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Case Report / Olgu Sunumu

# Management of patients developing axillary pseudoaneursym after ductal stenting: Report of three cases

*Duktal stent sonrası aksiller psödoanevrizma gelişen hastaların yönetimi: Üç olgu sunumu*

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***ABSTRACT***

Pseudoaneurysms develop as a result of disruption of the arterial wall due to trauma or iatrogenic reasons such as catheterization, and it is important due to the high risk of bleeding and rupture. Until recently, the main treatment of pseudoaneurysms was surgical repair. However, in recent years, minimally invasive methods such as ultrasound-guided compression and percutaneous thrombin injection have been used more frequently. In this article, the clinical course and findings of three different cases who developed pseudoaneurysm as a result of stenting the ductus arteriosus via the axillary artery were discussed.

***Keywords:*** Complication, ductus arteriosus, pediatric catheterization, pseudoaneurysm, thrombin.

***ÖZ***

Psödoanevrizmalar travma veya kateterizasyon gibi iatrojenik nedenlerle arter duvarının bozulması sonucu gelişir ve kanama ve rüptür riskinin yüksek olması nedeniyle önemlidir. Yakın zamana kadar psödoanevrizmaların ana tedavisi cerrahi onarımdı. Ancak son yıllarda ultrason eşliğinde kompresyon ve perkütanöz trombin enjeksiyonu gibi minimal invaziv yöntemler daha sık kullanılmaya başlanmıştır. Bu yazıda duktus arteriozusun aksiller arter yoluyla stentlenmesi sonucu psödoanevrizma gelişen üç farklı olgunun klinik seyri ve bulguları tartışıldı.

***Anahtar sözcükler:*** Komplikasyon, duktus arteriozus, pediatrik kateterizasyon, psödoanevrizma, trombin.

The axillary artery approach is often preferred as an alternative to the femoral artery or carotid artery for stenting of the ductus arteriosus (DA) in patients with a vertical, tortuous duct or for balloon angioplasty/valvuloplasty in neonates with critical aortic stenosis with or without aortic coarctation.[1,2] Stenting of the DA is used as an alternative to Blalock-Taussig shunt palliation in newborns with ductus-dependent pulmonary circulation.[1]

Pseudoaneurysms occur as a result of disruption of arterial wall continuity due to mechanical trauma or iatrogenic reasons such as catheterization.[3] It differs from true aneurysms since it does not contain all three arterial wall layers. There is a relatively high risk of rupture and major bleeding, and posttraumatic pseudoaneurysms are rare in children.[3]

In this article, we aimed to share three different cases and treatment modalities that developed pseudoaneurysm after stenting the DA via axillary artery.

## CASE REPORT

***Case 1–*** A male patient born via cesarean section (C/S) at 39 weeks with a birth weight of 2950 g was diagnosed with ventricular septal defect and pulmonary atresia during echocardiographic evaluation. A written informed consent was obtained from the parent of the patient. Four days later, the patient underwent DA stenting in the angiography unit. The echocardiogram revealed a vertical, tortuous, 2-3-folded DA. The right axillary artery was accessed under ultrasound guidance for the first puncture, and a 4F sheath was placed. A 3.5×15 mm

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coronary stent was successfully positioned in the DA, resulting in an increase in oxygen saturation from 60-65% to 95% after the procedure. The intervention lasted 50 min, with 24 min of fluoroscopy time. After achieving hemostasis, the patient received intravenous heparin infusion for 48 h, with dosage adjustments based on activated clotting time measurements. After two days of uneventful intensive care follow-up, heparin infusion was stopped, low-molecular-weight heparin (LMWH) and acetylsalicylic acid was started, and the patient was transferred to the cardiology service. Four days later, a 30×30 mm pulsatile mass was observed in the right axillary region. Doppler ultrasound and computed tomography (CT) confirmed the presence of a 33×27 mm pseudoaneurysm (Figure 1). As there was no reduction in pseudoaneurysm dimensions on the second day, the patient underwent primary repair by evacuation of the pseudoaneurysm sac. The patient, whose normal flow pattern was observed after the surgery, was discharged after acetylsalicylic acid and clopidogrel treatments were administered, and LMWH treatment was discontinued. The patient was scheduled for outpatient follow-up. At the six-month follow-up, there was no evidence of nerve damage.

