Original Article

A2-PULLEY RECONSTRUCTION: A COMPARISON OF THREE TECHNIQUES IN A CADAVERIC MODEL AND REVIEW OF THE LITERATURE

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ABSTRACT

Reconstruction of flexor tendon pulleys presents serious problems for the hand surgeon. The clinical result after reconstruction efforts in a flexor tendon-pulley unit depends on restoration of grip strength and active range of motion of the finger. Eight forearms were used to evaluate excursion resistance by range of motion and strength resistance of three different A2 pulley reconstruction (Bunnell's, modified Odobescu technique, pulley venting). The results of the in vitro study of excursion resistance and strength resistance of pulley reconstruction demonstrated that the three techniques have similar results, Bunnel's one just brought a better flexion for PIP joint flexion but pulley venting can be considered a second choice for that patients who has the anatomical absence of palmaris lounges (15% of people). The triple loop can be considered as the best choicefor A2 pulley reconstruction, in terms of strength and articular range of motion, in that cases where the techniques in not affordable, pulley venting can be considered a solid second choice for its results similar to the triple loop techniques, for the absence of tendon sacrifice and for the fast execution time.

KEYWORDS: *pulley, pulley venting, free tendon loop, tendon gliding, cadaveric model*

INTRODUCTION

The digital flexor sheath pulley system (Fig. 1) is a structure that permits normal and efficient flexor tendon function (1).

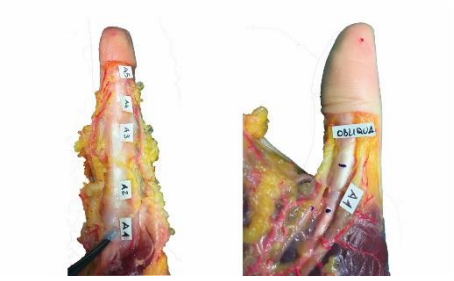

Fig. 1. *Pulley system anatomy.*

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This system is composed of a superficial retinacular and a deep synovial component. The pulleys are made of fibrous tissue, which encircle the flexor tendons forming a fibre-osseous channel which keeps the tendons adjacent to the phalanges, this for transferring a translational force generated from the muscle-tendon unit into a rotational moment on the phalanges. In the fingers there are 5 annular (A1-A2-A3-A4-A5) and 3 cruciate (C1-C2-C3) pulleys in descending order from proximal to distal. The A2 and A4 pulleys insert directly into bone, the A1, A3, and A5 insert mostly into volar plate, and with the cruciate pulleys, permit compression without impingement and expansion during finger flexion and extension. A1, A3, and A5 pulleys are located over the metacarpophalangeal (MCP), proximal interphalangeal (PIP), and distal interphalangeal (DIP) joints, respectively.

The cruciate pulleys C1, C2, and C3, lie between the annular pulleys. Proximal to the A1 pulley lies the palmar aponeurotic (PA) pulley, which is composed of transverse ligament of the palmar aponeurosis attached to the underlying septa of Legueu and Juvara, forming an arch over the flexor tendons superficial. For the thumb 4 pulley system components were described: A1, Av, oblique, and A2 pulleys. The A1 pulley lies over the MCP joint, the oblique pulley runs from proximal ulnar to distal radial over the proximal phalanx, and the A2 pulley is located over the interphalangeal joint. The Av (variable annular) pulley, has now been characterized. First reported in 2012, Schubert and colleagues (2) found this pulley to be present in 93% of cadaver specimens with 3 possible orientations: transverse, oblique, or continuous with the A1 pulley. Because of the historic importance of A2 and A4 pulleys, most surgeons suggest their preservation, repair, or reconstruction. Many surgical techniques have been described for pulley reconstruction to restore hand function (3). The reconstruction of pulleys, both in trauma injuries and tendon surgery, is necessary to avoid bowstringing (4) but above to preserve the natural tendon excursion and to preserve the tendon power. In general, conservation and reconstruction of A2 and A4 pulleys are recommended even if the venting of part of A2 pulley or the entire A4 pulley is now a key point to achieve an optimal result in the tendons sliding (5, 6). In this study, we want to report the effect of three different types of A2 pulley reconstruction on cadaveric specimens and evaluate if there are or not difference in sense of degrees of flexion reached which can lead us to the choice of one respect another.

MATERIALS AND METHODS

Cadaveric dissection

An anatomic study of the pulley was undertaken using 8 forearms from fresh cadavers. None of the specimens had any history of trauma. Dissection was conducted under x3.5 loupe magnification to identify the pulley system under the supervision of support personnel. The arm was dissected from the proximal volar part of the wrist to the pulley, the proximal radio-ulnar joint was secured to prevent rotation of the forearm. The forearms were dissected until the level of the musculotendinous junction, to identify the flexor digitorum profundus (FDP) and flexor digitorum superficialis (FDS) tendons associated with each digit.

Model of pulley reconstruction

Three methods of pulley reconstruction were studied (Fig. 2):

1. Pulley Reconstruction Using Free Tendon Graft (Triple loop) (7, 8) (Fig. 2A). This technique is made using tendon material, taken usually by the palmaris lungus, when present. In his original description, the pulley graft was placed superficial to the extensor apparatus in the middle phalanx and deep to the extensor mechanism. Although this type of pulley may be bulky, it does not seem to have an adverse effect on the extensor system.
2. Pulley Reconstruction Using Flexor Digitorum Superficialis Tendon with a maintained distal attachment (FDS slip). We modified the original technique of Odobescu and colleagues, that use the FDS for A4 pulley reconstruction (9) (Fig. 2B). If the FDS tendon tail is long enough, it may be used as a pulley. In this technique, the distal attachment is preserved, and the free proximal end is sutured over the implant onto the contralateral side. It may be sutured to either periosteum or the original pulley rim or fastened via small holes drilled into bone.
3. Pulley Venting (10, 11) (Fig. 2C) is made with enlargement of pulley to improve the gliding of repaired flexor tendon. Venting incision is made with an incision on the proximal ulnar half parallel to the bone and. Another incision is distally on the opposite side. The central part of the pulley is cut in a vertical “Z” shape and then the two part of the pulley are sutured.

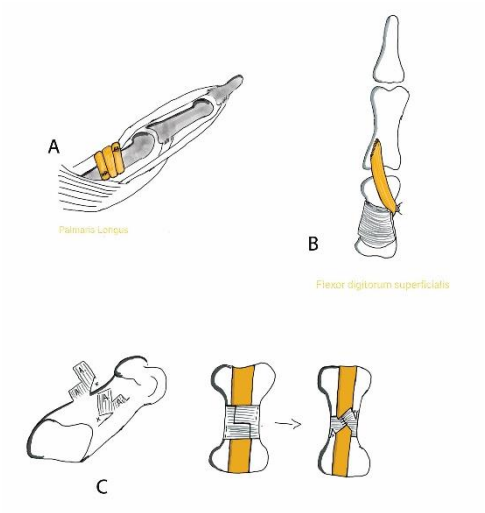

Fig. 2. *Three pulley reconstruction procedures (A) A tendon passed around the proximal phalanx as a triple loop; (B) Flexor digitorum superficialis with distal attachment; (C) Pulley venting.*

Technique details

To standardize the reconstruction technique, all pulleys were sutured with 4-0 non-reabsorbed monofilament. 24 fingers, including 8 index fingers, 8 middle fingers, 8 ring fingers from 8 cadaver specimens, were used in each in vitro model. A cross incision was made through the synovial sheath from the A2 pulley to the A5 pulley with the fingers in full extension. The FDP tendon was then pulled up to obtain the complete flexion of the PIP and DIP to verify the correct scrolling of the tendon. With the finger in full extension, the FDS and FDP tendons of each finger were sutured side to side and attached to the S-shaped hook of the dynamometer (Fig. 3).

Fig. 3. *The FDS and FDP tendons sutured side to side and attached to the S-shaped hook of the dynamometer.*

After, we cut the A2 pulley of the II-III-IV finger and reconstruct each finger in the following techniques):

- II FINGER:A length of another tendon passed around the proximal and middle phalanges as triple loop (Fig.4a);
- III FINGER: slip of flexor digitorum superficialis with a maintained distal attachment (Fig.4b);
- IV FINGER: Direct suture with Pulley Venting (Fig.4c).

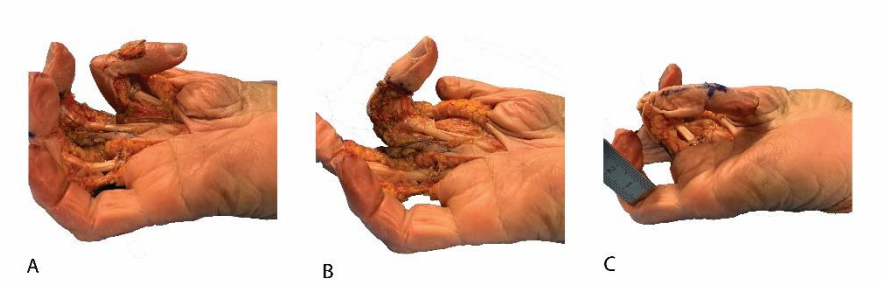

Fig. 4. *Cadaver Specimen of three pulley reconstruction (**A**): triple loop technique; (**B**): flexor digitorum superficialis slip with a maintained distal attachment (**C**): pulley venting.*

Biomechanical analysis

For each finger was evaluated the degree in flexion with intact pulley, with pulley cut and after the pulley reconstruction. The range of motion was measured using a finger goniometer. The angles of the MCP, PIP, DIP joints were measured in maximum flexion and extension, with the forearm and the wrist in neutral position. The Total Active Motion (TAM) of the American Surgery Society of the Hand (ASSH) and both Strickland classification systems (12-15) were applied. Before and after the A2 pulley reconstructions, we applied a constant strength of 5Newton using a dynamometer (Dr.Meter ES-PS01) (Fig. 3) and we evaluated if the pulley resist to a strength >10 N (more than the strength needed for transportation of a six pack of waters bottle).

Statistical analysis

Measurements were performed before and after reconstruction and data were evaluated according to the ANOVA test. The differences before and after the reconstruction were analyzed for statistical value via the t-test. The statistical analysis threshold was set at p-values, significant at <0.05. Statistical Analysis Software (Xlstat add-in excel) was used to conduct two-tailed Student's T, Pearson index, Kolmogorov-Smirnov test.

RESULTS

A total of 24 fingers, for 72 articulations were evaluated. We investigated which structure failed at the maximum load and we got only 1-vented pulley broken because it was too close to the bone, with a calculated bowstringing of 2mm. The other 7-vented pulley were sutured leaving a 3mm bowstringing which showed to be enough to avoid the pulley rupture. The PIP joint was the most affected by the A2 pulley cut, in venting and tendon graft reconstruction it got a range of motion in flexion comparable to the normal pulley. In the 8-pulley reconstructed with triple loops technique it was showed a constant bowstringing of 2 mm for each finger treated. The 8-reconstructions made with FDS slip showed the worst result ending in an important bowstringing of 6-7 mm , losing his containing function (Table I).

Table I. *Degree in flexion of all fingers with an intact pulley and after A2 pulley reconstruction with different techniques.*

Hand #1	MF pre	MF post	IPP pre	IPP post	IPD pre	IPD post	BOWSTRIN GING	Rupture at 10 N application
IInd	0/55	0/55	0/60	0/60	0/40	0/40	2mm	No
IIIrd	0/60	0/60	0/65	0/65	0/80	0/80	6mm	No
IVth	0/40	0/40	0/45	0/45	0/40	0/40	3mm	No
Hand #2	MF pre	MF post	IPP pre	IPP post	IPD pre	IPD post	BOWSTRIN GING	Rupture at 10 N application
IInd	0/70	0/80	0/120	0/110	0/45	0/65	2mm	No
IIIrd	0/70	0/90	0/90	0/100	0/45	0/50	7mm	No
IVth	0/90	0/115	0/110	0/100	0/65	0/90	2mm	Yes
Hand #3	MF pre	MF post	IPP pre	IPP post	IPD pre	IPD post	BOWSTRIN GING	Rupture at 10 N application
IInd	90	90	110	110	75	60	2mm	No
IIIrd	100	85	110	105	105	60	6mm	No
IVth	110	95	115	115	75	50	3mm	No
Hand #4	MF pre	MF post	IPP pre	IPP post	IPD pre	IPD post	BOWSTRIN GING	Rupture at 10 N application
IInd	70	65	120	110	45	45	2mm	No
IIIrd	0/70	0/70	0/90	0/80	0/45	0/45	6mm	No
IVth	90	90	85	80	75	70	3mm	No
Hand #5	MF pre	MF post	IPP pre	IPP post	IPD pre	IPD post	BOWSTRIN GING	Rupture at 10 N application
IInd	0/80	0/75	0/120	0/115	0/90	0/80	2mm	No
IIIrd	0/80	0/75	0/110	0/105	0/90	0/80	6mm	No
IVth	0/100	0/100	0/90	0/90	0/90	0/80	3mm	No
Hand #6	MF pre	MF post	IPP pre	IPP post	IPD pre	IPD post	BOWSTRIN GING	Rupture at 10 N application
IInd	0/70	0/80	0/120	0/110	0/45	0/65	2mm	No
IIIrd	0/70	0/90	0/90	0/100	0/45	0/50	6mm	No
IVth	0/90	0/115	0/110	0/100	0/65	0/90	3mm	No

Hand #7	MF pre	MF post	IPP pre	IPP post	IPD pre	IPD post	BOWSTRING	Rupture at 10 N application
IInd	100	100	120	120	85	80	2mm	No
IIIrd	100	95	120	110	85	75	6mm	No
IVth	110	105	130	120	100	95	3mm	No
Hand #8	MF pre	MF post	IPP pre	IPP post	IPD pre	IPD post	BOWSTRIN GING	Rupture at 10 N application
IInd	0/90	0/90	0/110	0/110	0/75	0/60	2mm	No
IIIrd	0/100	0/85	0/110	0/105	0/105	0/60	6mm	No
IVth	0/110	0/95	0/115	0/115	0/75	0/50	3mm	No

Comparing the degrees of flexion before and after reconstruction, we checked if there were statistically differences for type of reconstructive techniques and we obtained for Triple loops reconstruction not significative difference in MCP flexion (p=0.1), neither in DIP flexion (p= 0.45), significative PIP flexion (p=0.02). For reconstruction with FDS slip we got not significative difference in MCP flexion (p=0.45), neither in PIP flexion (p= 0.26) or DIP flexion (p=0.06). For reconstruction with pulley venting we got not significative difference in MCP flexion (p=0,41), neither in DIP flexion (p= 0.36), significative PIP flexion (p=0.007). The TAM and Strickland test also didn't show statistically significative difference for FDS strand and venting techniques, while showed significative result in tendon loop technique (p=0.05) (Table II).

Table II: *Tam and Strickland test before and after A2 pulley reconstruction*.

	FREE TENDON LOOP				FDS SHEET				VENTING			
	STRICKLAND		TAM		STRICKLAND		TAM		STRICK LAND		TAM	
HAND N°	PRE	POST	PRE	POST	PRE	POST	PRE	POST	PRE	POST	PRE	POST
1	57.1	57.1	58.5	58.5	82.8	82.8	77.3	77.3	48.5	48.5	47.1	47.1
2	94.2	60	88.63	62.2	77.1	85.7	77.3	90.57	100	108.5	100	115.1
3	105.7	97.1	103.7	98.1	122.8	94.2	118.8	94.3	108.5	88.5	113.2	94.3
4	94.2	88.5	88.6	83.0	77.1	71.4	77.3	73.5	91.4	85.7	94.3	90.5
5	120	111.4	109.4	101.8	114.2	105.7	105.6	98.1	102.8	97.1	105.6	101.8
6	94.2	100	88.6	96.2	77.1	85.7	77.3	90.5	100	114.2	100	118.8
7	117.1	114.2	115.1	113.2	117.1	105.7	115.1	105.6	131.4	122.8	128.3	120.7
8	105.7	97.1	103.7	98.1	122.8	94.2	118.8	94.3	108.5	94.3	113.2	98.1
p-value		0.05		0.07		0.07		0.2		0.2		0.3

At 10N load, 23-pulley did not break out, the only broken pulley was the 1 reconstructed with the pulley venting techniques which got the bowstringing of 2mm.

DISCUSSION

The Pulley system reconstruction is a fundamental aspect for a successful reconstruction of the flexor tendon injury (16, 17). The muscular belly of flexor profundus has a maximum shortening capability, because their excursion is constant, the pulley system is charged with maximizing the tendons' ability to generate flexion. Intact pulleys prevent tendon translation to palmar side. If the pulley is resected, the tendons displace volarly, and the maximum range of motion in flexion is decreased.

