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What Lies Beneath: Sub-diaphragmatic causes of subcutaneous emphysema – A Review

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ABSTRACT

Air in the subcutaneous tissues, commonly described as subcutaneous emphysema, creates a distinctive clinical sign on palpation. Usually, this results from an air leak from the airways or lungs forcing air into the mediastinum and in between soft tissue planes. This review aims to draw attention to bowel perforations below the diaphragm as an alternative origin of subcutaneous emphysema. Reference to numerous case reports illustrate a variety of presentations and causes. Delayed recognition and treatment of bowel perforation can have serious, potentially life-threatening, consequences. Patients presenting with subcutaneous emphysema without an obvious intra-thoracic cause should lead the clinician to consider acute intra-abdominal pathology, such as bowel perforation, especially after endoscopic procedures. Early recognition of this phenomenon can be life saving.

Introduction

Subcutaneous emphysema (SCE), sometimes known as "surgical emphysema", describes gas within the subcutaneous tissue planes of the body. It is a rare but striking clinical sign characterised by swelling and crepitus under the skin. As subcutaneous emphysema is usually found in the neck and thorax, acute thoracic or cervical pathology is often suspected. Indeed, it is most commonly caused by air escaping from disrupted pleurae or airways into the mediastinum, such as in acute pneumothorax or thoracic trauma[1-3]. Similarly, it is somewhat expected in cardiothoracic surgery or after thoracic interventions, such as the insertion of intercostal chest drains[4]. In these instances, the origin of the air leak is predictable and anticipated so it can normally be managed during the immediate post-operative period. Air from thoracic organs is normally not contaminated so does not typically pose a threat of sepsis and will dissipate with minimal consequences to the patient. However, another potential source of air in the mediastinum is from perforation of the oesophagus, often rapidly leading to subcutaneous emphysema[5]. Oesophageal perforations may occur after violent vomiting (Boerhave's syndrome), forced Valsalva manoeuvre or during upper gastrointestinal endoscopy. Hence, the recent medical history may suggest oesophageal perforation as a cause of subcutaneous emphysema, which can then be confirmed by imaging, such as a water soluble contrast study.

What is less known, however, is that visceral air from sub-diaphragmatic gastrointestinal perforations can track into subcutaneous tissue planes from the abdomen, probably entering the mediastinum via the diaphragmatic hiatus and causing subcutaneous emphysema[6,7]. Retroperitoneal perforations can be particularly difficult to identify as they may not initially cause the classic clinical signs of intraperitoneal visceral perforations, such as acute peritonitis, leucocytosis or pyrexia[8]. Instead, they may first present non-specific pain, sometimes radiating to the back, with subcutaneous emphysema and subsequently "sepsis of unknown origin"[9]. Further diagnostic confusion can occur because air from within the abdomen may also move to parts of the body anatomically distant from its origin, such as the neck[10] and even the orbit[11]. One recent case report even describes a patient presenting to otolaryngologists (ear, nose and throat surgeons) with a hoarse voice secondary to disseminated subcutaneous air arising from a perforated sigmoid colon[12].

The anatomical basis for subcutaneous air movements from within the abdomen is not always clearly understood as it does not always pass into the upper body via the mediastinum. For instance, several case reports have described bowel perforations causing subcutaneous emphysema in the scrotum[13-15] and lower limbs[16-18]. In these cases, the air may have passed out of the abdominal cavity via the deep inguinal ring in a similar way to an inguinal hernia. It could also be envisioned that if the patient is mainly supine following an intestinal perforation, the gas could track equally inferiorly as superiorly, depending on the path of least resistance. Unsurprisingly, such atypical presentations can cause diagnostic confusion and delays in treatment[19]. At its most serious, such delayed diagnoses of bowel perforation can result in life-threatening consequences, such as advanced sepsis or tension pneumothorax[20, 21].

Radiological diagnosis of subcutaneous emphysema

In most developed countries, there is less reliance on simple radiographs but an appropriately exposed radiograph can reveal subcutaneous emphysema clearly (see Figure 1) and may suggest a cause.