***Case 2–*** A female patient born via C/S at 39 weeks with a birth weight of 2900 g was diagnosed with ventricular septal defect and pulmonary atresia during echocardiographic evaluation. A written informed consent was obtained from the parent of the patient. Ductal stent placement was planned for the fourth postnatal day in the angiography unit. The right axillary artery was accessed under ultrasound guidance for the third puncture, and a 4F sheath was placed. A 3.5×15 mm coronary stent was successfully positioned in the DA, resulting in an increase in oxygen saturation from 74 to 94%. The procedure lasted 20 min, with a fluoroscopy time of 12 min. After achieving hemostasis, intravenous heparin infusion was administered for 24 h, followed by a transition to intravenous LMWH and oral acetylsalicylic acid. Heparin infusion was stopped. On the third day of follow-up, the patient was transferred to the inpatient service. On the fifth day, a pulsating, nonfluctuating firm mass in the right axillary region prompted an arterial and venous Doppler ultrasound, which revealed a cystic lesion measuring 48×26 mm with yin-yang flow (bidirectional flow due to the circulation of blood within the pseudoaneurysm) in the right axillary-subclavian region. Anticoagulant treatments were halted, and pulse and hematocrit levels were monitored. Due to an increase in hematoma size, a CT angiography was performed, showing a hematoma filling and expanding the right axillary fossa, along with contrast extravasation and contour irregularity in a segment of the axillary artery (Figure 2a, b). The patient was transferred to the intensive care unit for clinical follow-up and underwent surgery on the second day of follow-up due to further hematoma enlargement and decreased hematocrit levels. The pseudoaneurysm sac was evacuated, and primary repair was conducted. Postoperatively, normal flow patterns were observed, and LMWH treatment was resumed two days after surgery. After seven days of intensive care follow-up, the patient was transferred to the ward and commenced oral acetylsalicylic acid and clopidogrel while discontinuing LMWH treatment. The patient was discharged for outpatient follow-up. At the four-month follow-up, there was no evidence of nerve damage.

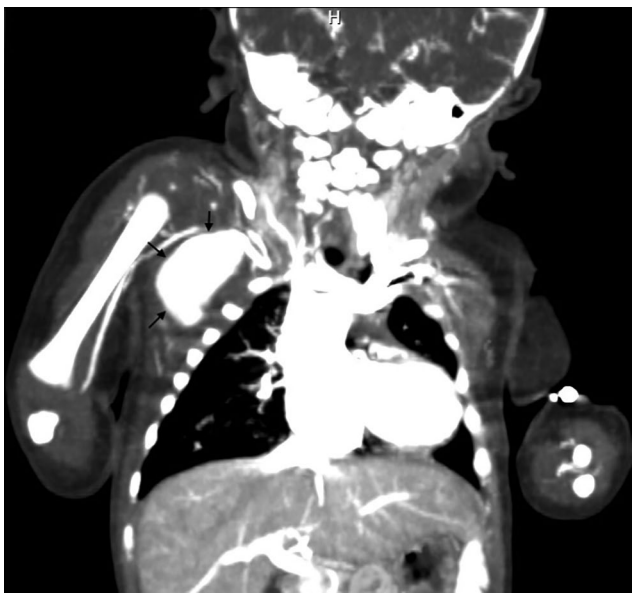


**Figure 1.** Contrast-filled pseudoaneurysm image in the right axillary region on computed tomography angiography.

***Case 3–*** A female patient born at 38 weeks by C/S with a birth weight of 3300 g was hospitalized in the pediatric cardiac intensive care unit with a diagnosis of complete atrioventricular septal defect and pulmonary atresia on echocardiographic evaluation. A written informed consent was obtained from the parent of the patient. Ductus arteriosus stent placement was scheduled for the fourth postnatal day in the angiography unit. The right axillary artery was accessed under ultrasound guidance for the first puncture, and a 4F sheath was inserted. A 4×15 mm coronary stent was successfully positioned in the DA, resulting in an increase in saturation from 74 to 95%. The procedure lasted 32 min, with a fluoroscopy time of 11 min. Due to prolonged bleeding, an activated clotting time