Reconstruction of flexor tendon pulleys is an important problem for the hand surgeon. Most pulley ruptures can be successfully treated conservatively with full return to preinjury activity. When surgery is necessary, pulley

reconstruction results in the best outcomes. Nowadays there is still not actual evidence of which techniques is the best option. In addition to the pulley reconstruction techniques studied in this study, other techniques such as the Kleinert/Weilby technique that is a technique involving weaving a tendon through the ‘‘always-present fibrous rim’’ of the pulley being reconstructed. Usually is used the tail of the superficialis tendon, but if not available, a tendon graft may be used instead. This technique has the advantage that it affords the surgeon good control over setting tension in the reconstructed pulley (18). The Karev—belt-loop is a technique that performed two transverse incisions in the volar plate and sliding the flexor tendon through the so called ‘‘belt loop’’ formed between the two incisions. Because the tendon must be passed through the belt loop, this technique can be used only in the presence of an adjunctive flexor tendon repair or tendon graft/implant and not for simple pulley reconstruction around an intact tendon (19). Moreover, the Lister’s technique harvests a segment of the extensor retinaculum, which is reversed and then passed around the phalanx (20). The major disadvantage of this technique is that a normal portion of the extremity must be violated to harvest the retinaculum. The main advantage is that the retinaculum provides a smooth gliding surface producing the lowest amount of resistance among the reconstructive techniques. In this study, we want to report the effect of three different types of A2 pulley reconstruction on cadaveric specimens and evaluate if there are or not difference in sense of degrees of flexion reached which can lead us to choose of one respect another.

The main purpose of this study was to compare the range of motion of each finger joint before and after the A2 reconstruction and to compare the biomechanics activity to evaluate which techniques gives the best result. Our study showed that Pulley venting has a different strength depending on how tight the suture is made, we showed that a laxity that leads on a bowstringing of 3 mm preserves the pulley, such as the strength goes on the A1 intact pulley, what make us think that in A1 and A2 reconstruction, a double pulley venting could not be the best choice, however, in the A2 isolated pulley reconstruction the techniques showed similar results through the evidence of not significant difference among various construction tecniques. The modified Odobescu techniques gave worse result due to a more mm bowstringing because the distal part of the FDS strand left attached to his insertion on F2, while the tendon graft techniques gave results similar to the venting, with a lower grade of bowstringing and a better result for IPP joint flexion. Nishida et al., in a 1998 (21) compared different reconstructive methods intrasynovialand extrasynovial, and found out the best reconstructive method, from the point of view of friction, is to loop the pulley reconstruction around bone, as in Lister's or Bunnell's technique. The around-bone method is also the strongest reconstruction (22, 23). Considering our result, there is not an evidence that one techniques is better than another, the only significative result is about TAM score in tendon loop repair, that appears in our experience as the most valuable technique. Considering that the tendon graft requires the presence of palmaris longus tendon, pulley venting can be considered like a solid choice for A2 reconstruction in that 10-15% of people who does not have the palmaris longus. We also showed that the flexor digitorum superficialis reconstruction is not a reliable procedure, although the easy execution and the small tendon sacrifice.

The limit of the study is due to the cadaveric experiments, in which is not possible to evaluate the follow up; there is no possibility in cadavers to evaluate the effectiveness of bulky reconstruction such as the tendon graft; and it’s not possible to evaluate if a continues stimulation due to the tendon glidingcan bring to pulley rapture, in particular in the pulley vented.

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Original Article

EFFICACY OF CAPSULAR REPAIR IN PARTIAL HIP HEMIARTHROPLASTIES: OUTCOME OF 100 CONSECUTIVE CASES

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ABSTRACT

In the last years, bipolar hemiarthroplasty has become the gold standard for the treatment of femoral neck fractures in elderly. The role of joint capsule as a passive stabilator of the hip and the importance of its reconstruction after hip replacement is known in the postero-lateral access as one of the key points for the success of the surgery procedure, due to its role as a protection factor from post operative dislocations. The lateral direct approach is burden of less episodes of hip dislocation, nevertheless there is no evidence of the possibility of capsular reconstruction after this procedure. The aim of this study is to compare the incidence of post-operative hip dislocations in patients underwent to bipolar hemiarthroplasty procedure with lateral direct approach with or without capsular reconstruction. We retrospectively analysed all patients underwent to hemiarthroplasty for a femoral neck fracture in our institute from July 2021. Inclusion criteria were bipolar hemiarthroplasty intervention, lateral direct approach to the hip, minimum follow up was 1 year. Incidence of post operative dislocation and the necessity of reintervention have also been calculated. 100 hip hemiarthroplasty with lateral direct approach were performed in patients with medial femoral neck fracture (64 women and 36 men). The mean age is 82.4 years (65-96). The mean duration of the operation was 79 min (20-150). In 50 cases the capsule was preserved (Group B: capsulotomy + capsulorrhaphy), while in the other 50 cases (Group A) only selective capsulectomy was performed. The groups are homogenous in terms of age, sex, duration of surgery and type of implant (monobloc vs modular stem, neck, biarticular cup, cementation ant head size). During the study period under analysis, there were 1 cases of post-operative dislocation, in the group of patients in which capsulorrhaphy was not performed and it was equipped with a modular stem. The dislocation occurs in 75 days; in this case a reduction manoeuvre of the new implant was performed which proved unsuccessful, since the residual instability of the implant made it necessary to undergo partial revision with a dual mobility cup, but leaving the stem and neck in situ.

Articular capsule plays a fundamental role in hip hemiarthroplasty performed also with lateral direct access, resulting in a further protective factor against post-operative dislocations.

KEYWORDS*: hemiarthroplasty, capsular repair, capsulorrha*

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INTRODUCTION

In the last years, bipolar hemiarthroplasty has become the gold standard for the treatment of femoral neck fractures in elderly. This population has been increased since the end of the XX Century and is estimated that its number will even further increase in the next years (1).

The goal of the treatment is to restore the articular function, to reach the pre injury activity level in the early post operative period and to reduce complications due to prolonged bed-rest and the consequent mortality rate (1).

According to NICE guidelines there is no clinical evidence in the choice between total hip arthroplasty and hemiarthroplasty (HA) in displaced femoral neck fractures (Types III and IV according to Garden classification) in elderly (2). In fact, although some studies showed that total hip arthroplasty has better clinical results measured using Harris Hip Score than HA beyond 2 years, clinical knowledges and experiences demonstrate that the long-term outcomes considered in the health economic model were similar to HA (3).

Hemiarthroplasty is usually recommended for patients > 80 years or those who have a limited predicted life expectancy due to lower peri-operative blood loss and lower dislocation rates during the first year after surgery (linked to the use of larger bipolar cups that allows an increased jump (4). Despite the low incidence, HA dislocation is the most frequent complication and consequently the main concern (5). It is showed by literature that this risk is independent from the surgical approaches but it has been demonstrated that joint capsule plays a key role in the prevention of dislocations (6).

In fact its role as a passive stabilator of the hip and the importance of its reconstruction after hip replacement is known in the postero-lateral access as one of the key points for the success of the surgery procedure. On the other hand the lateral direct approach is burden of less episodes of hip dislocation, nevertheless there is no evidence of the possibility of capsular reconstruction after this procedure.

The aim of this study is to compare the incidence of post-operative hip dislocations in patients underwent to bipolar hemiarthroplasty procedure with lateral direct approach with or without capsular reconstruction.

MATERIAL AND METHODS

We retrospectively analysed all patients underwent to hemiarthroplasty for a femoral neck fracture in our institute from July 2021 to December 2021. Inclusion criteria were bipolar hemiarthroplasty intervention, lateral direct approach to the hip, minimum follow up was 1 year. Incidence of post operative dislocation and the necessity of reintervention have also been calculated.

All patients were operated by skilled orthopaedic surgeons. Duration of surgery, size of the components, age and sex of the patients were noted. We further determined incidence of post operative dislocation and the subsequent treatment (closed reduction vs open revision).

RESULTS

The study cohort consisted of 100 patients (64 women and 36 men) who underwent hip hemiarthroplasty using the lateral direct approach for medial femoral neck fracture. The average age of the participants was 82.4 years, ranging from 65 to 96. The average duration of the operation was 79 minutes, with a range of 20 to 150 minutes. Among the participants, the capsule was preserved in 50 cases (Group B: capsulotomy + capsulorrhaphy), while in the remaining 50 cases (Group A), only a selective capsulectomy was performed. Thus, the study encompassed two distinct groups: Group A, which underwent selective capsulectomy, and Group B, which underwent capsular preservation (capsulorrhaphy) (Table I).

A senior surgeon, skilled in replacement surgery, performed all the hemiarthroplasty in Group A, while the interventions characterized only by selective capsulotomy (Group B) were conducted by an heterogeneous team of skilled surgeons.

Among 100 cases, a monobloc stem was utilized in 43 cases, with 20 cases in Group A and 23 cases in Group B. Conversely, the remaining 57 cases employed a modular stem, with cases in Group A 30 and 27 cases in Group B. As described by Solarino et al. in previous literature, in the hybrid implants, a distal cement restrictor was used, the medullary canal was cleaned with saline lavage and an injection gun was employed, together with digital pressurization of the cement (4).

Among the cases with a modular stem, 53 had a short straight neck (27 in Group A and 26 in Group B), 25 had a long straight neck (13 in Group A and 12 in Group B), 18 had a long retroverted neck (8 in Group A and 10 in Group B), and 4 had a short retroverted neck (2 in Group A and 2 in Group B).

All the heads used in the study were made of alumina material. Among the cases, 77 utilized heads with dimensions of "28" (37 in Group A and 40 in Group B), 20 cases used heads with dimensions of "22" (10 in Group A and 10 in Group B), 2 cases used heads with dimensions of "32" (all in Group A), and 1 cases used heads with dimensions of "36" (in Group A). Additionally, the stem was cemented in 87 cases, with 67 cases in Group A and 20 cases in Group B.

Table I. *Intraoperative factors*.

Intraoperative factors		**Group A (n=50)**	**Group B (n=50)**
Surgical approach		Direct lateral	Direct lateral
Sex (n)			
	Male	22	19
	Female	28	31
Age		82,7 (58-101)	81,92 (55-100)
Mean duration of surgery		78	80
Type of stem			
	Monobloc	20	23
	Modular	30	27
Neck			
	Short straight	27	26
	Long straight	13	12
	Short retroverted 8°	2	2
	Long retroverted 8°	8	10
Head			
	Short (-2,5mm)	27	26
	Standard (+0mm)	28	26
	Long (+2,5mm)	5	8
Biarticular Cup		46 (42-57)	46 (42-57)

The average dimensions of the biarticular cup were 46 mm, ranging between 42 and 57 (Table I). In the years taken into analysis, 1 cases of post-operative dislocations were registered, occurring in group A and it was equipped with a modular stem. This case occurred 75 days after surgery to a lady of 87 y.o. who referred an atraumatic event, while she was trying to sit on her sofa. The size of the components were: a cemented modular stem, a long retrograde (8°) neck, head of 28mm short and a bipolar cup of 44 mm (Table II).

Table II. *Features of dislocated implant and data of the patient.*

Group	**Patient A**
Surgical approach	Direct lateral
Sex (n)	F
Age	87
Duration of surgery	60min
Type of stem	Modular cemented
Neck	Long retrograde 8°
Head (mm)	28; -2.5
Biarticular Cup (mm)	44
Time of dislocation (days after suergery)	25
Reducible by closed manouvres	yes
Stable after reduction manoeuvre	no

DISCUSSION

Hip fractures are a significant health issue in the elderly population, with a high rate of morbidity and mortality (7). Hemiarthroplasty is a commonly used surgical option for the treatment of femoral neck fractures in the elderly population, and the use of a bipolar cup turned out to be an effective addition in the improvement of clinical outcomes (8-10).

Despite its effectiveness, post-operative dislocations can occur, leading to revision surgery and additional related complications. There are many variables that may contribute to an increased risk of post-operative dislocation; these include components type and size of the stem, the neck and the bipolar cup, along with the use of cement and repair of the capsule.

It is described by one of our authors that modular stems are frequentely used by young surgeons and residents to make an easier learning curve (8).

This study aimed to compare the incidence of post-operative hip dislocations in patients who underwent bipolar hemiarthroplasty procedures with a direct lateral approach.

Nowadays, the role of surgical approach that could lead to a major or minor risk of dislocation is debated. In the literature, lateral and posterior surgical approach are the most used for HA and BHA and between them posterior approach has showed better clinical and functional outcomes (2). But on the other side lateral approach has been characterized by lower dislocation rates (9). A cohort study by Jobory et al. in 2021 confirmed that direct lateral approach is already considered a safer option for reducing the risk of post-operative dislocations compared to the posterior approach, with a dislocation rate of 2.7% compared to 7.2% of the latter (10).

On the other hand, Gaurlich et al. in their study observed no differences between the two approaches in terms of dislocation rates (5, 6).

Talking about capsular preservation, there is a lack of studies that investigate the impact of capsular repair during hemiarthroplasty using the direct lateral approach.

The importance of capsular repair is showed in a review of the literature published by Miranda et al. in 2021(11). In their study, the authors showed that the dislocation rate after capsular repair in THA is lower dislocation than the one after capsulotomy (11). In addition, these data changed over different approaches: in the anterior approach the dislocation rate in case of capsular repair was 0.69% against 3.7% in capsulotomy group, in the lateral approach was 0.64% against 3.89% and in the posterior approach was 0.64% and 2.4% respectively (11).

Notably, this study marks the first in vivo investigation about capsular preservation in HA using lateral direct approach. In literature this aim as primarily been studied through cadaveric studies.

In 2015, researchers have measured the mean peak torque required to dislocate hip joints that underwent hemiarthroplasty through a direct lateral approach with capsular repair and compared it to hip joints also treated with HA via direct lateral approach but only undergoing capsulectomy. The “capsule repaired” group presented a mean peak torque at dislocation of 22.96 Nm at a mean angle of 94.9°. In the “capsule not repaired” group the mean torque at dislocation was merely 5.6 Nm at a mean angle of 57.5°. Both the evaluated parameters, mean torque applied at dislocation and mean angle, display statistical significance and support the proposition that surgical repair of the capsule provides a significant increase in the mean peak torsional forces, responsible for anterior dislocation of the hip (12).

The study yields valuable information that we couldn't otherwise obtain from an in vivo study. However, although insightful, it is important to acknowledge the limitations associated with cadaveric studies. The results should be interpreted with caution due to the inherent biomechanical disparities between live tissues and cadaveric specimens.

Our study holds significant importance as it seeks to address a critical gap in the existing literature by attempting to overcome the lack of in vivo studies on the subject matter, with the goal of minimizing the morbidity and complications commonly associated with hip fractures and their surgical interventions.

The study at hand comprised a cohort of 100 patients who underwent bipolar hemiarthroplasty using the lateral direct approach to treat medial (intracapsular) femoral neck fractures. The patients were divided into two groups: Group A, which underwent selective capsulectomy, and Group B, which underwent capsular preservation (capsulorrhaphy).

The study results indicated that, over a five-year period, 1 case of post-operative dislocations was reported, occurred in Group A, the group that did not receive capsular preservation, and it was equipped with a modular stem. The timing of dislocation was 75 days, and the age of the patients was 88 year. In this case, a reduction maneuver of the new implant was performed, which proved unsuccessful, necessitating partial revision with a dual mobility cup, leaving the stem and neck in situ. There is still concern about the management of HA dislocation, because the are no guidelines in literature.

While in our experience, the only case of dislocation demonstrated a failure in closed reduction, the studies published by Gill et al. (13) and Falsetto et al. (3) showed that revision surgery was needed only in 50% of dislocations. The statistical analyses of the study showed no statistically significant differences between the two groups with respect to age, sex, duration of intervention, cement use, and dimension of the head.

The limits of our study include the small sample and consequently the short period of follow up. In addition, it may be matter of discussion the fact that while in group A HA were performed by different skilled surgeon, only one senior surgeon was the author of the HA in group B.

CONCLUSIONS

Capsular restoration in bipolar hemiarthroplasty for femoral neck fractures is an additional stabilization to the hip joint and it may be considered a crucial factor in reducing the incidence of post-operative dislocations.

Despite the small sample size and significant variability in implant type, our study highlights the importance of exploring the feasibility of repairing the capsule during hemiarthroplasty procedures using a direct lateral approach. Further researches with larger cohorts and longer follow-up periods should be conducted to verify our findings. Ultimately, such knowledge could lead to improved outcomes for patients undergoing hip hemiarthroplasty.

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Original Article

NON-UNION OF TIBIAL SHAFT FRACTURE: FRACTING SCORE ANALYSIS IN A RETROSPECTIVE MULTI-CENTER STUDY

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ABSTRACT

Delayed union, malunion and non-union are serious complications in the healing of fractures. Predicting the risk of non-union before or after surgery is challenging. We analyzed FRACTING score for each patient. The aim of this study was to find out if this score is accurate to predict this complication.

We collected tibial shaft fractures undergoing surgery from January 2016 to December 2020 in three different trauma hospitals. In a retrospective multicenter study, we considered only fractures treated with intramedullary nailing, calculating FRACTING score at the time of definitive fixation.

Of the 130 patients enrolled, 89 patients (68.4%) healed within 9 months and were classified as union, 41 patients (31.5%) healed in more than 9 months or underwent other surgical procedures and were classified as non-union. For each patient, FRACTING score was calculated, and based on the clinical outcome the score was compared in a statistical analysis.

