C A S E R E P O R T S

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Surgical treatment of pleurocutaneous fistula in a patient 20 years after radiation therapy and breast-conserving surgery

Hirurško lečenje bolesnice sa pleurokutanom fistulom 20 godina posle poštedne operacije dojke i zračne terapije

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Abstract

Introduction. Radiation therapy after breast-conserving surgery substantially reduces the risk of local disease recurrence and moderately reduces the risk of cancer-related death. However, long-term side effects are related to numerous factors, including patient age, comorbidities, total radiation dose, fraction size, and extent of disease. **Case report.** We present the case of a 61-year-old female patient who developed an ulceration at the center of a postoperative scar, accompanied by progressive tissue destruction that eventually reached 6 × 3 cm in size, resulting in rib exposure. Twenty years ago, she had a conservative surgery of the right breast due to invasive ductal carcinoma with postoperative radiation therapy to the right chest wall and the draining lymphatics. A simple mastectomy with operative removal of all sclerotic and calcified tissue, with reconstruction of the right thoracic wall, was performed by a multidisciplinary team comprising a surgical oncologist, thoracic surgeon, and plastic surgeon. The right rib cage defect of 12 × 12 cm was reconstructed with Mersilene® mesh. A large 21 × 15 cm island musculocutaneous *latissimus dorsi* flap was raised and transposed for reconstruction of the soft tissue defect. One thoracic and one wound drain were set in place. The postoperative course was uneventful, and the patient was discharged on the 12th postoperative day. She had no complications after a six-month follow-up. **Conclusion.** Long-term complications of radiation therapy are rare, but they are usually accompanied by severe consequences that require a multidisciplinary approach in complex surgical treatment.

Key words:
breast neoplasms; fistula; radiotherapy; surgical procedures, operative.

Apstrakt

Uvod. Radioterapija nakon poštedne operacije dojke smanjuje rizik od ponovnog lokalnog pojavljivanja bolesti i umereno smanjuje rizik od smrti izazvane rakom. Međutim, njeni dugoročni neželjeni efekti povezani su sa mnogobrojnim faktorima, uključujući starost bolesnika, komorbiditete, ukupnu dozu zračenja, veličinu frakcije, i stepen bolesti. **Prikaz bolesnika.** Prikazujemo bolesnicu staru 61 godinu koja je razvila ulceraciju u sredini postoperativnog ožiljka, praćenu progresivnom destrukcijom tkiva koja je na kraju dostigla veličinu 6 × 3 cm, što je rezultiralo ekspozicijom rebra. Pre dvadeset godina imala je poštednu operaciju desne dojke zbog invazivnog duktalnog karcinoma, sa postoperativnom terapijom zračenjem desnog zida grudnog koša i pripadajućih limfnih čvorova. Jednostavnu mastektomiju sa operativnim uklanjanjem svih sklerotičnih i kalcifikovanih tkiva, sa rekonstrukcijom desnog torakalnog zida, uradio je multidisciplinarni tim sačinjen od onkohiruga, torakalnog hirurga i plastičnog hirurga. Defekt desnog rebarnog luka 12 × 12 cm rekonstruisan je Mersilene® mrežicom. Za rekonstrukciju defekta mekog tkiva podignut je i premešten „ostrvski“ muskulokutani *latissimus dorsi* režanj veličine 21 × 15 cm. Postavljeni su jedan torakalni dren i jedan dren za ranu. Postoperativni tok protekao je uredno i bolesnica je otpuštena 12. postoperativnog dana. Nije imala komplikacije nakon šestomesečnog praćenja. **Zaključak.** Kasne komplikacije terapije zračenjem su retke, ali su obično praćene teškim posledicama koje zahtevaju multidisciplinarni pristup u kompleksnom hirurškom lečenju.

Ključne reči:
dojka, neoplazme; fistula; radioterapija; hirurgija, operativne procedure.