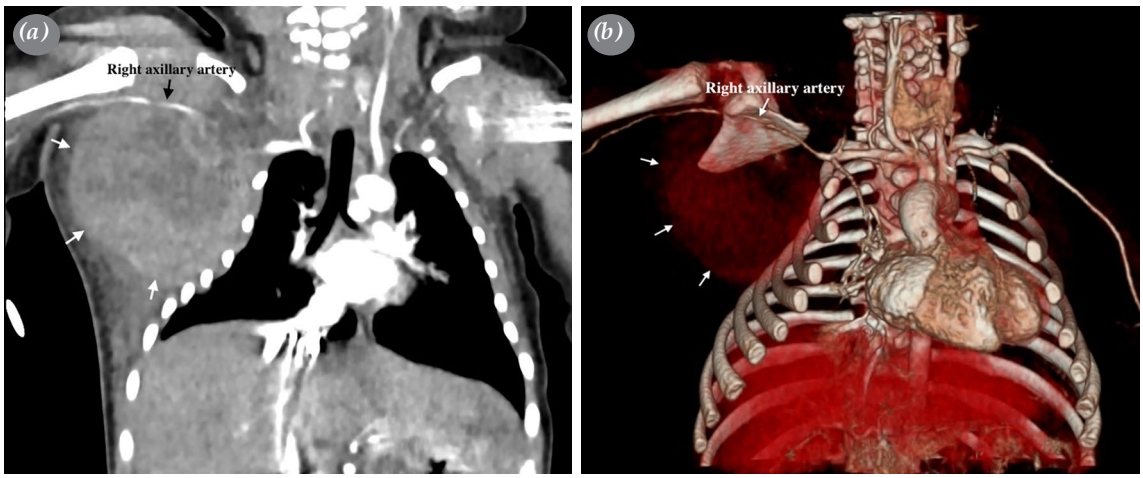
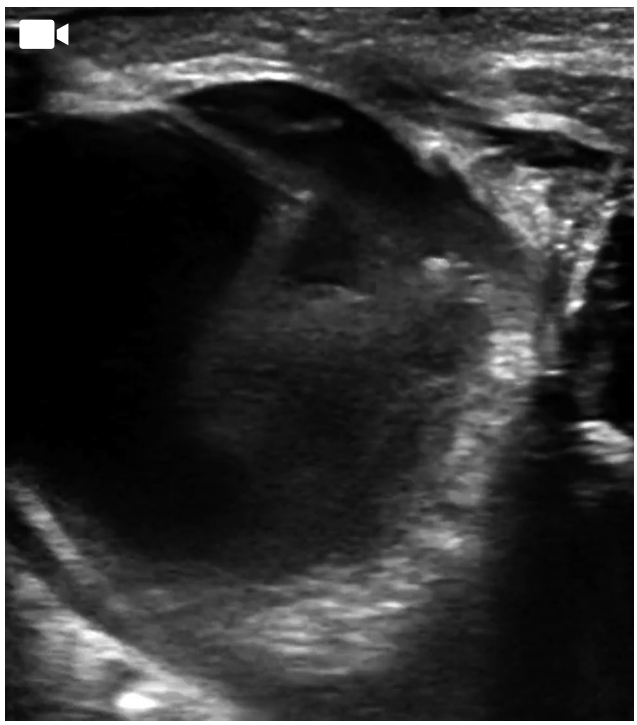



**Figure 2.** Contrast extravasation in the right axillary region and contour irregularity in the axillary artery on computed tomography angiography. **(a)** Two-dimensional coronal section, **(b)** three-dimensional image.

>600 sec and activated partial thromboplastin time >174 sec, heparin infusion was not initiated after the procedure, and LMWH was started instead. During patient follow-up, a small hematoma of approximately 10×10 mm was observed in the right axillary region. Hematoma size and pulse were monitored daily. On the seventh day after the procedure, there was a sudden increase in hematoma size and a 2 g/dL decrease in hemoglobin levels. Doppler ultrasound revealed a 35×33 mm pseudoaneurysm formation and active hemorrhage into the pseudoaneurysm. Despite ultrasound-guided compression for 10 min, bleeding in the aneurysm persisted. The patient was evaluated by the Interventional Radiology Department, and they offered thrombin injection as a treatment option. Subsequently, 1 mL of human thrombin with a concentration of 500 IU/mL (Tisseel lyo; Baxter International Inc., Deerfield, IL, USA) was injected into the pseudoaneurysm with a 22-gauge (G) needle under ultrasound guidance, resulting in thrombus formation (Video 1). The patient's distal circulation and pulse were closely monitored without any apparent issues. Unfortunately, the patient died suddenly 16 h after the procedure due to pulmonary hemorrhage, whose underlying pathology we could not understand. The patients' demographic, procedural, and treatment characteristics are summarized in Table 1.



**Video 1.** Thrombin injection into the neck of the pseudoaneurysm.

## DISCUSSION

Pseudoaneurysm formation in infants is rare and most commonly iatrogenic.[4] Potential risk factors associated with pseudoaneurysm formation are anticoagulation, large sheath size, short-term manual compression, simultaneous catheterization of the artery and vein, difficult compression of the site, and superior femoral artery puncture.[3] Furthermore, hematoma, dissection, bleeding, and nerve damage are the most common complications of axillary intervention.[2]

Karmegaraj et al.[3] observed pseudoaneurysm formation in 2.6% of 77 patients who underwent