FRACTING showed a good performance predicting the non-union risk. From our data is clear that male gender and greater age is a risk factor for non-union.

KEY WORDS: *trauma, bone, tibial fracture, nonunion, scores, prediction model.*

INTRODUCTION

Bone fracture healing is one of the most important and debated issues in olden and modern orthopedics. "Pseudarthrosis" (the Greek stem "pseudo-" means false and "arthrosis" means joint) is nowadays a less common word than "delayed union" and "non-union", which constitute two variations of it. Although in the literature there are a lot of different definitions (1), we speak of a delayed union when the fracture healing time does not correspond with period of time expected for a specific site and type of fracture. Instead, "non-union" is the failure of a fracture to heal after twice the period of healing time (which usually takes at least 6 months after trauma).

Non-union is currently defined (according to the Food and Drugs Administration - FDA) as a fracture older than nine months that presents no signs of healing in the last three months (2). Conversely, Brinker et al. define non-union as a fracture that, in the opinion of the treating physician, has no possibility of healing without further intervention (3).

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Delayed union or non-union represents one of the most challenging complications for modern orthopedics. Among the long bone fractures, non-union rate is estimated to be between 5% and 10% (4).

However, these data could raise in the next future because of the increasing high-energy trauma and the improvement of Basic Life Support (BLS) techniques. These types of trauma often involve diaphyseal fractures of several limbs, serious muscle and tendon injuries as well as causing damage to the parenchymal organs (4, 5).

A recent population-based study from Scotland estimated the incidence of non-union at 13 per 1000 pelvis and femur fractures per year, 30 per 1000 humerus fractures per year, and approximately 55 per 1000 tibia and fibula fractures per year (6). The management of these long fractures is complex, and the risk of mal-union, delayed union and non-union remains high. That contributes to considerable patient disability, reduced quality of life, and significant treatment costs (5).

Naturally, delayed union and non-union (like fracture healing) are multi-factorial events, so their complications are not easy to predict. Many risk factors contribute to non-union. Calori et al. identified gender, age, diet, diabetes, osteoporosis, muscular mass, smoking and alcohol habits, nonsteroidal anti-inflammatory drugs (NSAIDS) use, fracture personality, type of fracture, exposure, infection, multiple fractures as parts of this multi-factorial events (7).

Nevertheless, some studies in the literature trying to foresee the risk of non-union with some different score, calculated after the surgical treatment.

These scores aim to quantify non-union high-risk patients (8-10). We analyzed only FRACTING score, in order to find out if this score is accurate to predict the risk of non-union in tibial shaft fracture. The aim of this study was to evaluate in a group of patients, the risk of non-union after the surgical treatment of tibial fracture.

MATERIAL AND METHODS

We retrospectively reviewed all consecutive cases of tibial shaft fractures undergoing intramedullary nailing surgery from January 2016 to December 2020. We collected data from three different hospitals: Azienda Ospedaliera Universitaria delle Marche (via Conca 71, Ancona (AN), 60126, Italy), Ospedale Carlo Urbani Jesi (via Aldo Moro 52, Jesi (AN), 60035, Italy) and Azienda Ospedali Riuniti Marche Nord, Pesaro (piazzale Cinelli Carlo 1, Pesaro (PU), 61121, Italy).

All patients gave their informed consent at the enrolment and were included in a retrospective observational database. Both pre- and post-operative data, including sex, age, type of surgical procedure, and others more were collected from the hospital database and patient medical records. Declaration of Helsinki and Guidelines for Good Clinical Practice were applied.

We included open and closed tibial no-articular fractures (42-A-B and C, 43-A) according to AO/OTA classification (11) in patients older than 18 years. We excluded patients that reported articular fractures, periprosthetic fractures, open fractures IIIC according to Gustilo et al. classification (12) as well as patients with active neoplasia, doubt of pathological fracture and genetic disorders with bone involvement (i.e. Paget, Osteogenesis imperfecta). Pregnant women and patients younger than 18 years were excluded. Patients who underwent amputation or who died because of complications related to the trauma were also excluded from the study. We excluded the polytrauma patients according to the definitions reported in the literature (13, 14).

To obtain a homogeneous sample we had to value only the tibial fractures treated by intramedullary nailing. The patients underwent clinical examination and bi-projective X-Rays to assess the type of fracture. Tibia FRACTure prediction healING days (FRACTING) score was applied after the definitive fixation.

This score can be used both to predict fracture healing time-span, both to identify prolonged healing risk patients immediately after surgical treatment. FRACTING score was validated in a prospective, multicenter, observational study (9). FRACTING score parameters include age, malnutrition, smoking status, diabetes, use of NSAIDs, fracture exposure grade, location (diaphysis, metaphysis or epiphysis), synthesis device (nail, plate, external fixator, angular stability plate), instability, misalignment (>5°), bone graft use, type of fracture, loss of bone substance, bone diastasis (> 2mm), surgery duration (> 2 hours), cast and blood loss before and after treatment (Hb<10g/dl). The values of the score ranged from 3 to 18.

FRACTING score is able to predict fracture healing time in five-time intervals: ≤3, 4, 5, 6, and >6 months from trauma. All the patients underwent follow-up for at least 12 months. We collected patients' data about age, gender, type of fracture, surgery approach and pseudoarthrosis scores (Table I).

Table I. *Example of pre-release data collection.*

PATIENT	AGE AT THE SURGERY	SIDE	GENDER	TYPE OF FRACTURE (AO/OTA)	FRACTING SCORE	UNION	NON-UNION	FOLLOW-UP (months)
1	22	RIGHT	M	4.2C	9		X	18
2	28	LEFT	M	4.2A	6	X		12
3	82	RIGHT	M	4.2A	6		X	12

We used FDA definitions of non-union (2). The end-point of fracture healing was radiological and usually clinical: the patient can handle full weight-bearing without pain. The most common clinical features used for the definition of non-union were pain over fracture site, pain during weight bearing and mobility at fracture site.

By the way, all the fractures underwent to reoperation were considered non-union as well, according to Brinker definition (3).

All the fractures were nailed within 21 days at most from the injury (range 1-21 days). 14 patients with open fractures, soft tissue wide injuries, or life-threatening polytrauma were treated according to the damage control orthopaedics (DCO) principles and received a temporary external fixation.

Conversion to definitive surgery was performed as soon as soft tissue conditions allowed and when the patient overcame the immunodeficiency period after trauma (15). All nailing procedures were performed prior to antibiotic prophylaxis and with the patient in the supine position on a fracture table with fluoroscopic-guided imaging. The surgical technique was performed both by infra-patellar and supra-patellar incision according to the type of fracture and the surgeon's preference.

Tibial shaft was both reamed and unreamed, and a guide wire was used for all procedures. All nails were of the same brand and type, with different length and diameter. All the nails were locked with at least 2 proximal and 2 distal locking screws. In both tibial and fibular fractures, fibular was never fixed. There were no intraoperative complications. Patients were weight-bearing as tolerated postoperatively.

Participating surgeons did not offer stimulation modalities to promote bone growth (such as ultrasound and electrical stimulation) during the follow-up.

RESULTS

A total of 174 patients with tibial shaft extra-articular fractures surgically treated were assessed for eligibility, 17 did not satisfy the inclusion criteria, 11 did not accept to be enrolled, and 16 were lost during follow-up. Finally, 130 patients with tibial shaft fractures were entered into the database and completed the follow-up (Fig. 1).

Overall, 23 patients (17.6%) had open fractures; 9 patients (6.9%) occurred loss of bone tissue. 109 patients (90.8% of fractures), sustained both bone fractures (tibia and fibula). According to AO classification, 64 (49.3%) of fractures were type 4.2A, 31 (23.8%) were type 4.2B, 17 (13.07%) were typed 4.2C, and 8 (6.6%) were 4.3A type.

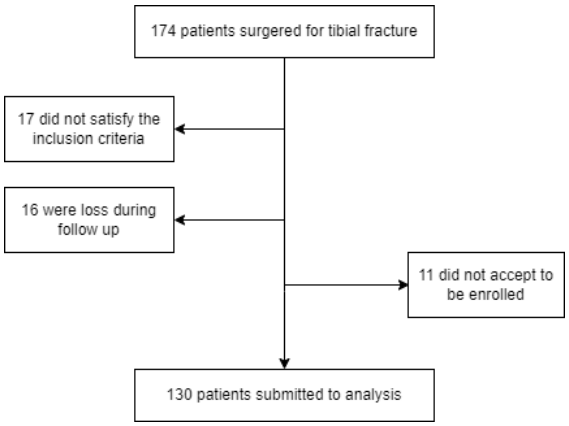

Fig. 1. *Case selection flow chart.*

Among the 130 patients with tibial shaft fractures, 89 (68%) healed within 9 months and were classified as Union, 41 fractures (31.5%) healed in more than 9 months or underwent other surgical intervention and were classified as Non-union.

The second surgery intervention included: nail dynamization, bone grafting, re-nailing, compression plating, and external fixation. Among the non-union group, male patients have a mean age of 45, with FRACTING score avg. 7.8 ± 1.8. Females patients, non-union group, instead have a mean age of 52, with FRACTING avg. 7.7 ± 2.1. Remember that the score is directly proportional to the risk of non-union.

Therefore, the higher the score, the lower the chances of healing the fracture. Scores owned their decision rule which depends on the threshold value. The cutoff value for FRACTING score was ≥ 8, as suggested by the authors.

Statistical analysis

The descriptive analysis is computed on following samples: the total size is first divided into gender (Female = 47; Male = 83); subsequently, each gender sample is divided into NON UNION and UNION.

Results for each sample size, as minimum and maximum value, median, IQR (interquartile range) and mean, are summed up in Table II.

Table II. *Descriptive statistics for scores and AGE on outcome of NON-UNION and GENDER.*

	Female (N=47)		Male (N=83)		Total
	Non Union (N=7)	Union (N=40)	Non Union (N=34)	Union (N=49)	
Age					
Min / Max	32 / 84	18 / 86	19 / 82	18 / 87	18 / 87
Med [IQR]	52 [43;55.5]	54 [42.2;62]	45 [28.2;57.8]	45.0 [27;60]	46.5 [34;60]
Mean ± St. Dev.	52.1 ± 16.4	52.5 ± 17.5	44.6 ± 17.9	44.5 ± 20.0	47.4 ± 18.7
Fracting					
Min / Max	5.0 / 10	1.0 / 10.0	5.0 / 10.0	2.0 / 9.0	1.0 / 10.0
Med [IQR]	8.0 [6.0;9.5]	5.0 [3.0;7.0]	8.0 [6.0;9.0]	4.0 [3.0;7.0]	6.0 [4.0;8.0]
Mean ± St. Dev.	7.7 ± 2.1	5.2 ± 2.3	7.8 ± 1.8	5.1 ± 2.2	6.0 ± 2.4

As described, a non-union score assumes an integer value calculated as the sum of risk factors, clinical parameters, and/or demographic variables observed. From Table III, it turned out that the increasing age does not affect the percentage of non-union patients.

Table III. *Percentage proportion of patients grouped by age ranges.*

Age	Non Union	Union	Total
18 - 45	21 (34%)	41 (66%)	62 (48%)
46 - 60	13 (35%)	24 (65%)	37 (28%)
> 60	7 (23%)	24 (77%)	31 (24%)

By applying the decision rule of FRACTING score to the patients, which depends by cutoff value, the prediction is computed and results are listed as confusion matrix (Table IV, V), Then, the score performances were evaluated in order to compare the reliability of decision rule.

Table IV. *FRACTING - Confusion matrix.*

	Union	Non Union	Total
Predicted Union	77	15	92
Predicted Non Union	12	26	38
Total	89	41	130

Table V. *Score evaluation performance metrics results.*

Score	Sensitivity	Specificity	PPV	NPV	F-measure
FRACTING	63.41%	86.52%	68.42%	83.70%	67.00%

In Fig. 2, it is shown the distribution of scores values for non-union and union patients, grouped into three groups where the last group has values equal and greater than the cutoff.

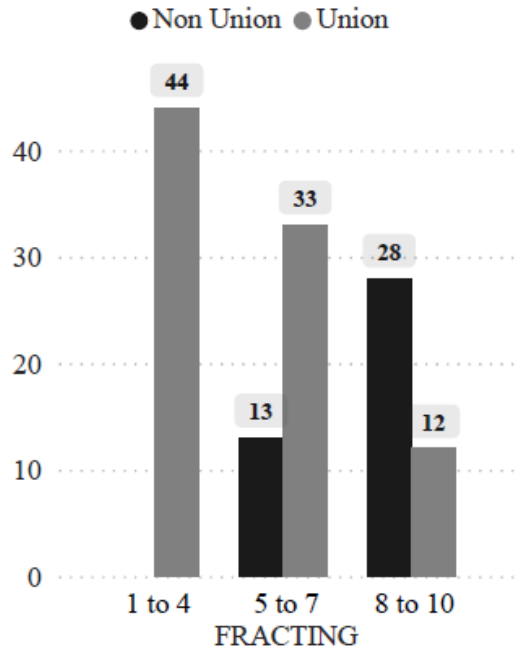

Fig. 2. *Histograms of score distribution.*

Considering the presence of class imbalance ratio (16) equal to 41,07% which determines a greater ease of predicting union patients, we have calculated Sensitivity, Specificity, Positive Predictive Value (PPV), Negative Predictive Value (NPV), and F-measure.

DISCUSSION

Malunion or non-union of long bones are one of the most challenging complications for the orthopaedic surgeons. This condition involves residual pain, lameness, use of aids for walking and the inability to lead a normal lifestyle, thus causing a great impact on the quality of life (17). Moreover, the healing time of tibial fractures are very variable for each person and may be affected by many factors.

Among the long bones fracture, a comprehensive review of studies reported non-union rates of 0–12% in femoral fractures, 0–33% in humeral fractures, and 1–80% in tibial fractures (18). It is often necessary a second surgery to obtain healing. Reoperations included bone grafts, implant exchanges or removal for hardware failure. In case of infected non-union: irrigation, débridement and soft tissue coverage procedures.

Numerous clinical factors were documented as having a prognostic value for delayed bone healing or non-union of tibial shaft fractures. For this reason, there are some different non-union scores (19).

The FRACTING score was born to predict the time of healing of tibial fractures with parameters analyzed in a retrospective study, called ARRCO (Algoritmo Rischio Ritardo Consolidazione Ossea), and later validated in the prospective, multicenter observational study, called FRACTING (20).

This is certainly the score with more parameters considered, different for each type of surgery and internal fixation. Moreover, it considers clinical, patient-related, fracture-relates surgical, and peri-surgical parameters. Bhandari et al in an observational study had identified that a set of three simple prognostic variables (open fracture, transverse fracture, and postoperative fracture gap) that can assist surgeons in predicting reoperation following operative treatment of tibial shaft fractures (21).

By the way, the presence of a large fracture gap and lack of cortical continuity after reduction is maybe the best single variable associated with delayed healing and non-union (19).

The topography of the fracture affects the fracture healing, according to the vascular anatomy and the different blood supply of the shaft. Santolini et al. divided the femur and tibial shaft into three zones, defined as zones of high, moderate, and poor vascularization (22). They argued that the tibial shaft vascularization is divisible into sections of thirds.

The upper third has a high degree of vascularization, the middle third has a moderate degree of vascularization, and the lower third has a poor degree of vascularization. Among the three scores, only the FRACTING score kept into account the localization of the fracture. Deep or superficial infections are significantly associated with tibial non-union (23, 24). FRACTING did not count infection, but other authors proposed a non-union prediction score at six weeks after surgery that included infection (25).From our results emerged that the male gender is a non-union risk factor.

Also the literature shows that male gender is a risk factor (4), maybe because males are more likely to suffer high-energy trauma for the type of sporting activity (26, 27). Smoking habit is associated with non-union in several studies (28, 29).

By the way, FRACTING score present smoking in own aims. Some drugs can involve fracture healing: not only NSAIDs (30, 31) and corticosteroid (32, 33), but also anticoagulant (34) and anticonvulsant (35). Indeed, FRACTING consider NSAID use as a conditioning factor.

The diagnostic accuracy demonstrated greater accuracy by FRACTING in low score values, this, could be explained not only for a wider range score but also because in our patient cohort, we consider only nail fixation, which potentially keeps a low score (FRACTING score assigned 3 points for external fixation and 2 point for plate and screw versus just 1 point for nailing).

The limitations of our study included the retrospective and the multicentric nature. When a multicenter study is conducted, especially in the surgical field, it is easy to have bias related to the surgeon's experience, surgical technique, postoperative treatment, and definition of healing. For example, there is no consensus about use of skeletal traction while waiting for surgery and allowing full or partial weight bearing after the surgery (36), and the score did not keep that in consideration.

Radiographic healing of the fracture was determined by the investigator based on his experience, clinical well-being and evidence of 3 out of 4 welded cortices. Moreover, there has been no utilization of any objective radiographic scoring to ensure fracture healing. In the broad panorama of leg fractures surgery, there are some additional variables that make standardization of population impossible.

