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IMAGE

Editorial

Medical Education in Safety Culture

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Abstract

Safety culture in healthcare has been a practice priority in recent years. It is also one of the main directions of medical education. In a healthcare organization, most safety related events that occurred are a consequence of all workers' daily actions, habits, and lifestyles. As a result, incorporating a culture of safety into medical education is crucial for healthcare professionals and providers. Employee's competence, the role of a leadership team, knowledge management system, and evaluation process are four foundational pillars in the culture of safety education.

Key words: medical education, culture of safety education

Providing a quality medical service and patient safety are primary goals of medical organizations[1]. However, a high quality medical service is built upon the safety culture of a hospital. A culture of safety includes the entire personnel's attitude held in a workplace. A real safe and secure environment would be for both patients and workers in a hospital. Safety culture does not grow automatically; it is rooted in an effective system of safety culture education[2]. All employees should be aware of the vulnerabilities of a system, and the whole medical team must have sufficient medical knowledge, skills and attitude to promote patients' safety[3,4]. In 2012, the Next Accreditation System (NAS) was revealed by the Accreditation Council for Graduate Medical Education (ACGME) in the U.S.A.; it indicated that the focus of residency training has been shifted from educational processes to educational outcomes[5,6]. Training hospitals are engaging trainees in six important areas that affect patient outcomes; two of them are patient safety and the quality of healthcare. Thus a leadership team must establish a monitoring system for eliminating errors in patient care and foster an environment for emphasizing accountability, trust, and mutual respect among all workers. In order to let all stakeholders gain updated information in their professional field, IPE (inter-professional education) and IPL (inter-professional learning) through a real life practical teaching by total patient care would be vehicles to lead them reach this goal. In addition, an ongoing evaluation system is an instrument to measure the level of safety culture and it can serve as indicators to direct the progress of safety culture for a hospital. This article will discuss how these four essential elements (Table 1), including developing employee's competence, identifying the responsibility of a leadership team, creating an effective knowledge management system and implementing an meaningful evaluation process, should align to medical education system to support in forming safety culture in a hospital.

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Developing Employee's Competence

The capacity of building safety culture in a hospital depends on the ability of all workers to apply their safety knowledge in the workplace. Educating employees and letting them become knowledgeable in safety culture is vital. It is critical to recognize that mistakes can happen across healthcare procedures. In the culture of safety education, workers not only need to reinforce their medical knowledge and skills from clinical case studies and a real life practical teaching, but also have to acquire and apply the concept of CUS

Table 1. Pillars of safety culture education

Four Foundational Pillars of Safety Culture Education			
Employee's Competence	**Leader's Responsibility**	**Knowledge Management**	**Evaluation Process (Ongoing process)**
Belief & Attitude **CUS Model** **Clinical case study** **Real life situation teaching** **IPE & IPL protocol** **Communication skills**	**Committee foci:** Define employees' responsibility Identify hospital's priority Provide report, cross professional support system Create rewarding policy Develope measurement tools **Culture:** Trusty Blame free Self assessment or reflection	**For employees** E-library E-forum (IPE & IPL) Webinar **For patients & patient's families** Hospital web page E-media Video Safety education booklet	**Process** Pre-assessment Identify affected factors Post-assessment Intervention **Assessment Tools:** Safety documents Observation matrix Surveys Interviews

CUS: concern, uncomfortable, safety, E: electronic, IPE: interprofessional education, IPL: interprofessional learning.

model ("I am Concerned"; "I am Uncomfortable"; "STOP this is a Safety issue") in their practice. Workers participating in culture of safety education will gain updated medical information from the training of IPE and IPL as well. Communication skills on talking with other healthcare providers about the hazards and related jeopardizing issues, and the methods for protecting hospital's employees from injury or infection in their workplace, are also fundamental contents in all associated training. Culture is the atmosphere, which is made up by beliefs, values, and attitudes. Therefore, building a belief in safety and starting with a shift towards safety need to be a theme and norm for all participants in the education. A well-designed and structured culture of safety education is a foundation for maximizing the capacity of safety culture in a hospital. In addition to the contents and topics, a hospital educational department also needs to organize all different level of training programs to meet every single employee's needs based on the his or her job responsibility.

Identifying the Responsibility of A Leadership Team

A safety culture requires a committed and strong leadership, along with the assurance and empowerment of all workers. Participants of safety culture education are not just the frontline workers but also leaders and administrators from different management levels of a hospital. In such education, topics about how to enhance safety management system for a hospital should be the focus for the leadership team. It includes organizing a safety committee, defining safety responsibilities for all levels of employees, identifying priority for the hospital, providing a clear report system and cross-professional support system, creating a rewarding policy, and developing measurement tools. Leaders also need to learn how to help employees build habits to always self-assess how their daily actions are influencing a true culture that embraces safety; and how to cultivate a transparent and blame-free system of communication and establish accountability for all employees. Leadership team plays a central role in shaping a safety culture; it is important to build trust between leaders and frontline workers. By receiving sufficient culture of safety education, leadership team members are trained in their important roles as safety leaders to implement these skills in their positions. They will create a shared vision of safety culture and share the vision with all employees to involve them for promoting a positive safety culture.

Creating an Effective Knowledge Management System

Knowledge management is the process of taking, developing, distributing, and efficiently using organizational knowledge. As we understand that

culture of safety education is not a one-day or one-week workshop; employees need to keep refreshing all knowledge and skill needed. In order to maintain the atmosphere of safety culture and keep the consistency among all employees' daily work, all knowledge and resource should be accessible and available 24 hours a day, 7 days a week. E-learning is using all types of technology, including electronic and digital technology, to enable learners to access information whenever and wherever they need it. Information and message can be updated and distributed uniformly over the web or network almost instantly. Additionally, IPE and IPL can happen for medical team within webinar trainings, and feedback can be received promptly through an e-forum. Since safety culture is influenced by all individual's actions in a hospital, patients and their families also need to get appropriate safety information while they are in the hospital. Besides traditional safety education booklets, electronic media is a comprehensible guide to help educate them to be involved in promoting patients' safety. As a result, videos or multimedia slide presentations can be displayed on waiting rooms or wards' TVs. In addition, a hospital's website can deliver safety related information for patients and their families before they enter the hospital. To create an effective knowledge management system for a hospital, a wide range of e-learning formats are strongly recommended for a hospital in the twenty first century.

Implementing a Meaningful Evaluation Process

An evaluation system measures types of essential indicators that can stop a safety program or make intervention programs. A completed evaluation process includes administering pre-assessments for evaluating the readiness for safety culture change, categorizing influential elements, processing post-assessments to evaluate the effectiveness of safety education program or its related policies, and choosing proper areas for intervention. The entire process and data collection are ongoing. Usually an assessment tool will contain certain parts such as analysis of safety related documentation, matrix for workstation observation, employee pre/post training survey forms, workers safety culture survey questionnaire, and safety committee member interview forms…etc. In the culture of safety education, people who are involved in the evaluation process should get training about the usage of assessment tools and the skill to organize and analyze the assessment data. Furthermore, The leadership team needs to learn how to interpret the data and use it to redirect the management program or make an intervention or action plan for improving the culture. An evaluation can be a catalyst for transformational change. Implementing a completed evaluation process will maintain an effective program and policies, and continuously make safety culture improvement.

Conclusion

Nowadays safety culture is the pursuit of self-improvement for most hospitals. A comprehensive education system is the keystone for fostering a safety culture in a hospital[7]. It will increase all employees' readiness and ability, provide the leadership team for an adept role, and build a high quality culture of safety. So as to provide updated and accessible information, developing an e-learning platform is recommended for a hospital's knowledge management system. Finally, a training to implement an evaluation process fully will help the leadership team develop a clear map to lead the culture of safety program in the right track.

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醫學教育之安全文化教學

遲景上

童綜合醫院

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摘要

病人安全及安全文化是當代醫院追求的目標，也是醫學教育推廣過程中，愈趨重要之教學變革方向。此文化之建立需藉助教學逐漸形成，而教學過程中，醫院需倡導及落實安全文化之環境，進而建立員工之安全工作方式、態度及信任。而一個良好的全院性安全文化教學架構須包括以下四大重點支柱：1. 員工及學員安全文化行為能力之培養、2. 領導者之管理系統規劃、3. 知識之管理系統設立、4. 教學安全文化之評估，最終讓教師及學員在醫療教學過程中，一起工作、一起學習、一起成長，而達到提升全院安全文化的最終目標。

關鍵詞：醫學教育、安全文化教學

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Review Article

Amniotic Fluid Embolism

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Abstract

To date, amniotic fluid embolism (AFE) is an unpredictable and unpreventable complication of pregnancy; furthermore, it is the major cause of maternal morbidity and mortality. In this review the current knowledge about AFE is outlined, including its incidence, risk factors, etiology, pathophysiology, clinical manifestations, diagnosis, and management. It is crucial that all members of the obstetric and emergent health care team have complete knowledge of AFE to facilitate early recognition and aggressive management of the syndrome, and to improve the chance of survival of both the mother and child. Much needs to be done to further reduce the morbidity and mortality associated with AFE.

Key words: Amniotic fluid embolism, cardiogenic shock, respiratory failure, disseminated intravascular coagulation

History

The presence of amniotic fluid in the maternal circulation was first identified in 1926 by a Brazilian pathologist JR Meyer[1]. The clinical and pathologic features were described and linked in 1941 by Steiner and Lushbaugh[2]. Eight maternal deaths were reported and autopsies revealed fetal and amniotic debris in the pulmonary circulation. The deaths were attributed to the obstruction of pulmonary blood flow by debris in the lungs. Since then, amniotic fluid embolism (AFE) has been recognized as a rare but major cause of maternal mortality. In 1995, Clark et al.[3] suggested that a humoral reaction by inflammatory mediators in amniotic fluid in the maternal circulation could result in a process similar to anaphylaxis or sepsis. Evidence that amniotic fluid can be found in the circulation of asymptomatic pregnant women has led to a shift of the research focus away from the presence of amniotic or fetal material as the sole cause of AFE toward an immune reaction in susceptible individuals[4].

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Incidence

The AFE syndrome is rare. Most studies indicate that the incidence rate is between 1 and 12 cases per 100,000 deliveries[3,5-8]. The lowest incidence rates are reported in countries using validated case identification, whereas highest incidence rates are reported by retrospective analysis of large population discharge databases, particularly if no additional criteria are applied to exclude false positive cases.

Risk Factors

Several factors have been associated with AFE. They include precipitous or tumultuous labor, advanced maternal age, cesarean and instrumental delivery, placenta previa and abruption, grand multiparity, cervical lacerations, fetal distress, eclampsia, and medical induction of labor[5,8,9]. These factors are probably associated with AFE, but may not be its direct cause. AFE remains unpredictable and unpreventable.

Etiology

The pathogenesis of AFE is not completely understood. Amniotic fluid can enter maternal circulation via endocervical veins, lesions of the uterus, or the site of placental attachment. Although previously proposed explanations of the development of AFE featured a purely mechanical obstruction of the pulmonary vessels by amniotic fluid components[10], today humoral and immunological factors are considered to be responsible[11,12] because in addition to insoluble fetal components (e.g., squames), amniotic fluid contains numerous vasoactive (bradykinin, histamine, and others) and procoagulant substances that can lead to endothelial activation and a massive inflammatory reaction[11,13].

These and other immunological and clinical similarities to anaphylactic shock have led to the anaphylactoid reaction hypothesis[3,10]. However, this hypothesis is controversial[12]. There is currently no clear reason why some women tolerate the transfer of amniotic fluid or its components with no clinical symptoms or complications and others do not[12].

Pathophysiology

Once amniotic fluid enters the maternal circulation, it can precipitate cardiogenic shock, respiratory failure, and inflammatory response. As in sepsis and other causes of distributive shock, a number of reactive mediators have been implicated in the circulatory changes seen in AFE. These include prostaglandins, leukotrienes, histamine, serotonin, and proteolytics. Key among these is endothelin, a powerful vasoconstrictor found in high levels in amniotic fluid. Endothelin also constricts the pulmonary bronchi and causes depression of myometrial and myocardial contractility[14,15].

Cardiogenic shock

A biphasic pattern of cardiogenic shock in AFE has been proposed[16,17]. According to this hypothesis, the initial onset of severe pulmonary hypertension as a result of vasospasm leads to acute right ventricular failure, with a left deviation of the interatrial and interventricular septums. Moreover, there is severe tricuspid regurgitation. These events then lead to left ventricular failure due to reduction of preload and result in systemic hypotension. Supporting this biphasic hypothesis, studies that used transesophageal echocardiography to measure hemodynamic parameters during early AFE reported diffuse vasospasm of the pulmonary vasculature, elevated pulmonary arterial pressure, and right ventricular failure[18,19]. The mechanism of myocardial depression is more complicated than expected. It may be the result of hypoxic injury to the myocardium, the release of maternal inflammatory mediators, or a direct depressant effect of amniotic fluid on the myocardium[20].

Respiratory failure

Hypoxemia is the most common manifestation of respiratory failure among patients with AFE. Severe ventilation/perfusion mismatching appears to be the primary cause of hypoxemia. Factors that contribute to the ventilation/perfusion mismatching include acute pulmonary hypertension during the first phase of cardiogenic shock and cardiogenic pulmonary edema during the second phase[21]. Other contributors may include bronchospasm and noncardiogenic pulmonary edema[3,22]. Noncardiogenic pulmonary edema occurs in up to 70% of patients who survive the first several hours[22]. It generally develops as left ventricular dysfunction improves. Evidence for damage to the endothelial-alveolar membrane and a capillary leak syndrome includes the high protein concentration in edema fluid and the presence of amniotic fluid debris in the septum and the alveolar spaces.

Coagulopathy

Coagulopathy develops with severe bleeding resulting from disseminated intravascular coagulation (DIC). To date, the cause of DIC remains unclear. Amniotic fluid contains many procoagulant substances (including tissue factor and phosphatidylserine), which can lead directly or indirectly (via cytokines or complement activation) to DIC with consumptive coagulopathy and secondary hyperfibrinolysis via activation of the extrinsic coagulation cascade[11,21,23,24]. Furthermore, there is a controversial hypothesis that coagulopathy may be the result of massive hyperfibrinolysis because amniotic fluid also contains increased concentrations of urokinase-like plasminogen activator and plasminogen activator 1[25]. Current coagulation studies using rotational thromboelastometry show signs of hyperfibrinolysis

and massive hypofibrinogenemia in as early as the initial phase of AFE[26]. There are contradictory conclusions as to whether the amount of procoagulant in clear amniotic fluid is sufficient to cause clinically significant coagulopathy[16,27]. High levels of tissue factor pathway inhibitors found in amniotic fluid during late pregnancy would inhibit procoagulant activity and may actually contribute to the rarity of this condition[28].

Inflammation

It seems unlikely that obstruction of the pulmonary vasculature is the sole cause of AFE because there can be a lag of many hours between the entry of amniotic fluid into the maternal circulation and the onset of the symptoms and signs of AFE. It has been proposed that the lag may reflect the evolution of an immunologic response or inflammatory reaction to the amniotic fluid. This is supported by reports of decreased complement and increased inflammatory markers in some patients with AFE, including increased pulmonary mast cell activity[11,12,29]. According to this hypothesis, fetal antigens enter the maternal circulation via the amniotic fluid. The severity of the clinical manifestations may be related to the degree of immunologic stimulation or the balance of leukotrienes and other arachidonic acid metabolites in the amniotic fluid[30,31].

Clinical Presentation

The onset of the symptoms and signs of AFE most commonly occurs during labor and delivery or immediately postpartum[21]. Rarely, it has been reported as late as 48 hours after cesarean delivery or postpartum as well as following a first or second trimester abortion, amniocentesis, or abdominal/uterine trauma[3,32,35]. According to data from the United Kingdom (UK) Obstetric Surveillance System, approximately 56% of AFE cases occurred prior to birth and 44% of cases occurred shortly after delivery, generally within 30 min[14]. Forty-seven percent of the women were found to have premonitory signs or symptoms prior to collapse, including numbness, tingling, agitation, pins and needles in their fingers, lightheadedness, chest pain, breathlessness, feeling cold, nausea, and vomiting. In 32% of patients, a concern about fetal distress prior to a change in maternal condition prevailed[14].

The major clinical findings are the abrupt and fulminant onset of hypotension secondary to cardiogenic shock, hypoxemia and respiratory failure, DIC, and coma or seizures. The frequency of various signs and symptoms is outlined in Table 1. There appears to be a less severe presentation of AFE (i.e., partial amniotic fluid embolism), in which only some of the major symptoms and signs occur[16,36,37]. Such patients generally present with a sudden onset of milder dyspnea and hypotension. The clinical course tends to be abbreviated and the prognosis is much better compared with women who have the full syndrome.

Diagnosis

There are numerous causes of hypotension, hypoxemia, and/or hemorrhage in women who are pregnant or postpartum. They can be divided into obstetric, anesthetic, and nonobstetric causes[21]. Obstetric causes include placental abruption, uterine rupture, uterine atony, eclampsia, and peripartum cardiomyopathy. Anesthetic causes include high spinal anesthesia and local anesthetic toxicity. Nonobstetric causes include pulmonary embolism, air embolism, anaphylaxis, septic shock, massive aspiration, transfusion reaction, and myocardial infarction. Diagnosis of AFE is based on clinical symptoms after other causes/diagnoses have been excluded. AFE should

Table 1. Signs and symptoms of amniotic fluid embolism

	Frequency
Hypotension	100%
Fetal distress	100%
Pulmonary edema or ARDS	93%
Cardiopulmonary arrest	87%
Cyanosis	83 %
Coagulopathy	83 %
Dyspnea	49 %
Seizure	48 %
Uterine atony	23%
Bronchospasm	15%
Transient hypertension	11%
Cough	7%
Headache	7%
Chest pain	2%

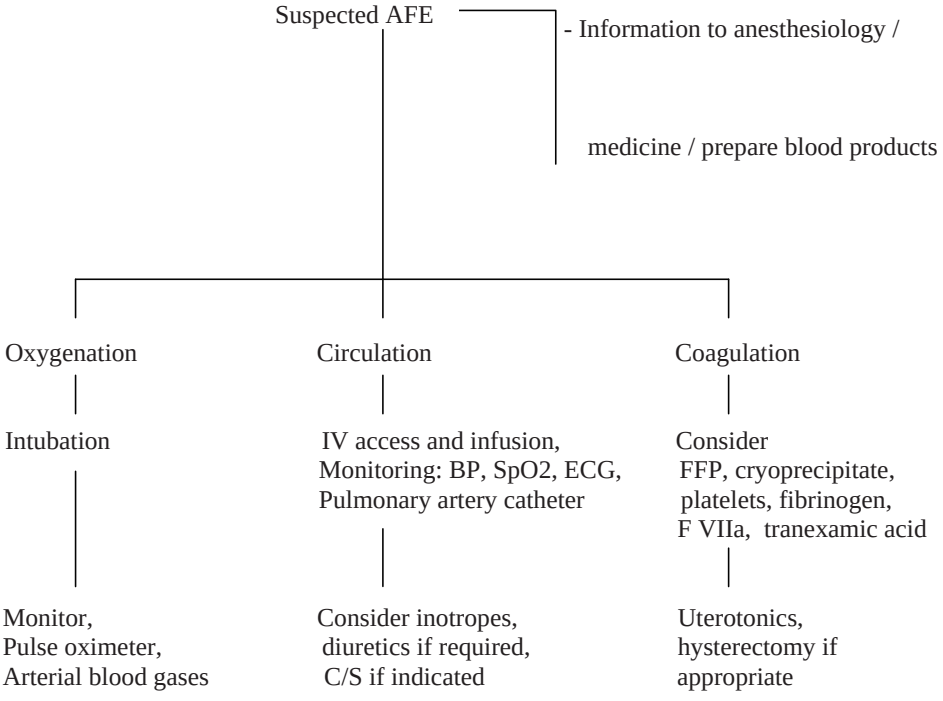
Adapted from Clark SL[3]; ARDS: adult respiratory distress syndrome.

be considered in every case of sudden maternal cardiovascular collapse and/or maternal death in childbirth with unexplained etiology. To date, there are no specific laboratory tests to diagnose AFE. Evidence of fetal cells in the pulmonary vessels is not a reliable diagnostic criterion and is not pathognomonic for AFE because fetal cells can be detected in 21%–100% of pregnant women without AFE[38]. Serologic assays that detect TKH-2 (a fetal antigen that can also be observed in maternal lung samples) or insulin-like growth factor binding protein-1 appear to have a high sensitivity for detecting AFE[39,40]. However, these methods have not been fully validated and cannot be recommended for routine clinical practice.

Management (Figure 1)

The management of women with suspected AFE remains supportive rather than therapeutic, with emphasis on maintaining oxygenation, supporting circulation, and correcting coagulopathy. In the immediate presentation of collapse, the basic ABCD of resuscitation should be applied, wherein D represents the delivery of baby. A multidisciplinary approach with early involvement of senior obstetricians, anesthetists, critical care team, hematologists, and neonatologists is key.

The airway must be maintained and protected usually by intubation, and high flow oxygen with monitoring of oxygenation by pulse-oximetry is essential. Blood gases are important to identify and correct acidosis. The circulation should be supported by securing intravenous access and administering a rapid infusion of crystalloids to maximize preload. The patient must be placed in a tilt or wedged position because aortocaval compression will severely worsen cardiovascular collapse. Continuous cardiac monitoring is required to detect potential arrhythmias and to allow treatment. If cardiac arrest occurs, cardiopulmonary resuscitation (CPR) must be started immediately. If CPR is unsuccessful after 3–4 min, plans should be made to achieve delivery of fetus



Modified from Tuffnell DJ[14] and Rath WH[41]; FFP: fresh-frozen plasma; BP:blood pressure; SpO2: partial oxygen saturation; ECG: electrocardiography; F VIIa: recombinant factor VIIa

Fig. 1 Management of a suspected AFE patient

within 5 min of arrest. There is evidence that the outcome for the fetus is time dependent and intact survival is most likely if the interval from collapse to delivery is less than 5 min[20]. Regardless of fetal condition, this approach is also warranted to maximize effectiveness of CPR for the mother.

Maintaining cardiac output may be difficult and may require vasopressors and inotropes. In the case of pulmonary edema, diuretics may be indicated. The changing hemodynamics mean that establishing the balance between supporting circulation with fluid and avoiding overload can be very difficult. Invasive monitoring is usually indicated but may require some correction of coagulopathy first. A pulmonary artery catheter can be considered to closely monitor cardiac output, central venous pressure, pulmonary capillary wedge pressure, and pulmonary artery pressure. This also provides a good entry point for blood collection. However, in the early phase of presentation it may not be possible to establish monitoring.