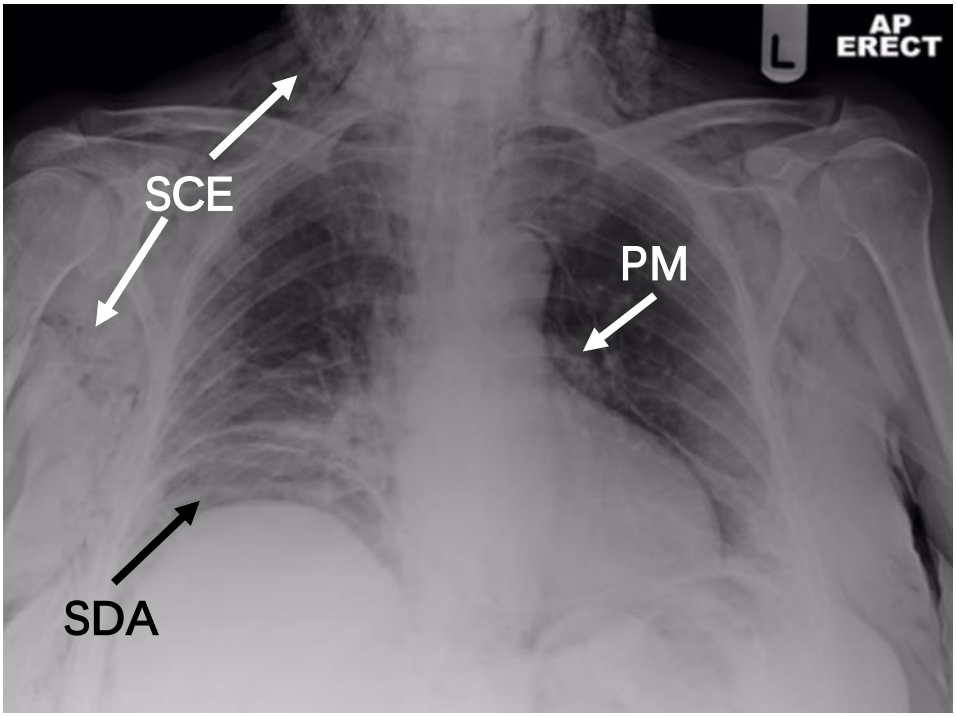

Figure 1. Chest radiograph demonstrating subcutaneous emphysema (SCE), pneumomediastinum (PM) and subdiaphragmatic air (SDA).

However, computed tomography (CT) is becoming a much more common early diagnostic tool as the rapidity, yield and specificity of radiological information is significantly greater. In the case of subcutaneous emphysema, CT imaging may reveal its anatomical site and extent, it may still be extremely challenging for the reporting radiologist to identify its origin in the absence of relevant clinical information or intra-abdominal stigmata. Indeed, subcutaneous emphysema of the head, neck and thorax may not even prompt the clinician to request inclusion of the abdomen. Occasionally, however, the information provided by an adequate CT scan is invaluable and can lead to a rapid diagnosis (see Figure 2). This review aims to encourage clinicians to include sub-diaphragmatic causes of acute subcutaneous emphysema within their differential diagnosis.