These variables are: intramedullary canal reaming or not (37), fibular osteotomy vs fibular fixation vs no touch fibular fracture focus (38), the use of a Poller screw, and the time of wound closure. The use of local prophylactic antibiotic (39), like Antibiotic-Coated Nail (40) in open fractures could be a solution to prevent septic non-union.

Therefore, any intra-operative or post-operative treatments (like biophysical stimulation with pulsed electromagnetic fields), that promotes bone healing should be used (41).

CONCLUSIONS

Our multicenter study analized the predictive value of FRACTING score. From our data is clear that male gender and greater age is a risk factor for non-union. FRACTING score showed good reliability from statistical analysis. For this reason, we recommend the use of this predictive score in clinical practice, because it can change the surgeon's operative approach and the choice of adjuvant therapy (ultrasound, pulsed electromagnetic fields, coated nails, or application of growth factor).

In the future would be interesting to make a comparison of the scores cited by the recent literature, in order to find out which is more suitable to predict delayed union or nonunion.

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Original Article

NEW APPROACHES IN THE TREATMENT OF RHIZOARTHROSIS: RESULTS OF PROSTHETIC REPLACEMENT AND COMPARISON WITH TRAPEZIECTOMY AND SUSPENSION ARTHROPLASTY

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ABSTRACT

Rhizoarthrosis is a degenerative disease of the first carpometacarpal joint; it causes pain and functional limitation of the thumb that progressively worsens until it affects the whole hand. The diagnosis is both clinical and radiological and the treatment in the early stages is conservative. When nonoperative measures fail, surgery is the only chance. There are many surgical options in the management of thumb arthrosis. The aim of this study is to compare the clinical and functional outcome after a prosthetic replacement vs trapeziectomy and suspension arthroplasty. From January 2020 to June 2021, 18 patients with diagnosis of rhizarthrosis (Eaton's grade II and III) were recruited from our unit. Eight patients were treated with prosthetic replacement while ten patients with trapeziectomy and suspension arthroplasty. The parameters evaluated were the first closure force, the index thumb grip force, the mobility of the first ray with the Kapandji score and the ROM of all the joints of the first ray. The follow up was performed at 3, 6, 12 months. The group of patients undergoing prosthesis replacement showed a statistically significant difference both in the force of the thumb-index forceps and in the Kapandji score following the removal of the post-surgical splint. Comparing the groups at 6 months there was no statistically significant difference in strength, while there was a statistically significant difference in range of motion in favor of the prosthesis (9+-0.76) over trapeziectomy (7.38+-1.32). Comparing the groups at 12 months there was no statistically significant difference in range of movement, while there was a statistically significant difference strength in prosthetic group over trapeziectomy (10.37+-2.41). Our study demonstrated a significant improvement in mobility and index thumb gripper strength in patients undergoing prosthetic replacement compared to patients undergoing trapeziectomy. The latter remains a valid therapeutic option in patients in whom prosthetic replacement is contraindicated, with a considerable improvement in pain symptoms and thumb functionality.

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KEYWORDS: *rhizoarthrosis, osteoarthritis, trapeziometacarpal joint, surgical treatment, prosthetic replacement, trapeziectomy, suspension arthroplasty*

INTRODUCTION

Rhizoarthrosis is the second most frequent site of osteoarthritis in the hand after osteoarthrosis of the distal interphalangeal joints, affecting about 33-66% of women over 55 years of age with radiographic diagnosis, with pain symptoms in about 22% of the population over 50 years of age (1).Prevalence increases with age and at post-menopausal age in women, with an F:M ratio of 6:1. Rhizoarthrosis most commonly affects the nondominant hand joint. Despite the presence of radiographic signs frequently detected on radiographs, a fair proportion of patients will never develop symptoms such that treatment for the condition is required. In symptomatic cases, loss of function of the first ray results in up to 50% loss of function of the upper limb.

There is a close correlation between excessive laxity of the trapeziometacarpal joint and the subsequent development of arthrosis; increased laxity leads to incongruity of joint surfaces and thus cartilage and altered force transmission at the joint level (2, 3). Forces 13 times higher develop at the trapezius metacarpal joint than at the apex of the thumb, furthermore, compressive forces at the TMC joint increase 12 to 20 times during thumb-index clamping compared with the resting state (4). Ligamentous laxity is more common in women, even at a young age, and repeated overloading of a lax and subluxated joint explains the higher prevalence of rhizoarthrosis in women over 50 years of age. Comparison of bone and joint morphology between the two sexes showed no significant differences, implying that the increased risk depends on physiological and functional factors. The hypothesis for this ligamentous laxity centers on the role of the hormone relaxin, which in women plays a key role in pelvic ligament laxity during pregnancy and childbirth; it is likely responsible for generalized laxity via an extracellular matrix metalloprotease signaling pathway (5).Pathologies involving soft tissues with ligamentous laxity also predispose to rhizoarthrosis, such as Ehlers-Danlos syndrome; just as a higher Beighton score is associated with increased TMC mobility and increased risk of developing TMC arthrosis (6).

The most important ligaments in stabilizing the TMC joint are the anterior oblique ligament, the posterior oblique ligament, and the dorso-radial ligament. Previous studies placed more emphasis on the anterior oblique ligament, which in more recent anatomical studies has been found to be less important in stabilization than the posterior oblique ligament because of its less structural organization and less thickness.

The anterior oblique ligament with its intimate relationships with the Tenar eminence muscles, rich in mechanoreceptors, thus contributes to the stabilization of the joint. The tendons of the long thumb abductor, the long and short thumb extensors, and the long thumb flexor provide extrinsic stabilization. Intrinsic muscles that stabilize the first ray joints include the thumb short abductor, thumb adductor, thumb short flexor, and thumb oppositor (1). Prior trauma and fractures of the base of the I MC may predispose to rhizoarthrosis, as well as jobs with repetitive and traumatic use of the thumb increase the risk 12-fold compared with the general population (7).

Inflammatory arthritis, first among them rheumatoid arthritis, are predisposing conditions. Approximately one third of patients with rheumatoid arthritis will develop clinically manifest rhizoarthrosis.

The pain is predominantly activity-related, especially when using the index thumb clamp. It is a localized pain at the base of the first ray, often radiating to the Tenar eminence and toward the first metacarpophalangeal. With the progression of the pathology there is an increase in pain symptoms to the onset of pain even at rest, atrophy of the Tenar eminence muscles, subluxation of the first carpometacarpal joint, loss of the strength of the index thumb pincer, decrease in the range of motion of the first ray, and related loss of hand function. With further progression comes the inability to abduct the thumb, with complete loss of thumb opposition with the other fingers of the hand, until thumb collapse and a swan-neck deformity with adduction of the first metacarpal and hyperextension of the first metacarpophalangeal (8).

The conditions most frequently associated with rhizoarthrosis are carpal tunnel syndrome, STT arthrosis, De Quervain's tenosynovitis, and trigger thumb. Of these, the most frequent is carpal tunnel syndrome, which is associated with rhizoarthrosis in 43% of cases, more frequent in women and diabetic patients. Inflammation of the trapeziometacarpal joint can extend to the carpal tunnel, and conformational changes at the carpal bones reduce the tunnel space by compressing the median nerve.

MATERIALS AND METHODS

Our study includes 18 patients referred to the Department of Orthopaedics and Traumatology of the Bari Polyclinic, operated between January 26, 2020 and June 1, 2021. Patients were evaluated with a minimum follow-up period of 12 months. The sample includes adult patients of both sexes, diagnosed with Eaton's Grade II or III, with worsening symptomatology and functional limitations in daily activities.

At the first visit and follow-up visits, fist closure strength was measured with an electronic dynamometer and thumb-index pincer strength. In addition, the mobility of the first ray was assessed with the Kapandji score and an evaluation of the ROMs of all joints of the first ray was performed. During the pre-admission examination were collected the following data:

- Biographical data
- Pathologic history and any prior trauma
- Patient's comorbidities
- Staging Rhizoarthrosis
- Objective examination and strength testing

Data collected at follow-up visits:

- Follow-up RX
- Objective examination and strength testing

The study includes two groups, patients operated with prosthetic replacement and patients operated with trapeziectomy and suspension plastic.

Prosthetic replacement was performed with Touch prosthesis with truncated acetabular component to avoid possible component loosening during thumb opposition movements. The above prosthesis requires a preoperative study for bone stock assessment for implantation of the acetabular component. Postoperative immobilization was maintained for 15 days with an antibrachio-metacarpal cast, and physiotherapy was initiated upon removal of the cast.

Trapeziectomy with suspension plastic was performed with the technique according to Ceruso, using the thumb long abductor tendon, using the most dorsal bundle if the thumb long abductor had a tendon consisting of multiple bundles.

The bundle of the thumb long abductor tendon was attached to the radial flexor of the carpus. Postoperative immobilization was maintained for 4 weeks, followed by use of the first ray brace exclusively at night and physical therapy was initiated 4 weeks after surgery.

In order to reach a correct diagnosis, all patients underwent a careful objective examination and instrumental exams (x-ray in 3P and EMG only in cases of altered sensitivity in the median nerve territory).

In the objective examination we considered: grind test (rotation of the base of the first metacarpal by applying a force in axial compression), distraction test or torque test(performed by rotating the base of the first metacarpal while applying a force in axial traction), the thumb-index pincer, weakness in the grip (1, 9).

In this recent study, an association between grip weakness and TMC arthrosis was demonstrated, even in the absence of radiographic signs (10). It must also be added that the Grind test does not always correlate with the degree of arthrosis shown by the X-ray.

The clinical diagnosis is confirmed by radiographs, taken in the three planes (PA, lateral, oblique). In addition, stress projections of the joint can be performed to demonstrate joint laxity by performing a posteroanterior projection of both trapeziometacarpal joints while the patient forcefully pushes the distal and radial ends of the thumbs against each other (11). It must be remembered that the degree of radiological staging does not always correlate with symptoms. After a careful objective and instrumental examination, we classified our patients according to Eaton's 4-stage classification, which is based only on radiographic changes (Fig 1).

Staging	Radiographic Characteristics
Stage I	Normal or slightly widened trapeziometacarpal joint Normal articular contours Trapeziometacarpal subluxation (if present up to one third of the articular surface)
Stage II	Decreased trapeziometacarpal joint space Trapeziometacarpal subluxation (if present up to one third of the articular surface) Osteophytes or loose bodies less than 2 mm in diameter
Stage III	Further decrease in trapeziometacarpal joint space Subchondral cysts or sclerosis Osteophytes or loose bodies 2 mm or more in diameter Trapeziometacarpal joint subluxation of one third or more of the articular surface
Stage IV	Involvement of the scaphotrapezial joint or less commonly the trapeziotrapezoid or trapeziometacarpal joint to the index finger

Fig 1. *Eaton's Classification.*

Although the most widely used, Eaton's classification is flawed in that it correlates only moderately with clinical and treatment protocols and has substantial inter-observer and intra-observer differences.

Conservative treatment

In the first instance, the pathology is approached with treatment conservative, based mainly on modifying the activities in which the use of the first ray of the hand is prevalent, rest, use of the specific brace for rhizoarthrosis, strengthening of the Tenar eminence muscles and anti-inflammatory therapy (8). At the first visit the patient is generally in the acute phase with marked pain and functional impotence, which is why the continuous use of the brace for 3 weeks, combined with anti-inflammatory therapy, is suggested. Subsequently the brace will be used intermittently, during the heaviest activities or when the pain is most present, associated with kinesitherapy, activity modification and joint protection education, possible physical therapy with magnetotherapy. Recent studies have examined the possible benefits of therapy with molecules with chondroprotective action in repairing joint damage and slowing cartilage degeneration.

The two most promising molecules are glucosamine sulphate and chondroitin sulphate (12). Corticosteroid infiltrations can be performed with medium-term benefit on pain symptoms, with actual risks only for repeated infiltrations with damage to the joint surface and capsule (6). Only in Eaton stage I was a response noted in 100% of cases with regression of symptoms at 18 months, the response to corticosteroid infiltrations in stages II and III drops to around 46% (13). Hyaluronic acid infiltrations have demonstrated long-term efficacy, with a slight initial worsening at 4 weeks with an increase in pain symptoms, but with improvement in pain symptoms at 12 and 26 weeks, with improvement in grip strength of the first finger (14). New conservative treatments are being studied, with promising results at short- and medium-term follow-up. The use of autologous adipose tissue obtained by liposuction and processed to increase the cellular component has proved effective at 12-month follow-up (15).

PRP (platelet-rich plasma) infiltration has also shown promising results, with improvement in the VAS scale and function as measured by different scores, both at 3 and 6 months after administration (16).

If conservative treatment does not bring any benefit in about two months, there is a debate about the surgical treatment to be performed.

Surgical treatment

The primary indication for surgical treatment is no tolerable pain, not responsive to conservative treatment. There are various surgical approaches for rhizoarthrosis, including the
trapeziectomy with or without ligamentous reconstruction and plastic interposition (LRTI), trapeziometacarpal joint arthrodesis (TMA), arthroscopy with debridement, trapeziometacarpal prosthesis (17). The indication for the choice of surgical treatment to be performed depends not only on the experience of the surgeon, but also on the staging of the rhizoarthrosis.

In patients under 50 years of age, with a need for joint stability and high-impact work activities, it is useful to consider TMA, with the downside of the increased risk of re-intervention, the risk of pseudo-arthrosis and the reduced mobility of the first radius, with subsequent development of arthrosis of the STT (6).

In early-stage rhizoarthrosis with minimal joint damage, joint debridement and capsulorrhaphy has also been proposed, however evidence is still limited (18).

In more advanced stages but with isolated arthrosis at TMC, there is no clear indication of the surgical procedure to be performed. At this stage, prosthetic replacement with a trapeziometacarpal prosthesis has shown excellent results

with essentially equal long-term functionality but with an earlier return to activity and a shorter rehabilitation path compared to trapeziectomy (19). In advanced stages, with involvement of the entire trapezius surface and diffuse arthrosis at the STT, treatment options mainly include trapeziectomy, with possible additional surgical management such as stabilisation with k-wires or tendon interposition (Fig. 2) (20).

Fig 2. *61-year-old woman before and after prosthetic replacement for Rhizoarthrosis (Eaton 3).*

RESULTS

The dependent continuous variables of the two groups were expressed as mean and standard deviation or as median and interquartile range and compared using T test or Mann Whitney U for non-normal distributions. A significance threshold $p<0.05$ was considered in all comparisons.

In the study, 18 patients referred to the Orthopaedics and Traumatology of the Bari Polyclinic. The patients were assessed pre-operatively after conservative treatment and post-operatively, conservative and in the post-operative period, comparing the two surgical techniques with the pre-operative parameters and comparing them with each other.

No differences emerged in the two groups with regard to age, gender, pre-operative assessment of index thumb force and first ray mobility measured with the Kapandji score. In the prosthetised patients at the removal of immobilisation, a statistically significant difference emerged with a lower force in the operated group and a higher Kapandji score in the operated group.

In patients operated with suspension plastic upon removal of the immobilisation, a statistically significant difference emerged with lower levels of index thumb forceps, with a statistically significant improvement in ROM as measured by the Kapandji score. When comparing patients operated with the two surgical techniques at the removal of immobilisation, a statistically significant difference emerged in both index thumb clamp strength and Kapandji score in favour of prosthetic replacement (Fig. 3).

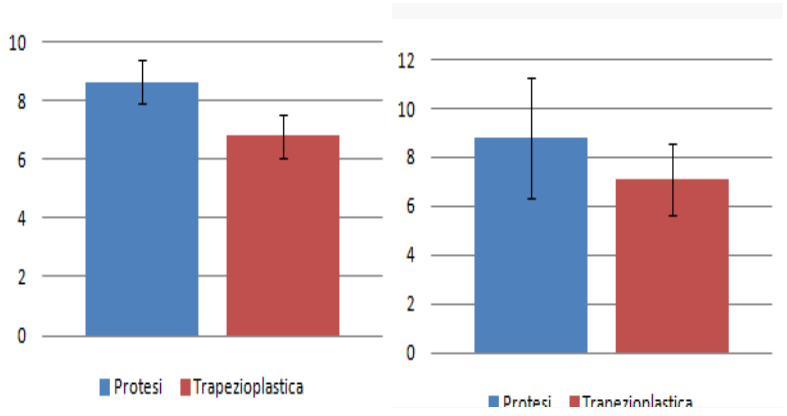

Fig. 3. *Kapandji score and strength in two groups upon removal of immobilisation.*

At the 6-month follow-up in the prosthetised patients, a statistically significant difference emerged with a non-full recovery of strength, but increased mobility as measured by the Kapandji score.

At the 6-month follow-up in patients operated with suspension plastic there was a progressive recovery of strength, but with statistically significant values in favour of pre-operative, and a further improvement in range of motion with a statistically significant difference.

Comparing the two groups at 6 months there was no statistically significant difference in strength, while there was a statistically significant difference in range of motion in favour of prosthesisation (Fig. 4).