Correcting hemorrhagic problems is urgent because there can be potentially massive blood loss as a result of coagulopathies and uterine atony. Initial administration of tranexamic acid to treat hyperfibrinolysis and the use of fibrinogen concentrate (for fibrinogen levels below 2 g/L) are essential and if possible should be performed using rotational thromboelastometry[26]. Early aggressive correction of coagulopathy and replacement of blood loss with blood products, including fresh frozen plasma, cryoprecipitate, and platelets, may help prevent further loss. Factor VIIa procoagulant, which increases thrombin formation, has been used in a number of cases, but strong data supporting its use is lacking. It is important to remember this agent is expensive and is only likely to be successful if other clotting factors have been replaced before factor VIIa is administered[14]. Ensuring uterine contraction by standard methods such as uterine massage and medical management using oxytocics, ergometrine, and prostaglandins may also help. Uterine artery embolization is not always practical in the acute phase but may be considered later on. Approximately 25% of these women require hysterectomy to arrest the bleeding[14].

There are some novel interventions that have been successfully used in patients with AFE. Inhaled nitric oxide and a right ventricular assist device have been used in patients with pulmonary hypertension and right ventricular failure[42,43]. Cardiopulmonary bypass, intraaortic balloon pump counterpulsation, and extracorporeal membrane oxygenation (ECMO) have been used in patients with severe left ventricular failure and hypoxemia[19,44].

Outcome

Estimates of maternal mortality vary greatly and appear to be largely dependent on criteria required for the inclusion of cases as true AFE. Series restricted to patients in whom all classic signs and symptoms of AFE are present suggest mortality rates exceeding 60%[3]. Maternal morbidity is also high and only 15% of survivors may be neurologically intact. More recent data suggest a lower mortality rate can be achieved; 27% in a population-based study performed in 1999[45], and 37% from the UK registry in 2005[6]. However, it is unclear if the improved mortality rate is related to better critical care management or an artifact related to different reporting techniques yielding a larger denominator. Neonatal outcome is generally poor with a mortality rate of 20–25%, and of the survivors, only 50% may be neurologically intact[3,6].

Summary and Recommendations

- AFE is a catastrophic condition that occurs during pregnancy or shortly after delivery. It is characterized by the abrupt and fulminant onset of hypotension secondary to cardiogenic shock, hypoxemia, respiratory failure, and DIC.
- AFE is unpredictable, unpreventable, and rare.
- AFE is a clinical diagnosis that is based on a constellation of clinical findings.
- Clinicians should suspect AFE whenever shock or respiratory compromise develops during labor and delivery or immediately postpartum.
- There is no specific treatment for AFE. The goal of therapy is to expeditiously correct hypoxemia and hypotension so that ischemic consequences in the mother are prevented and adequate oxygen supply to the fetus is ensured.
- Maternal mortality because of AFE remains high, although it is currently less than that in previous years. This most likely reflects early recognition and appropriate aggressive therapy. However, those who survive generally have a poor outcome, and many suffer neurologic injury as a result of cerebral hypoxia. Neonatal outcomes are also poor,

although they improve with prompt delivery.

- A team approach among obstetricians, anesthetists, intensive care specialists, and neonatologists is necessary for a successful outcome.
- Because the passage of some fetal tissue into the maternal circulation appears to be ubiquitous during delivery, even early in pregnancy, future efforts to prevent AFE may rely on identification of women at risk for this abnormal response.
- The number of new AFE cases in Taiwan is estimated at only 5–10 per year. To increase knowledge about AFE among ethnic Chinese, cooperative research with mainland China is warranted.

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Amniotic Fluid Embolism

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摘要

懷孕併發羊水栓塞症，發生之特性，非僅勢如雷霆之不可抑，亦且無迹可循而難逆料，是致產婦死傷之最兇惡疾。本文宏觀此症新知，包括發生率 \ 病因 \ 致病生理變化 \ 臨床表現 \ 診斷 \ 及處理，以期有益於同仁對此症之認知。面對此惡，搶救產婦，以目前科技能力雖還有諸多不及，然而早覺察，速應變，仍是唯一可著力且可能有效之要點。而覺察，應變，皆賴知識指引。無知常亂，真知乃有方。

關鍵詞：羊水栓塞症、心因性休克、呼吸衰竭、瀰漫性血管內血凝病變

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Original Article

Stabilization in Surgical Treatment of Comminuted Distal Radius Fracture: Clinical Comparisons Between Bone Graft Substitute and Locking Plate

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Abstract

Distal radius fracture is extremely common; its clinical outcome is worse when the fracture is severely comminuted. In such cases, surgical stabilization of distal radial articular fragments is important. The different surgical techniques used for this purpose provide different stability and clinical outcomes. Improvement in the bone graft substitute provides convenience in surgical repair and may offer effective bone stability. Although injectable bone graft substitutes have demonstrated sufficient compression strength in biomechanical models, clinical experience with them has not been elucidated.

In the present study, we prospectively analyzed the surgical outcomes in 62 patients (mean age at surgery, 62.4 years; range 30–89 years) with comminuted distal radius fracture. We performed open reduction with locking plate fixation in 30 patients and closed reduction with Kirschner wire fixation using injectable bone graft substitute [Minimally Invasive Injectable Graft (MIIG)] in 32 patients. We followed-up the patients for an average of 11.8 weeks (range, 8–26 weeks). We compared radiographic findings of radial inclination, palmar tilt, and ulnar variance between the two groups.

Compared with the patients treated using locking plate fixation, the patients who received closed reduction with MIIG showed bone healing with statistically significant greater decrease in joint height (radial inclination and ulna variance) two months post surgery. Wrist range of motion between the two groups was equivalent postoperatively.

In our clinical treatment of comminuted distal radius fracture, the bone graft does not provide sufficient stability when used by itself during bone growth as compared with the locking plate. In order to improve functional outcome, use of a bone graft substitute must be combined with other mechanical fixation methods.

Key words: distal radius fracture, bone graft substitute, locking plate

Background

The prognosis for displaced fracture of the articular surface of the distal radius is worse than that for extra-articular fractures due to the incongruity and arthritis of the radio-carpal and distal radio-ulnar joints, carpal subluxation, and associated inter-carpal ligament injuries. Treatment of intra-articular distal radius fractures has mainly focused on stable fixation to permit early motion and avoid prolonged immobilization.

Several surgical techniques are used for management of distal radius fractures, including open reduction with internal fixation, percutaneous pinning with cast, and extra-skeletal fixation. Internal fixation creates a more rigid mechanical scaffold, supporting fixation to enable early mobilization. Open techniques, however, generally require more extensive soft-tissue dissection and have been associated with higher complication rates than closed techniques.

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Bone grafts can accelerate healing across metaphyseal bone defects created by realignment of fracture fragments and offer support to non-impacted articular fragments. Autogenous bone grafts require an open reduction, which disrupts soft tissue stability. Synthetic bone graft substitutes, including coral, solid or injectable hydroxyapatites, allograft bone, and bone derivatives, offer convenient surgical alternatives without causing major soft tissue damage.

Herein we discuss our clinical experience of performing limited internal fixation using a calcium sulfate bone graft in the treatment of distal radius fracture. We compare this approach with a locking-plate fixation procedure. Postoperative radiographic joint height (ulnar variance) and radial inclination are important predictive factors for wrist function; therefore, in the present study we investigated surgical methods of maintaining articular reduction during bone union that the surgeon can assess postoperatively using follow-up radiographs.

Materials and Methods

Patients

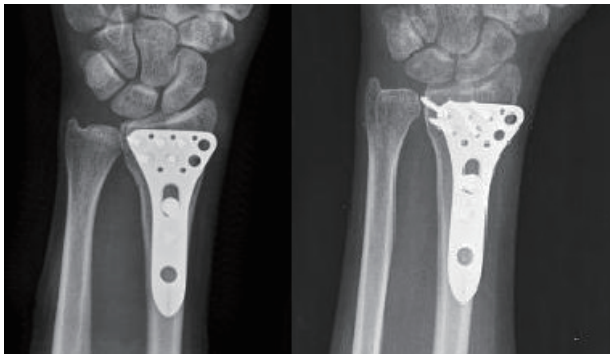
We recruited patients with Type C (complete articular fracture) distal radius fracture according to the AO (Arbeitsgemeinschaft fur Osteosynthesefragen) Comprehensive Classification of Fractures, which subdivides articular fracture into three groups and nine subgroups on the basis of the extent of articular and metaphyseal fragmentation and the orientation of the articular fracture lines. Patients with Type A (extra-articular) and Type B (partial articular) distal radius fractures were excluded. In addition, only Type C patients whose fractures were treated surgically were included in our study.

Surgical procedures

Between May 2013 and April 2014, 62 patients (mean age, 62.4 years; range 30–89 years) underwent surgical treatment for an AO Type C distal comminuted radius fracture in our orthopedic department. As shown in Figure 1, 30 patients (Group B) received palmar-approached open reduction with A-plus volar anatomy locking plate. As shown in Figure 2, 32 patients (Group A) received closed reduction with percutaneous Kirschner wire fixation and an injectable bone graft substitute. In all cases, surgery was performed under general anesthesia on a hand table; a tourniquet was applied during surgery for reduction of blood loss.

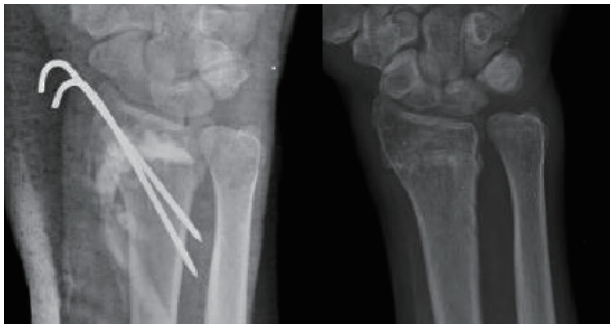
Group B patients underwent open reduction surgery via a palmar approach, performed through Henry's interval (a longitudinal incision between the flexor carpi radialis tendon and the radial artery). The surgeon then made an L-shaped elevation of the pronator quadratus muscle with a longitudinal incision close to the volar lip; followed by fracture reduction by manipulation of the bone fragments using a periosteal elevator. The surgeon checked the post-reduction joint line during the procedure by means of portable fluorography. With the patient's upper extremity in neutral rotation, the surgeon began closure of the volar wound by reinsertion of the pronator quadratus muscle.

Group A patients underwent closed reduction surgery without soft tissue disruption. Multiple smooth Kirschner wires temporarily maintained fixation of the reduced fracture. After satisfactory reduction and distraction, the surgeon made a short



Postoperative first day two moths later

Fig. 1



Postoperative first day two moths later

Fig. 2

longitudinal incision on the radial side of the wrist. The surgeon next injected the bone graft substitute into the fracture cavity. We used MIIG X3 (Minimally Invasive Injectable Graft) , composed of surgical grade calcium sulfate. Its compressive strength in vitro reaches 45 MPa 24 h after injection, which is close to that reported of polymethylmethacrylate (60 MPa).

Measurements

We obtained standard preoperative posterior-anterior and lateral radiographs of the fracture to demonstrate the extent and direction of the initial displacement.

Radiographic measurements used to assess the stability of the surgical fracture reduction include radial inclination, palmar tilt, and ulnar variance. The standard radiographic parameters of the intact distal radius include the following values: radial inclination, 23 degrees; average palmar tilt, 12 degrees; and ulnar variance, more than 2 mm. Successful surgical fracture reduction of AO Type C distal comminuted radius fracture is defined by the following postoperative radiographic measurements: articular incongruity, no greater than 1 mm; dorsal tilt, 10 degrees; and increase in ulnar variance, 2 mm. Failure to achieve these values may lead to adaptive carpal instabilities, ulnar impaction, and post-traumatic arthritis.

In order to identify residual deformity during bone union, we compared the mean degree of radial inclination, palmar tilt, and ulnar variance, taken at time of reduction and again at the final follow-up visit between groups A and B. We also compared the groups according to wrist range of motion.

Postoperative treatment

Initiation of motion as soon as possible after surgery is the ultimate goal in the treatment of patients with intra-articular distal radius fractures. The treated arm of patients in Group A were encased in a short cast that allowed free finger mobility for one month postoperatively. Patients in Group A did not receive any external fixation hardware.

Postoperative rehabilitation was the same between the two groups. Passive arm motion began three weeks after surgery; active non-weight-bearing arm motion began six weeks postoperatively. Full weight-bearing arm motion began two months after surgery.

Results

The average follow-up time of all study patients was 11.8 weeks (range, 8–26 weeks). All patients achieved complete bone union.

Radiographic values are contained in Table 1. The average radial inclination in Group A was 20.5 degrees immediately postoperatively and 17.9 degrees two months later. Average radial inclination in group B was 22.1 degrees immediately postoperatively and 21.4 degrees two months later. Average ulnar variance in Group A was 1.22 mm immediately postoperatively and 0.47 mm two months later; in Group B, average ulnar variance was 1.57 mm immediately postoperatively and 1.3 mm two months later in group B. The average palmar tilt in Group A was 9.88 degrees immediately postoperatively and 7.78 degrees two months later. Averaged palmar tilt in Group B was 10.37 degrees immediately postoperatively and 9.87 degrees two months later.

Radial inclination and ulnar variance, which can predict the postoperative range of motion, showed decrease in the two groups during the period of bone ingrowth. At the two-month postoperative follow-up, average decrement of radial inclination between groups A and B was statistically significant: 2.56 degrees in Group A and 0.7 degrees in Group B. Ulnar variance declined 0.81 mm in Group A and 0.27 mm in Group B. This result indicates that joint collapse occurred during bone healing in the patients in Group B, demonstrating that the injectable bone graft substitute cannot maintain joint height alone without rigid internal fixation.

As shown in Table 2, the average wrist range of motion following bone union was nearly equivalent in both groups. However, ulnar deviation was more limited in the patients in Group B. This finding supports the hypothesis that decreased ulnar variance will impinge wrist motion directed toward the ulna. Although joint collapse is more severe in Group B, the same wrist range of motion indicates that soft tissue preservation using the closed approach contributes to postoperative restoration of wrist mobility.

No postoperative infection, including pin tract infection, occurred during bone healing in either group. No patients experienced complications such as wound hematomas or stitch abscesses. In addition, no patients experienced neurologic complications such as median nerve injury or superficial radial

Table 1. The analyses for outcome measurements

Measurements *Measuring time*	Descriptive statistics						The t-statistics values and p-values for t-tests							
	Group A			Group B			Time effect within group			Group effect given fixed measuring time			Group effect on the difference between two measurements	
	n	*mean*	*SD*	*n*	*mean*	*SD*	*t*	*p*		*t*	*p*		*t*	*p*
Radial inclination (degrees)														
Post-surgery	32	20.50	02.55	30	22.1	01.27	-11.92	<.0001	*	-5.89	<.0001	*	3.15	0.0028	*	7.58	<.0001	*
Follow-up	32	17.94	02.79	30	21.4	01.52							6.11	<.0001	*			
Palmar tilt (degrees)																		
Post-surgery	32	09.88	01.50	30	10.37	00.67	0-6.21	<.0001	*	-4.78	<.0001	*	1.69	0.0988		4.52	<.0001	*
Follow-up	32	07.78	02.15	30	09.87	00.86							5.07	<.0001	*			
Ulnar variance (mm)																		
Post-surgery	32	01.22	00.87	30	01.57	00.50	0-7.13	<.0001	*	-3.25	0.003	*	1.94	0.0579		3.84	0.0003	*
Follow-up	32	00.41	01.16	30	01.30	00.60							3.85	0.0004	*			

Table 2. Range of motion of Wrist

	Group A	Group B
Dorsiflexion	60.5 ± 11.5	63.7 ± 10.6
Palmarflexion	55.8 ± 9.7	53.9 ± 9.4
Radial deviation	5.4 ± 1.2	7.5 ± 1.8
Ulnar deviation	20.8 ± 2.6	32.2 ± 3.9
supination	61.3 ± 10.1	55.4 ± 9.5
pronation	58.1 ± 10.9	53.6 ± 10.5

nerve branch injury.

In Group B, the surgeons used oblique volar incisions across the transverse wrist crease to limit scar contracture. The branches of the superficial radial nerve, the dorsal cutaneous branch of the ulnar nerve, and the palmar cutaneous branch of the median nerve were well protected.

Although monitoring of superficial radial nerve branches injury during insertion of Kirschner pins and bone graft substitute injection trocar is challenging, we observed no obvious nerve injury in the patients in Group A.

Discussion

Although comminuted distal radius fracture is a common injury, many factors influence the clinical outcome, including bone quality, severity of fracture, and postoperative stabilization. Preservation of anatomical reduction is crucial; in particular, successful reduction and rigid fixation during bone ingrowth are the most important factors for the best functional outcome. A successful outcome is possible via strong fixation implant, bone graft along with mechanical scaffolding for strength and rigidity, or prolonged immobilization by brace.

Several surgical techniques are used for the management of comminuted distal radius fractures, including open reduction with internal fixation, percutaneous pinning with cast, and extra-skeletal fixation. Internal fixation creates a more rigid mechanical scaffold, supporting fixation to enable early joint mobilization. Open techniques, however, generally require more extensive soft-tissue dissection and have been associated with higher complication rates than closed techniques. In order to avoid soft tissue injury associated with open techniques, limited-incision approaches and low-profile modular implants

are used. However, many surgeons doubt the long-term stability of limited fixations.

Bone grafts can accelerate healing across metaphyseal bone defects created by the realignment of fracture fragments and offer support to non-impacted articular fragments. However, at the start of bone healing, bone grafts are absorptive. This significantly decreases the rigidity of the bone and leads to the collapse of the joint line. In addition, clinicians must be aware that although autogenous graft is preferred, it requires an open reduction surgical procedure, which disrupts soft tissue stability. Therefore, synthetic bone graft substitutes, including coral, solid or injectable hydroxyapatites, allograft bone, and bone derivatives offer convenient surgical alternatives without causing major soft tissue damage. Some injectable allografts provide rigidity and stability of the same level as that offered by internal fixation during the early stage of bone healing. However, evidence of rigidity is reported only in a biomechanical model. Clinical reports are now required to validate its efficacy.

In the present study, we investigated two types of surgery to treat AO Type C comminuted distal radial fracture in adults. Specifically, we compared the efficacy of limited internal fixation using a calcium sulfate bone graft to a locking-plate fixation procedure. Using postoperative radiographic films, we measured joint height (ulnar variance) and radial inclination.

One of the goals of intra-articular distal radius fracture treatment has been to provide stable fixation that permits early postoperative motion and avoids prolonged immobilization of the wrist. Although many new surgical fixation techniques are now available, preservation of joint alignment remains a daunting challenge in the surgical treatment of comminuted distal radius fracture.

Postoperative joint alignment is critically important for a successful reduction. Knirk and Jupiter[3] found that the development of wrist arthritis strongly correlated with postoperative articular incongruity and that the magnitude of the incongruity positively correlated with the severity of arthritis.

Locking plate-and-screw systems have been shown to provide more rigid fixation and better clinical outcomes than other internal fixation implant procedures; however, an open reduction procedure is still used in this method, and the resulting soft tissue injury and compromise of the blood supply are associated with an increase in the nonunion rate. Therefore, some modified minimal invasive fixation techniques have been recommended to avoid these disadvantages.

In our study, we wanted to learn whether the bone graft substitute provides enough rigidity during bone healing.

High-energy impact injuries in young patients as well as moderate-energy impact injuries in osteoporotic individuals can create very complex articular and metaphyseal fractures that present particular treatment challenges. A major concern with high-energy intra-articular distal radius fractures is the presence of metaphyseal bone deficiency caused by impaction and cortical comminution.

Because of the high level of force causing this type of fracture, the articular surface is commonly displaced and impacted. Using an open or a closed surgical technique, the surgeon elevates and reduces the depressed articular surface and restores radial length. This procedure causes a large metaphyseal defect, resulting in a loss of structural support for the reduced articular surface. If no mechanical support is available during bone healing, maintaining the anatomical structure of the wrist is less likely.

Leung et al[1] defined displacement as greater than 10 degrees of dorsal angulation or 2 mm or more articular incongruity (i.e., radial shortening compared with the opposite side). They reported that displacement required surgical intervention.

Three major categories of intra-articular fractures generally require open reduction and internal fixation: shear fractures of the dorsal or palmar articular lip of the distal radius, displaced radial styloid fractures, and fractures that involve depression of the articular surface that cannot be adequately reduced using closed manipulation. May et al.[19] showed that distal radius fractures complicated by distal radio-ulnar joint instability were accompanied by an ulnar styloid fracture. Significant displacement of an ulnar styloid fracture was associated with an increased risk of distal radio-ulnar joint instability.

Failure to repair the fragmentation of the articular margin is a significant cause of bone loss during bone healing. It may be more difficult to manipulate ulnar fragments using a closed pinning technique. Therefore, an open surgical technique is preferable in the reconstruction of joint anatomy. If the locked

plate fails to secure this small ulnar fragment adequately, it may slip around the plate; subsequently, the entire carpus may become unstable and result in poor reduction.

Injectable bone graft substitutes have the same effect in supporting joint fragments. In our prospective study, we excluded patients treated surgically without initial successful reduction. All patients in our study treated using open or closed reduction received satisfactory reduction from the beginning, avoiding the effects of initial bone loss.

Catalano et al[21] reviewed 21 young patients an average of 7.1 years after articular fractures of the distal radius were treated surgically. They confirmed that the development of an arthrosis is strongly associated with residual articular incongruity. However, in contrast to the study by Knirk and Jupiter, Catalano's study found that arthrosis did not correlate with the functional result; it is likely that these findings were observed because more advanced techniques were used than those observed in the Knirk and Jupiter study. The results of Catalano's investigation suggest that restoration of the extra-articular alignment was more important than residual articular incongruities and arthrosis in determining the functional result. We interpret these results as indicating that the quality of the articular reduction likely is less important than other factors, such as the severity of the injury, associated injuries, and the quality of the extra-articular reduction.

As early as 1997, Lindau et al.[20] found that intra-articular distal fractures of the radius in young adults were associated with chondral and ligament lesions. The most frequent ligament tear seen was the triangular fibrocartilage complex (TFCC) injury; they concluded that chondral and ligamentous lesions were frequent sequelae of intra-articular distal fractures of the radius, and these complications may explain poor outcomes following well-healed distal radius fractures. They noted that when TFCC injury happened, supination and pronation of the wrist were associated with nerve impingement.

High-energy impact injuries in young patients and moderate-energy injuries in osteoporotic individuals can create very complex articular and metaphyseal fractures that are especially difficult to treat. After reducing these fractures, surgeons commonly find articular fragments located in the soft tissues of the forearm and cortical bone fragments located in the joint. If there is extensive articular fragmentation, anchoring the small fragments reliably with wires is challenging.

Bone graft substitutes with sufficient biomechanical strength can strengthen the joint line during the initial bone healing (4–6 weeks postoperatively). However, if bone healing is delayed, the joint would collapse after resorption of the bone graft substitute. Conversely, a locking plate system could provide long-term stability in patients with delayed bone ingrowth and preserve the joint functional structure.

