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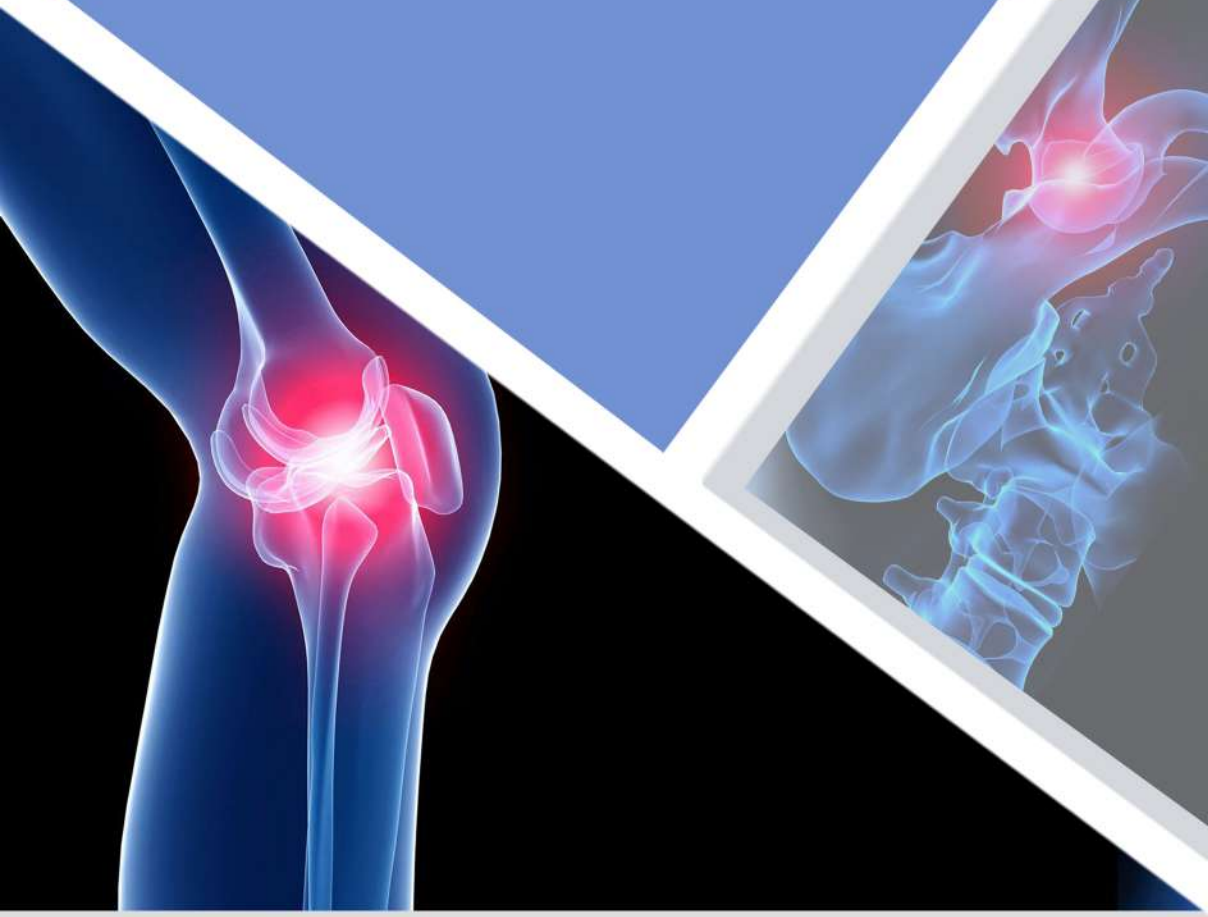