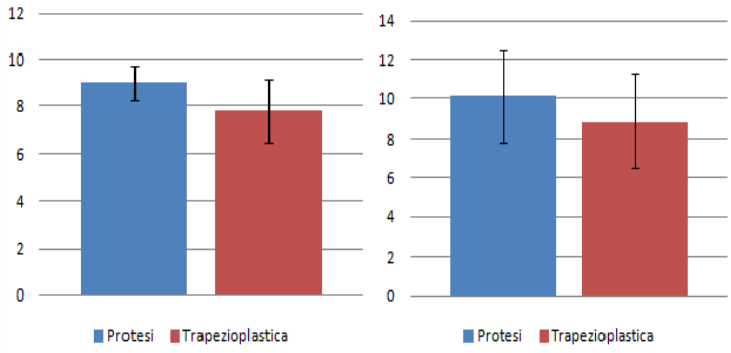

Fig. 4. *Kapandji score and strength in two groups at 6 months follow-up.*

At the 12-month follow-up in the prosthetised patients comparing pre-operative and post-operative values, mobility further improved with a statistically significant difference compared to pre-operative, and the recovery of strength was complete.

In the patients who underwent suspension plastic surgery, a further improvement in strength emerged at the 12-month follow-up, but a statistically significant difference remained in favour of the operated patients with regard to ROM as measured by the Kapandji score.

When comparing the two groups at 12 months, there was no statistically significant difference in range of motion, while there was a statistically significant difference in favour of the prosthetised patient group with regard to strength (Fig. 5).

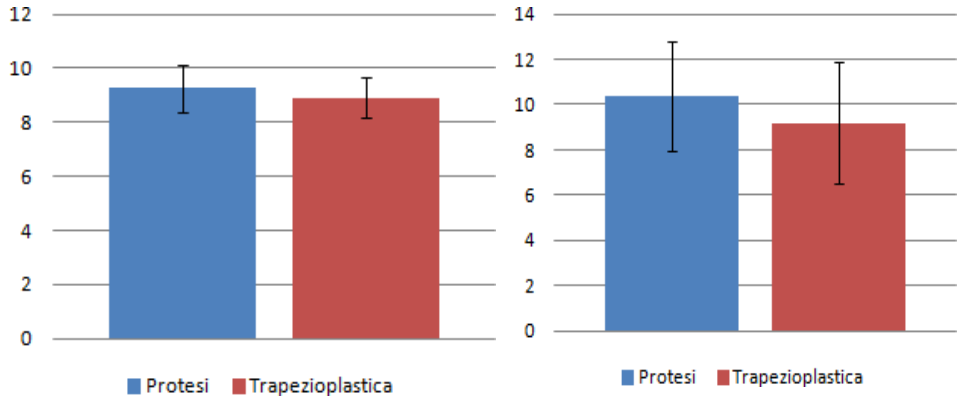

Fig. 5. *Kapandji score and strength in two groups at 12 months follow-up.*

Hand function was also assessed pre-operatively, at 6 and at 12 months with a modified DASH score using items that significantly involved the first ray.

There was a marked improvement in both groups with a statistically significant difference at 6 months and 12 months compared to pre-operative.

The difference was greater than 10 DASH score points, which is the smallest clinically significant difference visible to the patient. Comparing the DASH score between the two groups of operated patients, at 6 months there was no statistically significant difference, while at 12 months there was a difference in favour of prosthetic replacement, but with a net difference of less than 10 points, the smallest clinically significant difference visible to the patient (Table I-II).

Table I-II. *Difference in DASH score between the two groups at 6 and 12 months follow-up.*

Table I.

	Prosthetic replacement at 6 months (n=8)	**Trapeziectomy and suspension arthroplasty at 6 months (n=10)**
DASH score	20	21
Range interquartile	18-22	19-28

Table II.

	Prosthetic replacement at 12 months (n=8)	**Trapeziectomy and suspension arthroplasty at 12 months (n=10)**
DASH score	10	15
Range interquartile	8,5-13,5	12-20

DISCUSSION

In this study carried out in our Operating Unit it was confirmed the treatment course previously studied in the literature, with an initial conservative approach and a surgical approach as function deteriorated and symptoms worsened.

The patient sample was followed throughout the conservative treatment period, in which objective measurements of function and control X-rays were taken in addition to therapy. Subsequently, the patients underwent surgery for prosthesisation of the trapeziometacarpal joint and trapeziectomy with suspension plastic according to Ceruso.

They were subsequently followed up for 12 months. Objective examinations with objective measurements and control X-rays were performed.

What emerged from the study was the substantial improvement in the function of the joint following surgery, with a large improvement in range of motion. The improvement was mainly visible at the trapeziometacarpal joint, with maintenance or slight improvement of range of motion at the MCF and FI. This improvement was not reflected in other parameters; the strength of the thumb-index clamp and the strength of the other four fingers remained virtually unchanged.

Differences emerged between the two surgical techniques with regard to mobility and strength on removal of post-operative immobilisation, in favour of prosthetic replacement.

At follow-up at 6 months the group operated with suspension plastic had recovered strength but the gap in range of motion in favour of prosthesis was still marked. The DASH score was superimposable at 6 months. At one year, mobility was significantly improved in both groups, with total recovery of strength in the group operated with the Touch prosthesis and partial recovery of strength in the patients operated with trapeziectomy and suspension plastic, with a better DASH score in the group operated with the prosthesis.

CONCLUSIONS

The present study performed at our OU, comparing the two most common surgical approaches, demonstrated a significant improvement in DASH score, mobility and index thumb grip force in patients operated with joint prosthesization, more markedly than in the control group operated with trapeziectomy and suspension plastic, which remains a feasible procedure in patients in whom prosthetic replacement is contraindicated, with marked improvements in patient symptomatology and first ray function.

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Original Article

EVALUATION OF SURGICAL TIMING IN PATIENTS WITH SPINE METASTASES AND ACUTE SEVERE NEUROLOGICAL DEFICIT

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ABSTRACT

Spine metastases are one of the main causes of non-traumatic spinal cord injury (SCI). The acute onset of a neurological deficit in cancer patients with metastases makes it necessary to perform a rapid decompression of the vertebral canal, generally within 48 hours of the onset, to improve the clinical and neurological outcome, avoiding the onset of edema, venous congestion and secondary vascular damage. However, the optimal timing for performing this procedure is a matter of debate in the literature. The aim of our study is to evaluate whether an early surgical intervention in patients with vertebral metastases debut with acute neurological deficit influences the clinical outcome. The study includes a series of 40 patients with vertebral metastases and acute onset of neurological deficit, who underwent decompressive laminectomy and vertebral arthrodesis from September 2013 to March 2021. For each patient has been collected information relating to age, sex, of admission to the emergency room and time of surgery, level of the lesion, pre- and post-operative neurological function evaluated with the Frankel scale and histology of the neoplastic lesion. In the population studied, the pre-operative Frankel value was distributed as follows: 17 grade A patients, 19 grade B patients and 4 grade C patients. As regards the surgical timing from the onset of the deficit, 15 patients were operated on in the first 12 hours, 9 patients between 12 and 24 hours, 2 patients between 24 and 48 hours, while 14 patients were operated after 48 hours. A statistically significant difference was found ($p < 0.05$) in terms of neurological outcome between those who were operated within 12 hours from symptom onset and patients operated subsequently. All subjects operated within 12 hours had a clinical improvement. On the basis of the collected and analyzed data, it is advantageous a surgically strategy within 12 hours from the onset of neurological symptoms in patients with vertebral metastases. In agreement with the literature, also in the series of this study, the early approach proved to be favorable. Therefore, it is reasonable to assume that an early management of the lesion allows to interrupt the cascade of events responsible for the secondary damage, with a consequent better residual medullary function.

KEYWORDS: *spinal tumors, surgical intervention, timing*

INTRODUCTION

Spinal Cord Injury (SCI) is a lesion of the spinal cord, partially or completely, that compromises its function, for a limited period of time or permanently. SCI can be due to various causes, which can be roughly divided into traumatic and

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non-traumatic. Among the non-traumatic causes, the most important are the neoplastic one (linked to primitive neoplasms of the spinal cord or metastases) and mechanical/degenerative ones. Other causes are vascular, infectious, inflammatory and deficient (for example vitamin B12 deficiency) (1, 2).

The main therapeutic and neuroprotective treatment, therefore, should be implemented in the initial phase, to prevent the onset of secondary damage (3).

There are various mechanisms responsible for the secondary damage, in particular, among all of them, the alterations of the vascular microcirculation, which consist in the appearance of intravascular thrombi, hemorrhagic petechiae and vasospasm.

One of the major causes, in epidemiological terms, of non-traumatic SCI is neoplastic metastasis. Bones are among the main sites of metastatic localization of neoplasms; more than 90% of metastases are extradural and involve the bone component (4). The portions of the spine most frequently affected, based on frequency, are the thoracic tract, the lumbar tract and finally the cervical tract (5). The most common symptom is rachis pain, which typically gets worse in the supine position and improves in orthostatisn. After back pain, in order of frequency, there are motor disturbances, such as weakness, gait disturbances and paraplegia, but also sensory deficits, such as paraesthesia and anesthesia, and neurovegetative disturbances (6).

The specific treatment of myelolesion includes surgery, a medical treatment based on steroids, the management of complications (for example, heparin prophylaxis is used to prevent episodes of deep vein thrombosis) and any pharmacological devices for neuroprotection.

The therapeutic management of these pathologies is based on radiotherapy, often used as adjuvant therapy, or in case of absolute contraindication to surgical treatment. Conventional radiotherapy involves the irradiation of the affected vertebrae, including in the field also one or two vertebrae above and below the pathological one. Surgery is also a tool of decompression.

For thoracic and lumbar lesions, the standard surgical approach involves decompression and stabilization of the spine with screws and bars via a posterior approach. For cervical lesions, on the other hand, decompression and corpectomy is performed via a ventral approach, with placement of a prosthesis (7).

Surgery remains the fulcrum of therapy, especially in cases with spine instability and spinal cord involvement, which aims at pain management, improvement of neurological symptoms, stabilization of the spine and therefore improvement of quality of life. The treatment of vertebral metastases requires a multidisciplinary approach, personalized for each patient (8).

In literature, the debate regarding the optimal timing for surgery is still open. In general, the Authors suggest intervening as early as possible, generally there is approval in the 48-hour cut-off, to improve the clinical and neurological outcome, avoiding the onset of edema, venous congestion and secondary vascular damage (9).

Considering the high discordance of the studies in the literature, it is difficult to establish the optimal surgical timing in patients with vertebral metastases starting with neurological deficit. The aim of our study is to evaluate the timing in wich an early surgical intervention in patients with vertebral metastases starting with acute neurological deficit influences the clinical outcome prognostically.

MATERIALS AND METHODS

Patient population

The study includes data of 40 oncological patients, operated on between September 2013 and March 2021 at the Department of Neurosurgery of the Ospedale Riuniti Ancona. All the patients accessed the emergency room with an acute onset stenic deficit. Retrospectively, clinical information was gathered for each patient, in particular relating to age, gender, time of admission to the emergency room and time of surgery, level of lesion (cervical, thoracic, lumbar or sacral spine), pre- and post-operative neurological functions, surgical technique used, any post-operative complications and histological examination of the primary neoplasm. Furthermore, in the context of pre-operative planning, each patient was evaluated through MRI imaging study.

Frankel scale

The Frankel scale was used to evaluate the neurological symptoms and, in this study, was used to compare neurological functions before and after the surgery. All the patients in the study, in fact, presented a certain degree of neurological deficit, which was measured and monitored through this clinical score.

Data analysis

A statistical analysis of the data was performed, in order to demonstrate any significant difference in the neurological outcome of the patients operated on with different timing. For this purpose, the analysis of variance system ANOVA was used, in order to compare differences between certain groups. A result with p-value < 0.05 was considered significant.

RESULTS

Study population

The sample population consists of 26 men and 14 women, with an average age of 66 ± 14 years. Table I below summarizes the histology of the primary lesions of all the patients. In agreement with literature data, the tumors most frequently found in our series are lung, prostate, breast, hematological neoplasms and those of the gastro-intestinal tract.

Table I. *Histology of study's tumors*.

Primary lesion	Num. of patients
Lung	11
Prostate	6
Breast	3
Multiple Myeloma	3
Colorectal	3
DLBCL	2
Stomach	2
Pancreas	2
Follicular B Lymphoma	1
Thyroid	1
Klatskin's tumor	1
NET	1
Sarcoma	1
Ewing's sarcoma	1
Seminoma	1

In the population studied, the pre-operative Frankel value was distributed as follows: 17 patiens grade A, 19 patients grade B and 4 patients grade C. In the post-operative period, however, the values tend to improve. Indeed, 5 patients remained classifiable as A, 8 patients as B, 15 patients as C, 11 patients as D and one patient as E.

In our study, the most frequently affected tract was found to be the thoracic one, with 35 patients; the lumbar spine was involved in 3 patients, while only 2 patients had a cervical localization.

The surgical techniques used are: arthrodesis with vertebral decompression via laminectomy, laminectomy alone or vertebral stabilization alone.

During the postoperative period, most patients had no post-operative complication, while one patient developed a paravertebral hematoma and one patient developed a CSF fistula.

Finally, with regard to surgical timing, the patients in the study were divided according to the time between the onset of symptoms and surgery. As shown in the Table II below, 15 patients were operated on in the first 12 hours, 9 patients between 12 and 24 hours, 2 patients between 24 and 48 hours, while 14 patients were operated on after 48 hours.

Table II. *Surgery timing*.

Surgery timing	Num. Of patients
Within 12 h	15
12 - 24 h	9
24 - 48 h	2
After 48 h	14

Through statistical analysis, a statistically significant difference ($p < 0.05$) was found, in terms of neurological outcome, between those who were operated on within 12 hours of the onset of symptoms and patients operated on later. In fact, all subjects operated on within 12 hours had a clinical improvement, assessed by Frankel grade.

On the other hand, no statistically significant differences ($p > 0.05$) were found when evaluating the time cut-off of 24 hours and 48 hours.

Then, it was evalueted the link between the age of the patients and the change in the Frankel grade, to verify any improvement associated with particular age groups. However, these results did not prove to be statistically significant.

DISCUSSION

Given the high incidence and the important clinical, psychological, economic and social impact of spinal cord injuries, over the years we have tried to define the best way to manage patients with SCI, to restore their functionality and quality of life.

Specifically, patients with metastasis of vertebral column arising with neurological deficits, we tried to define the optimal surgical timing for maximun benefit, in terms of recovery the neurological functionality. Various authors propose different cut-offs, based on the data they have collected. In fact, in some studies are set relatively early deadlines, while in others is also accepted a more permissive time frame.

In general, there is a tendency to consider and recommend an approach as early as possible. In fact, knowing the pathophysiology of acute SCI, it is considered that early management of the lesion allows interrupting the cascade of events responsible for the secondary injury, with a consequent better residual spinal cord function.

As in other works, in this study the improvement of neurological function (assessed by Frankel grade) in relation to the time elapsed between the onset of weakness and surgery was retrospectively evaluated. In our series, the difference between patients operated on within 12 hours of the onset of symptoms and patients operated on later was statistically significant. On the other hand, the cut-offs of 24 and 48 hours were also evaluated, but were not found statistically significant differences. These results are in agreement with the general tendency of the literature to suggest an early and timely approach.

The main limitations of this study were the number of patients and the high heterogeneity of the patients examined. Furthermore, the significant variability of the subjects under examination could have influenced the statistical evaluation.

In fact, the patients studied have variable demographic characteristics, are affected by different neoplasms, with various metastatic localizations and have undergone different surgical interventions.

All these variables can influence the final outcome, therefore further studies are useful, which allow the evaluation of larger and more homogeneous population samples for demographic and clinical characteristics.

CONCLUSIONS

Based on the analyzed data, it is advantageous to intervene surgically within 12 hours of the onset of neurological symptoms in patients with vertebral metastases. In agreement with the literature, in fact, also in the series of this study, the early approach proved to be favorable, allowing to improve the neurological prognosis. The timing that is more advantageous is placed on the 12-hour cut-off.

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Case Report

SCOLIOSIS IN MAINZER-SALDINO SYNDROME: A CASE REPORT AND REVIEW OF THE LITERATURE

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ABSTRACT

Mainzer-Saldino Syndrome (MZSDS) is a rare autosomal recessive disease caused by mutations in gene IFT140 encoding intraflagellar transport protein, a subunit of the IFT-A complex involved in retrograde ciliary transport. MZSDS is a multisystem disorder characterized by chronic renal disease and skeletal abnormalities: phalangeal cone-shaped epiphyses, short stature, short-ribs thoracic dysplasia, pelvic deformities, maxillofacial and proximal epiphysis and femur metaphysis abnormalities. Scoliosis has never been described as one of the features of this syndrome. We present the case of a 26-year-old Italian Caucasian man affected by MZSDS with a severe scoliosis surgically treated. Cobb angle of major thoracic curve was 120° and Cobb angle of major lumbar curve was 110°. Curve flexibility was evaluated on the preoperative standing lateral bending X-ray and side-bending X-ray. By means of CT images, we obtained a Three-Dimensional (3D) model of spine used for the preoperative study. A single-posterior spinal arthrodesis extending from T2 to L5 vertebrae was performed. No intraoperative and early postoperative surgical complications occurred. Postoperative radiographs demonstrated main thoracic correction from 120° to 56° (53.7% correction rate), main lumbar correction from 110° to 52° (52.7% correction rate). In conclusion, our hypothesis is that scoliosis may be a skeletal feature of MZSDS. It can produce a severe deformity needing for major surgical treatment. Preoperative multidisciplinary assessment is necessary. Scoliosis correction and maintenance at follow-up result in the improvement of pulmonary function and high patient satisfaction.