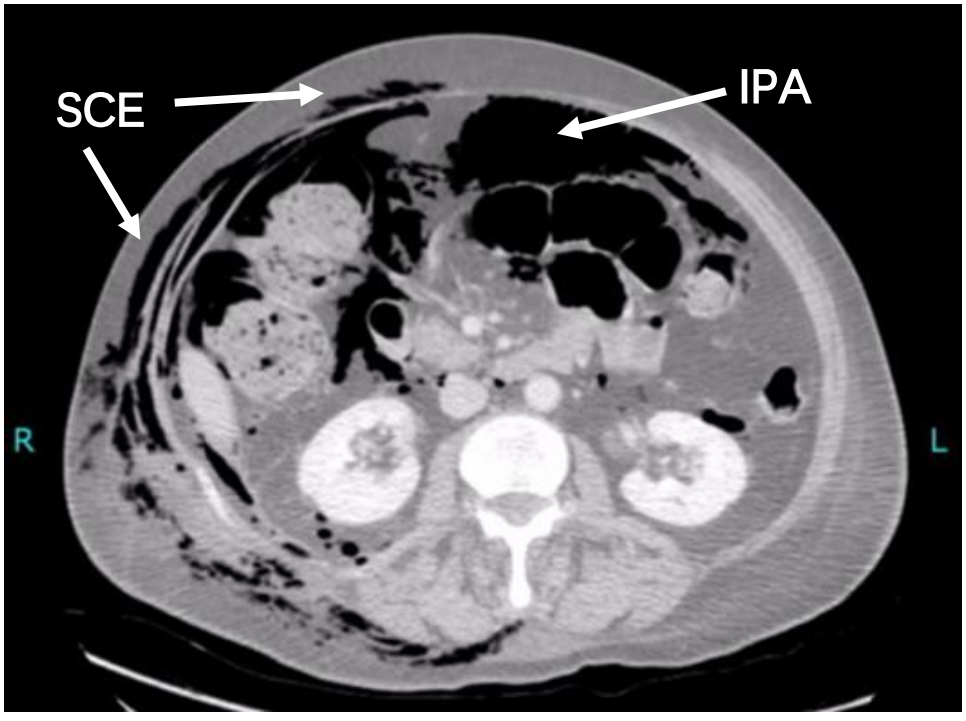

Figure 2. Saggital computed tomography (CT) image showing subcutaneous emphysema (SCE) and intra-peritoneal air (IPA) secondary to colonic perforation.

Spontaneous bowel perforation and subcutaneous emphysema

Spontaneous bowel perforations can result from trauma (accidental or iatrogenic), ischaemia, diverticulae or carcinomas[18,22,23]. Although it is a rare phenomenon, subcutaneous emphysema develops more commonly from colonic than small bowel perforations. This is probably due to the greater volumes of gas within the colon, high intra-luminal pressures and a significant proportion of the colon lying in a retroperitoneal position. Left sided colonic perforations account for the vast majority of case reports of subcutaneous emphysema secondary to perforation, especially involving the sigmoid[22,24-26]. Rarely, however, subcutaneous emphysema has been reported from small bowel perforations, such as the description of a 14 year old patient in whom a retained peritoneal shunt catheter led to erosive perforations of the jejunum[27]. Diop et al. report the case of a patient presenting with subcutaneous emphysema associated with a liver abscess, both of which were found to be secondary to perforation of the adjacent transverse colon[28]. Even rectal perforations can cause this phenomenon, where it is more likely to occur outside the pelvis[29,30].

Spontaneous sigmoid colon perforations are commonly secondary to diverticular disease, especially in the elderly, as diverticulae are natural weak points along the colon wall and may be dilated by the pressure of constipated stool and excessive flatus. As a result, it is unsurprising that there are numerous reported cases of subcutaneous emphysema arising from sigmoid diverticulosis[23,31-33]. Inflammation of the bowel can lead to weakness and localised ischaemia of the bowel wall. This is reflected in case reports of subcutaneous emphysema arising in patients with inflammatory conditions, such as ulcerative colitis[34] and appendicitis[35, 36].

Colonoscopy and Subcutaneous emphysema

Colonoscopy is an endoscopic procedure performed for screening, diagnostic or therapeutic purposes. It is known that trauma from the endoscope or from polypectomy can lead to bowel perforation, especially in the presence of diverticular disease, strictures or polyps/tumours[37,38]. Bowel perforations during colonoscopy occur in 0.1% - 0.2% of cases, rising to 1% - 2% where polypectomy is performed, especially in the elderly or where large polyps are involved. Such perforations are notoriously under-recognised at the time and may take hours or even days to be diagnosed as most colonoscopies are performed as day cases and it is often hard to distinguish between the usual discomfort of the endoscopy and the symptoms from a small perforation. Perforations causing subcutaneous emphysema have been reported secondary to endoscopic balloon dilatation of colonic strictures[39], endoscopic mucosal resection of polyps[40] and polypectomy[20,37]. However, this can also occur from diagnostic colonoscopies even without interventions[38,41-46].