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Introduction

Radiation therapy (RT) after breast-conserving surgery substantially reduces the risk of local disease recurrence and moderately reduces the risk of cancer-related death. RT also affects healthy tissue, causing early or long-term side effects. The latter ones mostly engage superficial skin layers and seldom progress to deeper structures, causing fistula with the respiratory tract, with only a few patients described [1].

Late consequences of radiation occur in a small percentage of patients with breast cancer who were irradiated and operated on, most often after more than 10 years from the radiation therapy [2]. The frequency of rib fractures is 1.8%, while tissue necrosis occurs in only 0.18% of patients who had tissue necrosis with a breast defect and also received chemotherapy before radiation [3].

Case report

We present a 61-year-old woman with pleurocutaneous fistula (PCF) after RT. Initially, in 1997, she underwent conservative surgery on the right breast due to invasive ductal carcinoma. The patient was followed up for the first 5 years after radiation and surgery, and she did not come for check-ups after that period. Postoperatively, she was treated with right chest wall (CW) and draining lymphatics RT. The telecobalt unit was used in a tangential field technique on the CW and parallel opposed fields to the supraclavicular and axillary regions. She was prescribed a dose of 40 Gy in 15 fractions over 15 days and an additional 10 Gy in 4 fractions. Bolus 0.3 mm was used to treat the CW. The axilla and supraclavicular regions were treated on the second day using two parallel opposing fields, prescribing a mid-plane dose of 40.0 Gy in 15 fractions over 15 days. After RT, she developed sclerosis of the right breast and paresis of the right brachial plexus.

Twenty years later, in July 2017, a small ulceration developed in the middle of the postoperative scar with progressive tissue destruction. By the time of admission to the Institute for Oncology and Radiology of Serbia, the lesion had reached a size of 6 × 3 cm, with rib exposure, and the remaining breast tissue was extremely sclerotic (Figure 1). The histopathology report of the ulceration biopsy showed no sign of malignancy.

Head, spine, pelvis, abdominal, chest X-ray, ultrasonography, and multislice computed tomography (MSCT) showed no sign of metastatic disease. Chest MSCT revealed fractures of the third to sixth ribs (Figure 2A), along with large breast calcifications and a partial pneumothorax (Figure 2B). The patient had no other comorbidities. Bacterial and fungal culture tests were negative.

Fig 1 – Ulceration in the middle of the postoperative scar with tissue destruction and rib exposure.

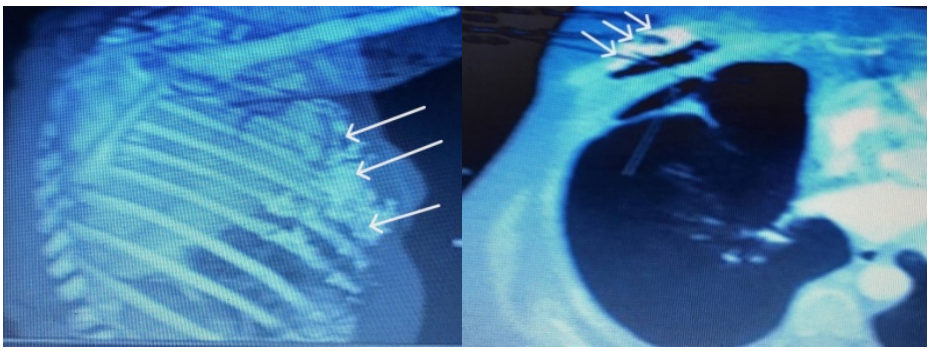
A)  **B)**

Fig 2 – Multislice computed tomography of the thorax shows A) fractures of the third to sixth ribs (indicated by white arrows), B) a calcified breast (indicated by white arrows) with a partial pneumothorax.