KEYWORDS: *posterior spinal fusion, case report, spinal deformities, MZSDS, 3D printing*

INTRODUCTION

Mainzer-Saldino Syndrome (MZSDS) is a rare autosomal recessive disease caused by mutations in gene IFT140 encoding intraflagellar transport protein, a subunit of the IFT-A complex involved in retrograde ciliary transport (1). Its prevalence is unknown. MZSDS is a multisystem disorder characterized by phalangeal cone-shaped epiphyses (PCSE) and chronic renal disease. Occasional features are: retinal dystrophy, cerebellar ataxia, hepatic fibrosis, bilateral hearing difficulties and skeletal abnormalities, suggesting that MZSDS is a skeletal ciliopathy (2). Skeletal abnormalities are:

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short stature, short-ribs thoracic dysplasia, pelvic deformities, maxillofacial and proximal epiphysis and femur metaphysis abnormalities. Diagnosis occurs when children have develop kidney failure – usually between the ages of 10 and 14. Electroretinography supports the diagnosis. Genetic testing (sequence analysis of the entire coding region of IFT140) is essential. In the past, the disease was burdened by a relatively high mortality rate, mainly due to renal insufficiency that quickly evolved into end-stage renal disease. The remarkable improvements recorded in the field of kidney transplantation have drastically reduced the mortality from renal syndrome (3).

Scoliosis has never been described as one of the features of this syndromic condition. In this article, we report the first described case of severe scoliosis associated with MZSDS requiring surgical treatment. Spinal deformities and medical conditions in syndromic population affect complication rates significantly.

CASE PRESENTATION

We present the case of 26-year-old Italian Caucasian man affected by MZSDS with a severe scoliosis surgically treated. The diagnosis of MZSDS was performed on the basis of clinical features and genetic testing when he was 9-years-old.

The patient was born by spontaneus delivery, at term, from healthy parents with normal stature and body build. He was the second of three children. The oldest brother died when he was a teenager because of renal failure in MZSDS, while the youngest is unaffected. Prenatal echography had not revealed any growth failure. At birth no abnormalities were detected. There was no family history of scoliosis.

At 4 years, progressive bilateral valgus knees arose. Physical and radiological examination of the knees carried out at 7 years showed worsening painful knees after long walking, and valgus angle equal to 12° on right and 15° on left. The patient underwent corrective surgery by bilateral femoral epiphysiodesis with good result. Two years later, removal of staple was performed.

At 9 years, because of acidosis with reduced glomerular filtration, he underwent renal biopsy that diagnosed focal segmental glomerulonephtitis. Later the patient developed stage V renal insufficiency requiring hemodialysis treatment. Consequently he had hypertensive heart disease, anemia, hyperparathyroidism and osteoporosis. Furthermore he had chronic liver disease with splenomegaly, retinitis pigmentosa and respiratory failure.

At 10 years, scoliosis was evidenced and it was first treated with global postural rehabilitation and brace but the patient had low compliance. Subsequently to scoliotic curves progression, surgical treatment became necessary but the patient and his family refused.

At 12 years, the patient underwent bilateral flat feet surgical correction.

In 2016 the patient was placed on waiting kidney transplant list at the Department of Nephrology of Riuniti Hospital in Ancona, but three years later he was excluded because of the development of severe respiratory failure.

In July 2019 the patient came to our attention and it seemed appropriate to proceed with surgical correction. On February 2020 he was admitted to Clinical Orthopaedics of Riuniti Hospital in Ancona. He was 155 cm tall and he had low BMI equal to 14.8. Physical examination showed deformity of the back, waistline asymmetry, unequal shoulder and anterior superior iliac spine levels. The Adam's forward bend test revealed right rib hump and left lumbar prominence. The assessment of posture and gait detected listing to left side due to leg length discrepancy with right leg 2.5 cm longer than left leg, clinically measured (Fig. 1).

Fig. 1. *Preoperative **(a, b, c, d)** clinical photographs show the severity of spine deformity at the clinical examination and clinical photographs at 2-years follow-up **(e, f, g, h)** show good overall balanced spine in the coronal and sagittal planes.*

No skin or soft tissue abnormalities were noted overlying his spine. Neurological examination confirmed no apparent current vascular and nervous deficits in his upper and lower limbs. F5 muscle strength was evaluated using MRC (Medical Research Council) Scale. Tendon reflexes were symmetrically elicited (Fig. 1).

The patient underwent brain and spine magnetic resonance imaging (MRI) before surgery in order to evaluate the presence of spinal cord malformation including occult syrinx, spinal stenosis, spinal cord tumors, Arnold-Chiari malformations or neuromuscular disorders (4).

Cobb angle measurement of coronal curves and coronal balance were achieved on standing posteroanterior X-ray (Fig. 2).

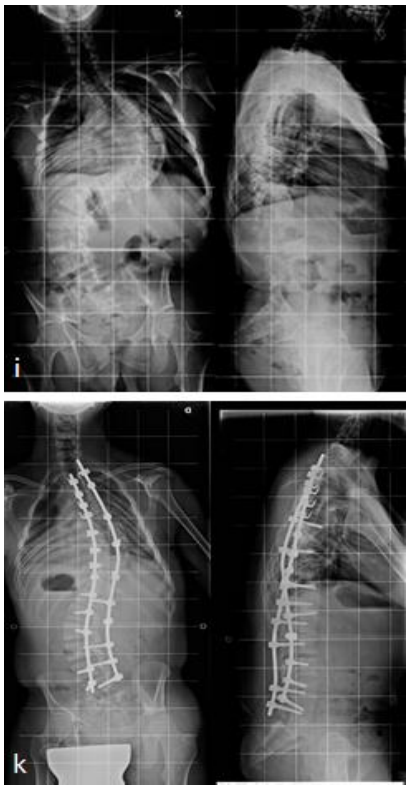

Fig. 2. *Preoperative posteroanterior and lateral X-ray examinations show the severity of scoliotic curves (i), and postoperative posteroanterior and lateral X-ray examinations show a satisfactory correction of the spine deformity (k).*

Rightward major thoracic curve extending from T4 to T11 with T8 apical vertebra, leftward major lumbar curve extending from T12 to L5 with L2 apical vertebra and leftward minor cervical curve extending from C5 to T3 with T1 apical vertebra were revealed. Measurement of pelvic obliquity (PO) was equal to 12°, right side elevated (5) (Fig. 3).

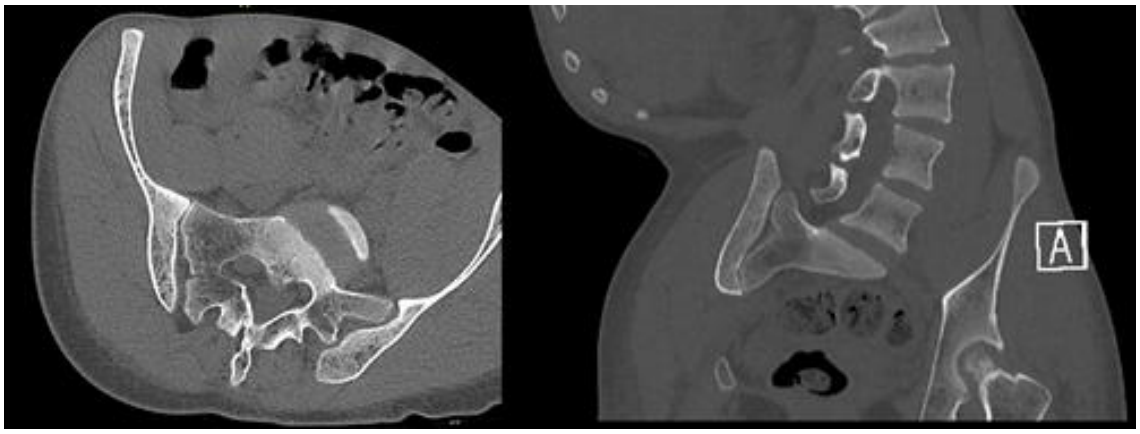

Fig. 3. *Computed tomography (CT) images show the rotation of sacrum and pelvis obliquity (PO).*

The Risser grade was 5 indicating his final skeletal growth. Sagittal curve and sagittal balance were determined on standing lateral X-ray (6). Curve flexibility was evaluated on the preoperative standing lateral bending X-ray and side-bending X-ray. In Table I, there is a recap of radiographic coronal measures on preoperative time.
Computed tomography scan with reconstruction according to the axial plane of each vertebra and with 3D reconstruction was performed to evaluate dystrophic features.

Table I. *Radiographic coronal measures on preoperative time (T0).*

	Thoracic curve (°)	Lumbar curve (°)	Coronal imbalance (cm)
Before surgery (T0)	120	110	-2
Standing lateral bending	100	95	-
Side-bending	84	-	-

According to the classification system proposed by Li Y et al., on a total of 34 pedicles, evaluated on CT images with reconstruction according to the axial plane of each vertebra, the results were: A: 10; B: 13; C: 11; D: 0; E: 0 (7). Wedging of vertebral bodies is present in six vertebrae at the apices of the major curves. According to Ho et al. method, rotation was assessed on CT images with reconstruction according to the axial plane of the apical vertebra (8, 9). The rotation angle of T8 was 38°, the rotation angle of L2 was 59°.

During the hospitalization, a preoperative assessment was organized. The patient underwent anesthesiological evaluation which required specialist investigations. Echocolordoppler of the supra-aortic vessels and the venous lower limbs detected no steno-obstructive diseases. High-grade restrictive spirometry pattern and high DLCO reduction were observed. Cardiological evaluation detected an estimated PAPs equal to 43 mmHg secondary to restrictive pneumopathy.

Preoperative planning

The patient underwent spine and pelvis CT, by whose images we obtained a Three-Dimensional (3D) model, which was thereafter printed by using a fused deposition modeling printer (Fig. 4).

Fig. 4. *Three-Dimensional (3D) printed model was used in preoperative planning.*

8 hours were spent for software production of the 3D printed model, while the printing required 175 hours. The amount of costs for materials was about 300 Euros (10).

Surgical time

A single-posterior spinal arthrodesis extending from T2 to L5 vertebrae was performed. Intraoperative spinal cord monitoring was performed during the surgical time recording somatosensory and motor evoked potentials and the signal was always normal.

A subperiosteal exposure of the spine with extensive facetectomies was performed in order to further mobilise the curves. Posterior instrumentation was hybrid and it included laminar and pedicle hooks, sublaminar bands, pedicle screws and rods. During the procedure for inserting hybrid instrumentation, the surgeons noticed a reduced bone quality, especially of the pedicles. At first, a temporary titanium rod on thoracic curve concavity from T2 to T12 was used (Fig. 5).

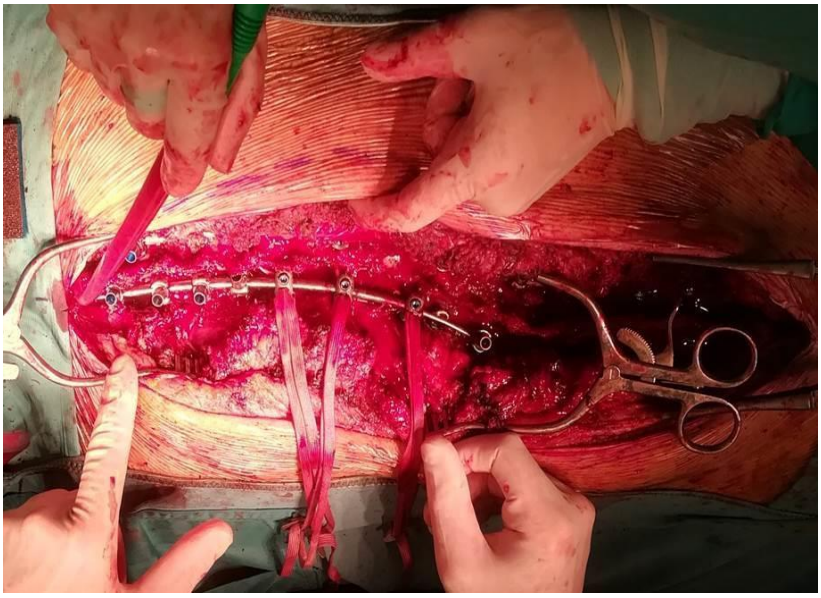

Fig. 5. *Temporary titanium rod on thoracic curve concavity used during the surgical procedure to perform corrective manoeuvres.*

Corrective manoeuvres included apical segmental translation, rod derotation and proximal/distal distraction of the construct. Later, a cobalt-chrome rod on the right-side was placed from T2 to L5. The correction of the thoracic convexity was achieved using compression manoeuvres while the correction of the lumbar concavity was achieved using apical segmental translation, rod derotation and proximal/distal distraction manoeuvres. At last, temporary titanium rod was removed and a cobalt-chrome rod on left-side was placed from T2 to L5. The rods were attached tightly to spine by hybrid instrumentation (11). Locally harvested bone from the spinous processes was used to achieve interfacetal and intertransverse arthrodesis. It was supplemented by freeze-dried bone allograft.

The total operative time was about 12 hours. Total amount of blood loss was 2000 mL. Intraoperatively the patient underwent hemodialysis.

Postoperative time

Postoperatively, the patient was monitored at the postoperative intensive care unit (ICU). The day after, he was extubated and transferred to Orthopaedics Clinic.

During the initial postoperative period, there were no neurological abnormalities and no sign of respiratory dysfunction. There was no sign of wound infection. On the 5th postoperative day, he mobilised out of bed with walking frame.

Postoperative radiographs demonstrated main thoracic correction from 120° to 56° (53.7% correction rate), main lumbar correction from 110° to 52° (52.7% correction rate) with a persistent imbalanced spine in the coronal and sagittal planes. The measures are reported in Table II and Table III (Fig. 2).

Table II. *Radiographic coronal measures on preoperative time (T0), postoperative time (T1) and 2-years follow-up.*

	Thoracic curve (°)	Lumbar curve (°)	Coronal imbalance (cm)
Before surgery (T0)	120	110	-2
After surgery (T1)	56	52	-6.4
2-years follow-up (FU)	57	54	+0.5

Table III. *Radiographic sagittal measures on preoperative time (T0), postoperative time (T1) and 2-years follow-up.*

	Kyphosis (°)	Lordosis (°)	Sagittal imbalance (cm)
Before surgery (T0)	43	50	+2.5
After surgery (T1)	25	28	+6.5
2-years follow-up (FU)	29	29	+ 3.7

Leg length discrepancy equal to 2.5 cm persisted and it was corrected by uniform heel-to-toe plateau on the left foot. The patient was discharged on the 10th postoperative day. No intraoperative and early postoperative surgical complications occurred.

Follow-up

In the third postoperative month, he returned to his daily activities. Radiographs of his spine at latest follow-up 2 years after surgery demonstrated a satisfactory maintenance of surgical correction in the coronal and sagittal planes. No late postoperative surgical complications occurred (Fig. 1). The spirometry at 2-years follow-up noticed high-moderate-grade restrictive pattern with FVC improved from 41% of predicted to 56% of predicted and a moderate reduction in DLCO (47% of predicted). The comparison of preoperative and 2-years follow-up pulmonary functions is shown in Table IV. SRS-24 questionnaire was administered to assess the patient's clinical satisfaction in 2-year follow-up (12) (Fig. 6). The results are recapped in Table V.