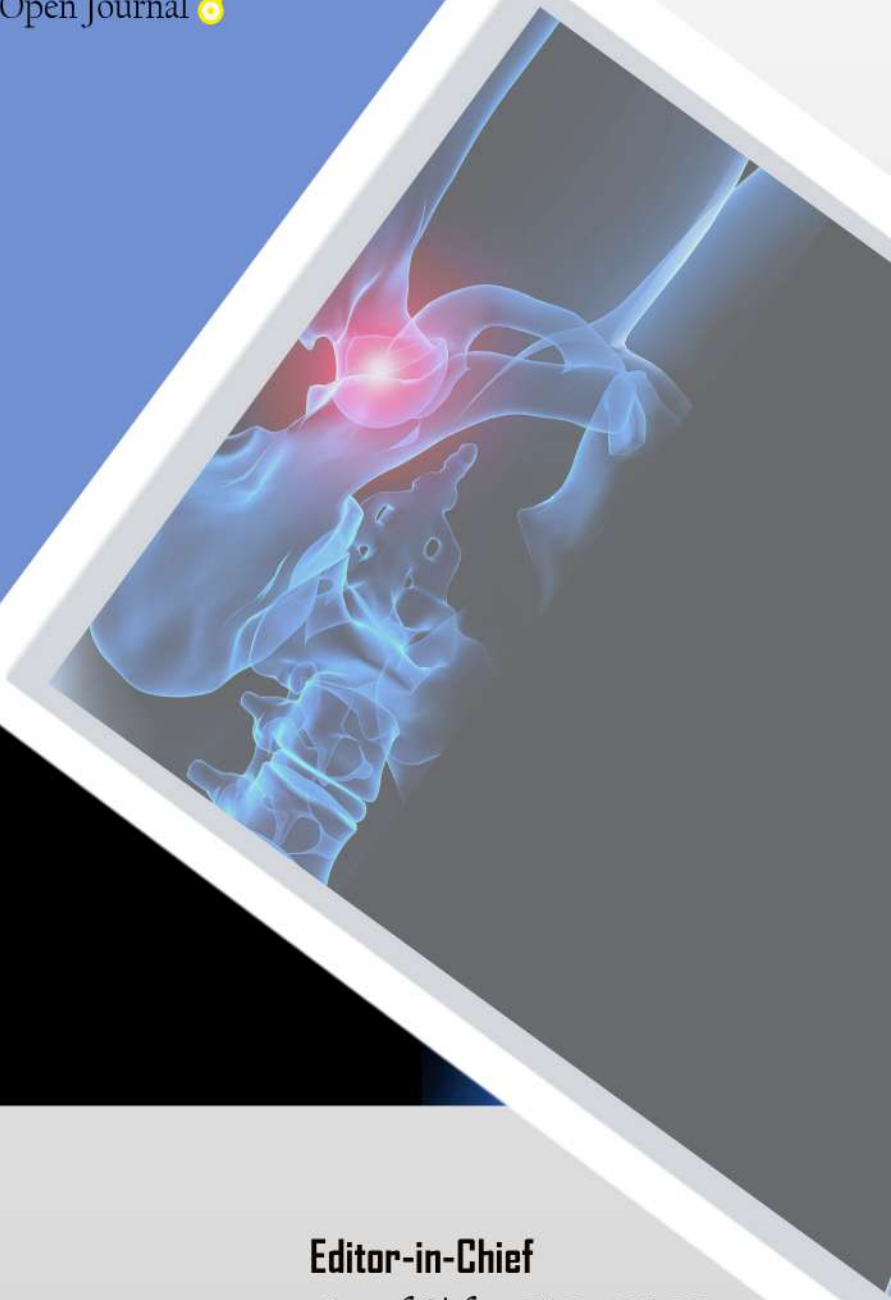

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Case Report

Giant Iatrogenic Lumbar Pseudomeningocele: A Case Report and Literature Review

Abolfazl Rahimizadeh, MD*; Housain Soufiani, MD; Shahrzad Rahimizadeh, MD; Naser Asgari, PhD; Mahan Amirzadeh, MS

Pars Advanced and Minimally Invasive Medical Manners Research Center, Pars Hospital, Iran University of Medical Sciences, Tehran, Iran

**Corresponding author*
Abolfazl Rahimizadeh, MD
Pars Advanced and Minimally Invasive Medical Manners Research Center, Pars Hospital, Iran University of Medical Sciences, Tehran, Iran; Tel. +98 21 8895 2035; Fax. +98 21 8898 9690; E-mail: a_rahimizadeh@hotmail.com

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ABSTRACT

Post-discectomy iatrogenic lumbar pseudomeningoceles are an uncommon complication. This pathology is an extradural, encapsulated, cerebrospinal fluid collection which develops at the site of previous surgery as the consequence of an unnoticed or unrepairable dural tear. A pseudomeningocele is defined as giant if it grows beyond 8 cm in length. Giant iatrogenic pseudomeningoceles with detailed information are quite rare in the literature with only 21 reported cases including the current case. Herein, we present a middle age woman with a giant pseudomeningocele which had developed subsequent to L4-L5 discectomy.

Keywords
Iatrogenic lumbar pseudomeningocele; Pathology; Discectomy.

INTRODUCTION

Post-discectomy pseudomeningocele was reported first in 1947 by Swanson and Fincher.[1] Three-years later in 1950, Winkler et al[2] reported two additional cases of a pseudomeningocele after a lumbar discectomy. In 1968, Miller et al[3] reported 10 new cases and classified pseudomeningoceles into congenital, iatrogenic and traumatic. Congenital pseudomeningoceles have been described in the patients with Marfan disease and neurofibromatosis mostly in the thoracic and lumbosacral regions respectively[4,5]

Majority of the traumatic pseudomeningoceles develop subsequent to blunt traumatic events and are mostly seen in the cervical spine, but seldom in the lumbar region or with penetrating injuries.[6-8]

Iatrogenic pseudomeningoceles may occur in the lumbar, cervical or thoracic regions in decreasing frequency.[9-13] In the lumbar region, they are mostly seen following the laminectomy for lumbar disc herniation or canal stenosis.

The cause of these cystic cerebrospinal fluid-containing lesions are incidental dural tears.[9-13] The growth of the iatrogenic lumbar pseudomeningoceles is limited but in rare occasions it may continue to grow, till the pressure of the cyst's contents and the surrounding tissues reach to equilibrium.[9-13] Most of the pseudomeningoceles remain relatively minute (below 5 cm in size) and a small number will grow above 5 cm in size which are classified as large. Very rarely this pathologic sac may grow beyond 8 cm being classified as giant subtype.[14-16]

In 1963, Miller et al[3] were the first to report 3 cases with giant iatrogenic pseudomeningocele and since then several cases with detailed information have been published so far.[17-27] Herein, we present a middle age woman with giant pseudomeningocele which had developed after L4-L5 discectomy. With consideration of the previously published cases of giant pseudomeningocele with detailed information, the current case will be the 21st in the literature. The detailed information of 21 cases with iatrogenic giant lumbar pseudomeningoceles including the current case are presented separately (Table 1).

