syncope, produced only in situations associated with the Valsalva's manouver [57] [58].

Postprandial hypotension (9.6% of our cases) is common in geriatric patients and is a little-known cause of syncope [59] [60]. Patients with neurological diseases, autonomic failure, diabetics, hypertensive or dialysis patients are at risk [59] [60]. It is due to an inadequate sympathetic response to splanchnic vasodilation produced during feeding.

Syncope due to compression of the carotid sinus, is usually mentioned as a cause of syncope in the elderly [11] [61] [62], but at least in our sample, was rare.

In 6 patients (11%), despite follow-up, their syncope was unexplained.

Three of them consulted for a single syncope and the others for a maximum of 2 or 3 episodes. We believe that in these patients, circumstances like ambient

heat, dehydration or prolonged standing came together at a certain moment: But during follow-up, they have not been repeated [63] [64] [65].

In this subgroup, it is mandatory to determine if they have a heart condition, since they have up to 30% more mortality than the population of the same age [2] [26]. So, our protocol includes carrying out a complete cardiological evaluation previous to HUT.

21. Study Limitations

First, it is difficult to obtain statistically valid conclusions due to the small number of patients studied in our sample. We understand that a greater sample is necessary in the future.

Second, we depend on the good memory of the patient or his relatives to recall that they are consulting for the first syncopal episode. In some cases, the patient, after a second interrogation, remembers having suffered episodes during childhood or adolescence (66).

Third, altough an exhaustive interrogation and pre-HUT exams, it is not always possible to completely rule out other causes such as drowsiness, vertigo, orthostatic dizziness or accidental falls.

22. In Sum

It is not common to begin with syncopes over age of 50.

Finding the ultimate cause for syncope in these patients can be difficult given the multiplicity of interacting factors. They are mainly male and exhibit different causes than those found at a young age.

Its study is more exhaustive and requires a good anamnesis, knowing the drugs used by the patient, knowing the concomitant diseases and careful monitoring to get closer to the diagnosis. We observe the high frequency of orthostatic hypotension as a cause for syncope, which is attributable in many cases to advanced age, failure of baroreflexes, cardiovascular and neurological diseases, and polypharmacy [29] [66] [67].

Acknowledgements

Cardiology technician personnel: Erika Collío, Pilar Altamirano, Francisco Osorio and Liliana Huerta. Cardiology nurses: Leonor Acevedo and Clotilde Sanhueza. Cardiology Counseling Dr. José Pardo Gutiérrez.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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International Journal of Clinical Medicine, 2020, 11, 454-464
https://www.scirp.org/journal/ijcm
ISSN Online: 2158-2882
ISSN Print: 2158-284X

Rhomboid Flap vs. Keystone Perforator Island Flap (KPIF) in the Treatment of Pilonidal Sinus Disease: Comparison of Short-Term Results

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How to cite this paper: Roatis, M.D. and Georgescu, A.V. (2020) Rhomboid Flap vs. Keystone Perforator Island Flap (KPIF) in the Treatment of Pilonidal Sinus Disease: Comparison of Short-Term Results. *International Journal of Clinical Medicine*, **11**, 454-464.
https://doi.org/10.4236/ijcm.2020.117038

Received: June 4, 2020
Accepted: July 24, 2020
Published: July 27, 2020

Abstract

Background: Pilonidal disease is a chronic inflammatory disease of the sacrococcygeal region that mainly affects young people. Its incidence is 26 cases per 100,000 persons. Although many techniques have been described, there is no consensus on the treatment of pilonidal sinus disease (PSD). **Materials and Methods:** This study included 30 patients with PSD who were treated between May 2014 and September 2017. All cases underwent excision and flap reconstruction. The operative time, postoperative complications, the length of hospital stay, painless sitting and walking time, patient satisfaction and recurrence were evaluated prospectively. **Results:** The results of this prospective, randomized and comparative study are based on experience of a single surgical centre. All patients were followed up 18 months after discharge from the hospital. There is a difference in surgery durations (minutes) between the two groups (33.86 ± 2.89 min. in "keystone" flap vs. 41.26 ± 4.19 in the "rhombic" flap group) ($p = 0.001$). There were no significant differences in the length of hospital stay, painless sitting and walking time or patient satisfaction. The total complication rate was 66.6% after rhomboid flap compared with 6.6% after keystone flap. There was no flap necrosis. **Conclusions:** Both of these methods have shown to be successful in treatment of PSD. The KPIF is associated with the advantages of very simple design, abundant blood supply from the perforator vessels and lower rate of complication.