Table IV. *Preoperative and 2-year follow-up pulmonary functions.*

		Preoperative time		2-year follow-up	
	Ref	Meas	%Ref	Meas	%Ref
FVC (Lt)	4.08	1.67	41	2.27	56
FEV1 (Lt)	3.55	1.28	36	1.86	52
FEV1/FVC (%)	83	77		81.9	
FEF25-75% (Lt/sec)	4.65	1.11	24	1.92	41
FEF 50% (Lt/sec)	4.83	1.63	34	2.05	42
PEF (Lt/sec)	8.73	4.45	51	5.13	59
VC (Lt)	4.26	1.67	39	2.27	53
TLC (Lt)	5.54	2.42	44	3.11	56
RV (Lt)	1.41	0.75	53	0.84	60
EVR (Lt)		0.42		0.64	
RV/TLC (%)	24	31		27	
DLCO (mL/mmHg/min)	29.3	11.0	37	13.7	47
DLCO/VA (mL/mHg/min/Lt)	5.29	6.44	122	6.20	117
VA (Lt)	5.54	1.70	31	2.21	40
DL Adj (mL/mmHg/min)	29.3	11.0	37	13.7	47
DL/VA Adj (mL/mHg/min/Lt)	5.29	6.44	122	6.20	117

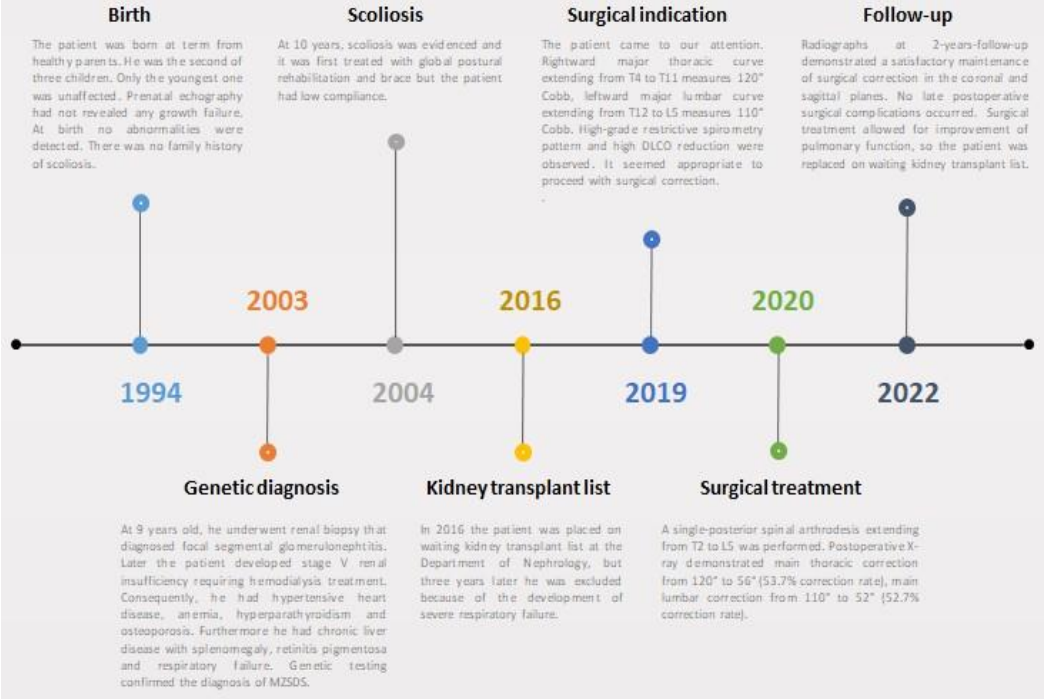

Fig. 6. *The timeline shows the key events of our case.*

Table V. *SRS-24 questionnaire results.*

Domain	Score Pt/Possible (max)
Pain	32/35
General self-image	13/15
Self-image after surgery	14/15
Function after surgery	10/10
General function	12/15
Function-activity	14/15
Satisfaction with surgery	14/15
Total	**109/120**
90.8%	

Consent

Written informed consent was obtained by the patient for publication of this case report and any accompanying images.

DISCUSSION

The term “syndromic scoliosis” is used to describe scoliosis associated with a systemic disease. Scoliosis in Ehler-Danlos syndrome (EDS), Marfan syndrome (MF), Down syndrome, Achondroplasia (AP), Prader-Willi Syndrome (PWS), and Osteogenesis Imperfecta are representative of this category. Syndromic population has a greater rate of developing scoliosis than idiopathic population. This article focuses on the analysis of the literature regarding the outcomes of surgical management in syndromic scoliosis patients. In a recent study of 2019, Andrew S. Chung et al. compared the hospital outcomes and surgical trends of patients with syndromic scoliosis and patients with idiopathic scoliosis undergoing spinal fusion. There was a significant decline in the number of anterior approaches performed in both cohorts, whereas the number of posterior approaches increased. Rates of major complications (Neurologic injury and pulmonary embolism) were 3-times higher in syndromic scoliosis cohort. Similar findings were reported in procedural and device-related complications (13). Levy et al. reviewed the literature and noted high rates of postoperative infections

(5%), implant failures (20%), and pseudoarthrosis (25%) in the syndromic scoliosis population (3). These results are in line with existing literature (14, 15).

Mainzer-Saldino Syndrome (MZSDS) is a multisystemic disease characterized by bony development disorders. Scoliosis has never been described in literature as one of the features of this syndrome. We reported the first described case of severe scoliosis associated with MZSDS requiring surgical treatment.

In a recent review, Li X. et al. summarized the role of primary cilia and ciliary proteins in the pathogenesis of skeletal diseases including scoliosis. In this review, the authors showed evidence that the primary cilia may be a promising target of clinical intervention for skeletal diseases (osteoporosis, bone/cartilage tumor, osteoarthritis, intervertebral disc degeneration, spine scoliosis, and other cilium-related skeletal diseases) (16). Schlösser et al. reported that the prevalence of scoliosis and significant spinal asymmetry in 198 patients affected by primary ciliary dyskinesia was 8 and 23%, respectively (17).

Our patient was first noted at the age of 25 years. He was affected by a severe spine deformity which required major scoliosis surgery.

3D printed model permitted to study and exhaustively comprehend the complexity of the deformity: we could have the visual and tactile experience of the entity of curves, sagittal and coronal balances, pelvis rotation, pedicles dimension and position. The 3D printed model was so used to evaluate vertebral dystrophy, in particular pedicles, allowing us to identify the type of surgical approach and to choose the better position of hooks, screws and sublaminar bands. It was useful to reduce the fluoroscopy exposition for both surgeons and the patient. The 3D printed model was also an instrument for better explaining the operative planning of this complex deformity to patients and younger surgeons.

A single-stage posterior deformity correction was managed. According to Dobbs MB et al., posterior-only approach has the advantage of providing the same correction as an anterior/posterior spinal fusion, without the need for entering the thorax and more negatively impacting pulmonary function (18). The posterior-only approach performed using high-density hybrid instrumentations (implant density equal to 0.72) produced a satisfactory correction of the severe deformity (19).

Operative time and intraoperative blood loss were compatible with the greater complexity of the surgery. Motor and sensory traces recorded intraoperatively were unchanged throughout the procedure.

The coronal and sagittal imbalance did not improve on immediate postoperative time, probably due to the severe impact of the surgical trauma, but it decreased in the 2-year follow-up leading to an acceptable overall balance.

Curves measurements on postoperative time and on the follow-up demonstrate no loss of surgical correction across the instrumented levels. There was also no evidence of detected pseudoarthrosis and no add-on junctional deformity proximally or distally the levels of the spinal fusion. Instrumentation was inside.

Surgical treatment allowed for improvement of pulmonary function, so the patient was replaced on waiting kidney transplant list at the Department of Nephrology of Riuniti Hospital in Ancona.

As SRS-24 questionnaire showed, surgical treatment positively impacted patient's life quality: no experience of severe pain, better rating of his self-image, no limitations in daily activities, walking without fatigue, practicing moderate sport.

The clinical case described in this paper was an important challenge for orthopaedic surgeons. Because of the serious comorbidities a detailed planning of each phase was essential, from the preoperative preparation of the patient, to the surgical procedure, up to the postoperative care and monitoring over time and follow-up. A multidisciplinary approach was the key of the success of the surgical treatment. Surgeons approaching a rare syndrome should fully understand its complexity. In addition to meticulous surgical planning, the patient must be accompanied and supported during all stages of care.

CONCLUSIONS

In conclusion, our hypothesis is that scoliosis may be a skeletal feature of MZSDS. It can produce a severe deformity needing for major surgical treatment. Surgical correction of scoliosis restores spinal balance, prevents severe pulmonary and cardiological complications and mechanical back pain, as well as improves cosmesis. Because of severe comorbidities, preoperative multidisciplinary assessment is necessary to avoid surgical morbidity. Scoliosis correction and maintenance at follow-up result in the improvement of pulmonary function and high patient satisfaction. Given the paucity of existing data regarding deformity correction outcomes in syndromic scoliosis population, further researches are necessary for a better management of these conditions.

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Case Report

MEDULLARY COMPRESSION FROM SPINAL ARACHNOID CYST: A CASE REPORT AND REVIEW OF LITERATURE

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ABSTRACT

Arachnoid cysts, extradural or intradural, represent a fairly rare cause of compression of the spinal cord. They are mainly intradural cysts, mainly located in the cervico-thoracic sector. The onset is frequently characterized by motor deficits (i.e. tetraparesis, paraparesis), dysesthesia and pain. Here we describe the case of a 51-year-old man who showed a sensory-motor deficit secondary to a spinal arachnoid cyst (SAC) diagnosed by MR investigation and surgically removed with clinical improvement. We also present a review of literature about terminology, presentation, recommended investigations, management and outcomes of patients with Spinal Arachnoid Cyst. The patient was clinically evaluated at the admission, after the surgical treatment and with one year follow-up. We recorded clinical scores such as Visual Analogue Scale (VAS) and Roland Morris Disability Questionnaire (RMDQ). The radiographic investigations for the definition of the pathology were X-ray and MRI of the spine, electrophysiological investigations was also performed. The evaluation and treatment of the patient was carried out with a combined approach between an orthopedic specialist and a neurosurgeon. For the review of the literature a PubMed, NCBI and Google Scholar search was performed with keywords 'Spinal Arachnoid Cyst' and 'spinal arachnoid cyst with spinal cord compression'. After surgical treatment there was a positive progress with recovery. At three months follow-up the limitation of movements and weakness of the upper limbs were recovered. At one year follow-up the patient had already resumed normal daily activities without any reported limitations. For the review of the literature we found 7 articles about the arachnoid cyst of the dorsal spine, of which 6 are single case reports and only one collects two cases. SACs are rare lesions that can present with various neurological symptoms: paraesthesia, neuropathic pain, paresis, and gait disturbances. Conservative treatment with careful observation may be an acceptable option for asymptomatic patients. However, in patients with progressive or recurrent symptoms, surgery is the gold standard of treatment.

KEYWORDS: *spinal arachnoid cyst, dorsal arachnoid cyst, cord compression, medullary compression, arachnoid cyst adult, decompression arachnoid cyst.*

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INTRODUCTION

Arachnoid cysts are cerebrospinal fluid-filled cavities that can develop between the brain surface and the skull base or on the arachnoid surface, which is one of the three meningeal membranes that surrounds the central nervous system and the initial nerve tract (1-5).

Arachnoid cysts have a higher male incidence (6). Many cases begin in childhood, but onset can be delayed until adolescence (7).

Arachnoid cysts can be located at the brain or spinal level, the latter being rarer than the former (7). There are two types of arachnoid cyst: the primary arachnoid cyst, which is present from birth and due to an incorrect development of the arachnoid, and the secondary arachnoid cyst, caused by other pathologies such as trauma, tumours, infections, etc. (8, 9).

Spinal arachnoid cysts (SACs) are rare entities and are classified into primary (congenital or idiopathic) or secondary (acquired). Acquired SACs are the most frequent. They most frequently occur in the mid-to-lower thoracic levels (7, 10-12).

The arachnoid cyst tends to be asymptomatic when its size is small. It becomes symptomatic when it reaches a significant size (1-5, 7, 13-15).

SACs can present with progressive myelopathy if they compress the common spinal cord. The most common manifestations are paraesthesia, neuropathic pain, paresis, and ataxia of gait (7, 11).

CASE REPORT

Patient M.A., a 51-year-old male, presented with right low back pain and mild weakness of the right lower limb approximately from 2014. He was initially treated with medical therapy, physiotherapy and ozone therapy with partial benefit. In 2019, left lumbosciatalgia appeared, associated with hypoesthesia of the left lower limb, dorsalgia and right cervicobrachialgia, and worsening the weakness of the right lower limb. ROT Patellar, Achilleus and Middle plantar were normally evocable bilaterally. On physical examination, the patient also complained of weakness in the upper limbs and slowed thoracic movements with severe limitation of flexion movements. The patient was also evaluated with clinical scores for pain with Visual Analogue Scale (VAS) (16) and with the Roland Morris Disability Questionnaire (RMDQ) to investigate the degree of disability given by the existing pathology; VAS value was 7, the RMDQ (17) score was 13.

The patient underwent appropriate diagnostic investigations (X-ray, MRI, electromyography, motor evoked potential, somatosensory evoked potentials) which showed degenerative disc disease L3-L4, L4-L5 and a dorsal cystic neoformation D3-D6 with a mild-severe neurological impairment.

The magnetic resonance showed the presence of a cystic formation, probably intradural, at the mid-dorsal level, in the posterior sector of the spinal canal. This formation causes a discrete compression of the spinal cord, which appears thinned and displaced anteriorly (Fig.1-3).

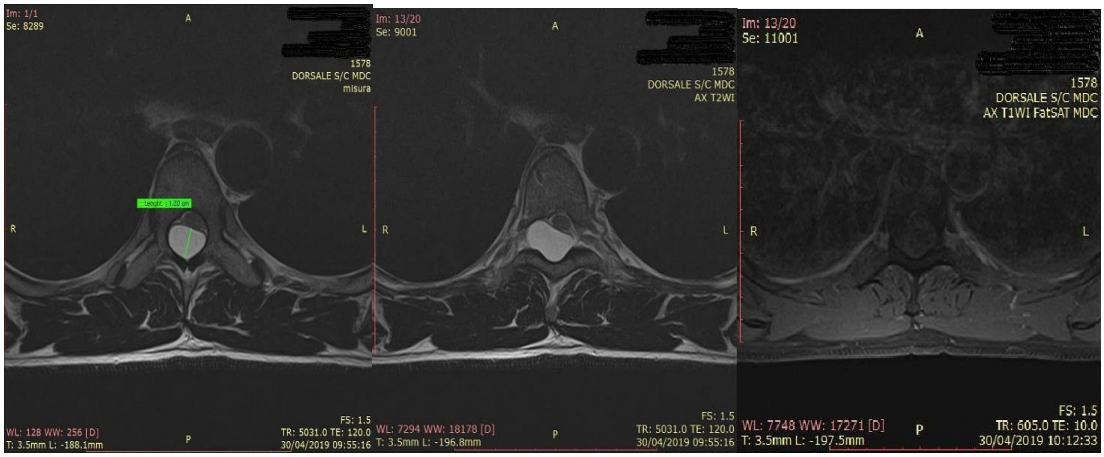

Fig. 1. *Images from the RM performed by the patient at the time of diagnosis (April 2019). From the axial cuts in the T1-T2 weights, an anteroposterior diameter of the cyst was found of 1.20 cm*

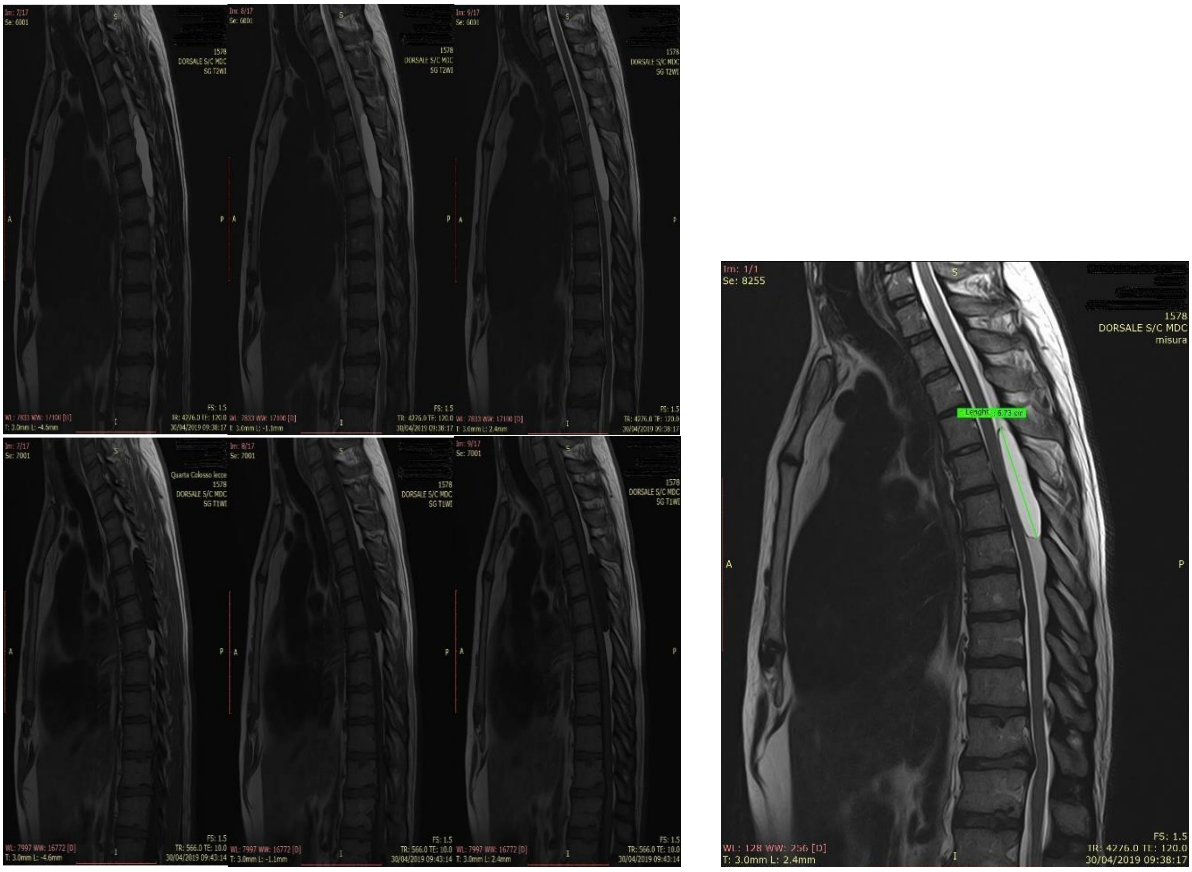

Fig. 2 -3. *Images from the RM performed by the patient at the time of diagnosis (April 2019). From the sagittal cuts in the T1-T2 weights, an cranio-caudal length of the cyst was found of 6.73 cm*

Following a specialist orthopedical and neurosurgical evaluation, the patient was advised to undergo surgery to remove the neoformation in the dorsal tract of the column. On June 2019 he underwent decompression surgery with a posterior approach by means of central laminectomy D3-D7 during which a cystic neoformation in tension D3-D6 was found. The wall of the cyst was incised posteriorly and the content was aspirated. (Fig. 4).