Fixed-angle anatomy locking plates can adequately stabilize articular fragments and both volar and dorsal comminution, even in osteoporotic bone. Optimal positioning of the plate and locking screws will restore joint height as well as radial and palmar tilt. In patients with osteoporosis, locking screws can provide a stronger stability than any other type of mechanical fixation. Therefore, the locking plate system is still the gold standard surgical treatment for comminuted distal radius fracture.

Clearly, the best possible articular reduction is desirable. However, the clinician must balance this aim against the benefits and risks of the available surgical techniques. In addition, precise restoration of articular congruity may not be possible in some complex articular fractures; but this does not necessarily preclude restoration of wrist function. In our study, the patients treated using bone graft substitutes whose radiologic joint line was worse postoperatively did not experience poor wrist function. This result is likely caused by the restoration of soft tissue structures in the patients who received closed surgical fixation.

Effective bone healing happens in the first three months following the injury. During the two-month postoperative follow-up, all patients in our study were found to have complete bone union, but the individuals who received closed reduction with bone graft substitute injection were shown to have lost more bone following fracture reduction surgery than those patients who received locking-plate fixation surgery. This outcome indicates that the rigidity and strength of the MIIG graft in vitro is insufficient for clinical use in standard fracture reduction.

From our experience, injectable bone graft substitutes only provide initial stability. At the two-month follow-up, the radial inclination and ulnar variance measurements of the patients treated using bone

graft substitutes decreased compared with those of the patients treated using locking plate. In addition, maintenance of the joint line was found only in the patients who were treated with locking plate fixation. Nevertheless, the wrist range of motion two months postoperatively was similar between the two groups, indicating that closed mechanical fixation is better for preservation of soft tissue structures. Therefore, to see better clinical results in patients with comminuted distal radius fractures, we recommend using bone graft substitutes in conjunction with more rigid fixation methods.

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比較注射型人工骨和鎖定式骨板對於遠端橈骨粉碎性骨折治療的穩定性差異

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摘要

遠端橈骨骨折為各年齡層均常見的骨折，當粉碎程度增加，臨床治療的成效就越差。以手術穩定骨折碎片，對於粉碎性遠端橈骨骨折相當重要；不同的手術固定方式也產生不同的臨床治療成果。

許多因素都會影響到遠端橈骨骨折的治療成果，近年來因為人工骨骼替代物技術的進步，可以提供粉碎型骨折更好的支撐力。我們提出相關的臨床資料，探討注射型人工骨骼替代物的力學穩定度在治療粉碎型遠端橈骨骨折的成果。

以回溯型研究分析六十二位因粉碎型遠端橈骨骨折於本院接受治療的病人；平均年齡為 62.4 歲（30 至 89 歲），平均追蹤時間為 11.8 週（8 至 26 週）。其中 30 位病人接受開放式復位及鎖定式骨板固定，另外 32 位病人接受閉鎖式復位及注射式人工骨骼替代物固定。並比較兩種治療方式術後追蹤的橈側傾角、掌向斜角以及尺骨關節高度的差異。

比較兩組病人的追蹤結果可以發現，使用注射式人工骨骼替代物治療的病人較容易發生橈骨關節面下沉的狀況；而且治療後的橈側傾角及掌向斜角都比使用鎖定式骨板固定的病人有顯著差異。而術後的關節活動度在兩組病人之間是沒有差異的。

雖然注射式人工骨骼替代物在生物力學上有足夠支撐強度，但在臨床上仍缺乏足夠的病人數據證明其力學強度。在我們的臨床數據發現，注射式人工骨骼替代物無法提供與鎖定式骨板相同的生物力學強度。因此在臨床上，仍建議將注射式人工骨骼替代物與更強的固定裝置合併使用，以達到較佳的治療成果。

關鍵詞：遠端橈骨骨折、人工骨骼替代物、鎖定式骨板

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Original Article

Bladder Volume Determination: A Comparison of Two-Dimensional and Three-Dimensional Transvaginal Ultrasound

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Abstract

Background and purpose: To assess the accuracy of *in vivo* measurement of bladder volume using three-dimensional ultrasound (3DUS) and compare the results with those of two-dimensional US (2DUS).
Methods: In this prospective study, 40 female patients (mean age 51.3 years, range 30–82 years) were scanned after obtaining informed consent. Each patient's bladder was initially scanned using the conventional transvaginal 2DUS method and then scanned using transvaginal 3DUS, and finally sterilized catheterization was used to obtain the true bladder volume. The 2DUS bladder volume was estimated by measuring the height and depth of the bladder and using the formula of Haylen et al. [estimated volume = (5.9 × height × depth) − 14.6]. The 3DUS volume was masked using a manual determination of the bladder outline to isolate the bladder for volume measurement. The time from initial transvaginal US to final catheterization was less than 10 min.
Results: The bladder volumes by catheterization varied from 30 mL to 590 mL. Using transvaginal 2DUS and 3DUS, the mean values of the error measurement estimation were 15.7% and −8.48%, respectively, and the correlation coefficients were 0.90 and 0.97, respectively.
Conclusion: Although 3DUS is more accurate than 2DUS for estimating bladder volume, either method could be used instead of catheterization for scanning bladder urine volume. However, the 3D transvaginal scan volume generally underestimates the actual volume, whereas 2D generally overestimates it.

Key Words: bladder volume, transvaginal ultrasound, two-dimensional ultrasound, three-dimensional ultrasound

Introduction

Measurement of post-micturition bladder volume is important in the assessment of voiding disorders, and serial measurements may indicate clinical progress. Traditionally, such measurement involves the passage of a urethral catheter to empty the bladder, which can be uncomfortable for the patient and may cause urinary infection or urethral trauma. A simple, noninvasive method for the determination of bladder volume would be highly desirable.

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Ultrasound (US) imaging now provides a possible method for estimating bladder volume noninvasively. Traditionally, bladder volume has been estimated with two-dimensional US (2DUS) using a transabdominal or transvaginal approach. 2DUS volume estimates are based on the assumption of a regular geometric shape for the organ and use ellipsoid or spherical equations to calculate the volume[1]. However, volume estimates for irregularly shaped objects can often be inaccurate, and various techniques have been proposed to improve the accuracy of 2DUS volume measurement[2]. Recently, three-dimensional US (3DUS) has been introduced into clinical use; 3DUS volume measurements have been shown to be accurate for *in vitro* objects[3,4]. The present study was undertaken

to assess the use of 2DUS and 3DUS imaging for measuring bladder volume.

Methods and Materials

Between November 2002 and March 2005, 40 female inpatients were recruited from the author's unit. Written informed consent to participation was obtained from all patients.

In the operation room, prior to anesthesia, the patient laid in the supine position with their knee flexed and then the bladder volume was estimated by a single ultrasonographer using a 7.5-MHz transvaginal transducer with 3D facility (VOLUSON 730, Kretz-Technik AG, Zipf, Austria). The 2DUS bladder volume was estimated first by measuring the height and depth. The data was stored in a computer and calculated by the formula of Haylen et al[5].

$$\text{Estimated bladder volume (mL)} = (5.9 \times H \times D) - 14.6$$

where H: maximum bladder diameter in the horizontal (superoinferior) axis (cm) and

D: maximum bladder diameter in the vertical (posteroanterior) axis (cm).

After measuring the height and depth, the ultrasonographer immediately measured the bladder volume by 3DUS using the same vaginal probe. The patient was asked to remain as still as possible and every effort was made by the ultrasonographer to limit inappropriate movements of the transducer. The truncated sector defining the region of interest was adjusted and the sweep angle set to 85° to ensure that the complete bladder was obtained. The volumes were captured through an automatic sweep of the transducer over the region of interest when the "scan" button was depressed. The 3D scanning procedure required <20 s. The images data were digitally stored in the computer and the estimated volume calculated later.

Indwelling catheterization, a routine procedure for every patient who undergoes a major gynecological surgery in our hospital, was performed by a registered nurse with a short plastic catheter after sterilization. The nurse pressed the suprapubic area at least three times to ensure complete urine collection. The urine remaining in the catheter was drained into the collection device. The volume of the urine specimen was measured in a cylinder with milliliter graduation, and this was considered to be the actual bladder volume. The time from initial transvaginal US to final catheterization was <10 min.

On activation of the 3D image data stored in the computer, three orthogonal planes were simultaneously displayed onscreen. The orientation of these planes was maintained throughout any translation or rotation. The 3D bladder volume was measured by outlining the edge of the bladder on each plane manually using a trackball (Figure 1). Twelve ultrasonic images, spatially interlocked, were generated at 15° angles to one another. A volumetric model of the whole bladder was constructed from these individual images and the 3D volume was calculated by the built-in computer software.

Statistical analysis

Results are presented as mean values ± standard deviation (SD) and the correlation coefficient (r). A *P*-value < .05 was considered statistically significant. Normality was tested with the Kolmogorov–Smirnov test. Statistical analyses were performed using computer software Statistical Package for Social Sciences (SPSS^{TM}) for Window XP version 11.

Results

Forty patients (mean age, 51.3 years; range, 30 to 82 years) were scanned. Of these, 18 (45%) had been admitted for anti-incontinence surgery and the remainder were all admitted for benign gynecological surgery. No patients with cancer were included. Their

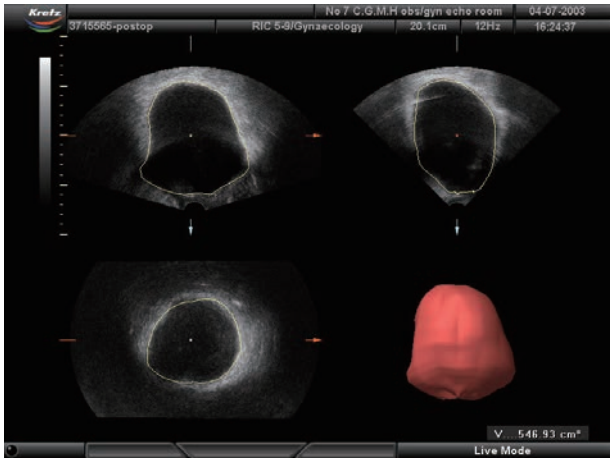

Fig. 1 The 3D bladder urine volume was measured by outlining bladder border of each plane through the mouse ball manually, and was calculated by the computer program.

demographic data is shown in Table 1. Their bladder volumes by transurethral catheter varied from 30 mL to 590 mL. Figure 2 shows the scan volumes plotted against the catheter volumes. Both the 2DUS and the 3DUS bladder volumes correlated with the catheter volumes, with correlation coefficients of 0.90 and 0.97, respectively.

Table 1. Characteristics of patients(n=40)

	Mean ± SD	Range	*n*(%)
Age (yr)	51.3 ± 10.3	30-82	
BMI (kg/m^2)	25.7 ± 3.5	18.6-34.3	
Parity	3.3 ± 1.7	1-7	
Menopausal status			
Premenopausal			27(67.5)
Postmenopausal			13(32.5)
Diagnosis			
Uterine and ovarian mass			20(50)
Urinary incontinence			14(35)
Urogenital prolapse			6(15)
Bladder volume (mL)			
2D ultrasound	254.6 ± 157.8	11-740	
3D ultrasound	198.1 ± 127.2	25-547	
Catheter	223.6 ± 148.7	30-590	

SD=standard deviation; BMI=body mass index; 2D=two-dimensional; 3D=three-dimensional.

On comparing the estimated volumes from 2DUS and 3DUS with the actual bladder volumes, we obtained the mean values of the error measurement, which for 2DUS was 15.7% ± 27.7% (range, −62.1% to 69.4%), and for 3DUS was −8.4% ± 16.3% (range, −31.7% to 43.0%; Figure 3).

Reliability was assessed as follows: For intraobserver variability, another 12 patients' bladder volumes were estimated by transvaginal 2DUS and 3DUS without catheterization, showing good correlation of the consecutively repeated measurements with

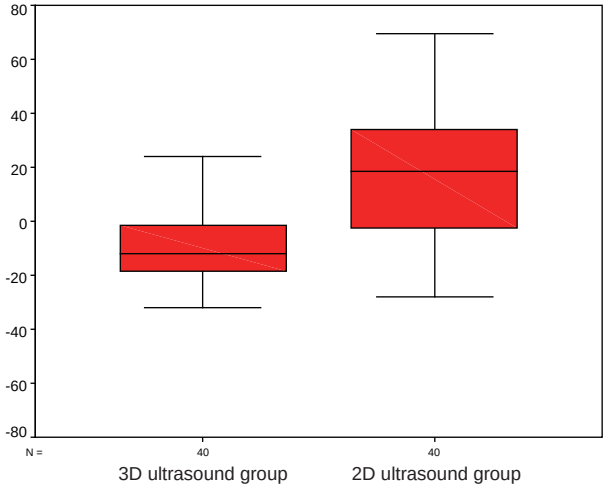

Fig. 3 The mean value of the error measurement between 2D and 3D ultrasound group.

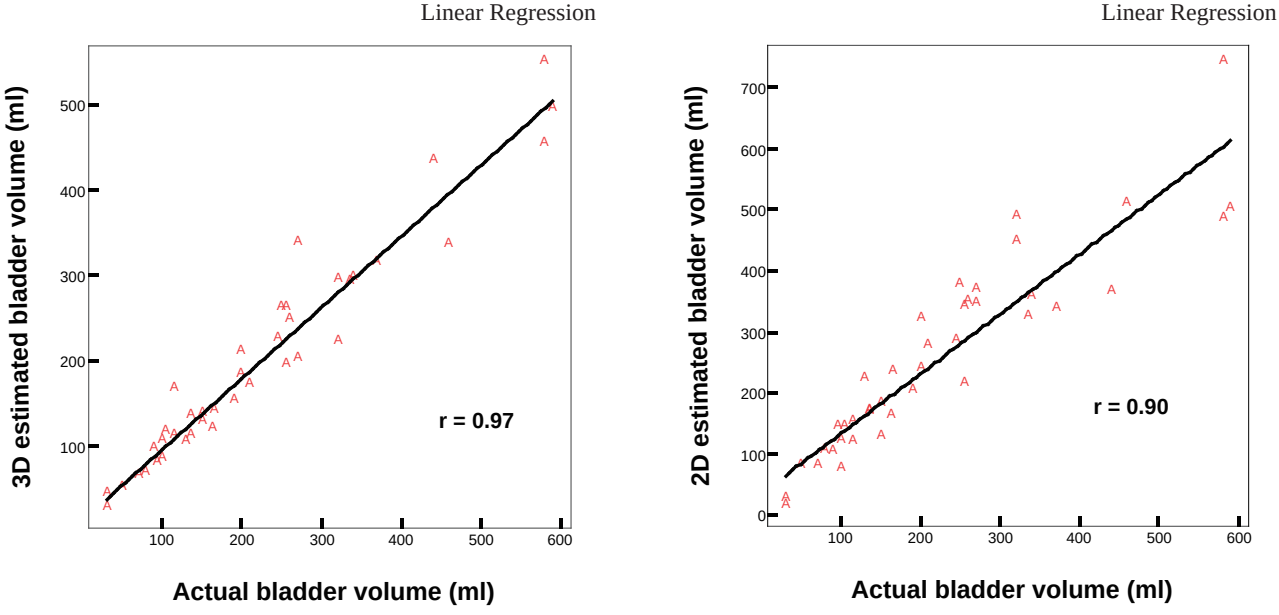

Fig. 2 Scattergram of catheter urinary volume versus transvaginal ultrasound scan volume with 2D (A) and 3D (B).

no significant difference between the two repeated measurements ($P > .05$). All the measurements were performed by the same ultrasonographer in this study; hence, there was no interobserver variability.

Discussion

Checking the residual bladder urine volume is important for women following anti-incontinence surgery, bladder training after radical hysterectomy, or postpartum urinary retention. Catheterization, the traditional method of measuring bladder volume, entails a risk of infection estimated to be at least 2%[6]. In patients with structural abnormalities of the urinary tract or otherwise predisposed to infection, this risk is higher[7]. In hospitalized patients, treatment of such infections may increase the length of stay and overall cost of care[8]. Avoidance of catheterization procedures through the use of bladder scanning can prevent patient discomfort and many urinary infections.

US imaging now provides a method for estimating bladder volume noninvasively. Traditionally, 2DUS has been used to estimate bladder volume clinically. It is easily available, but the estimated volume is based on the assumption of a regular geometric bladder shape. Consequently, volume estimates in irregularly shaped objects are often inaccurate. Recently, 3DUS has been introduced into clinical use and has demonstrated a high degree of reproducibility and accuracy of volume estimation, both *in vitro* and *in vivo*[9,10]. Unfortunately, due to the high cost involved, not every clinic or hospital has a 3DUS machine. In contrast, a 2DUS machine is easily available in our daily work.

Here we compared bladder volumes estimated by 2DUS and 3DUS with transurethral catheterized volumes. Both 2DUS and 3DUS volume estimates showed a high level of correlation with the actual volumes, and either method could be used instead of catheterization for scanning bladder urine volume. Although 3DUS appeared to be more accurate than 2DUS for the estimation of bladder volume, there was no statistically significant difference between them.

We found that, on an average, 3DUS underestimated the actual bladder volume (mean −8.4%), similar to that reported previously[11,12]. It could partly be explained by continued bladder filling during the delay before catheterization or failure of the scan to include all parts of the bladder because of the high urine volume. The bladders of five patients in the present study had an actual volume > 400 mL. Conversely, we found that, on an average, 2DUS overestimated the actual bladder volume (mean 15.7%). The reason for this may be the irregular bladder shape of the bladder, with the bladder volume estimated only using the measured height and depth.

According to previous studies, 3DUS in vivo bladder volume measurements are more accurate than those obtained using 2DUS[13]. However, in our study, although 3DUS was more accurate than 2D for estimating bladder volume, either method could be used for scanning bladder urine volume instead of catheterization, which could be particularly useful when 3DUS was not available. However, in our study, the 3D transvaginal scan volume underestimated the actual volume, whereas 2D generally overestimated it.

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膀胱容積的測定：經陰道 2D 超音波及 3D 立體超音波的比較

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摘要

背景及目的：利用超音波檢查人體膀胱的尿液容積，比較 3D 立體超音波和傳統 2D 超音波，分析兩者準確度的比較。

方法：本研究為前驅性研究，收集 40 位女性且事先已告知病患並同意檢查，先用 2D 超音波從陰道掃描膀胱容積後，再用 3D 超音波一樣由陰道再掃描膀胱容積，最後利用單次無菌導尿方式導出膀胱尿液並記錄實際體積，前後時間不超過 10 分鐘。2D 超音波測量膀胱高度（H）及深度（D），利用 Haylen 公式：[5.9 X 高度（H）X 深度（D）] 得到估算容積。3D 超音波則是掃描後，利用手繪方式畫出膀胱邊緣線，電腦則自動算出估算容積。

結果：40 位女性病患（平均 51.3 歲，從 30 到 82 歲）被掃描，實際膀胱容積為 30 毫升至 590 毫升，2D 超音波測量膀胱容積平均誤差為正 15.7%，相關係數為 0.9。3D 超音波測量膀胱容積平均誤差為負 8.48%，相關係數為 0.97。

結論：雖然 3D 超音波在測量膀胱尿液容積比 2D 超音波較準確，但這兩者皆可以用來測量膀胱尿液容積，代替使用侵入性的單次導尿。而 3D 超音波測量結果較可能低估實際膀胱容積，相反的 2D 超音波卻是高估。

關鍵詞：膀胱容積、經陰道超音波、2D 超音波、3D 超音波

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Tungs' Medical Journal 9 (2015) 26-29

Case Report

Airway Obstruction Caused by a Congenital Epiglottic Cyst

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Abstract

A congenital epiglottic cyst is a rare affliction associated with potential life-threatening airway obstruction. A 1-month-old baby boy was referred to our hospital for evaluation of respiratory distress and inspiratory stridor in December 2012. An obstructive upper laryngeal cyst was diagnosed on the basis of the results of transnasal fiberoptic laryngoscopy, magnetic resonance imaging, and computed tomography. Microlaryngoscopy was immediately performed to completely remove the lesion. Two days after surgery, the patient resumed a normal breathing pattern, showed significant improvement in feeding, and had no further episodes of stridor or airway obstruction. There was no recurrence after the 1-month follow-up visit.

Key words: epiglottic cyst, stridor, airway obstruction

Introduction

A congenital laryngeal cyst (CLC) was first reported by Abercrombie in 1881 and the incidence of this rare neonate disease is 1.82 per 100,000[1]. Occurring predominantly at the vallecula, aryepiglottic fold, and saccule of the ventricle, CLC is rarely observed at the epiglottis. Neonatal epiglottic cysts comprise 10% of all CLCs[2,3]. Congenital epiglottic cysts in infants often show clinical symptoms within a week after birth. Because the size of the respiratory tract in the infant is smaller than that in the adult, the symptoms of CLC are more pronounced with stridor, feeding difficulties, chest wall retraction, or cyanosis but less often with airway obstruction[4-6]. However, a few of the afflicted infants present only with growth retardation but with little or no airway symptoms. They are often ignored and misdiagnosed, and should be referred to an otolaryngologist for full endoscopic evaluation and possible intervention.

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This case of 1-month-old baby boy was referred to our hospital for evaluation of respiratory distress due to an obstructive upper laryngeal cyst. We report this case and discuss the treatment because only a few cases of upper laryngeal cyst have been seen in Taiwan.

Case Report

A full-term baby boy was delivered in a local obstetrics clinic with a bodyweight of 3200 g. This 1-month-old infant came to our hospital for respiratory distress, coughing with sputum, poor appetite, oligouria, and hysterical crying in December 2012. He had a bodyweight of 3240 g and milk volume of 30 ml at admission to hospital. A physical examination showed that he had a body temperature of 36.1°C, pulse rate 134/min, respiratory rate 30/min, crying with a hoarse voice, coughing with sputum, oral candidiasis, thick saliva with drooling, and difficulty with swallowing. In addition, the use of accessory muscles for breathing, retraction of chest wall, and funnel chest were also observed.

Under the transnasal fiberoptic laryngoscopy, a severe laryngomalacia and a laryngeal cyst were seen

(Fig. 1). A circular cyst was observed as a hypodense lesion in the foramen cecum using contrast computed tomography (CT) (Fig. 2) and 1.4 × 1.5 cm in diameter at the base of tongue by magnetic resonance imaging (Fig. 3)

Microlaryngoscopy was performed on the patient after explaining the treatment to the family. A 21-gauge needle was first inserted into the cyst and the content of cyst was withdrawn before resection by carbon dioxide (CO_2) laser. The cyst was identified as an epiglottic cyst consisting of squamous epithelium surrounded by lymphoid follicles. Two days after surgery, the patient resumed a normal breathing pattern and had no further episodes of stridor or airway obstruction. His activity and appetite increased significantly. His bodyweight reached 3700 g and milk volume increased to 100 ml upon discharge. There was no recurrence as examined by transnasal fiberoptic laryngoscopy after the 1-month follow-up visit.