Keywords

Pilonidal Sinus Disease, Keystone Perforator Flap, Rhomboid Flap, Surgical Treatment

1. Introduction

Pilonidal sinus disease is a common and acquired entity of young adults and has an estimated incidence of 26/100,000 in the general population [1] [2]. It is seen more commonly in men than in women (male:female ratio 3 - 4:1) [3]. It usually affects the skin overlying the natal cleft at the sacrococcygeal area [4] and was first described by Anderson in 1847 [5]. Several short- and long-term outcomes have been published comparing different methods of treatments in PSD. Most of these studies compare Limberg rhomboid flap with other surgical or non-surgical techniques. There are no studies within the literature comparing rhomboid flap versus keystone perforator island flap (KPIF) although KPIF has been widely used in clinical practice since it was introduced. The aim of the present study was to compare the short-term results of the management of sacrococcygeal PSD with the keystone flap vs. the rhomboid flap (Limberg or Dufourmentel) in a randomized controlled trial as regards recurrence and complications rates, operative time, duration of hospital stay, time to walk without pain, time to sitting on toilet without pain, duration of the incapacity to work and cosmetic satisfaction of the patients.

2. Materials and Methods

2.1. Patients

Between May 2014 and September 2017, we included a series of 30 patients, prospectively and consecutively, which randomly divided into two groups. Fifteen patients (group 1) undergone elliptical excision and keystone perforator island flap (KPIF) and others 15 (group 2) undergone a rhomboid excision and the rhomboid flap to cover loss of substances after pilonidal cyst excision. The nature of surgical procedures was explained to the patients and informed consent had been obtained. Both surgical procedures were performed by the same surgeons. The surgeries were performed under spinal anesthesia and the patients were placed in the prone jack-knife position. All patients received a single intravenous dose of 2 g Sulcef (Cefoperazone sodium 500 mg + Sulbactam sodium 500 mg, Novartis India Ltd.) with 30 min. before surgery. Hair of the sacral and gluteal regions was shaved a few days preoperatively, and rectal cleansing with enemas was performed the night before the operations. The buttocks were separated with adhesive tape to allow wide exposure of the operative field. In the case of the keystone perforator island flap (KPIF), the parasacral perforators were identified and marked on the skin, using an acoustic Doppler technique, for safety only, although this is not absolutely necessary because the integument covering this region has plenty of perforators and vascular connections from lumbar and gluteal arteries. The skin was prepared with 10% povidone-iodine solution. In both techniques, methylene blue was injected in the pilonidal sinuses to guide the excision [6]. Dissection and hemostasis were performed by using electrocautery.

2.2. Randomization

Patients were randomly assigned to undergo surgery with either the rhomboid flap or the keystone perforator island flap. Randomization was performed by using a sealed envelope that contained a paper with "Rhomboid flap" or "Keystone flap" which were opened before surgery.

3. Surgical Technique

3.1. Keystone Perforator Island Flap (KPIF) (Figure 1) (Figure 2)

The keystone perforator island flap (KPIF) was described by Behan in 2003 as a curvilinear trapezoidal shaped flap, representing the architectural shape of the keystone in Roman arches, relying on randomly fasciocutaneous or musculocutaneous perforators. It representing two opposing V-Y flaps joined together, with a flap width at a 1:1 ratio to an elliptical defect [7] [8]. Its length is determined by the size of the wound. An elliptical incision, including the pilonidal sinus, was made and a keystone flap was designed adjacent to the wound. The KPIF was incised along its sides without leaving a skin bridge intact and dissected down to the deep fasciae which was incised for further advancement, without or with minimal undermining to preserve the integrity of the vascular perforators [9]. Then, the flap padle was elevated and advanced to cover the defect. When we suture the flap on the defect-side it is very important to avoid dead space formation. Wound closure is achieved in a single layer using 1 Dafilon (B|Braun, Aesculap). The donor site was closed primarily. If the flap exceeds the 1: 1 ratio then more perforators can be incorporated into its structure thus creating a multiperforator advancement flap [8].