Operation plan

A simple mastectomy with the removal of calcified and sclerotic tissue was planned. In order to reconstruct thoracic wall defects, several options were discussed. The pectoralis major muscle could not be utilized due to *en bloc* adhesions with fibrotic and calcified breast tissue. The use of a transverse rectus abdominis muscle flap was precluded by the patient's obesity, and free flap reconstruction was not feasible due to the lack of technical support. A pedicled *latissimus dorsi* (LD) myocutaneous flap was planned for tissue defect coverage, while a Prolene™ mesh was selected for reconstruction of the thoracic wall defect.

Intraoperative finding

The procedure was performed by a multidisciplinary team that included a surgical oncologist, thoracic surgeon, and plastic surgeon. During breast excision, extensive areas of calcified tissue were identified, predominantly in the central and lower lateral poles. Parts of the pectoral major and minor muscles were also resected due to severe fibrosis (Figure 3A, B). Calcification and fractures were identified in the proximal portions of the excised third to sixth ribs, as well as part of the seventh rib (Figure 4A, B). No evidence of communication with pulmonary tissue was observed. Histopathological analysis of the excised fibrotic parietal pleura (Figure 5) revealed necrotic

A)   B)

Fig. 3 – A) and B) a simple mastectomy and resection of calcificated and fibrotic tissue.

A)   B)

Fig. 4 – A) and B) the resection of calcified ribs.

Fig. 5 – Excision of parietal pleura.

granulation tissue. The right rib cage defect measuring 12 × 12 cm was reconstructed using a polyester fiber mesh (Figure 6). A large island musculocutaneous LD flap, measuring 21 × 15 cm, was raised and transposed to reconstruct the soft tissue defect. One thoracic and one wound drain were placed accordingly (Figure 7A, B).

The postoperative course was uneventful, and the patient was discharged on the 12th postoperative day. A six-month follow-up showed no complications (Figure 8).

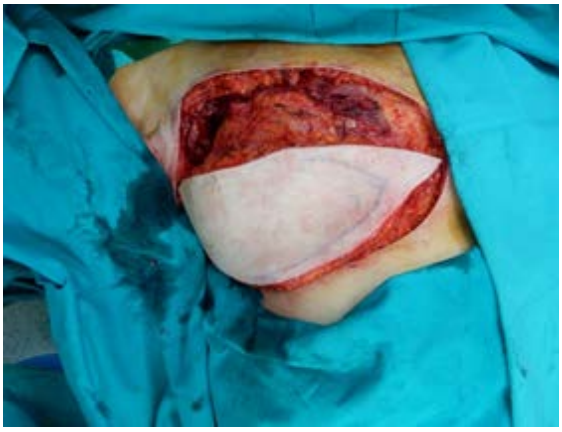
Discussion

PCF is a very rare late RT complication (after mastectomy) [4]. Our patient is the first case in the literature of PCF as a late RT complication after breast-conserving surgery.

RT is the most commonly used therapy for local disease control after breast-conserving surgery. Twenty years ago, when our patient was treated, a high daily radiation dose

Fig. 6 – Reconstruction of 12 × 12 rib cage defect with polyester fiber mesh.

A)



B)

Fig 7 – A) and B) reconstruction with pedicled myocutaneous *latissimus dorsi* flap.

Fig. 8 – The patient's appearance 6 months after the operation.

regimen was employed, consisting of 40 Gy delivered in 15 fractions, with an additional 10 Gy administered to younger and higher-risk patients. Overlapping of tangential fields that irradiate the chest or breast and the supraclavicular fields that radiate the lymphatics may occur. In the presented case, the tissue defect was located at the overlap of the supraclavicular and tangential fields, suggesting that it resulted from an excessive radiation dose delivered to the tissue. The importance of fraction size [1, 5] became apparent over time. Large fractions are no longer considered acceptable for this type of radiation for patients. Nevertheless, this therapy was routinely applied for many years in the past, and late side effects can manifest months, years, or even decades after RT. Pulmonary fibrosis, along with pleural and parenchymal changes, is recognized as a late effect of RT; however, these findings are mostly asymptomatic and are typically detected only on computed tomography scans [6]. In the presented case, RT left consequences on both skin and pleura, with additional calcifications that may represent a late manifestation of RT effects [7, 8]. Different responses to RT can be related to age, menopause, blood pressure, or smoking, but genetic differences are likely to represent most of the variations seen among patients [9].