CASE PRESENTATION

A 49-year-old female who had undergone a lumbar laminectomy for a central L4-L5 disc herniation eight months earlier was referred to our facility after the appearance of a large subcutaneous midline lumbar mass which was associated with severe low back

Table 1. *Data on the Surgical Hardware Used in the Course of Surgical Treatment of Patients with Fracture-Dislocations of the PH*

Author	Year	Age	Sex	Lumbar Procedure	Clinical Picture	Outcome
Miller et al[3]	1963	F	40	Low lumbar laminectomy	Low back and sciatic pain	Good
Miller et al[3]	1963	M	35	L5- S1 laminectomy	S1 radiculopathy	Good
Miller et al[3]	1963	M	31	L4-L5 laminectomy	Headache & LBP relieved by lying down	Good
Rinaldi et al[17]	1969	50	M	L3-L4 hemilaminectomy	Radiculopathy	Good
Rinaldi et al[17]	1969	42	F	L5-S1 discectomy	LBP, bilateral S1 radiculopathy	Good
Rinaldi et al[18]	1970	49	M	Subsequent to discectomy for recurrent L4-L5 disc	LBP, Unilateral, radiculopathy	Good
Schumacher et al[19]	1988	39	M	L3-L4 hemilaminectomy+ discectomy,	Radiculopathy	Good
Lee et al[20]	1992	18	M	Right L4-L5 discectomy+ PLIF	Persistent LBP, Right radiculopathy	Good
Lee et al[20]	1992	25	F	Right L4-L5 hemilaminectomy	Palpable mass, L5 Paresthesia	Good
Pavlou et al[21]	2005	59	F	L4-L5 discectomy	Weakness of dorsiflexion	Good
Hamilton et al[22]	2009	51	M	3 time surgery for L3-L4 spondylolisthesis	Retroperitoneal mass on left ureter	Good
Weng et al[23]	2010	26	F	L4-L5 laminectomy discectomy	Not described	Good
Liu et al[24]	2011	40	M	L4-L5 laminectomy& Screw-rod fixation	Radiculopthyy & Postural headache	Good
Alvarez et al[25]	2018	39	M	L4-L5 interlaminar-Laminectomy + discectomy	LBP & radiculopathy	Good
Alvarez et al[25]	2018	92	M	L3-L4 & L4-L5 laminectomy	LBP, Right leg radiculopathy	Good
Eneke et al[15]	2018	64	F	L4 &L5 laminectomy, L4-L5 discectomy	LBP, Radiculopathy, N claudication	Fair
Hamdan et al[26]	2018	26	F	L5-S1 discectomy	Episodes of black out while lying	Good
Rahimizadeh et al[14]	2019	30	F	L5-S1 interlaminar laminectomy+ discectomy	LBP & left S1 radiculopathy	Good
Rahimizadeh et al[14]	2019	52	F	Multilevel laminectomy	Neurogenic claudication	Good
Jah	2021	38	M	L4-L5Inter- laminectomy, discectomy	LBP & subcutaneous lump	Good
Current case	2021	49	F	L4-L5Inter- laminectomy, discectomy	LBP & Left L5 radiculopathy, Lump	Good

Figure 1. *A Photograph of the Lumbar Region; Shows a Giant Subcutaneous Lump*

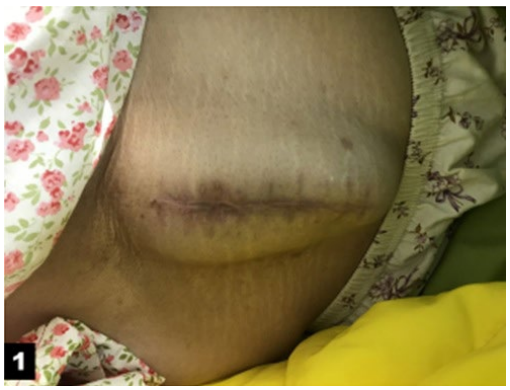
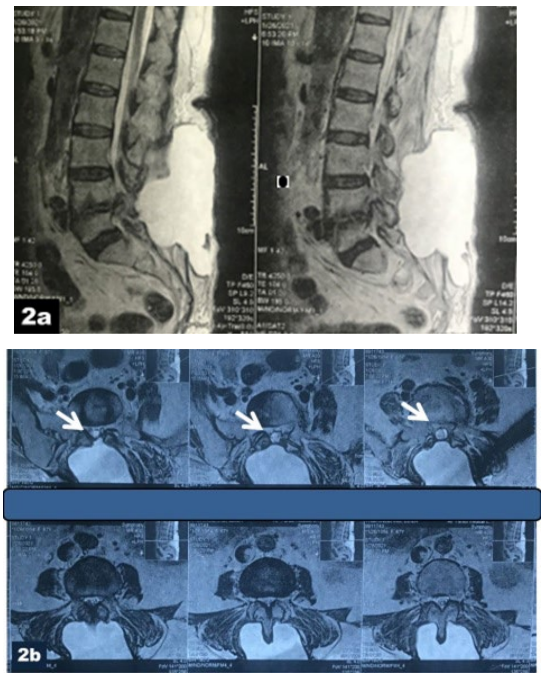


Figure 2. (a) *T2 Weighted Sagittal MRI, Shows a Giant Pseudomeningocele Extending from L2 to S4, (b) T2 Weighted Axial Images Showing the Narrow Stalk of the Pseudomeningocele (white arrows)*



pain for a duration of 3-months (Figure 1). Neurological examination of motor and sensory nerves was normal. With the diagnosis of a pseudomeningocele, magnetic resonance imaging (MRI) was performed and displayed a very large pseudomeningocele at the site of the previous surgery. The pseudomeningocele was extended from L2 to S4, with a total length of 13 cm (Figure 2).