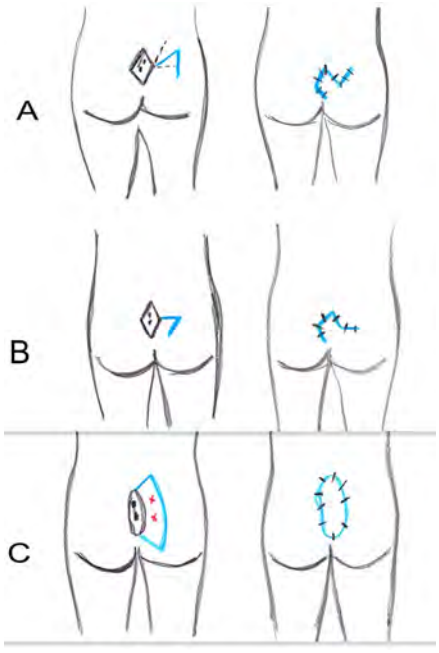

Figure 1. Diagram of surgical procedure (A) Dufourmentel flap (rhomboid); (B) Limberg flap (rhomboid); (C) Keystone perforator island flap.

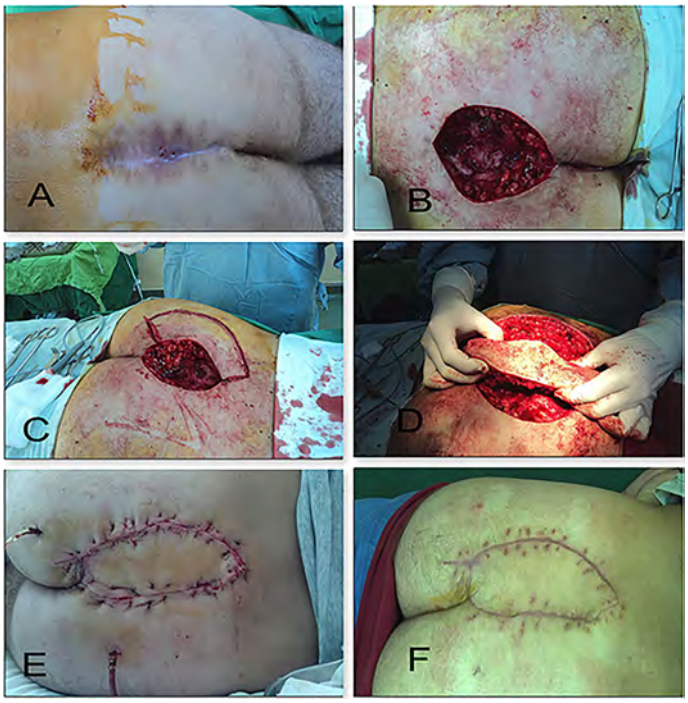

Figure 2. Keystone perforator island flap-clinical aspect (A) pilonidal sinus disease in sacrococcygeal area; (B) post-excisional defect; (C) the design of the flap; (D) the island-form and advancement of the flap; (E) postoperative clinical photography immediately after the flap inset; (F) aspect at 2 months postoperative.

3.2. Rhomboid Flap (Figure 1) (Figure 3)

Rhomboid flap was first described by Alexander Limberg in 1946 as a transposition flap with a random blood supply and it was modified by Dufourmentel in 1962 [10] [11]. In the Limberg flap, all angles are 60˚ and 120˚ and all sides are equal. The Dufourmentel flap has a safer blood supply than the Limberg flap because its base is wider [12]. The pilonidal sinus was included in a rhombic form and the flap was marked on the skin. Afterwards, the lesion was excised down to the presacral fascia under the guidance of methylene blue, and the fasciocutaneous flap was transposed medially to the defect [6]. A 2-layer closure was performed. The subcutaneous layers were approximated with 2 - 0 Vicryl (polyglactin 910, Ethicone, Johnson & Johnson) interrupted sutures that fix the undersurface of the flap to the presacral fascia and avoid tension and obliterate the dead space. The skin was closed with 2 - 0 Dafilon (B|Braun, Aesculap) interrupted sutures [13]. In both procedures, the suction drain was placed beneath the flap through 1 or 2 separate stab incision.

