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CASE REPORT

Samter's triad in childhood: a warning for those prescribing NSAIDs

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Summary

Aspirin-exacerbated respiratory disease (AERD) has been recognized in adults with chronic asthma. Samter's triad is a subset of AERD where adult patients develop nasal polyps, asthma, and sensitivity to aspirin. This condition is thought not to occur before the third decade of life. We report a 13-year-old boy with nasal polyps who suffered a life-threatening exacerbation of asthma during a graded aspirin challenge. Resuscitation required positive pressure ventilation and inotropic support. Our observations confirm that classical Samter's triad can occur in children. We suggest that graded aspirin challenges in children are undertaken in a facility with equipment and staff trained for resuscitation. Consideration should be given to this rare complication when prescribing nonsteroidal anti-inflammatory drugs in the perioperative period. Suspicion of this condition merits referral to an immunologist for desensitization to aspirin.

Introduction

Aspirin-exacerbated respiratory disease (AERD) is a well-recognized clinical phenomenon in adult patients. Up to 21% of adult patients with chronic asthma experience exacerbations after consumption of aspirin or other nonsteroidal anti-inflammatory drugs (NSAIDs) (1). A subset of adult patients with AERD have Samter's triad, an association of nasal polyps with chronic asthma and aspirin sensitivity (2). These patients undergo frequent and extensive upper respiratory tract surgery because of recurrence of nasal polyps and chronic sinus disease.

In the absence of a convincing history of a reaction to aspirin or NSAIDs, the diagnosis of AERD involves a carefully supervised challenge with graded doses of aspirin. While aspirin sensitive asthma can occur in children, it is less frequently diagnosed because long term aspirin is rarely prescribed outside rheumatology practice. NSAID-sensitive asthma has occasionally been reported in children (3,4), although Samter's triad, diagnosed by graded aspirin challenge, has not been previously reported in childhood.

Case report

A 13-year-old asthmatic boy presented with chronic upper respiratory tract symptoms. He had undergone seven myringotomies with placement of ventilation tubes. Nasal polyps were identified by computerised tomography (CT), and he underwent a polypectomy, although a subsequent CT scan revealed pansinusitis (Figure 1). He suffered ongoing nasal obstruction with anosmia. Inhalant allergy tests were negative, and he had normal immunoglobulins. A sweat test was normal. He was using a budesonide nasal spray and was undertaking sinus lavages.

In the absence of an explanation for his nasal polyps, he underwent a graded aspirin challenge (5). Soon after ingestion of 80-mg aspirin, he became wheezy, but without rash or cardiovascular compromise. Initial management involved administration of three doses of intramuscular (IM) epinephrine (0.5 mg doses), two salbutamol (albuterol) nebulizers (5 mg doses), the establishment of intravenous access, and administration of 1-l 0.9% sodium chloride. Oxygen was supplied by a Hudson mask at 8 l·min^{-1}. He was transferred to a

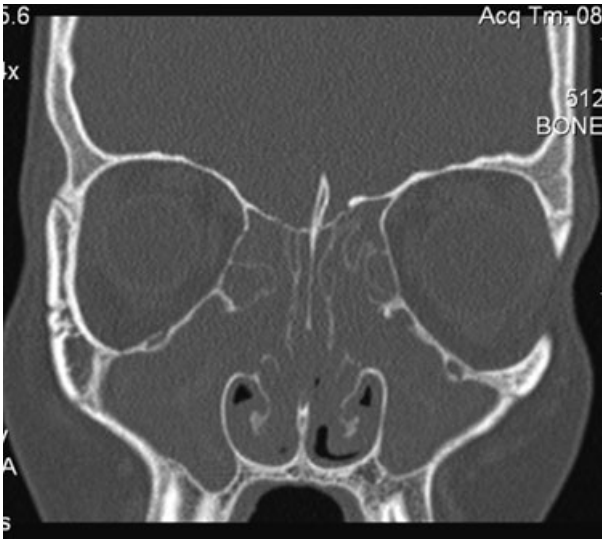

Figure 1 Coronal computed tomography of sinuses showing involvement of the maxillary sinuses and nasal polyps.