At surgery, the abnormally thick wall of the sac was widely opened. After evacuation of cerebrospinal fluid and at the depth of the cavity; a small breach with slow flow of cerebral spinal fluid

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(CSF) was found (Figure 3). The defect was subsequently enlarged and closed tightly with interrupted silk sutures. This was subsequently covered with free fat graft being reinforced with suturing of the lower part of the pseudomeningocele's fibrotic wall. This was followed by a placement of a drain and closure of the wound in three layers. The pathological result of the wall of the pseudomeningocele was composed of connective tissue, mainly fibroblasts being aggregate with foamy macrophages (Figure 4). The patient recovered well-during the 4-days hospital stay and all her complaints had ceased at the time of the one month follow-up encounter. The patient had complete relief of her back pain and was doing well 5-months post-operatively. Successful excision and closure of the pseudomeningocele was confirmed in a MRI taken at six months follow-up (Figure 5).

Figure 3. *(a) An Intraoperative Photograph Showing a Breach is Demonstrated at the Bottom of the Surgical Scene. (b) An Intraoperative Photograph showing a Pediatric Nasogastric Tube Inserted intrathecal via the Breach Only to Show its Communication with Thecal Sac, so was Later Removed. (c) After Closure of the Breach, Reinforced with a Second Layer Composed of the Fibrotic Cyst's Wall*

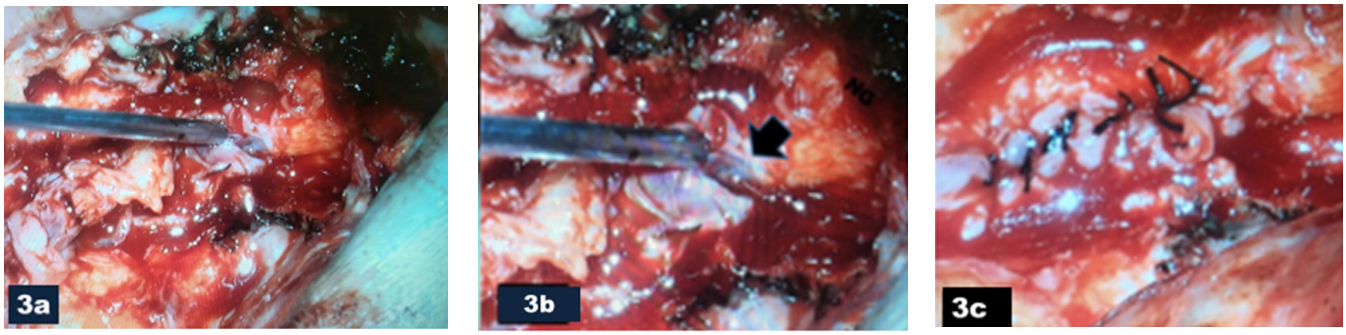


Figure 4. *Histopathologic Result of the Cyst's Wall (a) Note an External Layer being Composed of Fibroblasts. This Layer is Aggregated with Foamy Macrophages. No Epithelial Lining is Seen. (b) Histopathologic Result of the Cyst's Wall with Higher Magnification*

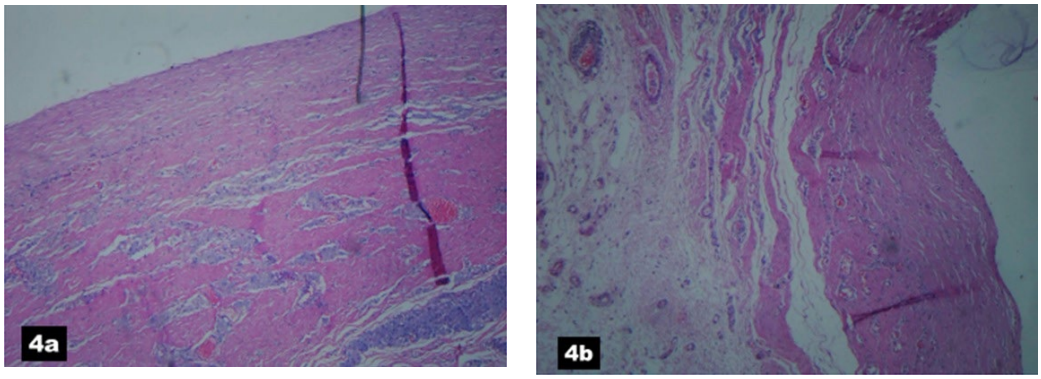
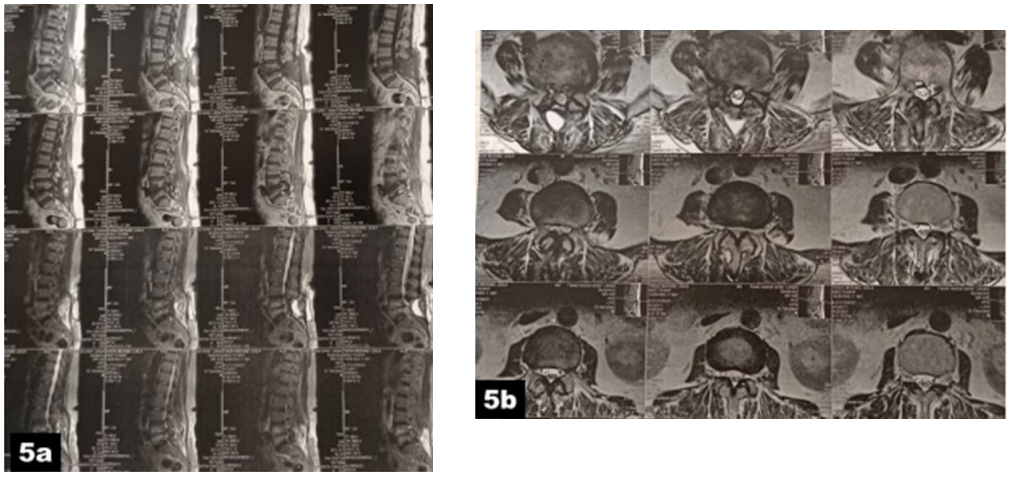