3.3. Postoperative Follow-Up

All patients were followed-up in the outpatient clinic weekly in the first month following discharge from the hospital for wound assessment and then every 3 months to one year. The skin sutures were removed after 14 postoperative days. The suction drain was removed when the effluent was less than 20 ml per day.

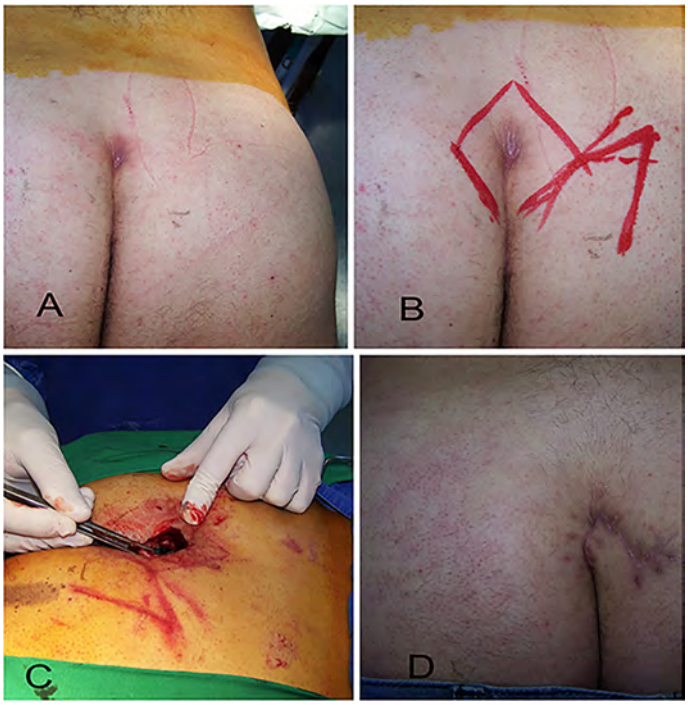

Figure 3. Rhomboid flap-clinical aspect (A) pilonidal sinus disease; (B), (C) Dufourmentel (rhomboid) flap; (D) Final result with stable coverage six weeks after surgery.

3.4. Statistical Analyses

All statistical analyses were performed using IBM SPSS Statistics for Mac. Student's t-test was used to compare continuous variables (quantitative data) between groups and Chi-squared or Fisher's exact test was used for categorical variables (qualitative data). The results are presented as mean plus or minus standard deviation for quantitative data or proportion, as appropriate. A two-sided p values of < 0.05 were considered to indicate statistical significance.

4. Results

Of the 32 patients enrolled initially, 2 were lost to follow-up within 4 months of the procedure and were not included in the study. The remaining 30 patients: 24 males (80%) and 6 females (20%) were included in this analysis.

The average age of the included patients was 35.4 years (range: 20 - 50 years) in the "keystone" group and 27 years (range: 17 - 48 years) in the "rhombic" group.

Operation time is important for the effectiveness of a surgical technique. It was defined as the time from the start of skin incision to the end of the last stitch. In terms of operative time, there was a slightly difference between the two groups: 33.86 ± 2.89 min. in the "keystone" flap group vs. 41.26 ± 4.19 min. in the "rhombic" flap group ($p = 0.001$) (Table 1).

The seroma, which is defined as the formation of non-infected serous fluid collection beneath the flap, has a formation rate significantly higher in the

rhomboid group (3 patients-20%) than in the keystone group (0 patients) (Table 1).

No patient in the keystone group had wound infection, but this complication occurred in 2 patients (13.3%) in the rhomboid flap group (p = 0.526). These two patients were treated with oral antibiotic therapy for 7 days (Table 1).

The rates of wound dehiscence in group 1 was 6.66% (1 patient and in group 2 were 20% (3 patients). All of these patients healed by second intention without requiring additional surgical procedures (Table 1).

The duration of hospital stay is defined as the number of days from the operation day until the day of discharge.

