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Case Report

Pneumorrhachis and Pneumocephalus Following Epidural Analgesia: Case Report and Systematic Review

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Abstract

We present a case of pneumorrhachis (PR) and pneumocephalus (PC) following labor epidural analgesia and conducted a systematic review of published studies on PR or PC after epidural anesthesia. A 36-year-old female, gravida 4, para 1 (one uncomplicated term pregnancy and two abortions), received epidural anesthesia for pain control during labor at 40 weeks of gestation with an uncomplicated vaginal delivery. About 12 hours after the epidural, the patient experienced muscle and upper back pain that progressed to a positional headache. Imaging revealed an air collection within the intraspinal and intracranial space. The patient underwent a series of conservative management and hyperbaric oxygen therapy. Our systematic review of 20 case reports from 1982 to 2023 included 24 patients who developed PR or PC post-epidural analgesia/anesthesia. Mean age was 46 ± 18.6 years, and 83.3% of the patients were female. 54.2% described obstetric patients, 76.9% of which underwent vaginal delivery. Loss of syringe technique was used in 62.5% of cases, of which 93% used loss of resistance to air. PR or PC following epidural anesthesia is rare, however obstetric patients have higher risk given the use of epidural anesthesia in labor.

Introduction

Lumbar epidural anesthesia is an effective and increasingly popular option for labor analgesia.[1] Pneumorrhachis (PR) or pneumocephalus (PC) is a rare complication of epidural anesthesia in which air collection presents in the intraspinal canal and intracranial space respectively. The condition is typically asymptomatic and air is reabsorbed spontaneously, though it can rarely be symptomatic and associated with discomfort, pain, and neurological deficits.[2, 3] Anesthesiologists administering epidural anesthesia typically utilize loss of resistance to air (LORA) or loss of resistance to saline (LORS) technique to identify the epidural space. At present, there is no established superiority of either technique.[4] We are presenting a case study and performing a systematic review to identify potential risk factors associated with PR or PC following epidural anesthesia.

Case Presentation

A 36-year-old woman, gravida 4, para 1 (one uncomplicated term pregnancy and two abortions) at 40 weeks gestation with a history of anemia and asthma, was admitted in labor. The patient received lumbar epidural anesthesia for labor analgesia at the L3-L4 level with an 18-gauge 3.5-inch Weiss needle using the LORA technique. The patient underwent an uncomplicated spontaneous vaginal

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delivery two hours later. Anesthesia evaluation immediately following delivery was within normal limits and patient was hemodynamically stable with no pain.

Approximately nine hours after delivery, the patient reported a positional headache with relief while in supine position and neck and upper back pain since delivery that was nonresponsive to NSAID and muscle relaxant therapy. The patient's initial diagnosis was a post-dural puncture headache (PDPH) given the patient's symptoms and recent epidural anesthesia procedure. Subsequent MRI exams of the cervical spine and brain showed large longitudinal air collection predominantly in the ventral epidural space and in the extra-axial space, suggestive of PR and PC respectively (Figure 1). The patient was placed in supine position, given continuous high flow oxygen, and acetazolamide 250-500 mg twice a day to relieve intracranial pressure and reduce meningeal inflammation, without resolution of symptoms. The patient was deemed a candidate for hyperbaric oxygen therapy in treatment of PR and PC.

A clinical decision was made to treat the patient with Modified Navy Dive Table 5, as the pathology underlying venous air collection was compared to an arterial gas embolism. Furthermore, the patient was treated in a monoplace center with limited access to Navy Dive Table 6. Following hyperbaric oxygen therapy, the patient's pain decreased from a 9/10 to 1/10 with resolution of headache and improvement in neck pain. Repeat CT head/spine without contrast continued to show PR throughout the cervical and thoracic spine despite symptom resolution. The patient was discharged and completed 3 outpatient hyperbaric oxygen treatments with continued neurology follow up.