Figure 5. *Post-Operative Sagittal and Axial Lumbar Spine MRI at 6-months Follow-up, (a & b) both T1 and T2-Weighted Sagittal and Axial Images after Excision of the Pseudomeningocele, the Small Hyperintense Mass is Probably Free Fat Graft*



DISCUSSION

Lumbar pseudomeningoceles are uncommon complication of lumbar disc surgeries which develop as a consequence of an incidental or unrepairable dural tear. The true incidence of post-discectomy lumbar pseudomeningocele is difficult to ascertain because many remain asymptomatic.[16] However, it is estimated to be between 0.1 and 2%.[14,19]

A small dural tear with concomitant arachnoid layer perforation results in a gradual extradural accumulation of the cerebrospinal fluids.[14,16,28] According to one theory, the small dural tears leads to a higher probability of pseudomeningocele formation with respect to ball-valve mechanism.[28,29] With subsequent reactive fibrosis, the fluid will be enveloped resulting in a cystic mass with false walls or pseudomeningoceles.[16-27]

Unnoticed dural tear with intact arachnoid and a ball valve mechanism will result in the development of a true cyst lined with arachnoid.[29] This type of iatrogenic cyst is called true meningocele where surrounding connective tissue might reinforce the arachnoid capsule with time.[14,16,28]

Higher frequency of the pseudomeningoceles in the lumbar region in comparison to thoracic and cervical region is due to the relatively higher CSF pressure in the caudal thecal sac and the more frequency of lumbar disc surgeries.

The size of the lumbar pseudomeningoceles in majority remain minute, but in a small number of the cases, in a time frame which varies from a few months to a year after laminectomy, the pathology continue to grow till it reaches to an equilibrium. In rare occasions, a pseudomeningocele might grow beyond 8 cm.

Weng et al[23] have attributed the giant size of a pseudomeningocele to high body mass index. However, we believe that intra-operative extensive dissection of the paravertebral muscles and fatty degeneration of these muscles might be the other predisposing factors.

Clinically, most of the lumbar pseudomeningoceles remain small and asymptomatic, this is in contrast to symptomatic ones which are relatively large. Large pseudomeningoceles might present a large subcutaneous lump. Nonetheless, in symptomatic cases, low back pain (LBP) which characteristically tends to be aggravated with straining and Valsalva maneuver is the most frequent clinical feature.[30] If a rootlet is extruded through the breach and trapped within, radiculopathy may coexist.[16,31-33] Such manifestation is clinically quite similar to a recurrent lumbar disc herniation.[16,31-33] Rarely, in those with anterior dural breach, some rootlets might be trapped in corresponding collapsed intervertebral disc space.[34] Occasionally, lower limbs motor dysfunction and incontinence and even *cauda equina* syndrome might occur.[35] Headache as well as syncope and hypotension may be caused by compression of the subcutaneous lump of the pseudomeningocele. Positional headache has been also described in an iatrogenic lumbar pseudomeningocele.[36] Positional syncope is another rare presentation of pseudomeningoceles which has been reported in a case report. The patient's symptom disappeared following the excision of the pseudomeningocele.[37] Headache due to chronic subdural hematoma is another rare complication of the pseudomeningoceles.[38] Clinical features of meningitis have been reported in an infected pseudomeningocele.[39,40]

An abdominal mass due to retroperitoneal growth of a pseudomeningocele is another rare presentation of the scenario.[41] Hydronephrosis secondary to ureteral obstruction caused by a retroperitoneal growth of a giant pseudomeningocele has been reported in a rare case report.[22]

On plain radiographs, erosion of the surrounding bones might be seen in long standing cases.[42] Ossification of the cyst's wall is an infrequent scenario.[43-47]

The degree and extent of ossification can be best demonstrated in reconstructed computed tomography (CT) images.[47]

Computed tomography myelography can detect the small pseudomeningoceles, even in those tiny ones that grow intraosseously.[48]

Magnetic resonance imaging remains the most useful diagnostic tool for the demonstration of a pseudomeningocele and its short fistulous tract. A pseudomeningocele displays low signal intensity in T1-weighted and high signal intensity in T2-weighted MRI images.[49] Furthermore, the measurement of the length of the pseudomeningoceles and their classification to minute, large and giant became possible with the aid of MRI.[16-27] Generally, this specific CSF-containing mass is located posterior to the dural sac; although in rare instances it might grow into the intervertebral disc space and even progress into retroperitoneal space.[22,50]

Owing the high chances of spontaneous regression, conservative treatment is recommended for asymptomatic cases.[51] Spontaneous regression may occur within 3-months to a few years following the diagnosis.[51] According to Solomon et al[51] healing of the dural defect with the gradual resorption of cerebrospinal fluid is the possible mechanism for the spontaneous resolution of pseudomeningoceles.[16,52] Surgery for a symptomatic lumbar pseudomeningocele starts with widely opening the cyst and closure of the dural breach itself.[14-20] For closure of the breach, both interrupted suture and titanium U shape clips can be used.[14-16,52,53] In the case of a radiculopathy where entrapment of a rootlet is responsible; reduction of the rootlet into the thecal sac through the breach is the key to adequate treatment.[16,31-33]

Following the closure of the breach, lumbar myofascial flap was introduced by Misra et al.[54] Myofascial flap which can be achieved with advancement of lumbar paravertebral muscles has been advocated in those with large dead space.