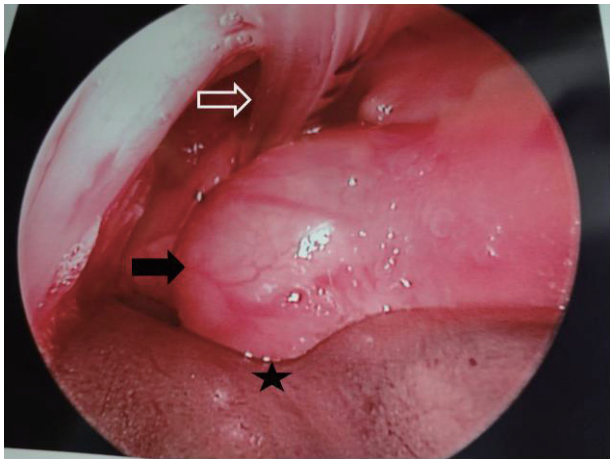

Fig. 1 Transnasal fiberoptic laryngoscopy shows a laryngeal cyst (solid arrow) in the larynx. Endotracheal tube (empty arrow) and the base of tongue (star).

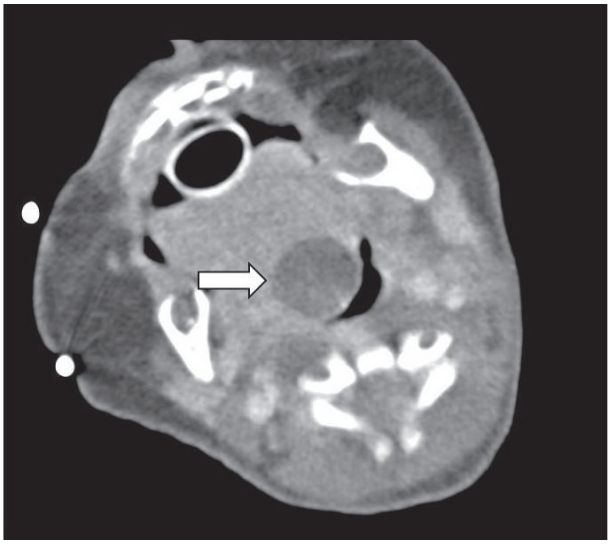

Fig. 2 A contrast computed tomograph (axial section) shows a hypodense circular cyst (arrow) in the foramen cecum.

Discussion

The cause of epiglottic cysts is not clear. Ductal cysts are common and are retention cysts resulting from obstruction of the submucosal collecting ducts of the salivary glands in the larynx. Collecting duct obstruction may occur from congenital occlusion, or from inflammation, trauma, or tumor[7].

The symptoms of an epiglottic cyst may vary from asymptomatic to a lump in the throat, easy choking, difficulties in swallowing, talking, and breathing. Symptoms depend on the cyst's size, location, and the age of the patient. In infants, clinical presentation of symptoms includes dyspnea, cyanosis, stridor, and hysterical crying. The respiratory distress syndrome is more pronounced when a patient is supine or feeding, making feeding difficult and leading to poor growth and development and low body weight

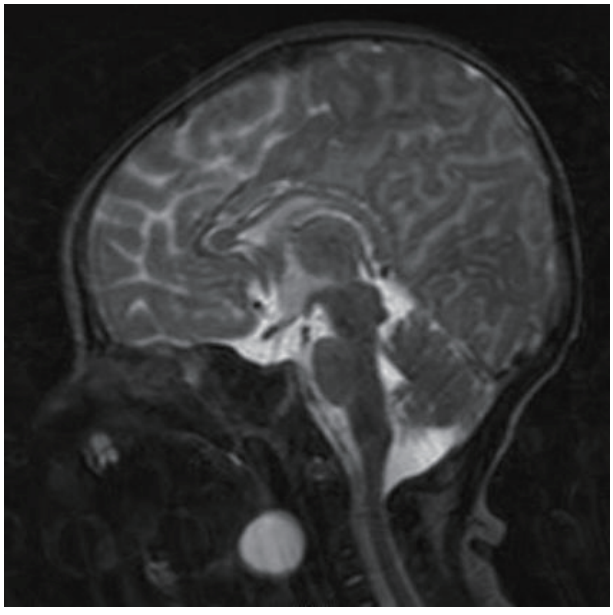

Fig. 3 Magnetic resonance imaging (T2W sagittal section) shows a 1.4 ×1.5 cm cyst in diameter (arrow) in the base of tongue.

in the infant. According to DeSanto's classification, there are three kinds of laryngeal cysts, based on the location, size, and association with the mucosa: saccular, ductal, and thyroid-cartilage foraminal cysts[7]. This classification is frequently used. Ductal cysts are common forms of laryngeal cysts. According to Newman's classification, cysts are classified on the basis of histological diagnosis: epithelial, tonsillar, and oncocytic cysts[8]. However, histological morphology cannot fully cover all forms of laryngeal cysts clinically. According to Arens' classification, cysts are classified on the basis of the origin and development of the cysts: congenital, retention, and inclusion cysts[9]. Except for oncocytic cysts, the anatomical classification is more important for the treatment. However, a complete classification would help a clinician better understand the development of laryngeal cysts.

When a cystic lesion is found in the throat area, it is necessary to perform a differential diagnosis for a laryngeal cyst from the lingual thyroid, thyroglossal duct cyst, thyroid remnant cyst, lymphangioma, hemangioma, teratoma, cystic hygroma, or dermoid cyst in order to reach an early diagnosis[5,6]. Neck ultrasonic examination accompanied with a thyroid scan would differentiate a laryngeal cyst from a functional ectopic thyroid gland. CT and magnetic resonance imaging (MRI) can distinguish a lesion as a cyst or a solid tumor, the lesion range, and the relationship between the cyst and surrounding tissue. Congenital stridor is often seen with laryngomalacia. Laryngeal cysts cause 0.9–4.3% of stridor but when combined with laryngomalacia, they can reach up to 64–91%[10]. Transnasal fiberoptic laryngoscopy can differentiate a simple laryngeal cyst or one combined with laryngomalacia. This case was diagnosed as a laryngeal cyst combined with laryngomalacia.

Treatments for an epiglottic cyst consist of suction, deroofing, or total resection. Although suction is simple to perform, it leads to a high recurrence rate and is infrequently used now. Currently, deroofing is the standard treatment for epiglottic cysts and it leads to a rapid recovery and low recurrence rate[10]. Therefore, microlaryngoscopy was performed to remove the lesion with a carbon dioxide laser. The laser has a precise and coagulating characteristic, avoiding the cumbersome maneuver of instruments in the narrow space. The cyst epithelia can be vaporized by the laser during deroofing and this prevents any side effects caused by soft tissue depositing in the trachea[11]. Total resection is done under the C-type microlaryngoscopy with a side opening.

In summary, a congenital epiglottic cyst is a rare affliction, but treatable, associated with potential life-threatening airway obstruction. This case was a 1-month-old baby boy who came to our hospital with respiratory distress and low appetite. After the diagnosis of epiglottic cyst by transnasal fiberoptic laryngoscopy, MRI, and CT, microlaryngoscopy was performed to completely remove the lesion. After surgery, the patient resumed a normal breathing pattern with no complications. There was no recurrence after the 1-month follow-up.

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先天性會厭囊腫引起的呼吸道阻塞

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摘要

先天性會厭囊腫伴隨呼吸道阻塞是一種罕見的疾病。本院於 2012 年 12 月經歷一名出生後 1 個月之男嬰，主訴呼吸窘迫和吸氣性喘鳴。經由喉纖維內視鏡、核磁共振攝影及電腦斷層掃描檢查，診斷為阻塞性上喉囊腫。立即進行顯微喉鏡手術，徹底清除病灶。手術後兩天，患者恢復正常的呼吸模式，餵食明顯改善，並且沒有再出現喘鳴或呼吸道阻塞。術後一個月門診追蹤無復發跡象。

關鍵詞：會厭囊腫、喘鳴、呼吸道阻塞

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Case Report

Transient Splenial Lesions in Children with Influenza-associated Encephalitis/Encephalopathy: A Case Report and Literature Review

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Abstract

Influenza-associated encephalitis/encephalopathy (IAEE) in children often has a grave prognosis. We report a 2-year-old boy with IAEE whose magnetic resonance imaging (MRI) showed a transient splenial lesion on diffusion-weighted images. His neurological manifestations quickly improved and were consistent with the resolution of the splenial abnormality on follow-up MRI. Transient splenial lesions in children with IAEE are rarely reported, particularly in children under the age of 5 years. Here, we report the youngest case and review the clinical features of this novel type of encephalopathy.

In conclusion, we suggest that when a clinical diagnosis of IAEE is made, and simultaneously, a transient focal lesion in splenium is found on MRI, further investigation is unwarranted if the clinic course and outcome are favorable.

Key words: Influenza, encephalitis, splenium.

Introduction

It is well known that influenza virus infection may invasive central nervous system (CNS) involvement. The clinical spectrum of the disorder includes Reye's syndrome, acute necrotizing encephalopathy, and other severe CNS diseases[1,2]. Influenza-associated encephalitis/encephalopathy (IAEE) is a complex clinical syndrome that may cause a rapidly progressive clinical course and poor outcome[1-4]. We herein report a 2-year-old boy with IAEE whose magnetic resonance imaging (MRI) showed a transient ovoid lesion in the central splenium of the corpus callosum on diffusion-weighted images (DWI). His neurological manifestations quickly improved, and consequently, the splenial abnormality resolved on 1-month follow-up MRI.

Case Presentation

A previously healthy 2-year-old boy initially presented with a 1-day history of prodromal illness consisting of high fever, myalgia, and vomiting before admission. On the evening of day 2 of his illness, he suddenly experienced loss of consciousness followed by upward gaze and a tonic posture with flexed arms and extended legs, evolving to a generalized clonic seizure. The seizure lasted for a few minutes and was repeated twice in 20 min. He was sent to our pediatric emergency department. On arrival, physical examination showed the following vital signs: blood pressure, 70/48 mmHg; heart rate, 107/min; respiratory rate, 22/min; and a body temperature of 38.0°C. He was lethargic and experienced delirium; his throat showed injected, but chest and abdominal

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findings were unremarkable. Neurological examination revealed normal muscle power in all extremities and the trunk and normal deep tendon reflex without Kernig's or Babinski's sign.

Laboratory data included white blood cell counts of 8.0 × 10^9/L, platelet counts of 411 × 10^9/L, and hemoglobin levels of 12 g/dL. Serum electrolytes, C-reactive protein, urea nitrogen, creatinine, glucose, aspartate aminotransferase, alanine aminotransferase, ammonia, and lactate levels were within the normal range. Cerebrospinal fluids (CSF) examination revealed protein and glucose levels of 14 mg/dL and 67 mg/dL, respectively, and no pleocytosis. No microorganisms were isolated. Virus identification from his throat swab showed influenza A, and a diagnosis of IAEE was made. Electroencephalography (EEG) showed slow background activity, and no paroxysmal discharge was found. Computed tomography of the brain was normal. MRI was performed on day 3; T1-weighted, T2-weighted, and fluid-attenuated inversion recovery (FLAIR) images demonstrated no abnormal findings. DWI exhibited a solitary, ovoid, and well-circumscribed lesion with a high intensity in the central splenium of the corpus callosum, and an apparent diffusion coefficient (ADC) map demonstrated low ADC values at the corresponding lesion (Fig. 1 A, B). His neurological manifestations quickly improved, and he completely recovered on day 6, and no antiviral medication or corticosteroids were prescribed. He was discharged without subsequent neurological sequelae. One month later, he underwent follow-up MRI and demonstrated complete resolution of abnormality on previous lesion (Fig. 2A, B).

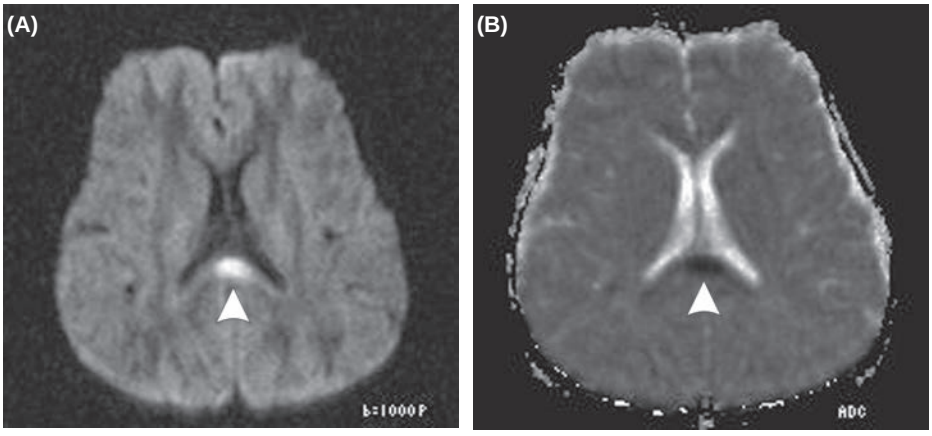

Fig. 1 (A) Axial diffusion-weighted images (DWI, spin-echo planar imaging; TR=4521; b=1000) and (B) apparent diffusion coefficient (ADC) map. An ovoid lesion (arrow head) in the central splenium of the corpus callosum exhibited high intensity in DWI and reduced in ADC.

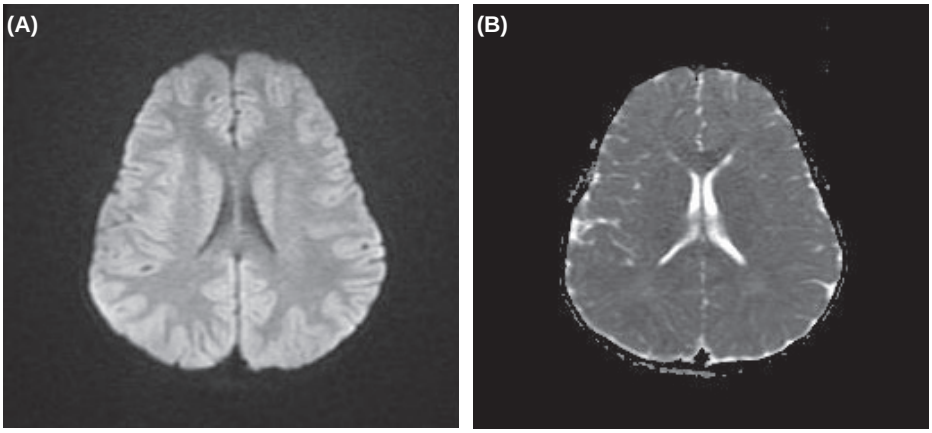

Fig. 2 Follow-up DWI (A) and ADC map (B) obtained one month later did not show the lesion.

Discussion

IAEE is complex and notorious clinical disease in which influenza virus may invade CNS causing fulminant inflammation with edematous changes in the brain, leading to considerably high mortality and disability[1-3]. In an effort to better classify this complex disease, IAEE with a transient splenial lesion in the corpus callosum was described as new disease entity[5]. This rare and novel type of IAEE with a transient lesion in the splenium of the corpus callosum was first described in two pediatric patients by Takanashi et al.[5] in 2004. Only five additional pediatric cases have been reported, in which all children were older than 5 years.

Table 1 summarizes the six known IAEE patients with transient splenial lesions[5-8], including our case. All patients were previously healthy and none had a history of seizures or took antiepileptic drugs. Analysis of neurological manifestations showed that seizures were the most common symptoms of IAEE with a transient splenial lesion in the corpus callosum, which was prominent in three patients between days 2 and 3. Moreover, in three seizure-free patients, one presented with muscle weakness and somnolence, one with hemiparesis and ipsilateral facial paralysis, and one with had disorientation and hallucination. EEG showed slow background activity in four patients and normal background activity in the remaining two. Laboratory data on blood counts, routine biochemistry, and CSF analysis in all six cases were within the normal range. Importantly, nasopharyngeal swab specimens collected from all patients tested positive for influenza, although the virus could not be detected in CSF samples. Among these cases, influenza A was identified in four patients and influenza B in two. All patients had focal splenial lesions without contrast enhancement, none of which were detected on T1-weighted images. It should be noted that splenial lesions were best visualized on DWI than on T2-weighted and FLAIR images. These splenial lesions showed a high intensity on DWI and a decreased intensity on the ADC map.

Treatment options varied between the cases; two received antiviral medication, three received symptomatic treatment, and one was administered corticosteroids intravenously. Although, all patients suddenly developed neurological symptoms, they all fully recovered between days 3 and 6. This suggests that the course of IAEE with a transient splenial lesion is relatively benign and self-limiting. Furthermore, the duration of this disease is short, lasting less than 1 week, and there were no residual neurological sequelae. The clinical manifestations of our patient and the other five reported cases show that the clinical course and outcome of IAEE with a transient splenial lesion are completely different from those of other severe forms of IAEE, which have a high rate of mortality and physical impairment[1-4].

The differences in pathophysiologic features of IAEE with or without a temporal lesion in the corpus callosum remain unclear. However, based on clinical

Table 1. Clinic profiles of pediatric patients of IAEE with transient splenial lesion

Reference	Takanashi et al.[6]	Takanashi et al.[6]	Tada et al.[5]	Bulakbasi et al.[1]	Matsubara et al.[3]	Present case
Pathogen	Influenza A	Influenza B	Influenza A	Influenza A	Influenza B	Influenza A
Age/Sex	7/F	11/M	5/M	6/F	12/F	2/M
CNS symptoms (day of onset)	Disorientation, hallucination(2)	Left facial palsy, right hemiparesis(4)	Seizure, delirium(2)	Seizure(3)	Muscle weakness, somnolence(2)	Lethargy, delirium seizure(2)
Day of Clinic recovery	4	5	3	6	5	6
EEG	Normal BA	Normal BA	Slow BA	Slow BA	Slow BA	Slow BA
Normalization on follow-up MRI	No available	Yes	Yes	Yes	Yes	Yes
Therapy	Antiviral (Amantadine)	Antiviral (oseltamivir)	No	No	Intravenous Methylprednisolone	No

CNS: central nervous system; EEG: electroencephalography; MRI: magnetic resonance imaging; M: male; F: female; BA: background activity.

observations, it was proposed[2,6,9] that the novel form of IAEE shows reversal of restricted diffusion and lacks both significant contrast enhancement and cerebral edema of fulminant hepatic failure. Collectively these findings suggest that influenza virus infection in these patients is limited. Therefore, viral infection is not sufficient to cause intense immunological responses that result in the breakdown of the blood–brain barrier, leading to brain damage that is commonly seen in other severe types of IAEE. Furthermore, negative CSF examination suggests that influenza virus is rapidly cleared from CNS, which may explain the relatively benign clinical course and favorable outcome of this disorder. Our findings also highlight the fact that antiviral or corticosteroid therapy may not be clinically warranted for treatment of the disease. However, further investigation is needed to fully understand the processes involved.

In conclusion, we report a pediatric patient with IAEE who presented with a transient lesion in the splenium of the corpus callosum. We found that DWI is the best tool to identify abnormal splenial lesions in patients. Furthermore, the hallmarks of this disorder in affected children are rapid resolution of clinical symptoms and favorable outcome, and invasive intervention is unwarranted.

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兒童之流行性感冒病毒所致腦炎之暫時性胼胝體壓部病兆：病歷報告及文獻綜述

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摘要

流行性感冒病毒所造成之腦炎或腦病變，在兒童常造成嚴重的傷害。此篇文章報告一位罹患流行性感冒病毒所造成之腦炎的 2 歲男童，其腦部核磁共振在擴散權重影像顯示暫時性胼胝體壓部病兆。病童於神經學上的病徵表現快速地改善，並在後續追蹤之腦部核磁共振顯示胼胝體壓部病兆消除。兒童胼胝體壓部病兆案例報導甚少，且尤其是年齡低於五歲之案例。此篇文章報告最年輕之案例，且對此型腦病變其臨床上特殊表現作總結性回顧分析。

於本文結論，我們建議當臨床上已診斷流行性感冒病毒相關腦炎，且腦部核磁共振同時只顯示胼胝體壓部之病兆，其臨床上會呈現較佳之病程及好的預後，其進一步侵犯性的檢查可能是不需要的。

關鍵詞：流行性感冒、腦炎、胼胝體壓部

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Case Report

Malignant Thyroid Peripheral Nerve Sheath Tumor: A Case Report

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Abstract

Primary peripheral nerve sheath tumors of the thyroid gland (TPNST) are exceptionally rare tumors that can be classified as benign or malignant. Benign TPNSTs include neurofibromas and schwannomas. Only two cases of isolated neurofibroma of the thyroid have been reported [1].To date, three cases of malignant TPNSTs have been reported in the literature. Malignant thyroid peripheral nerve sheath tumors (MTPNST) can be confused with anaplastic carcinomas, Riedel's thyroiditis, or other soft tissue tumors that may occur in the thyroid region and that usually present as an asymptomatic neck mass in adults. Here we describe a case of MTPNST in a 77-year-old woman who presented with a left stony thyroid mass, which rapidly grew over the course of a few weeks and was painless. The patient underwent surgery and received radiotherapy; she died 17 months after the surgical intervention.

Key words: Malignant Thyroid Peripheral Nerve Sheath Tumor (MTPNST), Thyroid peripheral nerve sheath tumor (TPNST), malignant thyroid tumor

Introduction

Malignant primary peripheral nerve sheath tumors (MPNSTs) are rare neoplasms with an estimated incidence of 0.1 per 100,000 per year in the general population. These tumors account for approximately 5–10% of all soft tissue sarcomas and have a strong association with neurofibromatosis type 1 (NF-1), also known as von Recklinghausen's neurofibromatosis[2,6,10].

Clinically, an MPNST typically presents as a progressively enlarging painless mass[3-5]. Majority of MPNSTs arise either de novo or from preexisting neurofibromas, with an estimated incidence of malignant transformation ranging from 3% to 30%. MPNSTs rarely arise as schwannoma, ganglioneuroma, or phaeochromocytoma[4,6,19]. Despite advances in diagnostic techniques and ultrastructural analysis, controversies about the natural history of these uncommon neoplasms remain[5,6]. The general term MPNST is utilized because these tumors tend to be histologically diverse[16,19]. Microscopically, MPNSTs are highly cellular, focally polymorphic tumors composed of spindle cells arranged in bundles or fascicles with high mitotic rates; they have indistinct cytoplasmic borders and a variable degree of nuclear pleomorphism[6,8].

Currently, identification of Schwann cells by electron microscopy and immunohistochemical analysis are useful in diagnosing MPNSTs. Immunohistochemically, the majority of MPNSTs express the neuroectodermal marker S-100 protein and the mesenchymal marker vimentin, whereas cytokeratin and desmin are rarely expressed. The p53 tumor suppressor protein, a marker for tumor aggressiveness, is frequently expressed. Ki-67 (MIB-1) immunoreactivity is used to evaluate the tumor growth percentage and typically ranges from 5% to 65%[4,7,14].