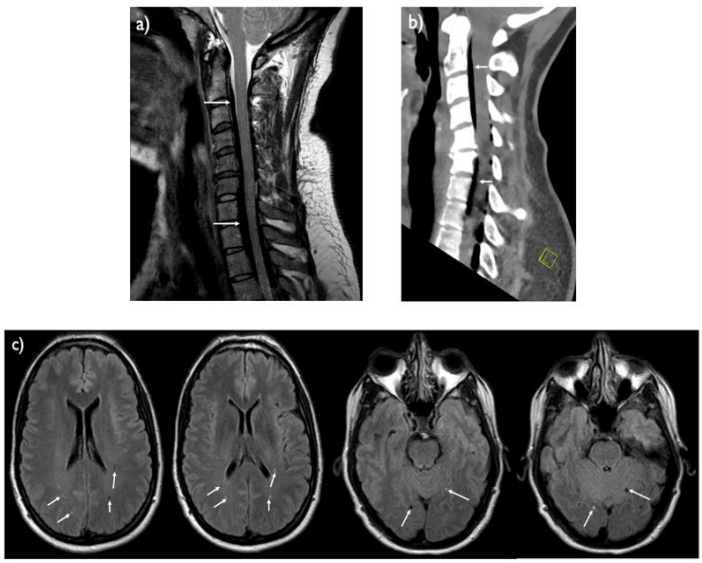

Figure 1: *Sagittal plane MRI, T2 sequence, white arrows illustrating air collection in the ventral spinal canal suggestive of PR (a); Sagittal plane CT cervical spine, white arrows illustrating hypointense air collection in the ventral spinal canal suggestive of PR (b); Axial plane MRI, FLAIR sequence, white arrows illustrating bubbles within the extra-axial space suggestive of PC (c).*

Methods

We conducted a systematic review using the PubMed engine in English from 1982 to 2023 (inclusion criteria: pneumorrhachis, pneumocephalus, aerorachia, intraspinal pneumocele, epidural emphysema, and epidural pneumatosis) to identify patients with PR or PC following epidural anesthesia/analgesia. Our exclusion criteria included patients with traumatic brain injury, brain or spine injury, and pneumomediastinum. We identified an additional 20 case reports with 23 patients that fit our criteria, for a total of 24 patients assessed.

From the case reports identified, we collected patients' sex, age, past medical history, procedure for which epidural anesthesia/analgesia was indicated, technique of administering epidural anesthesia/analgesia, symptoms of PR and/or PC, and treatment of PR and/or PC. Notable events mentioned regarding their procedure or post-procedure were also collected.

Results

We identified 24 patients diagnosed with PR or PC following epidural anesthesia/analgesia (Table 1). There were 20 female patients (83.3%) and 4 male patients (16.7%). Ages ranged from 21 to 78 years; the mean age was 46 ±18.6 years. Thirteen cases described obstetric patients (54.2%), with 10 (76.9%) undergoing vaginal delivery and 3 (23.1%) undergoing cesarean section. The loss of resistance technique was used in 15 (62.5%) case reports, of which 14 (93%) used LORA and 1 (7%) used LORS. There were 9 cases in which technique for administering anesthesia/analgesia was not mentioned or LORA/LORS were not stated. Thirteen (54.2%) patients presented with complaints of headache; 8 (33.3%) were positional in nature. Nine (37.5%) patients showed other neurological symptoms, including diplopia, dysarthria, vision loss, paraparesis, and tinnitus. Two (8.3%) patients showed signs of seizure-like activity and 5 (20.8%) patients presented with abnormal vitals. Four (16.7%) patients exhibited loss of consciousness, two of which resulted in cardiac arrest, and one resulted in comatose state.

Eleven (45.8%) patients were managed only with conservative treatment, including observation, positioning, pain control with analgesics, and supplementary oxygen. Two (8.3%) patients were managed with an epidural blood patch, with gradual resolution of symptoms. Four (16.7%) patients underwent hyperbaric oxygen therapy with symptom resolution after one session. Two of these four patients with hyperbaric treatment were obstetric patients, including our case with epidural analgesia using LORA

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Table 1: *Systematic review of patients diagnosed with PR or PC following epidural anesthesia.*