CONCLUSION

Incidental dural tears with CSF leakage during lumbar laminectomy should be properly addressed. Lumbar pseudomeningocele should be suspected in patients with a delayed reappearance of

lower back pain or radiculopathy within a few months to several years after the initial laminectomy. Appropriate surgical intervention should be decided upon and undertaken once the diagnosis is reached. There remains no difference in the management of large and giant pseudomeningoceles.

CONSENT

The authors have received written informed consent from the patient.

CONFLICTS OF INTEREST

The authors declare that they have no conflicts of interest.

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Case Report

Adipofascial Turnover Forearm Flap for a Large Defect in Hand after Squamous Cell Tumor Extirpation. A Case Report

Miot Boncy D, MD*

Orthopedic and Hand Surgeon, Centro Médico Docente Los Altos, Av El Lago, Municipio Carrizal 1203, Miranda, Venezuela

Corresponding author
Miot Boncy, MD
Orthopedic and Hand Surgeon, Centro Médico Docente Los Altos, Av El Lago, Municipio Carrizal 1203, Miranda, Venezuela; Tel. +58424-1560993;
E-mail: davemiot@gmail.com

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ABSTRACT

Squamous cell carcinoma is a cancer that is rare in the hand. The extirpation of this cancer can leave an important cutaneous defect in many cases. The cutaneous covering at the back of the hand is a challenge for any surgeon, especially when there are large skin defects with exposure of tendons, bones or neurovascular structures. Here, we describe a clinical case where an adipofascial turnover forearm flap and full thickness skin graft were performed, after the extensive extirpation of the well-differentiated squamous cell carcinoma has been made.

Keywords
Flap; Adipofascial; Forearm; Hand; Skin defect; Carcinoma; Tumor; Squamous cell.

INTRODUCTION

The skin carcinomas[1] are a common cancer in the world; squamous cell carcinoma (SCC) is derived from the intermediate layer of the epidermis. The frequency is 2:1 men and women, which is very rare before 50-years-old but more frequently around the 70-years-old.[2] This is an infiltrating tumor which can spread by contiguity, lymphatic system but in frequent by hematogenous route.[3] SCC metastases are very rare in the hand,[4] but predominant at the dorsal aspect of it.[5] For the treatment, various choices are available, depending on the tumor type, size, location and depth, as well as the age and overall health. The options include: excisional surgery, Mohs surgery, cryosurgery, electrosurgery, laser surgery, radiation, photodynamic therapy (PDT), topical medications.

The significant defect in the dorsal aspect of the hand is always a challenge for a surgeon, because it is very easy to have exposure of tendon, muscles, bones and neurovascular structures. There are several options for soft tissue coverage, some of them sacrifice a major artery; the most common of these arteries are based on the radial artery with or without skin.[6] There are also soft tissue flaps which do not sacrifice major arteries, among them the posterior interosseous flap, dorsal ulnar flap or the adipofascial flap with skin graft.[7-10] The last one is based on the vascular anatomy of the upper limb where there are vascular arches around the joints. There are perforators arteries which anastomose the posterior and the anterior interosseous arteries, and also others arteries from the subcutaneous adipose tissue,[7,11,12] which provide blood supply to the dorsal fascia of the forearm. The advantages of this flap are less aesthetic problems and do not depend on a single vascular pedicle.

CASE PRESENTATION

A 64-year-old male, with a history of hypertension under treatment, was an active smoker for forty years, with no previous skin lesions. After a trauma with abrasion on the dorsum of the hand, he began to show the growth of an exophytic mass. The patient was unable to give any details about the matter. Two-months later, a biopsy was performed by a dermatologist, and the pathology report showed well-differentiated SCC, which is why he was referred to a hand surgery service. He was evaluated 10-weeks after the onset of symptoms in conjunction with a medical oncologist, where an exophytic lesion with areas of necrosis on the surface was observed. The size of the exophytic lesion was approximately 3×4 cm (Figure 1), with normal neurovascular examination, and