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Management of MPNST is often challenging, particularly when localized in the head and neck. Once the diagnosis is confirmed, wide surgical excision is the mainstay of treatment and prolongs survival[5,15]. Because regional lymph node metastases are notably rare, prophylactic neck dissection is generally not recommended[6,9]. Distant metastases are noted in up to 80% of patients with MPNST, primarily in the lung[7,14]. The role of radiotherapy and chemotherapy in the treatment of this tumor remains controversial[8,9]. MPNST has been described as being highly radioresistant. Although some recent reports recommend the use of postoperative radiotherapy, there is no clear evidence of benefit[8,10,12,13].

Case Report

A 77-year-old Chinese woman was referred to our department in March 2008 with a 5–6-cm painless mass on her anterior lower neck that she had noticed approximately 2 weeks previously. She felt a foreign body sensation but had neither dyspnea nor dysphagia. She presented with no signs of hyperthyroidism or other symptoms. Her medical history included cervical carcinoma treated with a complete course of radiotherapy, diabetes mellitus, and chronic renal failure, for which she underwent regular medical follow up. There was no documented history of von Recklinghausen's disease in her family. Physical examination revealed a left stony enlarged thyroid mass without tenderness. Neck sonography revealed a 4.0 × 3.1-cm irregular solid nodule in the left thyroid gland associated with macro- and microcalcifications (Fig. 1). Computed tomography (CT) scan showed an ill-defined left thyroid gland mass of approximately 5.0 × 5.3 × 4.5 cm in size, with a tracheal deviation (Fig. 2). The laboratory analyses determined the following titers: free T4 = 4.300 mcg/dL, thyroid-stimulating hormone = 2.880 mU/L, and thyroglobulin = 2693.000 ng/mL. Surgical intervention was planned because of signs of compression

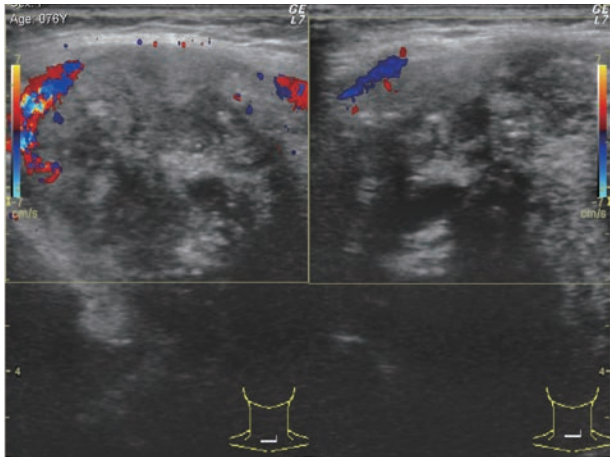

Fig. 1 Neck ultrasonography with color Doppler: Showed a 4.0 x 3.l cm irregular solid mass is observed in the thyroid associated with some macro- and microcalcifications.

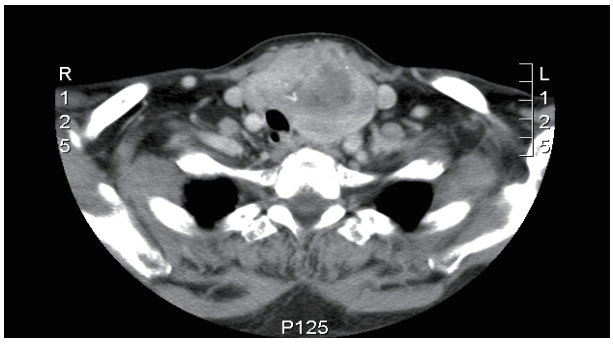

Fig. 2 Cmoputed tomography scans of the head and neck after intravenous contrast:Enlargement of bilateral thyroid glands mainly at left lobe, measuring approximately 5.0 x 5.3 x 4.5 cm. The mass caused external compression of trachea (arrow) and cervical esophagus with deviation to the right side.

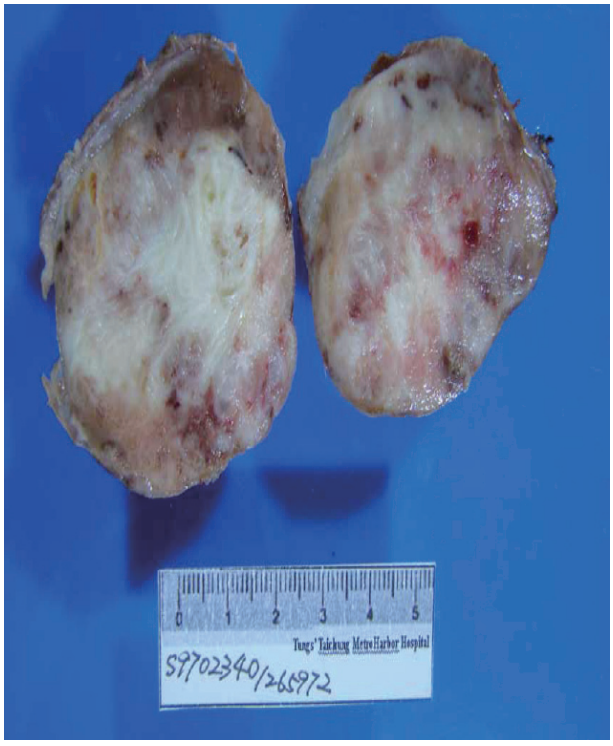

Fig. 3 Grossly, the tumor mass7.0 x 5.8 x 4.5 cm measured. The out surface shows brown gray in color and firm to focal hard in consistency.

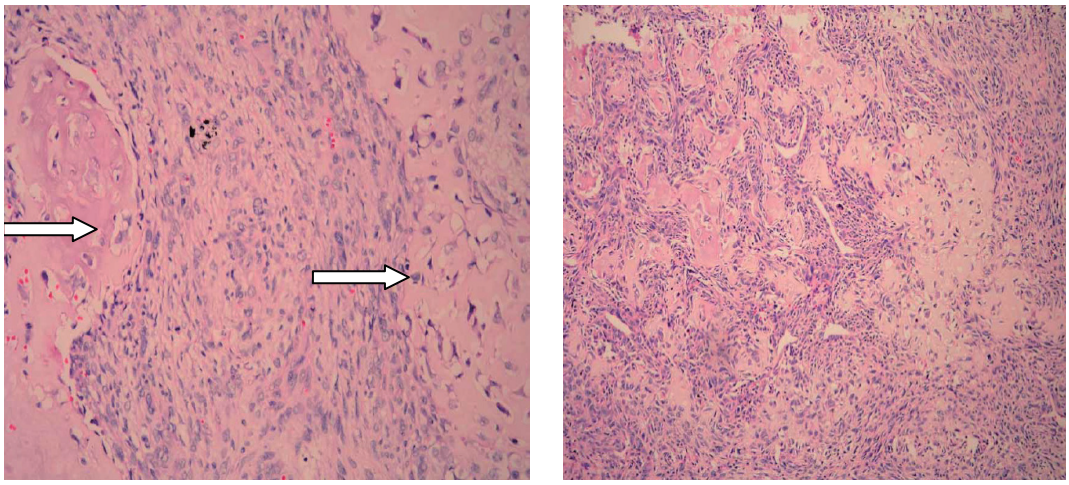

Fig. 4 Microscopic findings: (right) Low-power view of the tumor showed invasion into the surrounding soft tissue (hematoxylin-eosin, 10x). (left) High-power view showing proliferation of spindle-shaped cells with elongated, pleomorphic nuclei and brisk mitosis. Focal osteogenic sarcoma differentiation (arrows) was also show (hematoxylin-eosin, 200x).

and total thyrodectomy was performed in March 2008. The thyroid mass weighed 78 g and measured 7.0 × 5.8 – 4.5 cm in size. The tumor mass appeared brown–gray on the surface and had a firm to hard consistency. (Fig. 3). Histology revealed that the tumor consisted of spindle cells with focal osteogenic sarcoma differentiation and abundant mitosis (Fig. 4), and positive staining for S-100, neuron-specific enolase, actin, and Ki-67 (Fig. 5). In contrast, staining for cytokeratin and thyroid transcription factor 1 was negative. Pathological examination revealed a malignant peripheral nerve sheath tumor with an osteogenic sarcoma component, which is consistent with the macro- and microcalcification of the ultrasonographic finding. However, the patient died 17 months postsurgery due to an infiltrating urothelial carcinoma of the urinary bladder (T3N1Mx with left orifice invasion) and sepsis induced multiple organ failure.

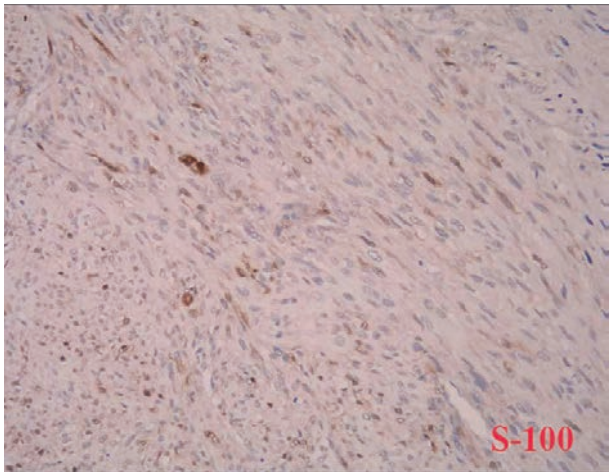

Fig. 5 Immunohistochemical stains shows tumors positive staining for S-100 (IHC stain, x 200).

Discussion

MPNSTs are uncommon malignant tumors that tend to show aggressive behavior and have a 5-year survival rate of approximately 40–60%. Among the three reported cases of MPNST[1,17,18], the common symptom was the presence of a mass accompanied by pain.

The recommended primary treatment entails surgical excision, with or without adjuvant radiation or chemotherapy. Survival appears to be related to the extent of tumor resection and the size of the tumor. Ducatman et al.[6] were among the first to report improved survival upon complete resection of the tumor. In their study, only patients who had undergone total excision of a small tumor showed a statistically significant improved survival. Patients with large, completely excised tumors showed no difference in survival from those with small or large incompletely excised tumors. The addition of radiation therapy and chemotherapy did not appear to alter the prognosis. In a study by Wanebo et al. in 1993[16], aggressive surgery significantly improved disease-free survival; however, overall survival was

not affected.

Chemotherapeutic treatments of soft tissue sarcomas were only moderately efficacious in patients with MPNST; only doxorubicin and ifosfamide have shown appreciable efficacy. Currently, considerable controversy surrounds the use of adjuvant chemotherapy in adults with soft tissue sarcomas.

The most important prognostic factors affecting the survival of patients have been reported to be tumor size and location. High-grade and large MPNST appear to have a particularly aggressive behavior. Overall, no other consistent prognostic factors have been identified. The prognosis of patients with MPNSTs of the head and neck is often relatively poorer than that of patients with MPNSTs of the extremities and the trunk, with documented 5-year-survival-rates ranging from 15% to 35%[6,10,11]. However, these differences were observed mainly because of a difference in local control. Our report is aimed to provide additional data to better understand these uncommon malignant tumors.

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甲狀腺惡性周圍神經鞘膜瘤：病例報告

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摘要

甲狀腺之原發性周圍神經鞘膜瘤，不管是良性或惡性都是非常罕見的。到目前為止甲狀腺的惡性周圍神經鞘膜瘤的報告案例僅僅只有三例，而甲狀腺的良性周圍神經鞘膜瘤可分為神經纖維瘤及許旺細胞瘤二種形態。在這當中，也只有兩例的甲狀腺神經纖維瘤曾被發表過。甲狀腺惡性周圍神經鞘膜瘤為一無症狀的頸部腫瘤也常常與成人的頸部其他腫瘤如頸部的未分化癌，立得氏甲狀腺炎或其他的軟組織腫瘤混淆。在這，我們發表一個甲狀腺惡性週圍神經鞘膜瘤的案例，個案為一名 77 歲女性，自覺左側前頸區有一無痛性的腫塊，在短短的數週變大許多，病人在診斷後接受了手術及術後放射性治療，但病人仍於術後 17 個月死亡。

關鍵詞：甲狀腺周圍神經鞘膜瘤、甲狀腺惡性周圍神經鞘膜瘤

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Case Report

Severe Hypokalemia and Rhabdomyolysis Associated with Conn's Syndrome: Clinical Approach to Aldosterone-Related Kaliuresis

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Abstract

Background: Profound hypokalemia, accompanied by rhabdomyolysis, often makes rapid and accurate differential diagnosis challenging.
Methods: We used the potassium-to-creatinine ratio in freshly voided urine to differentiate the etiology of hypokalemia.
Results: A 27-year-old man with a 5-year history of hypertension presented with an initial biochemistry indicating marked hypokalemia (1.8 mmol/L) and metabolic alkalosis with low urinary potassium excretion. The patient did not have a history of abusing alcohol or diuretics. Persistent hypokalemia (3.0 mmol/L) continued despite aggressive potassium supplementation. Hyperaldosteronism was confirmed, and computed tomography revealed a 2.4-cm-diameter mass over the left adrenal gland. After undergoing laparoscopic left adrenalectomy, the patient's condition improved without further medication.
Discussion: It is feasible to use the potassium-to-creatinine ratio in freshly voided urine for the differential diagnosis of hypokalemia. However, this case highlights that the initial kaliuretic effect of aldosterone may be restricted in severe hypokalemia.

Key words: hypokalemia, rhabdomyolysis, hyperaldosteronism, Conn's syndrome

Introduction

Primary hyperaldosteronism (PHA), the most common cause of secondary hypertension, most commonly occurs because of bilateral idiopathic adrenal hyperplasia. Although aldosterone mediates urinary potassium excretion, most patients with PHA are normokalemic[1]. Hypokalemia can be noted in a minority of cases and may induce severe complications, such as lethal cardiac arrhythmia, ascending paralysis, and probable respiratory arrest, particularly if serum potassium levels fall to <2.0 mmol/L[2-5].

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Assessment of urinary potassium excretion helps in the differential diagnosis of hypokalemia. Collecting 24-h urine samples is the preferable method for determining potassium excretion; however, it is often not feasible. On the other hand, random measurement of freshly voided urine is considerably simpler to perform. Both potassium-to-creatinine ratio ($U_K/U_{creatinine}$) and fractional excretion of potassium (FeK^+) are also useful diagnostic tools[3,6-8]. High $U_K/U_{creatinine}$ (>1.5) and FeK^+ (>6.5%) are considered indicators of high urinary potassium loss[7-9]. We present a case of severe hypokalemia complicated by rhabdomyolysis that was induced by Conn's syndrome. High urinary potassium excretion was noted, but the preliminary kaliuretic effect was not obvious. We suggest that comparison of repeated $U_K/U_{creatinine}$ values can yield faster and more accurate diagnosis.

Case Report

A 27-year-old man presented with limb pain and a 2-day history of progressive weakness. Initially, he experienced slight pain in his lower limbs that increased in severity and gradually extended to his upper limbs, with a gradual decrease in his muscle strength over 2 days. The patient had a 10-year history of ankylosing spondylitis for which he had undergone treatment with sulfasalazine. Diclofenac and acetaminophen/orphenadrine were frequently prescribed for back and limb soreness. He had a 5-year history of hypertension treated with bisoprolol and amlodipine; he did not take glycyrrhizic acid or diuretics. Upon admission, the patient's blood pressure (164/98 mmHg), pulse (83 beats/min), body temperature (36.8°C), height (173 cm), and weight (95 kg) were recorded during physical examination. The

Table 1. Patient laboratory values upon admission

	Study	Reference range	Value
Blood			
	White blood cell count, /uL	4800–10,800	10,270
	Red blood cell count, /uL	4.7–6.1 × 10^6	4.9 × 10^6
	Hemoglobin level, g/dL	14–18	15.4
	Platelet count, /uL	130–400 × 10^3	306 × 10^3
	Sodium level, mmol/L	136–145	141
	Potassium level, mmol/L	3.5–5.1	1.8
	Urea nitrogen level, mg/dL	7–20	13.9
	Creatinine level, mmol/L	0.044–0.1056	0.0748
	eGFR[a], mL/min	-	114.1
	AST[b] level, U/L	10–30	34.4
	ALT[c] level, U/L	2–32	14.1
	Calcium level, mg/dL	8.8–10.6	9
	Phosphate level, mg/dL	2.7–4.5	1.67
	Magnesium level, mg/dL	1.8–2.55	1.86
	Creatine kinase level, U/L	<167	5638
	Glucose level, fasting, mg/dL	70–115	96
	Total cholesterol level, mg/dL	110–200	178
	Triglyceride level, mg/dL	21–175	98
	Uric acid level, mg/dL	2.4–7	4.95
	pH	7.35–7.45	7.521
	Bicarbonate level, mmol/L	22–28	27.2
Urine			
	Sodium level, mmol/L		66
	Potassium level, mmol/L		7
	Chloride level, mmol/L		59
	Creatinine level, mmol/L		7.48

[a]estimated glomerular filtration rate, [b]aspartate aminotransferase, [c]alanine aminotransferase

patient's heart beat was regular without a murmur or abdominal bruit, but electrocardiography revealed presence of U-waves and flattening of T-waves. The results of his laboratory studies are shown in Table 1. Severe hypokalemia (1.8 mmol/L) and high creatinine kinase (CPK, 4971 U/L) levels were the prominent findings. His urinary potassium excretion ($U_K/U_{creatinine}$, 0.94; FeK^+, 3.89%) was low.

The patient's muscle symptoms (weakness and myalgia) were alleviated after administration of intravenous potassium chloride and oral potassium citrate. In addition, his CPK levels normalized within 1 week of treatment. However, low serum potassium levels (approximately 3.0 mmol/L) persisted despite aggressive potassium administration (84 mmol of potassium on the first day, 98 mmol on the second day, and 74 mmol per day since the third day) (Figure 1). Further investigation showed an aldosterone level of 37.7 ng/dL (normal range, 1.0–15 ng/dL) and a renin level of <0.13 ng/mL (normal range, 0.15–2.33 ng/mL), indicating a high ratio of plasma aldosterone to plasma renin activity. Other laboratory values were normal, including analyses of thyroid function, urinary vanilmandelic acid and cortisol levels, and abdominal Doppler ultrasound. Abdominal computed tomography (CT) revealed a 2.4-cm-diameter mass over the patient's left adrenal gland.

The patient underwent laparoscopic left adrenalectomy, after which his clinical symptoms were alleviated and blood pressure and serum potassium levels were normalized. Further antihypertensive medication, spironolactone, and potassium replacement therapy were not necessary after surgery. Histological examination of the mass showed adrenal cortical adenoma.

Discussion

Our patient was eventually diagnosed with Conn's syndrome and severe hypokalemia. Excessive potassium excreted into the urine is normally noted in cases of excessive aldosterone secretion[3]. However, the preliminary urinary potassium loss in the present patient was relatively low. Using the potassium-to-creatinine ratio in freshly voided urine ($U_K/U_{creatinine}$) or fractional excretion of potassium (FeK^+) for evaluating urinary potassium loss appeared to have limitations.

Aldosterone has a strong kaliuretic effect, activating the apical epithelial sodium channels of the principal cells in the connecting tubule and cortical collecting duct, resulting in sodium absorption. A luminal negative potential difference, generated by the selective absorption of sodium, drives passive potassium excretion via the renal outer medullary potassium channel (ROMK). Thus, measurement of urinary potassium excretion is a useful diagnostic tool for differentiating the etiologies of hypokalemia[10]. Although collection of 24-h urine samples to determine urinary potassium excretion is the preferable method, it is not practical in many cases. On the other hand, $U_K/U_{creatinine}$ or FeK^+ measurements provide useful assessments[7,8].

The initial kaliuresis was not obvious in our patient. This may have been because a reduction of ROMK activity in the principal cells induced a drop in potassium excretion, particularly in the hypokalemic situation. In addition, the H^+/K^+-ATPase in the intercalated cells is independent of the aldosterone effect and contributes to the reabsorption of potassium in the outer medullary collecting ducts. Therefore, the initial urinary potassium loss may not have been apparent because serum potassium levels were extremely low. The kaliuretic effect of aldosterone was noted after potassium administration (Fig. 1).

Based on our patient's metabolic alkalosis, history of refractory hypertension, and high renal potassium loss, we were able to narrow down the possible cause of the hypokalemia to an excess mineralocorticoid-like state (Fig. 2)[6,9]. Conn's syndrome, resulting from an aldosterone-producing adenoma in the adrenal gland, is a subtype of PHA. Patients with PHA may present with hypertension but most

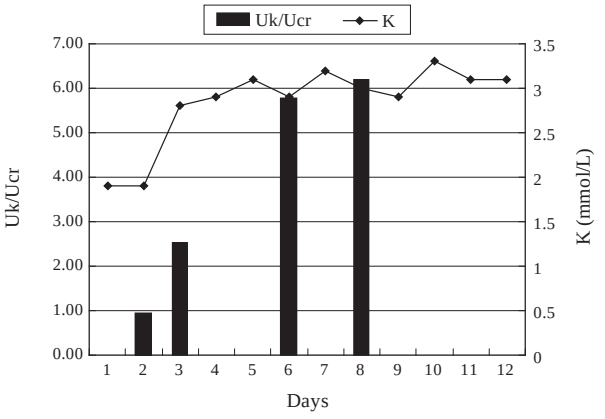

Fig. 1 The association between the serum potassium level and urinary potassium-creatinine ratio ($U_k/U_{creatinine}$)

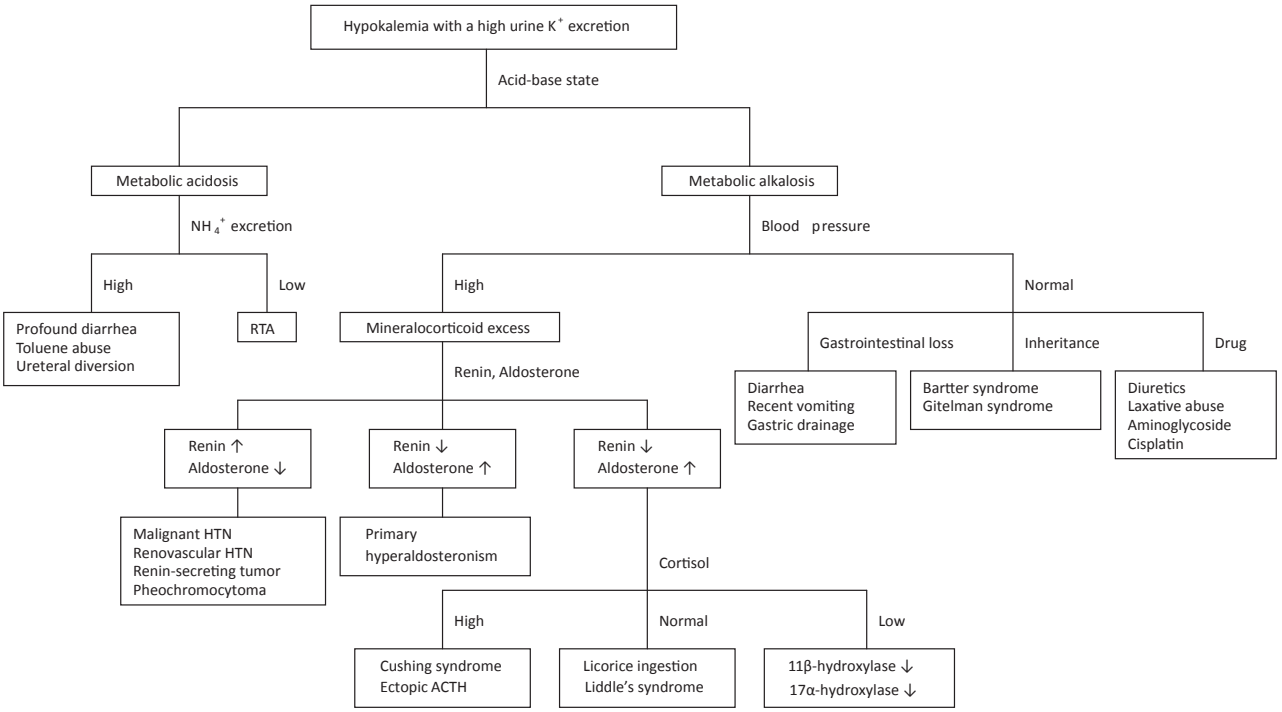

Fig. 2 Approach to hypokalemia exhibiting high urine potassium excretion [6].
RTA, renal tubular acidosis; HTN, hypertension; ACTH, adrenocorticotropic hormone

are normokalemic. Hypokalemia is noted only in approximately 9%–37% of patients with PHA [1,11,12]. However, severe hypokalemia may cause life-threatening clinical manifestations, including lethal cardiac arrhythmias or respiratory arrest [2,4,5]. Rapid and adequate treatment of hypokalemia can prevent the development of such complications. When an adrenal mass is found on CT, the next step to diagnose PHA should be adrenal vein sampling (AVS). However, AVS may be bypassed in young patients with marked PHA because aldosterone-producing adenoma is highly prevalent in these such patients [1,13].