The average length of hospital stay for the "rhombic" group was longer than "keystone" group (4 ± 1.98 days versus 2.33 ± 0.487 days, p = 0.003), suture disruption and infections are likely to increase the duration of hospitalization (Table 2).

There were no statistically significant differences between the two groups regarding time to walk pain-free (in days): 9.06 ± 1.48 in group 1 and 9.60 ± 1.45

Table 1. The distribution of complications within the 2 groups.

Complication	Total (n = 30)	Group 1 (n = 15)	Group 2 (n = 15)	P value
Minor complications	9 (30)	1 (6.66)	8 (53.33)	
Seroma	3 (10)	0	3 (20)	0.224
Wound dehiscence	4 (13.33)	1 (6.66)	3 (20)	0.598
Wound infection	2 (6.66)	0	2 (13.33)	0.483
Major complications				
Total or partial flap necrosis	-	-	-	
Recurrence	2 (6.66)	0	2 (13.33)	0.483

Values are presented as number and (%).

Table 2. Patients VAS score, time to walking pain-free and time to sitting on toilet without pain, incapacity for work (days) and hospital stay (days).

Parameters	Group 1 (n = 15)	Group 2 (n = 15)	P value
VAS score	7.13 ± 1.18*	7.0 ± 1.0*	0.742
Time to walking pain-free	9.06 ± 1.48*	9.60 ± 1.45*	0.329
Time to sitting on toilet without pain	10.26 ± 1.03*	11.8 ± 2.17*	0.032
Incapacity for work (days)	21.93 ± 1.94*	23.4 ± 3.83*	0.017
Hospital stay (days)	2.33 ± 0.487*	4 ± 1.98*	0.003

*mean.

in group 2, (p = 0.329) and slight difference as for time to sitting on toilet without pain (in days), 10.26 ± 1.03 and 11.8 ± 2.17, respectively (p = 0.032) (Table 2).

Recurrent disease occurred in 2 patients (13.3%) who undergone rhomboid flap (group 2) in the 11th and 14th postoperative months. No patients had recurrence in the "keystone" group (group 1). All cases were followed up for 18 months.

Time away from work was defined as the date on which patient returned to employment from the date of surgery. The postoperative duration of incapacity for work (days) was longer in the "rhomboid" group than in the "keystone" group (23.4 ± 3.83 days vs. 21.93 ± 1.94 days) (p = 0.017) (Table 2).

The visual analogue scale (VAS) score from 1 - 10 (0 for "very bad" to 10 for "very good") was used in order to assess the cosmetic satisfaction of the patients.

The cosmetic satisfaction was determined at the end of the third postoperative month by asking patients to describe their satisfaction with the operation scar due to pilonidal sinus disease.

The VAS score for satisfaction with the cosmetic appearance of the scars in "keystone" group was 7.13 ± 1.18, whereas it was 7.0 ± 1.0 in the "rhomboid" group (p = 0.742) (Table 2).

No patients developed flap necrosis in the two groups.

There were no donor site complications.

5. Discussion

The etiopathogenesis of SPD is still under debate, based on two principal theories: "congenital" theory (the absence of coalescence of the primitive ectoderm) and "acquired" theory (the hair follicle has the primordial role) [14].

The ideal surgical procedure for sacrococcygeal PSD should be simple and quick, should not require a long hospital stay, should have low recurrences rate and postoperative complications, low cost and high patient satisfaction [15] [16].

Many surgical procedures for SPD have been developed, but none of them fulfill all of these features. All of surgical techniques have advantages and disadvantages.

Excision of the cyst is not technically difficult but how to cover the surgical defect after excision of sacrococcygeal pilonidal sinus may be sometimes a dilemma.

Of all the surgical procedures, the local flaps fill the defect, avoid a midline scar, provide tension free closure and can flatten the natal cleft and thereby offer advantages over direct closure techniques [17].

The rhomboid flap is one of the most common surgical techniques used in PSD forasmuch is easy to design, raise and inset.

It has gained popularity because it is associated with low complication and recurrence rates [15].