tertiary children's hospital emergency department where he continued to deteriorate despite further doses of IM epinephrine, intravenous hydrocortisone (4 $mg \cdot kg^{-1}$), and magnesium (50 $mg \cdot kg^{-1}$). Ketamine (2 $mg \cdot kg^{-1}$) and succinylcholine (1.5 $mg \cdot kg^{-1}$) were used to facilitate intubation. Vocal cords were easily visualized on direct laryngoscopy, and there was no obvious supraglottic or glottic edema. Mechanical ventilation was instituted, an epinephrine infusion (0.5 $mcg \cdot kg^{-1} \cdot min^{-1}$) was started, and sedation was maintained with a propofol infusion (6 $mg \cdot kg^{-1} \cdot h^{-1}$). He was transfered to a pediatric intensive care unit (PICU). A chest radiograph revealed hyperinflation with no focal abnormality. Ventilation improved quickly over the next 90 min, there was complete resolution of his clinical wheeze and active expiratory effort, tidal volumes improved from 4 to 10 $ml \cdot kg^{-1}$ with reducing ventilator pressure support. His epinephrine infusion was rapidly weaned off, and extubation was possible 4 h after presentation. He remained in PICU a further 24 h with no return of symptoms.

Our patient made a full recovery, was commenced on a leukotriene receptor antagonist (montelukast) and received advice on a low salicylate diet. He will be cautiously desensitized to aspirin in the future.

Discussion

The occurrence of a severe anaphylactic reaction to aspirin is of concern to pediatric anesthetists who commonly prescribe nonsteroidal anti-inflammatory drugs in the perioperative period, particularly after otolaryngological surgery. This response was traditionally thought rare in children. Most adult patients experience mild symptoms including nasal congestion, mild exacerbations of asthma, and sometimes gastrointestinal symptoms during graded aspirin challenge. Severe reactions are rare.

The pathogenesis of the disorder is unclear but may be related to an imbalance of prostaglandins and leukotrienes (5). It is thought that there is a reduction in prostaglandin E2 and an increase in leukotrienes leading to inflammatory airways disease. This imbalance is exacerbated by aspirin and NSAIDs. NSAIDs selective for the prostaglandin H_2 synthetase (PGHS)-inducible cyclooxygenase type 2 site (PGHS-2 or COX-2) such as etoricoxib may be tolerated by most patients with Samter's triad but also need a graded challenge under expert supervision (5).

Once AERD is established, upper and lower airways inflammation continues, possibly as a result of salicylate-containing foods in the diet. Some patients who are highly sensitive to salicylates may experience upper or lower respiratory tract symptoms, with foods containing high concentrations of salicylates such as honey. Desensitization to aspirin may reduce the frequency and severity of nasal polyps and improve asthma control. It also allows liberalization of the diet in highly sensitive individuals who experience reactions to foods and substantially reduces the need for frequent upper respiratory tract surgery in these patients. Gastrointestinal adverse effects of long-term aspirin therapy are mitigated with proton pump inhibitors.

This patient had not previously been exposed to aspirin and there was no history of reactions to high salicylate–containing foods. His reaction to aspirin in the context of nasal polyps and asthma confirms that classical Samter's triad can occur in childhood. This single observation suggests reactions to aspirin in children may be at least as severe as those in adults. The use of montelukast prior to aspirin challenge in adults may potentially reduce the risk of severe lower respiratory tract exacerbations. We suggest that graded oral aspirin challenges in children should be undertaken in facilities that can provide full resuscitation. Further, those practicing pediatric anesthesia should be aware of this adverse reaction to NSAIDs. They are trained for resuscitation and can refer to immunologists for further management if the disorder is suspected.

Learning points

1. Aspirin-exacerbated respiratory disease (AERD) is a well-recognized clinical phenomenon that is described

after the third decade of life and is thought rare in children.

2. Children suffering asthma with nasal polyps may exhibit aspirin sensitivity (Samter's triad). A graded aspirin challenge may be needed to confirm diagnosis.
3. This sensitivity can extend to other nonsteroidal anti-inflammatory drugs, and these should be used with caution in children with asthma who also have nasal polyps.

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Conflict of interest

No conflicts of interest declared.

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