Surgical treatment of such a patient was challenging due to the extensive CW defect. The challenge is not only the reconstruction of the CW defect but also the restoration of CW rigidity with minimal deformity, preservation of pulmonary mechanics, and prevention of air leakage and herniation of pulmonary tissue [10]. The type of reconstruction is determined by the size, location, and complexity of the defect, the viability of surrounding tissues affected by prior surgery and RT, as well as the patient's comorbidities, taking also in consideration the experience of the multidisciplinary team. In the current literature, there is no consensus regarding the indications for CW reconstruction. One might expect that reconstruction depends mainly on the extension and location of the full-thickness defect. Defects > 5 cm in diameter or including ≥ 4 ribs, situated in the anterolateral part of the CW, that cause paradoxical motion and have a high risk of lung herniation should be reconstructed. In contrast, reconstruction may not be necessary for some large apicoposterior defects, up to 10 cm in size, due to support provided by the scapula and shoulder girdle [11].

The first step in CW reconstruction is to re-establish skeletal stability. The ideal material should be flexible enough to conform to the shape of CW yet rigid enough to prevent paradoxical motion. Additionally, it should be biologically inert and radiolucent [12]. Today, we have a large number of different synthetic, biological, and metallic materials, but none of them have proven to be clearly superior. In our case, we used polyester fiber mesh. Following the restoration of CW stability, soft tissue and mesh coverage must be achieved. The best type for soft tissue reconstruction is the flap, which contains muscle in its composition. A muscle flap provides a good blood supply that can prevent possible infection of the prosthetic material, bulkiness for dead space obliteration, and, when necessary, contributes to the restoration of both structure and function. Although various options are available in the era of microsurgery, technical limitations in our setting made microsurgical reconstruction unfeasible, making flap selection particularly challenging. The optimal solution was the use of a pedicled LD myocutaneous flap, often considered the workhorse of CW reconstruction. It is the largest single flap that can be harvested. It can be tailored to any size and shape, while its long vascular pedicle makes the flap especially suitable for large skin defects [13].

Necrosis of the chest as a late effect of radiation therapy after breast cancer occurs most often on the skin or cartilage and can rarely appear as a fistula that communicates with the lungs or bronchus [14, 15]. In those cases, ST of the fistula is very challenging. Azuma et al. [4] demonstrated an effective approach for managing fistulas, where the defect was reconstructed using an omental graft. In their report, they presented a patient who, 25 years earlier, underwent a Halsted mastectomy and was irradiated postoperatively, and subsequently developed a bleeding fistula. A free omental flap with vascular anastomosis and a skin graft was implanted in the neck region, successfully closing the pulmonary cutaneous fistula. Postoperatively, emphysema remained inside the omentum, but it resolved spontaneously over time [16].

There are different techniques of flap surgery using healthy tissues, but each has some disadvantages. Han et al. [16] described cases in which the defect was reconstructed using a muscle-sparing vertical LD incision. In two patients with bronchopleural fistulas, reconstruction was performed using a muscle-sparing vertical LD flap, and thus, there was no need to change the patient's position.

A chest defect is often accompanied by a chronic infection. In their paper, Wang et al. [17] showed how they solved the operative fistula with chronic osteomyelitis. In the first act, a resection of the fistula, ribs, and clavicle was performed, as well as a partial resection of the sternum, and then the infection was repaired. In the second act, a reconstruction was made by myocutaneous incision with pedicles, with a satisfactory clinical effect. All these studies indicate various possibilities for solving chest defects, as well as the complications and risks that accompany surgical treatment. New techniques have enabled a greater choice of treatment and improved efficiency in treatment.

Conclusion

The treatment of radiation therapy complications is challenging due to extensive tissue destruction, engagement of other chest organs and structures, and severe tissue sclerosis. Surgical solutions should be individually tailored based on a multidisciplinary team assessment.