Rhabdomyolysis, defined as skeletal muscle damage with the release of toxic intracellular material, is a rare complication of PHA-related severe hypokalemia. It is most often caused by trauma, ischemia, drugs, toxins, metabolic disorders, infections, or electrolyte disorders[14-17]. Potassium mediates vasodilation in muscle tissues and helps to provide sufficient blood flow during exercise. Thus, diminished blood flow to the muscle resulting from severe hypokalemia may lead to muscle ischemia during exercise. Prolonged ischemia leads to muscle necrosis that may result in rhabdomyolysis, particularly when serum potassium levels reach <2.0 mmol/L[2,18]. A review of the literature showed that severe PHA-induced hypokalemia with rhabdomyolysis commonly leads to a diagnosis of Conn's syndrome[13,16, 19-22]. Moreover, rhabdomyolysis may lead to acute kidney injury and influence serum and urinary creatinine levels. However, no gross hematuria was noted in our patient, and his serum and urinary creatinine levels were normal. The interference of rhabdomyolysis in this patient should be limited.

In conclusion, measurement of urinary potassium excretion is useful for the differential diagnosis of hypokalemia. Repeated $U_K/U_{creatinine}$ measurements should always be considered to avoid any masking effects. In patients with hypokalemia-related rhabdomyolysis induced by hyperaldosteronism, Conn's syndrome should be the first consideration.

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Conn's Syndrome 相關之嚴重低血鉀及橫紋肌溶解症：Aldosterone 相關尿鉀排泄作用之臨床探討

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摘要

背景：嚴重低血鉀合併橫紋肌溶解症，通常帶來需要迅速及正確地做出鑑別診斷的挑戰。
方法：我們使用剛排出尿液中鉀離子及肌酸酐的比例，來鑑別低血鉀的致病原因。
結果：個案本身已有 5 年的高血壓病史，初始血液生化值呈現明顯的低血鉀（1.8 mmol/L）及代謝性鹼血症，同時尿液中鉀離子排出是低的，而個案本身並無濫用利尿劑之情形；經由積極的鉀離子補充後，個案仍呈現持續性的低血鉀（3.0 mmol/L）。隨後高醛固酮血症被證實，左側腎上腺內一直徑 2.4 公分的腫塊亦被發現;經過腹腔鏡手術作左側腎上腺切除後，個案恢復的情況良好，並且無需後續其他藥物治療。
討論：測算剛排出尿液中鉀離子及肌酸酐的比例，作為低血鉀鑑別診斷的工具是有效的方法，然而，在嚴重低血鉀的情況下，醛固酮造成的尿鉀排泄作用可能被侷限。

關鍵詞：低血鉀、橫紋肌溶解症、高醛固酮血症、康氏症

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Case Report

Postobstructive Negative Pressure Edema Associated with Pulmonary Hemorrhage in an Intubated Patient During General Anesthesia: One Sporadic Case Report and Literature Review

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Abstract

Negative pressure pulmonary edema accompanied by hemorrhage as a manifestation of upper airway obstruction secondary to lung cancer is an uncommon problem that is potentially life-threatening. This report presents a case of significant pulmonary hemorrhage and negative pressure pulmonary edema in an intubated patient during general anesthesia. Bronchoscopy showed tumor rupture with bleeding in the right lower lung lobe. We believe that in our case, pulmonary bleeding was due to disruption of the tumor capillaries during general anesthesia. Early diagnosis of pulmonary edema is very important because it affects the patient's postoperative morbidity and mortality. We discuss the possible causes and therapeutic approach, and we briefly review of literature.

Key words: negative pressure pulmonary edema, upper airway obstruction, lung cancer, general anesthesia

Introduction

Negative pressure pulmonary edema (NPPE) after acute upper airway obstruction is a well-described event[1]. This results in a typical clinical picture, with frothy, pink sputum and easily recognizable clinical and radiological findings. However, pulmonary hemorrhage as a manifestation of upper airway obstruction arising in associated with negative pulmonary edema has rarely been reported[2-3]. We report a case in which significant pulmonary hemorrhage and pulmonary edema arose in an intubated patient, resulting in upper airway obstruction. Most cases are treated by mechanical ventilation with positive end-expiratory pressure (PEEP) or CPAP without any other therapy. The prognosis is favorable, with most cases recover within the first 24 h. The aims of this case report are to increase awareness about this condition and review the criteria for its definitive diagnosis and treatment.

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Case Report

A 45-year-old woman (body weight: 45 kg, height: 150 cm) with no history of smoking but with a medical history of hepatitis underwent emergency surgery for stump formation of the left index finger. She had suffered from chronic cough for 2 years. The preoperative chest radiograph showed bronchiectasis over right zone of lung (Fig. 1), and electrocardiography showed no remarkable abnormalities. Blood tests and electrolyte data were within the normal limits.

General anesthesia was induced with fentanyl

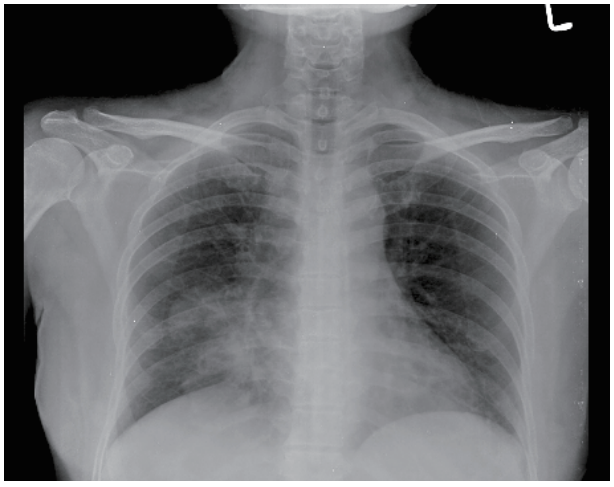

Fig. 1 A chest radiograph showing patchy infiltration at both lower lung lobes (Bronchiectasis)

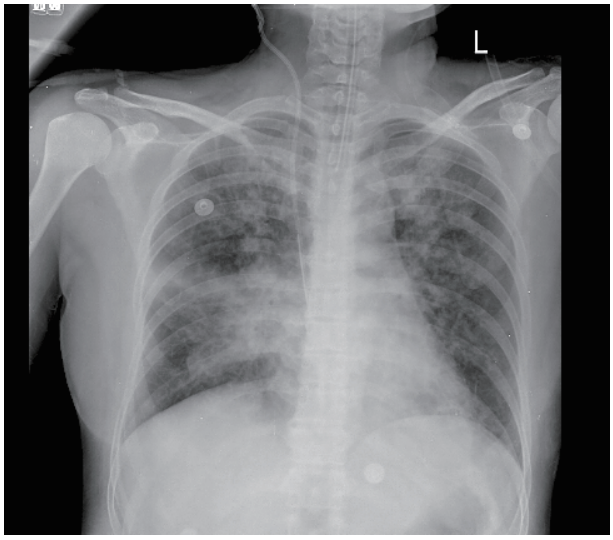

Fig. 2 A chest radiograph showing ill-defined opacification over both lungs.
Pulmonary edema, pneumonia or occult pathology are ruled out.

(100 μg) ,thiopental (250 mg) and maintained with 2% sevoflurane and oxygen. The patient was given rocuronium (40 mg) prior to an endotracheal intubation. Clear bilateral breath sounds were noted, and the capnography trace was normal after induction of general anesthesia. There were no wheezing or urticaria on the body, and the patient's oral cavity was dry. It was a short (30 min) and uneventful surgery, during which the patient received 600 ml intravenous normal saline solution.

Approximately 30 min after the induction of anesthesia, towards the end of surgery, the patient began to spontaneously breathe, but mechanical ventilation became difficult. The patient's peak inspiratory pressure suddenly increased to 40 cmH_2O, and the patient's oxygen saturation decreased to 60-80% immediately afterwards, and a large volume of frothy, pink sputum with bloody secretions appeared exclusively in the endotracheal tube. Heart rate was 155 beats/min and blood pressure was 85/50 mmHg. Arterial blood gases showed pH 7.11, PaO_2 70 mmHg and $PaCO_2$ 110mmHg. Auscultation of lungs at the time revealed bilateral moist rales. The patient was treated with rocuronium (30 mg) to induce neuromuscular blockade, and mechanical ventilator support with 100% O_2 was provided. Adjuvant therapy included intravenous furosemide (20 mg), hydrocortisone (100 mg) and dopamine (5 μg/kg/min). The patient showed rapid improvement.

Based on our impression that the patient was suffering from acute pulmonary edema with unstable blood pressure, she was shifted to the surgical intensive care unit. She was placed on ventilatory support with CMV, with a tidal volume 450 ml, frequency of 15/min, and PEEP of 5cmH_2O on 100% O_2. Chest radiographs, taken 2 h after symptoms began, showed rapidly worsening of diffuse bilateral pulmonary infiltration suggestive of pulmonary edema (Fig. 2). Arterial blood gas data showed pH of 7.29, PaO_2 of 80 mmHg, $PaC0_2$ of 48 mmHg, $HC0_3$ of 20 mmol/L, base excess of 3 mmol/L, $Sa0_2$ of 95%, suggestive of acute respiratory distress syndrome. We performed electrocardiography; a cardiologist was consulted, and we diagnosed the parient with non-cardiogenic acute respiratory distress syndrome. The patient was sedated with a continuous infusion propofol (50 μg/kg/min) and morphine (1 mg bolus) every 4 h.

Clinically, the patient's pulmonary edema and oxygenation resolved rapidly over next 24 h. On the third postoperative day, a chest physician performed bronchoscopy on the patient. There was tumor rupture with bleeding in the tracheobronchial tree and a collection of dark blood in the right lower lung lobe. Biopsy was performed of the right lower lung lobe, and the histopathologic diagnosis was non-small-cell of lung cancer. The patient had progressive improvement in pulmonary edema and was discharged 2 weeks later.

Discussion

NPPE has been defined as a non-cardiogenic edema with transduction of fluid to the pulmonary interstitium in response to an increase in negative intrathoracic pressure secondary mainly to obstruction of the upper airways after extubation[4]. Symptoms and signs of respiratory distress are often present, but frothy, pink sputum is the hallmark sign of NPPE. The incidence of NPPE was 0.094%, and complications develop few minutes after airway obstruction. This obstruction was common after extubation (73.5%) and is caused by laryngospasm in all cases[1,5]. This can affect, on rare occasions, intubated patients in the absence of protection with oral airway when they bite the endotracheal tube during emergence of the anesthesia[6].

NPPE is into two types[7]. Type I NPPE is secondary to acute obstruction of the airway caused by postextubation laryngospasm, obstruction of the endotracheal tube, epiglottis and postoperative vocal cord paralysis. Type II NPPE is caused by the treatment of chronic airway obstruction performed in patients with, for example, tumors, adenoidectomy and obstructive sleep apnea syndrome[8] .

The pathophysiolgy of NPPE includes three mechanism[2]: negative intrathoracic pressure, increase systemic and pulmonary capillary hydrostatic pressure, and mechanical stress on the alveolar-capillary membrane. Negative intrathoracic pressure increases the venous return due to the reduced pressure in the right atrium, with a concomitant increase in systemic and pulmonary capillary hydrostatic pressure. This is complicated by a reduction in interstitial perivascular hydrostatic pressure. This causes an increase in the transcapillary pressure gradient, favoring transudation of fluids to the interstitial space. An increase in central venous pressure is also seen, which prevents the flow in the lymphatic vessels. The increase in pulmonary blood flow is due to increased systemic blood pressure secondary to norepinephrine release in response to hypoxia, hypercapnia, and agitation. This leads to right ventricular distension, causing deflection of the interventricular septum to the left and decreases in the cardiac output. The increased systemic vascular resistance increases left ventricular wall tension, which contributes to a decrease in the left ventricular ejection fraction. In addition, pulmonary capillary rupture with an associated loss of protein is responsible for frothy, bloody, or pinkish secretions. The third mechanism shows that the loss of capillary integrity can result from the mechanical stress on the alveolar-capillary membrane, leading to alveolar edema and hemorrhage. However, any one of the mechanisms mentioned above, in isolation or in combination, can contribute to the development of pulmonary edema.

It is very uncommon for pulmonary hemorrhage to be the presenting feature in a patient subjected to severe negative pulmonary pressure, as was seen in our case presented here. In fact, only one case has been reported : Nam YS et al. recently described a case in which pulmonary hemorrhage was associated with negative pressure edema in an intubated patient.

It is unclear why pulmonary hemorrhage only rarely ocurrs in upper airways obstruction, whereas NPPE is relatively common. It is also uncertain where the bleeding originates[6]. In this case, there was no fluid overload, and cardiogenic pulmonary edema was ruled out because there was no pre-existing heart disease, and the electrocardiogram remained absolutely normal throughout the patient's stay. Furthermore, as per the cardiologist's opinion, the patient was diagnosed as NPPE. The pathophysiological of NPPE was mechanical stress on the alveolar-capillary

Table 1. Causes of NPPE

Type 1 NPPE
Postextubation laryngospasm
Epiglottis
Croup
Choking/foreign body
Strangulation
Hanging
Endotracheal tube obstruction
e.g., biting, secrections
LMA blockage
e.g., biting, displacement
Laryngeal tumor
Goiter
Mononucleosis
Postoperative vocal cord paralysis
Migration of Foley catheter balloon used to tamponade epistaxis
Near drowning
Intraoperative direct suctioning of endotracheal tube
Type II NPPE
Posttonsillectomy/ adenoidectomy
Postremoval of upper airway tumor
Choanal stensois
Hypertrophic redundant uvula

membrane, intrathoracic pressure alternation, hemodynamic alternation, and arterial hypoxemia[9]. We believe that pulmonary bleeding in our case was due to disruption of the tumor capillaries during general anesthesia.

The most common site of lung cancer is the wall or epithelium of the bronchial tree. Early lung cancer may cause no symptoms; thus, the disease may be advanced when it is diagnosed. Lung cancer tend to spread very early, and only 15% are detected in their early stage[10,11]. Chest radiography usually shows an advanced lesion and can detect a lesion up to 2 years before signs and symptoms appear. Bronchoscopy can identify the tumor site and provide materials for cytologic and histologic study.

After the diagnosis of NPPE has been made, treatment is directed toward reversing hypoxia and decreasing the fluid volume in the lungs. Maintaining the airway and providing supplemental oxygen are usually the only measures required for a positive outcome.

Mechanical ventilation is particularly useful in the treatment of patients with noncardiogenic pulmonary edema. If oxygenation does not improve in the intubated patient, PEEP should be administered to promote alveolar expansion. In these conditions, we usually at 5-20 cm H_20. This actually increases functional residue capacity and avoids the risk for further pulmonary injury[11].

Pulmonary edema has many causes, methods of prevention, and treatment. In the case described here, the symptoms and signs were clearly evident, the treatment was rapidly instituted . Prompt diagnosis and treatment markedly improves patient prognosis and significantly decreases morbidity and mortality, which may occur if NPPE is left untreated.

Table 2. Differential diagnosis for NPPE based on initiating mechanism of pulmonary edema

Imbalance of starling forces
Increased pulmonary capillary pressure
Increased pulmonary venous pressure without left ventricular failure
e.g., mitral stenosis
Increased pulmonary venous pressure secondary to left ventricular failure
Increased pulmonary capillary pressure secondary to increased pulmonary arterial pressure (so-called over perfusion pulmonary edema)
Decreased plasma oncotic pressure
Hypoalbuminemia
Increased negativity of interstitial pressure
Rapid removal of pneumothorax with large applied negative pressures (unilateral)
Large negative pleural pressure as a result of acute airway obstruction alone with increased end-expiratory volumes (i.e., asthma)
Lymphatic insufficiency
After lung transplant
Lymphangitic carcinomatosis
Fibrosing lymphangitis (e.g., silicosis)
Altered alveolar-capillary membrane permeability (acute respiratory distress syndrome)
Infectious pneumonia-bacterial, viral, parasitic
Inhaled toxins(e.g., phosgene, ozone, chlorine, fumes, nitrogen dioxide, smoke)
Circulating foreign substances (e.g., snake venom, bacterial endotoxins)
Aspiration of acidic gastric contents
Acute radiation pneumonitis
Endogenous vasoactive substances (e.g., histamine, kinins)
Disseminated intravascular coagulation
Immunlogic-hypersensitivity pneumonitis, medications (nitrofurantoin)
Shock lung in associated with nonthoracic trauma
Acute hemorrhagic pancreatitis
Idiopathic or incompletely understood
High altitude pulmonary edema
Neurogenic pulmonary edema
Narcotic overdose
Pulmonary embolism
After cardioversion
After anesthesia

Conclusion

Pulmonary hemorrhage as a complication of upper airway airway obstruction is uncommon, and in this situation, the usual adverse result is NPPE. We believe that our case provide some further insight into the possible mechanisms underyling the development of hemorrhage (i.e., tumor vessel bleeding) in this situation. The early detection of the signs of this syndrome is vital to successful the treatment and positive patient outcome.

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插管病患在全身麻醉中發生肺出血造成呼吸道阻塞性負壓肺水腫：偶發病例報告及文獻回顧

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摘要

插管病患在全身麻醉中，因肺癌血管破裂出血造成上呼吸道阻塞繼而誘發負壓性肺水腫病例並不常見，但其產生的急性肺水腫常伴隨著高死亡率。我們提出一位 45 歲女性病患，因工作時不慎被機械壓碎其左手食指，經手術醫師詳細檢查後，立即安排進行左手食指殘端形成術。麻醉前評估病患過往無抽煙習慣，但有肝炎和慢性咳嗽病史，術前胸部 X 光檢查於右肺近中隔處有明顯陰影，但比較去年的 X 光片，並無明顯差異（去年 X 光報告診斷為支氣管擴張）。給了病患施行氣管內插管之常規全身麻醉，麻醉過程順利。聽診時雙側呼吸音正常。手術進行 30 分鐘後，快接近手術結束時，發現病患已有自行呼吸現象，但突然病患之氣道壓力升高至 40 ㎝ H_20，給予氣管內抽吸發現有大量粉紅色泡沫和血水。由於後續血氧濃度偏低，血壓偏低，發生不明原因之上呼吸道阻塞所產生的負壓肺水腫，該病患經積極治療肺水腫並於情況穩定後，送至加護病房照顧。後續追蹤其後發生原因，胸腔科醫師在支氣管鏡檢查發現術前 X 光片呈現於右肺近中隔處陰影，其實是肺腫瘤浸潤合併出血因而發生阻塞性負壓肺水腫。全身麻醉手術期間所發生非預期性急性肺水腫，由於病情變化快，早期快速正確的診斷和有針對性的治療措施是挽救病患生命的關鍵。在適當處理下，大多數病患在最初 24 小時內恢復，預後良好。本文旨在報告此一偶發病例，出現在全身麻醉氣管插管病人因顯著肺出血造成上氣道阻塞繼而引發負壓性肺水腫並做相關文獻回顧與討論。

關鍵詞：負壓肺水腫、上呼吸道阻塞、肺腫瘤、全身麻醉

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Case Report

Bilateral Extracranial Carotid Artery Aneurysms Presenting with Acute Cerebral Infarction

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Abstract

Extracranial carotid artery aneurysm (ECCA) is an uncommon cause of embolic stroke. Patients with ECCA present with a variety of clinical symptoms. Careful inspection of the skull base of cross-sectional brain computed tomography (CT) is important for screening high cervical ECCAs. Further cervical magnetic resonance (MR) angiography or CT angiography is necessary to confirm the final diagnosis. Treatment options include open surgery, endovascular intervention, or medical management. Here, we report the case of a patient with bilateral atherosclerotic ECCAs who presented with acute cerebral infarction. The cause of stroke was documented as artery-to-artery emboli from a giant aneurysm of the left high cervical internal carotid artery (ICA).

Key words: extracranial carotid aneurysm, internal carotid aneurysm, cerebral infarction

Introduction

Extracranial carotid artery aneurysms (ECCAs) are uncommon, accounting for 0.4%–4% of all peripheral artery aneurysms[1-3]. The most common causes of ECCA include atherosclerosis, trauma, fibromuscular dysplasia, and prior surgery[4]. Other reported causes of ECCA include neurofibromatosis[5], Takayasu's arteritis[6], infection[7], and radiation[4,8]. The common presenting symptoms of ECCA include a palpable neck mass, acute stroke, transient ischemic attack (TIA), visual symptoms, and aneurysm rupture[4]. Bilateral ECCAs are rare and account for less than 10% of all ECCAs[4]. Here we report the case of a patient with bilateral ECCAs who presented with symptoms of acute stroke.