**Table 1. Demographic, procedural and treatment characteristics of the patients**

| Diagnosis | VSD and pulmonary atresia | VSD and pulmonary atresia | Complete AVSD and pulmonary atresia |
|---|---|---|---|
| Age at intervention (day) | 4 | 4 | 4 |
| Age at PSA treatment (day) | 12 | 11 | 11 |
| Weight (g) | 2950 | 2900 | 3300 |
| Puncture site | Right axillary | Right axillary | Right axillary |
| Number of punctures | 1 | 3 | 1 |
| US guidance | Yes | Yes | Yes |
| Sheath profile | 4F | 4F | 4F |
| Procedure time (min) | 50 | 20 | 32 |
| Fluoroscopy time (min) | 24 | 12 | 11 |
| Pseudoaneursym size (mm) | 33×27 | 48×26 | 35×33 |
| Treatment | Surgery | Surgery | Thrombin injection |

VSD: Ventricular septal defect; AVSD: Atrioventricular septal defect; PSA: Pseudoaneurysm; US: Ultrasound.

axillary stent placement within six years. Breatnach et al.[1] observed pseudoaneurysm in one of 20 patients who had stented axillary intervention. In the 2.5-year experience of our center, the axillary route was preferred in 34 of 149 patients with ductal stent placement, and pseudoaneurysm formation was observed in three (8.8%) patients.

Pseudoaneurysm treatment methods are surgery, ultrasound-guided compression, endovascular methods (coil embolization, stent graft placement in the pseudoaneurysm neck), and percutaneous thrombin injection.[5] Conservative treatment may be attempted in small-sized pseudoaneurysms.[3] We also initially followed our patients clinically but planned an intervention due to the decrease in hematocrit levels and significant increase in pseudoaneurysm dimensions. The surgical approach is end-to-end anastomosis after resection of the lesion or repair with a donor artery, with high success rates increasing with advances in microsurgical techniques.[4] However, due to the surgical difficulty brought on by small arteries, particularly in infants, it is increasingly being replaced by minimally invasive procedures, such as ultrasound-guided compression or percutaneous thrombin injection.[3,4]

Rapidly expanding pseudoaneurysms, pseudoaneurysms larger than 18 mm, infected pseudoaneurysms, distal ischemia caused by local pressure of the pseudoaneurysm on the artery, neuropathy caused by local pressure on the nerve, and ischemic soft tissues and skin caused by local pressure are among the indications for pseudoaneurysm treatment. Surgery is preferred to noninvasive techniques in some patients with pseudoaneurysms that are infected, rapidly expanding, and cannot be treated percutaneously.[3,5]

Ultrasound-guided compression was first described by Fellmeth et al.[6] in 1991 and became a preferred procedure in the treatment of pseudoaneurysms until the widespread use of thrombin injection.[5] However, it is less preferred in children since the procedure is painful, requires sedation and analgesia, and has high recurrence rates in patients using anticoagulants.[4] We tried this method in one of our patients but later administered thrombin injection in the same patient since it was unsuccessful.

Cope and Zeit[7] used thrombin injection to treat pseudoaneurysm in adult patients for the first time in 1986, and it required a while for the method to become widespread.[5] The first successful treatment with thrombin injection for the treatment of pseudoaneurysm in children was used by Frush et al.[8] in 2000 for femoral artery pseudoaneurysm. In the last 20 years, many successful cases have been reported, and thrombin injection has been used as an alternative to surgery in both children and adults for the treatment of pseudoaneurysm.[3,8] The most common complication after thrombin injection is distal embolization, and its incidence was reported as 2%.[5] To avoid embolization, the needle tip should be directed away from the pseudoaneurysm neck during

injection, and a small size of the syringe should be used.[3,8]

In our first two patients, the surgical method was preferred due to the relatively large size of the pseudoaneurysm, the limited experience of thrombin, and the presence of an experienced surgical team. Our patients, who underwent pseudoaneurysm resection and end-to-end anastomosis, had no complications during their follow-up. We consulted the third patient with the Interventional Radiology Department, and they suggested thrombin injection as a treatment option. Since the neck of the pseudoaneurysm was suitable for injection and we had never tried it before, we decided to try thrombin injection as an alternative treatment option to surgery. Successful and rapid thrombus formation was observed after 1 mL of thrombin injection. There were no complications related to the distal circulation of the extremity during the one-day clinical follow-up. However, since the patient died, no information on the long-term follow-up is available.

In conclusion, as a result of increased arterial interventions in the pediatric population, an increase in the incidence of pseudoaneurysm is unavoidable. Although surgery is the standard treatment for pseudoaneurysms, ultrasound-guided percutaneous thrombin injection should be considered in appropriate cases.

**Data Sharing Statement:** The data that support the findings of this study are available from the corresponding author upon reasonable request.

**Author Contributions:** Idea/concept, design, control/supervision: İ.C.T.; Data collection and/or processing, references and fundings: A.N.G.; Analysis and/or interpretation, literature review, writing the article: D.K.; Critical review: A.C.H.; Materials: S.B.

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