Figure 1. *Squamous Cell Carcinoma at Dorsum of the Hand*

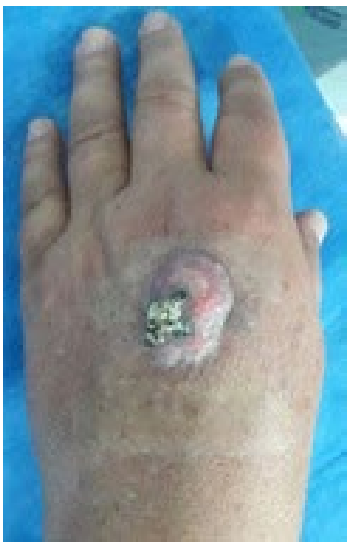


Figure 2. *MRI T1 Axial Plane of the Hand*

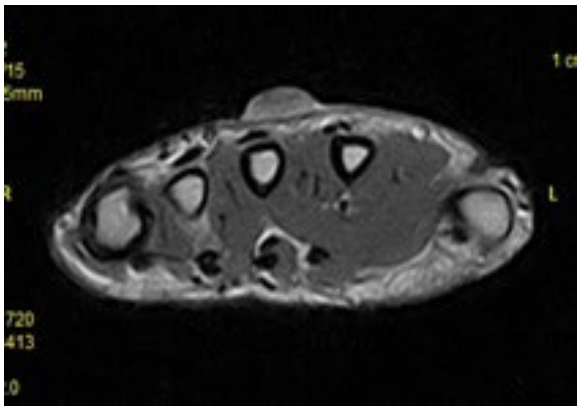
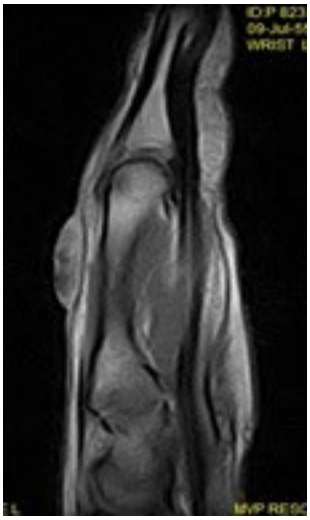


Figure 3. *MRI T1 Sagital Plane of the Hand*



no clinical observation of adherence in deep planes. An magnetic resonance imaging (MRI) study showed tumor growth in depth, without tendon involvement (Figures 2 and 3).

Under general anesthesia, a pneumatic tour niquet applied. A wide excision was made including skin, subcutaneous tissue and the tendon sheath of the extensor tendons which were under the SCC, leaving a skin defect of approximately 8×7 cm., with exposure of the common extensor tendons of the index, middle and ring fingers (Figure 4). A frozen biopsy was made and reported that 8 mm negative margin was obtained in all edges. A turnover adipofascial flap of the forearm was performed, in which an "H" incision is made on the dorsal side of the forearm (Figure 5), the dorsal skin is carefully lifted away from the adipofascial layer. The dorsal antebrachial fascia with adipose

Figure 4. *Wide Excision of the Squamous Cell Carcinoma with Margin Free Edges and Exposure of the Extensor Tendons*

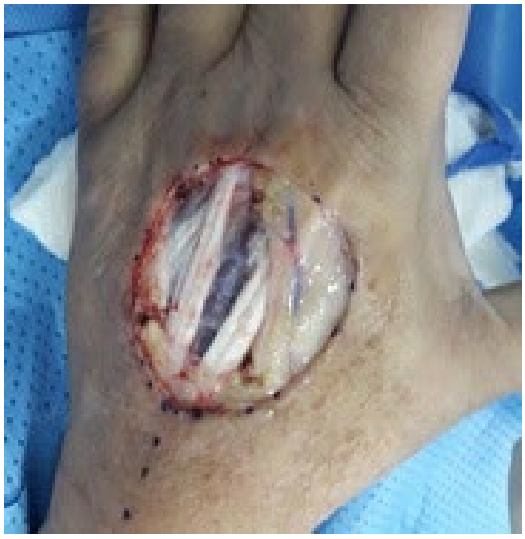
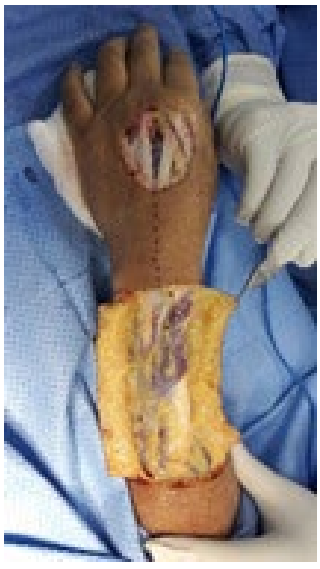


Figure 5. *"H" Forearm Incision with Exposure of the Adipofascial Layer*



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component was marked proximal after measuring the necessary length of the flap to cover the defect. To measure the length of the flap it is important to take into account the distance from the pivot point, which is around 4 to 6 cm., proximal of the radial styloid to the distal edge of the defect; and also we have to add a few millimeters more in order not to have tension on the pedicle. The adipofascial flap was dissected free from the underlying paratenon or muscles from proximal to distal. The proximal arteries and veins were ligated and the perforators vessels were coagulated. The flap was passed through a skin tunnel in the dorsal wrist (Figure 6). After flap suture, it was covered with a full thickness skin graft (Figure 7); a Penrose drain was placed in the closure of the forearm, and finally immobilization was made with an antebrachial digital splint. The d rain was removed on the third post-operative day, the splint 14-days after, and physical therapy started the third week. Outpatient follow-up was performed every two-weeks, the skin graft had a partial necrosis which did not need any further procedure (Figures 8A and 8B). The patient showed a satisfactory post-operative outcome after two-months, with integration of the skin graft, and full motion of the fingers and wrist was recovered (Figure 9). The complete follow-up took one-year, when he was discharged from medical oncology service.