Case Report

An 86-year-old woman with a history of hypertension was brought to our emergency department on account of sudden onset of right limbs weakness. She had no history of diabetes mellitus or head or neck trauma, and she had not undergone any endovascular procedure previously. There was no associated double vision, slurred speech, swallowing disturbance, or neck pain. On examination, her blood pressure was 132/87 mmHg. Physical examination showed no palpable mass or carotid bruits in the neck. Muscle power in the right limbs were grade 4/5 according to the scale proposed by the Medical Research Council of Great Britain. The patient had a right limping gait owing to weakness of her right lower limb.

Emergency brain computed tomography (CT) did not show abnormal intracranial lesions but revealed prominent dilated bilateral high cervical internal carotid arteries (ICAs) with extensive peripheral calcification (Fig. 1). She was admitted to the neurological ward under a diagnosis of pure motor lacunar stroke. No diabetes mellitus or hyperlipidemia was found by laboratory tests. Electrocardiography showed normal sinus rhythm. A brain magnetic resonance imaging (MRI) study found scattered tiny acute infarcts in the left frontal, temporal, and parietal regions,

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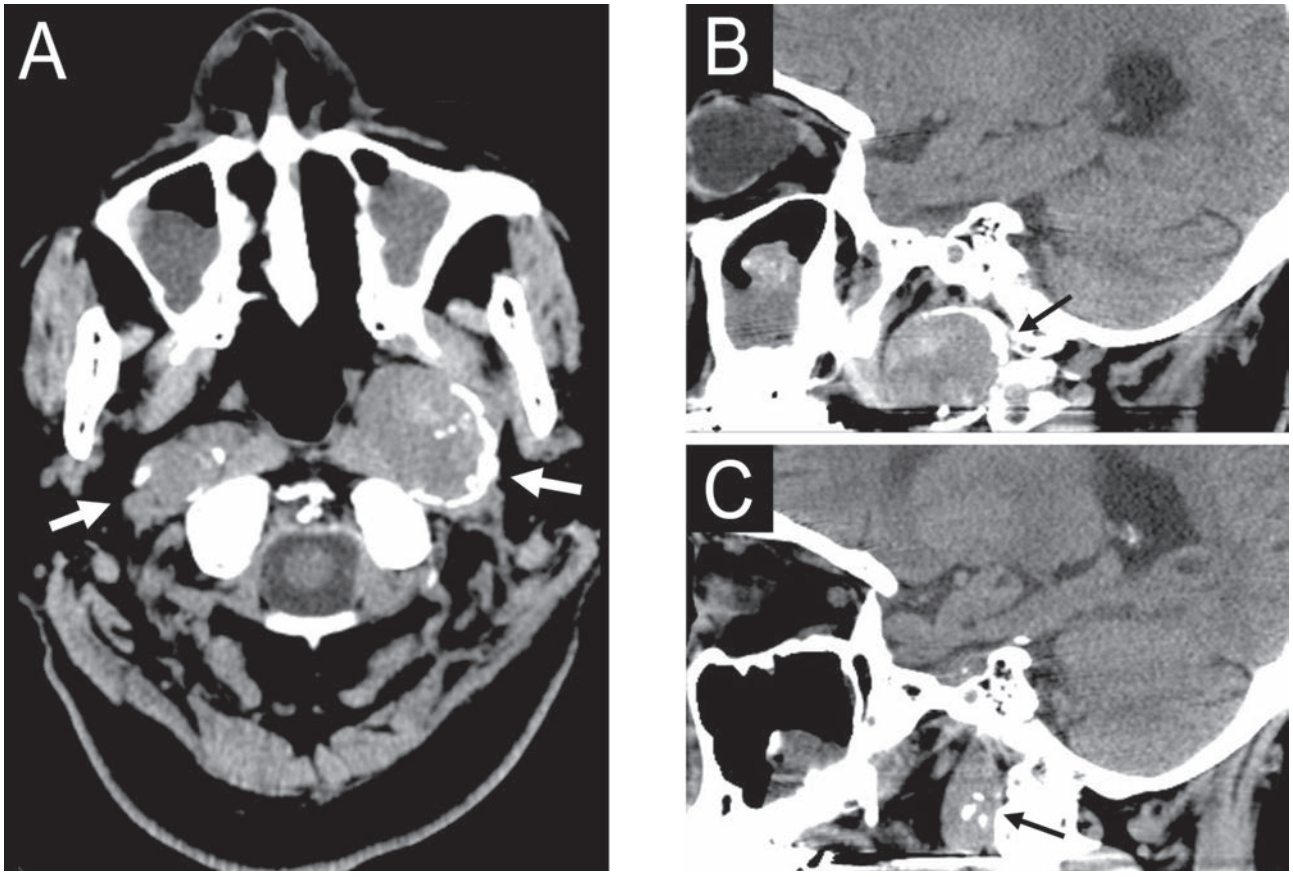

Fig. 1 (A) Axial CT shows well-defined heterogenous soft tissue mass lesions in the bilateral parapharyngeal spaces with irregular peripheral calcification (arrows). The two mass lesions measured 37 mm × 36 mm × 26 mm on the left with ovoid shape on sagittal view (B, small arrow), and 26 mm x 20 mm x 16 mm on the right with elliptic shape on sagittal view (C, small arrow), respectively.

suggestive of embolic stroke in the area of the left middle cerebral artery (Fig. 2). Contrast-enhanced brain MR angiography revealed large aneurysms in bilateral cervical ICAs, with a larger aneurysm on the left side (right: 1.9 × 1.4 cm; left: 2.2 × 1.5 cm) (Fig. 3). Both carotid arteries were tortuous but patent. Color-coded carotid duplex sonography (CCD) showed only a mild degree of atheromatous lesions in the bilateral carotid bifurcations. The Doppler flows of bilateral ICAs were normal. Further echocardiography showed no abnormal intracardiac thrombus or prominent valvular heart disease.

Considering the patient's age as well as the distal location of the ICA aneurysms, we recommended further endovascular treatment rather than surgical management. After discussion with the patient and her family members, conservative medical treatment with aspirin was employed for stroke prevention. The patient recovered gradually and was able to walk with the help of a walker upon discharge. At the 4-month follow-up, the patient had been well without further TIA, stroke, or cardiovascular symptoms.

Discussion

The patient suffered from an embolic stroke resulting in right side hemiparesis. Serial studies showed no evidence of cardiogenic origin of the emboli. Bilateral ECCAs were suspected from the initial brain CT and subsequently confirmed by cervical MR angiography. The cause of stroke was documented as artery-to-artery emboli from a giant aneurysm of the left high cervical ICA.

ECCA is an uncommon cause of acute cerebral infarction. ECCA is typically caused by embolization of a thrombus from the aneurysm[9,10]. The presenting symptoms of ECCA include a painless or painful neck mass, stroke, TIA, aneurysm rupture, visual symptoms,

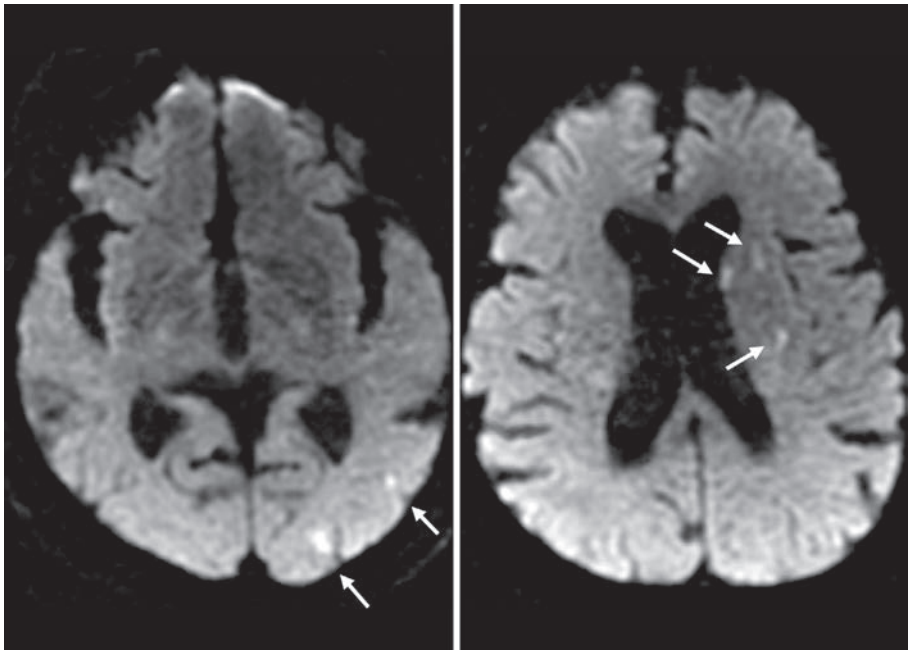

Fig. 2 Axial Diffusion-weighted MR images disclose multiple scattered acute embolic infarcts in the left middle cerebral artery territory (arrows).

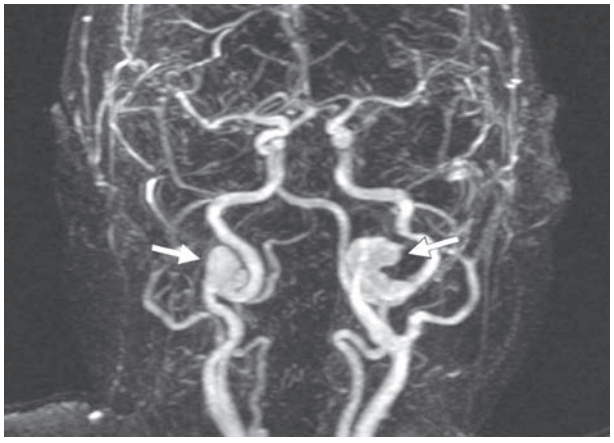

Fig. 3 MR angiography demonstrates large aneurysms of bilateral high cervical internal carotid arteries with irregular contour (arrows).

dysphagia, tongue weakness, and bruit. In a recent review of 141 ECCAs during a 15-year study period by Fankhauser et al, 48% of the ECCAs were asymptomatic and were diagnosed by imaging studies[4]. Diagnosis was most commonly made by MRI or CT[4]. A thorough search for possible ECCAs is usually performed when a patient presents with a pulsatile neck mass or aneurysm rupture. However, ECCAs tend to be incidental findings on cross-sectional imaging or carotid ultrasound examination in most patients presenting with stroke, such as the patient reported here.

Although CCD can detect an aneurysm located in the common carotid artery or proximal portion of the ICA and identify the presence of an intraluminal thrombus[11], ultrasound study has limitations in revealing lesions in the distal ICA. Non-enhanced cross-sectional brain CT is the main diagnostic tool for patients with acute ischemic stroke. An ICA aneurysm showing as a soft tissue-like mass lesion at the skull base may be overlooked on routine brain CT, particularly when the aneurysm is not large. MR angiography is more valuable than CT in differentiating ECCAs from possible tumors, abscesses, or ectatic or kinked vessels. Axial images can also allow assessing the true extent of the aneurysms, thrombus, and residual lumen. CT angiography also provides further information for the diagnosis of ECCAs. In more complex cases, conventional angiography is required for better investigation and visualization of the extent of aneurysm, particularly before endovascular or surgical intervention[12]. In this case, we were not able to find the abnormal bilateral distal ICA aneurysms by CCD. There were no abnormal Doppler flows in bilateral ICAs. Abnormal mass lesions with peripheral

calcification at the skull base revealed by brain CT led to a suspicion of vascular anomaly. Further cervical MR angiography confirmed the final diagnosis.

The most common causes of ECCA are atherosclerosis, trauma, fibromuscular dysplasia, and prior carotid endarterectomy[1,3,13,14]. Younger patients tend to have ECCA from trauma or connective tissue diseases and in older patients, ECCA tends to be associated with atherosclerosis or prior endarterectomy. True aneurysms are more commonly described as atherosclerotic or degenerative; other causes include arterial dissection, fibromuscular dysplasia, infection, and neck irradiation. Pseudoaneurysms may be found as complications of endarterectomy or other traumatic situation. Identifying the exact cause of aneurysm formation can be a challenging task. Although tissue proof was not obtained in our patient, the older age, history of hypertension, and extensive irregular calcification of the aneurysm walls suggest atherosclerosis as the most likely cause of ECCAs.

Once diagnosed, ECCAs pose a clinical challenge for appropriate management. Historically, ECCAs have been successfully treated using open surgery or endovascular procedures. The 5 basic options for open surgical intervention include aneurysm clipping, excision with primary anastomosis, excision with interposition graft, extracranial–intracranial bypass, and carotid ligation[15]. Open surgical interventions have been preferred when patients present with mass effect or bleeding[4]. An endovascular approach has gained acceptance for the treatment of ECCA, particularly for pseudoaneurysms, aneurysms of the distal cervical ICA, or hostile necks due to prior surgery or radiation[4,16]. A retrospective review by Zhou et al indicated that endovascular intervention is an effective alternative to open surgery for the treatment of ECCA, resulting in fewer complications (cranial nerve injuries, wound complications, 30-day stroke rate, and 30-day death rate) as well as shorter recovery time (3.5 vs. 9.4 days)[14]. Although endovascular stenting has gained popularity on account of its lower invasiveness, it is not free of complications. Li et al showed that the procedural success of endovascular stenting was 92.8%, whereas the overall incidence of in-hospital mortality, stroke, and cranial nerve injury was 4.1%, 1.8%, and 0.5%, respectively[16]. Open surgical intervention was more common in patients with symptoms, true aneurysms, and larger aneurysms. Patients with pseudoaneurysms were more likely to undergo endovascular intervention than patients with true aneurysms.

Atherosclerotic ECCAs have been reported to be associated with an exceptionally high stroke rate if treated non-operatively. Three out of 6 (50%) ECCAs resulted in ipsilateral stroke during a mean follow-up period of 6.3 years in Zwolak's series[17]. However, a more recent study involving a larger population by Fankhauser et al suggested that selected patients may also be managed safely using antiplatelets or anticoagulants[4]. At a mean follow-up of 33.9 months, no patient who received non-operative treatment suffered death or major morbidity associated with the aneurysm. Varying causes and severities of ECCAs pose different risks of morbidity and mortality. Patient treatment should be based on the presenting symptoms, underlying cause, location and size of aneurysm, underlying risk factors, and patient consent. The preferred management in our patient was endovascular intervention in the symptomatic left ECCA owing to the older age of the patient and the high cervical ICA location of the aneurysm. However, the final consensus of the patient, her family members, and the physicians was conservative medical treatment with antiplatelet agents. There has been no subsequent cerebral vascular event, such as cerebral infarction or bleeding, since her first episode of stroke and during the 4-month follow-up period.

In conclusion, bilateral ECCAs are uncommon causes of embolic stroke. Careful inspection of the skull base of cross-sectional brain CT is important for screening high cervical ECCAs. Further cervical MR angiography or CT angiography is necessary to confirm the final diagnosis. Management of ECCAs includes open surgery, endovascular intervention, or conservative medical treatment.

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以急性腦梗塞為表現的兩側性顱外頸動脈血管瘤

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摘要

顱外頸動脈血管瘤是少見造成血栓性腦梗塞的原因，顱外頸動脈血管瘤病人的臨床表現是多樣化的。仔細的審視腦部電腦斷層顱底部構造是篩檢顱外頸動脈血管瘤很重要的方式。進一步頸部核磁共振血管攝影或電腦斷層血管攝影是確定診斷的必要檢查。治療的方法包括外科手術、血管內介入性治療及藥物治療。我們報告一例此少見的以急性腦梗塞為表現的兩側性顱外頸動脈血管瘤。造成腦梗塞的原因是來自於高位性內頸動脈巨大血管瘤產生的動脈 - 至 - 動脈的血栓。

關鍵詞：顱外頸動脈血管瘤、內頸動脈瘤、腦梗塞

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Case Report

Mortality Resulting from Inability to Ventilate after Tracheostomy Bleeding: A case report

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Abstract

We report the case of a patient who experienced difficulty breathing after a tracheostomy and was checked for hemorrhaging. The patient eventually died.

An 81-year-old male patient received a small bowel resection and adhesion lysis due to jejunal diverticulitis and adhesion ileus. Two weeks later, a tracheostomy was scheduled because mechanical ventilation could not be discontinued. Hemorrhaging from the tracheostomy site occurred 3 days later. When patient was sent to the operating room, a large number of blood clots were noted over his neck and bed. Ventilation could not be achieved. A suction catheter was inserted into the tracheostomy tube, and a small amount of fresh blood was suctioned out. Ventilation resumed, and the tracheostomy tube was changed to an oral endotracheal tube. After insertion of the endotracheal tube, we were unable to reestablish ventilation. This time, asystole soon developed, and cardiac massage was started. Due to the inability to restart ventilation, the patient eventually died.

Prompt diagnosis and management are important in the condition of inability to ventilate. Bronchoscopy is helpful in the diagnosis and in the removal of the obstructed lesions.

Key words: tracheostomy bleeding, airway total obstruction

Background

Tracheostomy is a procedure commonly performed on patients requiring prolonged mechanical ventilation. The advantages of this procedure are the ease of secretion removal and a reduction in the breathing workload followed by the ability to wean the patient off the ventilator. Life-threatening complications such as tracheal tube obstruction and hemorrhaging can occur[1,2].

We present the case of an elderly patient who experienced bleeding from the tracheostomy site, which resulted in life-threatening sequelae due to the inability to ventilate. The patient eventually died.

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Case Report

An 81-year-old male, height 168 cm and weight 69 kg, with hypertension, chronic obstructive pulmonary disease, and chronic kidney disease had been admitted 16 days earlier due to perforated jejunal diverticulitis, adhesion ileus, and peritonitis. Resection of the small intestine and lysis of adhesion had been performed soon after admission. Attempting to wean the patient off the ventilator failed 13 days after the operation. A tracheostomy was performed, but 3 days later, tracheostomy bleeding was observed. Wound compression was tried for 4 hours after the bleeding was noticed. The cuff of tracheostomy tube was not hyperinflated. Unfortunately, 4 hours later, the ICU doctor found that wound compression did not work and informed the surgeon. An hour later, the surgeon arrived and decided to check the

bleeding in the operating room. The patient was sent to the operating room nursing station in 30 minutes, and the anesthesiologist who visited the patient there noted a large number of blood clots around the patient's neck and on the bed. The patient could breathe spontaneously with the aid of manual ventilation with oxygen. His Glasgow Coma Scale score was E4M6Vt. The breathing rate was approximately 20–30/min. Bilateral breathing sounds were coarse and weak. The patient was sent to the operating room, and pulse oximetry revealed only 90% when breathing 100% oxygen. Three minutes later, his lungs could not be ventilated because airway resistance was very high. We inserted a suction catheter into the tracheostomy tube and suctioned a small amount of blood, after which we reestablished ventilation, but the peak airway pressure was still at 45 cmH_2O and end-tidal CO_2 was only 15 mmHg. At that point, the patient's blood pressure was 106/72 mmHg and pulse rate was 65/min. Pulse oximetry revealed 91%. The surgeon then requested changing the tracheostomy tube to an oral endotracheal tube in order to have a larger working field. We removed the tracheostomy tube and re-intubated orally without any sedation due to the patient's critical condition. However, the ventilation still could not be reestablished. End-tidal CO_2 was only 2 mmHg. Pulse oximetry soon dropped to zero, and several seconds later, the heart rate also decreased from 65 to zero. Cardiac massage was started. Insertion of suction catheter was reattempted, but it was unsuccessful. The surgeon removed some blood clots around the tracheostoma but in vain. Due to the inability to ventilate, the patient died.

Discussion

Complications from tracheostomies may occur immediately after insertion of the tracheostomy tube or may occur at a later date. One immediate complication is usually hemorrhage from transection or avulsion of aberrantly situated vessels such as an elevated right subclavian artery or a heavily vascularized thyroid gland[3]. Later complications may include mucus plugging with life-threatening occlusion of the tracheostomy tube and/or trachea. Delayed hemorrhaging is an uncommon event and is usually due to tracheovascular fistula formation[4,5]. It occurs in only 0.1%–1% of cases with the innominate artery being the most vulnerable site[4,6]. The vessels that may also be involved are the right and left subclavian arteries and the aorta[7,8].

In most cases, tracheostomy bleeding usually can be controlled with a pressure dressing or suture ligation[9]. A massive bleed needs immediate management, which involves resuscitation, hyperinflation of the cuff of the endotracheal tube, or digital compression of the artery anteriorly against the sternum through the tracheostomy wound[10]. Hyperinflation of the cuff could not only exert pressure on the bleeding source but also prevent blood flow into the lower trachea and bronchi. If bleeding cannot be controlled by the above maneuvers, surgical intervention is needed. The tracheal tube must not be removed during bleeding from the tracheostomy site until surgery. It had been reported that one case died due to large amount of blood flow into the trachea because the tracheal tube was removed during bleeding from the tracheostomy site[11]. Angiography may or may not be helpful and it can dangerously delay definitive treatment[12].

Tracheal tube obstruction has been reported in 2.5% of surgical tracheostomies[13] and up to 3.5% of percutaneous tracheostomies[14]. Obstruction may result from blood clots, mucus casts, granulation tissue, foreign bodies, cuff herniation, or tube malposition. The inability to ventilate in the case of our patient, when bleeding from the tracheostomy site occurred, indicated the possibility of airway obstruction from blood clots. The blood clots could be tenacious and may not be easily removed by suctioning.

After we re-intubated orally, the ventilation could not be reestablished. We did not use a flexible bronchoscope to inspect the subglottic region prior to changing endotracheal tube. Neglecting this procedure could be serious. Because there was the possibility of blood clots in the subglottic region; these blood clots might be pushed into the lower trachea or bronchus by tracheal intubation, resulting in complete airway obstruction[15]. This may explain why we could not reestablish ventilation again after re-intubation. Changing to the endotracheal tube from a tracheostomy tube should be considered more seriously especially when airway obstruction had been suspected previously.

The obstruction may also act by a ball-valve effect[16]. This could explain why insertion of the suction catheter into the endotracheal tube the

second time did not reopen the airway. Perhaps when the catheter passed through the blood clot, it penetrated the blood clot but could not create a permanent opening, and the clot formed a valve; this caused a significant amount of trapped air, which could have resulted in a tension pneumothorax. A hemothorax could also happen during bleeding from the tracheostomy site if blood flowed into the pleural cavities[16]. Definite diagnoses of pneumothorax or hemothorax needs a chest x-ray[16]. This patient was deteriorating very rapidly, so taking a chest x-ray was impossible. Emergency needle decompression still could have been be tried but was not performed, as the patient was near death. Using a fiberoptic bronchoscope, if possible, is highly suggested for such conditions to establish diagnosis and to remove any obstructed lesions[15]. A large amount of fresh blood might be present in the airway and would obscure the visual field making the effort futile.

In conclusion, any hemorrhaging condition after tracheostomy might be a serious, potentially life-threatening problem. Airway obstruction caused blood clots should be considered in conditions when patients present with respiratory distress. Bronchospasm, sputum retention, pneumothorax, and hemothorax can also mimic the same clinical scenarios. Prompt diagnosis and management are crucial because major sequelae might develop or death may occur. In such circumstances, airway evaluation with a bronchoscope would be helpful in making the diagnosis and removing obstructed lesions. A chest x-ray also could aid the diagnosis if the patient's condition did not deteriorate too rapidly.