However, in the rhomboid flap large tissue should be displacements and the

flap is usually a random pattern, which limits the size of flap that can be raised. As it looks like Arpaci *et al.* the lower pole of the rhomboid flap can be a weak point in terms of recurrence [18]. On the contrary, the keystone flap does not require extensive mobilization and it does not require fixation at the sacral fascia and thus the operative time is lower than then when we use rhomboid flap (33.86 ± 2.89 min vs. 41.26 ± 4.19 in our study).

In KPIF the wound tension is effectively reduced and redistributed and thus the healing process is facilitated [19]. The KPIF has been widely used in clinical practice since it was first introduced for the reconstruction of defects located on the head and neck, trunk, and extremities. The KPIF can be divided into four subtypes: type I the classical flap, in which the flap is superficially disected of deep fascia; type IIA requires division of the deep fascia; type IIB that cannot be closed without a skin graft due to the tension, type III, which consists of two KDPIFs; and type IV that is a rotational keystone flap [7].

KPIF is a multiperforator advancement flap based on random fasciocutaneous or musculocutaneous perforator but in the sacral area, these may be parasacral perforators arise from the lateral sacral artery or the internal pudendal artery as described Koshima and Ahmadzadeh [20] [21].

The most important factors to consider when evaluating the results of the surgical treatment are: early complications and delayed relaps [6].

Ersoy *et al.* reported wound infection rate after the Limberg flap (8%) [22]. Altintoprak *et al.* had 3.3% (11 patients) rate of infection on 324 patients after Limberg flap procedure. In our study, no patients have wound infection after KPIF but infection rate in rhomboid flap was 13.3%.

The main aim of the surgical treatment of pilonidal sinus disease remains prevention of recurrence.

A meta-analysis of more than 100 randomized controlled trials found that the recurrence rate at 5 years overall surgical therapies for pilonidal disease was 20.3% [3] [4] [23].

Topgül *et al.* on 200 operated patients reported 2.5% recurrence rate for Limberg flap and Daphan *et al.* on 147 operated patients noted 4.8% recurrence [24].

We had no recurrence with keystone flap but in rhomboid group we had 2 patients with recurrence at 18 months.

The hospital stay is also an important criterion in determining the success of a surgical technique.

Mentes *et al.*, in their study, on 353 patients operated, reported 4.51 ± 2.85 days of hospital stay, Topgül *et al.*, 3.1 days on 200 patients, Erdem *et al.*, 3.5 ± 1.16 days on 40 patients and Eryilmaz *et al.*, 3 days on 63 patients [24].

In our study, we operated 30 patients, and the mean length hospital stay was 4 ± 1.98 days for "rhomboid" group and 2.33 ± 0.487 days for "keystone" group.

In contrast to the direct suture, in the case of cutaneous flaps, the scars are larger and, therefore, its can be sometimes unpleasant for the patient. The cosmetic appearance of the scars may be more important for women than men.

Eryilmaz *et al*., reported that as many as 40% of patients were not pleased with the appearance of Limberg flap scars [17] [25]. In our study, the cosmetic appearance of the scars at the operation site was accepted by the majority of the thirty patients. No significant difference was found between patients treated with the keystone flap technique and those treated with the rhomboid flap technique. This was an issue especially for female patients, that expressed their dissatisfaction with the cosmetic outcome for both procedures. All these results encourage to use keystone flap technique in the surgical management of PSD.

6. Conclusions

The keystone and rhomboid flaps are useful techniques to close wounds after pilonidal cyst and both of them achieve off-midline closure and flattening the natal cleft.

The KPIF, unlike rhombic flap techniques, avoids extensive mobilization of tissue and maintains good blood supply with minimal tension. Where a larger excision is required, we can use the modified KPIF.

Although, both of these methods have shown to be successful, the keystone flap technique seems to be a safer method. We successfully managed PSD with excision and KPIF reconstruction, without recurrence and minimal postoperative complications. In conclusion, we believe that KPIF is a good alternative choice in the management of PSD because its elevation is easy, fast, and safe.

Limitations and Future Work

The mean follow-up time for patients in this study was 18 months and this comprised 30 patients. This period is short for the evaluation of recurrences. A larger patient population and a longer follow-up time are required.

Consent

All included patients provided their written informed consent, and the study's protocol was approved by the research ethics committee of Emergency County Hospital Satu Mare.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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