Figure 6. *Adipofascial Turnover Flap Passed Through a Skin Tunnel at the Wrist*

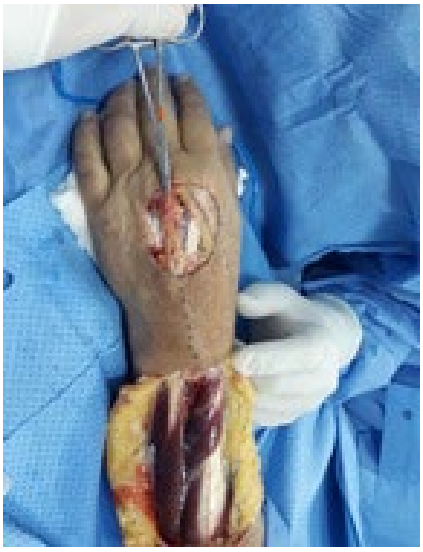


Figure 7. *Adipofascial Turnover Flap Covered with a Full Thickness Skin Graft*

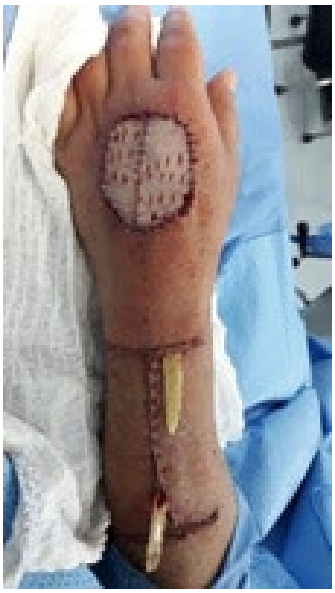


Figure 8. *A) Post-operative Evolution at 10-days, B Partial Necrosis of Skin Graft at 5-weeks*

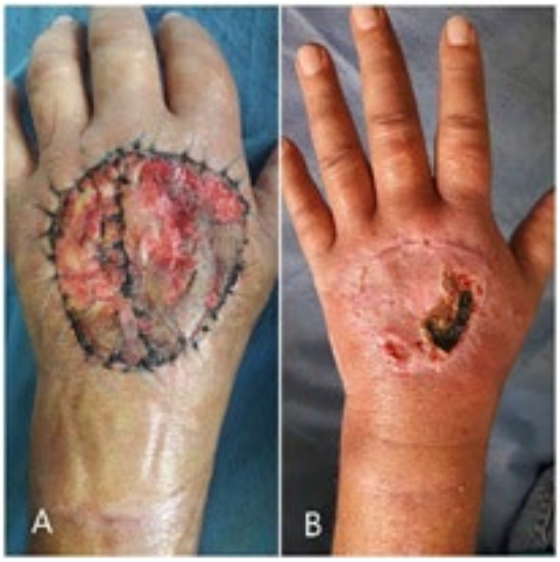
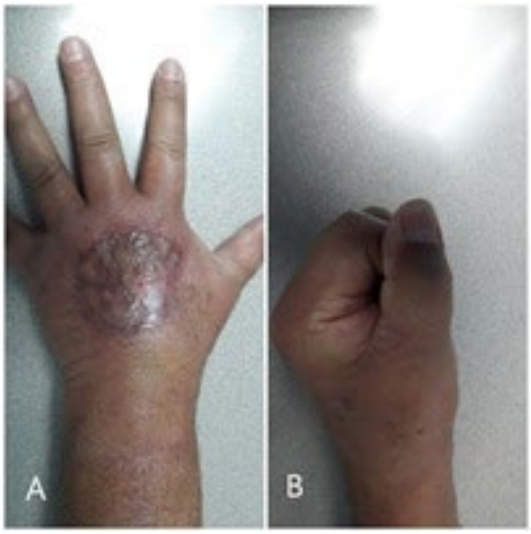


Figure 9. *A) 8-weeks Post-operative with Finger Extension and Integration of the Skin Graft, B) Fingers Flexion*



DISCUSSION

In this case, there has been a rapid growth of the SCC which is not very common because, generally, the growth is slower. That is

why it was necessary to make a wide excision in order to extirpate the tumor, which left a large cutaneous defect, which could not be covered with local flaps.

When there is exposure of the bone or tendons without their sheath, we cannot place skin grafts directly because it compromises the tendon gliding, and/or the viability of the skin graft.[13] In such cases, the options for the cutaneous coverage are used: regional, distant or free flaps. The majority of these procedures require prior microsurgical training. Most regional flaps are based on an axial pattern of vascularity of minor arteries. In older patients or smokers, when the vascular pedicle of the posterior interosseous or dorsal ulnar flap is rotated, it may present problems in blood supply. Other options are flaps based on major arteries, but sacrificing the radial or ulnar arteries.[6,14] Lai et al[15] in 1991, was the first one to describe adipofascial turnover flap based on the vascular arches around the joints, for the reconstruction of the skin defect in the dorsal aspect of the hand and finger, and is frequently used to cover the back of the fingers more than the back of the hand.[16,17] In the forearm, the vascularity, not only depends on the anastomosis of the interosseous arteries, but also on the contribution of the dorsal arch of the wrist, reducing then the chances of necrosis. The skin graft can be performed at the same time of the adipofascial flap or in a second surgery.

In the literature, there are few cases of SCC at the back of the hand, but we did not find any article in which the coverage of the skin defect was performed with this adipofascial turnover flap after oncological extirpation of SCC.

This flap is reliable, straightforward, and allows a good coverage surface, with good cosmetic result. It can be used safely to cover the complex wounds with exposed tendon, bone, or neurovascular structures, and since it does not require microsurgical training, this is a flap that can be performed by Orthopedics or Hand Surgeons without experience in microsurgery, for traumatic and non-traumatic reconstruction.

CONSENT

The written and verbal consent of the patient could not be obtained to reproduce the information and photographs that appear in this article despite multiple attempts to contact him. The author confirms that the submitted article is not being considered or previously published.

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