If one considers that as much as 8 litres of carbon dioxide are insufflated in a typical colonoscopy [44], ongoing intracolonic insufflation following an unrecognised perforation can lead to very large volumes of gas being pumped into the peritoneum or retroperitoneal space [41]. This can lead to life-threatening complications, such as tension pneumothorax [20, 21]. There has been an increase in performing colonoscopy under general anaesthesia over the past 25 years, from approximately 8% to 30% in some centres. However, while this eliminates the pain and discomfort of colonoscopy for the patient, it increases risks and can further delay the recognition of visceral perforation, especially as anaesthesiologists are more likely to consider subcutaneous emphysema under general anaesthesia as arising from ventilator-induced lung trauma.

Endoscopic retrograde cholangio-pancreatography and subcutaneous emphysema

Endoscopic retrograde cholangio-pancreatography (ERCP) is a highly skilled endoscopic procedure used to visualise the duodenal ampulla and the

biliary tree for diagnostic and therapeutic purposes. The technique is used to biopsy lesions of the bile duct or ampulla and to retrieve or release gallstones in the common bile duct. Strictures of the peri-ampullary ducts may also be dilated or widened by sphincterotomy, with or without deploying a biliary stent[47].

Although there is a risk of duodenal perforation during ERCP, it occurs in less than 1% of procedures and often retroperitoneal. As with other retroperitoneal perforations, clinical signs may be subtle but may include the development of subcutaneous emphysema. Although it is a rare complication following ERCP, case reports suggest that subcutaneous emphysema in these patients typically tracks to the thorax, neck or face and may be sufficiently extensive to cause airway compromise [48] or pneumothorax [49]. It has even been reported, bizarrely, in the lower extremities after ERCP, but the anatomical mechanism of this is obscure [50]. Other complications of ERCP, such as pancreatitis, may also be present and may complicate the diagnosis [51]. Nevertheless, it would appear that the development of subcutaneous emphysema following ERCP, as with other endoscopic procedures, should be considered diagnostic of a perforation.

Management

Rapid diagnosis of a bowel perforation is key to successful treatment. Once a visceral perforation has been diagnosed, the typical management involves proceeding to emergency laparoscopy or laparotomy with washout of intraperitoneal contamination and drainage. There is an overall mortality of 7% rising to as much as 19% within the first 28 days if faecal peritonitis is also present [52]. Up to 30% of these patients require a permanent colostomy but early diagnosis reduces the risk of a stoma by approximately 70%. These data mainly come from the commonest causes of perforation, especially perforated diverticular disease or stercoral perforations, where spillage of faeces and intra-colonic bacteria rapidly lead to intra-abdominal sepsis. Importantly, however, most reports of subcutaneous emphysema in association with colonic perforations involve leakage of isolated intra-luminal air in the absence of significant faecal contamination. Moreover, in the cases discussed above involving colonoscopic perforations, patients will have taken bowel preparative purgatives, reducing or eliminating the presence of intra-luminal faeces. This implies that such perforations are either relatively small or rapidly sealed by omentum. In the case of retroperitoneal perforations, it is possible that the restricted space between tissue planes acts to tamponade luminal leaks against faeces but allow the passage of gas.

Therefore, although the subcutaneous air comes from within the intestinal lumen, most bowel perforations causing subcutaneous emphysema can be managed conservatively. Most of the gas within the subcutaneous tissue planes will gradually resolve within days. The early use of antibiotics is recommended to prevent potential sepsis, although there is not published evidence for this at present. Indeed, where localised septic collections develop subsequently, that may be amenable to drainage by interventional radiological approaches without surgery.

In those cases of subcutaneous emphysema of abdominal origin which require operative management, either for sepsis control or bowel resection, the additional risks of anaesthesia should be considered. General anaesthesia may increase the risks of pneumothorax and tension pneumothorax during artificial ventilation of these patients [53]. In these circumstances, the patient should have unilateral or bilateral intercostal chest drains inserted prior to the anaesthetic in order to mitigate the risks of these complications.

Summary

The early recognition of the signs and symptoms of bowel perforations and prompt intervention is key to improving patient outcomes. Subcutaneous emphysema should prompt a full examination and, especially in the absence of pneumothorax, should raise the possibility of a visceral perforation as a

significant differential diagnosis. We hope that this review raises awareness among physicians and emergency practitioners in order to prevent sepsis and save lives.

Conflict of Interest Statement:

The authors have no conflicts of interest to declare.

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