Conflict of interest

The authors declare no conflict of interest.

R E F E R E N C E S

1. *Fletcher GH.* Hypofractionation: lessons from complications. Radiother Oncol 1991; 20(1): 10–5.
2. *Fehlauer F, Tribius S, Höller U, Rades D, Kuhlmey A, Bajrovic A,* et al. Long-term radiation sequelae after breast-conserving therapy in women with early-stage breast cancer: an observational study using the LENT-SOMA scoring system. Int J Radiat Oncol Biol Phys 2003; 55(3): 651–8.
3. *Lichter AS, Lippman ME, Gorrell CR, d'Angelo TM, Edwards BK, de Moss EV.* Adjuvant chemotherapy in patients treated primarily with irradiation for localized breast cancer. In: *Harris JR, Hellman S, Silen W,* editors. Conservative management of breast cancer. Philadelphia: J. B. Lippincott Co.; 1983. pp. 299–310.
4. *Azuma R, Kajita M, Kubo S, Kiyosawa T.* Radiation-induced thoracic necrosis with pulmonary cutaneous fistula repaired using a free omental flap: a case report. BMC Surg 2019; 19(1): 14.
5. *Powell S, Cooke J, Parsons C.* Radiation-induced brachial plexus injury: follow-up of two different radiation schedules. Radiother Oncol 1990; 18(3): 213–20.
6. *Schratter-Sehn AU, Schurawitzki H, Zach M, Schratter M.* High resolution computed tomography of the lungs in irradiated breast cancer patients. Radiother Oncol 1993; 27(3): 198–202.
7. *Davis SP, Stomper PC, Weidner N, Meyer JE.* Suture calcification mimicking recurrence in the irradiated breast: a potential pitfall in mammographic evaluation. Radiology 1989; 172(1): 247–8.
8. *Robertson JM, Clarke DH, Pevzner MM, Matter RC.* Breast conservation therapy. Severe breast fibrosis after radiation therapy in patients with collagen vascular disease. Cancer 1991; 68(3): 502–8.
9. *Turesson I, Nymn J, Holmberg E, Odén A.* Prognostic factors for acute and late skin reactions in radiotherapy patients. Int J Radiat Oncol Biol Phys 1996; 36(5): 1065–75.
10. *Seder CW, Rocco G.* Chest wall reconstruction after extended resection. J Thorac Dis 2016; 8(Suppl 11): S863–71.
11. *Netscher DT, Baumholtz MA.* Chest reconstruction: I. Anterior and anterolateral chest wall and wounds affecting respiratory function. Plast Reconstr Surg 2009; 124(5): 240e–52e.
12. *Le Roux BT, Shama DM.* Resection of tumors of the chest wall. Curr Probl Surg 1983; 20(6): 345–86.
13. *Serafin D.* The latissimus dorsi muscle—musculocutaneous flap. In: *Serafin D,* editor. Atlas of microsurgical composite tissue transplantation. Philadelphia: WB Saunders; 1996. p. 208.
14. *Yoshida J, Ishimaru T, Ekimura M.* Bronchocutaneous fistula after treatment for breast cancer: a case report. Nihon Kokyuki Gakkai Zasshi 1999; 37(10): 851–3. (Japanese)
15. *O'Neill A, Beddy P.* Bronchopleural cutaneous fistula. AJR Am J Roentgenol 2008; 190(5): W315.
16. *Han SJ, Kim J, Kim S, Ha Y.* Case Report: Vertical muscle-sparing latissimus dorsi flap in the reconstruction of chronic radiation-induced chest wall ulcers after breast cancer surgery: a case series. Front Surg 2024; 11: 1397233.
17. *Wang L, Liu Z, He Z, Zhang C.* Autologous myocutaneous flap implantation for chronic refractory chest wall sinus with infection: a case report. J Cardiothorac Surg 2023; 18(1): 121.

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