Case	Report	Year	Age	Sex	Technique	Symptoms	Procedure	Treatment
1	Krishna and Gupta[13]	2015	34	F	LORA	Posterior postural headache and posterior neck and right shoulder pain	Vaginal delivery	Epidural blood patch
2	Liu et al[14]	2022	34	F	Unknown	Right lower extremity paralysis, dysesthesia, allodynia, decreased deep tendon reflex of the right knee and ankle	Cesarean section	High dose steroid therapy
3	Shaikh et al[15]	2021	26	F	LORA	Tachypnea and oxygen desaturation	Cesarean section	Conservative treatment
4	Shin et al[16]	2018	78	F	LORA	Hypotension and loss of consciousness, followed by cardiac arrest	Back pain relief	ICU post-arrest interventions
5	Shin et al[16]	2018	69	F	LORA	Loss of consciousness followed by cardiac arrest	Back pain relief	ICU post-arrest interventions
6	Hsieh et al[17]	2015	72	F	LORA	Persistent drowsy consciousness followed by generalized convulsant	Total knee replacement	Conservative treatment followed by emergent decompressive craniotomy
7	Romano-Ribeiro et al[5]	2022	32	F	LORS	Motor blockade	Cesarean section	Hyperbaric oxygen therapy
8	Reddi et al[18]	2015	30	F	Unknown	Positional right frontal headache and neck pain. Neck stiffness, binocular vertical diplopia on leftward gaze	Vaginal delivery	Conservative treatment
9	Reddi et al[18]	2015	32	F	Unknown	Positional right frontotemporal headache radiation to back of head and neck pain	Vaginal delivery	Conservative treatment
10	Reddi et al[18]	2015	28	F	Unknown	CSF leakage, positional holocephalic headache, lightheadedness and unresponsiveness, hypotension	Vaginal Delivery	Conservative treatment
11	Nistal-Nuño et al[19]	2022	34	F	LORA	Occipital headache	Vaginal Delivery	Conservative treatment
12	Avila[20]	2016	33	F	Unknown	Headache	Vaginal Delivery	Conservative treatment
13	Pires et al[21]	2020	35	F	LORA	Positional headache worsened with movement	Vaginal Delivery	Conservative treatment
14	Gómez-Ríoz and Fernandez-Goti[22]	2013	34	F	LORA	Frontal headache	Vaginal Delivery	Unknown
15	Jagia et al[23]	2016	52	F	LORA	Bilateral frontotemporal headache, nausea, hypertension, tachypnea	Hip replacement	Conservative treatment
16	Avellanal et al[12]	1996	71	M	Neither	Disorientation, dysarthria, deterioration of consciousness	Inguinal hernia repair	Unknown
17	Chew et al[24]	2017	75	M	LORA	Unresponsiveness, seizure-like activity, postictal drowsiness	Knee arthroplasty	Conservative treatment
18	Bolster et al[25]	2013	44	F	LORA	Headache	Colectomy	Unknown
19	Marino et al[26]	2017	78	M	LORA	Confusion, agitation, vertical nystagmus, vision loss, left paraparesis	Nephrectomy	Hyperbaric oxygen therapy
20	Thongtan et al[27]	2021	44	F	Unknown	Positional headache, nausea	Hysterectomy	Epidural blood patch
21	Yin et al[28]	2013	61	M	Unknown	Comatose state, left upward eye deviation	Aortic valve replacement	Mechanical ventilation
22	Rafiq et al[29]	2021	21	F	LORA	Headache, photophobia, tinnitus, nausea, neck stiffness	Vaginal Delivery	Conservative treatment
23	Castedo et al[30]	2021	51	F	Unknown	Drowsiness, slurred speech, altered consciousness, persistent left central facial paresis	Knee arthroplasty	Hyperbaric oxygen therapy

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technique for vaginal delivery, and the case by Romano-Ribeiro et al using LORS technique for epidural anesthesia for cesarean delivery. [5]