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氣切術後流血造成無法通氣而死亡：個案報告

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摘要

我們報告一個病例，本預進行氣管切開手術術後出血之止血，卻因為通氣困難最終導致死亡。

這是一位 81 歲病患，因為空腸憩室炎與沾黏性腸阻塞，而接受小腸切除與沾黏撥離手術，兩周後，病患仍無法脫離呼吸器輔助，於是安排了氣管切開手術，又過了三天後，氣管切開的地方開始出血，當病患送至手術室時，發現在病患的頸部與床上有大量的血塊，沒多久之後變成完全無法通氣，我們將抽吸管放入氣切管內，只抽吸出少量的新鮮血液，但是通氣又恢復了，於是開始換插經口的氣管內管，然而換管完成後，又變成無法通氣，這次心臟很快的就停止跳動了，我們開始體外心臟按壓，因為通氣一直無法恢復，病患最終還是死亡。

呼吸道無法通氣時診斷與處置是很重要的，軟式支氣管纖維鏡除了可以幫助診斷無法通氣的原因外，若有阻塞物亦可嘗試直接移除。

關鍵詞：氣切術後流血、呼吸道完全阻塞

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Pathology Page

A Malignant Peripheral Nerve Sheath Tumor on the Right Elbow

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Abstract

A 37-year-old man presented with an extremely large tumor (15 × 12 × 9 cm) over his right elbow that had developed over a period of 6 months. A physical examination revealed multiple tumors in the bilateral limbs, axilla, and back that had developed over the previous 10 years. Computed tomography chest scans revealed multiple nodules in both lungs, indicating metastasis. The patient underwent wide excision of the tumor on his right elbow, which was diagnosed as a stage IV malignant peripheral nerve sheath tumor (MPNST). The patient received radiotherapy for local control of the elbow tumor and chemotherapy for the multiple lung metastatic lesions. The patient died 7 months after surgery due to multiple lung metastasis and respiratory failure. A MPNST is highly aggressive and approximately 50% of cases are associated with neurofibromatosis type 1. MPNST cases have an overall 5-year survival rate of around 34%.

Key words: elbow, malignant peripheral nerve sheath tumor, respiratory failure

Pathology Page

A 37-year-old man with a 3-year history of neurofibromatosis type I characterized by multiple neurofibromas over the extremities, chest, and back, and a Lisch nodule of the iris presented with an extremely large tumor (15 × 12 × 9 cm) over his right medial elbow region that developed over a period of 6 months. He also had multiple subcutaneous tumors of the bilateral limbs, axilla, and back that had developed over a period of 10 years. Magnetic resonance imaging on admission (Figure 1, left panel) revealed an extremely large encapsulated and lobulated tumor over the right elbow joint. Computer tomography (CT) of the chest revealed multiple nodules in both lungs measuring up to 7.7 cm in diameter, indicating metastasis.

The patient underwent wide excision of the right elbow tumor and the specimen was sent for pathology examination. A gross examination showed that the tumor (15 × 12 cm) was solid, well encapsulated, and gray in color with focal necrosis. Microscopically, the tumor cells were arranged in sheet-like patterns combined with spindle-like cells (Figure 1, right panel), showing hyperchromatic nuclei, necrosis, and frequent mitoses. Immunohistochemical analysis with the nerve marker S100 demonstrated focal cytoplasmic staining of the tumor cells (Figure 2, panel A). The malignant muscle tumors were negative on staining of the smooth muscle actin, Myo D1, and desmin. Synovial sarcoma was ruled out by negative staining for cytokeratin and epithelial membrane antigen (Figure 2, panels B and C, respectively). Malignant melanoma was ruled out by staining for human melanoma black 45 (Figure 2, panel D). The final diagnosis was a clinical stage IV malignant peripheral nerve sheath tumor (MPNST, malignant schwannoma). The patient received radiotherapy for the local control of the elbow tumor and chemotherapy (doxorubicin and ifosfamide) for multiple metastatic lung lesions. The patient died 7 months after surgery due to multiple

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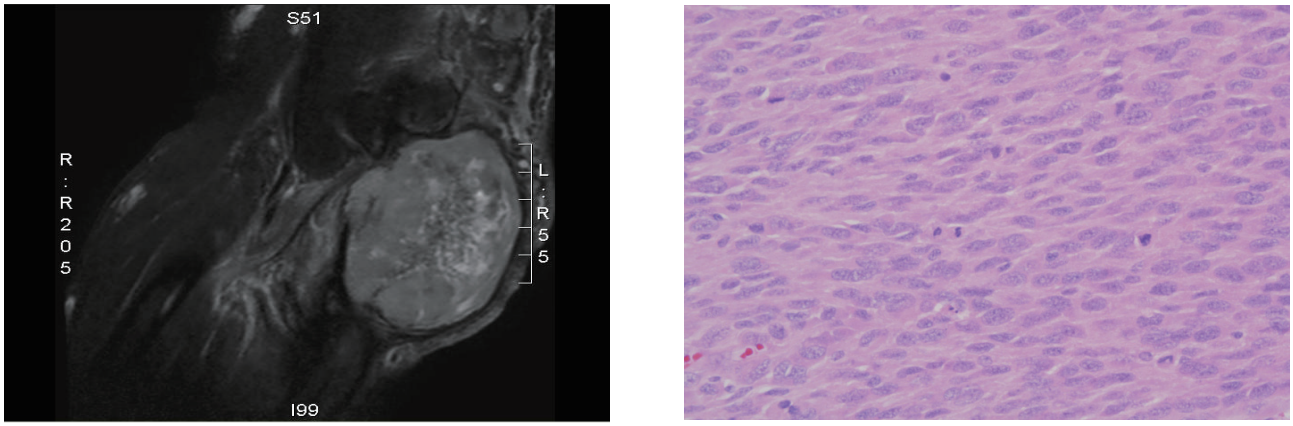

Fig. 1 MRI findings of MPNST (15 x 12 x 9 cm) in right elbow (left panel), and histopathologic features of spindle tumor cells (right panel, Hematoxylin-Eosin stain, 400x). Arrows indicate area of mitoses.

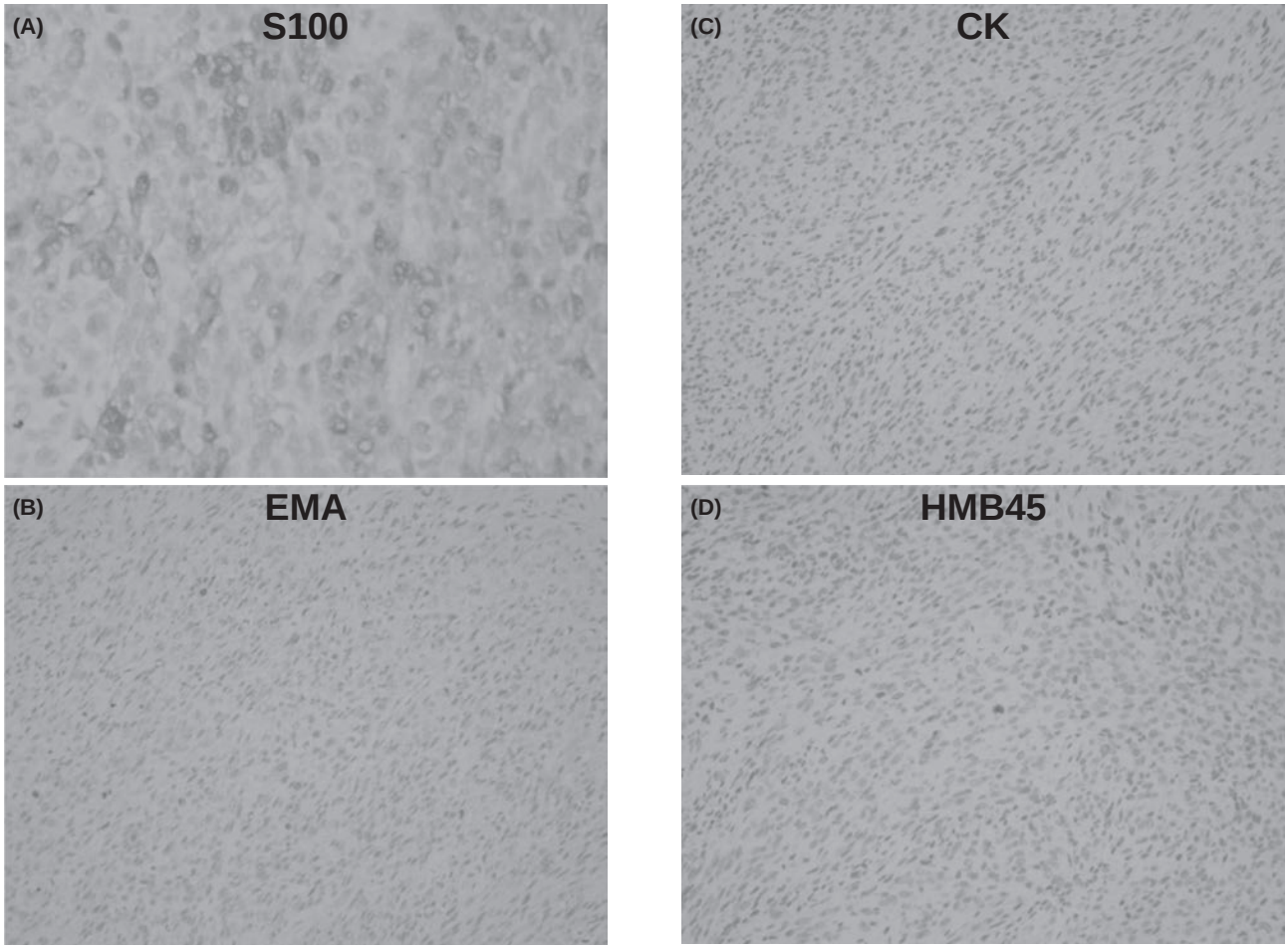

Fig. 2 Immunohistochemistry stains of tumor show positive staining to S100 (panel A, 400x); and negative staining to EMA (panel B, 400x), CK (panel C, 400x), and HMB45 (panel D, 400x).

lung metastasis and respiratory failure.

A MPNST is a rare type of sarcoma with one of the poorest prognoses of all soft tissue sarcomas. Such growths arise from the peripheral nerve sheath and approximately 50% of cases are associated with neurofibromatosis type 1 (NF1). The median age of patients associated with NF1 is around 28–36 years, which is younger than that of sporadic cases (40–44 years). The histologic subtypes of MPNSTs include epithelioid and glandular types. MPNST cases have an overall 5-year survival rate of around 34%.

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右手肘惡性周邊神經包被瘤：病例病理報告

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摘要

一位 37 歲男性因右手肘腫瘤（15 x 12 x 9 公分）已生長六個月而求診，理學檢查發現病患同時有雙側肢體、腋下、及背部多顆腫瘤，已有十年的病史。入院胸部電腦斷層檢查顯示二側肺葉有多顆腫瘤，比較像是轉移性腫瘤。病患接受右手肘大範圍腫瘤切除術後，病理診斷為惡性周邊神經包被瘤，臨床分期為第四期。患者於術後接受手肘放射線治療以防止局部復發，以及化療以控制肺部轉移性腫瘤。病患於七個月後因肺部轉移性腫瘤及呼吸衰竭而過世。惡性周邊神經包被瘤是一高侵犯性腫瘤，百分之五十的瘤伴隨第一型神經纖維瘤，平均五年的存活率約百分之三十四。

關鍵詞：手肘、惡性周邊神經包被瘤、呼吸衰竭

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Tungs' Medical Journal 9 (2015) 66-69

Image

Successful Medical Treatment of Pulmonary–Gastric Fistula

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Abstract

Pulmonary–gastric fistulae are extremely rare and can occur secondary to infection or malignancy invading through the lung into the pleural space, through the diaphragm, and into the stomach. A 40-year-old man presented to our facility with left lower lobe lung abscess and thoracic empyema; further investigation revealed a pulmonary–gastric fistula. There was no history of trauma or operation. Upper gastrointestinal endoscopy demonstrated no gastric malignancy or perforated ulcer. Spontaneous closure of the fistula was observed in our patient after thorough conservative management of infection with appropriate antibiotic and parenteral nutrition. Upper gastrointestinal series revealed a healed defect in the gastric fundus without leakage of dye.

Key words: pulmonary–gastric fistulae, lung abscess, thoracic empyema, parenteral nutrition

A 40-year-old man presented to the pulmonary outpatient department with fever and dyspnea on exertion for four days. On physical examination, he was consciously clear and oriented with no signs of cardiorespiratory distress. His temperature was 39°C, heart rate 102 beats/min, respiratory rate 20 breaths/min, and blood pressure 159/83 mmHg. Diagnosis on admission was left lower lobe pneumonia with pleural effusion. Standing chest X-ray demonstrated a lesion with air fluid level over the retrocardiac area with pleurisy (Panel A). Empiric antibiotic therapy was initiated. Left pleural fluid analysis revealed empyema. Lung abscess was suspected, and chest CT demonstrated pneumonia with pleural effusion over the left lower lobe and gastric distension (Panel A). The patient was placed on NPO status, and nasogastric tube decompression was initiated. Pleural fluid culture revealed *Streptococcus viridans*. The patient symptomatically improved while on NPO and nasogastric tube decompression. When oral feeding was resumed, he developed a productive cough with purulent, foul-smelling sputum, and high-grade fever. Therefore, a fistula of the esophagus or stomach was suspected. Esophagography and non-contrast enhanced abdominal CT demonstrated intact esophagus without obstruction or perforation. A defect in the superior aspect of gastric fundus with erosion into the left diaphragm was observed (Panel B). There were no septic or peritoneal signs and medical treatment was done. Total parenteral nutrition was initiated. Later, a nasojejunal tube was inserted to bypass the gastric area. Three weeks after placement of the nasojejunal tube, endoscopy revealed localized crowding of gastric folds at the fundus (near high body, posterior wall), no apparent fistula or holes were observed on separation of the folds by biopsy forceps (healed fistula, lymphoma was unlikely due to soft consistency of the folds). Upper gastrointestinal series revealed a healed defect in the gastric fundus without leakage of gastrografin, total non-opacification of the gastro–diaphragmatic–pulmonary fistula, and left lower lobe pneumonia (Panel C). Feeding by nasojejunal tube was tolerated, and the patient was shifted to a soft diet. After conservative management, the patient's condition improved, and he was discharged after 33 days in the hospital. Follow-up standing chest X-ray demonstrated resolution of air

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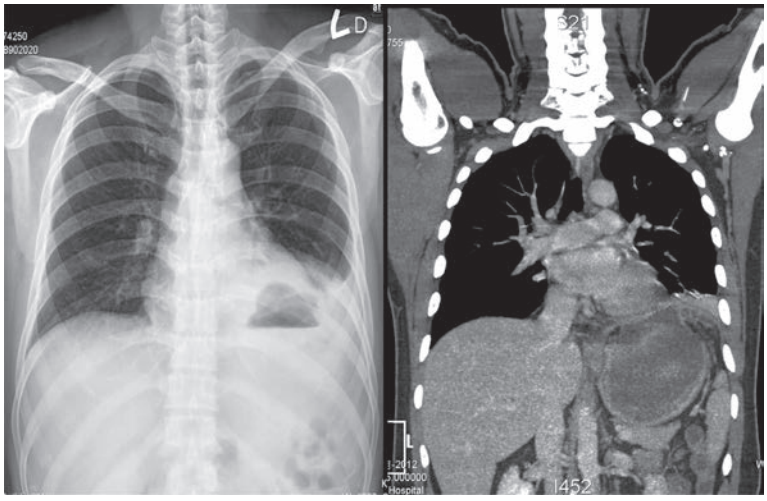
fluid level over the left lower lobe and left pleural effusion (Panel D).

Pulmonary–gastric fistulae can occur secondary to penetrating or blunt trauma, pneumonia, lung abscess, empyema, gastrointestinal or pulmonary malignancy with local invasion, foreign body aspiration, or pulmonary surgery. Fistulous connection between the lung and stomach is extremely rare in patients who have not undergone previous gastric or esophageal surgery[1]. Because our patient displayed no direct evidence of blunt trauma; foreign body ingestion; gastric perforation; malignancy, such as lymphoma or previous pulmonary; esophageal; or gastric surgery, the potential cause can only be speculated. Left lower lobe pneumonia or lung abscess with direct invasion of the diaphragm to the gastric fundus is more likely. Our patient had no history of epigastric pain, nausea, vomiting, or weight loss to provide clues to upper gastrointestinal pathology. Pulmonary–gastric fistula was suspected because of no improvement in lung infection with antibiotic treatment, and symptoms worsened when the nasogastric tube was removed and oral feeding was initiated.

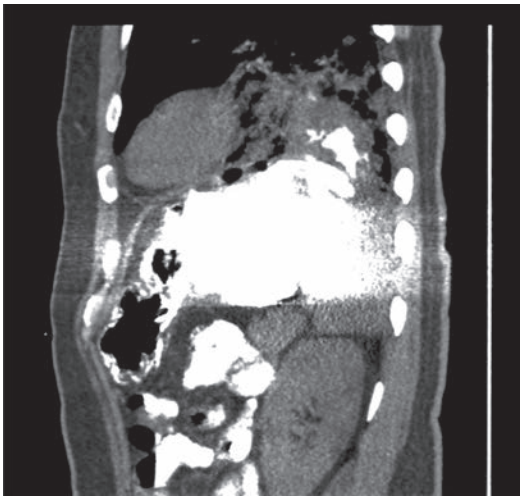
The presence of pulmonary–gastric fistula in a patient who has not undergone previous pulmonary, esophageal, or gastric surgery should be suspected when pneumonia or a lung abscess is unresponsive to conventional treatment. Other diagnostic indicators of pulmonary–gastric fistula are presence of bile or food particles in the sputum or pleural fluid. Bronchoscopy may reveal bilious gastric secretions and/or food particles in the airway; however, the fistulous tract is more readily visible on flexible esophagogastroscopy[2].

Therapeutic strategies are based on the severity

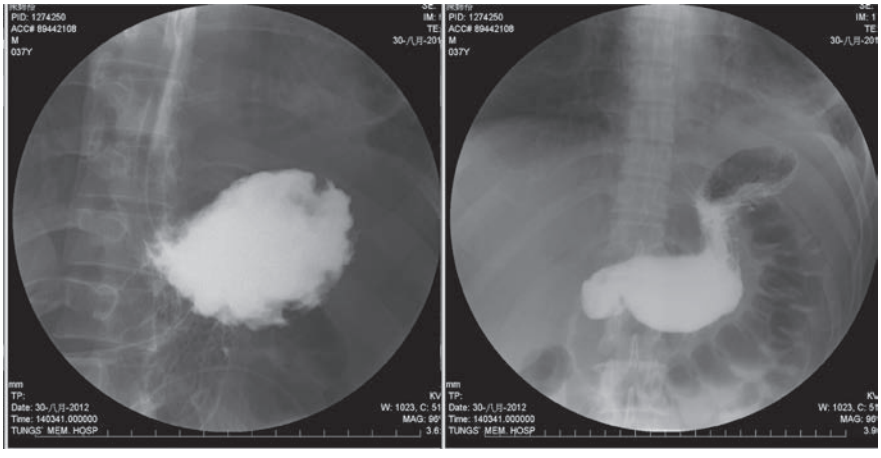
Panel A



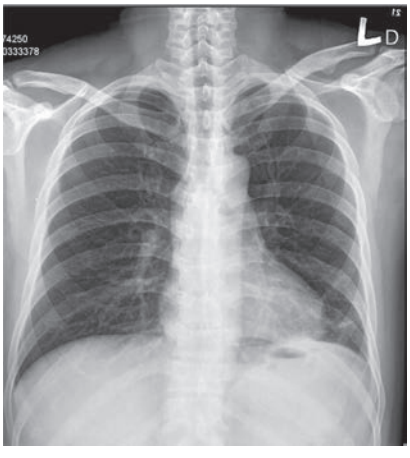
Panel B



Panel C



Panel D



of the patient's clinical condition. After placing our patient on NPO status; nasogastric tube decompression; and broad spectrum antibiotic, his toxic signs, such as fever and cough with purulent sputum, subsided. Our patient did not show signs of peritonitis, which would have required emergent surgical intervention. Probable risks of conservative treatment are sepsis with shock, peritonitis, and acute respiratory failure. If the patient reveals no clinical improvement after medical treatment, surgical intervention is necessary. Follow-up upper gastrointestinal endoscopy revealed no apparent fistula or holes in the fundal area, indicating that our conservative treatment was successful. Upper gastrointestinal series demonstrated a healed defect in the gastric fundus and total non-opacification of the gastro–diaphragmatic–pulmonary fistula.

A literature review suggests that appropriate antibiotics should be initiated, and a bronchial blocker bronchoscopically placed in the offending segmental bronchus can be used to stabilize the patient until surgical correction is possible. Surgical treatment of pulmonary–gastric fistulae comprises resection of the fistulous tract, the abnormal area of pulmonary parenchyma and/or bronchus, and the abnormal portion of the gastrointestinal tract with interposition of a vascularized, pedicled tissue flap (muscle flap, omentum, and pericardial fat pad)[2]. Our patient who had a small fistula between a bronchus and the gastrointestinal tract without sepsis or peritonitis responded well to medical treatment.

References

1. Osaki T, Matsuura H. Thoracic empyema and lung abscess resulting from gastropulmonary fistula as a complication of esophagectomy. Ann Thorac Cardiovasc Surg 2008;14: 172-174.
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保守治療成功治癒肺胃瘻管

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摘 要

肺胃瘻管是極為罕見的疾病。這個疾病是因為肺部感染或惡性腫瘤侵入到肋膜，穿過橫隔膜到胃部。我們提出一例 40 歲男性病患因發燒及膿痰，被診斷左側肺炎併肺膿瘍與膿胸，進一步檢查發現肺胃瘻管。病患無外傷或胸腔、食道、胃部手術病史。上消化道內視鏡無胃癌，潰瘍穿孔，也無腹內敗血症。使用保守治療例如 : 禁食，抗生素，靜脈營養。病患臨床症狀改善，成功治癒。上消化道攝影也顯示胃底部缺損已癒合，無顯影劑滲漏。

關鍵詞：肺胃瘻管、肺膿瘍、膿胸、靜脈營養

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(2) Yang KTA, Chen HD: A semi-automated method for edge detection in the evaluation of left ventricular function using ECG-gated single-photon emission tomography. Eur J Nucl Med 1994;21:1206-11.

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(2) Levinsky NG: Fluid and electrolytes. In: Thorn GW, Adams RD, Braunwald E, lsselbacher K, Petersdprf RG eds. Harrison’s Principles of Internal Medicine, 8th ed. New York: Mcgraw-Hill, 1977:364-75.

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