It apparently had a clear appearance and was not stocked. The neoformation, whose margins appeared not in continuity with the perimedullary dura mater, was incised longitudinally and completely excised. Finally, synthetic dura was affixed to protect the dural sac and at the sampling site we proceeded to posterior arthrodesis using reconstruction plates and screws of suitable length. After placing the plates, no possible instability of the segment subjected to laminectomy was detected, which is why no further stabilizations were carried out (Fig. 5-6).

The collected material was sent to a laboratory for histological examination, revealing fibrous tissue with islets of meningothelial cells associated with psammomatous bodies. These features suggest an arachnoid cyst.

After surgery, there was positive progress with recovery, and the patient underwent adequate follow-up and a new diagnostic investigation. Three months after the operation, the pain, assessed with the VAS scale, was around 6 – probably due to the degenerative disc disease of the lumbar spine – and the RMDQ score was slightly decreased at 11. The difficulties at the thoracic level such as limitation of movements and weakness referred to the upper limbs, were completely recovered.

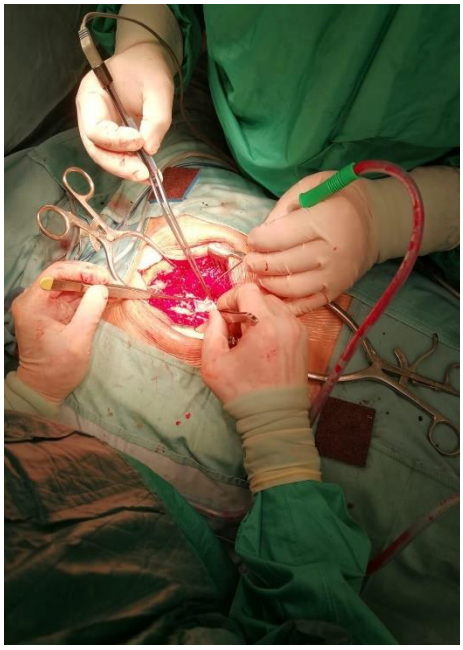

Fig. 4. *Intraoperative detail after the incision of the cyst.*

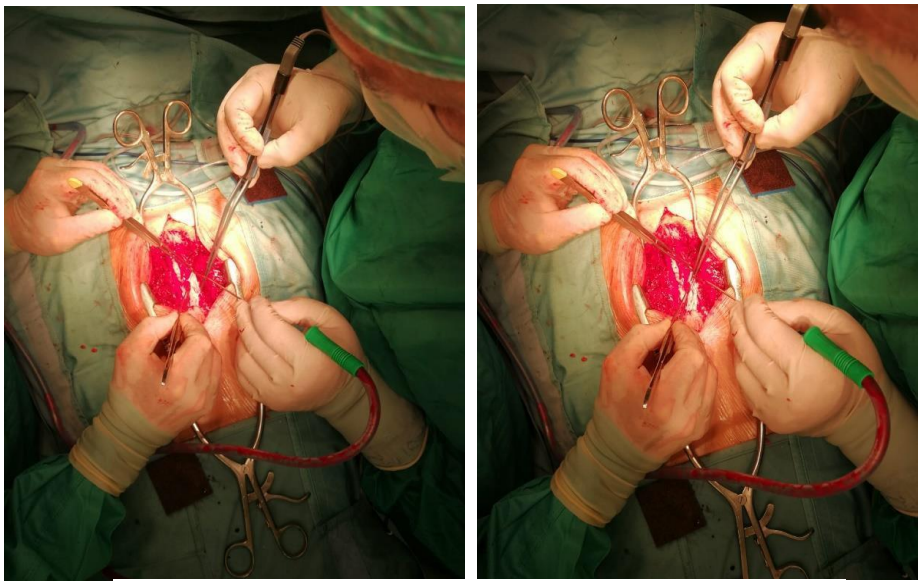

Fig. 5-6. *Intraoperative detail after excision of the cyst.*

Subsequently, in October 2019, with the worsening of the lumbo-cruralgic symptoms, the patient underwent a specialist orthopedic evaluation that suggested and then performed surgery for lumbar spinal stenosis with degenerative disc disease L2-L3, L3-L4. Before this new treatment, an MRI was repeated as a follow-up of the dorsal spine and to better evaluate the stenosis in the lumbar tract (Fig. 7-8).

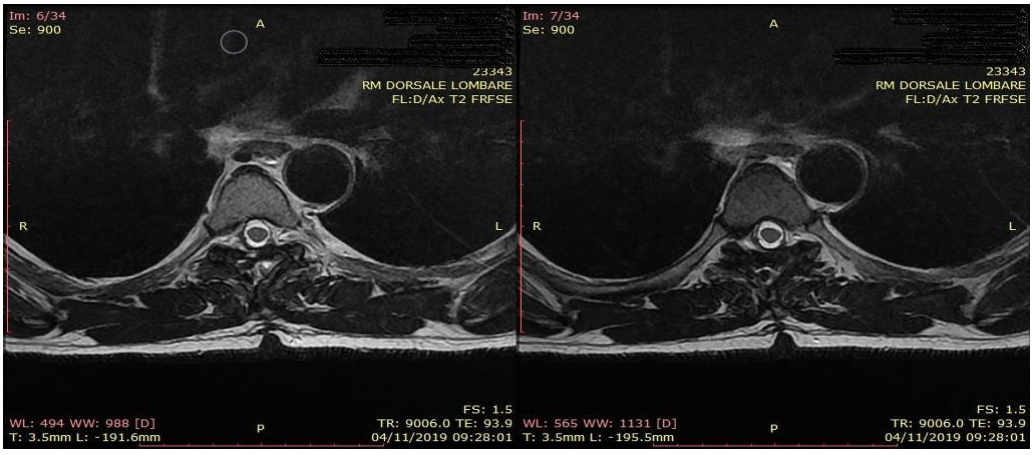

Fig. 7. *MRI performed by the patient in November 2019. Axial cuts at the level of the removed cyst.*

Fig. 8. *MRI performed by the patient in November 2019. Saggital cuts at the level of the dorsal spine. As in the axial cuts the spinal cord is completely decompressed with no other prominent signs of the dorsal tract.*

The patient underwent surgery on December 2019 for hemilaminectomy L2-L3, left L3-L4 with foraminotomy L3, and left L4 with a posterior approach and lumbar spine fusion with screws and rods at the same level. After the second surgery, the patient was evaluated at one month, three months, six months, and one year follow-up.

At the last clinical and radiographic check, the clinical scores reported the following values: VAS 2/3 and RMDQ 4, and the patient had already resumed normal daily activities without any reported limitations. A one-year follow-up check electromyography was also performed, as well as motor-evoked potential and somatosensory-evoked potentials. The mild-severe neurological impairment showed at the neurological tests was almost completely regressed and showed up a 60% improvement in results compared to pre-operative controls.

Review of literature

Due to the rarity of this pathology, it is very difficult to conduct case series, retrospective, and prospective studies. However, it is possible to find a wide variety of reported cases in the literature.

A PubMed, NCBI, and Google Scholar literature review was performed on case reports with search items 'Spinal Arachnoid Cyst' and 'spinal arachnoid cyst with spinal cord compression'. Of the articles found in the literature we selected only those reporting patients treated with arachnoid cysts at the level of the dorsal spine in the adult population. To the best of our knowledge, Table I reported results that showed different types of dorsal spinal arachnoid cysts in the literature (7, 10, 12, 18-21).

Table I. *Review of the literature of patient with arachnoid cyst of dorsal tract of spine in adult population.*

Authors and year	Age/Sex	Clinical presentations	Cyst type and location	Therapy	Follow-Up
Marrone et al 2022	70-year-old male	Lower limbs hyposthenia, Paraesthesia	Spinal Extradural Arachnoid Cyst T11-T12	Laminectomy T11-T12	In 3-months complete symptoms resolution
Ebot et al 2020	80-year-old female	Back pain progressively worsening, lower limbs weakness, incontinence	Spinal Intradural Extramedullary Arachnoid Cyst T4-T10	Selective laminectomies T4-T5 and T9-T10, duble microsurgical fenestration and decompression	In 1-month lower improve of clinical symptoms
Himes et al 2018	60-year-old female	Back pain progressively worsening	Spinal Intradural Arachnoid Cyst T3-T7	Selective laminoplasties T3-T7, double microsurgical fenestration.and decompression	In 7-months complete symptoms resolution
Velz et al 2018	51-year-old female	Back pain, neck pain, headaches	Spinal Intradural Arachnoid Cyst T4-T7	Hemilaminectomy T4-T6 marsupialization and	In 6-months improve of

				placement of an intradural shunt	clinical symptoms
Lee et al 2018	51-year-old male	Gait difficulties, urge incontinence, lower limbs hyperreflexia	Spinal Extradural Arachnoid Cyst T7-T9	Hemilaminectomy T8-T9; In 2-weeks recurrence; Laminoplasty T7-T9, marsupialization and complete excision	In 8-months improve of clinical symptoms
Ghimire et al 2020	83-year-old female	Acute heaviness of lower limbs, Upper motor neuron syndrome	Spinal Intradural Arachnoid Cyst T3-T4	Laminoplasty T2-T4, marsupialization and complete excision	Complete symptoms resolution
Raes et al 2021	38-year-old female	Progressive hyposthenia, hyperreflexia, dysesthesia of the right leg, mild urinary incontinence.	Spinal Intradural Extramedullary Arachnoid Cyst T2-T7	marsupialization of the thoracic cyst and laminectomy at T7; 1-month later recurrence: placement cysto-peritoneal shunt; 8-months later recurrence: selective laminectomy T1-T3;	Complete symptoms resolution
	45-year-old male	complete paraplegia, ascending sensibility loss, allodynia	Spinal Intradural Arachnoid Cyst T4-T11	Laminectomy T9-T11 and marsupialization. Persistent pain post-operative, 2 months later, placement of cysto-peritoneal shunt	Slight improvement left with incomplete disappearance of the neuropathic pain

DISCUSSION

Pathophysiology

Spinal arachnoid cysts are rare spinal tumours that do not expand within the spinal canal. They can develop as cystic formations caused by dural defects and can grow due to the flow of cerebrospinal fluid originating from the intradural arachnoid space (1).

A SAC can grow extraspinal or intraspinal and is typically located on the posterolateral portion of the spinal canal, compressing the spinal cord towards the anterior side (1).

As the enlargement continues, a SAC can worsen the compression of the spinal cord or nerve root, leading to the development of symptoms such as pain and weakness (1).

The pathophysiological explanation of the formation of arachnoid cysts is still unknown; however, various theories have been proposed.

The main hypothesis is that the SAC derives from a diverticulum or a dissection in the septum posticum; a thin fibrous membrane that joins the arachnoid and the pia mater along the posterior line of the spinal cord (7, 22).

However, reports of ventral cysts in the spinal cord suggest that there may be another origin. For example, the incarceration of arachnoid granulations can produce cerebrospinal fluid that becomes trapped in the arachnoid diverticula. These sequestered fluid sacs lead to further disruption of normal cerebro-spinal fluid (CSF) flow and are therefore capable of producing or expanding arachnoid cysts (7).

For nearly all cases of arachnoid cysts, communication between the subarachnoid spaces and the cysts has been reported. This communication can be induced by a congenital anomaly, or secondary to a traumatic injury to the spinal cord, postsurgical arachnoiditis, meningeal infection, haemorrhage, and other insults that cause inflammation and subarachnoid adhesions (7-12). The possibility of a dural injury increases if patients are suffering from conditions such as. Marfan syndrome or dural ectasia or if they experienced movements of the dural sac such as trauma or loss of cerebrospinal fluid (1, 2, 5, 15).

The dural lesion appears to act as a ball valve allowing CSF flow to form a hernia of the arachnoid membrane, while active secretion of CSF from the residual arachnoid membrane plays a role in the expansion of the cyst, which can enlarge up to cause compression myelopathy (1, 2).

Clinical presentation

Arachnoid cysts have a male prevalence with an average peak incidence around 45 years of age.
Most SACs are found in the thoracic region (7, 12, 18, 22, 23). They can be solitary or multiple. SAC can be asymptomatic (mostly congenital cysts), but as they grow, they can present with progressive myelopathy and show signs of upper motor neuron disease. The most common presenting disorders are paraesthesia, neuropathic pain, paresis and ataxia of gait. Hyperreflexia, muscle hyper or hypotonia, bowel dysfunction urinary incontinence, and sexual dysfunction can also occur (7, 11, 21, 24).

Anterior localization of the cyst is associated with more likely to cause weakness and myelopathy, while thoracic cysts are more commonly associated with neuropathic pain and numbness (7, 11).

Diagnosis

Clinical presentation suggests upper motor neuron syndrome and requires radiological instrumental confirmation. The diagnosis of SAC is given by imaging of the spinal cord.

The gold standard is magnetic resonance imaging (1, 5, 8), which will show a mass occupying spinal extramedullary space consisting of a collection of CSF (7, 12, 22); in fact, the T1 and T2-weighted signals of the arachnoid cyst are identical to those of the cerebrospinal fluid. There is no improvement with gadolinium contrast liquid. If possible, MRI with CISS-3D sequences should be performed, allowing for high-resolution visualization of the subarachnoid space and detection of septa, trabeculae, and intradural cystoid formations with a more precise size than T2w imaging (7, 22).

MRI flow study or CT / MRI myelography are valid alternative investigations (7, 12). Myelography should be reserved for unclear cases to show or exclude the communicating sites of the cyst with the subarachnoid space or, in case of consideration of new surgery for insufficient narrowing, when MR imaging appears insufficient (7, 12).

The differential diagnosis is made with neurenteric cysts, dermoid cysts, epidermoid cysts, teratomas, ependymal cysts and parasitic cysts (8, 25).

Therapy

The average duration of symptoms before initiation of therapy is 12-17 months (7, 24). There are several treatment options for SAC. If the cysts are not clinically evident, conservative treatment with careful observation may be a justifiable option, especially for children or high-risk surgical patients (7, 12, 18, 22, 23).

Symptomatic SAC may be related to the high pressure of fluid within the cyst pressing on surrounding structures, therefore treatment with acetazolamide (AZM), a carbonic anhydrase inhibitor known to reduce CSF production, has been proposed. Limiting the amount of fluid in the cyst simulates surgical decompression of the cyst, however its effectiveness is still questionable (7, 23).

Surgery is the gold standard of treatment and should be considered in patients with progressive worsening of symptoms or relapse (7, 16, 18, 22, 24). Surgical marsupialization with or without laminectomy is the most commonly used technique (1, 7, 11). However, large arachnoid cysts that span multiple spinal segments require extensive longitudinal exposure and bony removal increasing the rate of complications, including surgical site infection, hematoma, and spinal instability (19).

In these cases selective bony windows at the margins of the cyst and placement of a cystoperitoneal drain can be considered and it represents a viable, less invasive alternative approach to effective cyst decompression (7, 11, 19, 22). During this procedure is possible to use an ultrasound to directly visualize the cyst, to monitor CSF flow across the area and to confirm disappearance of the cyst (20).

After surgery, 60-70% of patients experience an improvement in symptoms (7, 11, 22). Motor ability has the highest response rate (71%) and the least pain (50%) (7).

Revision surgery was indicated in 12.5% of cases (7). Long-term follow-up shows no differences in quality of life between the different surgical techniques (7, 22).

The case reported here underwent exeresis of the arachnoid cyst with laminectomy and found an considerable improvement of the symptoms.

CONCLUSIONS

SACs are rare lesions that can present with various neurological symptoms: paraesthesia, neuropathic pain, paresis, and gait disturbances, much like a progressive spinal cord injury due to spinal cord compression. The SAC can also be acquired, as the result of a traumatic injury to the spinal cord. The pathogenesis remains unknown.

Conservative treatment with careful observation may be an acceptable option for asymptomatic patients. However, in patients with progressive or recurrent symptoms, surgery is the gold standard of treatment.

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