Discussion

In this study, we report an obstetric patient case of PR and PC following epidural anesthesia administration and included our findings to a pooled analysis of similar cases published in the literature. Our findings suggest that approximately half of patients reported with PR or PC presented with symptoms of headache; a majority were obstetrical patients. Headaches following epidural anesthesia are not uncommon; secondary causes of headache in obstetrical patients most commonly include post-dural puncture headache (PDPH) which occurs in 0.5% to 1.7% of patients who receive epidural anesthesia.[6] PDPH is classically characterized as a headache or backache developing within days after an epidural procedure as a result of excessive CSF leakage from dural puncture. The pain is exacerbated by head movement and relieved by supine positioning[7]. The PR or PC is much rarer and also commonly presents with headaches; however, it may also have other nonspecific symptoms such as altered mental status, seizure, hemodynamic instability, cerebral cortex focal deficits, lethargy, and loss of consciousness. As routine imaging is not commonly indicated in headaches that occur in the peripartum period, it is possible that obstetrical patients with PR or PC following epidural anesthesia administration are misdiagnosed with PDPH given its more common prevalence. The PR or PC should therefore be considered as a possible etiology of headache in the setting of epidural anesthesia to aid in early diagnosis and treatment for quicker recovery.

Multiple theories regarding the etiology of PR and PC have been proposed, including iatrogenic air administration through epidural anesthesia/analgesia and spontaneous air uptake through the Dandy "ball valve" theory and the Horowitz and Lundsford "inverted-soda-bottle-effect" theory. The Dandy theory hypothesizes that air moves unidirectionally from the atmosphere to inside the cranial cavity.[8] Horowitz and Lundsford suggest that loss of CSF creates a negative intracranial pressure which results in a vacuum formation that traps air inside the cranial space. The loss of CSF as described by Horowitz and Lundsford includes physiologic causes as well, such as during Valsalva maneuvers.[9, 10] During the Valsalva maneuver, intra-abdominal and intrathoracic pressure is increased, which elevates intracranial pressure (ICP) and intrathecal pressure (ITP). The magnitude and duration of ICP and ITP increase depend on the duration and intensity of the maneuver. ICP is then transiently decreased following a Valsalva maneuver, and when paired with a lumbar puncture, causes air to be sucked inside the cranium, resulting in equalization of ICP. [11] This hypothesis regarding air trapping due to increased intra-abdominal pressure may explain the predominance of obstetric patients with PR or PC in the current literature, as patients are instructed to perform Valsalva maneuvers and deep inspiratory breaths during the delivery process for long periods of time. This may also explain the findings of Avellanal et al, whom described a patient with findings of PC during a dural puncture with no air administered by syringe; the patient had a short series of coughs with vigorous inspiratory efforts which may mimic the Valsalva maneuver.[12]

Our study had several strengths, which includes detailing a large series of patients with PR or PC after epidural anesthesia/analgesia. We identified a lack of clarify on establishing causality of the outcomes related to epidural anesthesia/analgesia techniques utilized in obstetric and non-obstetric procedures. Our study also has limitations that should be acknowledged. Firstly, PR or PC were underreported, which may result from selection bias, publication bias, and observer bias. The limited number of case reports available for analysis prevented us from performing a thorough statistical analysis. Secondly, inconsistencies in reporting may have arisen due to the authors' varying interests and the heterogenicity of patient cases, thus resulting in difficulty in synthesis of evidence and establishing causality. Thirdly, we lacked access to raw data and detailed information on each case, including unknown technique for epidural anesthesia/analgesia administration in 8 of 23 cases and unknown treatment in 3 of 23 cases. Finally, as we only reviewed literature available in English, our findings may not be representative of non-English language sources.

In conclusion, based on our systematic review of the literature from case reports, and including our own case, we have demonstrated that obstetric patients are associated with a higher risk of PR or PC following epidural anesthesia/analgesia. Vaginal delivery is associated with higher risk than cesarean section due to the prevalence of Valsalva maneuver during vaginal delivery. The LORA epidural technique is the more commonly used technique in all cases with PR or PC complications. Further research is needed to explore and identify the optimal labor analgesia techniques and delivery methods.

Data availability: The authors confirm that the data supporting the findings of this study are available within the referenced material of this article and openly accessible through PubMed at https://pubmed.ncbi.nlm.nih.gov.

Conflict of interest: The authors declare that there is no conflict of interest regarding the publication of this article.

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